“HOW I VIEW THE WORLD”: GENERATING EVIDENCE TO INFORM A PSYCHOEDUCATION INTERVENTION FOR YOUNG PEOPLE WHO HAVE HAD RETINOBLASTOMA

NICOLA RACHEL O’DONNELL

BSC (HONS), PGCERT., MSC., CPSYCHOL.

DOCTOR OF PHILOSOPHY

UNIVERSITY OF YORK

DEPARTMENT OF HEALTH SCIENCES

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ABSTRACT

Retinoblastoma (Rb) is a rare eye cancer typically diagnosed between 0-5 years. Rb can be heritable due to a genetic mutation, or non-heritable, arising from spontaneous mutations. The heritable form carries a higher risk of second cancers and can be passed on to future generations. Survival rates are good in high-income countries but vary globally due to differences in access to medical care and public health resources.

This thesis aimed to understand the experiences of teenage and young adult (TYA) Rb survivors, to generate evidence for a future psychologically informed intervention. Existing research has highlighted that survivors can experience high levels of lifelong distress, yet there are no specific, evidenced-based tools to address the condition's nuances. This thesis is grounded in qualitative methods, highlighting the value of survivors' voices and Patient and Public Involvement. Study one was a reflexive thematic analysis that considered the experiences of survivors. Study two, a systematic review, considered interventions for wider childhood cancer survivors, enabling assessment of the components that are evidenced to be beneficial. The third study was a retrospective content analysis of desired Rb intervention content. These studies generate robust evidence to inform the design of a future psychosocial intervention.

This work found that there is a gap in psychological support for this specific group of cancer survivors within existing healthcare systems. It identified psychosocial needs and challenges faced by Rb survivors, their views on any support received, desired interventions content and tools and will be used to inform future interventions. Findings emphasise the necessity for integrated psychosocial support in routine long-term follow-up care, aligning with the NHS long-term plan. This considers the potential for international collaboration and implementation, and application of the findings to other conditions, with the goal of improving psychosocial care for young survivors of cancer and genetic conditions globally.

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LIST OF ABBREVIATIONS

2-WAY SSS: 21-item 2-way Social Support Scale

ACT: Acceptance and Commitment Therapy

AH: Ann Hewison

AIP: Adaptive Information Processing

ARC: Adversity Restoration Compatibility framework

AWAKE: The Achieving Wellness After Kancer Intervention

AYA STEP: Adolescent and Young Adult Self-Management via Texting, Education, and Plans for Survivorship

BA: Behavioural Activation

BADS-SF: Behavioural Activation for Depression Scale – Short Form

BP: Bob Phillips

CA: Content Analysis

CASP: Critical Appraisal Skills Programme

CBT: Cognitive Behavioural Therapy

CDRISC-10: Connor Davidson Resilience Scale

CES-DC: The Centre for Epidemiological Studies Depression Scale for Children

CHECT: Childhood Eye Cancer Trust

CNS: Central Nervous System

COVID-19: Coronavirus Disease

CSM: Common-Sense-Model of Illness Representations

CTT: Craniosacral Therapy Technique

DH: Debra Howell

DM: Dorothy McCaughan

EBRT: External Beam Radiotherapy

EUA: Examination Under Anaesthetic

FACT-G: Functional Assessment of Cancer Therapy-General

GAD-7: 7-item generalized anxiety disorder scale

GCP: Good Clinical Practice

GOSH: Great Ormond Street Hospital

GP: General Practitioner

GSES: The General Self-Efficacy Scale

HBOC: Heritable Breast and Ovarian Cancer

HLC: Health Locus of Control

HPPS: Heritable Pheochromocytoma and Paraganglioma Syndrome

HRA: Health Research Authority
IAC: Intra-Arterial Chemotherapy
IcamC: Intracameral Chemotherapy
ICRB: Intraocular Classification of Retinoblastoma
ID: Identification
IES-R: Impact of Events Scale—Revised
IRSS: International Retinoblastoma Staging System
ISF: Investigator Site File
IV chemotherapy: Intravenous chemotherapy
IVF: In Vitro Fertilisation
IvitC: Intravitreal Chemotherapy
JLA: James Lind Alliance
LE: Leila Ellis
LFS: Li-Fraumeni Syndrome
LiSat-9: The Life Satisfaction Questionnaire
LTFU: Long-Term Follow Up
MHC-SF: 14-item Mental Health Continuum-Short
MDT: Multi-Disciplinary Team
MMSE: The Mini Mental State Examination
MUIS-C: Mishel Uncertainty in Illness Scale–Community
NHS: National Health Service
NICE: National Institute for Health and Care Excellence
NIHR: National Institute for Health Research
NOD: Nicola O'Donnell
p-IVitC: Precision Intravitreal Chemotherapy
PANAS-X: Positive and Negative Affect Schedule
PCSP: Personalised Care and Support Plan
PEDSQL: Pediatric Quality of Life Inventory
PHQ-9: Patient Health Questionnaire-9 item
POMS: Profile of Mood States
PPI: Patient and Public Involvement
PRISM: Promoting Resilience in Stress Management
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROMIS: Patient Reported Outcomes Measurement Information System
PSP: Priority Setting Partnerships

PSQI: The Pittsburgh Sleep Quality Index

PSS-10: Perceived Stress Scale

PTGI: Posttraumatic Growth Inventory

PTSD-RI: The UCLA Child/Adolescent PTSD Reaction Index for DSM-5

QoL: Quality of Life

Rb: Retinoblastoma

RB1: Retinoblastoma Protein, a tumour suppressor gene

RCMAS: Revised Children’s Manifest Anxiety Scale

RCT: Randomised Controlled Trial

RENEW: RElaxation aNd Exercise for Wellness

RNIB: Royal National Institute of Blind People

RoB: Risk of Bias

RS: Rebecca Sheridan

RSES: Rosenberg Self-Esteem Scale

RTA: Reflexive Thematic Analysis

SCCIP: The Surviving Cancer Competently Intervention

SEQ-C: The Self-Efficacy Questionnaire for Children

SF-36: RAND Medical Outcome Study 36-Item Short Form Health Survey

STAI: State–Trait Anxiety Inventory

STEM: Science, Technology, Engineering, and Mathematics

TA: Thematic Analysis

TTT: Transpupillary Thermotherapy

TYA: Teenagers and Young Adults

UK: United Kingdom

USA: United States of America

YACS: Young adult cancer survivors

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AUTHOR'S DECLARATION

I declare that this thesis is a presentation of original work, and I am the sole author. This work has not previously been presented for an award at this, or any other, University. All sources are acknowledged as references.

The following have been published from the work described in this thesis:

Articles in peer-reviewed journals

- O'Donnell, N., Satherley, R., Dave, E., & Bryan, G. (2023). Fraudulent participants in qualitative child health research: identifying and reducing bot activity. *Archives of Disease in Childhood* 2023;108:415-416. doi: 10.1136/archdischild-2022-325049.

- O'Donnell, N., Phillips, B., Morgan, J.E., & Howell, D. (2024) 'It's not meant to be for life, but it carries on': a qualitative investigation into the psychosocial needs of young retinoblastoma survivors. *BMJ Open* 2024;14:e082779. doi: 10.1136/bmjopen-2023-082779

- O'Donnell, N., Ellis, L., Morgan, JE., Howell, D., Axél Gregersen, P., Willard, V., & Phillips, B. (2025). Psychosocial interventions to improve wellbeing in teenage and young adult post-treatment survivors of childhood cancer: a systematic review. Psycho-Oncology. https://doi.org/10.1002/pon.70081

Articles in press

- O'Donnell, N., Howell, D., Morgan, J.E., & Phillips, B. (in press). "That's all anyone needs really, support": A content analysis of retinoblastoma survivors' psychosocial support needs. Pediatric Blood and Cancer.

- O'Donnell, N., Noret, N., & Phillips, B. (in press). "Words Stick with You": The Lasting Impact of Bullying on Retinoblastoma Survivors. International Journal of Bullying Prevention.

Conference abstracts

- **Children's Cancer and Leukaemia Group Annual Meeting 2023:** 'It's not meant to be for life, but it carries on': A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

  Children with Cancer UK Conference 2023: 'It's not meant to be for life, but it carries on': A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

- Psychosocial interventions to improve wellbeing in teenage and young adult (TYA) post-treatment survivors of childhood cancer: a systematic review

- **SIOP International Conference 2023:** 'It's not meant to be for life, but it carries on': A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

- **Multinational Association of Supportive Care in Cancer (MASCC) International Conference 2024:** 'It's not meant to be for life, but it carries on': A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

- Psychosocial interventions to improve wellbeing in teenage and young adult (TYA) post-treatment survivors of childhood cancer: a systematic review

- **SIOP International Conference 2024:** Generating evidence to inform a psychoeducation intervention for young people who have had retinoblastoma

- Psychosocial interventions to improve wellbeing in teenage and young adult (TYA) post-treatment survivors of childhood cancer: a systematic review

Invited conference talks

- **Division of Health Psychology Conference 2023:** 'It's not meant to be for life, but it carries on':

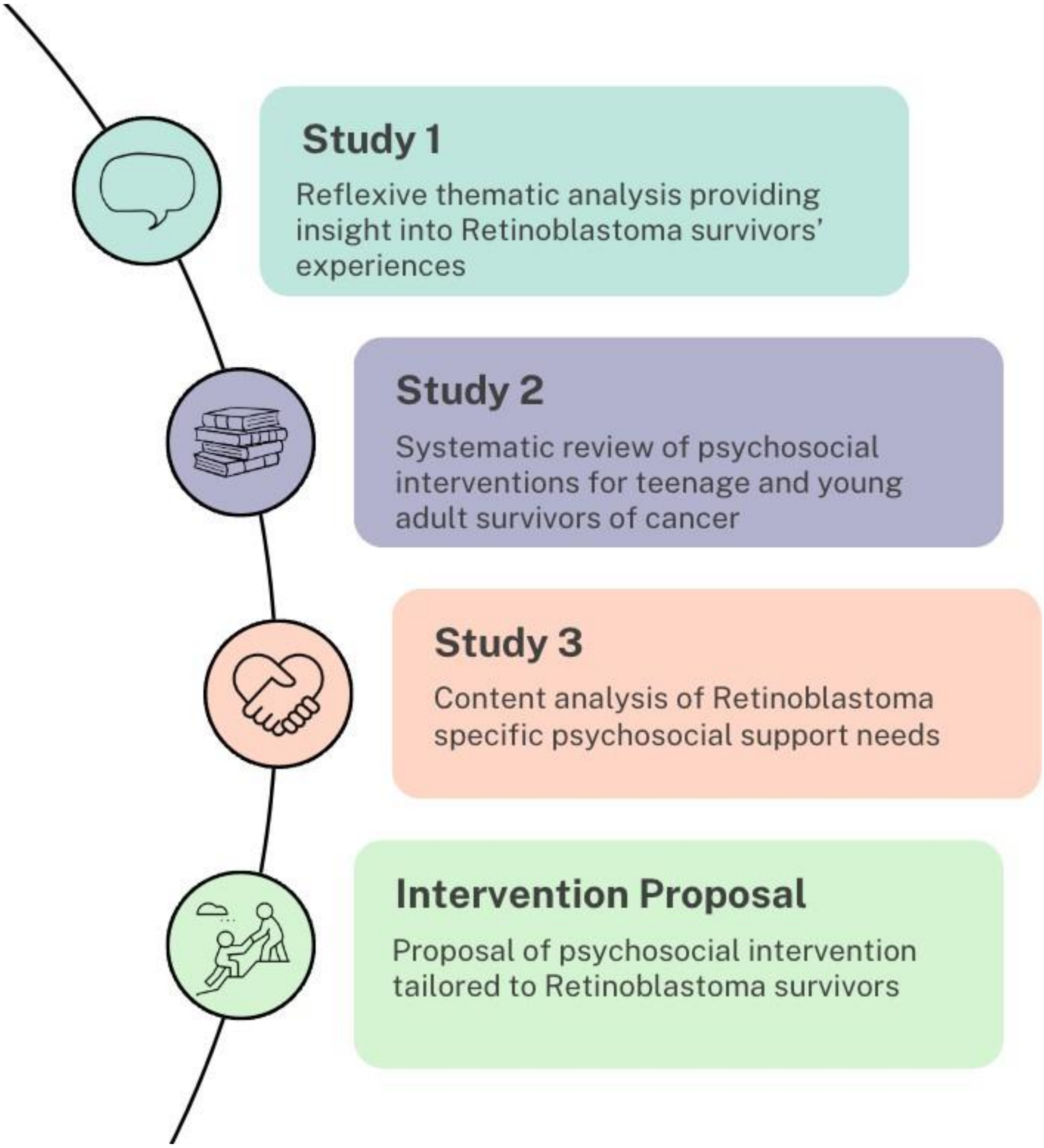
A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

- **British Psycho Oncology Society Conference 2023:** ‘It’s not meant to be for life, but it carries on’: A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

Media reports

- ‘Bullying significant issue for young people in aftermath of eye cancer, study shows’ – Optometry Today, May 2024
- ‘Bullying and retinoblastoma: “I came home one day and asked, ‘What’s a cyclops?’’ – Optometry Today, May 2024
- ‘Bullying significant issue for young people in aftermath of eye cancer, study shows’ – University of York Press, May 2024
- ‘Exploring the Psychological Impact of Retinoblastoma: A Path to Bespoke Interventions for Young People’ – Know the Glow, August 2023
- ‘The psychological and social impact of Retinoblastoma on teenage and young adult survivors: A CPD course’ – Optician, August 2023
- ‘Communicating the Retinoblastoma Experience’ – Optician, February 2023
- ‘Retinoblastoma researcher reaches finals’ – Dispensing Optics, January 2023
- ‘Retinoblastoma PhD researcher becomes finalist in York Talks competition’ – January 2023
- ‘Work of CHECT retinoblastoma researcher highlighted in University of York PhD competition’ – Optometry Today, January 2023

AN OVERVIEW OF THESIS STRUCTURE



Study 1

Reflexive thematic analysis providing insight into Retinoblastoma survivors' experiences

Study 2

Systematic review of psychosocial interventions for teenage and young adult survivors of cancer

Study 3

Content analysis of Retinoblastoma specific psychosocial support needs

Intervention Proposal

Proposal of psychosocial intervention tailored to Retinoblastoma survivors

CHAPTER 1: RETINOBLASTOMA: HISTORY, CURRENT RESEARCH PRIORITIES, AND THESIS OVERVIEW

1.1 WHAT IS RETINOBLASTOMA?

Retinoblastoma (Rb) is a rare cancer of the retina that is diagnosed annually in approximately 40-50 children in the UK, and 8,000 children worldwide (Jenkinson, 2015; Fabian, Rosser and Sagoo, 2018). The median age of diagnosis is between one and two-years (Aerts *et al.*, 2016), with the vast majority of instances diagnosed before the age of five (Jenkinson, 2015). This range is compounded by children with a known family history of Rb, who may be offered prenatal genetic screening (Gerrish *et al.*, 2020) and retinal screening from birth (Wijsard *et al.*, 2021). Despite this, about 10% of children are diagnosed later in life, the likelihood being that tumours were previously undetected (Aguirre Neto *et al.*, 2007; Soliman *et al.*, 2017). Rb survival rates are >95% in high-income countries like the UK, but globally are historically as low as <30%, with areas such as Asia and Africa experiencing up to 70% mortality in children diagnosed (Dimaras *et al.*, 2015). This is due to factors such as sparse treatment access, lack of modern medical intervention that is offered in higher-income countries, and less access to public health resources to raise awareness of early symptoms (Waddell *et al.*, 2015). Having said this, it is hoped that approaches to improving these outcomes through sharing of resources is continually improving survival rates worldwide (Waddell *et al.*, 2021).

1.2 HISTORY OF RETINOBLASTOMA

The first known case of Rb was recorded via autopsy in the Netherlands in 1579. It would be another 150 years before the second case was recorded by Scottish surgeon James Wardrop in 1809. Wardrop pioneered the use of enucleation (surgical removal of the eye) for treating Rb, with alternative treatments at the time including the use of leeches, iodine, mercury, and laxatives (Grossniklaus, 2014). Despite their enucleation treatment, it was the high mortality rate of individuals which prompted further investigation into the role of the optic nerve in Rb.

By the end of the 19th century, treatments had moved on due to the invention of the ophthalmoscope, a tool which allows professionals to see into the structures of the eye, allowing for increased understanding of Rb symptomology and earlier detection (Moschos, 2017) (figure 1). It was this medical progress which allowed doctors to accurately examine Rb tumours for the first time. This advanced understanding and led to the use of external radiation in 1903, then internal radiotherapy via plaque brachytherapy and chemotherapy treatments (Alzahem, 2019), which will be discussed in more detail later in this chapter.

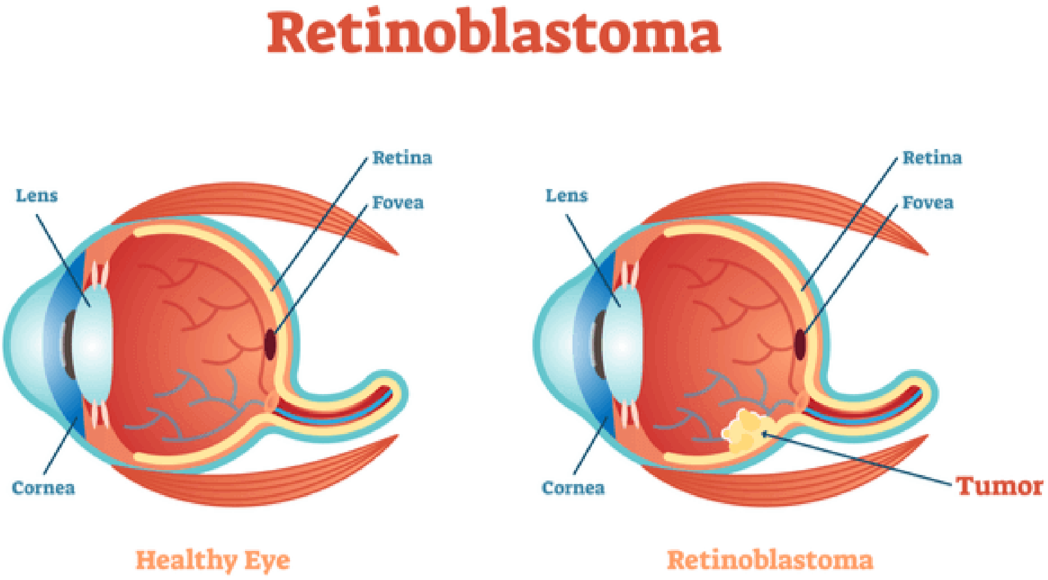


FIGURE 1: ANATOMY OF THE TYPICAL EYE AND AN EYE WITH RETINOBLASTOMA, RETRIEVED FROM: HTTPS://WWW.MEDICAL-SOLUTIONS-BCN.COM/EN/RETINOBLASTOMA-CHILDREN-EYE-CANCER/

1.3 GENETICS

Extensive research into Rb led to the discovery that retinal tumours were derived from retinoblasts, cells which multiply to form the retina in early development, leading to the cancer being named Rb (Albert, 1987). Rb can present unilaterally (in one eye; approximately 60%), bilaterally (in both eyes; approximately 40%), or rarely, trilaterally (in one or both eyes and with a pineal/midline neuroectodermal tumour) (Soliman *et al.*, 2017). The latter two forms are always heritable (de Jong *et al.*, 2014); however, approximately 10% of unilaterally affected patients with Rb have the heritable form. In Rb approximately 45% of patients have a heritable variant of the condition due to a constitutional alteration (genetic mutation) in the *RB1* gene in all cells of the body. This result in a high risk for Rb in both eyes (bilateral disease) (Hülsenbeck *et al.*, 2021). In non-heritable Rb, the *RB1* gene mutation is only occurring in the retinal cells of the affected eye. *RB1* is a tumour suppressor gene, meaning that if it is not functioning correctly due to mutations, Rb develops. Carrying an *RB1* mutation in all cells of the body, as is the case in individuals with heritable Rb, poses numerous challenges including passing on the altered *RB1* gene to future children and having a greater risk of developing second primary cancers later in life, notably sarcomas and melanomas (Meadows and Leahey, 2008; Gregersen *et al.*, 2021). The genetic components of the different types of Rb are discussed in more detail below.

Heritable and Non-Heritable Retinoblastoma

One key theory of the development of Rb is Knudson's 'two hit' hypothesis (Knudson, 1971). This concept describes that mutations of both alleles (two hits) of the *RB1* tumour suppressor gene are necessary for Rb to develop (Karaoui, 2013; Gaikwad *et al.*, 2015). This is the case in both heritable as well as non-heritable Rb. In heritable Rb, the first 'hit' is constitutional (i.e. in all cells of the body), and the second 'hit' is the occurrence of an *RB1* mutation in the retinal cells. In the non-heritable form, Knudson suggested that both *RB1* mutations occur randomly during the development of the child's retina (thus remaining contained to this area of the body). It is possible that *RB1* mutations could occur anywhere, but mainly affect the retina. This is because the *RB1* gene primarily is expressed in the developing retina, meaning that the two 'hits' there invariably cause the development of a tumour.

Risks Associated with Heritable Retinoblastoma

Knudson's theory has also advanced understanding of Rb as it has led to the explanation as to why those with the heritable form of Rb are more likely to develop second cancers (Martínez-Sánchez *et al.*, 2021). Second cancers refer to a new and unrelated cancer occurrence, most often in a different area of the body. It is these second cancers, often bone and soft-tissue sarcomas (Kleinerman *et al.*, 2019), which pose the highest mortality risk for bilateral Rb survivors (Temming *et al.*, 2017).

Unilateral

Unilateral Rb, or tumours in one eye, are usually non-heritable, with only ~18% deemed heritable (Richter *et al.*, 2003; Chen *et al.*, 2014). The non-heritable instances are caused by the alterations of both *RB1* alleles in the tumour (Soliman *et al.*, 2017). For this variant of Rb to occur, the two random 'hits' must occur in a retinal cell's *RB1* gene, resulting in the development of a tumour (Price *et al.*, 2014). As these occurrences are by chance, there is little risk of individuals going on to develop second, unrelated tumours (Price *et al.*, 2014). Having said this, some individuals with inherited unilateral Rb have underlying germline mutation, meaning that they are at risk of tumour development in the unaffected eye (Shields *et al.*, 2008). For these reasons, it is also important to offer genetic testing to individuals who are unilaterally affected to ensure that they are aware of the form of Rb that they have, and any risks that these pose (Chinnery, 2018).

BILATERAL

In children with bilateral disease, a single *RB1* allele is mutated in every cell of their body (constitutional *RB1* alteration) (Fabian, 2018). This mutation not only puts individuals at risk of losing vision and/or their eye(s) but also predisposes them to developing second cancers throughout their lifetime (Francis *et al.*, 2018; Hill *et al.*, 2018). Individuals with *RB1* mutation also have a 50% risk of passing this genetic sequence to each future child, meaning that there are future implications for this group to consider when reproducing in adulthood. Other patients with Rb have mosaicism for an *RB1* mutation. In these individuals, the *RB1* mutation occurs sporadically after conception during embryonic development, and as a result, the *RB1* mutation is only present in specific cells of the body, not the whole of the body.

Although all patients with bilateral Rb have heritable disease, most children diagnosed are the first in their family to present with the condition, this is known as *de novo* occurrence (AlAli *et al.*, 2018). Approximately only 5-10% of individuals knowingly having a family member who was also treated for Rb (inherited, heritable disease) (Hill *et al.*, 2018). Children who have a parent with heritable Rb are estimated to have around a 45% chance of being affected themselves, as inheriting the *RB1* gene mutation does not inevitably cause a tumour despite high penetrance (AlAli *et al.*, 2018). As described above, the remaining 90-95% of individuals have a '*de novo* mutation'; a genetic *RB1* alteration that occurs in the germ cell from either the mother (egg), the father (sperm) or during fertilisation (Kato *et al.*, 1994). Therefore, despite having heritable disease, they are the first (*de novo)* within a family to develop tumours as opposed to being an unaffected carrier in instances of reduced penetrance *RB1* alterations (Mallipatna, Marino and Singh, 2016). It is vitally important that all *RB1* mutations are identified and classified as heritable, as this will provide clinicians and parents with the information needed to make important treatment and life choices for the affected child and future relatives.

TRILATERAL

Individuals with heritable Rb have an approximately 5% susceptibility of developing what is known as an intracranial midline primitive neuroectodermal tumour, meaning that their Rb is considered 'trilateral' (de Jong *et al.*, 2014). This variant of Rb was first considered in 1977, where medics noticed that some individuals with bilateral Rb also had intracranial tumours (tumours in the central nervous system) (Jakobiec *et al.*, 1977). Of all the Rb variants, children diagnosed with trilateral Rb have the worst outcomes, accounting for the majority of Rb deaths in the West (Shields, Spahr and Slavich, 2020). A systematic review of 211 instances of trilateral Rb between 1977 and 2015 found that the median survival time post-diagnosis was 10.3 months, with a 5-year survival rate of 15.7% (Yamanaka, Hayano and Takashima, 2019). Despite this, with advancing treatments including combination chemotherapy, and improved awareness leading to earlier diagnosis, the prognosis for children diagnosed with trilateral Rb is improving (Ortiz and Dunkel, 2016).

CLINICAL FEATURES

As the most common paediatric eye cancer, Rb is unique in that it is the only central nervous system (CNS) tumour that is visible to lay people without the use of in-depth imaging (Dimaras and Corson, 2019). The most common presenting clinical feature of Rb is whitening of the pupil (leukocoria) (Vempuluru and Kaliki, 2021). This is created by the reflection of light from the retinal tumour, often noticed for the first time by parents or carers when looking at their child in certain light. It is also commonly visible when photographing a child, although there are concerns that this will become less common due to the rise in use of phone photography as opposed to flash photography. Secondary to this, many parents notice that their child has a ‘squint’ (strabismus) (Balmer and Munier, 2007). A lesser proportion of children are deemed to have ‘atypical’ features of Rb, showing symptoms of ‘red eye’ (orbital cellulitis due to infection of the eyelid) (Abramson *et al.*, 2003), inflammation of the eye (uveitis or hypopyon) (All-Ericsson *et al.*, 2007; Blitzer *et al.*, 2021), a pool of blood in the white of the eye (hyphema) (Zafar, Zaheer and Khan, 2017), change of colour in the iris (heterochromia) (Abu-Ain *et al.*, 2019), visible blood vessels in the white of the eye (rubeosis) (Pe’er *et al.*, 1997), damage to the optic nerve (glaucoma) (Rao, Honavar and Reddy, 2019), or severe eye damage causing the eye to shrink (phthisis bulbi) (Mullaney *et al.*, 1997; Balmer and Munier, 2007). Individual or a combination of these presentations are often reasons for parents to take their child for an optician’s or general practitioner’s (GP) consultation, where the possibility of Rb may be considered for the first time.

STAGING

When a child is diagnosed with Rb, one of the first tasks of the clinical team will be to ‘stage’ the level of disease in one/both eyes. Staging involves the use of eye examination and imaging tests to determine how extensive the cancer is, particularly whether it has spread outside of the retina. Staging is important because it provides a standardised process which informs a child’s treatment plan and allows doctors to establish the prognosis of saving a child’s eye, their vision, and ultimately their life (Tomar *et al.*, 2020).

The first classification system was proposed in the 1960s (Reese and Ellsworth, 1963), with the primary purpose of assessing the likelihood of being able to save the child’s eye following external beam radiotherapy. At this time, treatments for Rb were radically different to what they are now, with intravenous chemotherapy not introduced for another ~30 years (Fabian, 2018). In 2006, a newer staging system was introduced, considering modern treatment options and cultural differences to produce an international guide for use in the assessment of the spectrum of Rb conditions (Chantada *et al.*, 2006). The International Retinoblastoma Staging System (IRSS) classifies Rb instances from stage 0, where disease is located within the eye only, to stage IV, where the primary cancer has metastasised, often to the lymph nodes, bone, bone marrow, and CNS (table 1) (Dimaras and Corson, 2019). Alongside this classification, the Intraocular Classification of Retinoblastoma (ICRB) (Shields *et al.*, 2006) groups Rb instances from A (very low risk of spread of disease) to E (very high risk) (table 2). This grouping is based on the size of the tumour, where it is located, and whether there are additional factors such as seeding (where the tumour disperses out of the retina into other parts of the eye) (Munier, 2014). If a child is impacted in both eyes, each eye will be staged separately.

TABLE 1: THE IRSS CLASSIFICATION AND STAGING FAMEWORK (FABIAN, 2018)

Stage	Clinical Description
0	Patient treated conservatively
I	Eye enucleated, completely resected histologically
II	Eye enucleated, microscopic residual tumour
III	Regional extension
	a. Overt orbital disease
	b. Preauricular or cervical lymph node extension
IV	Metastatic disease
	a. Haematogenous metastasis (without central nervous system involvement)
	1. Single lesion
	2. Multiple lesions

b. Central nervous system extension (with or without any other site of regional or metastatic disease)

1. Prechiasmatic lesion
2. Central nervous system mass
3. Leptomeningeal and cerebrospinal fluid disease

TABLE 2: ICRB A-E FRAMEWORK (SHIELDS, 2006)

Group	Clinical Description
A (very low risk)	Retinoblastoma ≤ 3mm (in basal dimension or thickness)
B (low risk)	Retinoblastoma >3mm (in basal dimension or thickness) or - Macular location ≤ 3mm to foveola - Juxtapupillary location ≤1.5mm to disc - Additional subretinal fluid ≤ 3mm from margin
C (moderate risk)	Retinoblastoma with: - Subretinal seeds ≤ 3mm from tumour - Vitreous seeds ≤ 3mm from tumour - Both subretinal seeds and vitreous seeds ≤ 3mm from tumour
D (high risk)	Retinoblastoma with: - Subretinal seeds > 3mm from tumour - Vitreous seeds > 3mm from tumour - Both subretinal seeds and vitreous seeds > 3mm from tumour

E (very high risk)	Extensive retinoblastoma occupying >50% of globe or with: - Neovascular glaucoma - Opaque media from haemorrhage in anterior chamber, vitreous or subretinal space - Invasion of postlaminar optic nerve - Choroid (>2mm), sclera, orbit, anterior chamber

1.4 TREATMENT

Treatment for Rb has historically involved radiotherapy, chemotherapy, local ophthalmic therapies and enucleation, otherwise known as surgical removal of the eye (Balasopoulou *et al.*, 2017; Gündüz *et al.*, 2020). These treatments have been, and are, largely very effective, resulting in a cure in around 95% of instances in Western societies (Wong *et al.*, 2014). Despite this, many factors including these treatments, the cancer itself, and genetics, can produce long lasting effects such as reduced vision, facial changes, and second cancers (Sethi *et al.*, 2014; Temming *et al.*, 2016). Even as recently as the 1980s, children were often treated with external beam radiotherapy, which had high success at curing Rb, but often left individuals with wide-ranging and life-long side effects (Shields and Shields, 2010). The causes of these late effects are complex and difficult to separate, making it hard to know the true cause and thus any associated risk factors. Furthermore, due to the change in treatments for Rb over the decades, with more attempts to save the eye in modern interventions, psychological challenges may have changed (Gündüz *et al.*, 2020). For these reasons it is important to outline the various treatment modalities that a child may be offered, as this will impact their cancer experience and could potentially influence risk of developing late effects. This in turn may influence how they think and feel about themselves, others, and the world as they grow older, shaping the psychosocial support they might require.

A recent review of modern Rb treatments (1990-2020) (Ancona-Lezama, Dalvin and Shields, 2020) outlined the following widely used treatment modalities:

INTRAVENOUS CHEMOTHERAPY (IV CHEMOTHERAPY, CHEMOREDUCTION)

First developed in 1953, IV chemotherapy is still deemed a primary treatment option for Rb treatment, with multiple chemotherapy drugs (often Carboplatin, Vincristine, and Etoposidel JOE) administered in up to six cycles (Chen *et al.*, 2018). IV chemotherapy is praised for ascertaining control over Rb tumours, retaining vision, and preventing trilateral disease, seemingly with no increased risk of second cancers (Kaliki and Shields, 2015). IV chemotherapy is often used as a first line treatment for individuals with bilateral disease, with a large cohort study showing that the majority of individuals treated in this way remained stable and cancer-free over the following 20 years (Shields *et al.*, 2020).

INTRA-ARTERIAL CHEMOTHERAPY (IAC)

Utilising the same or varied chemotherapeutic agents as IV chemotherapy, IAC is a highly successful treatment (Marr *et al.*, 2012) which involves injections into the ophthalmic artery (Abramson *et al.*, 2012). The benefits of this over IV chemotherapy are around the direct delivery of drugs to the site of the tumour, minimising the potential of side-effects including second cancers (Shields and Shields, 2010; Turaka *et al.*, 2012). Having said this, IAC is not successful in treating Rb for all individuals, and it does not have a preventative role for trilateral disease. Individuals undergoing IAC must be monitored for systemic toxicity, ensuring that the eye(s) are not damaged (Manjandavida *et al.*, 2019).

INTRAOCULAR CHEMOTHERAPY

INTRAVITREAL CHEMOTHERAPY (IVITC)

IvitC is performed by injecting chemotherapy (often melphalan and topotecan) directly into the tumour (Ancona-Lezama, Dalvin and Shields, 2020). This treatment option was introduced in 2003 as an additional treatment for individuals with higher risk tumours who would otherwise have had their eye(s) removed (Ancona-Lezama, Dalvin and Shields, 2020). IvitC has been shown to be particularly successful for individuals with recurrent, dispersed tumour seeds, achieving 91% cancer stability and preserving 84% of eyes (Munier *et al.*, 2012; Manjandavida and Shields, 2015).

PRECISION INTRAVITREAL CHEMOTHERAPY (P-IVITC)

A variation of IvitC, p-IvitC is a recent treatment developed in 2018 as an alternative treatment for tumour seeds and involves the localised injection of chemotherapy into these directly (Yu *et al.*, 2019). Although shown to be helpful in treating more complex tumours, the risk of retinal toxicity is higher than alternative treatments (Ghassemi and Shields, 2012; Camp, Lally and Shields, 2019).

INTRACAMERAL CHEMOTHERAPY (ICAMC)

As with the other treatments in this category, IcamC was developed to treat individuals with Rb seeding, specifically aqueous seeding; the move severe form of seeds that a decade ago, would have likely resulted in enucleation (Kaliki and Shields, 2015). In a review of IcamC in 11 Rb patients, 100% had tumour control at 7 months post-treatment, but 45% had required enucleation due to relapse at 24-month follow-up (Kaliki, 2021).

FOCAL THERAPIES

CRYOTHERAPY

Often used to treat smaller Rb tumours or seeds, cryotherapy involves a triple-freeze-thaw technique which kills malignant cells through freezing at temperatures up to -90°c (Warda *et al.*, 2022). Although rarely a standalone treatment (Ancona-Lezama, Dalvin and Shields, 2020), the success rate is high, with one review highlighting a cure rate for 90% of tumours that are less than 3mm in diameter (Chawla, Jain and Azad, 2013).

TRANSPUPILLARY THERMOTHERAPY (TTT)

TTT is a focal treatment devised in the last 30 years, which like Cryotherapy, is often used to treat smaller (up to 1.5mm diameter), lower grade Rb tumours (Cieślik *et al.*, 2021). TTT uses thermal energy to the affected area, delivering up to 60°c heat to the tumour (Shields *et al.*, 1999). Often used in conjunction with chemotherapy, TTT has been shown to be successful in treating small tumours, with one review of 42 children finding 79% efficacy at achieving tumour control over two years, and 100% success in preserving the affected eye across 30 children, who had no further incidences of cancer (Murat *et al.*, 2015).

EXTERNAL BEAM RADIOTHERAPY (EBRT)

Although not the current primary treatment of choice, EBRT has historically been a well-utilised treatment for Rb (Ancona-Lezama, Dalvin and Shields, 2020). This treatment targets high-dose radiation at Rb tumours through an external radiotherapy machine (Kim and Park, 2015). As mentioned above, EBRT is largely successful in treating Rb, with a large retrospective case series reporting that 81.4% of Rb patients treated with EBRT were tumour-free and had preserved eye(s) at 1 year follow-up, and 53.4% at 10-year follow-up (Abramson *et al.*, 2004). Despite this, EBRT can led to a multitude of late side-effects including dry eye syndrome, cataracts, radiation retinopathy (retinal disease), optic neuropathy (damage to the optic nerve), facial deformity. Most concerning is the high risk of second cancers, with a recent review reporting this to occur in 53% of EBRT-treated patients by the age of 50 (Yousef *et al.*, 2021). For these reasons, EBRT is now only considered as a last resort prior to enucleation (Kim and Park, 2015).

PLAQUE RADIOTHERAPY

Often used as a secondary treatment for higher-grade, multiple tumours, plaque radiotherapy treats Rb through placing fragments of radioactive material on or near the tumour site to deliver continuous, concentrated radiation (Shields *et al.*, 2001). The rationale behind choosing plaque radiotherapy over EBRT is to spare wider eye tissue and treat seeding near the front of the eye. This procedure has also shown high levels of efficacy at treating more complex Rb but is also used sparingly due to the side-effects mentioned above, with a recent study reporting over 50% risk of post-treatment haemorrhage (Ancona-Lezama, Dalvin and Shields, 2020). Furthermore, it is of note that plaque radiotherapy involves two operations (to implant and then remove the plaque), meaning it is a highly invasive option for the child (Abramson, 2005). Not only can this be physically challenging but can leave children with high levels of distress and procedural anxiety that, if left unsupported, can last beyond the period of treatment, and remain detrimental to wellbeing throughout life (Nunns *et al.*, 2018).

ENUCLEATION

Enucleation, the surgical removal of the eye, is a long-standing treatment option for Rb that has been utilised since the 1800s (Jenkinson, 2015). Enucleation is always performed under general anaesthetic and involves the insertion of an orbital implant to the eye socket. Although radical, this treatment is indicated for two different reasons. Primary enucleation is offered for high grade, advanced, and/or unilateral Rb, where there is significant risk to a child’s life or where the vision in that eye is irredeemably lost. The alternative is secondary enucleation which is utilised when other treatment options have failed or where tumours are difficult to control in an eye with very poor vision (Bowman and Bowman, 2018). Although primary enucleation is less common in the UK, it remains the most common first line treatment for Rb worldwide, likely due to the lack of extensive treatment options offered in the West (Gibbs, Reynolds and Shea Yates, 2022). Losing an eye(s) is a distressing and life-changing experience, and it is plausible that the type of Rb diagnosed may impact parental choices about having their child’s eye enucleated. For parents who have prior experience of Rb, for example if they themselves have been treated, they may decide to enucleate their child’s eye earlier to avoid undergoing extensive medical procedures that can lead to late effects. In addition, if a child has very limited or no vision in an eye, this can also influence parental treatment choice. In this circumstance, many parents choose enucleation as first-line treatment, avoiding other treatments in the attempt of saving a non-functioning eye.

OCULAR PROSTHESIS

First established in 1884, prosthetic or artificial eyes have been used after an enucleation to improve comfort and cosmetic appearance (Mourits *et al.*, 2018). Prosthetics have developed hugely over time, with heavy and painful ‘Ekblephara’ eyes (comprising of a metal band worn around the head) used in the 16th century (Danz, 1990) to 20th century glass eyes (Chinnery, 2018) and 21st century custom-fit, acrylic, or even 3D-engineered robotic eyes which dilate in a way that mimics a typical pupil (Cafiero-Chin, Marques and Danz, 2015; Strathearn, 2021). Living with a prosthetic can be challenging, requiring regular removal and cleaning of the eye and socket to avoid infection. An additional issue (albeit one that may diminish as quality of implants and prosthetics improves) is the way that prosthetic eyes move, with many individuals feeling self-conscious about its’ functional deficits (Korani *et al.*, 2020). Research suggests that individuals living beyond Rb who have experienced enucleation struggle with particular psychosocial challenges, with increased issues related to appearance, vision, and peer relations. It has therefore been recommended that specific psychosocial support is needed for this subset of Rb survivors (Banerjee *et al.*, 2020).

1.5 TREATMENT CENTRES

Due to the rarity of Rb diagnoses, there are only two specialist treatment centres in the United Kingdom (UK): The Royal London Hospital shared with Great Ormond Street Hospital (GOSH) and Birmingham Children’s Hospital. Children across the whole of the United Kingdom are treated here, with around 20 children a year receiving treatment in London and 30 in Birmingham (NHS, 2013), mostly due to the travel times from much of the UK being shorter to the Midlands than to the capital, particularly from Wales, Scotland, and Northern Ireland. Both hospitals contain specialist Rb teams which are classified as being ‘supra-regional multidisciplinary teams’ (Jenkinson, 2015) containing professionals from a range of disciplines including paediatric oncologists, ophthalmologists, geneticists, clinical nurse specialists, and more. The clinical teams work closely with the Childhood Eye Cancer Trust (CHECT), who fund a family support worker at each hospital who is available to provide emotional and practical assistance to every family whose child is diagnosed, equating to over 2,000 hours to 525 families each year (CHECT, 2022).

Worldwide, there are 180 Rb treatment centres (figure 2), these range from hospitals in high income areas such as St Jude Children's Research Hospital in Tennessee, USA, SickKids Hospital in Toronto, Canada, Sant Joan de Déu in Barcelona, Spain, and Helsinki University Hospital in Helsinki, Finland. Hospital sites in upper middle-income areas include Beijing Tongren Hospital in Beijing, China, S. Fyodorov Eye Microsurgery Federal State Institute in Moscow, Russia, and Hospital de Pediatria Garranhan in Buenos Aires, Argentina. Finally, centres in lower middle-income countries include Ruharo Eye Center in Mbarara, Uganda, King Hussein Cancer Center in Amman, Jordan, Narayana Nethralaya Eye Hospital in Bengaluru, India, and University of Ghana Medical School in Accra, Ghana (Tomar *et al.*, 2021).

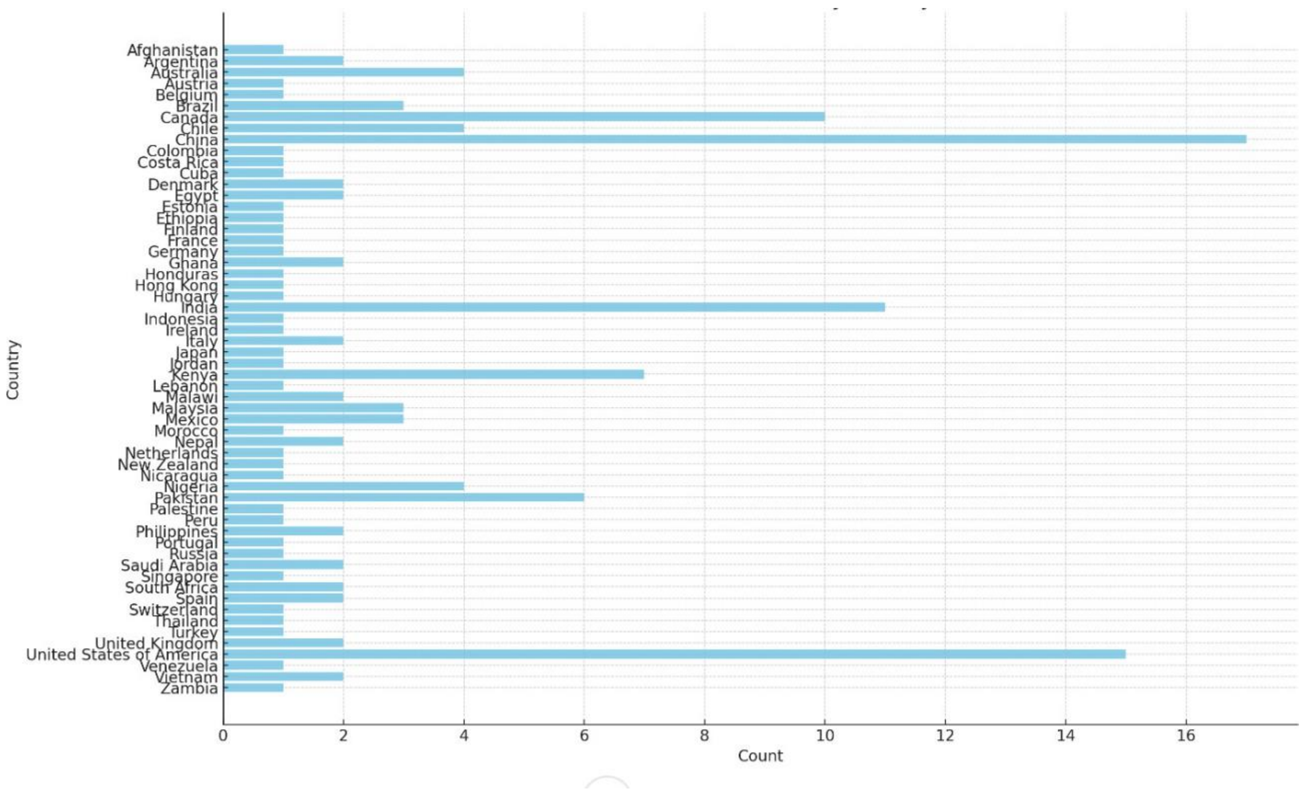


FIGURE 2: RETINOBLASTOMA TREATMENT CENTRES BY COUNTRY

1.6 UK SERVICE STRUCTURE AND TREATMENT MANAGEMENT STRATEGY

As the focus of this PhD will be around young people treated in the UK, the service structure and treatment management strategy utilised in this area will be discussed in more detail. If a child is suspected of having Rb, they will most often have been seen by their GP or health visitor and referred to a specialist centre via their local hospital (NHS, 2013). Having said this, if Rb is already known to be present within a child's family, screening will be arranged prenatally or shortly after birth. For unexpected instances, primary care clinicians are likely to conduct a red reflex test to determine ocular alignment and pupillary reflex (Li *et al.*, 2010). As per National Institute for Health and Care Excellence (NICE) guidelines, if 'normal' red reflex is not found in both eyes, a very urgent ophthalmologist appointment will be requested to occur within the following 48 hours (NICE, 2021). Once Rb is confirmed, children will be treated by specialist centres at the Royal London Hospital and Birmingham Children's Hospital.

After referral, further investigation can take place. This will always involve a detailed eye examination under anaesthetic (EUA), a short procedure involving retina photography and ultrasound. It is during this operation that a firm diagnosis, including staging of disease, can be made. If bilateral disease is confirmed, genetic screening will also be recommended for close relatives, who will be monitored until the age of 7 years (NHS, 2013). For the affected child, diagnosis will then be followed by the commencement of a treatment plan which may involve any and multiple options discussed above. For children whose treatment involves IVC, their care is often shared with their local paediatric oncology principal treatment centre, to allow care to be delivered closer to home. Once treatment is complete, children will be monitored with regular EUA until they are one year tumour free for unilateral Rb and two years for bilateral Rb.

It is recommended that individuals remain under the care of the National Health Service (NHS) long-term follow-up clinics (LTFU) until they are at least 16 years, but preferably for life. Here the individual will be monitored for both recurrence of Rb and, as time goes on, screen for any negative impact of treatment such as second cancers. LTFU is also an opportunity for individuals to be given education about their cancer, behavioural recommendations to prevent likelihood of second cancers (e.g., not smoking), genetic counselling (if applicable), as well as psychological support tailored to their age and stage of development.

1.7 PSYCHOLOGICAL RESPONSE TO RB CANCER DIAGNOSIS AND TREATMENT

The psychological and social impact of cancer has been referred to as a 'hidden toxicity', a result of diagnosis and treatment which is overlooked and understudied (Bangs, 2024). Rb is distinctive in that the vast majority of instances are diagnosed before a child reaches the age of five. Early childhood is a time of rapid development, with attachments with caregivers, cognitive and linguistic abilities, and emotion regulation evolving in line with the environment (De Young *et al.*, 2021). With Rb impacting very young children, most individual's treatment will be complete before they are old enough to process the experience with meaningful language, their developmental stage hindering their ability to verbalise their experiences of medical trauma. This begins in infancy, where children have minimal skills to communicate distress, to toddlers who are more aware of control and are prone to feeling powerless during threatening situations, and pre-schoolers who need support to regulate their emotions, and are prone to 'magical thinking' such as *"if I was good, I wouldn't have gotten ill".* All of these experiences have the potential to distort memories and complicate psychosocial outcomes as they become teenagers and young adults (Wizansky and Bar Sadeh, 2021). Trauma has long-been considered in terms of an integrated mind-body response, with the Adaptive Information Processing (AIP) model stating that the human brain stores unprocessed trauma in a dysfunctional way (Shapiro, 2001). This can result in psychological distress, maladaptive thoughts or behaviours, and even physical pain, with the stress of trauma overwhelming the nervous system with difficult memories remaining in the primitive, non-verbal, highly emotive part of the brain. Because treatment for Rb is conducted at pivotal time of cognitive development, this theory of trauma may be particularly relevant. This has been shown in children who have experienced traumatic medical intervention unrelated to Rb, with interpretations of medical procedures linked with high degree of threat, frightening associations with pain, and immature understanding of their experience including confusing feelings of betrayal towards parents, who can become associated with medical procedures (Nabors *et al.*, 2013). For these reasons, parents are usually the ones who experience strong psychological reactions to their child's cancer diagnosis at the time of treatment (Belson *et al.*, 2020), often feeling powerless, guilt, and trauma (Locatelli, 2020). Due to the small number of instances diagnosed each year, many parents will not have heard of Rb prior to their child's diagnosis (Wang *et al.*, 2016). Research suggests that even for parents who have knowledge of Rb or have been treated for it themselves, it is not uncommon for many to be unaware of many elements of the cancer and treatment. This is especially as they would have been treated at a very young age, and because treatments have changed so rapidly over time that protocols are likely to be very different (Hill *et al.*, 2018).

Although the stress and distress of parenting a child with cancer is widely documented (e.g., Jones *et al.*, 2018; Carlsson *et al.*, 2019; van Warmerdam *et al.*, 2019; Öhman, Woodford and von Essen, 2021) there are limited studies which specifically focus on the psychological impact of Rb within families, particularly on the diagnosed child themselves. Of the few studies which have investigated Rb specifically, findings are similar to those in the general cancer population, with parents and children experiencing a range of negative psychosocial outcomes. These include the broader anger, anxiety, and sadness experienced by many cancer diagnoses, but further explicit distress about losing an eye(s) through Rb (Ek, 2000; Wang *et al.*, 2016; Beddard *et al.*, 2020). It has been shown that parenting a child with Rb can also lead to poorer social outcomes, with isolation and loneliness common due to having to manage a stressful situation that many cannot relate to, as well as having to spend a lot of time away from ‘normal life’ situations due to frequent hospital attendance, often very far away from home (Hamama-Raz, Rot and Buchbinder, 2012). Despite an extensive literature search, only one paper could be found which uses qualitative methods to understand childhood Rb survivors’ views, although this too considered parental perspectives and only focused on the experience of living with a prosthetic eye (Gibbs, Reynolds and Shea Yates, 2022). In interviews with seven children between the ages of four and nine, most explained that despite being too young to have memories of enucleation, they valued play programs in which they learnt about their condition and were able to gain understanding of what had happened to them. Additionally, the children’s parents highlighted the need for their children to develop resilience strategies, cope with changes to self-image and manage peer interactions as they grew up. Overall, the paper highlighted the need to give children and young people a sense of control through providing them with age-appropriate information about Rb and how to live well beyond diagnosis and treatment. It also suggested that future research should aim to identify ongoing support and information for young people to facilitate coping.

Another area of literature focuses on the life-long impact of Rb on individuals as they progress through adulthood. However, this is limited and of the few studies conducted, all focus on adult survivors as opposed to young people. A recent review of literature considered qualitative views of health-related quality of life for heritable Rb survivors (Belson *et al.*, 2020). Findings suggested that early psychological intervention is needed, particularly highlighting the voice of the child. This came from findings that parents more often reported different perspectives on quality of life for their child than their child did for themselves, indicating that any psychological support provided may not be accessible or acceptable if based on parental experiences alone. This, along with the other studies presented below, highlight the need for further research into the experiences of young people who have had Rb, which is what this thesis aims to do.

1.8 PSYCHOEDUCATION INTERVENTIONS

WHAT ARE PSYCHOEDUCATION INTERVENTIONS?

Psychoeducation interventions involve therapeutically providing individuals with relevant and up-to-date information about their health to support them to live with and/or beyond a diagnosis. Interventions are likely to include facilitating peer discussion and social support and drawing upon the evidence-base to offer problem-solving and coping skills training (Barsevick *et al.*, 2002). This approach is seen as an important element in the treatment of many physical or psychological conditions such as oncology, anxiety, and trauma (Vassilopoulos *et al.*, 2013; Thompson and Young-Saleme, 2015; Kolaitis, 2017). Psychoeducation can take many forms and has typically involved app or website-based interventions, group, and individual workshops (Steinglass, Ostroff and Steinglass, 2011; Mak *et al.*, 2018; Seidel *et al.*, 2020; Harrer *et al.*, 2021; Jones *et al.*, 2021; Sekse *et al.*, 2021; O'Donnell *et al.*, 2022). There are, however, many barriers to the success of psychoeducation, and it is a crucial consideration for young people that this is provided in an age-appropriate and accessible way to ensure engagement and good psychosocial outcomes (Bekker, Griffiths and Barrett, 2017).

USE IN PAEDIATRIC AND ADOLESCENT CANCER SURVIVORSHIP POPULATION

Research suggests that children and young people who have had cancer experience more mental health challenges than the general population (Friend *et al.*, 2018). In an attempt to improve psychosocial outcomes, psychoeducation interventions are one tool that has been utilised. Although the potential late effects of child and adolescent cancers can vary dependent on disease type, age of diagnosis and many other key variables, survivors are generally at greater risk of anxiety and/or depression, difficulty re-integrating with peers, attention and cognitive difficulties, and later, problems with sexual function (Siegwart *et al.*, 2022). Below, I summarise three examples of intervention across these different variables to demonstrate impact. Many interventions are based on Cognitive-Behavioural Therapy (CBT) techniques, and a small study from the Netherlands utilising this approach found that it led to positive outcomes (Stam *et al.*, 2009). Over six sessions, young people benefited from peer support, had an opportunity to understand more about their diagnosis, took part in guided relaxation around medical procedures, and underwent role play exercises to build confidence. Findings indicated high efficacy in improving skills, social competence, and positive thinking amongst the group. Equally, a psychoeducation intervention with a similar format, but specifically for children on chemotherapy, found that even when delivered over a relatively short period of four sessions, anxiety symptoms reduced (Megasari and Wulandari, 2024). These findings were supported by self-report and questionnaire scores and were scheduled around each participants' chemotherapy session to assess efficacy. Importantly, improvements were found in children as young as 4 years, with 72% of all participants rating their anxiety levels as mild at the end of the sessions, comparative to 'severe' scores which were most common pre-intervention. Finally, a recent Randomised Controlled Trial (RCT) considered the efficacy of a 12-week, online intervention for young people (aged 19-40) who had survived childhood cancer (Fagerkvist *et al.*, 2024). This intervention was focused on sexual dysfunction following cancer and showed potential in reducing emotional distress in relation to sexual function. Having said this, adherence was low and recommended that future interventions must consider more tailored content to better address this needs of this specific group.

CHILDHOOD CANCER

A RCT investigated the efficacy of interactive group psychoeducation for children under 12 with leukaemia (Day *et al.*, 2021). A treatment group of 26 and delay (waitlist control) group of 32 were offered four, 2-hour sessions which addressed topics such as general understanding of leukaemia, treatment side effects and the importance of a healthy lifestyle. Findings suggested that children found the intervention helpful, to the extent that the review recommended that it be offered as part of routine hospital care. Focusing on the outcome quality of life (QoL), the treatment group showed higher scores post-intervention with no improvement in the delay group during the same period, highlighting the efficacy of the intervention. Despite this positive finding, the small sample size must be acknowledged, as well as the fact that all participants were treated in the North of the UK, reducing population validity. Furthermore, the inclusion of young people who were still on active treatment may have skewed findings. Another trial considered the acceptability and efficacy of a coping intervention for children with a variety of diagnoses and an average age of 14 (control group) and 13 (experimental group) (Wu *et al.*, 2014). Using a modular approach, participants were first taught to identify stressors related to their illness, then given self-help methods to generate coping, before providing the opportunity to share experiences of positive coping. The intervention was also supplemented by an online game and Facebook support page to encourage two-way communication. Results suggested that coping skills did not differ significantly between groups but considered that the small sample size may have been a reason for this. Equally, authors considered that individual differences, particularly when managing a distressing situation like cancer diagnosis and treatment, can make it likely for participants to disengage in positive coping strategies and use behaviours like distancing, avoidance, and distraction in their place. These challenges are likely to be apparent regardless of intervention, and therefore must be considered when supporting this group.

Teenage and Young Adults on treatment for Cancer

Aiming to improve cancer-related knowledge using a psychoeducational video game, 375 13–29-year-olds currently being treated for leukaemia, lymphoma, or solid tumours were recruited to either intervention or control groups. Findings highlighted that the intervention group had significantly larger increases in knowledge after receiving the ‘Re-Mission’ videogame. Despite this, it was found that the videogame was not used to its’ full potential, with the average participant only completing two of the 20 available levels. It may, therefore, be that the knowledge-increase was due to a combination of the game, but also through stimulating interest in carrying out individual research into cancer, treatment, and self-care strategies. Overall, researchers concluded that the effect size of the intervention on knowledge was too small to invest resources into the production of a similar tool for use in routine cancer treatment. It may be, however, that developing this study using a qualitative approach might allow greater understanding into the components that were or were not helpful, feeding into a future revised version.

A RCT exploring a novel psychoeducation intervention provided young adults with fertility-related distress following cancer with a 12-week, web-based programme (Micaux *et al.*, 2021). This used videos of survivor's stories, a discussion forum focusing on topics such as fertility distress and managing relationships, and quizzes around fertility after cancer. The intervention group of 64 participants received access to this immediately, whilst the control condition of 60 participants accessed standard care which may or may not have included fertility-specific support. Three months post-intervention, small differences were found in the distress levels of the intervention group, with this group showing lower distress and greater knowledge than the control group about fertility-related issues. However, overall findings showed that the website had little impact on fertility-related distress, suggesting that web-based interventions may be less effective than those delivered face-to-face or in combination. It is also important to note that although RCT is considered 'gold standard' evidence, this is debatable when used to measure psychosocial interventions due to the lack of control that is possible through lack of double-blinding. As the researchers acknowledge, this can lead to inconclusive assessment of interventions of this nature, and suggest the further testing is required to measure true effect.

USE IN PAEDIATRIC AND ADULT GENETIC CONDITIONS

HERITABLE CANCER PREDISPOSITION SYNDROMES

A recent study tested the efficacy of a psychoeducation resource for children with inherited cancer predisposition syndromes, meaning that individuals have a genetic mutation which increases their chance of developing cancer (Schlub *et al.*, 2021). Focusing on children with either Li-Fraumeni Syndrome (LFS) or Heritable pheochromocytoma and paraganglioma syndrome (HPPS), bibliotherapy (the use of stories and literature) was utilised to develop a story to help children understand their condition. Ages of children ranged from two to 18, and interviews found that the story's reframing of stressful situations and normalising of experiences was helpful. It was found that the story was not distressing to children of any age, with younger children identifying themselves in the story's characters and taking comfort in this. This psychoeducational technique also appeared to allow children to raise difficult conversations with their parents, opening up discussions about frustrations, blame, and uncertainty around their conditions. This study was the first of its' kind to use bibliotherapy as a psychoeducational intervention for children with heritable cancer predisposition syndromes. The positive findings highlight the benefits of using this technique within psychoeducation, but due to most of the participants being young children, it has been recommended that this approach is tested across older ages and with different conditions. Although a small study of 12 parents (albeit from a small pool of individuals at risk of rare cancer predisposition syndromes), this research clearly highlights the value of psychoeducation interventions. It also showcases how to modify risk information to be understood by children as young as five, providing a useful framework for Rb interventions.

A systematic review and meta-analysis of the efficacy of psychoeducation interventions for adults with Heritable Breast and Ovarian Cancer (HBOC) syndrome and Lynch Syndrome was conducted (Baroutsou *et al.*, 2021). Considering a range of interventions (e.g., booklets, websites, webinars, and apps) and theoretical frameworks (e.g., Health Belief Model, Motivational Interviewing, Theory of Stress and Coping, Ottawa Decision Support Framework), the primary outcome of all included studies was to improve family communication. Describing 14 papers, it was found that providing knowledge about cancer genetics was essential, with knowledge being linked to greater coping. Due to disparity in intervention design and delivery, there was not enough data to conclude whether the interventions for this population improved psychosocial outcomes. However, it was highlighted that there was no difference between online and face-to-face interventions, which widens access to many and creates greater possibilities for the creation of future, innovative interventions. As with many papers, a limitation was the lack of participants from ethnically and socially diverse backgrounds. This is problematic as these individuals remain unheard and potentially may be experiencing barriers to accessing psychoeducation support.

HERITABLE CONDITIONS UNRELATED TO CANCER

Two meta-analyses considered the effects of Cognitive Behavioural Therapy (CBT) and mindfulness-based psychoeducation to manage distress for individuals diagnosed with neurodegenerative conditions: Huntington’s disease, Parkinson’s disease, or multiple sclerosis (Ghielen *et al.*, 2019). Results from a total of 19 studies across all conditions found that psychoeducation had a small to moderate effect size on reducing distress, which was less effective than drug therapy. Critically, the studies included were of largely low quality, with risk of bias present in most and no RCTs for Huntington’s disease available for analysis. Additionally, it is vital to note that psychoeducation for these populations is vastly different to individuals with/who have had cancer. This is because neurodegenerative conditions are likely to reduce individuals’ ability to learn and benefit from techniques like CBT, therefore making it difficult to draw comparisons with Rb. When considering more similar conditions, such as Li-Fraumeni syndrome, existing research has highlighted a need for psychoeducation interventions (Werner-Lin *et al.*, 2020; Barnett *et al.*, 2022; Wilsnack *et al.*, 2022) yet there is a lack of them developed and available to test. This highlights the importance of developing tailored psychoeducation interventions for individuals with rare genetic disorders, as their specific cognitive challenges and needs are not sufficiently addressed by existing approaches, underscoring the need for higher-quality research and intervention development.

RECOMMENDATIONS FOR USE IN RETINOBLASTOMA

It is important to understand the psychological impact of living beyond both heritable and non-heritable Rb, with existing research highlighting the need for better psychosocial support in this population (Gregersen *et al.*, 2021). Previous research has indicated that anxieties in this population relate to personal health, survival, and impact on family (Belson *et al.*, 2020; Feng *et al.*, 2020). Equally, psychotherapeutic programmes for children currently undergoing treatment for Rb have been shown to be beneficial in helping children and their families to cope with recurring traumatic medical experiences, like examinations under anaesthetic (Rich *et al.*, 2024). However, as described above, little is known about the specific psychological challenges experienced by teenagers and young adults, who must manage the effects of Rb in a period of life when psychosocial support is crucial to the healthy development of individual, social, and sexual identity (Zebrack, 2011). This lack of knowledge is in part because of the small number of diagnoses made each year, meaning that there is a limited insight into the particular risks and challenges of teenage and young adult life after Rb. For those who have an additionally diagnosed cancer pre-disposition syndrome e.g. Rb1 mutation (Ketteler *et al.*, 2020) or Li-Fraumeni Syndrome (Kumamoto *et al.*, 2021), there is not only a fear of recurrence but also of second primary cancers (Kamihara *et al.*, 2017).

Wider literature exploring the psychosocial needs of young people living beyond cancer indicates that support must be given to support the creation of a new normal, even when future outcomes are uncertain (Gibson *et al.*, 2016). Considering illness and long-term effects of treatment at this crucial developmental stage can cause significant hindrance to identity and mental wellness (Bradley Eilertsen *et al.*, 2012; McArthur, Strother and Schulte, 2017). It is also recognised that young people who have experienced cancer are at heightened risk of identity distress. This concept describes concerns around how the self is perceived in terms of psychosocial development, independence, appearance, social relationships, and stigma, both now and in the future (Barbot *et al.*, 2021). The likelihood of distress is heightened when there have been associated changes in physical appearance due to illness and/or treatment, as for many individuals who have had Rb (Zebrack, 2011; Kearney and Ford, 2017; Pearce *et al.*, 2020). Whilst these are all important to consider, appropriate intervention for the unique psychosocial challenges associated with life after Rb requires further investigation.

1.9 CURRENT RESEARCH PRIORITIES

Determining the most acceptable and accessible support is important to patients and families, clinicians, policy makers and commissioners of services. Intervention development requires a thorough understanding of the challenges faced by people living beyond Rb, and a detailed examination of possible theoretical and evidence-based approaches to address the challenges and support them. There is widespread evidence to suggest that psychosocial support is warranted, and wanted, in the Rb population. Recent qualitative studies have found that both young people and adults would like access to psychologists who have specialist knowledge of Rb-related challenges, preferably offering psychoeducation at early stages to prevent potential Rb-related psychological difficulties (e.g., Belson *et al.*, 2020; Gregersen *et al.*, 2021).

1.10 CURRENT POLICY FOCUS

Published in 2019, the NHS Long Term Plan was developed (Kapur, 2020) to set out a framework for improving patient experience in many areas, including the provision of personalised care. The ambition of personalised care is to provide choice and control to the individual receiving treatment, moving away from a standardised approach towards health and considering individual differences. Within this plan was the more specific Long-Term Plan for Cancer, which aims to improve patient quality of life outcomes, increase positive patient experiences, and reduce variation and inequality. Working in partnership with the NHS is leading cancer support charity Macmillan, who are advocating that, off the back of the NHS' proposal, all individuals living with or surviving from cancer should be offered a Personalised Care and Support Plan (PCSP) (Macmillan, 2020). This states that individuals who are/have faced cancer should have access to supportive conversation, health and wellbeing resources, individual support needs, and digital tools and information regarding all aspects of cancer and the impact it can have. The current PhD would fit well into this NHS agenda, offering targeted, co-produced, psychosocial support based upon Rb survivors' experiences.

I have also considered the research priorities set by the James Lind Alliance (JLA), an organisation funded by the National Institute of Health and Care Research (NIHR) which sees clinicians, patients, and carers collaborating through Priority Setting Partnerships (PSPs), identifying research areas of need in healthcare. For each health condition, a 'top ten' list of priority areas for research is produced. In the last decade, several areas relating to the focus of this PhD have been identified as priorities, demonstrating the need for novel research in this area. This has included 'sight and vision' (2013), 'mental health in children and young people' (2018), 'living with and beyond cancer' (2018), 'detecting cancer early' (2019), and 'children's cancer' (2022) (James Lind Alliance, 2022). Within these areas, key questions relevant to this PhD have been raised, stated in figure 3 below.

FIGURE 3: JAMES LIND ALLIANCE PRIORITIES FOR RESEARCH

- *How can retinoblastoma be identified, prevented, and treated in children? (2013)*
- *How can young people be more involved in making decisions about their mental health treatment? (2018)*
- *What are the most effective early interventions or early intervention strategies for supporting children and young people to improve mental resilience? (2018)*
- *What are the most effective self-help and self-management resources, approaches or techniques available for children and young people with mental health issues? (2018)*
- *At what ages would it be most effective to start to educate children and young people about mental health? (2018)*
- *How can patients and carers be appropriately informed of cancer diagnosis, treatment, prognosis, long-term side-effects and late effects of treatments, and how does this affect their treatment choices? (2018)*
- *What are the short-term and long-term psychological impacts of cancer and its treatment and what are the most effective ways of supporting the psychological wellbeing of all people living with and beyond cancer, their carers and families? (2018)*
- *How can the short-term, long-term and late effects of cancer treatments be (a) prevented, and/or (b) best treated/ managed? (2018)*
- *What are the social, financial and economic impacts of living with and beyond cancer – how does it affect families, relationships, finances, work and use of NHS services? (2018)*
- *What are the best ways to cope with the fear and anxiety about cancer returning (combining self-management approaches, treatments and psychological support)? (2018)*
- *How can we predict who is at risk of developing mental health conditions in people living with and beyond cancer (e.g. depression) and what are the best ways of supporting those with mental health conditions? (2018)*
- *Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like? (2022)*
- *How can we make being in hospital a better experience for children and young people? (like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care)*
- *What are the best ways to ensure children and families get and understand the information they need, in order to make informed decisions, around the time of diagnosis, during treatment, at the end of treatment and after treatment has finished? (2022)*

- *What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive? (2022)*
- *What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)? (2022)*
- *What is the psychological and social impact of cancer and treatment on children and their families during treatment and in the long-term; what factors affect these impacts? (2022)*
- *What are the best ways of making sure people who had cancer as a child receive the information they need about the long-term effects of cancer and treatment? (2022)*
- *What are children's and survivors' experiences of the side effects and long-term effects of cancer treatment? (2022)*
- *What are the best ways to support children as they get older, and their needs change, to understand and take responsibility for their health, and to live with the long-term effects of cancer and treatment? (2022)*

1.11 AIMS AND OBJECTIVES OF THE PHD

The main aim of this thesis is to understand the experiences of young Rb survivors to generate evidence for a psychologically informed, educational intervention to support this group in the future. To achieve this, the following objectives will be met:

1) To understand the views of teenagers and young adult survivors of Rb regarding their psychosocial needs
2) To explore Rb-associated psychosocial challenges arising in adolescence/young adulthood
3) To seek the opinions of teenagers and young adults regarding the support they have had/would have liked
4) To identify potential content of a psychosocial intervention through qualitative interviews and existing cancer interventions
5) To draw upon the above to generate robust evidence to inform a novel psychosocial intervention

1.12 QUALITATIVE METHODS APPLIED TO THIS RESEARCH

To develop a psychoeducation resource that is meaningful to the Rb population, I feel strongly that this research should capture and be led by their voices. For this reason, I have chosen to use primarily qualitative methods, seeking out the views and experiences of young people who have survived Rb and working together to address the research aims above. Full details about the qualitative methodology utilised in this thesis can be found in chapter two.

As an overview, I aim to utilise a social constructionist approach, seeing the narratives of the young people I work with as a direct reflection of their experiences, whilst acknowledging the context that they inhabit (Willig, 2019). This is an approach that has been shown to be beneficial when working with survivors of cancer, gaining deep understanding of how individuals make sense of their experiences of being treated and living beyond their diagnosis. An example of this can be taken from a recent study exploring women's experiences of ovarian cancer (Staneva *et al.*, 2018). This paper acknowledged the various discourses that are present within the context of cancer, including the potential impact on identity, understanding, and ways of belonging in a society which prioritises futuristic planning and being 'well', despite being so unwell and facing a life of uncertainty. It struck me that authors of this paper recommended that healthcare professionals look for hidden stories of vulnerability, and how access to this is a privilege made possible through using qualitative methods.

As with any approach, there are criticisms of qualitative methods which must be acknowledged. Gaining true understanding of individuals' experiences takes time, and it is important that the resources are given to both the collection and analysis of data to ensure this is both accurate and appropriate. It is also true that due to the nature of qualitative work requiring researchers to dig deep into individuals' experiences, ethical issues must be considered. It is imperative that research participants, parents, and researchers are protected from harm and that distress protocols are in place, providing support if needed. Unlike quantitative methods, qualitative approaches often involve more subjective analysis, meaning great care is needed to ensure reliable and valid research is created. This can be helped through the use of qualitative research checklists such as that from the Critical Appraisal Skills Programme (CASP, 2018). These frameworks encourage researchers to consider key questions when conducting qualitative research, addressing key areas such as recruitment strategy and the relationship between the researcher and their participants. All of these factors will be crucially important when working qualitatively with young people who have had Rb, considering the role that I will have in understanding such a sensitive subject. This may be particularly poignant when aiming to recruit participants from additional minority groups, who may find it difficult to share their experiences or have the emotional energy to consider what they've been through (Morse, 2020).

1.13 THEORETICAL APPROACH

To date, there have been limited attempts to explore how young people who have had Rb may respond to this lifechanging event, both psychologically and socially. This gap in the literature might seem surprising, given the profound and life-altering nature of this diagnosis, which affects not only physical health but also psychological well-being and social functioning. Whilst much attention has been given to the medical aspects of Rb, less is known about how young survivors adapt to the emotional and social challenges they face throughout their lives, particularly as they transition through key developmental stages. This thesis aims to fill that gap by exploring how young individuals process and respond to their experiences with Rb, drawing on established frameworks such as the developmental psychopathology approach (Sroufe and Rutter, 1984) and the concept of health locus of control (Norman and Bennett, 1996).

DEVELOPMENTAL PSYCHOPATHOLOGY APPROACH (SROUFE AND RUTTER, 1984)

The developmental psychopathology approach, as articulated by Sroufe and Rutter (1984), offers a comprehensive framework for understanding the psychological and social responses of young individuals who have faced life-altering experiences, such as diagnosis and treatment of Rb. This theoretical model goes beyond static notions of development, emphasising the dynamic interplay between individual experiences and biological factors across the lifespan. By considering how individuals process their life events cognitively and affectively, this approach acknowledges the complexity of human development, recognising that major milestones, such as the onset of puberty and transition to adolescence, as well as the establishment of significant relationships, education and career development, and how these events continue to shape individuals well into adulthood.

Moreover, this perspective underscores the concept that development does not stop once adulthood is reached, but rather continues indefinitely, continually adapting throughout life. This perspective resonates particularly with the population of my PhD, as children who have experienced a cancer diagnosis during their early years may face long-lasting psychosocial impact that can manifest differently across various stages of life. By embracing a lifespan perspective, this approach highlights the nuanced ways in which individuals navigate and respond to significant life events, offering valuable insights into the experiences of young people who have experienced Rb and the subsequent challenges they may encounter throughout their development. Thus, considering the developmental psychopathology framework throughout this thesis provides a robust theoretical foundation for exploring the multifaceted impacts of Rb on individuals' psychological and social well-being across the lifespan.

HEALTH LOCUS OF CONTROL (NORMAN AND BENNETT, 1996)

In this thesis, the concept of health locus of control (Norman and Bennett, 1996), which pertains to individuals' beliefs regarding the extent to which they can influence their health outcomes and cope with challenges, is considered. Existing literature has consistently highlighted a prevalence of external locus of control among cancer survivors compared to the general population (Hodges and Winstanley, 2012; Brown *et al.*, 2015, 2017; Wilson *et al.*, 2018). This means that survivors are more likely to believe that their health outcomes are largely determined by external forces beyond their control, such as fate, chance, or the actions of others, rather than by their own behaviours or decisions. For these individuals, the rarity of a childhood cancer diagnosis combined with the unpredictable nature of cancer often reinforces a sense of powerlessness, making it difficult for them to feel in control of their health and future well-being. This phenomenon can be attributed, at least in part, to the pervasive fear of cancer recurrence, which often appears insurmountable and beyond individual control. This aspect is perhaps particularly pertinent to individuals with heritable Rb, where the inherent genetic predisposition amplifies concerns about recurrence and exacerbates feelings of helplessness. In addition to this, the very nature of a childhood cancer diagnosis usually means that locus of control is held by their parents and health care providers, meaning that there is never an opportunity for the diagnosed individual to be in ‘control’ of their disease experience. By holding in mind the health locus of control within the context of Rb survivors, this thesis aims to shed light on the unique psychological dynamics at play and contribute to a deeper understanding of how individuals with heritable Rb navigate their health perceptions and coping strategies in the face of uncertainty and perceived lack of control.

1.14 STRUCTURE OF THIS THESIS

Conventionally, PhD theses often follow a structure that starts with a systematic review outlining existing literature, followed by studies exploring the aims of the work. However, this approach is less suitable for this thesis because the lack of existing research on the psychosocial experiences and interventions for survivors of Rb, meaning that starting with a review would be an insufficient starting point. Additionally, the complexities and unique needs of this topic necessitate a structure that prioritises survivor voices and experiences and the iterative development of research objectives. Although this introductory chapter offers an overview of the existing research that situates this PhD, I decided to structure the thesis in the format outlined below. This approach ensures that the structure not only fills existing gaps in literature but also centres the lived experiences and voices of the Rb community.

Following this chapter is an in-depth consideration of the qualitative methodology used in this thesis, providing context to the subsequent studies. This focus on qualitative methodology is critical because it allows for a nuanced exploration of survivor perspectives, which are often underrepresented in the literature. By highlighting my chosen methodological framework, the thesis ensures that the research is grounded in approaches that prioritise the lived experience, aligning with the overarching goals of this work. Chapter three is a reflexive thematic analysis conducted as part of study one, followed by an overview of Patient and Public Involvement (PPI) throughout this PhD. I decided to start with this study as it was imperative to me that I structure this thesis with survivor's voices at the forefront, as this approach is both methodologically and ethically significant. Methodologically, centring survivors' voices ensures the research captures the lived realities and diverse perspectives of those directly impacted by Rb, fostering a deeper and more authentic understanding that can later inform an intervention. Ethically, it acknowledges and values their experiences, empowering survivors by prioritising their narratives in the research process. In addition, conducting an in-depth qualitative study allowed me to understand what is most important to the Rb community and subsequently informed my systematic review question. Chapter five is a systematic review of childhood cancer interventions, providing a narrative synthesis of findings. Following this is the third and final study of this thesis, a retrospective content analysis of data collected as part of study one. The aim of this analysis was to identify content for a future psychoeducation intervention for young Rb survivors. Chapter seven provides a discussion and conclusion of my findings, synthesising the results of the studies to demonstrate how the chosen structure supports the research objectives. This chapter ties together the unconventional approach by showing how each component builds on the previous to address gaps in the field. Chapter eight's final reflections and considerations situates this work within the wider literature and illustrates its contributions to future research, policy, and clinical practice, highlighting the importance of an iterative, survivor-focused methodology in shaping meaningful interventions.

By structuring the thesis in this way, I aimed to ensure that the research is directly informed by the foundational discussions in this introductory chapter. The decision to begin with a reflexive thematic analysis of qualitative data was directly informed by the gaps highlighted in this introduction, particularly the need to situate survivor perspectives in order to fully understand their psychosocial experiences. This introductory chapter establishes the theoretical and empirical context for these discussions, for example, common psychological responses to Rb, or wider childhood cancer, diagnosis and treatment. This later shaped the research questions and the topics explored in participant interviews and focus group discussions. Specifically, it provides novel insight into the emotional and psychological impact of living with and beyond Rb, as well as the absence of tailored psychosocial interventions. This led to a research design where I have prioritised open, survivor-led discussions to explore what aspects of their experiences were most meaningful and relevant to them. By using this qualitative approach as the foundation for the subsequent studies in this thesis, I aimed to ensure that survivor voices actively shape the direction of the research, rather than being secondary to pre-determined academic frameworks.

1.15 OVERVIEW OF STUDIES

STUDY ONE

The first study involved a two-phase qualitative process utilising focus groups with teenagers (age 13-19 years) and individual interviews with young adults (age 20-29 years) with a history of Rb. The aim of this study was to understand the psychosocial experiences of young Rb survivors, including any challenges that resulted from this.

STUDY TWO

The second study involved a systematic review undertaken to identify suitable cancer psychoeducational interventions to adapt for teenagers and young adults with Rb. Previous research has highlighted that psychoeducation is needed in Rb populations, so findings from study one, which provided Rb-specific knowledge, were used to inform the clinical questions to focus on in the review.

STUDY THREE

The third study was a retrospective content analysis on data collected as part of study one. This focuses on potential intervention content, as suggested by survivors and shaped by their experiences, wants, and needs. Originally study one was due to focus on potential intervention content too, but due to the large amount of rich and novel data obtained in study one, this led to the development of two separate research focuses.

FUTURE STEPS

Combining the findings from the three PhD studies, a proposal for a novel psychoeducation intervention will be co-developed with Rb survivors post-doctorally. These co-researchers may be a mix of participants from study one, and PPI experts. This will be designed to be signposted by late effects clinics and the Childhood Eye Cancer Trust (CHECT) and accessed by teenagers and young adults to support their emergence into adulthood. As the current research will also be designed for young people who may have visual impairments, it is imperative that practical implementation of psychoeducation is also considered (Yildiz and Duy, 2013; Elsman *et al.*, 2019). It is hoped that the proposed intervention can be tested using a feasibility RCT in the future.

CHAPTER 2: METHODOLOGY

This chapter describes the research methodology chosen to address the aims of this thesis, which is to generate evidence for a psychologically informed, educational intervention to support teenagers and young adults living beyond Rb. Below I will explain my choice of methodology, as well as the ontological and epistemological positions that I take.

2.1 QUALITATIVE RESEARCH

Qualitative research has been said to provide *'rich and compelling insight into the real worlds, experiences, and perspectives'* of individuals (p.1, Braun & Clarke, 2014). Qualitative research is concerned with words and spoken language as data, seeking to interpret patterns of meaning. This is in contrast to quantitative research, which focuses on the use of numbers, statistics, and relationships between variables (Braun and Clarke, 2013).

RESEARCH PHILOSOPHY PARADIGMS

> *'A paradigm is inclusive of several components that can be categorised as the following: Ontology, Epistemology, Methodology, and Methods'* (Scotland, 2012; as cited in Alharahsheh & Pius, 2020*).*

The purpose of much qualitative research is to produce rich, in-depth data that focuses in on a specific group of people; it's aim is not to be representative or generalisable to the wider population, but to contribute to understanding of the context and experiences of a set group. Encompassing a wide range of qualitative techniques, such as Interpretative Phenomenological Analysis, Ethnography, Grounded Theory, and Thematic Analysis to name a few, it can be utilised in different forms, on a paradigm that ranges from 'Big Q qualitative research' to 'small q qualitative research' (Kidder and Fine, 1987). The position of Big Q sees qualitative research as tools and research values that are aligned purely with qualitative values, including researcher subjectivity, context, and inference of meaning. Small q, on the other hand, is aligned with postpositivist research values that typically sit within quantitative research; hence valuing generalisable, objective and replicable data (Braun and Clarke, 2021a). I am more aligned with Big Q research and I understand that I bring my own context to this research topic. For this reason, I have included extensive discussion around my position as a researcher later on in this chapter, considering how this will influence how participants communicate with me, and influence how I analyse the data.

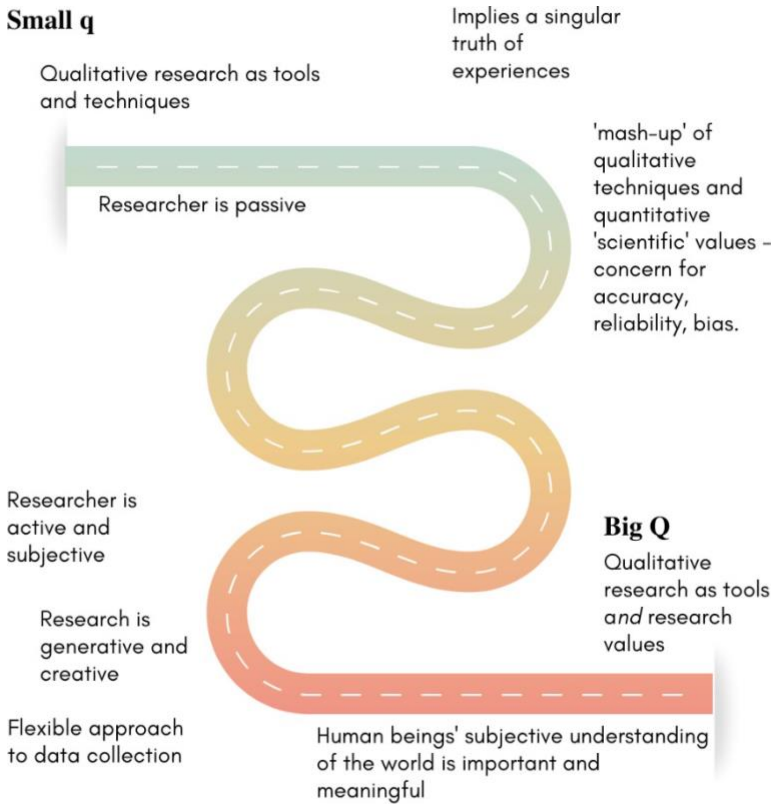


FIGURE 4: THE DIFFERENCES BETWEEN SMALL Q AND BIG Q IN QUALITATIVE RESEARCH, INFORMED BY A LECTURE GIVEN BY VICTORIA CLARKE AS PART OF THE 'FOUNDATIONS OF QUALITATIVE RESEARCH' SERIES (CLARKE, 2021)

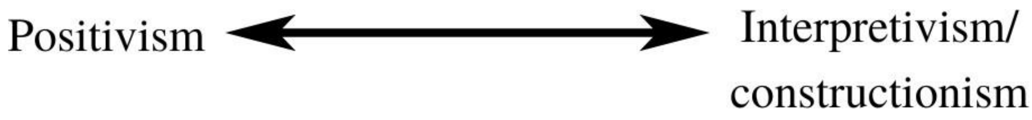
ONTOLOGY – WHAT CAN WE KNOW?

‘Ontology raises basic questions about the nature of the human being in the world’ (Denzin, 2005).

When reading about ontology, I was struck by the question of whether things exist outside of our minds, or whether the world is constructed by our thoughts. These two arguments represent the realist and relativist positions in ontology, with critical realism considering that reality exists independently of our minds (Ryan, 2018). Scientists who subscribe to this way of thinking attempt to identify phenomena to suggest that this is true (Bergin et al., 2010). Critical relativists on the other hand believe that reality is subjective and therefore there must be multiple interpretations and realities. Scientists who work in this way seek to understand these subjective experiences and do not subscribe to only one truth (Levers, 2013).

I can see the points of view of both realist and relativist ontology, acknowledging that reality exists independently of our perceptions but that we inevitably all have our own interpretations of this. This belief is grounded in the idea that, despite our varied personal experiences and interpretations (including our cultural norms, development, attachment styles, social reality, and more), there is an underlying reality that is consistent and observable. In my research with individuals diagnosed with the same type of cancer, Rb, I approach the study with the understanding that while each person's experience is unique, these experiences are grounded in a shared, objective reality of the disease and its potential impacts. By acknowledging this shared reality, I aim to uncover patterns and consistencies that contribute to a broader understanding of the condition and its effects on those affected.

EPISTEMOLOGY – HOW CAN WE KNOW?



‘Epistemology can be defined as how reality is being known by the researcher’ (Alharahsheh and Pius, 2020) .

Epistemology is concerned with how knowledge is gathered and from where; in particular, there is a focus on the world of and position of the researcher, giving insight into their interpretation of the data. For these reasons, their philosophical beliefs are important. Four commonly considered epistemological perspectives include: positivist/postpositivist (observing and measuring reality), interpretive/constructivist (multiple realities), critical (empower and identify information free from legal, social, or political restriction), and postmodern/post structural (deconstructing of previous truths) (Merriam, 2009).

It is important for me as a researcher to situate myself in an interpretivist position. This is because I believe that I am inherently part of my research through the fact that I am interpreting the data. My interpretations cannot be separated from my position and my context, and therefore my research will be influenced by this. Some might say that this makes my research subject to bias (Noble and Smith, 2015); it does, but that is acceptable as I acknowledge and own this position.

Bias refers to the subjective perspectives, preconceptions, and personal influences that a researcher brings to their study. Taking this approach values the subjective nature of knowledge and seeks to understand and articulate how the researcher's position impacts the interpretation of the data. I would also argue that psychology and the study of human beings is inherently biased and subjective (particularly when working qualitatively), and it would be unrealistic to claim otherwise. Equally, bias does not necessary equate to ‘bad’ if it is acknowledged and measures are taken to reduce this, and therefore prevents results from becoming misleading.

This also fits well alongside my ontological position, as interpretivism has an inherently relativist perspective, with the understanding that experiences are subjective and context is critical. In terms of my research topic, I am exploring the multiple experiences and realities of what it is like to grow up beyond a diagnosis and treatment for Rb. It would be inappropriate to take a positivist approach to this, as I would be presuming knowledge of my participants and not allowing for exploration of different truths. As someone who has not experienced a personal diagnosis of any cancer, including the one which I am researching, I aim for this thesis to be co-led by those with diverse lived experiences, not confining it to one perspective or understanding of what it is like to have had Rb.

Methodology – how can we find out?

‘a strategy or design by which the researcher maps out an approach to problem-finding or problem solving’ (Jamshed, 2014).

Thematic Analysis

Commonly utilised within health and social care settings and health psychology (Braun *et al.*, 2023), thematic analysis (TA) is an approach which involves identifying and developing thematic patterns within a data set (Campbell *et al.*, 2021). Originally developed in the 1970s by Gerard Holton (Clarke and Braun, 2014), TA became commonly used within social sciences after a 2006 paper by Victoria Clarke and Virginia Braun became highly influential (Braun and Clarke, 2006). Described as *'a starting point for your journey, not a map'* (Braun, Clarke and Hayfield, 2022), TA encompasses a broad selection of methods, providing suggestions for how you might conduct your analysis, rather than providing a step-by-step guide. Broadly speaking, TA methods can be categorised into three types: coding reliability approaches (e.g. Guest et al., 2020), codebook approaches (e.g. Brooks et al., 2015), and reflexive approaches (Braun and Clarke, 2021). The latter of these has been chosen for study one of this PhD, so will be focused on in more detail in chapter three. These approaches can best be thought of as being on a continuum, with coding reliability at one end, and reflexivity at the other.

This thesis is inductive by approach, seeking to generate meaning and understanding from the data that I collect. This is opposed to a deductive methodology, where an existing theory is sought to be tested. Inductive methods are commonplace in thematic qualitative research (Javadi and Zarea, 2016), as it allows for thematic development to be led by the participants themselves. I will also be utilising both semantic and latent coding, meaning that I will be analysing participant narratives at a 'surface level' (they say what they mean to say), and interpreting beyond the literal meaning of the words spoken to consider wider significance and connotations (Byrne, 2022). Latent coding is, therefore, aligned more with reflexive thematic analysis (described below), which actively encourages the researchers to bring their own experience to the meaning they are finding in participant themes.

STUDY ONE - REFLEXIVE THEMATIC ANALYSIS

More recently, Braun and Clarke have conceptualised a novel form of TA; reflexive thematic analysis (RTA) (Braun and Clarke, 2021). This approach is considered 'big Q' and is rooted in TA, however, as mentioned above, it also encourages continual development of themes developed from codes, as well as highlighting the positionality of the researcher with explicit reflection on this. It takes the stance that it is impossible to separate the researcher, the person conducting and analysing the data, from the data itself. The researcher is encouraged to consider the skills, experiences, values, and training that they bring, and be authentic about this (Braun and Clarke, 2021a). Since the approach values the individuality that the researcher brings to their work, there is no expectation that a colleague will interpret themes in the same way. It is therefore not seeking the typically 'scientific' goal of developing 'accurate' or 'reliable' coding, but about how the researcher is able to make sense of their own interpretations and embed this within their work.

In contrast, content analysis is a systematic and replicable method used to identify, categorise, and quantify patterns within qualitative data. Unlike the interpretative nature of reflexive thematic analysis, which emphasises researcher subjectivity and the co-construction of meaning, content analysis is considered more ‘small q’ in qualitative research. It follows a structured and often rule-based approach to ensure consistency and reproducibility in data interpretation (Kyngäs, 2020).

Content analysis is widely used in health research due to its’ ability to manage larger volumes of data in an organised and transparent way. It involves condensing raw data into distinct categories or themes, allowing the analysis not only of the presence of specific concepts but also their frequency, distribution, and contextual relevance within the data. Although a standalone approach, content analysis can be used flexibly in more than one form, sitting on a continuum of more quantitative to more qualitative, or by using a mixed methods approach to draw on both elements (Krippendorff., 2013). My analysis served two purposes; I wanted to understand the frequencies of young people’s opinion’s regarding intervention content, structure, and delivery, and I also wanted to focus on survivor’s voices and ensure that their words were reflected in the analysis. For this reason I drew on a mixed methods approach; I opted to create a coding system to break down the data into coding units, as well as using supportive quotations to illustrate each of these categories. I chose to do this using an inductive approach, where categories were determined from the data itself, rather than being pre-conceived (Vears & Gillam, 2022). This approach allowed me to explore both expected and unexpected patterns of survivor experiences, making it a valuable tool in terms of qualitative depth and quantitative rigour. By employing a mixed methods approach, I was able to capture both the nuanced, lived experiences of survivors with the overarching frequencies of their opinions on a future Rb intervention. The combination of frequency analysis and illustrative quotations provided a balanced representation of the data, ensuring that numerical patterns did not overshadow the richness of individual narratives and fitting in with the approach of this thesis. This dual approach also enhanced the transparency and credibility of my findings, as the coding framework provided structure while the direct quotations of participants grounded the analysis in real life and authenticity.

A key strength of content analysis is its’ capacity to bridge qualitative and quantitative methodologies. While it retains a focus on meaning and interpretation, it also allows for measurable comparisons across different data sources. This makes it particularly useful in health-related research like this work, where understanding patterns of communication, recurring themes in patient experiences, or the prevalence of specific ideas can be used practically to inform policy, intervention development, and clinical practice.

Given these advantages, content analysis was well suited to the aims of study three. The structured approach provided a systematic means of categorising key themes whilst also enabling an analysis of their relative prominence within survivors’ narratives. This was particularly important for ensuring that the findings of study three were both transparent and reproducible, aligning with the broader research objectives.

POLITICAL POSITIONING

Encouraged by Braun and Clarke, I have also considered the political standpoints which will inform my research and analysis (Braun *et al.*, 2023). It would be inappropriate for me to consider a phenomenological perspective, as this seeks to describe universal experiences of a particular phenomenon, in my case, Rb (Phillips-Pula, Strunk and Pickler, 2011). This is in direct opposition to the relativist perspective, and the idea that each individual can have a different interpretation of one reality. This is not to say that there will not be commonalities of experience, but I have given equal value to individual perspectives too. My approach fits with the values of feminist position, which poses that research should hold the understanding that knowledge is situated within individual subjectivity, particularly in marginalised groups, such as women (Freeman, 2019). This felt particularly important for my participant population, who by virtue of having had a rare childhood cancer that for many has led to differences in their appearance and vision, could be seen as marginalised. Post structural feminism in particular argues that one’s reality is constructed by language, culture and power (St Pierre & Pillow, 2000). This means that it is important to consider how discourse can contribute to the maintenance of marginalisation and is imperative that this is held in mind when considering my power as a researcher and the conversations I am facilitating. In traditional empirical research, the power imbalance between researcher and participant is often ignored. Feminist research acknowledges that power always influences research, therefore owning this position and considering the part it plays, as well as trying to minimise this imbalance where possible, is crucial. One such attempt to do this within the current thesis is to include a deeply reflexive account of my positionality as a researcher, considering how my identity influences power within the production of knowledge (Day, 2012).

2.2 INFORMATION POWER

When considering how many participants one ‘should’ recruit to their study, a popular concept is to use ‘data saturation’, an idea embedded within qualitative research methods (Constantinou, Georgiou and Perdikogianni, 2017). In line with Braun and Clarke’s outlined assumptions of RTA, this concept does not fit. As discussed above, relativist ontology presumes that each individual holds their own truth, and therefore may bring unique concepts to a piece of research. For this reason, it is argued that meaning is generated from interpretation of the data, and it would be inappropriate to assume that no new participant would bring anything different to the table. For these reasons, the concept of information power was used when considering the qualitative sample size for this thesis (Malterud, Siersma and Guassora, 2016). This idea considers that the aim of the study is an important factor in estimating how large a sample should be in order to gain sufficient information power. For this reason, a flexible target of participants was chosen for this thesis. Information power also argues that researchers should consider the range of experiences and knowledge that participants bring to the study. It is proposed that researchers monitor the demographic characteristics and experiences that participants have as the study continues. It is also important to consider the theoretical background of the study, with studies that have limited theoretical support requiring larger sample sizes and vice versa. Consideration of the quality of the dialogue between researcher and participants is also key, with information power stating that strong and clear communication mediates against requiring a larger sample; this can be quantified by using a topic guide and by being open and clear about the positionality you hold as a researcher. Finally, the type of qualitative analysis that the researcher chooses is important in determining if information power is an appropriate measure of sample size. As the aim of this study is not to uncover every possible experience of individuals growing up beyond Rb (I am of the belief that you could never uncover every eventuality even if you interviewed 5,000 participants), but rather to understand one group of individuals’ unique experiences. As discussed above, this makes RTA an appropriate methodology and information power is well known for being utilised within this (V. Braun and Clarke, 2021b).

2.3 REFLEXIVITY AND RESEARCHER POSITIONALITY

“The participants are trying to make sense of their world; the researcher is trying to make sense of the participants trying to make sense of their world.” (Smith and Osborn, 2004, p.53).

As discussed above, it is important to me that I consider the impact that my positionality has on my research. In order to demonstrate how my positionality aligns with the theoretical underpinnings of my approach, I have undertaken critical consideration of the different aspects of my identity, context, and experiences in order to *"Match the philosophy of the method, philosophy of the research as well as the world of the researcher"* (p.7, Davis, 2020). Additionally, as part of the reflexive process, I kept a reflexive research journal which noted my thought processes, arising feelings, interpretations, and how I responded to my research throughout the analysis process. As it will not be possible for me to include the full depth of my positionality in any individual papers that I publish, a positionality statement which fixes my identity will be incorporated, as well as signposting to the current thesis.

MY HISTORY AND BACKGROUND

The 'Social GRACES' (Burnham, 1993) is a systemic tool developed to encourage consideration of ones' social differences; gender, geography, race, religion, age, ability, appearance, class, culture, ethnicity, education, employment, sexuality, sexual orientation, and spirituality (figure 5).

FIGURE 5: BURNHAM'S SOCIAL GRACES (BURNHAM, 1993)

I have drawn upon this framework to consider the position that I bring to this research project and reflects my intention to bring a nuanced, systemic lens to this project. As discussed, the Social GRACES is an established framework that explicitly encourages reflection on key social dimensions. I believe that this is vital in ensuring sensitive transparency and inclusivity when engaging with my research population of Rb survivors. It is important to note that I have also considered other factors, such as context and life experiences, as well as the idea of intersectionality (Crenshaw, 1989). Intersectionality is the concept that each of these categories does not exist in isolation, acknowledging that it is a complex combination of these which shape a person’s identity and experience, both positively through privilege and negatively in terms of discrimination, bias, and oppression. When conducting qualitative research, the context in which it occurs must be considered; it is time and place-specific, informed by both the participants and the researcher and within their relationship to each other (Dodgson, 2019). It is vitally important to me that I consider the impact of these categories on my interaction with the Rb community, with participants in the study, in my interpretation of data, and analysis of findings. It is also imperative that readers of this research understand who is conducting this research, allowing greater understanding of the impact of their role, their biases, beliefs, and experiences on the study and increasing the credibility of the findings (Berger, 2015). Through engaging with this act of critical self-reflection, I hope to create an inclusive research environment where all stakeholders in this PhD (including participants and PPI group members) feel that their identities and perspectives are recognised and held in mind, including those whose voices may historically have been marginalised or disregarded.

GENDER

I am a female, identifying as the sex that I was born, and I use the pronouns she/her. As a psychologist, I work in a very female-dominated profession. As a health scientist, the gender balance is more equal, although working as a qualitative researcher my work is generally considered more ‘feminine’. It is important to acknowledge the role of male privilege in society and make reference to the fact that their still remains bias against women in Science Technology Engineering and Mathematics (STEM) professions (Dancy *et al.*, 2020). This has led to women feeling intimidated, out of place, and discriminated against.

It is also important to consider how my gender might influence my research with male, non-binary, or trans participants. Previous research has highlighted the lack of attention historically paid to the experiences of men in academic research, resulting in some men feeling marginalised, less-than, and unwelcome (Gatrell, 2006; Lefkowich, 2019). This will inevitably impact on their wellbeing and the quality of data collected. I am mindful that as a young female, it may be more challenging for young men to share their intimate thoughts and experiences with me. I am also aware of the influence of my gender on how I might approach conversations with men or people who identify as non-binary or trans.

GEOGRAPHY

I was born and raised in the United Kingdom and this upbringing comes with inherent Western privilege. I was born and lived in Surrey until I was five years’ old, living in a suburban town with both of my parents. When their relationship ended, I split my time between my Dad’s houseboat in Surrey, then his home in Norfolk, and my Mum’s home in Suffolk. As I spent almost all of my childhood in rural Suffolk, I consider this to be the place where I grew up. Living in a small village shaped my view of the world and meant that I initially found it difficult moving to a city for the first time when I went to university. Considering such transitional stages in development will be important in this thesis.

RACE

I am of Caucasian ethnicity and thus live my life with white privilege. Labelled as an *'invisible package of unearned assets'* (McIntosh, 2019), this unavoidable aspect of my identity impacts how I see others and how they see me. Utilising McIntosh's daily effects of white privilege checklist, I highlight just some of the ways in which this is true:

1. *If I wish I can arrange to be in the company of people of my race most of the time.*
2. *I can turn on the television or open to the front page of the paper and see people of my race widely represented.*
3. *I am never asked to speak for all the people of my racial group.*
4. *I can worry about racism without being seen as self-interested or self-seeking.*
5. *I will feel welcomed and "normal" in the usual walks of public life, institutional and social.*

This privilege shapes not only my lived experiences but also my ability to navigate societal structures with relative ease, compared to those who face systemic barriers related to race. Acknowledging this privilege is essential in fostering empathy, humility, and a deeper understanding of the experiences of others, both in life and in this PhD research. By reflecting on how these advantages influence my views and interactions, I aim to use this awareness as a foundation for equity and inclusion in the work of this thesis.

RELIGION

I do not practice a religion and did not grow up in an environment where religion was a focus, despite being christened when I was born. Not having a religion does not seem to be problematic in my life and society, and it is possible that my life may be ‘easier’ without any potential challenges as a by-product of being part of a particular religious community. Equally, my life may be considered more ‘difficult’ by not having the community of a faith group. Throughout this research, I will hold in mind that many individuals have differing beliefs that hold importance in their life and shape how they view the world and their coping skills.

AGE

I am a young woman who has conducted this PhD in her late twenties, submitting at the age of 30. I am often considered to look younger than I am, and I wonder how that impacts the way that people perceive me. Having grown up with older parents, and having half-siblings much older than me, I feel very used to spending time with and communicating with people older than me. I am aware that through this thesis, I will be spending a lot of time speaking to people who are younger than me, and perhaps a few who are the same age, given that the upper age limit for my recruitment is 29. I am very comfortable communicating with young people and owe that to the extensive experience I have of working in children and young people’s mental health services. I hope that I will be an approachable person for the participants in this study to speak with but acknowledge that my age could be a barrier in some instances, for example if it is wrongly linked to assumed lack of experience.

ABILITY

I am able-bodied and thus most of what I want and need to do is accessible to me. This also includes the ability to see, hear, and access all of my senses to communicate, which may not be the case for some of my participants. Even in areas where I have challenge, for example the need to wear glasses, I can do this in a societally acceptable way and have the resources to access the prescription that I require. I do not and never have experienced discrimination in this area and I am determined to make sure that my research does not discriminate against anyone, regardless of their ability. This includes providing information in a variety of formats and having open communication throughout the PhD to ensure that there are no barriers to participation.

APPEARANCE

I have a straight-sized body that enables me to find clothes in any shop that I would like to purchase from; my size is probably not the first thing that people notice about me. Despite this, I have often been pre-occupied with diet culture and the goal of being ‘smaller’, a privilege that I can afford due to having the means to action this. I see people who look like me represented in mainstream media. I am a long-term sufferer of cystic acne, a condition that dominated my adolescence and remains into my 20s. This is a condition which impacts the way that my face looks, and the way that others perceive it (or the way that I perceive others to see me). It is something that has been commented on in a negative light, and an aspect of myself that I have wanted to hide. I am aware that my participant group may have changes to their appearance due to their Rb and/or the treatment they received for this, despite being an entirely different issue and one that I do not make comparisons with.

CLASS

I was born to a working-class father and middle-class mother and have a typical southern accent, which in my experience, is deemed acceptable by others. Growing up, both of my parents were employed and, although they did not live together, I was able to access most of what I needed, despite most of the time being raised by a single parent on a low income. As with all of these factors, I am aware that the participants who take part in this research may be from a range of backgrounds and associate with different social classes. I endeavour to make everybody feel welcome.

CULTURE

I do not often consider my culture, nor place great interest or importance on contemporary British culture as a whole. I am writing this thesis at a time when Britain is a divided state, with unprecedented historical events happening at an alarming rate; the UK’s departure from the European Union (Brexit), increasing protests around women’s safety after the murder of Sarah Everard (amongst many others), Black Lives Matter marches influenced by the murder of George Floyd (amongst many others), climate change, the death of the Queen, Elizabeth II (and thus questions around the value of the monarchy), the COVID-19 pandemic and, in my opinion, the dire handling of this by the government, against a backdrop of broken systems underfunded and undervalued by over 12 years of Conservative leadership. In terms of my research, and in my dual role as a health psychologist as well as researcher, I see and experience the biggest impact of this within the National Health Service (NHS). At the time of writing, it is increasingly difficult to secure a doctor’s appointment, and there are waiting lists for hospital treatments longer than ever before. It is well documented that waiting for medical care can impact psychological wellbeing, and

I can testify to that in my personal life too. This may also be the case for my research participants, which may therefore impact how they are feeling when they speak to me.

ETHNICITY

Ethnicity differs from race through its' focus on culture, language, heritage, religion, and customs, as opposed to emphasising skin colour and physical characteristics (Bulatao and Anderson, 2004). As discussed in other areas, I do not feel that my ethnicity holds particular importance to my identity, and I am aware of the privileged position I have to be able to make this statement and am mindful of this when I am working with people from different backgrounds. I would like to capture the perspectives of individuals who have had Rb that come from a range of ethnic backgrounds. I am acutely aware, however, that individuals from non-white ethnic groups are often underrepresented in research. In other research, this has been attributed to cultural reluctance to disclose mental health struggles, or due to structural racism barriers and social inequalities that impact on an individual's access to research participation.

EDUCATION

I attended a comprehensive state school in rural England where I completed my education from the age of 11 up to when I had completed my A-Levels at 18. I immediately started an undergraduate degree at a University in the South of England, then a post-graduate therapeutic qualification at another Southern University, a Master's degree in London, before commencing my doctoral qualification in the North. Education is clearly an important element of my identity, and I place great value on academia. I acknowledge my privilege in this and understand that this may differ for individuals whose experience of education is very different to mine. This may be captured in the narratives in this thesis, particularly around the nature of bullying that is experienced by many individuals who have had Rb.

EMPLOYMENT

In one capacity or another, I have been employed since I was 14 years old. This was, in part, possible due to my rural upbringing, and access to local families who allowed me to babysit their children. For financial reasons, I worked throughout completing my degree, employed at a supermarket to supplement my income. Since graduating, I have worked continuously in many NHS roles. This is with exception to a six-week period when I left a job that I was deeply unhappy in. During this time I felt lost and fearful of the impact of unemployment on my job prospects. I wondered what others' might think of me, and in turn I wonder how much my judgements and preconceptions about the importance of employment impacted me during this time. This experience gave me a greater understanding of the complexity of feelings that can arise due to lack of employment, and I am mindful of the different ways in which this could influence an individual’s sense of self and how they view the world.

Sexuality and Sexual Orientation

I am a heterosexual individual in an opposite-sex marriage. I am able to be in an open and socially acceptable relationship which is the most ‘typical’ setup within society. This may not be the situation for some participants, and as in any situation in life, I hope that I create an inclusive environment that accepts individuals and their experiences regardless of their sexuality or sexual orientation.

Spirituality

As someone who does not have a religious practice, spirituality is an area that I do not relate much to. Having said this, I would like to develop more of a spiritual practice through yoga, an exercise which I enjoy and take relaxation from. I hope that the participants in this research feel able to bring their authentic selves, including any spiritual beliefs, into our conversations.

Context and Life Experiences

Clinical Background

Before moving into research, I spent five years working clinically in children’s services. I was always drawn to and most enjoyed working with children and young people with a combination of physical and psychological difficulty. I spent a lot of time working within eating disorders services, as well as in paediatrics, first as an assistant psychologist and later as a therapist. I found this work fascinating because I could see the impact that a physical health condition can have on a young person, and their family’s identity; both positively and negatively. I also found it interesting to consider the role of psychology in how people cope and manage living with physical health conditions. After these experiences, I went on to co-develop a psychoeducation intervention for parents of young people with type one Diabetes. This was my first experience of designing a psychosocial intervention and provided me with a great grounding in the skills that I would later draw upon in this PhD. In the years I have been working on my PhD, I have also gained many more years of clinical experience working psychologically in physical health services. So much so, that at the time of handing in this PhD, I have qualified as a chartered health psychologist and work part-time in the NHS. I believe that being dually qualified as a clinician and academic is an asset to my PhD work and gives me deeper insight into the psychological challenges that many experience when living with and beyond cancer. It also gives me a good grounding in the NHS context in which an intervention might be implemented, as well as realistic expectations about changing clinical care, policy, and practice.

PERSONAL EXPERIENCE WITH CANCER

My interest in this project also draws on the personal experience of my Dad being diagnosed with and then dying from cancer within the first six months of 2020. This inside view of how a medical diagnosis can shape a person's outlook, coping skills, and identity, really furthered my passion for working psychologically within cancer care. I hope that this experience makes me a better researcher, as it has given me a unique insight into the pain that an individual and family goes through when cancer enters their lives. Despite this, I am aware that I cannot generalise my experience to that of the individuals involved in this research project. I am mindful that the diagnosis of a childhood cancer is a uniquely distressing experience, and I am clear that I cannot blur my perspective on their stories with this.

COVID-19

Starting my PhD in 2021, it is impossible not to acknowledge the backdrop of the COVID-19 pandemic. Despite no longer being in government-enforced 'lockdowns' by this point, the pandemic was very much still present in the background, shaping University teaching, meetings, and more. It is also important to consider that should COVID-19 have not happened; I may have been unlikely to have been granted this PhD scholarship. Prior to the pandemic (and the developing norm of remote working), the department were looking to fund a scholar who was based in York. As this was not viable for me at the time, it is likely that I would have been ruled out of applying on geography alone. I am so grateful that one positive came out of this experience, through the University adjusting their policies and giving consideration to me enrolling on this PhD research from my home. I am privileged in that I was able to do this, with access to high speed WIFI, a supportive family, and a safe home to live in with the space for a desk and workspace. I acknowledge that this might not be the case for some of the participants in this research, and that many individuals may carry trauma from their experience of the pandemic.

In addition to the Social GRACES, I have also considered the LUUUTT Model. Conceptualised by Pearce and Pearce (1998), the LUUUTT model complements the Social GRACES by encouraging individuals to consider the types of stories we have lived, the stories that are untold, the stories that are unheard, and the stories that are made sense of through collective sense making (figure 5). It is a model that encourages open and honest sharing of narratives that may have been marginalised, and allows for new, shared meaning to be developed. By integrating both models, I aim to demonstrate the power of co-creating new meaning through the exploration of intersecting social identities and shared narratives. This allows diverse perspectives to be both acknowledged but also to actively inform the outcomes of this research. For example, through my choice of methodology throughout this thesis, I hope that I am co-creating a space for previously unheard and untold stories to be heard and told, leading to the opportunity to create new support tools for the Rb community. This is an approach that has been used within cancer care, and that I am interested in applying to survivorship (Goering and Krause, 2017). In summary, by considering both the Social GRACES and the LUUUTT model to inform this research, I sought to emphasis:

- **Collective Sense-Making**: Through self-reflection and facilitating discussions that allow participants to collaboratively make sense of their experiences, fostering a shared yet nuanced understanding of survivorship.
- **Hearing Untold Stories**: Creating a space where individuals from the Rb community could share their lived experiences, including those that may have been previously unheard or marginalised. This also includes sharing these experiences for others, through the dissemination of this work.
- **Developing Support Tools**: Using insights from these shared narratives to inform the design of new support tool tailored to the unique needs of the Rb community.

THE LUUUTT MODEL

Stories:

1. **L**ived – actions of our lives that cannot be changed e.g. having had Rb
2. **U**ntold
3. **U**nknown Stories that may not be conscious e.g. memories, bullying, inner critical voice
4. **U**nheard
5. **T**old – the meaning that we make of these stories, which inform future actions
6. **T**elling – the way in which people tell their stories

FIGURE 6: THE LUUUTT MODEL AS APPLIED TO RETINOBLASTOMA

CHAPTER 3: STUDY 1 – *“IT’S NOT MEANT TO BE FOR LIFE, BUT IT CARRIES ON”:* A QUALITATIVE INVESTIGATION INTO THE PSYCHOSOCIAL NEEDS OF YOUNG RETINOBLASTOMA SURVIVORS

3.1 CONTEXT

Study one comprises a qualitative study utilising reflexive thematic analysis to understand the experiences of teenage and young adult survivors of Rb. This method was chosen due to its’ flexibility to accommodate different methods of data collection across a varied population, including a large age range (13-29 years) and different Rb diagnoses (heritable and non-heritable, unilateral and bilateral). I made the decision to conduct this study prior to a systematic review, due to the known lack of research in this area. To address this, the current study aimed to explore, understand and describe the psychosocial needs of young people living beyond Rb, to combine with the other studies in this thesis to inform a future psychoeducation intervention.

3.2 INTRODUCTION

As described in chapter one, Rb is a rare childhood cancer that is diagnosed in approximately 40-50 UK children a year, 45% due to heritable factors (Jenkinson, 2015; Hülsenbeck *et al.*, 2021). Although highly curable, Rb can have a huge impact on the psychological wellbeing of individuals long after their treatment has ended (van Dijk *et al.*, 2009). For many their vision is impaired, and they may experience eye loss or facial changes (Sethi *et al.*, 2014; Temming *et al.*, 2016). For others, there will be lifelong anxiety about the development of second cancers and the possibility of future children developing Rb too (Hill *et al.*, 2018). Existing research has highlighted the need for psychosocial support in this population, yet we know that the nature of this support is under-researched and largely unavailable (van Dijk *et al.*, 2009; Ford *et al.*, 2015; Belson *et al.*, 2020; Gregersen *et al.*, 2021).

This study is the first of three projects that aims to understand the psychosocial needs of young people living beyond Rb. It will provide rich information about an individual's experiences that will inform future psychoeducation interventions. This will be of significant benefit as there is currently no known routine psychological support offered to young people as they transition from childhood and begin to navigate the vast impact of life beyond Rb, in the UK or Worldwide.

3.3 BACKGROUND AND RATIONALE

Due to the nature of Rb that is discussed earlier in this thesis, it is important to understand the psychological impact of living beyond both heritable and non-heritable Rb, with existing research highlighting the need for better psychosocial support (Gregersen et al., 2021). Previous research has indicated that anxieties in this population relate to personal health, survival and impact on family (Belson et al., 2020; Feng et al., 2020). However, little is known about the specific psychological challenges experienced by teenagers and young adults, who must manage the effects of Rb in a period of life when psychosocial support is crucial to the healthy development of individual, social, and sexual identity (Zebrack, 2011). This lack of knowledge is in part because of the small number of diagnoses made each year, meaning that there is a limited insight into the particular risks and challenges of teenage and young adult life after Rb. Furthermore, the change in treatments for Rb over the decades, with more attempts to save the eye in modern interventions, psychological challenges may have changed (Gündüz *et al.*, 2020). Wider literature exploring the psychosocial needs of young people living beyond cancer indicates that support must be given to support the creation of a new normal, even when future outcomes are uncertain (Gibson et al., 2016). Considering illness and long-term effects of treatment at this crucial developmental stage can cause significant hindrance to identity and mental wellness (Bradley Eilertsen *et al.*, 2012; McArthur, Strother and Schulte, 2017).

Whilst these are all important to consider, appropriate intervention for the unique psychosocial challenges associated with life after Rb requires further investigation. Determining the most acceptable, accessible, and effective support is important to patients and families, clinicians, policy makers and commissioners of services. Intervention development requires a thorough understanding of the challenges faced by people living beyond Rb, and a detailed examination of possible theoretical and evidence-based approaches to address the challenges and support them. There is widespread evidence to suggest that psychosocial support is warranted, and wanted, in the Rb population. Recent qualitative studies have found that both young people and adults would like access to psychologists who have specialist knowledge of Rb-related challenges, preferably offering psychoeducation at early stages to prevent potential Rb-related psychological difficulties (Belson *et al.*, 2020; Gregersen *et al.*, 2021). As the current study will also be designed for young people who may have visual impairments, it is imperative that practical implementation of psychoeducation is also considered (Yildiz and Duy, 2013; Elsman *et al.*, 2019).

3.4 STUDY AIMS

This research involves a two-phase qualitative study: (1) focus groups with teenagers (age 13-19 years) and (2) individual interviews with young adults (age 20-29 years), all with a history of Rb. The study aims to explore the experiences of living beyond Rb and views on challenges that they may face as a result. The specific objectives are as follows:

a) To understand the views of young people who have had Rb regarding their psychosocial needs

b) To explore Rb-associated psychosocial challenges that arise during teenage and young adulthood

c) To seek the opinions of individuals of who have Rb regarding what psychosocial/educational support would be/have been beneficial to them

3.5 METHODOLOGY

A QUALITATIVE APPROACH

As discussed in chapter two, Reflective Thematic Analysis (RTA) was my chosen qualitative methodology for this study. RTA is a flexible approach which allowed me the opportunity to combine different methods of data collection through conducting both interviews and focus groups. At the outset of this research, I did not intend to analyse the data as one, but it made sense to do so due to the heterogeneity of themes across adolescent and young adult participants. As an iterative processs, this approach allowed me to continually revisit and refine the themes over a long period, holding discussions with the wider research team to delve into the nuances of participant meaning. One of the significant strengths of this methodology is the emphasis on researcher reflexivity, which encourages continual, in-depth engagement with the data whilst simultaneously acknowledging my role in interpretating the narratives that I am collecting. Unlike other qualitative methods, this emphasis on reporting reflexive processes helps to mitigate against bias by acknowledging that it is inevitable, encouraging researchers to name how they are relating to the data and make this transparent. Ultimately, I hope that this enhances the credibility of the findings of this study and highlights the strength of using robust qualitative methods.

Despite its' popularity, the founders of the approach feel that it is often mis-used, and thus produced a 'best practice recommendation' to support the development of effective reflexive thematic analysis within health research (Braun *et al.*, 2023) (see table 3). I have found it incredibly useful to consider these recommendations and how I have tried to implement them into my research. I have included these reflections below, which also aligns well to my research method which values reflexivity.

TABLE 3: TWENTY BEST PRACTICE RECOMMENDATIONS FOR EFFECTIVELY CONDUCTING AND REPORTING THEMATIC ANALYSIS IN HEALTH RESEARCH (BRAUN ET AL., 2023)

Area	**Recommendation**	**Evidence within thesis – study one**
Selecting the most appropriate type of TA	1. Determine the goal/purpose of research. If this is quite open, reflexive TA is appropriate. If this is more delimited than open, then codebook or coding reliability approaches are more appropriate.	The overarching purpose of my PhD is to generate the evidence to inform a novel psychosocial intervention. My first study's aims are much more open, in that I want to find out the experiences of young people who have had Rb. There is limited existing knowledge on this, and therefore I deem RTA to be an appropriate method.
	2. Reflect on your paradigm/research values. If (post)positivity (e.g. concerns about coding accuracy/reliability, minimising bias, etc), use coding reliability TA. If not positivist, use codebook or reflexive TA.	I am aligned with an interpretivist paradigm; I value subjectivity and context; I do not believe that all realities are the same. I believe that each one of us, and of the research participants, will bring something unique to my study. All of my participants will have had the same form of cancer, but I do not believe that they will all have experienced the same things. Even if there are similarities, I do not believe that they will interpret these the same, nor do I necessarily believe that I will have the same interpretations. Therefore I will be using RTA.
	3. Reflect on theme conceptualisation. If the focus is on shared topics (topic summaries), select codebook or coding reliability TA.	My themes will not necessarily be aiming to find shared experiences, given that I believe that we all hold own our truths; there is no one truth. This therefore cements the idea that I should not use codebook or coding reliability TA.

	4. If considering using multiple analytic methods (e.g. TA and grounded theory), reflect on why, and whether it really is necessary. Read more around TA.	I will be using RTA only.
Methodology	5. Make clear what general type of TA you have used. Avoid citing divergent or incompatible approaches without clear explication of what is taken from each and why (but hold in mind the importance of methodological coherence and integrity).	I have found it useful to situate myself with the chosen methodology and paradigm. I hope that I have explained this clearly in chapter 2.
	6. Ensure any rational for your use of TA avoids any descriptors but connects to your research topic, theory and/or context.	It is important to continually link the methodology that I use to my research topic and position as a researcher.
	7. Make sure you specify the ontological and epistemological assumptions guiding your use of TA (and then enact these consistently).	As above, found in chapter 2.
	8. Discuss the explanatory/political theories and concepts informing the analysis (e.g. phenomenology, social cognition, feminism); avoid treating concepts as theoretically neutral (e.g. body image).	Chapter 2 contains extensive consideration of phenomenology, due to its' usefulness for understanding how Rb survivors perceive and make sense of their experiences. I have also discussed how societal norms and power structures impact health outcomes and access to care, referring to this throughout my discussion. Concepts have not been thought of neutrally, as to understand how survivors perceive their physical self's post-treatment, it was important to draw on numerous concepts.

9. Make clear your particular orientation to TA (e.g. semantic/latent coding, inductive/deductive analysis); ensure ideas like latent and deductive are conceptualised in a way that is consistent with the TA approach used.	I have explored this in chapters 2 and 3.
10. Clearly discuss what you actually did for your analytic process, rather than generically describing the process, such as listing six phases of reflexive TA.	This is described in detail in my methods chapter, in ‘methodology’ and the ‘summary of themes’ section of study one, and study one’s discussion.
11. Avoid confusing and conflating positivist notions of bias with researcher reflexivity. For reflexive and other Big Q TA, include some discussion of both the reflexive processes engaged in, and the professional/personal positioning of the researcher or the broader contexts shaping their experiences and perspectives. If small q TA, discuss the management of researcher bias/influence.	As I am using RTA and thus ‘big Q’ research, I have included extensive discussion around my position as a researcher.
12. Use language and a writing style consistent with your TA approach. For example, for reflexive TA, take care not to suggest that themes emerge, or were identified. Avoid language of bias and aim to write in the first person.	I hope to have done this; it has been challenging to reassure myself that I can write in the first person, when for years of early study you are continually told to write ‘scientifically’ and thus in the third person.

13. Use a reporting format and headings appropriate to your TA approach. For example, a combined results/discussion is often the best way to report analysis in reflexive TA.	I have reported my results in a way that fits best with my RTA and the data from the study.
14. Ensure your quality practices are theoretically consistent both with your approach to TA, and with your ontological and epistemological assumptions. Realism>respondent validation; triangulation. Positivism> multiple independent coders; interrater reliability; consensus coding/theme development. Big Q/nonpositivism>reflexive journaling; member reflections.	It has been crucial to me to understand the different ontological and epistemological perspectives and consider where I fit. As I am using RTA, I have kept a reflexive research journal and included member reflections.
15. Consider providing a clear overview of themes and thematic structure – such as a table or figure.	A diagrammatic overview of themes and subthemes can be found in the 'analysis' section of the study. This is accompanied by a written summary of themes and subthemes.
16. Make it clear how many themes (including any overarching themes and subthemes) will be reported.	As point 15.
17. Make sure themes are named appropriately. For example, names of shared meaning themes should ideally capture the key concept of the theme; in reflexive TA, avoid single word theme names.	The names of themes were chosen as they use participants' own words. Subtheme names were chosen to highlight the key features of the content. No single word theme names were used.

	18. Ensure what is reported within each theme aligns with the methods used. For example, in reflexive TA, themes need to be rich and complex, and capture more than one analytic insight/observation.	The themes and subthemes are rich and in-depth, capturing complex narratives.
	19. Ensure any fragmentation of thematic structure is appropriate to type of TA. If using reflexive TA, be wary of an overly fragmented thematic structure, as analytic quality requires depth in reporting, which fragmentation can preclude. Use subthemes only when desirable to highlight a particular facet of the central theme concept. Consider using supplementary materials or develop separate papers to report in depth.	I deliberately chose not to have many subthemes of each theme, instead capturing the rich and often overarching narratives in a comprehensive thematic structure.
	20. Make sure you appropriately use data to evidence themes and analytic observations and insights. Provide an analytic narrative that provides interpretation – in reflexive TA, for example, you ideally offer a rich, complex analytic narrative woven around the data extracts.	All insights are supported by quotes that are reported with participant number (P) and by self-reported Rb type (bilateral as B, unilateral as U, heritable as H, non-heritable as NH. E.g., P1, NH, U). In the results and discussion sections, I offered a rich interpretation of the data around each quote inside each theme and subtheme.

RECRUITMENT SOURCE

This qualitative study was set within the UK’s two specialist treatment centres: The Royal London Hospital shared with GOSH and Birmingham Children’s Hospital. Furthermore, one of the UK’s only charities dedicated to supporting individuals impacted by Rb (CHECT), acted as a third recruitment source. Please see appendices A for details of contracts set up with the above sites for the purpose of this research.

SAMPLE AND SAMPLING

Initially, a flexible target of participants was aimed to be recruited due to the small recruitment pool that participants were sought from, given the rare nature of Rb. This was also based upon the concept that there is not a set number of participants required to generate rich and meaningful data on the topic (Braun and Clarke, 2021). The concept of information power was used to guide sample size, considering the idea that the level of relevant information that a sample holds will determine the number of participants needed (Malterud, Siersma and Guassora, 2016). To ensure the inclusion of individuals with different cancer experiences and levels of visual impairment, and other demographic and family characteristics that may influence transitions, I proposed 2-4 focus groups for the younger participants. Groups were purposively sampled where possible to include individuals with different characteristics, such as type of Rb, visual impairment etc. Younger individuals were also planned to be separated by age (13-15 years and 16-19 years) due to life stage. This decision was informed by developmental psychology principles (Rutter & Sroufe, 2000), acknowledging that adolescents at different ages and stages experience distinct cognitive, emotional, and social challenges. The separation ensured that discussions were relevant to participants' lived experiences and life stage and that younger participants did not feel overshadowed by older peers. It was anticipated that individuals with heritable Rb would experience different challenges to those with non-heritable Rb. Therefore, these characteristics were continually monitored for representation. Focus group size was informed by established qualitative research guidance (Rabiee, 2004) and were structured around a theoretically informed topic guide, also devised through discussions with the PPI experts in the project proposal and existing research. Additionally, focus groups provided a setting where participants could engage in shared storytelling, potentially fostering a sense of belonging and validation amongst their peers.

For older participants aged 20-29 years, in-depth, semi-structured interviews were conducted after being recruited via the above procedure. Participants responded to either a social media advert or an NHS letter identifying that they were eligible to take part. Letters of invitation and patient information leaflets were sent by NHS sites to individuals to gauge interest in taking part. Nominated clinical staff within each department also provided information to eligible participants in clinic. These contained details of the research team for potential participants to make contact with, if they wish to take part. Initially, convenience volunteer sampling was utilised, drawing on the interest of individuals within the networks of the above settings (Andrade, 2021). Later, once initial data had been collected, purposive sampling was drawn upon to target participants whose characteristics had not yet been captured (Campbell *et al.*, 2020). This was done with the aim of deepening understanding of participant experiences, yielding richer insight into a range of Rb narratives. The decision to conduct interviews, rather than focus groups, with the older age group was informed by the likelihood that these participants could potentially have more complex thoughts and feelings shaped by their longer post-treatment trajectory. One-to-one interviews provided the flexibility for participants to share their experience in depth, including reflections on long-term impacts of treatment and their transition into adulthood. Additionally, this approach acknowledged the potential for greater variation in perspectives within this group, as older participants may have developed different coping strategies, identity shifts, and support needs over time. It was also anticipated that young adults may wish to discuss more sensitive topics, and therefore may benefit from the privacy of a 1-1 interview. The sampling strategy aimed to recruit roughly equal numbers of individuals with heritable and non-heritable Rb, unilateral and bilateral Rb, differing demographic characteristics and treated with a range of treatment options across the two NHS sites. By ensuring representation across these variables, the study sought to capture both commonalities and divergences in experience, offering a comprehensive understanding of how Rb impacts individuals over time and across life stages. Data quality and participant diversity was monitored during data collection to make a final decision regarding sample size, in line with the concept of information power.

3.6 INCLUSION AND EXCLUSION CRITERIA

For both participant interviews and focus groups, wide inclusion criteria were set. This was to encourage individuals with diverse experiences to take part, this included any form of Rb (heritable or non-heritable), bilateral or unilateral, any age of diagnosis, and any treatment regime. Specific criteria are summarised below:

INCLUSION CRITERIA:

- Prior diagnosis of Rb (heritable or non-heritable)
- Survivors who are off treatment
- Aged between 13-19 (phase one) or 20-29 (phase two) at the time of the study
- Willing and have capacity to provide consent and attend focus group (phase one) or individual interview (phase two) either online or on the phone
- Fluent in English

EXCLUSION CRITERIA:

- Those who lack capacity to participate in the study, guided by parents and clinical staff for those under 16 years old and the Mental Capacity Act (2005) for individuals over 16 years old

3.7 PARTICIPANT PROFILE

As discussed above, participants were teenagers aged between 13-19 years, and young adults aged 20-29 years, who had been diagnosed with any form of Rb at any stage of their childhood. All participants had been treated in the UK at one of the two national treatment centres in London or Birmingham. Social media recruitment to this study commenced via CHECT on 9th June 2022 and ceased on 8th December 2022. NHS site recruitment commenced on 12th September 2022 via Barts Health NHS

Trust, Great Ormond Street Hospital for Children NHS Foundation Trust, and Birmingham Women's and Children's NHS Foundation Trust. NHS recruitment ceased on 15th January 2023.

3.8 PATIENT AND PUBLIC INVOLVEMENT (PPI)

PPI involvement was informed by the GRIPP2 guidance on effectively involving experts by experience in health research (Staniszewska et al., 2017), as well as National Institute for Health Research (NIHR) guidelines (NIHR, 2021).The continuous involvement of CHECT members maintained the personal experience perspective throughout the project. Two adult survivors of Rb also inputted into the project proposal and study documents and expressed interest in continued involvement in the project through a regular PPI panel meeting. Overall, a total of five Rb survivors formed a PPI group for the purpose of this study. These individuals were primarily involved in reviewing the study findings and providing input on the interpretation and analysis of these (see appendix A21 for the results summary sent to the group, as well as A22 for the PowerPoint slides which formed the PPI results discussion meeting held on 25th July 2023). Additionally, I engaged with the Young People's Advisory Group (YPAG) in Leeds, which is part of the Generation R Alliance. This group includes young people with a range of health conditions including lived experience of cancer. The YPAG were consulted for feedback on all elements of this project. This was done by sending participants (and the PPI panel) a broad summary of findings to check their views on these.

All young people were provided with detailed information sheets and consent forms, with adapted versions available for individuals with visual impairment. These were informed by PPI experts and the Royal National Institute of Blind People (RNIB) guidance for creating accessible content for people with impaired vision (RNIB, 2014). For greater insight into the PPI involvement in this thesis as a whole, see chapter four. For insight into training completed to aid this study, please see appendices A.

This research is funded by the CHECT who were continually involved in the development, recruitment, and dissemination of this study. This included advising on recruitment and advertising for participants through the CHECT member network and youth panel. CHECT had no say in the findings from the study, so as to not cause conflict of interest.

3.9 ETHICAL CONSIDERATIONS

ETHICAL APPROVAL

Health Research Authority (HRA) approval was granted on 27th May 2022 (Research Ethics Committee reference: 22/NI/0082). See appendix A2 for HRA approval letter.

DATA MANAGEMENT AND STORAGE

In line with the 2018 General Data Protection Act and the Research Governance Framework for Health and Social Care Research, data (anonymised focus group and interview transcripts) were securely archived by the University of York for a minimum of 10 years. Personal data of participants has been stored for up to 3 years after the study has ended for the purpose of disseminating study findings.

All information collected during the study has been kept strictly confidential. Information has been held securely in electronic formats at the University of York. The University of York complies with all aspects of the 2018 General Data Protection Act and operationally this includes obtaining consent from participants to record personal details including name, postal and email address, and contact telephone numbers: and appropriate storage, restricted access and disposal arrangements for patient and carer personal details. Participants were assigned a study identifier so that their data remains anonymous. Personal data and pseudonymised data were stored separately in a restricted access folder on a secure university server and access is password protected.

PARTICIPANT CONFIDENTIALITY

Participants were informed of their right to confidentiality and what this meant if they disclosed information that suggested that they or others were at serious risk of harm. Participants were informed that they had the right to withdraw from the study at any time and that their data could be excluded from the study if not already analysed. All personal data were stored in a password-protected file, using a participant identifier (which was assigned at the point of consent) to link participants' details to their data (i.e. focus group/interview transcript). This information and all data were stored on the University of York servers and were not accessed by anyone outside of the research team. Codes and the corresponding names were kept in a separate document stored on a secure server and accessed by a password protected computer that only the research team had access to. Demographic characteristics of participants and quotations from participants may be used in research reports and other publications and presentations; however, care was taken to protect the anonymity of participants, including the use of pseudonyms, so that others are not able to identify them in any way. Complete transcripts were not shared with journals for publication. Participants were informed of this prior to consent and at the time of the focus group/interview.

Due to the uncertainty around the COVID-19 pandemic at the time of data collection and the geographical spread of potential participants/research team members, attention was given to the potential for utilising remote/digital methods for qualitative interviews and intervention development. It is arguable that remote delivery had the potential to make it harder for researchers to make a connection to those involved in the study (Tates *et al.*, 2009; Roberts, Pavlakis and Richards, 2021; Saarijärvi and E. L. Bratt, 2021). When conversing remotely, it may be that non-verbal cues are lessened or lost, and this could have impacted how comfortable people felt to discuss sensitive topics. However it may be that when discussing personal topics, participants could have felt more relaxed and able to share their experiences when they were not face-to-face, contributing to more rich and honest conversations (Powell *et al.*, 2017). This is further supported by the fact that, as a population, many of us are now used to communication via online platforms. It is also possible that risk issues could come have arisen during interviews, which would have been harder to manage remotely. This was mitigated against through conducting interviews/sensitive conversations under strict data governance and ethical guidance, signposting individuals to appropriate resources, such as CHECT's support line, Young Minds etc., if needed.

RISKS AND BURDENS TO PARTICIPANTS

Individuals were informed that the decision to participate was voluntary and would not affect any services that they receive. There was a risk that asking questions about cancer experiences and psychological wellbeing may identify psychological distress, which may not have been being managed by appropriate healthcare professionals. This raised ethical concerns of causing participant distress. For these reasons, an outline of potential areas that the focus group/interviews would cover was provided in the participant information sheet. This aimed to help make sure that participants were prepared for the focus group/interview ahead of time. If a participant became distressed during the focus group/interview, I asked the participant if they would like to take a break or stop; this was guided by the participant. In cases where distress was significant or sustained, I would suggest that participation is stopped, although the decision would be made with the participant. I would then offer the participant the opportunity to finish the interview on another date. Where needed, assistance was given to help the participant identify appropriate sources of support, including signposting to their Rb care team who could refer them to relevant mental health services if required. CHECT also agreed to act as a source of post-study support for any participant who would like this. Details of CHECT's email and phone number were provided to all participants via the participant information sheet given prior to the study, as well as in a debrief email at the end of the study. In this email, other sources of support were also listed. After taking part the participants were given a follow-up call 48 hours afterwards, to check that they did not have any additional questions or concerns. Should participants have disclosed any concerns of risk at any stage during the study, the research team would always try to discuss a plan of action with them first. If the participant was under 16 years of age their parents would be contacted to share the risk issue. If the participant was recruited via a hospital site their clinical team would be notified of the risk issue. If the participant was recruited via CHECT, they would be encouraged to contact CHECT's support team and their GP/Rb long-term follow up clinic if applicable. See appendix A14 for the distress protocol.

Risks to research team

To ensure researcher safety, I would have adhered to the University of York lone worker policy (Department of Health Sciences, 2016) should interviews have been conducted in person (in this study, the participants opted for remote interviews only). The research team had a buddy system where a colleague knew the whereabouts of the researcher conducting the interview at all times. The team also followed the procedure on risk management that is consistent with those used by the Trusts and Charity that we were working with. All research staff were Good Clinical Practice (GCP) trained (see appendix A27). Researchers debriefed with another member of the research team following focus groups/interviews to ensure that any psychological impact of conducting this study was minimised. Regular supervision and the use of a reflective diary throughout the research process also ensured researcher wellbeing was maintained.

3.10 Recruitment and Data Collection

After potential participants had expressed interest via email, they were sent a digital information sheet (in both written and video formats) and consent form (permitting audio and video recording and use of direct quotes) (appendices A3-A10). These were returned to the research team via email and acknowledged by providing a signed copy of the consent form for their records, and allocation of a participant identification (ID) number. At all stages of the recruitment process participants were given the opportunity to ask questions, as well as provided with reminders about confidentiality, anonymity, and the right to withdraw. It was agreed that any anonymised data would be included in analysis up until the point of withdrawal.

CONSENT PROCESS

Only individuals with the capacity to consent took part in this study. It was ensured that capacity was checked by the research team (who were trained in assessing capacity) at the outset of the research and were also guided by the Mental Capacity Act 2005 for those over 16 years. All participants under 16 deemed to have capacity were encouraged to discuss taking part with their parents. Participants recruited through research sites were only approached if clinicians deem them to have capacity. Consideration around accessibility of the study (including ability to consent) due to potential visual impairment of participants were paramount in order to avoid unnecessary barriers to participation. Ultimately, Gillick competence (assessment of capacity to give informed concept after listening to and understanding information) was used to make the overall decision for consent amongst young teenage participants (Wheeler, 2006; Parekh, 2007).

If individuals were interested in taking part, they made contact via email, using information provided by NHS teams or via the CHECT advert. I discussed the study with participants provided them with study information in video and written format, detailing the study's aims, objectives, and participation requirements. If the participant was happy to continue, an invitation to either a focus group or interview (depending on participant age and preference) was arranged. Both the researcher and the participant signed and dated the consent form to confirm that consent was obtained prior to the start of the interview or focus group. The participant received a copy of this document and a copy was filed in the Investigator Site File. Consent was monitored throughout the study. For example, I looked for disengagement or withdrawal and checked that the participant was happy to continue at every stage of communication. Participants were reminded that they could choose not to answer questions that were too distressing or that they would prefer not to answer. Participants were also informed of their right to withdraw at any time and without giving a reason. This was covered in the participant information sheet and was reiterated prior to starting the focus group/interview.

If a participant had chosen to withdraw from the study, they would have been informed that they would have been able to withdraw their information as long as it had not already been used (i.e. in the analysis). No further data would then be collected from participants who withdrew and all data able to be withdrawn (from their contribution) would be destroyed, unless consent was provided to include data collected up to the point of withdrawal in the study. Fortunately, this was not an issue in this study.

3.11 PROCEDURE

Despite separating data collection by age, these are not sequential and were facilitated at the same time. This allowed for an integrated approach where topics discussed in both phases could inform topic guides of each other (appendices A17 and A18). Please see figure 7 for a diagrammatic overview of the participant journey throughout the research process.

Different methods of data collection were employed for teenagers and young adults to address their unique needs and preferences. Teenagers participated in focus groups, while young adults were engaged through individual interviews. This methodological choice was informed by existing literature and consultations with clinicians, which highlighted that, for many teenagers, this research might represent their first opportunity to discuss their experiences of having had cancer in depth. Recognising the potential emotional challenges associated with such discussions, focus groups were designed not only as a data collection tool but also as a supportive environment. By fostering peer interaction and providing an opportunity to connect with others who had faced similar experiences, the focus groups aimed to reduce barriers to participation and create a sense of solidarity. This supportive setting was anticipated to empower teenage participants to share their thoughts and feelings more openly, enriching the quality of the data collected. In contrast, individual interviews were chosen for young adult participants to provide a private and flexible space for reflection, allowing for a deeper exploration of their personal experiences. Young adults are at a different stage of life in comparison to teenagers, with increased independence, diverse responsibilities, and potentially more complex emotional and psychological responses to their Rb experience. Individual interviews offered the opportunity to tailor discussions to each participant's unique context and to delve into sensitive topics that might be less comfortably addressed in a group setting. This was because it was anticipated that young adults may have been more likely to discuss sensitive topics, such as fertility concerns. This method was also intended to respect the young adults' likely preference for privacy and autonomy, thereby creating an environment where they felt safe to share openly and candidly.

CONDUCTING THE FOCUS GROUPS

Focus groups utilising semi-structured questions were conducted in person or via video-call. These were led by me and co-facilitated by a second member of the research team (BP) to ensure consistency and to reduce bias across groups. These sought teenager's views on Rb-related challenges they had faced in the past, faced at present, or may face in the future, and current psychosocial support they access or are aware of. Alongside conversational content, group dynamics were noted through a field diary.

Focus group participants were offered the opportunity to take part in-person or online. This was decided for two reasons; teenagers are likely to still be under the long-term follow-up care of their hospital site. This means that there is likely to be greater opportunity to convene a group of teenagers in one location, making the feasibility of an in-person meeting better. Additionally, it was felt that teenagers may need more support when discussing potentially distressing topics. It may therefore have been easier to provide support to them in person. Having said this, research suggests that virtual focus groups can offer advantages for participant diversity, particularly when participants may experience barriers to access (Rupert *et al.*, 2017). For this reason, virtual alternatives were also arranged and were subsequently opted for in all instances.

CONDUCTING THE INTERVIEWS

For young adult participants, in-depth, semi-structured interviews were conducted by telephone or video call, lasting approximately one-hour. Recordings were anonymously transcribed verbatim by an external transcription company then checked by the interviewer. Interviews utilised a narrative approach to explore the life-stories of individuals and aimed to gain clear understanding of individuals' transition to adulthood in the context of prior Rb experiences. Interviews also sought views on what has helped individuals to address challenges they have experienced, and what they would provide to their teenage selves to better prepare them.

All interview participants were offered the choice of taking part online via a video calling platform like Zoom, or via telephone. Since the COVID-19 pandemic, remote interviewing has become the norm in qualitative research (Pratama *et al.*, 2020; Sah, Singh and Sah, 2020; Foley, 2021). As such, I sought guidance from peer-reviewed publications on the best practice for doing this (Saarijärvi and E.-L. Bratt, 2021). All interviews were conducted in my private home office. Participants were often at home in a private space, but occasionally were in a quiet office at their place of work. The option for in-person interviews was considered, however due to the expansive geographical spread of participants, this option was not taken forward.

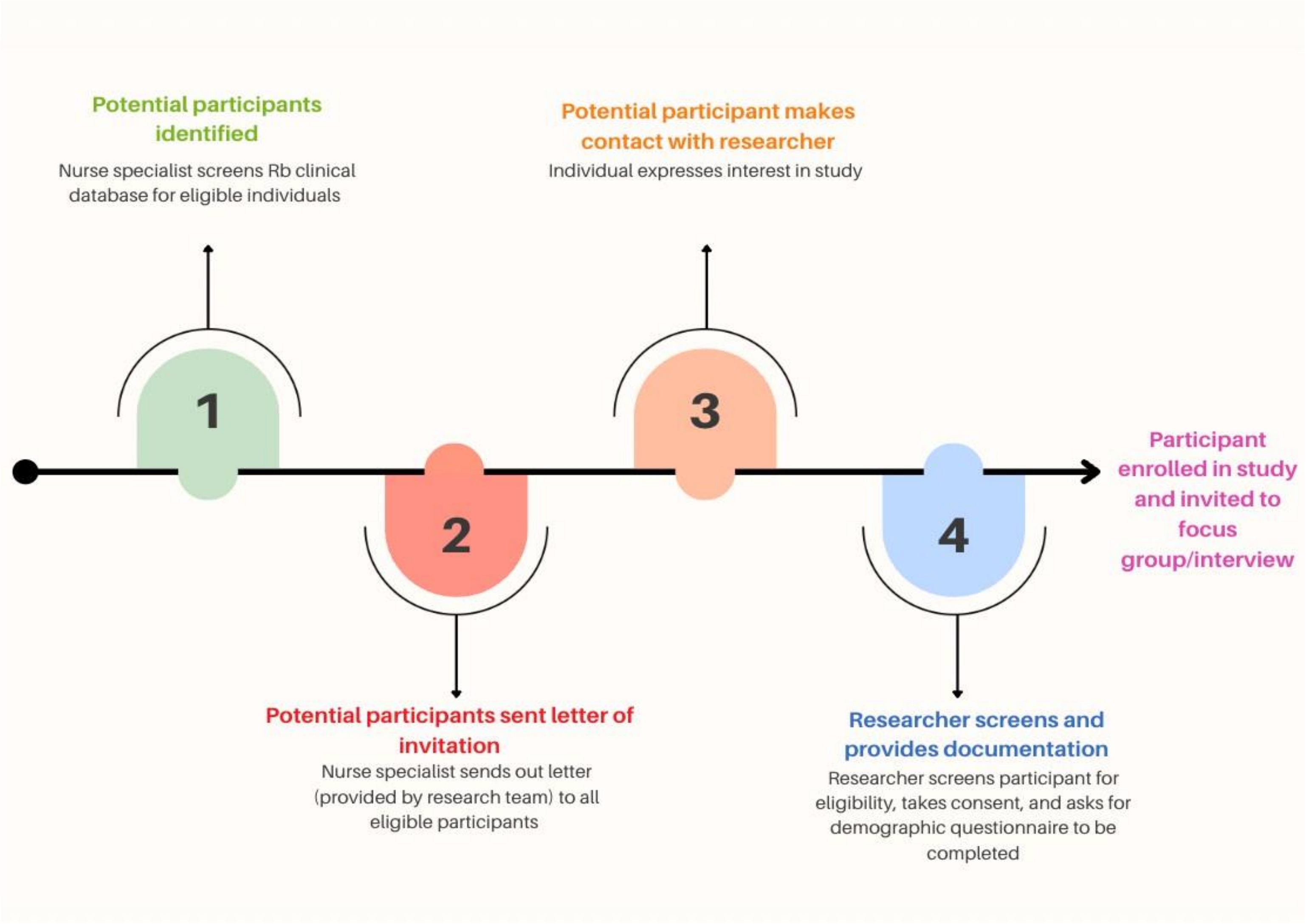


FIGURE 7: PARTICIPANT JOURNEY THROUGH THE RESEARCH PROCESS

TEENAGERS

For teenage participants, focus groups were conducted online through video call, lasting approximately two hours. These used topic guides (appendices A17 and A18) that sought teenager's views on challenges that they had faced in the past, faced in the present, or may face in the future, and current psychosocial support they access or are aware of.

Each group was facilitated by me and co-facilitated by another second member of the research team (Bob Phillips; BP) to ensure consistency and to reduce bias across groups. Alongside conversational content, group dynamics were noted through a field diary. Where necessary, prompts or probes were used to encourage participants to respond to issues elicited by others. The groups were audio and visually recorded to allow in-depth analysis of the discussions.

YOUNG ADULTS

For young adult participants, in-depth, semi-structured interviews were conducted by telephone, or video call, lasting approximately one-hour. These were audio and, where conducted online, video recorded. Recordings were anonymously transcribed verbatim by an external transcription company then checked by me. Interviews utilised a narrative approach to explore the life-stories of individuals and aimed to gain clear understanding of individuals' transition to adulthood in the context of prior Rb experiences. As above, topic guides (appendices A17 and A18) were used to structure interviews and were informed by PPI discussion and various psychological models that have previously been utilised within oncology and psychoeducation literature. The use of key themes ensured that challenges identified by teenagers are explored consistently within young adult interviews. Interviews also sought views on what has helped individuals to address challenges they had experienced, and what they would have liked to have had access to in terms of psychosocial support.

Additional questions were added iteratively as the interviews and focus groups developed, giving the participants the opportunity to discuss other relevant issues. All individuals were debriefed at the end and contacted two days post-discussion to provide after care. All participants were offered a £20 gift voucher for their participation to compensate them for their time. Researchers debriefed after each interview to discuss any inconsistencies in interpretation.

3.12 MEASURES

At the point of consent all participants were asked to complete a demographic and clinical characteristics questionnaire to ensure, where possible, the inclusion of a diverse sample.

INTERVIEW SCHEDULES

Topic guides were used to structure the interviews and focus groups (appendices A17 and A18); these were primarily developed through PPI discussion. These discussions played a pivotal role in shaping the topic guides and provided insight into the most salient concerns, challenges, and needs of children and young people affected by cancer (Rb and wider diagnoses), ensuring that the research captured meaningful and relevant perspectives. Through consultations with several groups, topics such as personal coping strategies, perceived control over health, emotional adjustment post-treatment, and the long-term psychological impact of cancer were identified as key areas to discuss with Rb survivors.

Various psychological models that have previously been utilised within oncology and psychoeducation literature also informed my thinking, but did not directly form specific questions in the guide. These approaches ensured that the study addressed both the lived experiences of participants whilst being influenced by key theoretical constructs associated with coping, adaptation, and well-being. The health locus of control and the extent to which an individual attributed their health and ability to cope to internal or external factors, were considered (Wallston *et al.*, 1976; Lima, Moret-Tatay and Irigaray, 2021). The Common-Sense-Model of Illness Representations (CSM) framed topic guides through its' focus on the relationship between illness cognitions, perceived ability to cope, and psychological wellbeing (Leventhal, Meyer and Lorenz, 1980). Finally, The Adversity Restoration Compatibility (ARC) framework was drawn upon due to its' focus on how individuals make sense of living beyond their cancer experiences (Le Boutillier *et al.*, 2019).

Using a semi-structured approach, questions were open and allowed participants to discuss topics that felt meaningful to them. As I have conducted qualitative interviews before, I felt confident in my ability. However, to ensure my confidence as a doctoral researcher, I practiced using my topic guides with my supervisory team to check the flow of questions, and that they made sense to others. Prompts were available if needed, but often participants spoke naturally and gave rich detail to their experiences, unaided.

3.13 CONDUCTING THE ANALYSIS

Data was coded primarily using NVIVO data analysis software (QSR International PTY LTD., 2020). As discussed above, RTA (Braun and Clarke, 2021) was chosen because it allows for a flexible and nuanced exploration of complex qualitative data, making it particularly well-suited for understanding the rich and varied experiences of Rb captured in this data. This method is divided into six steps which are outlined below:

Step 1: Familiarisation with the data. This began with reading all the transcripts multiple times to immerse myself in the content, ensuring I developed a deep understanding of the participants' experiences and perspectives. As part of this process, I noted recurring patterns, unexpected insights, and questions arising from the data. Debra Howell (DH) read 10% of the data (approximately four transcripts), and we compared notes during a follow-up discussion to align our interpretations. These initial notes helped shape my approach to coding and provided a foundation for identifying key areas of focus related to the research aims.

Step 2: Generating initial codes. Using NVIVO, I systematically coded the entire dataset by highlighting and labeling segments of text that represented relevant features of the data. This process involved breaking down the transcripts into manageable units while keeping my research questions in mind. DH independently coded the same set of transcripts, and we cross-checked our codes to ensure reliability and consistency. During this stage, I also began grouping similar codes and made notes on how they might relate to broader patterns or themes.

Step 3: Developing themes. The development of themes was a collaborative and iterative process. After the initial coding, DH and I met to compare our coded datasets, discussing areas of agreement and divergence. I then began grouping related codes into potential themes, carefully considering how each theme captured meaningful patterns in the data. This step involved repeatedly revisiting the original transcripts to ensure that the themes represented the participants' voices authentically. I also explored possible relationships between themes, identifying overarching categories and emerging subthemes that contributed to a nuanced understanding of the data.

Step 4: Reviewing themes. This was an iterative process where I refined themes and subthemes alongside wider qualitative research colleagues. This involved refining and validating themes through multiple cycles of review. I worked with qualitative research colleagues, including Ann Hewison (AH), Rebecca Sheridan (RS), and Dorothy McCaughan (DM), who independently reviewed two transcripts each to validate the initial coding and themes. We then held group discussions to assess whether the themes adequately reflected the coded extracts and the dataset as a whole. Themes or subthemes that lacked coherence or relevance were revised or excluded. This collaborative review helped ensure that the final themes were robust, credible, and aligned with the research objectives.

Step 5: Defining and naming themes. During this phase, I further refined the identified themes to ensure they were clearly delineated and captured the essence of the participants' experiences. Each theme was defined in terms of its scope and relevance, and subthemes were identified to represent more nuanced aspects of the data. I created detailed descriptions and summaries for each theme, considering their relevance to the research aims and their ability to provide insights into the Rb community's experiences. This process also involved drafting concise, evocative names for each theme and subtheme to communicate their central ideas effectively.

Step 6: Producing the report. The final step involved synthesising the themes into a coherent narrative that reflected the research findings. I selected illustrative quotes from the transcripts to support each theme and subtheme, ensuring that the participants' voices were at the forefront. These quotes were carefully contextualised within the broader analysis to highlight their significance. Throughout the write-up, I aimed to provide a comprehensive and accessible account of the findings, linking the themes back to the research questions and objectives. This step was pivotal in translating the analysis into meaningful insights that could inform future support tools for the Rb community.

3.14 ANALYSIS

DEMOGRAPHICS

Overall, 32 individuals took part. Four focus groups were conducted with 15 teenagers, each lasting two hours, and 17 young adults were interviewed, for between 22 to 65 minutes (mean=43.3). Interviews were conducted with 17 young adults between 25th June 2022 and 14th December 2022. These took place at a date and time chosen by the participant, and according to their preference of video call or phone call. To enable choice and maximise accessibility, participants were offered the opportunity to undertake these interviews over the phone. In this event, all participants opted for video call interviews. Online focus groups were conducted with 15 teenagers on 26th August 2022, 26th October 2022, 20th December 2022, and 18th January 2023; refer to table 4 for details of focus group composition.

TABLE 4: FOCUS GROUP COMPOSITION

Location	**Number of participants**	**Age**	**M:F ratio**	**Ethnicity (self-defined)**	**Heritable: Non-heritable**	**Bilateral: Unilateral**	**Composition notes**
Online, 2 facilitators	6	13-16	2:4	4 white British 1 white other 1 mixed white British and other	2:4	2:4	Median age of diagnosis = 21.5 months, range 1 month – 2 years. 4 of 6 individuals treated with unilateral enucleation, 1 with double enucleation and total visual impairment. 1 participant opted to have camera off. 1 participant had to leave the group 30 minutes early. 1 participant had heritable Rb and lived with a completely blind Father.
Online, 2 facilitators, 1 observer	2	17-19	0:2	1 white British 1 mixed white British and Asian	1:1	1:1	Median age of diagnosis = 36 months, range 1 year - 5 years. Both individuals treated with unilateral enucleation.
Online, 2 facilitators	3	13-15	1:2	3 white British	1:2	2:1	Median age of diagnosis = 24 months, range 3 months – 2 years 1 month. 2 participants treated with unilateral enucleation.
Online, 2 facilitators	4	16-19	2:2	4 white British	1:3	0:4	Median age of diagnosis = 22.5 months, range 5 months – 4 years. 2 participants treated with unilateral enucleation.

ADOLESCENT SAMPLE

Fifteen teenagers from various locations across the UK and one from Malta took part in four focus groups, each lasting two hours. All were treated within the UK at the time of diagnosis. The sample consisted of 10 females and five males aged between 13 and 19 years (*median=15).* Ten reported having had non-genetic Rb, and five having genetic Rb. Ten were impacted unilaterally, and five bilaterally. Eleven had been treated with enucleation and used a prosthetic eye, and four retained both of their eyes. Thirteen described themselves as ‘White’ and two as ‘Mixed ethnicity’. For full adolescent demographic details please refer to table 5.

TABLE 5: INDIVIDUAL PARTICIPANT CHARACTERISTICS - ADOLESCENTS

Participant ID	Gender	Current age range	Age at diagnosis	Type of Rb	Uni/bilateral	Treatment
1	Female	13-15	2 years 6 months	Non-heritable	Unilateral	Enucleation
2	Female	13-15	3 years 5 months	Non-heritable*	Bilateral	Enucleation, chemotherapy, laser
3	Female	13-15	19 months	Non-heritable	Unilateral	Enucleation
4	Male	13-15	2 years	Heritable	Unilateral	Enucleation
5	Male	16-19	2 years	Non-heritable*	Bilateral	Enucleation, chemotherapy, radiotherapy, cryotherapy, laser, Intraocular melphalan
6	Female	13-15	1 month	Heritable	Unilateral	Chemotherapy
7	Male	16-19	3 years	Non-heritable	Unilateral	Chemotherapy, enucleation
8	Female	16-19	5 years	Non-heritable	Unilateral	Enucleation
9	Female	16-19	1 year	Heritable	Bilateral	Cryotherapy, chemotherapy, enucleation
10	Female	13-15	2 years	Non-heritable	Unilateral	Enucleation
11	Male	16-19	5 months	Non-heritable	Unilateral	Cryotherapy, chemotherapy, radiotherapy, laser
12	Female	16-19	9 months	Heritable	Unilateral	Chemotherapy, radiotherapy
13	Female	16-19	4 years	Non-heritable	Unilateral	Chemotherapy, enucleation
14	Male	13-15	3 months	Non-heritable*	Bilateral	Cryotherapy, laser, chemotherapy, radiotherapy, enucleation
15	Female	13-15	2 years	Heritable	Bilateral	Laser, chemotherapy

*Note. An age range is given instead of a precise age, to prevent participants becoming identifiable, * is used where participant has reported an inconsistent type of Rb*

YOUNG ADULT SAMPLE

Seventeen young adults were included in the sample, recruited from various locations across the UK, as well as one from Malta and all took part in individual interviews. All were treated within the UK at the time of diagnosis. The sample consisted of ten females and seven males aged between 20 and 29 years (*median=25*). Ten reported having had non-genetic Rb, six having genetic Rb, and one was unsure of the form of Rb they had had. Thirteen were impacted unilaterally, and four bilaterally. Thirteen had been treated with enucleation and used a prosthetic eye, and four retained both of their eyes. Fourteen described themselves as ‘white’, one ‘Indian’, one ‘Hispanic’, and one as ‘mixed ethnicity’. For full demographic details please refer to table 6. For in-depth demographic details of all participants, see table 7.

TABLE 6: INDIVIDUAL PARTICIPANT CHARACTERISTICS - YOUNG ADULTS

Participant ID	**Gender**	**Current age range**	**Age at diagnosis**	**Type of Rb**	**Uni/bilateral**	**Treatment**
16	Male	25-29	1year 5months	Non-heritable	Unilateral	Chemotherapy, enucleation
17	Male	20-24	2 years 3 months	Heritable	Unilateral	Enucleation
18	Male	20-24	2 years	Unknown*	Unilateral	Enucleation
19	Female	25-29	6 months	Non-heritable	Unilateral	Enucleation
20	Male	25-29	1 year	Non-heritable*	Bilateral	Cryotherapy, chemotherapy, enucleation, iridium wire
21	Female	25-29	3 years	Non-heritable	Unilateral	Enucleation
22	Female	20-24	7 months	Non-heritable	Unilateral	Enucleation
23	Male	25-29	Birth	Heritable	Unilateral	Chemotherapy
24	Female	25-29	3 years	Non-heritable	Unilateral	Chemotherapy
25	Female	25-29	2 years 6 months	Non-heritable	Unilateral	Enucleation
26	Female	20-24	2 weeks	Heritable	Bilateral	Cryotherapy, chemotherapy, radiotherapy, ruthenium plaque
27	Male	20-24	Birth	Heritable	Bilateral	Chemotherapy, radiotherapy, radioactive plaque
28	Female	25-29	5 months	Non-heritable	Unilateral	Enucleation

29	Male	20-24	11 months	Heritable	Unilateral	Enucleation
30	Female	25-29	3 months	Non-heritable	Unilateral	Enucleation
31	Female	20-24	3 years	Heritable	Unilateral	Enucleation
32	Female	25-29	18 months	Non-heritable*	Bilateral	Cryotherapy, radiotherapy, Enucleation

*Note. An age range is given instead of a precise age, to prevent participants becoming identifiable, * is used where participant has reported an inconsistent type of Rb*

TABLE 7: DEMOGRAPHIC DETAILS

	Participants N [%]	Adolescents (13-19)	Young adult (20-29)
Total	32 [100]	15 [47]	17 [53]
Type of Retinoblastoma			
• Heritable	10 [31]	5 [33]	5 [29]
• Non-heritable	21 [66]	10 [66]	11 [65]
• Unknown	1 [3]	0 [0]	1 [6]
Eye(s) impacted			
• Unilateral	23 [72]	10 [66]	13 [76]
• Bilateral	9 [28]	5 [33]	4 [24]
Median age in years (range)	20 (13-29)	15 (13-19)	25 (20-29)
Age at diagnosis			
• <6 months	8 [25]	3 [20]	5 [29.4]
• 7-11 months	5 [15.6]	2 [13.3]	3 [17.6]
• 1 year – 1 year 11 months	5 [15.6]	2 [13.3]	3 [17.6]
• 2 years – 2 years 11 months	8 [25]	5 [33.3]	3 [17.6]
• 3 years – 3 years 11 months	4 [12.5]	1 [6.6]	3 [17.6]
• 4 years – 4 years 11 months	1 [3.1]	1 [6.6]	0 [0]
• >5 years	1 [3.1]	1 [6.6]	0 [0]

Treating hospital			
• Birmingham Children’s	13 [41]	8 [53.3]	5 [29.4]
• Royal London/Great Ormond Street	19 [59]	7 [46.6]	12 [70.6]
Treatment (in isolation or combination)	24 [75]	10 [66.6]	14 [82.3]
• Enucleation	7 [22]	4 [26.6]	3 [17.6]
• Cryotherapy	17 [53]	6 [40]	11 [64.7]
• Systemic Chemotherapy	6 [19]	1 [6.6]	5 [29.4]
• Intra-Arterial Chemotherapy	5 [16]	4 [26.6]	1 [5.8]
• Laser therapy	7 [22]	0 [0]	7 [41.2]

N=number of participants; %=percentage; *=at time of study

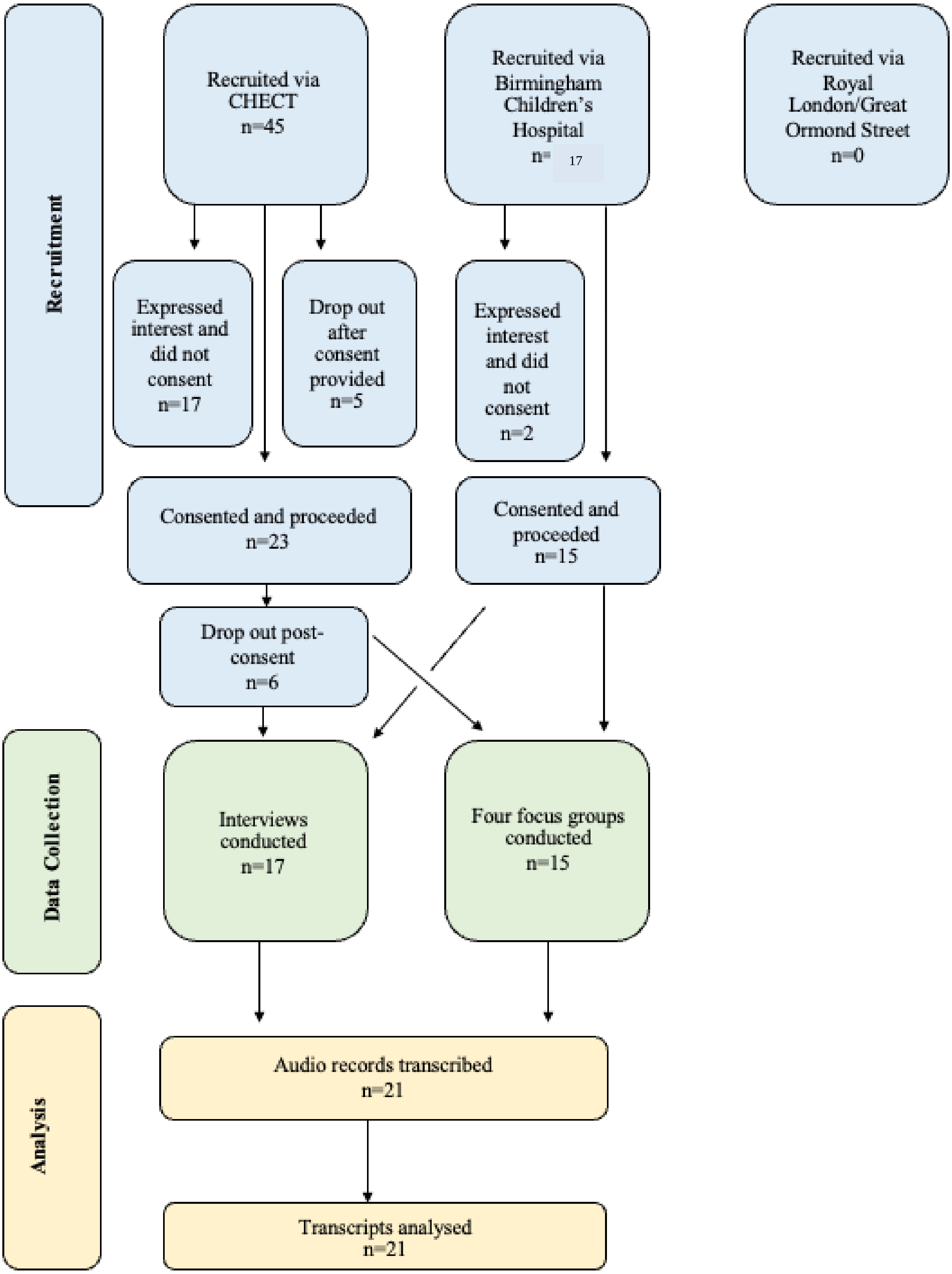


FIGURE 8: PARTICIPANT RECRUITMENT PROCESS

3.15 SUMMARY OF THEMES

Using Braun and Clarke's reflexive thematic analysis (Braun and Clarke, 2021), three themes and eight subthemes were identified:

1. Childhood 'the legacy of trauma' *(a) family experiences and survivor guilt, b) memories from treatment, c) life-long impact)*
2. Adolescence 'when you're a teenager, you feel like everything is the end of the world' *(a) psychological impact, b) identity, c) 'normal' for me)*
3. Adulthood 'it's not meant to be for life, but it carries on' *(a) acceptance, b) doing 'the work').*

The analysis was conducted inductively, with a decision made to broadly organise the themes around the concept of time, as much of the discussions were thinking back to childhood and remembering treatment and the impact on the family unit, adolescence and how having had Rb impacts your identity, and adulthood and the lasting psychosocial impact of Rb in the present. For in-depth details about the theory underpinning the methodology utilised, please refer to chapter two. All the themes and subthemes have been explored in greater depth within this chapter and accompanied by evidence of participant quotes.

Despite separating the participants by age and utilising different methods (interviews/focus groups) to elicit their experiences, I have synthesised all the study data into one analytical section. Originally, I had planned to do this in two separate sections, separated by age and method of data collection (focus group or individual interview). However, the themes are overarching and cross-cutting and do not differentiate by age groups. For example, the theme of 'identity' spanned the full age range of participants, with those who were teenagers at the time of interview stating that this was often a more challenging time than earlier in childhood, and young adults reflecting back that adapting to your condition gets easier as you mature. It therefore made more sense to provide a cohesive overview of these narratives, rather than separating them out by age. I do, however, often comment on the age of the participant in my analysis, as well as the method in which the data were collected, including the influence of others on this aspect of discussion if it was discussed as part of a focus group.

Quotes are reported with participant number (P) and by self-reported Rb type (bilateral as B, unilateral as U, heritable as H, non-heritable as NH. *e.g., P1, NH, U*). On occasion the Rb type reported by the individual was not in line with what we know to be biologically correct (e.g. it is not possible to have bilateral disease that is not heritable). This is discussed in further detail in the results and discussion sections of this chapter, and where there is a conflict of self-report this is marked with an Asterix (e.g. P2, B, NH*).

See figure 8 for a diagrammatic overview of themes and subthemes. To further support my analysis and evidence the processes used, coded transcript samples can be found in supplementary NVIVO files.

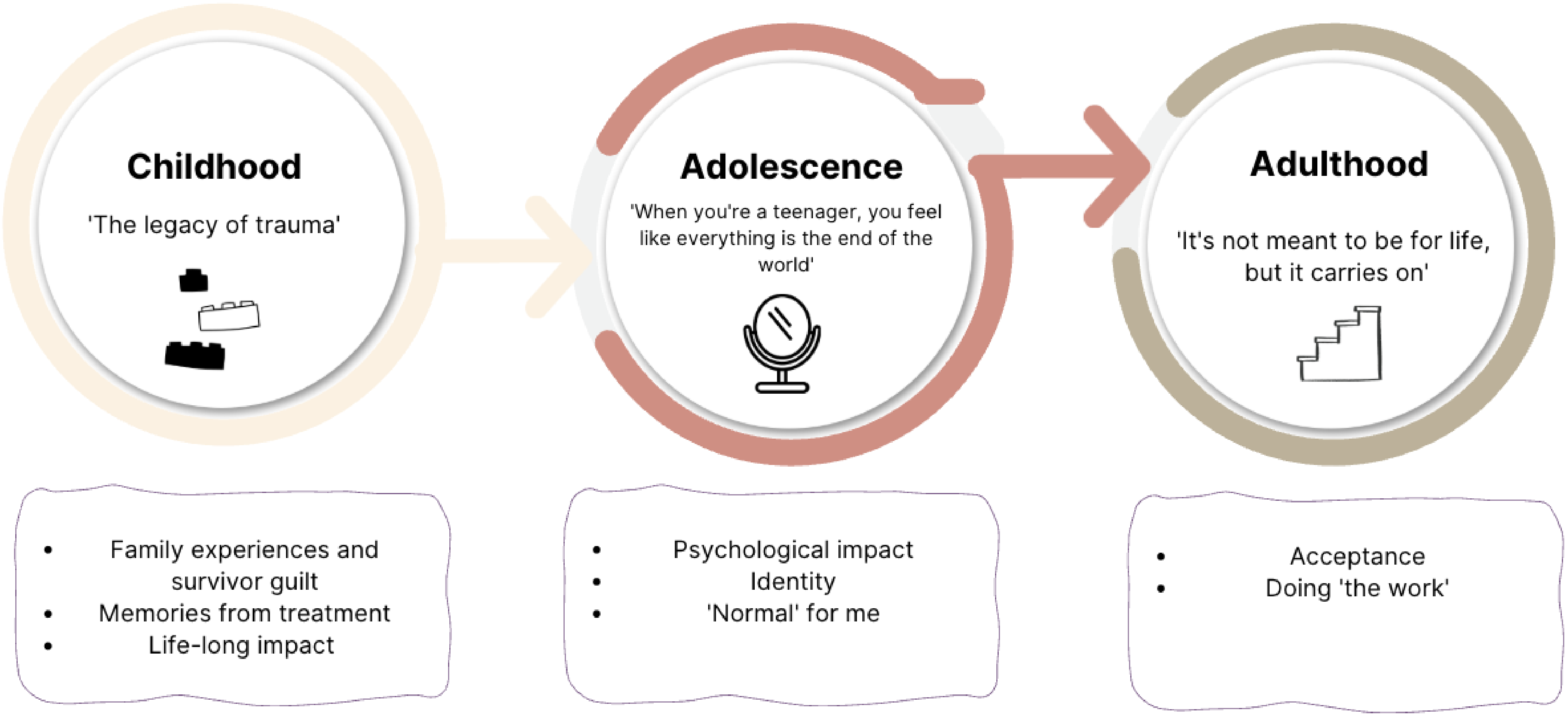


FIGURE 9: DIAGRAMMATIC OVERVIEW OF THEMES AND SUBTHEMES

THEME 1: CHILDHOOD – ‘THE LEGACY OF TRAUMA’ (P27, B, H)

One potential impact arising from a Rb diagnosis is life-long trauma, which is an issue that is clearly highlighted within almost all participant narratives. Variations of this legacy appear to be influenced by the age a child is diagnosed, the genetic nature of the diagnosis, wider family history of the condition, the severity of visual and facial impact, and late effects from the treatment received. The legacy of the trauma theme encapsulates ‘*family experiences and survivor guilt’*, which considers the role of parents and siblings in the trauma experience. It also considers ‘*memories from treatment’*, and how these link to the individuals’ emotional response and understanding of the world and others around them. Lastly, the combination of these trauma experiences and *‘the life-time impact’ on the individual* are considered. This is because individuals commonly spoke about continuing to carry the legacy of their experiences into adolescence and young adulthood. This understanding helps us to best conceptualise the level of psychosocial support that may be most beneficial.

1.1 FAMILY EXPERIENCES AND SURVIVOR GUILT

The subtheme ‘*family experiences and survivor guilt’* covers numerous factors incorporating individuals’ concern about the effect of their illness on their parents, generational trauma for those historically impacted by Rb themselves, as well as the shock of a diagnosis for those without a family history. It also considers the role of siblings on the affected and unaffected family members.

One young adult with a parent who was also a survivor of Rb spoke of the perceived *“emotional strain”* on his mother at having a child who was *“different [to] what you’d like” (P27, B, H).* As this participant was not the first in their family to have the condition, this suggests that even in these circumstances, there are complex feelings of guilt that linger from childhood experiences into young adulthood, as noted in the following quotation:

“it was the emotional strain that was on my parents having to look after this new-born child and having intravenous drips everywhere, and Mum not able to kind of have those first few weeks, with a child as you’d like, basically.” (P27, B, H)

Reflecting on this guilt felt important for many now-older individuals, empathising with the perceived suffering and stress of their parents. This view was shared amongst participants who were impacted by the heritable form of Rb, with one individual openly acknowledging that his parents' thoughts and emotional responses to diagnosis influenced his own. Referring to the perception that he took on all of his parents' thoughts from a young age indicated the vast influence that others may have on an individuals' thought processes. This is indicated in the quote below, in which death, dying, and serious health consequences were discussed, all of which are difficult concepts for an adult to comprehend, let alone a child.

"Obviously, I was impacted by my parents, so their thoughts would have been inflicted to me in a way…so ultimately that affected me, and I thought all the same things they were thinking about." (P17, U, H)

Despite this being more common in heritable instances, for some who were the only impacted family member (either through a non-heritable diagnosis or being the first heritable case in their family) these feelings were also apparent. For these individuals there was the added complexity of feeling *"bad [that they were] causing" (P32, B, NH*)* a difficulty that had not been faced by their family before. This sense of guilt was clearly evident in many conversations, with many individuals showing huge empathy for what parents must go through at the time of diagnosis and treatment. In this sample, the majority of participants had felt unable to share this perceived burden with their family members, largely keeping this to themselves to prevent causing further distress.

"They've [parents] never been through it. They can only imagine it…and I could only imagine that was a very hard job." (P32, B, NH)*

In focus group one, one teenager paid consideration to the fact that she was her parents' first-born child, feeling that this added weight to the significance of her diagnosis. As an individual with non-heritable Rb, these thoughts may have been more prominent because of the unexpected nature of a child being diagnosed with cancer. This individual was also diagnosed relatively 'late' at the age of five, meaning that they may have had more conscious understanding of what was happening and the emotional impact of this on the family. This suggests that it may be useful to take account of the age of the child at diagnosis and their stage of development, if psychosocial support is being considered at a later stage.

“I’m their first child and everything…there hasn’t been any history of my family or any of my relatives having cancer and… so I do think my parents did really have a hard time.” (P5, U, NH, focus group)

Interviewer: “Mm definitely – does that make it hard to, to talk about with them, or…?”

“I mean, not really but I think… ‘Cos obviously I have baby pictures of me…I didn’t have any hair, weirdly, I actually don’t like looking at those pictures, I don’t know why. I feel like it doesn’t remind me of something good. Even though I don’t remember it, like just subconsciously it’s just… yeah. But I think like sometimes it is okay to talk about because…the people who have supported me the most have been my parents…” (P5, U, NH, focus group)

The concept that Rb *“started with you” (P17, U, H)* was brought up by many participants, and survivor guilt and comparison to others was prominent. Many individuals used language such as “*difficult” (P24, U, NH)*, *“hard” (P5, U, NH, focus group),* or *“not easy” (P27, B, H)* when describing the impact that just being themselves had on others.

“I started this gene. So, my mother and father didn’t have it. My brother doesn’t have it…I’m the lucky one who started with this gene.” (P17, U, H)

This negative self-talk and the subsequent impact on self-esteem and behaviour is therefore important to consider.

“it makes you feel guilty a lot of the time when you’re not particularly easy.” (P27, B, H)

Many individuals also acknowledged empathy for their parents and the decisions that they had to make, and the wider impact on their own behaviour and feeling unable to talk to their parents about how they feel. This links to the need for young people to access information and support independently at an age and developmentally appropriate time, in a way that is autonomous from parents and family members.

"My Mum doesn't really like reliving it. She said that the few weeks between me getting diagnosed and my eye being removed, it's just a blur for her…. she doesn't really remember any of it, 'cos she was just going through the motions and stuff, so I don't really ask her about it." (P19, U, NH)

The dynamic of having siblings was also mentioned by many, both in families where more than one child was diagnosed with Rb, but also in those where the diagnosis was unique. For some, this led to a sense of resentment for those who were diagnosed, whilst unaffected siblings had the *"luxury [of feeling] relieved." (P31, U, NH)*

"My sister was like, "Oh well, do I have the genes for it?"… so she got tested and she doesn't have genes for it, which – you know, it was a relief for her." (P31, U, NH)

Siblings were not always mentioned in a negative light and were often appreciated for enabling a more 'normal' environment at home, instead of life revolving around hospital visits and treatment.

"I think I'm very lucky. My mum and dad kind of just treated me like they treated my brother and said, "You can do whatever the hell you want." (P22, U, NH)

For families with heritable Rb, the trauma of seeing your sibling experience treatment before you could be extremely difficult. For one young adult this was particularly prominent, as she also experienced the death of her sister (who had had Rb) from a second cancer. Individuals with heritable Rb are recognised as being at increased risk of further cancers, which inevitably raises difficult emotions for other family members.

"With my family and the treatment my sister went through, that did take a lot of toll…when you look back, you realise you did spend a lot of your childhood either in hospital for yourself or with her…and obviously when she passed away, that was really hard." (P26, B, H)

One first-born young adult with heritable Rb recounted a conversation that had been passed down through the generations of his family, relating to the possibility of him not surviving, due to a potential pregnancy termination because he might also have Rb. This highlights the added complexity introduced via religious and cultural beliefs, and family pressures around pregnancy and termination, emphasising the importance of reflecting on an individuals' wider social context when considering the implications on, and an intervention for, their psychosocial wellbeing.

"My mum had told me that she'd gone through this really distressing time, under pressure to not keep the pregnancy… I think they were really distressed and mum tells me… she had a kind of real breakdown moment where she was kind of like… should I/should I not? She's quite religious…she kind of said, "Okay, I think you know, God kind of told me no, you know, I've got to, I've got to keep the baby."" (P23, U, H)

1.2 MEMORIES FROM TREATMENT

As Rb is most commonly diagnosed in babies and young children, many individuals spoke about the expectation from others, and to some extent the reality, of not having fully formed memories of diagnosis and treatment. This feeds into the narrative of guilt and finding it hard to discuss with your parents, because of the perception that they "*had it worse" (P24, U, NH)* due to having more concrete memories of this time.

"Their [parents'] memory is vivid and it is that specific that they can remember every blood transfusion, every chemo…everything that went wrong, what day it was, what month it was, where they were at the time. So, I always just felt very guilty speaking about how I felt with them, because in my opinion, they'd had it worse than I had." (P24, U, NH)

Wide-ranging memories from treatment were discussed, countering the myth that having cancer at a young age means that you cannot recall it. These ranged from distressing, traumatic memories to more neutral or even fond recollections, particularly of the *"fun" (P25, U, NH)* and *"kindness" (P13, U, H, focus group)* that clinical staff instilled into procedures that are inherently traumatic, such as enucleation. In focus group two, one individual had particularly difficult memories that lasted beyond her treatment and expanded to other situations in which other people would get close to her face.

"I kind of remember kicking and screaming at the doctors, to be fair. I mean, I'm screaming at my mum when she had to remove herself 'cos she didn't wanna hurt me either, but I just remember absolutely throwing a fit because of it." (P5, U, NH, focus group)

Interviewer: "No, that sounds really difficult…do you remember how old you were?"

"Literally 5. And then 5 to the age of 10 that's when I started not liked anyone going near my face whatsoever." (P5, U, NH, focus group)

One young adult had more pleasant recollections, suggesting that individual differences will always play a part in how someone remembers their experiences.

"I have one memory from when I had my eye removed… I remember having my own little room with the curtain drawn around me, and a nurse trying to put a bandage over my face to cover – 'cos I don't think my prosthetic had been made yet. And I remember her trying to put this bandage across my face… and I remember continually pulling it off, and she was putting it back on, and we were laughing. And I remember my doctor coming into the room…he did my operation for me, and I remember running to him and him picking me up." (P25, U, NH)

Although many of the individuals in this sample received treatment whilst very young, key, often sensory memories, could be recalled vividly in adolescence and young adulthood. For some these memories were more dormant, being triggered by external sensory experiences. This was mostly focused on distinctive smells, which many stated elicited strong emotional reactions. For some this was combined with attending routine health appointments, which was more expected.

"certain smells set me off as well, like when you go to the dentist and you've got that anaesthetist kind of smell, that sort of is a memory smell for when I was then given eyedrops." (P24, U, NH)

For others it was more of a shock and seemed to come out of nowhere, with some describing feeling *'overwhelming sadness' (P7, U, H)* reminiscent of clinical trauma responses. In focus group one the teenagers shared several experiences where they found smells to be triggers to their Rb experience:

“There’s like some things with memories attached to them, so it’s kind of like – when I was younger, I used to have – have to have like check-ups - like the mask with anaesthetic, and I hated the mask. Like I was screaming and crying every time I had to have it.” (P1, U, NH, focus group)

“That happened with me as well.” (P7, U, H, focus group)

“so my dad had shoe polish or something, and it smells exactly the same, or metallic Sharpies, they smell the same, and they can make you feel like upset, ‘cos you associate it with that memory.” (P3, U, NH, focus group)

Interviewer: “Yeah. So – even the…it’s the smell of the thing that was connected to the memory, that brings it back and brings back those feelings, right. Does anyone else relate to that at all?”

“Yeah, the – the mask, especially – erm, they – they had to end up using a needle in the end ‘cos they just – the mask – I – it’s very – er, I wouldn’t say traumatic as such, but it – when like you do get like a sensation that reminds you of the things you went through, it does make your heart go a little bit, and you go like, “Oh alright then.”” (P5, U, NH, focus group)

Sometimes this experience was multi-faceted, particularly if it was combined with an experience of present distress, like experiencing teenage acne that impacts how you look.

“I was about 16, 17, getting the classic hormonal teenager spots … and so I bought some Clearasil to clean my face. I was in the bathroom and I put it on and I just immediately froze and freaked out a little bit because it triggered this memory of when I used to go in for a check-up or maybe it was surgery… I’d be held by one of my parents, and there would be this green mask with this green tube that they would put on me and I think it would knock me out and that had a very strong particular chemical smell and it was the exact same as the Clearasil, and I was kind of like, whoa! And that was always a real point of fear.” (P23, U, H)

For adolescents who were still at school at the time of this research, COVID-19 and safety measures brought in by the pandemic resurfaced memories that had been forgotten. This demonstrates that understanding is needed for both the individual and those around them, as if they are equipped with the knowledge that traumatic experiences can be triggered by external stimuli, coping mechanisms can be taught in advance. This was particularly apparent in focus group four:

"I can't remember the exact smell but I do have a recollection…it was always a certain type of hand sanitiser we used to have in school that always reminded me of the hospital 'cos you always had to put that on." (P8, U, NH, focus group)

"I was just going to say it's the same smell at like the hospital I think, and it reminds me." (P13, U, NH, focus group)

1.3 LIFE-LONG IMPACT

Understandably, many felt that the legacy of their experiences had shaped the person that they are today and will be throughout life. For some, this focused on feeling *"special" (P23, U, H)* and *"unique" (P32, B, NH*)* and this was most prominent for individuals when they were young children, before tapering off as they got older. For a few, however, this positive sense of self lasted throughout their lifetime due to the constant reassurance from their parents, instilling a confidence that it is ok to be yourself. In focus group one, one teenager spoke of the importance of external reinforcement:

"My mum just drilled it into me that, "Do you know what, you're unique, just go along with it, it's fine." (P5, U, NH, focus group)

Interviewer: "mm, yeah, so actually having people to support you is really important, absolutely."

"Yeah, pretty much." (P5, U, NH, focus group)

The idea that it is easier to *"be me" (P18, U, U*)* when you are young is reflective of a typical childhood experience, and it getting *"worse later on" (P25, U, NH)* is suggestive of the turbulence of adolescence, including transition to secondary school, hormonal changes, more complex social dynamics, seeking independence, and focus on appearance.

"When I was in junior school, up until the age of year six, I was fine, completely fine. In fact, I used to talk about it [Rb] all the time and bring it [prosthetic] into school – you know, bring my spare eyes into school and sort of show off a bit about it...it gets worse later on." (P25, U, NH)

Despite the best efforts of parents to instill a sense of pride in being themselves, for some the *"realisation" (P18, U, U*)* that you are not quite like "*everyone else*" *(P18, U, U*)* still leads to a sense of insecurity. This suggests that the onus of support should not just be left to families, but that the provision of a wider network of help is required.

"My parents definitely brought me up in the sense of, you're no different to anyone else, so I didn't really become aware of like what it [Rb] was or anything until I sort of realised myself." (P18, U, U)*

Some thought that the legacy of trauma had *"made me stronger" (P23, U, H)*, suggesting the possibility that individuals can be positively shaped from such a negative formative experience.

"If I didn't have RB when I was young, I would have been a totally different person. I would have different goals and aspirations in life." (P17, U, H)

This was sometimes linked to ambition and a desire to achieve, with some individuals setting themselves goals which they may otherwise have not aspired to.

"If I wanted to do something, I want to do it and I'll find a way."(P24, U, NH)

For others, it was the experiences of living beyond Rb that they felt were more important. For the majority of individuals with prosthetics this unfortunately included bullying and receiving unkind comments, which led to a variety of reactions. Individual differences were crucial here, with those identifying themselves as "*strong"(P31, U, H)* and seemingly more able to cope than those who identified as *"shy" (P10, U, NH, focus group)* or *"lacking confidence." (P28, U, NH)*

"There'd be the odd few [people] who would make a comment, but I was always quite strong in myself, in the fact that I wouldn't really let anyone say anything about it. Like I'd just bite back harder. So I shut them down pretty quickly, and no one really ever commented about it." (P31, U, H)

Comments from others were apparent at different stages, but were markedly less frequent in primary school, suggesting that insecurities begin during secondary school and beyond.

"When I was in primary school, I used to take my eye out and show people…and obviously it stuck with some people, and then when I joined secondary school, they obviously all remembered it…and they were just – it would just be awful, just like, you know, one eye, like a freak… all the like nasty things kids say." (P28, U, NH)

The negativity and bullying can be seen below, with others' actions directly impacting young Rb survivors' behaviour.

"In secondary school I was… I'm not gonna say like viciously – well, maybe – I was bullied 'cos I didn't fit in with everyone, 'cos obviously my confidence was quite low when I started. I was very much like a 'stay at home' kind of kid." (P29, U, NH)

Some individuals highlighted that verbal abuse can be just as harmful, with the words of bullies remaining in your mind and influencing self-esteem a long time after they were said.

"I've grown up basically getting abuse all throughout my whole life, so I've not ever been very confident about it. I went to an all girls' school, so it wasn't like very violent…it was always verbal, but I think verbal's worse sometimes…words stick with you more." (P19, U, NH)

These experiences and their impact on the individual also fed into ways of coping. The vast majority of participants had not accessed any psychological support to manage difficulties like bullying, and it was apparent across the sample that *"someone to talk to" (P26, B, H)* who wasn't a parent, or your doctor, would have been helpful from a young age. It was key that any individual providing support was specifically knowledgeable about Rb, as it was unanimously felt that this condition is so unique that *"generic" (P26, B, H)* support is not helpful. It was hoped by many that such support would also alleviate the sense of not managing difficult situations *"better" (P20, B, NH*)*, which is yet another burden.

"I would say to my younger self, you should have dealt with situations better and stood up to the bullies…I wouldn't be the person I am today if I had done that." (P20, B, NH)*

In terms of Rb-specific support, it was apparent that uniquely trained and supportive individuals with specialist understanding in prosthetic eyes and the specific difficulties that might arise were necessary. For example, many females who had been treated with enucleation found that, in an attempt to reduce comments from others, they would modify their hairstyle so that it acted as a *"security blanket." (P19, U, NH)*

"I had a huge thick fringe across half my face, and now when I look back at photos of myself, I'm like just, oh my god, why did anyone let me do that…I was just so anxious all the time that someone was gonna notice it [my eye]. It was a worry of mine throughout most of my childhood, I would say." (P28, U, NH)

This coping mechanism was shared across female participants, highlighting a common need to alter appearance in an attempt to hide their differences.

"I feel like it [my hair] made me feel a bit safer. Like it's kind of like a security blanket of, "Oh, no one can see it [my eye], so it's fine." But obviously they could see it, and my hair's dead thin, so you could always see it through it, but it just made me feel a bit better that it wasn't as noticeable 'cos it wasn't like on show. It was just like the odd little glimpse of it." (P19, U, NH)

For others, feeling different fuelled a determination to be seen as *"normal" (P22, U, NH)* and a desire to be someone other than a childhood cancer survivor. A need to not let Rb define who you are and the way that life is, was very common amongst the sample.

"I suppose there is a victim mentality, but I wanted to be judged on my own merits rather than as someone who had cancer as a child." (P16, U, NH)

For participants with comorbid conditions, this desire was particularly strong and appeared to lead to development of a tough exterior that prevented disclosure of other obvious health problems:

"I have asthma as well, and as silly as it sounds, I didn't want to be seen taking my inhaler, 'cos I thought that's another thing that's wrong with me." (P29, U, NH)

THEME 2: ADOLESCENCE – 'WHEN YOU'RE A TEENAGER, YOU FEEL LIKE EVERYTHING IS THE END OF THE WORLD' (P28, U, NH)

Adolescence was widely considered the most difficult period of life and a time when support skills are required, and this theme encapsulates three subthemes. The first, *'psychological, social, and behavioural impact',* reflects on participants' feelings of unfairness and powerlessness at both the perceived and actual restrictions caused by the Rb. It also reveals how being the survivor of a rare cancer can make it hard to express yourself, as many anxieties and difficulties are so unique that it is hard to find someone who has experienced the same or a similar trauma, who can relate to this. This ability to share feelings and be understood is a vital part of upholding good self-esteem, and without it left many feeling very *"lonely." (P22, U, NH)*

"I feel really lonely and I feel really weird." (P32, B, NH)*

The subsequent influence of these thoughts and feelings on personal behaviour are also considered. Finally, the conflict of feeling lucky to be alive and of downward comparison to those less fortunate are discussed. The second subtheme, *'identity'*, explores narratives common amongst teenagers, including discovery of who you are and how that fits with how others perceive you. Lastly, the subtheme, *'normal for me'*, considers how limited vision and/or wearing a prosthetic eye shapes life, the expectation that others openly and freely comment on what you look like without invitation, as well as asking for information about you, and what it is like to not know any different.

2.1 PSYCHOLOGICAL, SOCIAL, AND BEHAVIOURAL IMPACT

Across both the teenage and young adult participants, adolescence was named as the most difficult life stage, often being in *"sustained periods of stress mode" (P23, U, H)* as you manage all the 'normal' challenges of adolescence alongside the impact of being a childhood cancer survivor. Many felt that this period of life is when they felt most aware of how they looked, and experienced peers as being more critical too, adding to the sense of feeling *"abnormal." (P16, U, NH)*

"I guess when you're that age, the only thing you want to be is normal… it's on your face… like you're looking into people's eyes the whole time when you talk to them, so it's what you're greeted with. I guess being normal in that respect, it's the most important thing when you're that age." (P22, U, NH)

Thankfully these feelings were less frequent for many in adulthood, with the pressures of adolescence behind them.

"I was judged at that stage of my life, and I don't feel like I'm being judged now." (P20, B, NH)*

For some, these feelings led to a change in their behaviour, with many overcompensating for their eye(s) by trying to improve other areas of their image.

"I think there was a time where I was more worried about my appearance and whatnot and I was trying to compensate by getting in the gym and making myself look better." (P18, U, U)*

Others took a more self-deprecating approach, which was more common amongst current teenagers than young adults. This might reflect individual personalities, but equally could represent as shift in cultural thinking about body diversity, cancer, and mental health in the two decades between the oldest and youngest participants. In focus group three, many teenagers discussed that humour can be a helpful coping mechanism:

"I mean, you get like banter with people sort of saying stuff (laughs) but I actually quite enjoy that. I don't know, it's just like, you know, it's, it's something that happened, you might as well make it, have a joke about it. Erm, so I, I don't know, maybe that's just me but I quite enjoy (laughs) having banter with people about it really." (P14, B, NH, focus group)*

"Yeah I definitely agree... I've made so, so many jokes about my eye, it's unreal. Like, I always say to people if you don't laugh, you'll cry and that's the truth." (P13, U, H, focus group)

Interviewer: "has that always been something you've found quite easy to do, like sort of banter with people about it or has that got easier for you?"

"Yeah, no, I think so. I think I've always just found it, you know, just like it's something that happens and get on with it, you know." (P14, B, NH, focus group).*

Ironically, it was also widely discussed that adolescence was the time when you felt most unable to articulate your thoughts and to ask for or accept help.

"Like I do think I could have got help if I actually asked for it. I think my problem was that I probably just suffered a bit in silence." (P28, U, NH)

At the time when help was universally felt to be most needed (by both current teenagers and young adults reflecting back), it was equally the most difficult time to acquire support.

"if you had tried to tell me anything as a teenager, I would have told you to go away…I would not have listened at all." (P28, U, NH)

Participants suggested that the time at the end of primary school (approximately aged 10-11), before transitioning to secondary school might be the most useful to receive an intervention, offering coping strategies before they are needed.

"As I perceive it, I think the main benefit would be in the adolescence, the early teens, right through, probably starting from age 10." (P16, U, NH)

Accessing help appeared to be particularly problematic for the males in the sample, who often felt more isolated, and may reflect wider cultural norms and expectations that men *"just grit my teeth together and get through it" (P29 ,U, NH),* as well as wider stigma attached to men that expressing their emotions means you will be perceived as weak, as with the quote below.

"I feel like there's definitely a stigma attached to men nowadays, because there's like this whole thing of "Oh, you've got to be a man, you've got to be tough. Like you've got to be like Captain America…a strong, tough, alpha male, I guess."…That's what a lot of men want to be and what they think they have to be." (P29 ,U, NH).

This led to many male participants feeling that they had to manage difficult thoughts and feelings alone, compounding the idea that help is not available or accessible.

"I had that mentality in my head that, "Okay, I'm doing this alone." I always had my family and everything, but the main part is always dealing with it alone. So, honestly, I've never had the opportunity to speak to anyone relating to this circumstance." (P17, U, H)

For some this led them to engage in more maladaptive coping strategies, such as drug use:

"It would just feel so offensive to throw [my health] away and I did, I definitely have had periods across my life falling into quite heavy usage of marijuana during certain years of my life… in my teens." (P23, U, H)

and joining cult internet forums, often encountering men with views that would further reduce the likelihood of help-seeking.

"I found a lot of…how do I say it, relatability, online. And there were all these other angry young men who I thought, "Oh, these guys are the same as me," which they weren't… people

who've had bad experiences, when they feel inadequate, they'll eventually fall into this like circle or this group of less than pleasant people online… I was sort of like teetering on the edge of it." (P29 ,U, NH)

This did not differ across those with heritable and non-heritable Rb, and every male involved in this research stated that this was the first time they had felt able to speak about how Rb has impacted how they think and feel.

"*I've never spoken to anyone about anything to do with Rb." (P29 ,U, NH)*

This indicates that developing a culture of open discussion around mental health may be particularly crucial for boys before they enter adolescence. It also suggests that participants in this study may have received therapeutic benefits from taking part in this study, further highlighting the importance of providing a space to talk from a young age.

"I might ask just – just why the interest… Why – why did you choose Rb to do your research into?... it is a very, very kind of isolating for people that grow up with it… I was a bit hesitant at first to get back to you, I haven't spoken before…" (P27, B, H)

As these individuals reached the upper end of adolescence, they often behaved in the opposite way, with many appearing to develop a stronger responsibility to look after themselves than potentially that of their peers. Some of this appeared to have been as a result of having had many frank conversations with their medical teams, with some individuals having internalised the narrative given from doctor to patient in long-term follow-up (LTFU) clinic, usually attended between the ages of 16-18.

"Obviously, just keep an eye out for any like lumps and bumps and all that kind of stuff… they [LTFU doctors] always say, you know, "Make sure you wear sunscreen," and all that, because obviously, sarcomas are quite frequent." (P26, B, H)

This impact on behaviour change and self-care can of course be hugely positive in terms of living a healthy lifestyle:

"One of the hangover effects of RB is that I felt this sense of responsibility to look after myself, so I always felt quite strongly against things like smoking or excessive alcohol consumption, starting about 17 [years old] I suppose." (P23, U, NH)

but needs to be balanced in those who felt highly anxious and constantly surveying their bodies during adolescence:

"There was, umm, an incident where I had like a tiny lump somewhere …Obviously at that moment I thought, "Do I have it back?" Yeah, so having cancer again would be something that I wouldn't be really prepared for." (P5, U, NH, focus group)

Across both gender and age, many of the sample who had a prosthetic eye(s) described feelings of powerlessness to change their situation.

"Why was it me, why can't I have two eyes still?... Cos then I wouldn't have had people coming up behind me and smacking me in back of head expecting it [prosthetic eye] to fall out." (P21, U, NH)

The majority made particular reference to other people and their behaviour towards them, as well as the inherent comparison to peers who had not had Rb or did not have any visible differences.

"You know when people do comment on it, it's not something that you can change. So I think that's why sometimes it hits home a bit harder cos it's like, well I can't change it so if you could not comment on it that would be great. Whereas if someone doesn't like that you have long hair you could always cut your hair if it was that much of an issue but it's not something I can change, so I think that's why it probably stuck in my mind when people have said things." (P30, U, NH)

For some there was also an underlying fear that because of how they looked, other people would not care about them or their skills, dismissing them and making judgements based upon their appearance alone.

"It's more just like they're not really gonna care about what maybe I have to offer or what I'm saying or whatever, they could maybe just dismiss on the fact that oh, he's got wonky eyes, or he's got one eye or whatever … cos a lot of people judge a book by its cover, I don't blame them … but it's just, there's just more to a person than that if that makes sense." (P18, U, U)*

This fear of being seen as 'less than' others was also linked to the restrictions that having had Rb has caused, which became most apparent during the teenage years. Some of these were legitimate, particularly regarding careers that require a certain level of vision or being unable to pass the sight test needed to drive a car.

"All the military is complete write off, emergency services, all complete write off, I can't drive a car, some other things as well, so." (P14, B, NH, focus group)*

Learning to drive and exploring career choices are both key aspects of adolescence that, for many, are exciting and signal signs of adulthood and independence. For those who could not join their peers in these experiences, it was a disappointing and saddening time that further highlighted their differences.

"I sort of always knew that I couldn't do it, but I just held onto it like a pipe dream, if you know what I mean. I was thinking like, "Oh, I can maybe do it [drive], I can maybe do it." But it turns out I couldn't, obviously." (P29, U, NH)

For some with heritable Rb, they reflected that they were impacted by their family members' own experiences of restriction, including a male participant who grew up with a parent with the condition. This suggests that up-to-date guidance around the impact that reduced vision can have on life choices would be useful from a young age.

"I think that, especially in my teenage years, I took on this very pessimistic attitude towards work because of my dad's experiences." (P23, U, H)

Although it is important to acknowledge genuine restrictions that can result from Rb, there were many other instances where individuals had unnecessary restrictions put upon them by others, highlighting the negative impact that others' lack of understanding or excessive anxiety can have.

"It's just the fact that when I was younger they were like, "Oh, she can't do this, she can't do that, she won't be able to do this." ...you're already putting things in the way before I've started anything." (P21, U, NH)

In focus group four, two teenagers discussed this in depth:

"I do horse riding ...so people are like oh no, you can't do that, that's too dangerous, you won't be able to see straight." (P13, U, NH, focus group)

Interviewer: "it sounds like another example of people around you saying, that you can't do that or, worry about it when you know that there's ways around it"

"uh huh." (P13, U, NH, focus group)

"Well, I think I, when I done my driving test my mum was like, "Right, make sure you're looking," (laughs) like, "make no mistakes otherwise you'll probably fail," I was like, "Alright, okay," erm and yeah, no, I passed, with no minors and stuff, so…" (P8, U, NH, focus group)

For one adolescent in focus group one who lives with two prosthetics and complete blindness, the assumptions that people made about him were a particular challenge.

"I don't know if this is a side effect of just me being blind ...but a lot of people, for some strange reason, don't think I can use staircases…a lot of people talk to me like I'm a child…or what's even worse, if I'm out and about with somebody else, they talk to the person that's with me about me and not to me directly, which is very infuriating." (P6, B, NH, focus group)*

Sometimes, the restrictions were imposed by individuals themselves, with anxieties about what others might think or say about their appearance meaning that they did not allow themselves to get involved in valued activities.

"My drama teacher actually pulled me aside and she said to me "You have to make a decision." She said, "I totally understand why you have your fringe like that, and I'm not gonna tell you that you have to change it, but I am gonna tell you that if you want to go on stage, you need to have your eyes – you need to show your face, basically."" (P25, U, NH)

These inadvertently self-imposed barriers left many feeling out of place, isolated, and different.

"it just kind of made you feel on the side of things. I don't really know how to describe it. Like you weren't completely always part of a group, just because you're always aware there was maybe things that I couldn't do." (P22, U, NH)

High levels of anxiety were prominent across the sample, beginning in adolescence and remaining for many into adulthood. These were commonly linked to three broad areas: sex and relationships, having your own children, and developing second cancers. Sex and relationships were an important topic, particularly amongst young men. The perception of being ugly, or that others perceive you to be so, appeared to be devastating to confidence and the development of peer relationships.

"I never asked anyone out really at secondary school because I didn't have any confidence, and I felt like I was ugly and I always felt like women didn't like me." (P29,U, NH)

This was influenced by both peer groups and media perceptions of having one eye or reduced vision.

"It was really panic inducing… I just thought I look ugly. I thought people would think I was ugly or like different … you know when you're growing up, you're reading like Roald Dahl and watching pirate films, and it's all kind of this really ugly thing [one eye] that is attributed to people that are weird or mean, and so I think I didn't yet have the kind of separation from that. That wouldn't bother me now, but I think as a – as a teenager, you do feel more sensitive." (P25, U, NH)

In focus group one, there were differences in opinion and showed how the responses of others can vary, both across culture, how visible your differences are, and individual situations.

"I feel like the fact that I'm blind has also contributed to me having a harder time socialising…not many people talk to me." (P6, B, NH, focus group)*

"Oh, erm, I was just gonna say…it's very hit and miss with other people, especially, you know, during school. Some people will be okay with it. They'll not really bother. Like, you know, they'll just carry on as if you were normal. But some people will really, you know, kind of smoosh it in your face and that." (P1, U, NH, focus group)

For one young man in particular, he felt that having had Rb and living with a prosthetic had impacted his confidence and ability to engage in all of the typical activities that young people enjoy.

"So I hit 21 having no girlfriend, still a virgin…That's something that's been hard, you know, when everyone else seems to be having relationships, and you're just here wondering what's wrong with you… 'cos obviously, everyone in uni is about going out, clubbing, trying new things, having sex…and I didn't do any of that. Like another thing I'd say it's [Rb] had a big impact on is the relationship side of it." (P29, U, H)

These feelings were prolonged for two groups of individuals once they did meet a partner; those with heritable Rb and those who had a prosthetic eye. For the former, it was strongly felt that having Rb *"has an impact on relationships and who you choose to spend your life with" (P27B, H)*, feeling that they have to find a particularly understanding and generous person to have a relationship with.

"My partner now, who I live with, I didn't tell him until about a year into our relationship 'cos I was so worried about how he'd react, and he didn't give a crap, do you know what I mean? But I think it was just that worry, like, "Oh god."" (P28, U, NH)

This also included insight into the additional considerations that many with prosthetics have regarding how others view their eye(s).

"[as a teenager] you're more concerned about body image in terms of moving into romantic kind of settings, … and how people would view romantically your attractiveness…when you're talking to someone you're attracted to or things like this, trying to make sure you're sitting face on rather than having to turn, and maybe your eyes be convergent or divergent, which might be off-putting to other people." (P16, U, NH)

Decisions about having children was another common anxiety, particularly for young people with heritable Rb, who have a 50% chance of any future children also having the cancer. It was apparent that considerations about having children happened much earlier for this group than perhaps their peers, suggesting that childhood cancer forces individuals to make big life decisions at a younger age.

"I think the more pressing worry, especially as I've been with my partner for a number of years now... I think there's almost this feeling of placing a bet because it is, it is 50/50 whether that gene's inherited and that does weigh on my mind, it weighs on my mind about obviously that child but also the impact that that might have on my partner." (P23, U, H)

For some young men, this also linked into worries about considering themselves a *"burden" (P27B, H)* on others, if, as many young people do, he had a casual relationship or one night stand that ended with an unplanned pregnancy. The individual quoted here spoke at length about the impact and guilt that he felt he, and his condition, could have on others, including potential future children.

"I am less inclined to have casual flings, to be quite honest, because there's always the potential for that being an unfair burden for anybody to carry, basically, unless you're in a committed relationship." (P27,B, H)

This sense of morality and guilt was also brought up by teenagers, who despite not considering having children in the immediate future, were already aware of the potential risks. In focus group three, this concern was shared amongst both male and female participants, highlighting the need for both genders to be taught about reproduction after Rb.

“This is gonna sound really horrible, not horrible but weird, I don’t want my child to die ‘cos I know obviously some people can survive from it, like us, but like obviously some people do die from it and I feel like I’m just a bit worried if that was to happen to my kid.” (P10, U, NH, focus group)

“It’s the kind of moral question, is it right to bring something into the world that’ll have similar or possibly worse disabilities and effects because of what you’ve created.” (P14, B, NH, focus group)*

Interviewer: “Hmm, yeah, I mean really huge important questions and it sounds like the ones that certainly you two have both considered.”

“But erm, you know, there’s like IVF and all that, so.” (P14, B, NH, focus group)*

“Yeah, my mum tells me like it’s much better like how, how we have it, like back in, when she had me, erm, ‘cos now they can like check when the baby’s growing inside your stomach.” (P10, U, NH, focus group)

Although participants with non-heritable Rb are at population risk of inheritance and their children being impacted by the condition, this anxiety was also present for some.

“I just felt guilty more than anything that maybe that wasn’t a decision, to potentially put my child through something worse than what I ended up going through in the end.” (P24, U, NH)

This could partly be explained by a lack of understanding, particularly for teenage participants, signalling the need for young people to be given accurate information. As with focus group three, these concerns were echoed by teenagers in focus group one:

“You don’t wanna think, “Oh no, if I have kids, what if they have it as well and they have to go through it?” and things like that.” (P1, U, NH, focus group)

“Yeah, that’s definitely one I was like thinking, of – if – if you – if you were to want to have kids, would you pass it down to your child?” (P3, U, NH, focus group)

For others who appeared to have a good understanding of the genetics of Rb, the anxiety about the unknown was greater than the need for factual information. This feeling was particularly heightened for the individual in the example below, who was pregnant with her first baby at the time of interview.

"If I'm being really honest, it's a big worry. You're toying with the idea of whether it's a good idea to be pregnant in the first place and start a family, because it's so unknown… that's maybe what's worried me the most about all of it." (P24, U, NH)

Despite this, the importance and power of information was evident amongst teenagers who had recently attended long-term follow-up clinic, potentially the first appointment with their Rb team since they were discharged many years previously. The hope that could be instilled through the provision of up-to-date scientific information was powerful in calming previous anxious thoughts. In focus group four this concern was more prevalent amongst females:

"by the time I'm gonna have kids, science is evolving every day…cos when I went to Birmingham last time I had a meeting with a Doctor…and she was just telling me like there is things that you can do about it. So that's not really as much of a worry as it used to be." (P12, U, H, focus group)

One young man admitted this was not a consideration for him at this time, and may reflect general gender norms surrounding having children, or simply that females may be more likely to be spoken to about this from a younger age, as is the norm to place the onus of pregnancy prevention on young women.

Interviewer: "Does anyone else relate to either of those worries or have other worries?"

"I'm more worried about how I would do like driving…that's pretty much it." (P11, U, NH, focus group)

Interviewer: "So the other stuff's not really on your radar?"

“Erm not really.” (P11, U, NH, focus group)

Interviewer: “Uh huh, and that’s okay, there’s nothing wrong with that”

“I, I don’t really, I don’t plan on having kids anyway.” (P11, U, NH, focus group)

Understandably, the likelihood of being diagnosed with a second cancer was also highly worrying for many. Some of the language used about *“catching cancer” (P12,U, NH, focus group)*, as well as general anxiety about the likelihood of developing cancer again was indicative of the need for information.

“The side effects of like, catching cancer again is quite scary.” (P12,U, NH, focus group)

This was the case for both those in the general population (non-heritable Rb survivors) and for heritable survivors who have a higher risk of second malignancy. For those not at risk, there were misconceptions about their risk levels across both teenage and young adult participants.

“I’m not really too sure about – if it increases my risk of getting cancer in the future. I think it might do. I’m not really too sure.” (P29, U, NH)

The below quote shows a clear lack of information about risk and cancer re-occurrence, which may feed into unnecessary anxieties and self-restriction.

“I’m likely to have cancer again at some point in my life, it’s quite common for people that have had it once to have it again.” (P25, U, NH)

However some, again who had recently been to a long-term follow-up clinic, were more informed, highlighting the importance of good communication.

“I think the only thing for me is like obviously getting cancer back, but my doctors did say that I’ve got the same chance as anyone else now it’s not my major worry, it’s not the biggest worry I’ve got.” (P13, U, NH, focus group)

For the heritable survivors, this emphasises the need to provide an intervention that supports the management of this anxiety and tolerance of this uncertainty. In focus group two this anxiety was shared amongst both participants:

"'Cos we've got a high risk of it coming back compared to normal people, I think it's just the thought of that." (P9, B, H, focus group)

Interviewer: "Yeah, absolutely. I think it sounds like for both of you it's sort of in the background a little bit but it's not something that's troubling you every day, is that fair?"

"yeah" (P5, U, NH, focus group)

Young adults also expressed high levels of fear about what might happen in the future, cementing the need for continued support across the lifespan:

"Because unfortunately my sister was very unlucky, it kind of makes you think, "Oh well, surely, I must get something then." and as I say, 'cos my dad had a secondary one [cancer] – luckily, it wasn't too severe in his case. But yeah, you do think, "Is it inevitable or were they just really unlucky?" I don't know. So yeah, it does – does play on your mind a little bit." (P26, B, H)

This also included the well-documented fear that comes from hospital check-ups, a necessary part of post-treatment care:

"Perhaps there was a feeling that every time you go in [to hospital], you know, there's always the possibility that they say, "Oh no, it's gone – it's gone horribly wrong and they've come back, and you need to have all this treatment again."" (P27, B, H)

2.2 IDENTITY

For many young adults looking back to adolescence, there was a recognition that they had numerous questions about themselves, and how their history of having had Rb impacted their sense of self.

"it's just a time where you don't feel, you don't even know who you are, you're trying to figure out, you know, what kind of clothes am I supposed to wear, what's my style, to what do I want to do. Like, who am I?" (P32, B, NH)*

Individuals who were still adolescents at the time of interview expressed frustration that others around them made Rb a core feature of their identity, when they were in fact trying to hide this. This suggests that a level of acceptance of what has happened to you comes with age and 'doing the work', as discussed later.

In focus group four, teenagers discussed how they negotiate sharing information about their Rb experience:

"I tell people but don't make that the number one thing about me, like oh yeah, I had cancer." (P8, U, NH, focus group)

"I think, I think when I was younger, like, probably until I was 16, 17, I only used to tell people if I was like close with them, erm, but since then, I've like, if people like oh, like what's wrong with your eye and stuff, I'm just like well, I just tell them, just because I, I don't really care, like, if anyone thinks differently of me anymore, like, there's no point." (P12, U, H, focus group)

Similar conversations were had in focus group one, with greater emphasis on others' lack of understanding, which has resulted in judgemental questions and comments:

"Some people can be rather rude, especially with adults as well. They think, just because I'm a child or something, because I'm younger than them, they don't see me as equal to them. They don't really think that like, "Oh, perhaps this child has feelings because they've been through something."" (P1, U, NH, focus group)

Interviewer: "Yeah, yeah, we're getting agreement round the – er, round the screen for – for people who have similar experiences, by the looks of things."

"Erm, in school, 'cos I joined, er, high school later than some of my friends, what they did on my first day was they actually went around, erm, our year in the playground, saying, "Oh,

this is [name], she is partly blind." And then, erm, not many people judged. But I think they were waiting to hold their judgement after they asked like questions about it."(P2, B, NH, focus group)*

"To add on to what [name] said, like there's a lot of people that, when I'm having a conversation with, they'll ask me, erm, "Oh, am I allowed to say this?" or "Am I allowed to say that?" or – or something, something like – say – there's two people in front of me, and one of them's telling another person about my eye, they'll ask me, "How am I meant to say it?" And like, you know, "Am I allowed to say this? Will it – will it offend you?" and stuff." (P4, U, H, focus group)

When considering identity, many kept coming back to the idea that what they had been through was unfair, not only in terms of the diagnosis and having had childhood cancer, but for the impact that this had on perceptions of self, others, and the world around them.

"I always did ask why did it happen – this is a very rare disease, like why did it have to happen to me. I kind of still ask that now, like why me." (P29, U, NH)

This often included comparison with peers and others, which compounded the idea of difference.

"I'd go home and I'd say to my mum, like, "Why can't she [friend] get it, like why me? Why does it have to be me? It's so unfair."" (P10, U, NH, focus group)

Figuring out your identity in your teenage years was made more complicated by the nature of Rb, with many individuals doubting the legitimacy of their diagnosis and often hearing it described as one of the 'good cancers' to have due to the high survival rate. This was particularly prominent amongst people who had not been treated with enucleation and whom had no visible 'signs' of anything have ever been 'wrong', leading to them making downward comparisons between themselves and others.

"it would almost be a bit of a, I guess a joke that I'd say, "Oh, you know, I struggled as well," sitting around with all these people that aren't as fortunate as I was." (P24, U, NH)

Such comparisons were also noted as barriers to accessing support, particularly when individuals had been invited to generic cancer support groups with children who had cancers that they perceived to be ‘worse’ than Rb.

“It was kind of upsetting to me to be around other kids that also had cancer and had it worse than I did. And I think I was quite aware of that even when I was younger, that there were people that had it worse, and I felt kind of sad about that, so I didn’t engage with that [support] as much.” (P25, U, NH)

For some, such feelings extended to mental health. One example is the young person in the example below who questioned whether they had depression, or even the right to acknowledge depressed feelings as others had not had it ‘as bad’ as him.

“I’d say sometimes it’s a certain sense of like guilt that I had, ‘cos I’d be like, “Oh, even though I’m feeling bad…” I wouldn’t say I was depressed as such. Maybe I was, I don’t know. Maybe I am, I don’t know. But, I always thought there was a certain sense of guilt, like, oh, there’s people out there who have it a lot worse and, I don’t know, I’m just – I’m just moping about here.” (P29, U, NH)

This also had a complex interaction with feeling *“enormously lucky” (P27, B, H)* which appeared to be juxtaposed against legitimising the need for psychosocial support.

“I actually survived, you know, and I not only survived, but I can see.” (P32, B, NH)*

This was complicated further in individuals who had family members with the condition, as there was always a comparator of ‘luck’, meaning that many felt even more undeserving of support if they did not reach the self-imposed benchmark of ‘need’.

“I mean, I’ve always felt quite lucky. As I say, I’ve never had an eye removed and my sight’s been far better than theirs [family members] has, and I don’t know whether that was just luck of later developments and whatnot.” (P26, B, H)

As previously noted, appearance becomes a hugely important factor for many teenagers, and for survivors of Rb this is no different.

“I think, after the initial period of me feeling ashamed, which was when I was just starting to - you know, to hit puberty, which is when I had my fringe and I felt really insecure and really – I think it – it would have felt really bad then.” (P25, U, NH)

As discussed previously, this became most problematic for many around the start of secondary school, emphasising the need for support to be offered in advance of this time to aide coping skills.

“I remember like my first day of school, people being awful about it, and I think that’s ,probably where like the worries and stuff came from. That was when I like started covering it [eye] up.” (P28, U, NH)

For some (interviewed as adults), these formative experiences around appearance were long-lasting.

“I still have confidence issues now. They’re – they’re better, but they’re still not – I still don’t feel good about myself, I’d say, but, it’s not as bad as it was. Like throughout, secondary, I – I wouldn’t say I really had a good thought about myself, really.” (P29, U, NH)

This was particularly relevant for individuals who had been treated by others as if they were disabled, a label which the majority of the sample disagreed with. Many felt angry at others for putting this label on them, as well as others’ making assumptions about their capabilities, based upon factors such as their appearance or use of visual aids. This was discussed in-depth in focus group three, with teenagers discussing how school can often be a difficult and exposing environment:

“I don’t actually know why I don’t want people to know, but I don’t for some reason. I think it might be like the attention you get from it as well…you feel like your kind of, your secret’s out there…that’s why I didn’t use the cane anywhere near where I live.” (P14, B, NH, focus group)*

Interviewer: “Yeah. Hmm. (names), what do you think about people not knowing, do you feel similarly?”

"I wouldn't like to be asked questions all the time. So it's like I don't really want people to know." (P10, U, NH, focus group)

"It doesn't help fitting in when everything [school work] is like size 40 font and you have a teacher standing next to you the whole time…it's very necessary for school work to have a teacher stand next to you but it's also really annoying 'cos sometimes you feel like you're missing out on jokes or sitting next to people you want to because there's a teacher sat next to you the whole time." (P14, B, NH, focus group)*

For many adults this had a long-lasting impact on personal identity, that then had a knock-on effect on confidence and self-esteem.

"I knew people that had glasses, I knew people that were mentally disabled, but I was like why are you putting me in the same group? like no offence but I cannot relate". (P32U, NH)*

A small proportion of individuals expressed that their experience of Rb no longer bothers them – partially as a result of good social support but also due to maturity. It is important to consider how much of this attitude is genuine, and how much it may be a coping mechanism.

"The way I look I'm not really too fussed about anymore. Like you are who you are." (P29, U, NH)

It is of note that the majority of the quotes on this topic were from young men. This poses the question about the role gender has in coping and will be crucial to understand in terms of developing an accessible intervention.

"I used to care about how I look, but now I don't, I don't really know, I'm just a bit more, I don't really care anymore because it's not, it's not a big deal to people." (P10, U, NH, focus group)

2.3 'NORMAL' FOR ME

As Rb largely affects very young children, it is not a surprise that many individuals could not remember a life any different to what they know as a teenager or young adult. This was the same across individuals with and without prosthetics, as well as those who had facial changes as a result of their treatment.

"It's not really a big part of my life, to be honest, 'cos I was so young when I lost it [eye], I don't think about it much on a daily basis. It's just something that I kind of exist with." (P25, U, NH)

In focus groups two, one teenager was matter of fact about the norm of living as an Rb survivor:

"It's what you live with, it's what you get used to, do you know what I mean like, you don't know any different." (P5, U, NH, focus group)

Focus group four shared similar sentiments:

"I never remember being able to see out of two eyes, so you know, you just accept it." (P11, U, NH, focus group)

Interviewer: "Yeah, and I think from what we've heard from other people who've been in these situations it's often the other peoples' reactions and other people not knowing what to do rather than you kinda can just get on with it. Is that, would you say, is fair enough?"

"yeah." (P12, U, H, focus group)

"uh huh." (P13, U, H, focus group)

As previously discussed, it was often the influence of other people and comments that they made that reminded individuals of their differences.

"I came home one day and I was like, "Oh, what's a Cyclops?" And they [parents] were like, "Oh, it's a mythical creature with one eye." And I was like, "Oh, that's what this boy calls me at school."" (P30, U, NH)

Experiences of this were common throughout childhood and into young adulthood, from peers, friends, and strangers.

"So, one of my friends…It was New Year's Eve when we were all out in town. She said to me, "I can't believe how many guys are after you even though you only have one eye." And I just thought – I think she meant it as a compliment, but I did not take it as one." (P28, U, NH)

For individuals that chose to make a feature of and embrace their difference, in many cases through wearing a coloured or uniquely designed prosthetic, this often fuelled hateful comments and encouraged a culture of self-blame.

"I still get horrible comments … people tend to go, "Oh well, you're asking for it by wearing something like a coloured prosthetic." (P19, U, NH)

In line with this, many brought up the frequent questioning from others in response to any visible differences, suggesting that wider awareness around the condition might support survivors to avoid this issue. This was a topic brought up in three of the four focus groups, highlighting that this was a common experience amongst the youngest participants in the study.

In group one:

"Like sometimes, if they ask a lot of questions, it can be quite hassling, 'cos obviously you've been answering them for years and years." (P1, U, NH, focus group)

"Yeah, sometime I go to a group of people and tell them I had my eye scooped out, and then I'll see their reaction and know exactly what type of person they are." (P4, U, H, focus group).

"The hassle of explaining it to people made me just not wanna tell anyone when I went to secondary school… 'cos sometimes when people ask you questions you feel… a bit exposed… I don't really mind talking about it, but I don't really know why I don't want that many people to know." (P14, B, NH, focus group)*

In group two:

"I feel awkward, I'm not gonna lie, explaining "I had cancer when I was one." I dunno, it just feels a bit awkward to say it out loud because… I feel like before I go into like any new situation, for example, before I go to school… Obviously people have to know that I have an artificial eye for like school reasons and just, you know, generally but, if people do ask then I'm just like, "Yeah, I guess I… it is an artificial eye" Some people don't believe me, but yeah." (P5, U, NH, focus group)

"Yes, but, I mean, I don't know how to say it… I mean, you do get awkward 'cos then they start to sympathise and you don't know what else to say to it, you're just like, "Thanks, I really appreciate it," I guess. You don't know how to respond." (P9, B, H, focus group)

The realities of living with reduced vision and/or using a prosthetic was understandably problematic in several ways. Some felt obliged to wear prosthetics for the comfort of other people, rather than prioritising their own wellbeing. This was only discussed by teenage participants, which again suggests that increasing age is associated with a greater sense of self-assurance that you can be who you want to be regardless of what others might think or say. In focus group one, two individuals shared their difficult experiences:

"as a child, I used to wear it [prosthetic eye] a lot 'cos I didn't want to, you know, scare my classmates as such." (P1, U, NH, focus group)

"because both eyes are enucleated, what I have here are prosthetics, they get infected so goddamn easily, it's not even funny… and then it's just incredibly irritating when it happens." (P6, B, NH, focus group)*

In focus group two, one teenager shared a similar experience of feeling self-conscious:

"I felt like you could see that I had an artificial eye if I took it off or something like that. Like I would just never ever take it off in front of people. Obviously I took it off before I'd go to sleep but no one's gonna see me then." (P5, U, NH, focus group)

Different issues were considered in the adult participants, with many speaking more about the physical pain that they continued to live with as a result of having a prosthetic, but also from health conditions arising from having invasive procedures on their retained eyes.

"you get used to being in pain, as crazy as it sounds, you get used to it and you learn how to function and it's not easy, I'm not saying it's easy, I'm saying you get used to it and you figure out and develop ways around it to just survive because that's what it is, survival." (P32, B, NH)*

THEME 3: ADULTHOOD – 'IT'S NOT MEANT TO BE FOR LIFE, BUT IT CARRIES ON' *(P5, U, NH)*

Adulthood was both a time for reflection and projection, dependent on the life stage of the individual participant. This theme is divided into two subthemes, the first being *'acceptance'*, a state of being that was universally considered to be unachievable whilst still young. Reasons why are discussed below, and include a lack of choice, being able to validate yourself, and the power of seeing others who are like you. This led into the second subtheme, *'doing the work'*, encapsulating the need to seek out information to answer unknown questions, as well as the role of strategies such as therapy and peer support in improving psychosocial wellbeing.

3.1 ACCEPTANCE

In comparison to their teenage years, many young adults had developed the ability to accept themselves and their identity, acknowledging the experience of Rb without making it the only thing about them. It was clear that this required an understanding of the separation between acceptance and being grateful, and the distinct differences between the two.

"Don't compare yourself to anybody… You need to make your reality your possibility… figure out, write it down on a piece of paper what are you able to do... I started to accept, not appreciate, not be grateful, because I'm not grateful for what I've been through, I'm grateful for the strength that I developed to get out of it." (P32, B, NH)*

For some, adulthood brought a sense of discontent at feeling so negative about yourself and discovering a freedom that comes with embracing who you are. This was an active decision for many participants, who had worked hard to accept themselves and their differences.

“Probably when you’re just a bit of an adult, like you just don’t care as much. So, now I’m older like there’s literally nothing I can do to change it, do you know what I mean? Like I can mope about it or I can just kind of get on with it.” (P28, U, NH)

For some, this acceptance started slightly earlier in life, particularly if supported to accept their situation with therapy. This again emphasises that preventative care is crucial in helping children and teenagers to reach this stage as early as possible.

“Nowadays when I realised, you know, it’s nobody’s fault, it can’t actually be avoided, it’s just someone’s gonna get it so if it’s me, it’s me.” (P14, B, NH, focus group)*

The acknowledgement that the impact of having had Rb does not ‘go away’ seemed to come with time, with one individual highlighting the potential life-long impact of childhood cancer.

“It’s not meant to be life but it does carry on throughout your years.” (P5, U, NH, focus group)

The ability to validate your experience and reassure yourself was common, and for some this was attributed to making concerted effort to ‘make friends’ with Rb and how it has shaped your identity and your life, rather than fighting it and denying its’ existence.

“I’m gonna find my power in this thing, I’m gonna make this my motivation and I’m gonna be friends with myself. I’m gonna be friends with my illness… if you don’t present it as a weakness, people won’t take it as a weakness.” (P32, B, NH)*

For others, time was more impactful, providing learning experiences that adults are less likely than teenagers to comment on your differences.

"Because I worked in an office for like however many years…I didn't get one comment. I just – I was like, "Oh well, you know what, people don't actually like care", do you know what I mean? It just felt better." (P28, U, NH)

This was attributed to maturing and many adults being more tactful towards individual differences.

"In adulthood, I've never really had any issues. Obviously, people become more sensitive, especially in an age now, things are a bit more understanding, empathetic."(P16, U, NH)

In incidences where adults have commented on a survivor's appearance, many learnt that they were better able to cope than they had thought.

"I know people stare…if I'm meeting someone new and I know what they're thinking about while I'm trying to like talk to them about something and it's like the elephant in the room …then it usually comes out later on… "Oh yeah, I've got a prosthetic eye in one eye." But it doesn't bother me now." (P18, U, U)*

What appeared to be hugely important across the sample was the power that came from seeing someone who is like you. For many, peers were more important than professionals, having already spent a lot of time with clinicians throughout their lives. For teenagers and young adults this seemed particularly interlinked with social media and being able to connect with others in the Rb community, or even just being able to see a photo or a video of someone who may use a prosthetic, was incredibly powerful.

"I saw a girl on Instagram recently … she's really outspoken about it. It's the first time I'd ever seen anybody on social media take their eye out. And it was really shocking to me, 'cos I'd never seen anybody do that, you know…other than me, I've not ever seen it. So, I was like, "Woah, you're so brave." And it was even braver that she was doing it on the social – she had this platform." (P25, U, NH)

Much of the older participants in this sample grew up without the constant presence of social media, meaning that it had become particularly relevant in adulthood. Combined with the maturity to accept themselves and work on any difficult feelings about the impact of Rb, as well as a greater ability to move away from potential negativity found online, the power of social media felt most helpful for participants in the adult stage of life.

"I went to a meet up with people who've had RB [who I met online]... I found that amazing, because that really made me feel a lot better, just meeting other people who've gone through similar experiences...It made me feel so much better, 'cos I've never actually met anyone who had one eye before until that day." (P19, U, NH)

Many found it incredibly validating to feel part of a group, having spent much of adolescence feeling excluded and isolated from 'the norm'.

"It's nice that you've got someone who is in exactly the same position as you and has been through it, you know, can just tell you it's fine. I think just the whole telling you that it's gonna be fine is probably quite important, because at the time you're probably thinking – I know I definitely was, like, "this sucks"." (P22, U, NH)

This suggests that peer support may be an influential element of a future psychosocial intervention.

"I mean, [meeting others] it's kind of like a stress reliever and somewhere where you weren't judged, it was quite a nice little place, it was." (P5, U, NH, focus group)

3.2 DOING 'THE WORK'

"The work" (P32, B, NH)* is an overarching term that refers to many different coping strategies found to be helpful by people in the current sample. Firstly, the need to seek out information was hugely prominent in terms of developing personal understanding. The below quote that compares tumours to volcanos sums up the understanding that a child can have. Without the provision of accurate information, these beliefs can be carried into adulthood, alongside anxieties from childhood.

"I'd internalised that, that there's these tumours that could suddenly, you know, go off like a volcano." (P27,B, H)

There was also a huge focus on the need for *"information for me, not my parents" (P4, U, H, focus group).* Of course, in childhood cancers like Rb, it is often parents who are given the information and education at the time of diagnosis and treatment. Although there is now a greater push from the NHS for young people to take ownership of their health at long-term follow-up clinics, many of this sample felt that it was hard to know what you needed to know, what and who to ask for advice.

"The problem is I'm not aware of anything, and obviously now my mum's passed I don't really speak to my father, I don't really know… I need to like see a doctor probably to know if it's heritable or anything." (P18, U, U)*

This was also particularly prevalent in focus group one:

"it's small stuff like that that concerns me personally. Like basically, to sum it up in a word, how can I live my life without relying on other people to take my hand and – and gently guide my way through it, you know."(P1, U, NH, focus group)

Interviewer: "yeah"

"it's survival of the fittest." (P4, U, H, focus group)

"I – I think it's – it's really strange that they – like you just went from being in hospital, like completely cold, just to having to live." (P1, U, NH, focus group)

"For some reason, they [clinicians] told my parents the information that they had to know and they didn't tell me." (P4, U, H, focus group)

It was apparent across the sample that a sense of being proactive and having someone independent to talk to was important in terms of preventing difficulties later down the line.

"I didn't really understand it growing up. I think you need to be told early, then it prepares for the future if that makes sense, rather than just seek out the information when like you get curious or you need it." (P18,U, U)*

This in turn supports the idea that a preventative intervention may be crucial, offering information at an early stage which can then be built on as different situations occur.

"Because I was so young, it was a strange kind of process, and lots of things that were maybe a little bit more informative came later in life, so then it was trying to piece that together as I went along." (P24, U, NH)

One individual highlighted the fact that, in adulthood, therapy is now more widely accessible. As a child, particularly in generations older than current children, this is much more difficult unless there is support specifically offered to you.

"When you get to be an adult, you know, like if I need someone to speak to, I could go to like a therapist. Like I could seek that resource. But when you're a kid, you don't know what's available, do you? You just have no idea." (P28, U, NH)

This was perhaps particularly important to alleviate some of the anxieties discussed previously, as well as remove an element of the unknown.

"I don't know that much about it outside of what I've been told, which I don't feel like is a significant amount. I haven't done that much research into it. I don't really know how much I could do, how much is accessible for me. But yeah, I think it would be useful to do." (P25, U, NH)

For some, there was a fear that seeking out support could unearth old concerns, again suggesting that offering support from an earlier stage may prevent worries like this.

"I've never like just wanted to look for it, I don't think. Maybe I'm scared of some of the answers, I don't know." (P19, U, NH)

For some, this included learning how to navigate the healthcare system, suggesting that young people require further information about the NHS, what is available, and how to access this.

"I generally have no idea where to go or who I need to speak to or what. But it's just a case of like starting somewhere and then hopefully getting pointed in the right direction." (P18, U, U)*

Many adults found that 'the work' included accessing therapy, which many had integrated into their lives and found huge benefits from.

"I felt like I'd lost control of my life and I felt that therapy was like: oh, okay, yeah, you can, you can start living again." (P23, U, NH)

This also highlights that this may be useful in earlier life alongside peer support.

"It sort of feels like there could have been a few things nipped in the bud just by talking... psychological support in the sense of therapy, talking about my experience, talking about how I view the world; talking about how the impact of it on my family has impacted me." (P31, U, NH)

Others expressed a desire to have someone independent of your doctors and your parents to talk to but had not ever accessed this.

"Maybe having someone to talk to growing up – 'cos, you know, you have down days and you have up days, and if you're having a down day, maybe just talking to someone who isn't your parents might be helpful, who really, really gets it." (P31, U, H)

This is particularly important for those who chose not to or could not speak with their family members, offering a space to receive support without relying on parents.

"I even found it very difficult to talk about my worries to my parents when I was a kid. I – I just couldn't talk about them. I don't know why." (P29, U, NH)

It was common that individuals felt that this independent support should be linked into hospital appointments, meaning that you did not have to bring up the topic alone, as well as knowing that there was a space for you to talk if you needed it.

"It would be a good thing to offer, because then if you have a bad day and you want to, you can, or if you don't and you don't want to and you feel fine, then you don't have to." (P24, U, NH).

Even as adults, many found it hard to have the confidence or vocabulary to raise the topic with professionals, suggesting that having support available that could be flexibly tapped into throughout the life stages was important.

"When I was growing up, I always thought that I was the lone wolf sort of, you know. So, I always kept to myself. I always talked to myself... There could have been times where I should have talked to someone or something, expressed my feelings, but I didn't." (P17, U, H)

Some championed the idea of being taught coping strategies from as young as primary school age, avoiding ever having to *"battle" (P32, B, NH*)* in a way that many of the current participants have.

"Being the person that I am and having the complications or circumstances that I have because nobody taught me, I had to learn by finding it and by battling, and by figuring it out basically - I have learnt by myself and nobody gave me any tools how to deal with insecurities, how to deal with feeling different, or depression or you know, all that kind of stuff that I'm learning now, so yeah, support. That's all, that's all anyone needs really, support." (P32, B, NH)*

This was surmised by one adult who stated you *"just need someone to steer you in the right direction" (P23, U, H).* Poignantly, one young person gave an overview of how they would like psychosocial support integrated into routine clinical practice.

"It depends how old the child is., If they're young, like primary school age, you could bring it up like "how are your friends at school? Do any of the other kids say mean things about

you?" And then like as they get older, into secondary, maybe the end of primary, at least secondary, you could be like, "How is your changeover to secondary, are you coping? Like are you fitting in?" stuff like that. Then ask whether you're having problems with bullying, "Are people harassing you or assaulting you or being mean to you?"…making sure like no one's hurting you, 'cos that happened to me'…at the end [when you're discharged], maybe offer like a support service, if you want to come back, if you're still feeling like there's a reason to come back, if you want to talk to someone, I think that would be really good." (P29, U, NH)

3.16 DISCUSSION

Here, the findings of this study will be critically appraised and interpreted in relation to both the research aims and existing literature. I will provide my interpretations and observations of the data, highlighting their significance and implications within Rb research, clinical practice, and policy.

3.17 MAIN FINDINGS AND THEIR RELATION TO EXISTING LITERATURE

This study aimed to explore young people's experiences of living beyond Rb, understanding their psychosocial needs, any psychosocial challenges that arose during adolescence and young adulthood, and seeking their opinions on psychosocial support. For most, the experience of having had Rb caused long-term distress, heightened feelings of anxiety and lowered self-esteem. Despite this, many individuals also found that the experience developed their resilience and enhanced coping abilities, resulting in robust coping strategies that may not have otherwise been developed. My findings expand upon previous literature and identified themes which were unified across Rb type, therefore providing a unique contribution to the growing evidence base for psychosocial support requirements for young Rb survivors.

One potential impact arising from Rb diagnosis is life-long trauma, an experience that was clearly highlighted within almost all participant narratives. Variations of this legacy appear to be influenced by the age a child is diagnosed, the genetic nature of the diagnosis, wider family history of the condition, the severity of visual and facial impact, and late effects from the treatment received. This first theme encapsulated 'family experiences and survivor guilt', which considered the role of parents and siblings in the trauma experience. The experiences of family members, particularly parents, is much better understood than that of the individual affected by Rb themselves (Ek, 2000; Hamama-Raz, Rot and Buchbinder, 2012; Gibbs, Reynolds and Shea Yates, 2022). This may be because, as previously mentioned, the vast majority of Rb diagnoses are made when a child is very young, meaning that it is usually the parents or carers who are required to take on information and make decisions about treatment. The current findings explored survivors' experiences of feeling burdensome to their families, both when they were the first in their family to develop Rb or feeling that they were contributing to generational trauma for those with a family history of the disease. Many of the individuals who took part in this study shared complex thoughts about 'putting their parents through difficulty', which may have rooting in reality, as suggested by Hamama-Raz et al. through conversations with parents about emotional difficulties and anxiety associated with authorising medics to treat their child for Rb. This was also prevalent in Gibbs et al.'s recent paper, which suggested that parents found learning to adapt to a diagnosis, treatment, and in some cases enucleation and use of a prosthetic eye, stressful. Many individuals in the current study acknowledged empathy for their parents and the decisions that they had to make regarding treatment, and the wider impact on their own behaviour and feeling unable to talk to their parents about how they feel. This links to the need for young people to access information and support independently at an age- and developmentally appropriate time, in a way that is autonomous from parents and family members. Again, this shares the same recommendations as recent literature, which suggests that child survivors of Rb should have access to resilience strategies to help them manage living beyond cancer (Gibbs, Reynolds and Shea Yates, 2022). Having said this, research such as this often focuses on the needs of younger children as opposed to teenagers and young adults. Whilst it is positive to see recommendations of support for individuals whilst young (such as the implementation of 'Eye Club', a group intervention for young children living with an Rb-related prosthetic (Gibbs, Reynolds and Shea Yates, 2022)), my findings emphasise the gap in support for older survivors who must also manage the complexities of growing up in the context of being an Rb survivor.

It also considered 'memories from treatment', and how these link to the individuals' emotional response and understanding of the world and others around them. This is a phenomenon discussed in previous childhood cancer survivor literature (Molinaro and Fletcher, 2017; Hinton *et al.*, 2022), but has not previously been made specific to Rb and/or cancers affecting very young children. Findings from the current study highlight that Rb survivors who were as young as one month old at diagnosis have pre-verbal, often sensory, salient recollections of this period. This shares findings with research into individuals who experience cancer as teenagers or young adults, with cancer diagnosed earlier in life disrupting key developmental stages and shaping cognitive processes (Sansom-Daly *et al.*, 2018). This fits with the current findings considering trauma experiences and 'the life-time impact' on the individual. Many individuals spoke about continuing to carry the legacy of their experiences into adolescence and young adulthood. This understanding helps us to best conceptualise the level of psychosocial support that may be most beneficial. These findings also fit with wider trauma literature (discussed in chapter one) which suggests that negative emotional memories (such as distressing medical procedures) are stored differently to those that are neutral (Goodman *et al.*, 2019; Maddox *et al.*, 2019). Memories relating to traumatic experiences or threat can become intrusive and lead to maladaptive responses later in life, such as some of those discussed in the current study. This is the case even if the threat is perceived and not actual, such as the fear that a medical professional is trying to harm you, when they are trying to examine your eye. These findings provide novel evidence to the Rb literature and expansion to wider trauma understanding, highlighting the long-term impact that medical trauma can have on a child as they grow older.

Although there were numerous different experiences expressed in the current study, some participants thought that the legacy of trauma had "made them stronger" suggesting the possibility that individuals can be positively shaped from such a negative formative experience. This was sometimes linked to ambition and a desire to achieve, with some individuals setting themselves goals which they may otherwise have not aspired to. This again, correlates with existing childhood cancer survivorship literature, which recognises that many individuals feel positively about having had a cancer diagnosis (Duran, 2013; Koutná *et al.*, 2017; Marziliano, Tuman and Moyer, 2020), with Tedeschi and Calhoun terming this 'post-traumatic growth' (R G Tedeschi and Calhoun, 1996; Chen *et al.*, 2020). This can be particularly relevant to children diagnosed at a very young age, who can often adapt to their situation well as they cannot recall ever living differently; a position shared by many in the current study.

Theme two focused on experiences of adolescence, a time which was widely considered the most difficult period of life and a time when support skills are required, and this theme had three subthemes. The first, ‘psychological impact’, reflected on participants’ feelings of unfairness and powerlessness at both the perceived and actual restrictions caused by the Rb. It also revealed how being the survivor of a rare cancer can make it hard to express yourself, as many anxieties and difficulties are so unique that it is hard to find someone who has experienced the same or a similar trauma who can relate to this. Previous literature has referred to this as ‘negotiating the psychosocial challenges from two different worlds – ‘the healthy’ and ‘the ill’’ (Larsen *et al.*, 2022). The ability to share feelings and be understood is a vital part of upholding good self-esteem, and without this left many participants feeling very “lonely”. This contradicts previous work which suggested that Rb survivors report lower levels of depressive, anxious, and somatic symptoms than non-Rb childhood cancer survivors (Jennifer S. Ford *et al.*, 2015). Our findings could argue that this is not reflective of survivors’ true feelings but could be indicative of a reluctance to disclose complex emotional responses and distress through fear of being judged for feeling this way. This is more in line with findings from van Dijk et al. (2010) who found that adult survivors of Rb have 20% more psychological difficulties, such as anxiety and depression, than a healthy comparison group. This finding was echoed by Morse et al. (2023), who found that although Rb survivors report typical emotional health and quality of life, they are more anxious than a comparison group of peers who have not experienced cancer. The current study explored the sense that Rb is seen as a ‘good cancer’, meaning that this perception could act as a barrier to help-seeking and emotional expression. This sentiment has been shared by other cancer survivors who are told that they have a ‘good’ cancer, meaning that they are unlikely to lose their life from this, or may live ‘with’ cancer for an extended period of time and therefore should feel ‘grateful’ (Easley, Miedema and Robinson, 2013; Randle *et al.*, 2017; Hewison *et al.*, 2020; Howell *et al.*, 2022). These studies highlight the feeling that some cancer experiences can be downplayed by others, meaning that individuals felt that they were not ‘genuine’ survivors or ‘entitled’ to support. Hewison’s synthesis expanded on this further, with participants across nine studies living with chronic myeloid leukaemia fearing that health care professionals viewed their cancer as ‘low key’, which may discourage them from seeking support. These are important findings to follow up within the Rb literature, particularly when considering the development of a specific psychosocial intervention.

Ironically, it was widely discussed that adolescence was the time when individuals felt most unable to articulate their thoughts and to ask for or accept help. At the time when help was universally felt to be most needed, it was equally the most difficult time to acquire support. This suggests that the time at the end of primary school (approximately aged 10-11), before transitioning to secondary school might be the most useful to receive an intervention. This is a period of time generally considered to be challenging for any child (Hanewald, 2013), therefore it seems an appropriate time to offer coping strategies to childhood cancer survivors before they are needed and at an age where support could be more easily accepted. A high proportion of individuals in the current sample expressed feeling uncertain about many aspects of life, notably sex and relationships, having your own children, and developing second cancers. Feelings of uncertainty are well documented amongst cancer survivors of all ages, with individuals often over-appraising the likelihood that something ‘bad’ will happen again (Miller, 2014; Belpame *et al.*, 2019; Benedict *et al.*, 2020; Newton *et al.,* 2021; Shay, Allicock and Li, 2022). These feelings can be amplified further when access to information is limited, social support is reduced, and when encountering ambiguous health care systems (Donovan *et al.*, 2015).

As may be expected of adolescence, identity was a key theme and for many there was a recognition that they had numerous questions about themselves, and how their history of having had Rb impacted their sense of self. Individuals who were still teenagers at the time of interview expressed frustration that others around them made Rb a core feature of their identity, when they were often trying to hide this. As with other components of the current findings, this is shared amongst wider childhood cancer survivor literature (e.g. Granek et al., 2012; Ingersgaard et al., 2021). Having said this, Rb can differ to other childhood cancers in that it can leave individuals with a physical reminder of illness and/or treatment, most commonly through being treated with enucleation and using a prosthetic eye. This is a unique contribution to the literature, with most existing studies either focusing on temporary changes to appearance during cancer treatment (e.g. loss of hair from chemotherapy, or weight gain due to steroid treatment), permanent changes post-cancer that can be more easily hidden (e.g. breast cancer survivors treated with mastectomy that can be modified with implant surgery or a breast prothesis under clothing) or permanent changes related to non-cancerous illnesses or accidents (e.g. burns survivors, cleft palate, scarring, or inherited conditions like Neurofibromatosis), which come with different psychosocial impacts to cancer survivorship. Of course, any of the above conditions can be deeply distressing with vast psychosocial impact (Wallace *et al.*, 2007; Williamson *et al.*, 2010; Jenkinson *et al.*, 2015; Norman and Timothy P. Moss, 2015; Brierley *et al.*, 2019), but Rb (particularly when heritable e.g. Gregersen et al. (2021) or treated with enucleation e.g. Banerjee et al. (2020)) is unique and must be supported as such. Having said this, it is important to acknowledge that as treatment protocols change, fewer children diagnosed with Rb are expected to be treated with enucleation (Ancona-Lezama, Dalvin and Shields, 2020). As discussed, a large proportion of the participants in this study were treated with enucleation, as is common for survivors of their generation. Their experiences and need for support (e.g. bullying due to loss of an eye) may therefore not be relevant for survivors who are not treated in the same way. It is therefore important that any support that is developed is adaptable to the needs of many survivors; although this research highlights many overarching themes, a modular approach for a psychosocial support interventions is recommended.

The final theme focused on adulthood, which was both a time for reflection and projection, dependent on the life stage of the individual participant at time of discussion. This theme was divided into two subthemes, the first being ‘acceptance’, a state of being that was universally considered to be unachievable whilst still young. Reasons why include a lack of choice, being able to validate yourself, and the power of seeing others who are like you. This led into the second subtheme, ‘doing the work’, encapsulating the need to seek out information to answer unknown questions, as well as the role of strategies such as therapy and peer support in improving psychosocial wellbeing.

In comparison to their teenage years, many young adults had developed the ability to accept themselves and their identity, acknowledging the experience of Rb without making it the only thing about them. It was clear that this required an understanding of the separation between acceptance and being grateful, and the distinct differences between the two. This finding is shared by wider cancer survivorship literature, which emphasises the psychosocial challenges unique to adolescent cancer survivors who must manage ‘normal’ life whilst working through questions about their identity (Janin *et al.*, 2018), interpersonal relationships (Barnett *et al.*, 2016), education (Fardell *et al.*, 2017), and cancer-specific anxieties (Rourke, Samson and Kazak, 2015; Friend *et al.*, 2018). These are thought to ease as individuals transition to young adulthood, particularly if supported by specific psychological support, many of which have a specific acceptance component (Osborn, Demoncada and Feuerstein, 2006; O’Conner-Von, 2009; Hulbert-Williams, Storey and Wilson, 2015; Arch and Mitchell, 2016; González-Fernández and Fernández-Rodríguez, 2019; Arch *et al.*, 2021; Clarke *et al.*, 2021; Nilsson, Segerstad and Olsson, 2022). This fed into the final subtheme and doing “the work”; an overarching term that referred to many different coping strategies found to be helpful by people in the current sample. In the current study, many adults found that ‘the work’ included accessing therapy, which many had integrated into their lives and found huge benefits from. This is shared with wider cancer survivorship literature, with several randomised controlled trials finding that psychological therapies have a significant positive impact on wellbeing (van der Spek *et al.*, 2017; Holtmaat *et al.*, 2020; Lleras de Frutos *et al.*, 2020) These findings, combined with the current study, also highlights that structured psychological therapy may be useful in earlier life alongside peer support, another factor which was named as useful for Rb survivors.

In addition, the need to seek out information was hugely prominent in terms of developing personal understanding. Such information surrounded heritability of disease (and therefore implications on other family members, and having children in the future), and risk of second malignancies. There was a huge focus on the need for education and “information for me, not my parents”. This need was emphasised by the relatively high number of participants who were unsure or gave incongruent answers when discussing Rb and its’ impact on them. Of course, in childhood cancers like Rb, it is often parents who are given the information and education at the time of diagnosis and treatment (Landman-Parker, 2017; Sadak, 2017; Szalda *et al.*, 2017). Although there is now a greater push from the NHS for young people to take ownership of their health at long-term follow-up clinics (Michel *et al.*, 2019; Knighting *et al.*, 2020), many of this sample felt that it was hard to know what you needed to know, what and who to ask for advice. This sentiment was shared in a national cohort study of long-term childhood, adolescent, and young adult cancer survivors (Gianinazzi, Kiserud, Ruud, & Lie, 2022). Findings suggested that when asked, 50-60% of 5361 survivors felt that they did not have enough information about their cancer and the treatment received, or about late effects. This suggests that for childhood cancer as a whole, there needs to be a wider emphasis on the survivor themselves accessing information about their condition, supporting the concept that those who have better knowledge of their health are more likely to feel a sense of acceptance about their situation and are more likely to seek support.

3.18 STRENGTHS AND LIMITATIONS

To my knowledge, this is one of the first qualitative studies which has sought to understand wide-ranging psychosocial experiences of Rb survivors, regardless of whether they were impacted heritably or non-heritably or uni- or bilaterally. This research therefore provides in-depth insight into the psychological complexity of rare childhood cancer survivorship, highlighting the sensitive needs of individuals as they transition from childhood to adolescence, to young adulthood and beyond. It is a real strength of this study to have successfully recruited a large sample size of 32 individuals for a qualitative study. It was anticipated that, due to the rarity of the condition and thus the small recruitment pool, it may be difficult to recruit enough participants to adhere to information power guidelines. Despite this, I was amazed at the response and was fortunate to exceed my recruitment target very quickly, to the extent of having to turn away interested parties. Additionally, I was contacted by ineligible individuals who were older than the upper age limit (29 years). This further highlights the desire from the Rb community for psychosocial investigation into survivorship.

It is important to reflect on the strengths and limitations of my chosen method of analysis, reflexive thematic analysis. One of the significant strengths of this methodology is the emphasis on researcher reflexivity, which encourages continual, in-depth engagement with the data whilst simultaneously acknowledging my role in interpretating the narratives that I am collecting. Unlike other qualitative methods, this emphasis on reporting reflexive processes helps to mitigate against bias by acknowledging that it is inevitable, encouraging researchers to name how they are relating to the data and make this transparent for the reader. Ultimately it is hoped that this enhances the credibility of the findings and highlights the strength of using robust qualitative methods. In addition, reflexive thematic analysis is a flexible approach which allowed me to combine different methods of data collection through conducting both interviews and focus groups. At the outset of my research I did not intend to analyse the data as one, but as discussed previously it made sense to do so due to the homogeneity of themes across adolescent and young adult participants. As an iterative process, this approach allowed me to continually revisit and refine my themes over a long period, holding discussions with my wider research team to delve into the nuances of participant meaning.

Whilst this approach is effective, it is not without limitation. As described above, one key challenge is the subjective nature of interpretation inherent in qualitative analysis. It is well understood that different researchers will interpret data differently, meaning that different individuals will not present data in the same way. Whilst this could be a challenge, I continually engaged with the reflexive process and reflected on the assumptions and biases which could be interfering with the analysis. More can be read about this process in chapter two. Lastly, due to the open-ended nature of reflexive thematic analysis, combined with broad research aims, it was sometimes difficult to focus on the most prominent themes without becoming distracted by other interesting data that was not relevant to the main research question. Striking this balance could be challenging but utilising supervision and maintaining a reflective research journal was a helpful strategy.

Qualitative research typically attracts more female participants, with young men in particular known to be a difficult population to recruit in health research. Despite this, I was able to recruit a relatively even split of male and female participants (20 females, 12 males, 5:3 ratio). Although the aim of qualitative research is not to be generalisable, it is a strength of the current study to have such a high contribution from young men, deepening our understanding of male survivors' experiences and psychosocial needs. This is all important for the future development of psychosocial resources, ensuring that they are informed by and applicable for as many survivors as possible. Despite this, I am aware that this study was based in the UK only, and although included some participants from other countries and cultures, all of them were treated in England. It would be interesting to replicate this study across cultures, particularly in non-Western areas, to assess whether the experiences discussed in the current study are shared worldwide.

It is of note that despite aiming to recruit participants from both of the UK Rb treatment centres, none were enlisted from the Royal London Hospital/Great Ormond Street Hospital. The reasons for this were multi-faceted; firstly, there was a significant delay to the additional ethical approval process required by the trusts (alongside the standard approval granted in May 2022). Due to administrative staff being on summer break, the internal approvals process did not begin until September 2022. I was then required to present to the haematology/oncology committee for approval, which took place on 26th September 2022. Once approved to proceed to the next stage of the trust's processes, I was required to complete a Study Assessment Form for internal review; this was finally granted on 4th November 2023. Unfortunately, there was then a further delay within the clinical team, meaning that we did not get to discuss study recruitment until 22nd December 2022. Despite all of the agreements being in place, the clinical team were not able to respond until 15th

February 2023, where they confirmed that had sent out one batch of recruitment invites and only one person had expressed interest (who later did not consent and proceed to participate). At this stage recruitment had already finished at the other sites and the recruitment target had been exceeded. It was therefore decided to close the recruitment period for the London site as the analysis stage needed to begin. Although this was disappointing, I could understand that recruiting for the study placed an additional workload on to a small, specialist team. I also assessed that I had sufficiently captured the demographic of participants treated in London through the social media recruitment, meaning that this would not be detrimental to my sampling strategy.

The majority of the participants were recruited via CHECT’s social media, meaning that it is likely that the sample had self-selection bias. For participants to have become aware of the study, they, or someone that they know (several teenage participants informed me that their parents had seen the study advertised online) would have been following CHECT’s social media channels. It is therefore assumed that these individuals support the work of the charity, and have an inherent, vested interest in supporting CHECT. This was reflected in the data, whereby many participants spoke about their desire to ‘give back’ to the charity and the wider Rb community. It is also plausible that they saw the opportunity as a cathartic experience to process their survivorship journey.

Throughout the study, I encountered two ethical challenges in relation to a sub-section of participants. Firstly, when collecting demographic characteristic data prior to conducting the interviews/focus groups, I became aware of some participants self-reporting diagnoses that could not be biologically correct (e.g. an individual with bilateral disease cannot have non-heritable Rb). Upon noticing this irregularity, I raised it with my supervisory team and also had discussions with the medical team at Birmingham Children’s Hospital, without providing participant details. It was decided that the best course of action would be for me to notify the relevant individuals by email (see appendix A20). This email was sent to young adults directly if over the age of 16 years and copied in parents if the participant was between 13-16 years of age. After conducting a risk assessment with my supervisors, we deemed it appropriate to carry on with the planned interview/focus group prior to informing the relevant individual. This was because of the risk of this information impacting the data, and the likelihood that informing them after their participation would not cause any undue harm. It would have been inappropriate and unethical for me to inform the young people’s treatment centre of this error, due to participant confidentiality. For this reason, I encouraged the participants to contact their Rb team directly to clarify their diagnosis. I also had a separate conversation with the medical teams at London and Birmingham to inform them of this issue, and that individuals may be in touch. To ensure transparency and mitigate any potential distress, I worded the email carefully and sensitively, emphasising that the purpose was to support their understanding rather than challenge their lived experience. I also provided resources for further guidance and encouraged them to seek support from CHECT and other signposting services if they felt that this would be helpful. In follow-up discussions with my supervisors, we reflected on the ethical complexities of managing incidental findings in qualitative research, particularly when working with young people. This experience reinforced the importance of balancing ethical responsibility with participant autonomy, ensuring that safeguarding measures are in place while respecting confidentiality and the integrity of a study.

The second ethical challenge involved only one participant, who disclosed feelings of helplessness and suicidal ideation during the course of their interview. As I am a psychologist, I am well versed in managing such situations in a clinical context. However, I was aware of my role as a researcher in this situation and drew upon the distress protocol set out at the study's conception. At the time of disclosure, I expressed my concern for the participant and sought permission to raise this with their healthcare professional. The participant provided consent for me to do this, and I had a phone call with their general practitioner, outlining their disclosure as part of a research study. Once I knew that the participant was being supported by a clinician, I also provided the standard study debrief and ensured that they had access to adequate mental health signposting resources as well as the CHECT support details. In subsequent discussions with my supervisory team, we reflected on the challenges of navigating researcher-participant boundaries when managing distressing disclosures. This experience reinforced the importance of having a clear ethical framework in place, including well-defined distress protocols and escalation procedures, to protect both participants and researchers. It also highlighted the value of researcher training in handling sensitive conversations with empathy while maintaining appropriate professional boundaries.

Existing studies often find a correlation between volunteering for research participation and higher levels of education. Applying this to the current study, it is plausible that the participants in this study are more highly educated and may therefore be more inclined to volunteer their time for the purpose of research. Furthermore, they could hold a heightened awareness of scientific processes and how their contribution could significantly enhance knowledge about the psychosocial impact of Rb. It could also be that my study materials were better tailored to individuals with higher levels of education, with an assumed level of education required to read and understand the participant information sheets and consent forms. Despite this, as the youngest participants in the study were 13 years old, all materials were designed for this level of education, cognitive ability and maturity, hopefully providing an inclusive opportunity for anyone of varied educational ability to take part. It is also a strength of this study that I provided video information alternatives for any individual who wished to hear and/or see the research team discuss the study aims and procedure for taking part. This was set up primarily for the benefit of individuals who have visual impairment but was offered to all participants regardless of ability. Lastly, I did not explicitly request details of participants' educational attainment, meaning that I cannot comment on whether this phenomenon is true in this instance.

In terms of thematic development, my analysis led me to organise these around the concept of time, considering the impact of past experiences on present day thoughts, feelings, and behaviours, and the cumulative effect of both of these periods on the future and the need for support. It is possible that this inductive analysis occurred, in part, due to the structure of the topic guides used to structure interview and focus group discussions. When reviewing the guides, the conversation was opened by asking individuals about themselves and their experiences with Rb. Although this was left as an open question, prompts such as *'what do you understand about Rb?'* and *'can you tell me what you remember about this time and your treatment?'* were added to probe participants if necessary. Although these were not routinely asked to all participants, it may be that these questions could have encouraged participants to discuss their experiences in terms of the past, present, and future.

Lastly, as has become increasingly commonplace since the COVID-19 pandemic, all of the data collection took place over the remote videoconferencing software, Zoom. Whilst this enabled us to engage with individuals spanning a huge geographical distance (including abroad), there were some limitations specific to focus groups that were unique to this method. The aim of a focus group discussion is to encourage group interaction, a method which was felt would be useful when engaging teenagers in a sensitive conversation. It was hoped that this group approach would provide peer support and allow individuals to relate to one another, a process which would not be possible through individual interviews. Whilst I was pleased that the teenage participants engaged well with each other, there were a couple of issues which were more difficult to address. As discussed in the analysis, some of the participants who took part in the study had visual impairments, ranging from minor sight loss to complete blindness in one case. For some individuals impacted in this way, conversing remotely was sometimes difficult, particularly in instances where home WIFI delayed audio responses or where social cues and non-verbal interactions (such as nodding) were missed. To address this, all participants were reminded at the start of the group session to communicate via speech where possible to aid inclusion. At times when individuals did communicate non-verbally, such as through a nod or shake of the head, or through writing in the Zoom chat box, facilitators would name this e.g. *"I can see NAME is shaking their head* *there”* or *“NAME has written TOPIC in the chat box, does anyone have the same opinion?”.* Although a rare occurrence, there was a couple of occasions when an individual could not or did not want to have their camera turned on. This posed a challenge to group cohesiveness and made it difficult for us as facilitators to ensure that they were engaged in the conversation and included by other group members.

Unfortunately throughout the recruitment process for this study, I encountered a difficulty with ‘bots’ (malicious automated software) and fraudulent or imposter participants (people paid by third parties to undermine studies or for personal gain), a growing issue in qualitative health research (Quach *et al.,* 2013; Hydock, 2018; Glazer *et al.,* 2021; Jones *et al.,* 2021; Ridge *et al.,* 2023). The recruitment which took place online via social media channels used two versions of a recruitment poster (see appendix A11 and A12), one of which utilising a QR code that could be scanned for access to an information video about the study. This latter detail is important as it is thought that this addition increased bot activity hugely. I first noticed an issue when the steady flow of emails expressing interest became over 150 emails in less than 24 hours. It was initially not clear which emails were genuine, although I noticed that there were several similarities amongst emails, such as lack of salutations, vague or no information specific to the study or to Rb, medical details which are impossible to be true (e.g. claims of being diagnosed with Rb at the age of 24) and the mention of being treated at hospitals that do not exist. As to not falsely rule out any genuine participants, I spoke with my supervisory team to clarify my thoughts; this also involved cross-checking alleged hospital details with worldwide colleagues and engaging in further correspondence with suspicious participants. In addition, I posted on twitter to see if any other researchers were having similar issues. Within 24 hours I had got together a small group of researchers which led to a publication in the Archives of Disease in Childhood (O’Donnell *et al.*, 2023). This has led to so many opportunities that have allowed me to up skill myself, being invited to collaborate on international publications, peer review relevant articles for major journals, and network with colleagues whom I would never have come across otherwise. The ability to throw myself into novel situations means that I am constantly developing myself as a researcher, and I am pleased that some good has come out of a difficult issue encountered during my PhD. It is also positive that through my work, I continue to contribute towards the development of enhanced screening procedures to prevent this concerning issue, which is both unethical and has the potential to undermine the integrity of health research.

3.19 REFLEXIVITY

In chapter two I have extensively explored my influences on the research area but wanted to comment on a few features specific to the current study, notably the influence of my gender, my professional background, and the political and social context of the study.

GENDER INFLUENCE

A small proportion of individuals in this study expressed that their experience of Rb no longer bothers them; it is of note that the majority of the quotes on this topic were from men. This poses the question about the role gender has in coping, and I question my influence in this, as a woman, and how that could have fed into this narrative. For this reason, I made sure to carefully consider all participant transcripts, re-reading them at different time points to ensure that this was not influencing my analysis.

It will also be crucial to understand in terms of developing an accessible intervention. Exploring the potential gender-related differences in coping and outcomes can provide valuable insights into tailoring support that is sensitive to the diverse needs of individuals living beyond Rb. Moreover, by acknowledging the potential impact of my own gender and the bias this may have had on this research process, I can provide a more balanced and comprehensive understanding of the experiences of both the men and women in this study, ultimately leading to more effective interventions and support strategies for as many individuals as possible.

PROFESSIONAL INFLUENCE

Throughout the study I became aware of some individuals having negative feelings towards the lack of psychological support offered as part of their long-term care. I was very aware of my role as a psychologist and a researcher and the way in which I might have been perceived by participants during these conversations, both positively and negatively. There were times when by acknowledging participants' wishes for better psychological support, I did not want to be seen as being negative towards the medical teams who work so hard to provide excellent medical care. This was especially true as I had spent extensive periods of time shadowing the Rb clinics at Birmingham and London and had seen the extraordinary care and attention given to families at times of acute distress. I was also mindful that the individuals I was interviewing were treated more than ten, sometimes even twenty years ago, where psychological support was less widespread and possibly less acknowledged. Some participant experiences were at odds of what I had witnessed in current long-term follow-up clinics, where health care professionals took the time to ask about thoughts and feelings and had good knowledge around the potential long-term psychological effects of cancer. At other times I felt that participants could have wanted to please me, given my professional background, by advocating for the role of psychology in Rb care. Additionally, they may have volunteered to take part because they felt it was an important topic to further research. When the majority of participants had positive experiences of or recommended the integration of psychological support into routine care, I was mindful that I needed to remain neutral to their comments as to not influence their conversations. Despite this, on one occasion I had another difficulty, when speaking to a participant who was particularly anti-therapeutic support. In this interview a comment was made about therapists *"being paid to care",* and therefore not really caring at all. In this instance it was challenging for me not to provide the benefits of psychological input and correct his opinion that was so at odds with mine. Thankfully this was a one-off and I was able to remind myself that every individual is entitled to their own opinion and we can hold different experiences and viewpoints.

POLITICAL AND CULTURAL CONTEXT

At the time of these interviews and focus groups taking place (2022-23), the NHS was a complex and evolving environment, more so than when the participants were treated. There are ongoing debates surrounding healthcare funding and resource allocation, and there are continued attempts to provide comprehensive and accessible healthcare services in a notoriously underfunded system. The aftermath of COVID-19 pandemic and the addition of Brexit both meant that the political landscape of the UK was complicated, with a cost-of-living crisis and rising rates of mental health difficulties making it a challenging backdrop for the research study.

The NHS is deeply important to much of British society, with many considering that it is intertwined with our national identity, with any changes or developments met with intense scrutiny and debate. With this in mind, it may have been difficult for participants to both criticise care that they had received (or not, in terms of psychological support), and to contemplate a new model of support for the future. The deep attachment that many have to the NHS can sometimes lead to a reluctance to voice concerns or criticisms, even when it comes to vital aspects of care, such as psychological support for survivors of Rb. It was therefore important for me to recognise this narrative amongst participant transcripts, ensuring that I portrayed a balanced view of their past experiences and future wants and needs.

In terms of disseminating this work, it may also be challenging for other members of the Rb community when reading this study. This is likely to be particularly true for older individuals, some of whom I have got to know through my PPI group. Anecdotally, individuals who were treated for Rb thirty or more years ago can feel resentful for the lack of holistic care that they received, with some feeling that they have been left visually and physically impaired without any support to manage the complex feelings surrounding this. Within both my participant and PPI groups, there was understanding that treatment and therefore survivorship outcomes had changed over time. For example, fewer children diagnosed today with be treated with enucleation than those diagnosed in the past. It is therefore imperative that future research and intervention development accounts for this, contextualising the needs of the individual and adapting to meet them.

3.20 IMPLICATIONS

The implications of the current findings extend significantly to inform and influence NHS policy. Firstly, through providing a comprehensive understanding of the profound psychological and emotional impact that people treated for Rb can experience. This offers insight into the need for integrated, specific psychosocial support within long-term follow up. This would provide valuable contribution to the NHS Long-Term Plan, which set out aims to give patients more options for support, particularly where it is preventative and supporting psychological needs (NHS England, 2023). This also fits in with changes to healthcare systems post COVID-19, with the pandemic encouraging services to plan ahead for non-hospital based follow up (e.g. care in the community) and digital and remote support options. Despite the noted benefits, potential challenges must also be acknowledged. For example, access and equity is a known issue when it comes to remote support. It is difficult to ensure equitable access to support across geographic regions, especially in rural and underserved areas. Furthermore, digital support may not be accessible to all and assumes that individuals have access to sufficient broadband, modern technology (e.g. smart phones, laptops) and if they do, that they have the ability and desire to use these. For survivors of Rb with visual impairment it is also important not to rely on purely visual tools. Through recognising the emotional and behavioural impact that Rb can have, this research can provide recommendations to establish a dedicated care pathway shaped by survivors themselves. The third study of this PhD provides more in-depth analysis into what such care could look like, recognising recommendations made in the current study for peer support, preventative intervention and psychological therapy. Ultimately if this is achieved, the recommendations from this study can contribute towards a more comprehensive and individualised support system, providing coping strategies prior to individuals reaching crisis point and reducing burden on mental health services and medical oncology teams. This is already being done successfully with other aspects of Rb follow-up care, for example genetic counselling (where necessary) and routine eye examinations to monitor physical health changes. These can therefore act as a format that we can follow when it comes to implementing specific, widely available psychosocial care. This is not to say that psychological support is not offered in some services, with some services employing part-time psychologists and/or support workers, all of whom offer excellent care. However it is rare for this support to be full-time, is often ad-hoc, and is usually provided upon request from the young person or their family. My study highlights that it is often difficult to vocalise the need for support, let alone have to explicitly ask for help. I therefore propose that the creation of a routine psychosocial intervention that can be implemented across services in the UK and beyond.

3.21 SUMMARY

Ultimately, the findings of the current study highlight the vast influence of having had Rb. In some cases this can impact the way that you are able to see, or how you look, but often impacting the way you consider yourself and the world around you, frequently resulting in complex feelings that can be devastating. For those with the heritable form of the condition, these concerns can be even greater, regularly considering the impact of your behaviour and life choices on your future self, others, and any future children. For those with prosthetics, which a high proportion of this sample did have, the influence of others, especially if negative, can be hugely detrimental and affect peer and romantic relationships, and sense of identity. These experiences have the potential to vastly influence various aspects of an individuals’ and their family’s lives. This study therefore presents a novel exploration of the interplay between Rb, self-perception, interpersonal dynamics, and wider societal and healthcare context, which I hope can be combined with the wider studies in this PhD to inform a tailored psychosocial intervention for this group of cancer survivors.

CHAPTER 4: PATIENT AND PUBLIC INVOLVEMENT (PPI)

Throughout my PhD and my research career as a whole, it is imperative that I actively work in partnership with individuals with lived experience as much as I can. I am passionate about breaking down the ‘patient vs professional’ barrier and working ‘with’ rather than ‘doing to’. This felt particularly important for this PhD and generating evidence to inform a novel psychoeducation intervention, as individuals who have had Rb can bring an expertise that would be impossible for me to have. Although I discuss how patient and public involvement (PPI) is included in each of my studies in the respective chapters, this chapter will provide a comprehensive overview.

In health science research, the integration of PPI has become increasingly recognised as a crucial element in advancing understanding and developing impactful interventions (Jennings *et al.*, 2018; Biggane, Olsen and Williamson, 2019; Arumugam *et al.*, 2023; Colomer-Lahiguera *et al.*, 2023). The traditional model of research often placed healthcare professionals and researchers in a position of authority, with patients and the public viewed as subjects or recipients of interventions rather than active participants. Individuals with lived experience possess a unique understanding of Rb, its challenges and late effects, as well as the intricacies of navigating the healthcare system that it would be difficult to fully grasp as a researcher or clinician. My work reflects a paradigm shift towards more inclusive and participatory methodologies, and by acknowledging the invaluable expertise that individuals with lived experience bring, I hope that I am fostering a more equitable research environment whilst ensuring that the intervention that will be developed as a result of this PhD is relevant, effective, and sensitive to the needs of Rb survivors.

One of the key benefits of incorporating PPI in my research is the potential to co-create solutions that are evidence-based and contextually relevant. By engaging individuals with lived experience as active partners in the research process, I hope that their voices are both heard and valued in shaping interventions that directly impact their lives and lives of future generations affected by this rare cancer. This collaborative approach enhances the credibility and applicability of these research findings and also aims to foster a sense of ownership and empowerment among the individuals impacted by Rb.

Throughout my PhD and in all of my research moving forward, it is incredibly important to me to work side-by-side with young people with lived experience. As someone who has not had Rb myself, it would be remiss to assume that I know how survivors feel. My stance is always that I hope that by working with individuals who have been through Rb, they can bring their lived experience and combine it with some of my skills as a researcher. By doing this, I hope that I capture the wants, needs, and views of the very people who this research affects most, meaning that when we develop our intervention it will be both acceptable and helpful for them. it is important to note that all of the young people mentioned here consented to the inclusion of their names, acknowledging their contribution as partners in this work.

4.1 STUDY 1

I received a lot of interest in this work and the first academic paper which summarises the findings has now been published in the British Medical Journal Open (O'Donnell *et al.*, 2024). However not everyone wants to or can read an academic paper, especially if they are very young or if they are living with impaired vision or blindness. Therefore it was important to me to disseminate the findings of the study, and the powerful words of the young people who shared their experiences with me, more widely. For this reason, I got a group of Rb survivors together and hired a podcast studio in Birmingham. CHECT's support worker Sarah Turley and I met with six young people, James, Yoadey, Yuri, Tom, Katie, and Kieran (none of whom knew each other previously) to make my research as accessible as possible. Together will some illustrations of young people's experiences that I had commissioned by Natalie Harney (figure 9), the young people recorded the audio for a video summarising the research findings. They also sat down as a group (without Sarah and I) to create a podcast about what it is like to be a young person who had Rb (https://chect.org.uk/chect-tya-podcasting-day/). The group did an excellent job, and hearing their voices explaining the study as well as sharing their experiences was incredibly powerful. It's important to say that none of the young people who created the video and podcast were participants in the research; it is incredibly important to keep participants' identities private, and it is such a privilege to be able to highlight their experiences in order to improve Rb support in the future. The video has been disseminated widely and can be watched by linking the link or using the QR code below: https://www.youtube.com/watch?v=00XhTZMhbEU.

4.2 STUDY 2

Incorporating PPI in the process of writing a systematic review is invaluable but often not seen as a priority. It was important to me to recruit teenage and young adult survivors of Rb to act as lived experience co-researchers. This is because they can offer unique and personal insights that significantly enhance the relevance and impact of my research, ensuring that I am identifying and reporting data that is relevant to them. Their involvement ensured that my systematic review addressed the most pertinent issues in childhood cancer survivorship and support and reflects the actual needs and preferences of those it aims to benefit. This collaborative approach not only empowered my PPI co-researchers by valuing their experiences and perspectives but also enriched the research process by providing a deeper understanding of what psychological support strategies are effective from the standpoint of those who have lived experience.

Throughout the synthesis of the studies, the wealth of information gathered had to be carefully refined to focus on the most critical aspects. This was particularly difficult in that for my review, there was a lot of heterogeneity in the data, particularly in the outcomes reported. I engaged Rb survivors in this process by asking them to rate the importance of various topics. This participatory method ensured that the review did not merely rely on theoretical or clinical perspectives and is grounded in the real-world experiences of survivors. By prioritising my focus based on their input, the study can better identify what works and what does not in existing psychological support systems. Although this review was broader than Rb (due to the lack of specific psychosocial support currently available for this group), I hope that post-doctorally this will be combined with the other studies in this PhD to inform the design of specialised support tool tailored specifically for young Rb survivors.

4.3 STUDY 3

Throughout my three studies, the involvement of my PPI group members has been integral. From the very beginning, PPI members were engaged in developing the research questions, ensuring that they were relevant and meaningful to those directly affected by Rb. Their input helped shape the focus of my studies, aligning the research objectives with the real concerns and priorities of living beyond Rb. This collaborative approach not only enhanced the quality and relevance of the research questions but also fostered a sense of ownership and partnership among the PPI members, many of whom had never met each other or even anyone else effected by Rb before.

For study three, my content analysis, PPI members played a crucial role in the analysis and interpretation of findings. Their lived experiences provided valuable context and depth to the qualitative data, helping to identify nuances and insights that might have been overlooked by me as a researcher without personal experience of Rb. This involvement ensured that my content analysis was grounded in the realities of those who have been through such challenging experiences, leading to more accurate and empathetic interpretations of the data. Although I completed the initial analysis, their contributions were essential in recognising subtle yet significant aspects of the data that a purely academic analysis might miss. For example, I might place greater focus on clinical outcomes or treatment pathways, while overlooking the emotional impact on family dynamics or the long-term psychological effects of living with a visual impairment. Through their lived experiences, PPI members might highlight how small moments, such as their experience of dealing with social stigma, are just as important to consider. By sharing their personal insights and reflections, PPI members helped to contextualise the data within the broader framework of their lived experiences, ensuring that the themes identified were relevant and resonant with the experiences of other Rb survivors.

Finally, as with all of the studies in this PhD, PPI members were actively involved in the writing, publishing, and dissemination of the research findings. Their perspectives helped to develop the narrative and ensure that the findings were communicated in a way that was accessible and meaningful to a broader audience, including other survivors, healthcare professionals, and policy makers. By participating in the dissemination process, PPI members helped bridge the gap between research and practice, advocating for changes that could directly benefit their community. Their contributions were instrumental in ensuring that the research not only advanced academic knowledge but also had a tangible, positive impact on the lives of those affected by Rb.

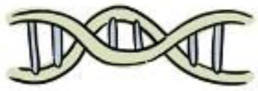
Understanding how having had Retinoblastoma (Rb) as a child affects how you think and how you feel as you grow up

Retino means from the retina, in the eye,

blast looks similar to cells in early development,

and oma refers to a group of cells or a tumour. Essentially, Rb is a rare eye cancer

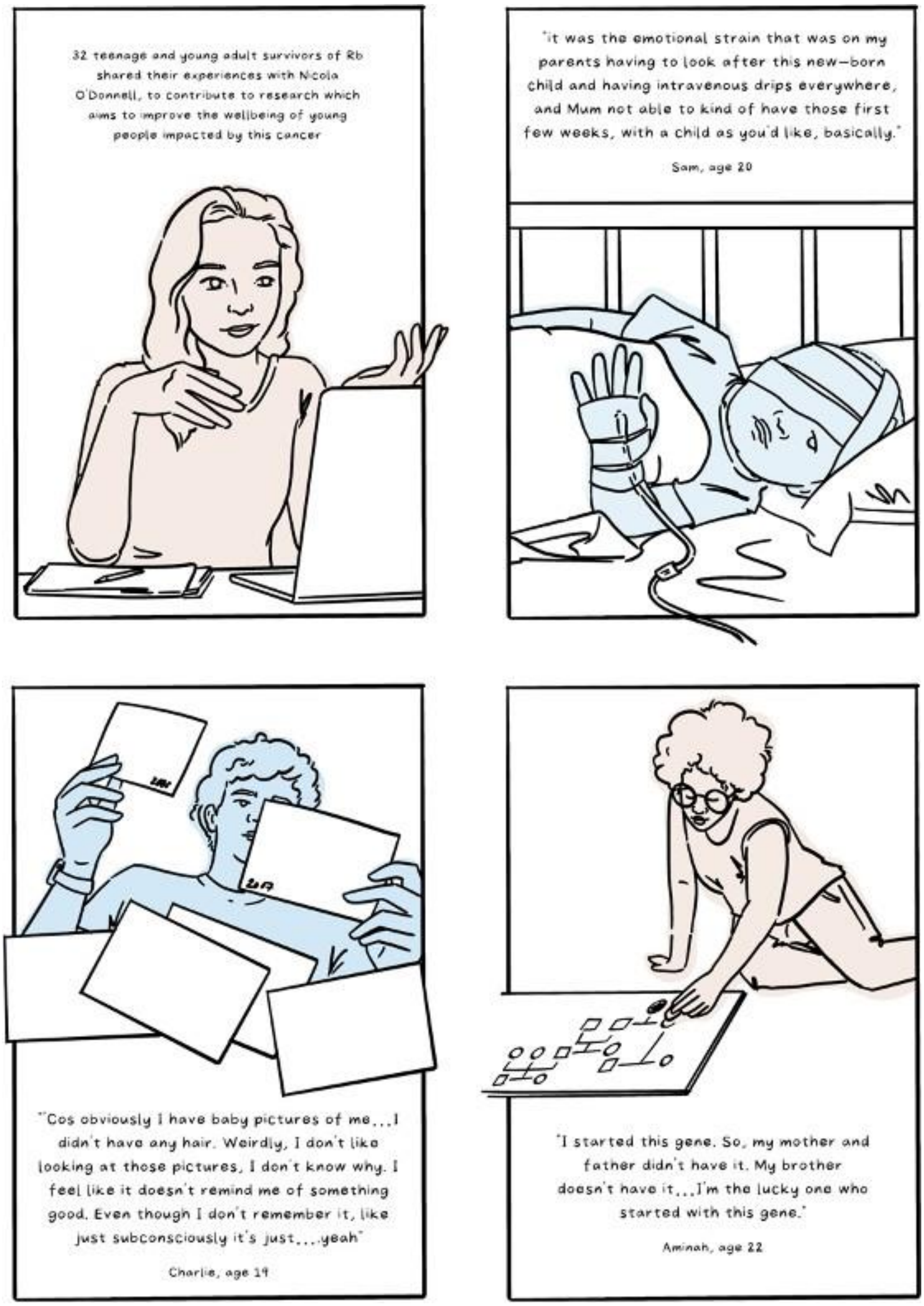
It is diagnosed in approx. 50 children a year, or one child a week, in the UK.

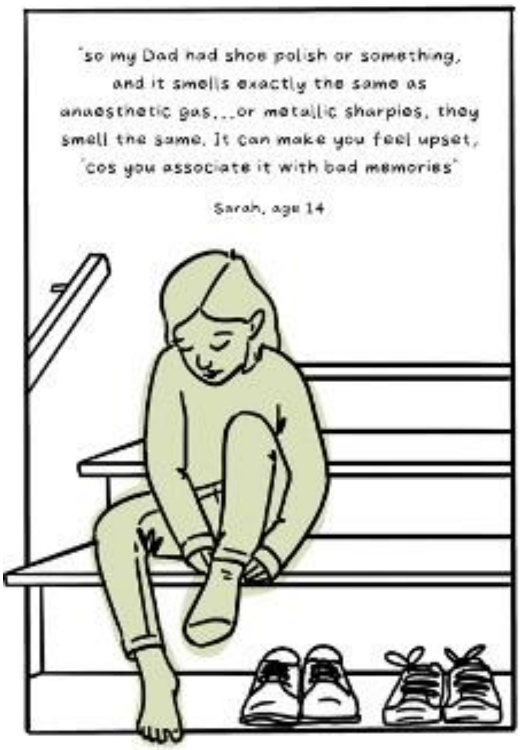


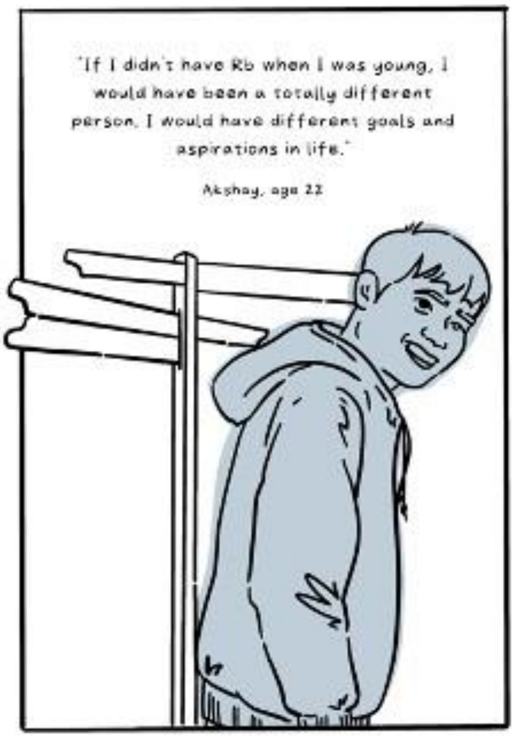
It's most commonly diagnosed in babies and young children and there are two forms of the condition, one is heritable (genetic) and the other is not.

It affects some people in one eye (unilateral) and some in both (bilateral). There is a very high survival rate in the West (98%) but children often undergo a range of intensive cancer treatments and sometimes enucleation (surgical removal of the eye or eyes).

We know that the impact of any cancer and/or treatment can have both long- and short-term effects on psychological and social wellbeing, some of which can last throughout a lifetime. For this group, it can also mean reduced vision, facial changes and identity related distress.

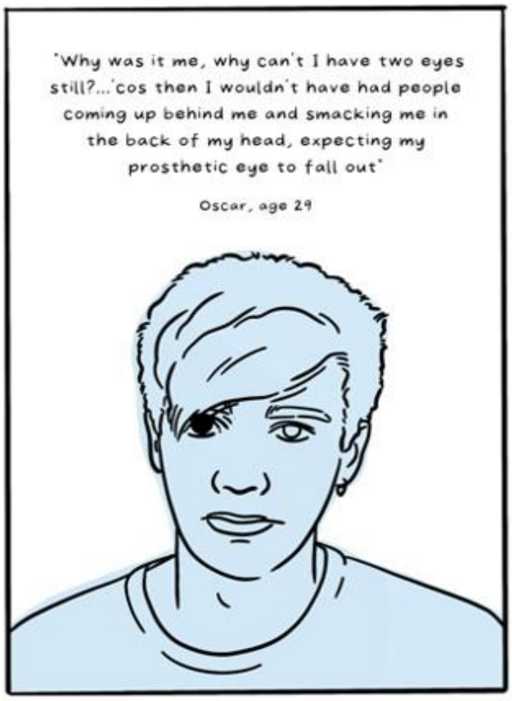

32 teenage and young adult survivors of Rb shared their experiences with Nicola O'Donnell, to contribute to research which aims to improve the wellbeing of young people impacted by this cancer
"it was the emotional strain that was on my parents having to look after this new—born child and having intravenous drips everywhere, and Mum not able to kind of have those first few weeks, with a child as you'd like, basically."
Sam, age 20
2017
"'Cos obviously I have baby pictures of me...I didn't have any hair. Weirdly, I don't like looking at those pictures, I don't know why. I feel like it doesn't remind me of something good. Even though I don't remember it, like just subconsciously it's just....yeah"
Charlie, age 19
"I started this gene. So, my mother and father didn't have it. My brother doesn't have it...I'm the lucky one who started with this gene."
Aminah, age 22


"so my Dad had shoe polish or something, and it smells exactly the same as anaesthetic gas…or metallic sharpies, they smell the same. It can make you feel upset, 'cos you associate it with bad memories"
Sarah, age 14


"If I didn't have Rb when I was young, I would have been a totally different person. I would have different goals and aspirations in life."
Akshay, age 22


"I guess when you're that age, the only thing you want to be is normal… it's on your face… like you're looking into people's eyes the whole time when you talk to them, so it's what you're greeted with. I guess being normal in that respect, it's the most important thing when you're that age."
Harriet, age 21


"Like I do think I could have got help if I actually asked for it. I think my problem was that I probably just suffered a bit in silence."
Will, age 27


"Why was it me, why can't I have two eyes still?...'cos then I wouldn't have had people coming up behind me and smacking me in the back of my head, expecting my prosthetic eye to fall out"
Oscar, age 29


"It's just the fact that when I was younger, other people were like, "oh she can't do this, she can't do that, she won't be able to do this"....you're already putting things in the way before I've started anything"
Carmen, age 29


"I never asked anyone out really at secondary school because I didn't have any confidence, and I felt like I was ugly and I always felt like women didn't like me".
Matt, age 29


"I feel like the fact that I'm blind has also contributed to me having a harder time socialising...not many people talk to me."
Niamh, age 20


"You don't wanna think, 'Oh no, if I have kids, what if they have it as well and they have to go through it?' and things like that."
Leah, Age 16
"Yeah, that's definitely one I was like thinking, of – if – if you – if you were to want to have kids, would you pass it down to your child."
Priya, Age 14
"I'm not really too sure about – if it increases my risk of getting cancer in the future. I think it might do. I'm not really too sure."
Tom, age 20
"It's just a time where you don't feel, you don't even know who you are. You're trying to figure out, you know, what kind of clothes am I supposed to wear, what's my style, what do I want to do. Like, who am I?"
Hannah, age 29
"I'd internalised that, that there's these tumours that could suddenly, you know, go off like a volcano"
Samson, age 20


"I actually survived, you know, and I not only survived, but I can see"
Kaye, age 29
"It doesn't help fitting in when your school work is like, size 40 font and you have a teaching standing next to you the whole time... I know it's necessary to have the teacher there, but it's also really annoying. Sometimes you feel like you're missing out on jokes, and you can't sit next to people you want to"
Rob, age 15
"I came home one day and I was like, "what's a cyclops?" and my parents were like, "it's a mythical creature with one eye". And I was like "oh, that's what this boy calls me at school"
Nicholas, age 25
"I'm gonna find my power in this thing, I'm gonna make this my motivation and I'm gonna be friends with myself. I'm gonna be friends with my illness... if you don't present it as a weakness, people won't take it as a weakness."
Imogen, age 29
N

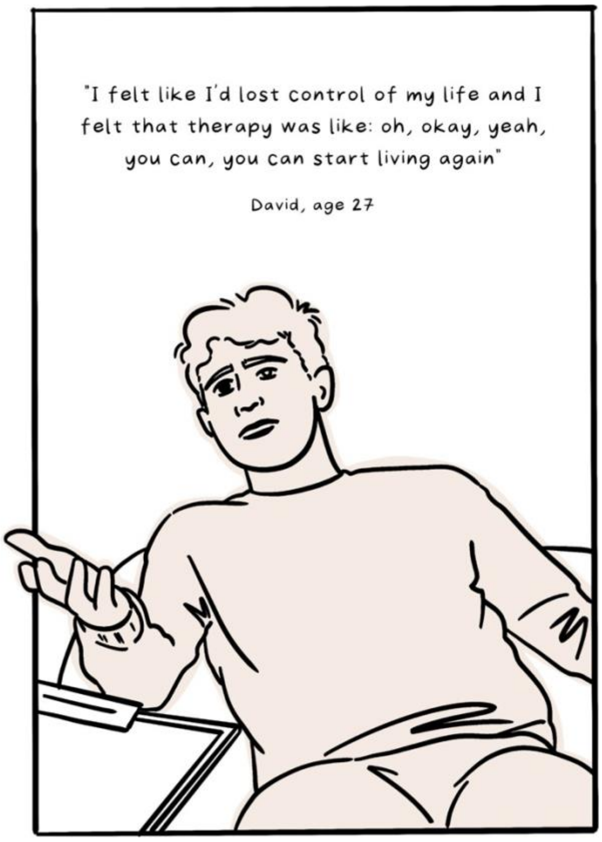


FIGURE 10: ILLUSTRATIONS FROM STUDY ONE, PRODUCED BY NATALIE HARNEY

CHAPTER 5: STUDY 2 – "YOU *JUST NEED SOMEONE TO STEER YOU IN THE RIGHT DIRECTION*" – A SYSTEMATIC REVIEW OF PSYCHOSOCIAL INTERVENTIONS DESIGNED FOR TEENAGE AND YOUNG ADULT SURVIVORS OF CHILDHOOD CANCER.

5.1 CONTEXT AND STUDY TEAM

Study 2 comprises of a quantitative systematic review which I led in collaboration with my PhD supervisors (Professor Bob Phillips, Professor Debra Howell, and Dr Jess Morgan), and researchers in the departments of Cancer Survivorship and Psychology at St Jude Children's Research Hospital in Memphis, Tennessee (USA), the University of Bristol (UK), and Aarhus Universitetshospital, Denmark. These collaborations were formed after I reached out to Dr Victoria Willard, a psychologist at St Jude, who works in a department that is world-renowned in the research and treatment of children with rare cancers such as Rb, as well as being highly skilled in this methodology. It is with sadness that Tori died shortly before the submission of this PhD and the publication of this systematic review. Throughout the review process she was always supportive and helpful, and this work will be published in her memory. Dr Leila Ellis is a medical doctor who became involved after we met at a conference where we discovered our shared interests in this research topic. Dr Pernille Axél Gregersen is a geneticist who I connected with due to our shared research interests and we are in the process of forming an international psychosocial Rb collaboration. I secured funding from the Childhood Eye Cancer Trust (CHECT) which enabled me to visit Pernille and her team in Aarhus in November 2023. I used this time to work with the team, discuss the search terms, and access their databases for articles. As per the objectives of my PhD, I proposed the review question, project managed the study, led all aspects of the review, and was first author on all written work. Leila (LE) acted as second reviewer, and Bob (BP) third reviewer to manage any disagreements between myself and Leila. To ensure that the systematic review was relevant and meaningful to the Rb population, individuals with lived experience were asked to collaborate on this work as project advisors and/or co-authors. This was particularly important given that this study does not focus specifically on Rb (due to a lack of existing interventions available for this group). The opportunity was advertised via the CHECT. Three individuals volunteered to take part: Katie Peller, Ana Perez, and Georga Gorrell. All individuals were compensated with a voucher for their time and are named as co-authors on the published review. Dependent on individual choice and availability, involvement was optional to be either continuous or one-time only, but all three individuals were involved throughout. Using the ‘Cochrane Involving People’ learning resource, I proposed that individuals were involved at the following stages of the review process (table 8). All three individuals were involved from the outset, including being actively involved in determining the most relevant outcome measures to report in the review, as well as providing general oversight of the findings from a survivor’s perspective. All PPI group members will be actively involved in the dissemination of this research.

TABLE 8: PPI INVOLVEMENT IN THE SYSTEMATIC REVIEW PROCESS

Stage of review	Aim of involvement	Proposed involvement
1. Developing the question	Clarifying the review question	Small group discussion
2. Planning the methodology	Clarifying the methods	None
3. Writing and publishing the protocol	Agreeing the protocol content for the review	None
4. Developing the search	Advising on the search strategy terminology	None
5. Running the search	Identifying relevant papers using the agreed search strategy	None
6. Selecting the studies	Providing opinion on whether selected studies meet the study aims	None
7. Collecting the data	Reading selected papers and extracting data	None
8. Assessing the risk of bias	N/A – little evidence of involvement of PPI in this stage	None
9. Analysing the data	Comment on findings	Small group discussion
10. Interpreting the findings	Providing consensus on the above stage and determining the clinical value of findings	Small group discussion
11. Writing the review	Providing feedback on drafts	Sent drafts for comment
12. Publishing and disseminating the review	Contributing to a plan of dissemination and potentially co-presenting findings	Co-authors and co-presentation of dissemination

INTRODUCTION AND RATIONALE

In the third chapter of this thesis, I reported the qualitative experiences of teenagers and young adults (TYA) who have had Rb. In this chapter I examine the systematic review exploring psychosocial interventions designed for TYA survivors of childhood cancer. I aimed to investigate the types of psychosocial interventions that exist for TYA survivors of childhood cancer, whether a specific type of psychosocial intervention provides higher efficacy in improving survivors' mental wellbeing, and whether psychosocial interventions positively influence the wellbeing and psychological health of TYA survivors', as well as considering if there are any possible negative impacts or 'adverse events'. This provides a robust synthesis of quantitative evidence that I can combine with my qualitative findings to propose a co-designed psychoeducation intervention for Rb survivors.

It has been documented that once treated for cancer and off treatment, individuals and their families can be left 'in limbo' (Lopez *et al.*, 2014). This captures the survivorship journey towards normality and life without active treatment, but with an impact on well-being, potential additions of fear of recurrence and social difficulties when reintegrating into 'normal life' (Wakefield *et al.*, 2010). Psychosocial interventions are increasingly used in clinical practice to reduce such difficulties. Despite this, little is known about their feasibility and efficacy, particularly for childhood cancer survivors, who are now TYA (Campo et al., 2017; van Dijk-Lokkart et al., 2016). TYA in cancer settings are classed as a unique group, situated in the middle of healthcare systems aimed at either children or adults (Palmer *et al.*, 2007; James Lind Alliance, 2018). For this reason, the psychological support needs of survivors of childhood cancer have been named as a JLA top 10 priority (Aldiss *et al.*, 2019, 2023; James Lind Alliance, 2022). The psychosocial impact of childhood cancer on this group can be vast, as many experience interrupted development, impacting cognitive and social outcomes (Patterson *et al.*, 2015). Therefore, tailored psychosocial care and interventions must be offered to respond flexibly to the needs of individuals at this life stage (D'Agostino, Penney and Zebrack, 2011). As described earlier in this thesis, psychoeducation interventions involve providing individuals with relevant and up-to-date information about their health to support them to live with and/or beyond a diagnosis.

Existing reviews highlight many possible long-term psychological consequences of childhood cancer, including depression, anxiety, behavioural difficulties, drug misuse and body image concerns (Friend *et al.*, 2018; Nicklin *et al.*, 2021, 2022; Hamilton-Smith, 2022), issues that were also highlighted in my first study. A 2023 review provided an overview of specific needs, focusing on identity, mental wellbeing, autonomy, relationships, and navigating the future (Neylon *et al.*, 2023). Furthermore, it is documented that these needs change over the course of adolescence and young adulthood, with TYA survivors experiencing many decades of survivorship in comparison to the average adult diagnosed with cancer and therefore needing long-term, flexible support. For these reasons, practical, evidence-based and effective psychosocial support is warranted and wanted (Pugh et al., 2020; Pugh et al., 2016; Walker et al., 2016). Despite this widespread view, many survivors and practitioners feel that there is a lack of targeted psychosocial support available. Having said this, it is worth noting that not all survivors will experience psychosocial challenges that might be recognised as needing such an intervention. For these reasons, we must consider how we help anyone who would like support, not just those who are presenting in a particular way or meeting certain ‘thresholds’. Researchers also call for RCTs to evaluate the benefit of such interventions, as well as considering feasibility, factors which prevent engagement, and the ‘cost versus benefit’ for the mental wellness of survivors (Pugh et al., 2016; Sansom-Daly et al., 2021; Walker et al., 2016). This is what this review aimed to focus on.

Mental wellness can be defined in several ways, with the World Health Organisation (WHO) referring to an individual realising their own ability, being able to cope with life stresses, and contributing to their work and community (WHO, 2021). The Faculty of Public Health expands upon this to include the capacity to form positive relationships with others, experience contentment and joy, have confidence, and take responsibility for oneself (Schramme, 2023). For the purpose of this review, psychosocial well-being and mental health are considered unique to each individual, encompassing the above definitions. In this context, the current systematic review aims to bridge existing gaps in the literature by synthesising and critically evaluating psychosocial interventions tailored to TYA survivors of childhood cancer. By considering the feasibility and efficacy of these interventions, this review seeks to provide evidence-based insights that inform clinical practice and contribute to the development of future targeted interventions.

5.2 METHODOLOGY

A systematic review is a comprehensive and structured method to synthesise existing scientific evidence on a specific research question or topic. Considered the ‘gold standard’ approach (Munn *et al.*, 2018), the process involves a rigorous literature search and systematic assessment of relevant studies. Following a predefined protocol, a systematic review, such as the one conducted for this thesis, aims to provide a clear summary of the current state of knowledge in a particular field.

Often conducted in conjunction with systematic reviews are meta-analyses; a statistical technique which assesses the strength and consistency of the presented studies (Pigott and Polanin, 2020). In the current review, a meta-analysis was not used due to the vast array of primary outcome measures used across the included studies.

5.3 AIMS AND OBJECTIVES

This systematic review aimed to explore psychosocial interventions designed for TYA survivors of childhood cancer. Specifically, it aimed to answer the following questions:

- What types of psychosocial interventions have been trialled for TYA survivors of childhood cancer?
- Is there a psychosocial intervention that provides higher efficacy in improving survivors' mental well-being?
- Do psychosocial interventions positively influence the well-being and psychological health of TYA survivors, and are there any possible negative impacts or 'adverse events'?

5.4 METHODS

A protocol was produced and registered prospectively on PROSPERO (CRD42023422933) (appendix B1). This review is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement and checklist (Page *et al.*, 2021) (see Appendix B2).

SEARCHES

The literature was searched extensively and systematically between 15th July and 11th August 2023, considering studies which evaluated any intervention which targeted TYA aged 13-39 post-treatment survivors of any type of cancer. This age range was determined to incorporate both the UK and USA definitions of ‘TYA’, as per National Cancer Institute (National Cancer Institute, 2024), Cancer Research UK (Cancer Research UK, 2023), and James Lind Alliance (James Lind Alliance, 2024). I met with a health sciences librarian at the University of York on two occasions to refine the search terms. We searched for studies published from any year in the databases of MEDLINE ALL, PsycINFO, Scopus, the Cochrane Library, CINAHL (EBSCO), British Nursing Database, PsycARTICLES, and EMBASE. Searches of PROSPERO and clinical trial registries such as ‘clinicaltrials.gov’ and ‘UK Clinical Trials Gateway’ were also conducted to identify unpublished or ongoing reviews and studies on similar topics. Forward and backward citation searches were then performed. Owing to time and resource constraints, only articles written in English were included. There were no restrictions on geographical location and studies published in any year were considered. Full database searches can be found in Appendix B3.

Search terms were exploded and chosen a priori through a breakdown of the research question. Terms that relate to the same concept were combined using the Boolean operator ‘OR’. Concepts were separated using the operator ‘AND’. Terms were searched in titles, abstracts, and keywords. Search terms were first devised for MEDLINE ALL and adapted for subsequent databases.

5.5 SELECTION PROCESS

STUDY SELECTION

As lead reviewer, I conducted all database searches (appendix B3) and imported these into Covidence reference management software (Veritas Health Innovation, 2023). LE replicated these searches independently to check for reliability. Any duplicate or irrelevant articles were identified, noted, and removed. I screened all remaining texts by title and abstract for inclusion suitability. The full text of all records deemed appropriate was retrieved and reviewed against inclusion criteria. Reference lists of all relevant articles were also searched. A second reviewer (LE) independently screened a sample of 10% titles and abstracts. Of the 847 records screened by both reviewers, there was 0.92 proportionate agreement, 0.77 random agreement probability, and a Cohen’s Kappa of 0.64 showing good levels of agreement. For the papers where there were conflicts, consensus discussion revealed that there was differing understanding of the ‘maybe’ function on Covidence, and agreement was made. There was no need for any papers to be referred to a third reviewer.

All full texts were considered for eligibility by two reviewers (LE and I), with both independently screening 100% of full texts. Any disagreements over inclusion were managed using consensus discussion (n=28) and through a third reviewer (BP, 1 study referred). For the papers where there were conflicts, consensus discussion revealed that many papers had been marked ‘conflict’ where both reviewers had excluded, but for differing reasons, meaning agreement was made. Of the 277 papers screened at full text by both reviewers, there was a 0.90 proportionate agreement probability, and a Cohen’s Kappa of 0.68 showed good levels of agreement.

5.6 INCLUSION AND EXCLUSION CRITERIA

Studies were included if they met the inclusion criteria shown in figure 11 and PICO criteria below.

Inclusion criteria:

- Studies that evaluate any psychosocial intervention targeting adolescents aged 13-39 post-treatment of any type of cancer (from day after completion of treatment onwards)
- Interventions that target the family as a whole if survivor well-being is a primary outcome
- All intervention types e.g. psychological, social, behavioural, educational if aimed at childhood cancer survivors
- Studies that consider long-term conditions as a whole if they meet other criteria and that oncology data can be analysed separately
- Studies with RCT design including mixed-methods studies

Exclusion criteria:

- Studies that are not published in English
- Interventions focused on individuals receiving palliative care

FIGURE 11: INCLUSION AND EXCLUSION CRITERIA

5.7 STUDY DESIGN

It was decided to include RCT only, to review interventions that have undergone the highest level of testing and to reduce bias (Burns et al., 2011). The PICO criteria for inclusion in the review are below:

PARTICIPANTS/POPULATION

Survivors of any type of childhood cancer who were aged 13-39 at the time of the study. This included survivors of central nervous system/brain tumours, of which I anticipated specific interventions will have been conducted. Post-treatment survivors refers to young people who are in complete remission (including those in remission post-treatment for relapse). This is defined as all signs and symptoms of cancer being completely absent (National Cancer Institute, 2019). In the UK, TYA are referred to as anyone aged between 13-24 years (James Lind Alliance, 2018; Cancer Research UK, 2023) and in the USA is defined as individuals aged between 15-39 (National Cancer Institute, 2023). If studies also included participants outside of these ranges, they would be included if the data was analysed separately. Additionally, if wider age ranges were used, if the majority of participants were in this study's defined age range, it was included.

INTERVENTION(S), EXPOSURE(S)

All interventions which aim to improve the wellbeing of childhood cancer survivors who are now teenagers or young adults. All intervention types (e.g. psychological, social, behavioural, educational if aimed at childhood cancer survivors) were considered. The intervention could include survivor-only interventions or whole family interventions, but survivor wellbeing must have been a primary outcome.

COMPARATOR(S)/CONTROL

Any, including usual clinical care and wait list controls.

MAIN OUTCOME(S)

Childhood cancer survivor wellbeing as measured by quality of life questionnaires. All study outcome measures can be found in the table in appendix B6.

MEASURES OF EFFECT

Standardised mean difference. Odds ratio and relative risk were also considered.

ADDITIONAL OUTCOME(S)

Other validated measures of psychological wellbeing e.g. Warwick-Edinburgh Mental Well-being Scale (WEMWBS).

5.8 DATA EXTRACTION AND QUALITY ASSESSMENT (RISK OF BIAS)

Data were extracted by me and independently checked by a second reviewer. Information was extracted from the selected studies and summarised in a data extraction table (Appendix B4). This was completed using Microsoft Word before being exported to Excel.

Risk of bias was assessed using version 2 of the Cochrane risk of bias for randomised trials (RoB 2) (Sterne *et al.*, 2019). This tool provides a judgement of 'low risk' to 'high risk' and determines whether there are 'some concerns' about specific bias across five domains: randomisation process, deviations from intended interventions, missing outcome data, measurement of the outcome, and selection of the reported result.

5.9 RIGOUR

Rigour was ensured by adhering to a comprehensive search strategy set out at the beginning of the research process. Multiple databases were searched using comprehensive search terms to gather a diverse range of relevant studies. To maintain high levels of interpretative validity, at least two reviewers (NOD and LE) were involved in the screening, data extraction, and risk of bias assessment, with a third reviewer (RSP) considering any studies where a consensus could not be made between reviewers one and two. To ensure that findings were transferable, a study characteristics table provided full context of the studies to enable readers to comprehensively assess the applicability of the findings. Through doing this, readers are able to evaluate the relevance of the study results to their specific contexts, enhancing the potential transferability of the findings beyond the original study settings.

5.10 RESULTS

STUDY SELECTION

A total of 11,952 records were identified through the initial database search. After removing duplicates, 8468 articles remained for title and abstract screening. Following this screening, 277 articles were selected for full-text review. Ultimately, 15 studies met inclusion criteria and were included in the final analysis. Of the 262 studies excluded at full text, some used study design other than RCT (n=180), used outcome measures that were not validated measures of QoL or psychological well-being (n=32), was a protocol or did not include study results (n=30), did not include TYA cancer survivors (n=16), used an inappropriate intervention (n=2), had an older adult population aged >39 years (n=1), or was an older version of an updated and included study (n=1). The study selection process is summarised in the PRISMA flow diagram (figure 11).

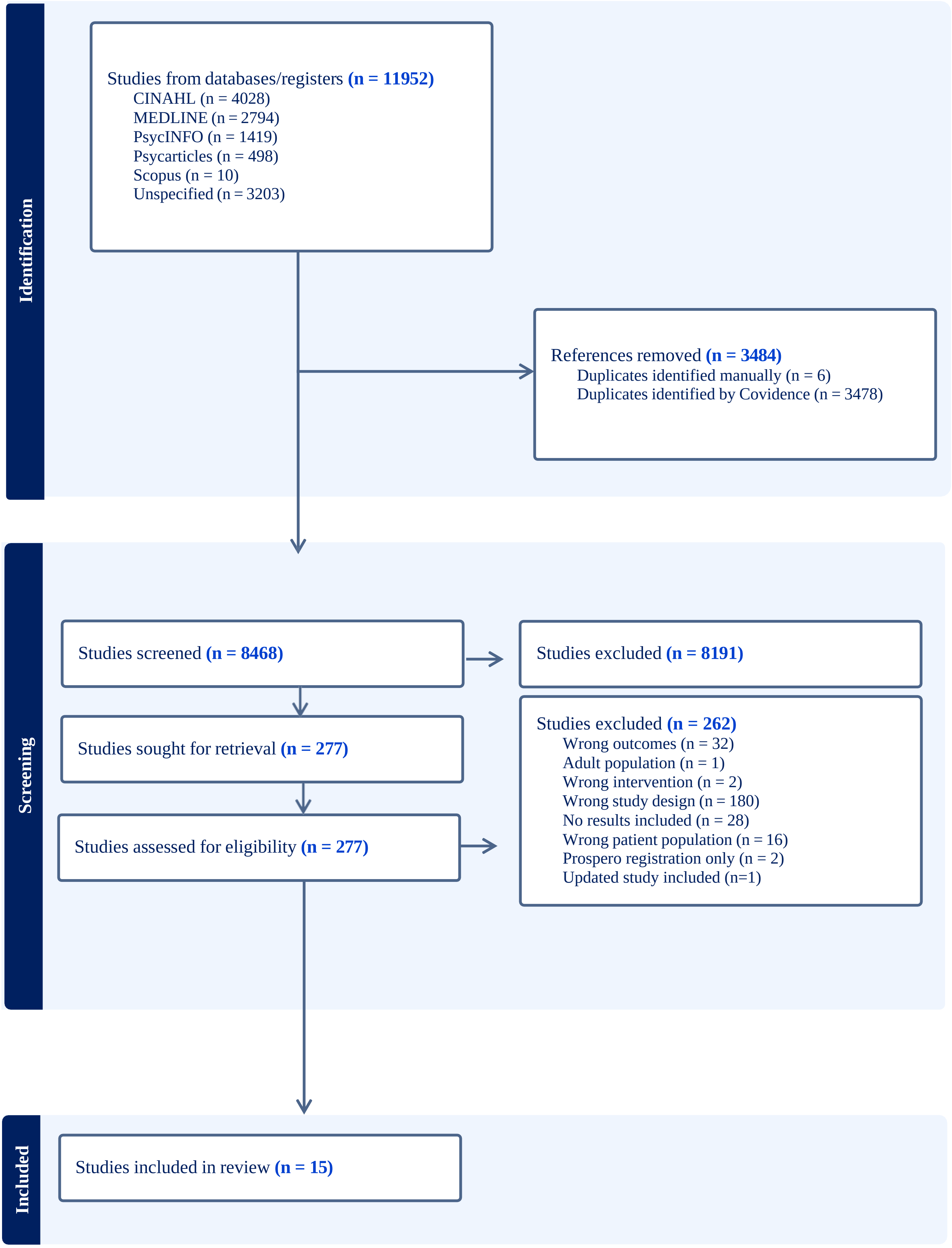
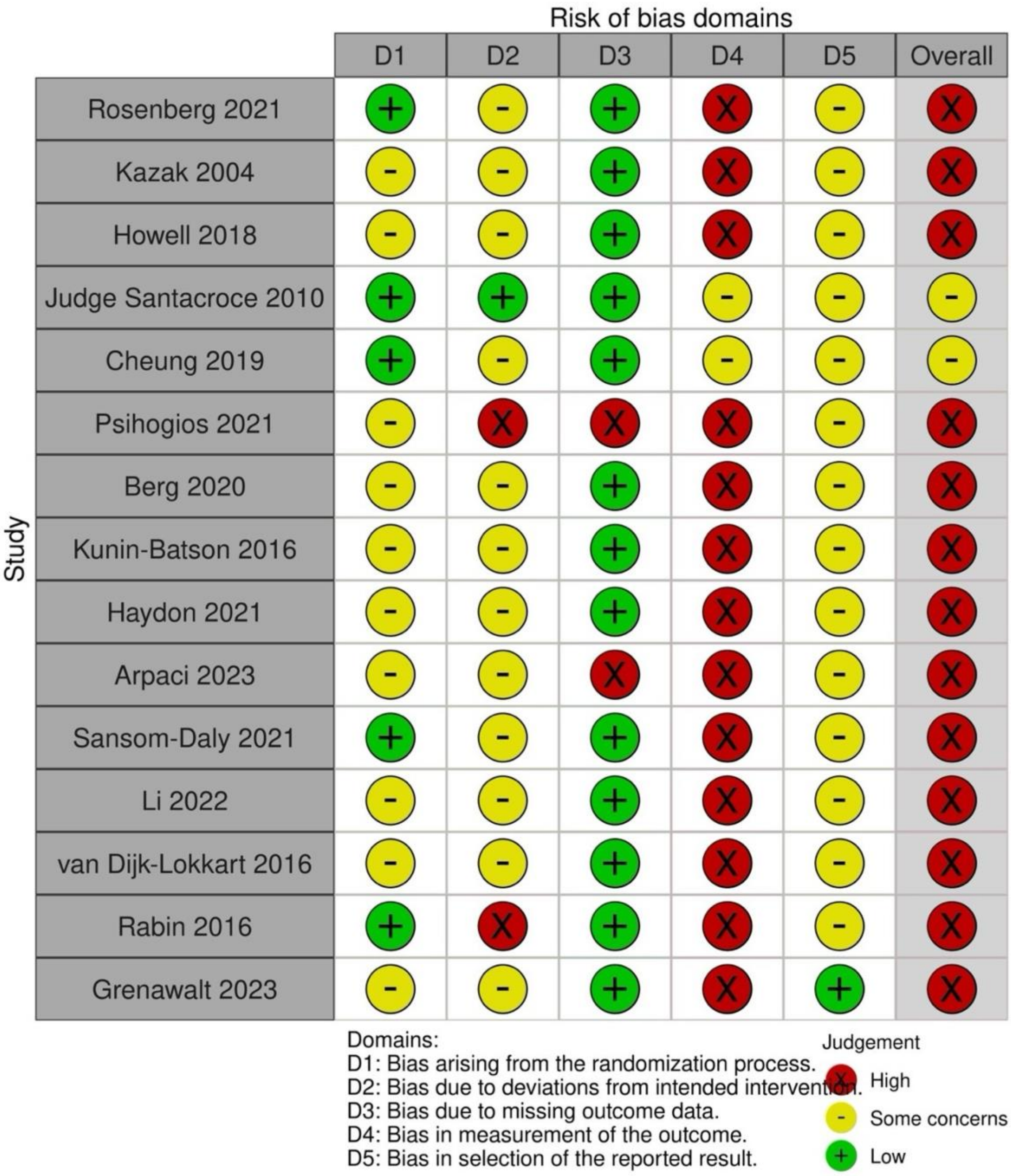


FIGURE 11: PRISMA FLOW DIAGRAM



Risk of bias domains

Study	D1	D2	D3	D4	D5	Overall
Rosenberg 2021	+	-	+	X	-	X
Kazak 2004	-	-	+	X	-	X
Howell 2018	-	-	+	X	-	X
Judge Santacroce 2010	+	+	+	-	-	-
Cheung 2019	+	-	+	-	-	-
Psihogios 2021	-	X	X	X	-	X
Berg 2020	-	-	+	X	-	X
Kunin-Batson 2016	-	-	+	X	-	X
Haydon 2021	-	-	+	X	-	X
Arpaci 2023	-	-	X	X	-	X
Sansom-Daly 2021	+	-	+	X	-	X
Li 2022	-	-	+	X	-	X
van Dijk-Lokkart 2016	-	-	+	X	-	X
Rabin 2016	+	X	+	X	-	X
Grenawalt 2023	-	-	+	X	+	X

Domains:
D1: Bias arising from the randomization process.
D2: Bias due to deviations from intended intervention.
D3: Bias due to missing outcome data.
D4: Bias in measurement of the outcome.
D5: Bias in selection of the reported result.

Judgement
X High
- Some concerns
+ Low

FIGURE 12: RISK OF BIAS ASSESSMENT RESULTS USING ROB2

5.11 Search Results

STUDY CHARACTERISTICS

The table in appendix B4 provides an overview of the characteristics of the studies included. Fifteen RCTs had sample sizes ranging from 21 to 253. Studies were published between 2004 and 2023, encompassing diverse populations and geographic locations; ten in the United States of America (USA) (Berg et al., 2020; Grenawalt et al., 2023; Haydon, 2021; Howell et al., 2018; Kazak et al., 2004; Kunin-Batson, Steele, Mertens, & Neglia, 2016; Psihogios et al., 2021; Rabin et al., 2016; Rosenberg et al., 2021), two in China (Cheung et al., 2019; Li et al., 2022), one in Turkey (Arpaci, Altay and Copur, 2023), one in Australia (Sansom-Daly et al., 2021), , and one in the Netherlands (van Dijk-Lokkart et al., 2016). Below is a brief summary of the key characteristics of each study.

Arpaci's (2023) study involved 62 participants aged 12-18 years who had completed treatment for leukaemia. The inclusion criteria required patients to have received chemotherapy and/or radiotherapy, be at least 2 years post-treatment, and be proficient in using technology and speaking Turkish. Exclusion criteria included treatment with stem cell transplantation, relapse, physical or mental disability, or severe psychiatric problems. The intervention was a Technology-Based Psychosocial Education and Counselling Programme delivered via telephone and website, with outcomes measured using the Pediatric Quality of Life Inventory (PedsQL 4.0) (Varni, Seid and Cheryl, 1999), Self-Efficacy Questionnaire for Children (SEQ-C) (Muris, 2001), and KIDCOPE (Spirito, 1996).

Berg's (2020) research included 56 participants aged 18-40 years, within 2 years of cancer treatment completion. Participants had to be English-speaking and able to use a smartphone. Exclusion criteria were cancer recurrence, cancers of the central nervous system, alcohol or drug dependency, psychosis, bipolar disorder, major depressive disorder, or receiving hospice care. The AWAKE intervention was delivered via an app-based platform with daily mood and health behaviour monitoring, with outcomes assessed using the Adult Trait Hope Scale (Feldman and Jazaieri, 2024), RAND Medical Outcome Study 36-Item Short Form Health Survey (SF-36) (Hays, Sherbourne and Mazel, 1993), Functional

Assessment of Cancer Therapy-General (FACT-G) (Brucker *et al.*, 2005), and Patient Health Questionnaire-9 item (PHQ-9) (Kroenke, Spitzer and Williams, 2001).

Cheung's (2019) study involved 60 participants aged 7-16 years who had completed treatment for paediatric brain tumours. Inclusion criteria required a Centre for Epidemiological Studies Depression Scale for Children (CES-DC) (Shahid *et al.*, 2012) score of 16 or above and a Mini Mental State Examination (MMSE) (Molloy and Standish, 1997) score of 18 or above. Exclusion criteria included evidence of cancer recurrence or a second malignancy. The intervention was Musical Training, delivered via weekly home visits by research assistants, with outcomes measured using the CES-DC, Rosenberg Self-Esteem Scale (RSES) (Gnambs, Scharl and Schroeders, 2018), and Pediatric Quality of Life Inventory 4.0 Generic Core Scale (PedsQL 4.0) (Varni, Seid and Cheryl, 1999).

van Dijk-Lokkart's (2016) research involved 68 participants diagnosed with various types of childhood malignancies, within 12 months of treatment completion. Inclusion criteria required diagnosis with any type of childhood malignancy and treatment with chemotherapy and/or radiotherapy. Exclusion criteria included the need for stem cell transplantation, growth hormone therapy, or being wheelchair-dependent, unable to read, write, self-reflect, or follow instructions due to learning difficulties. The FitSurvivor intervention included physical exercise training and psychosocial training, with outcomes measured using three versions of the Dutch PedsQL (Schepers *et al.*, 2017), the Child Behavior Checklist (Achenbach, 1999), and the Youth Self-Report (Song, Singh and Singer, 1994).

Grenawalt's (2023) study involved 127 participants aged 18-30 years, diagnosed with brain tumours before the age of 18. Participants had to be capable of consenting to research. The intervention was an Internet-based Behavioural Activation Intervention, with outcomes measured using the Life Satisfaction Questionnaire (LiSat-9) (Post *et al.*, 2012), Perceived Stress Scale (PSS-10) (Cohen, 1999), and Behavioural Activation for Depression Scale – Short Form (BADS-SF) (Manos, Kanter and Luo, 2011).

Haydon's (2021) research involved 203 participants aged 18-39 years, in remission from cancer. Inclusion criteria required a cancer diagnosis between the ages of 15-39 years, completion of primary treatment, fluency in English, and internet access. Exclusion criteria included currently receiving treatment for cancer recurrence or an inability to commit to the intervention. The intervention was an online prosocial intervention, including expressive writing and peer helping conditions, with outcomes measured using the Mental Health Continuum-Short Form (MHC-SF) (Lamers *et al.*, 2011), Centre for Epidemiologic Studies Depression Scale (Radloff, 1991), Generalized Anxiety Disorder scale (GAD-7) (Spitzer *et al.*, 2006), and various measures from the Patient Reported Outcomes Measurement Information System (PROMIS) (Cella *et al.*, 2010).

Howell's (2018) study involved 78 participants aged >11 and <15 years, in active follow-up care. The intervention was a Web-based Physical Activity Intervention, with outcomes measured using the Pediatric Quality of Life Inventory (PedsQL) (Varni, Seid and Cheryl, 1999).

Kazak's (2004) study involved 150 participants aged 8-18 years, who had completed treatment for various types of childhood cancer. Inclusion criteria required participants to be within the above specified age range and have a history of any childhood cancer. Exclusion criteria included relapse, learning difficulties, lack of fluency in English, or living more than 150 miles from the hospital. The intervention was the Surviving Cancer Competently Intervention Programme (SCCIP), including cognitive-behavioural and family therapy approaches, with outcomes measured using the Impact of Events Scale—Revised (IES-R) (Creamer, Bell and Failla, 2003), Post-Traumatic Stress Disorder Reaction Index (PTSD-RI) (Steinberg *et al.*, 2004), State–Trait Anxiety Inventory (STAI) (Marteau and Bekker, 1992), and Revised Children’s Manifest Anxiety Scale (RCMAS) (Reynolds and Paget, 1983).

Kunin-Batson's (2016) research involved 52 participants aged 15-29 years, with a history of hematologic malignancy or malignant neoplasm, off treatment, and who were in first remission. Participants had to be English-speaking and have access to a computer with internet access, with exclusion criteria including significant visual, neurological or cognitive impairments. The intervention was a Web-based Health Information and Support Intervention, with outcomes measured using the State Trait Anxiety Inventory (STAI) (Marteau and Bekker, 1992) and Multidimensional Health Locus of Control (HLC) (Wallston *et al.*, 1976).

Li's (2022) study involved 153 participants aged 15-39 years, diagnosed with various types of cancer. Inclusion criteria required participants to be within the specified age range, diagnosed with cancer, and willing to participate in research. Exclusion criteria included existing mental illness, other physical diseases, communication disorders, drug or alcohol dependency. The intervention was a Physical Activity Intervention and a Behavioural Activation (BA) Intervention, with outcomes measured using the Pittsburgh Sleep Quality Index (PSQI) (Carpenter and Andrykowski, 1998), Functional Assessment of Cancer Therapy Scale–General (FACT-G) (Cella *et al.*, 2024), Social Support Rating Scale (SSRS) (Malecki and Elliott, 1999), and General Self-Efficacy Scale (GSES) (Chen, Gully and Eden, 2001).

Psihogios' (2021) research involved 224 participants aged 15-29 years, who had completed curative treatment for cancer at a paediatric cancer centre. Inclusion criteria required participants to be English-speaking AYA, have completed treatment, and cognitively capable of completing the study procedures. The intervention was AYA STEP (self-management via Texting, Education, and Plans for Survivorship), with outcomes measured using the PROMIS Profiles (Cella *et al.*, 2010).

Rabin's (2016) study involved 35 young adult cancer survivors (YACS) aged 18-39 years. Inclusion criteria required participants to be within the specified age range, diagnosed with cancer in the past 10 years, completed all cancer treatment (with the exception of hormone treatment), in remission, able to read, write, and speak fluently in English, not regularly participating in relaxation activities, and described as sedentary. The intervention was RENEW (Reach out to ENhancE Wellness), incorporating Physical Activity and Mindfulness Meditation, with outcomes measured using the Profile of Mood States (POMS) (Andrade and Rodríguez, 2018).

Rosenberg's (2021) study involved 92 AYA participants aged 13-25 years, diagnosed with malignancies treated with systemic chemotherapy. The intervention was PRISM (Promoting Resilience in Stress Management), with outcomes measured using the Pediatric Quality of Life (PedsQL) (Varni, Seid and Cheryl, 1999), Hope Scale (Snyder *et al.*, 1997), Connor Davidson Resilience Scale (CDRISC-10) (Connor and Davidson, 2003), and Kessler-6 psychological distress scale (Mewton *et al.*, 2016).

Sansom-Daly's (2021) study involved 40 participants aged 18-39 years who had completed treatment for various types of cancer. Inclusion criteria required participants to be fluent in English and diagnosed with cancer, with no specific exclusion criteria mentioned. The intervention was the Recapture Life Intervention, a cognitive-behavioural therapy (CBT) online program designed to improve quality of life, psychological well-being, and coping strategies. The outcomes were measured using various tools, including the Impact of Cancer Scale AYA module (Husson and Zebrack, 2016) to assess the positive and negative impacts of cancer, the Depression Anxiety and Stress Scale-Short Form (Ali *et al.*, 2022) to measure depression and anxiety symptoms, the Centrality of Events Scale-Short Form (Gehrt *et al.*, 2018) to assess identity changes, a subset of 17 items from the Cancer Needs Questionnaire for Parents/Carers of Adolescents and Young Adults with Cancer (Carey *et al.*, 2012) to gauge unmet needs, and the KIDCOPE (Spirito, 1996) to measure coping strategies.

Santacroce's (2010) research involved 19 participants aged 15-25 years, with a history of any childhood cancer. Inclusion criteria required diagnosis with childhood cancer at least 5 years ago, completed treatment at least 2 years ago, and no current evidence of active cancer. The intervention was Telephone-Delivered Craniosacral Therapy Technique (CTT), with outcomes measured using the Mishel Uncertainty in Illness Scale–Community (MUIS-C) (Sharkey *et al.*, 2019), and State-Trait Anxiety Inventory–State subscale (Kendall *et al.*, 1976).

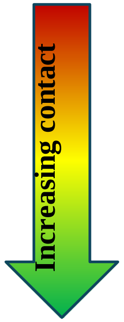
PARTICIPANTS

A total of 1,109 participants aged 8–39 years were included, with mixed populations of males and females included across studies. In eight of fifteen studies, more female participants were included than males, one study had a 50:50 gender split, and the remaining seven studies included more male than female participants. Participants were reported as survivors of many cancers, including: Blood **(**Leukaemia n=161; Lymphoma n=155; Blood non-specified n=237), Brain and Central Nervous System (CNS) (Brain Tumour n=148; CNS n= 24), Breast (n=41), Bone (n=22), Cervical (n=2), Colorectal (n=3), Germ Cell (n=1), Melanoma (n=10), Neuroblastoma (n=6), Retinoblastoma (n=11), Sarcoma (Soft Tissue Sarcoma n=5; Ewing Sarcoma n=1; Rhabdomyosarcoma n=4), Solid Tumours (n=115), Thyroid (n=26), Testicular (n=3), and other non-specified (n=134).

INTERVENTIONS AND COMPARATORS

No two studies evaluated the same intervention and all 15 interventions were evaluated in a single study. These were all modular and scheduled over multiple sessions, ranging from 4 to 52 weeks. They have been grouped into categories, highlighting similarities in delivery methods and area of focus (Table 9).

TABLE 9: INTERVENTION CATEGORIES



		Type of content		
		Integrated physical activity	**Educational**	**Stress management and coping**
Mode of delivery	**App**		Berg = AWAKE Intervention Web-Based Physical Activity Intervention Psihogios = AYA STEP (Self-Management via Texting, Education, and Plans for Survivorship)	
	Internet	Grenawalt = Physical Activity, Behavioural Activation, and Usual Treatment	Grenawalt = Internet-based Behavioural Activation Intervention Kunin-Batson = Web-Based Resource to Improve Cancer Knowledge	Haydon = Online Prosocial Intervention
	Telephone		Arpaci = Technology-Based Psychosocial Education and Counseling Programme Santacroce = Telephone-Delivered Craniosacral Therapy (CTT))	
	In person	van Dijk-Lokkart = FitSurvivor (Physical Exercise Training and Psychosocial Training) Rabin = RENEW (Reach out to ENhancE Wellness) Li = Physical Activity Intervention and a Behavioural Activation (BA) Intervention	van Dijk-Lokkart = FitSurvivor (Physical Exercise Training and Psychosocial Training	Rosenberg = PRISM (Promoting Resilience in Stress Management) Sansom-daly = Recapture Life Intervention Cheung = Musical Training Kazak = Surviving Cancer Competently Intervention Programme (SCCIP)

Note. Some interventions appear across multiple categories

5.12 DATA SYNTHESIS

A narrative synthesis was performed to provide an overview of main outcomes and trends. Meta-analysis was not conducted because of the heterogeneity in interventions and outcomes reported.

1. EFFICACY IN IMPROVING SURVIVORS' MENTAL WELL-BEING

Due to the wide variety of outcomes measured, the effectiveness of interventions is reported within the following categories of outcome; QoL, depression, anxiety, stress, mood, behaviour, self-efficacy, coping, and support. For summary of findings see table 10. All effect sizes can be found in the table in appendix B5.

TABLE 10: INTERVENTION INFLUENCE ON WELLBEING AND PSYCHOLOGICAL HEALTH

Outcome category	**Effect direction**	**Author**	**Type of content**			**Mode of delivery**				**Findings**
			Integrated physical activity	**Educational**	**Stress management and coping**	**App**	**Internet**	**Telephone**	**In person**	
QOL	Positive	Arpaci		✓				✓		Benefits at 12 months
		Cheung			✓				✓	
		Rosenberg			✓				✓	Benefits at 24 months
		van Dijk-Lokkart	✓	✓					✓	Short-term benefits in reducing pain and procedural anxiety
	Negative									Limited overall improvement
Depression	Positive	Berg		✓		✓				Potential efficacy
		Cheung			✓				✓	Reduced depressive symptoms in brain tumour survivors
		Grenawalt	✓	✓			✓			Improved life satisfaction and low mood symptoms in brain tumour survivors
		Haydon			✓		✓			Decreased depression over time from baseline to one-month follow-up
		Sansom-Daly			✓				✓	Peer support condition improved depressive symptoms
	Negative									Limited improvement in depressive symptoms
	No difference	van Dijk-Lokkart	✓	✓					✓	No significant changes in depressive symptoms
Anxiety	Positive	Kazak			✓				✓	Marginal effect on fathers' anxiety
		Santacroce		✓				✓		Reduced anxiety in survivors; control group remained stable or deteriorated
	No difference	Haydon			✓		✓			No significant differences between intervention and control groups but decreased anxiety over time
		Kazak			✓				✓	No significant changes in anxiety for survivors, mothers, or siblings
		Kunin-Batson		✓			✓			No significant differences between intervention and control groups on anxiety scales
Stress	Positive	Kazak			✓				✓	Reduced post-traumatic stress symptoms
		Santacroce		✓				✓		Reduced stress and post-traumatic stress in survivors and parents
		Rosenberg			✓				✓	Sustained improvements in stress and psychological distress
	No difference	Grenawalt	✓	✓				✓		No significant reduction in perceived stress

Mood	Positive	Rabin	✓			✓			Improved mood (POMS scale) associated with increased physical activity
		Haydon			✓			✓	Improved positive and negative affect over time
Behaviour	Positive	Santacroce		✓		✓			Improvements in behaviour, benefit finding, and health promotion
	No difference	van Dijk-Lokkart	✓	✓			✓		No significant differences in behavioural challenges between intervention and control groups
Self-Efficacy	Positive	Arpaci		✓				✓	Increased emotional self-efficacy and coping scores at 3-month follow-up
		Li	✓				✓		Physical activity group showed sustained improvements at 1-week and 3-month follow-ups compared to control and BA groups
Coping	Positive	Arpaci		✓				✓	Improved coping skills compared to routine follow-up
		Sansom-Daly			✓		✓		Increased use of adaptive coping strategies over time
Social Support	Positive	Li	✓					✓	Improved social support scores in the physical activity group
	No difference	Haydon			✓			✓	No significant improvements in social support over time

Note. **Positive** indicates improvement or beneficial outcome. **Negative** indicates limited improvement or other suboptimal outcomes. **No difference** indicates no statistically significant changes between intervention and control groups.

	• Santacroce = reduced stress and post-traumatic stress in both survivors and their parents. • Rosenberg = sustained improvements in stress and psychological distress.		
Mood	• Rabin = intervention group showed significant improvement in mood as measured using the POMS scale. This improvement was associated with increased minutes of 'at least moderate intensity' physical activity per week. Statistical analyses demonstrated significant interaction effects for the intervention group for both physical activity and mood improvement. • Haydon = used PANAS-X to report improvements in both positive and negative affect over time. The study's adjusted means further highlighted a main effect of time, with depressive symptoms and anxiety decreasing across various conditions from baseline to post-intervention and baseline to 1-month follow-up.		
Behaviour	• Santacroce = improvements in the behaviour of survivors and their parents, particularly in terms of benefit finding (ability to perceive positive outcomes or personal growth) and health promotion (engage in behaviours that actively seek to improve health and well-being).	• van Dijk-Lokkart = no significant differences in behavioural challenges between intervention and control groups.	
Self-efficacy	• Arpaci = significant increase in emotional self-efficacy scores for the intervention group over time, with higher scores than the control group at 3-month follow-up. The intervention group also exhibited higher active coping scores and lower avoidant coping scores compared to the control group. Overall, the findings suggest that the technology-based programme had a positive influence on the self-efficacy of survivors, with between-group comparisons indicating significant improvements. • Li = physical activity group demonstrated a significant difference in self-efficacy in comparison to the control group at 3 months post-intervention. This was in comparison to the BA group, who did not show a significant difference from the		

	control group at the same time point. This suggests that the impact of the physical activity intervention on self-efficacy was better sustained, compared to the BA intervention. Moreover, the physical activity group consistently had higher self-efficacy scores than the BA group and control group at both 1 week and 3 months post-intervention.		
Coping	• Arpaci = significantly improve coping skills of survivors compared to the routine long-term follow-up control group. • Sansom-Daly = participants in both interventions reported increased use of coping strategies 6-week post-intervention, but with survivors demonstrating more adaptive coping strategies at 12 weeks than those in the peer-support group.		
Social support	• Li = physical activity intervention group demonstrated statistically significant, higher scores compared to the behaviour activity group and the control group at both one-week and three-months post-intervention.	• Haydon = the intervention group did not demonstrate significant improvement over time.	

1.1 QoL

Four studies utilised the Pediatric Quality of Life Inventory (The PedsQL 4.0) (Upton *et al.*, 2005); (van Dijk-Lokkart *et al.*, 2016; Cheung *et al.*, 2019; Rosenberg *et al.*, 2021; Arpaci, Altay and Copur, 2023). Considering the four studies together, it became evident that the different interventions were reported to have varying impact on survivor QoL. Three interventions were reported to show positive effects on QoL in the longer term (Arpaci's technology-based psychosocial intervention n=12 months, Cheung's musical training n=12 months, Rosenberg's PRISM n=24 months) (Cheung *et al.*, 2019; Rosenberg *et al.*, 2021; Arpaci, Altay and Copur, 2023) whereas the other indicated limited overall impact, with minor short-term benefits (van Dijk-Lokkart *et al.*, 2016). However, each study used a different type of intervention, making it difficult to make direct comparisons. van Dijk-Lokkart et al. was the only intervention of the four to include physical exercise, and although found short-term positive effects on pain and procedural anxiety, this was reported by parents not the survivors themselves. Additionally, there was found to be no significant overall improvement in QoL or well-being.

1.2 Depression

Six studies used depression measures, but none utilised the same scale. Four interventions found clinically significant improvements; the AWAKE app-based intervention demonstrated potential efficacy, as did musical training which significantly reduced depressive symptoms in brain tumour survivors. Additionally, Grenawalt's internet-based BA intervention positively impacted life satisfaction and symptoms of low mood in young adult brain tumour survivors. Haydon found a main effect of time on depressive symptoms in peer helping and expressive writing intervention groups, indicating decreased depression from baseline to one-month follow-up. As with QoL, van Dijk-Lokkart's intervention did not show significant changes in depressive symptoms post-intervention between the intervention and control groups. Sansom-Daly's online CBT programme found that participants across groups reported higher levels of depression at 12-weeks and 12-months post-programme. This was in comparison to the peer support condition who had improved symptoms, highlighting the nuances of different psychosocial interventions.

1.3 Anxiety

Five studies used anxiety measures. Haydon's study of prosocial writing interventions demonstrated that whilst there was a decrease in anxiety over time, there were no significant differences in anxiety between intervention and control groups. Kazak found that although the intervention had a marginal effect on fathers of survivors’ anxiety, overall, it did not significantly impact anxiety levels in adolescent survivors, mothers, or siblings. Santacroce's pilot study highlighted that the HEROS PLUS coping skills training (CST) intervention led to reduced anxiety in TYA survivors compared to those in the control group who remained the same or deteriorated over time. Having said this, the findings were not stated to be statistically significant. Kunin-Batson did not find significant differences between intervention and control groups on anxiety scales.

1.4 Stress

Three of the four studies which measured stress outcomes found that their interventions had a positive impact. Kazak's study demonstrated the effectiveness of SCCIP, a CBT and family therapy approach, in reducing post-traumatic stress symptoms. Similarly, Santacroce's HEROS PLUS CST intervention provided promising results in reducing stress and post-traumatic stress in both survivors and their parents. Rosenberg's PRISM intervention also reported sustained improvements in stress and psychological distress.

In contrast, Grenawalt's BA intervention showed no significant reduction in perceived stress. Although it positively impacted TYA’s life satisfaction, it did not have significant effects on stress levels.

1.5 Mood

Two studies collected data on mood. Rabin's study explored the effects of a physical activity and meditation intervention on mood. The intervention group showed significant improvement in mood as measured using the POMS scale (Curran, Andrykowski and Studts, 1995). This improvement was associated with increased minutes of ‘at least moderate-intensity’ physical activity per week. Statistical analyses demonstrated significant interaction effects for the intervention group for both physical activity and mood improvement. Haydon used PANAS-X (Watson and Clark, 1999) to report improvements in both positive and negative affect over time. The study's adjusted means further highlighted a main effect of time, with depressive symptoms and anxiety decreasing across various conditions from baseline to post-intervention and baseline to 1-month follow-up. The improvements in positive and negative affect, as well as the decreases in depressive symptoms and anxiety, happened across all three conditions (two intervention, one control).

1.6 Behaviour

Two studies used two separate behavioural outcome measures; it is of note that the behaviours measured are varied, and therefore should be compared cautiously. van Dijk-Lokkart assessed behavioural difficulties (internalising, anxiety, depression, withdrawal. Externalising; aggression, ‘delinquency’, hyperactivity) in survivors. The results showed that at baseline, a notable percentage of parents reported clinically significant total, internalising, and externalising challenging behaviours. However, the study found no significant differences in behavioural challenges between the intervention and control groups after the intervention period. Santacroce focused on coping skills training delivered by telephone, finding improvements in the behaviour of survivors and their parents. Results showed that outcomes for AYA and parents who received the HEROS PLUS intervention improved, particularly in terms of benefit finding (ability to perceive positive outcomes or personal growth) and health promotion (engage in behaviours that actively seek to improve health and well-being).

1.7 Self-Efficacy

Two studies used two separate measures of self-efficacy. Arpaci’s technology-based intervention showed a significant increase in emotional self-efficacy scores for the intervention group over time, with higher scores than the control group at 3-month follow-up. The intervention group also exhibited higher active coping scores and lower avoidant coping scores compared to the control group. Overall, the findings suggest that the technology-based programme had a positive influence on the self-efficacy of survivors, with between-group comparisons indicating significant improvements. Li found that the physical activity group of their intervention demonstrated a significant difference in self-efficacy in comparison to the control group at 3 months post-intervention. This was in comparison to the BA group, who did not show a significant difference from the control group at the same time point. This suggests that the impact of the physical activity intervention on self-efficacy was better sustained, compared to the BA intervention. Moreover, the physical activity group consistently had higher self-efficacy scores than the BA group and control group at both 1 week and 3 months post-intervention. These combined findings suggest the potential efficacy of technology-based psychosocial education programmes and physical activity interventions in enhancing self-efficacy among TYA survivors.

1.8 Coping

Two studies examined coping outcomes, both (Sansom-Daly *et al.*, 2021; Arpaci, Altay and Copur, 2023) using the KIDCOPE (Spirito, 1996). Collectively, both studies emphasised the positive impact of their respective online CBT, peer support, and technology-based psychosocial education interventions.

Sansom-Daly's study focused on coping strategies in two different interventions: Recapture Life (CBT) and a peer-support group. Participants in both interventions reported increased use of coping strategies 6-week post-intervention, but with survivors demonstrating more adaptive coping strategies at 12 weeks than those in the peer-support group. Arpaci's intervention was also found to significantly improve coping skills of survivors compared to the routine long-term follow-up control group.

1.9 Support

Two studies assessed support outcomes, both (Haydon, 2021; Li *et al.*, 2022) using the 21-item 2-way Social Support Scale (2-Way SSS) (Shakespeare-Finch and Obst, 2011). Both studies highlight the positive impact of interventions on social support among survivors. Haydon's study focused on prosocial interventions, specifically peer helping and expressive writing with peer helping, evaluating effects on social support. Ultimately, the intervention group did not demonstrate significant improvement over time. Li's study investigated the impact of physical activity on social support. The physical activity intervention group demonstrated statistically significant, higher scores compared to the behaviour activity group and the control group at both one-week and three-months post-intervention.

1.10 Miscellaneous

Eleven studies utilised twenty-three different outcome measures that did not fall into any other category, therefore were omitted from the synthesis. For reference, the outcomes used are reported in the notes of the table in appendix B4.

2. PSYCHOSOCIAL INTERVENTIONS AND POSITIVE INFLUENCE ON WELL-BEING AND PSYCHOLOGICAL HEALTH OF TYA SURVIVORS

Three interventions were found to positively influence QoL: Cheung's music training, Rosenberg's PRISM stress management, and Arpaci's technology-based psychosocial programme. Additionally, van Dijk-Lokkart's physical exercise intervention showed minor short-term benefits but had limited overall impact on QoL. In terms of depression, Berg’s AWAKE app-based intervention, Cheung’s musical training, Grenawalt's internet-based BA, and Haydon’s peer helping and expressive writing interventions showed improvements. For anxiety, Haydon's prosocial interventions, particularly peer helping and expressive writing with peer helping, showed trends towards greater increases in social support and reductions in anxiety symptoms.

Focusing on stress outcomes, Kazak's SCCIP intervention, Santacroce's HEROS PLUS telephone-based craniosacral intervention, and Rosenberg's PRISM intervention positively impacted stress and post-traumatic stress symptoms. For mood, Rabin's physical activity and meditation intervention and Haydon's peer helping and expressive writing interventions showed improvements. Concentrating on behaviour outcomes, Santacroce's HEROS PLUS CST intervention positively impacted health promotion behaviour and benefit finding in survivors and their parents. For self-efficacy, Arpaci's technology-based intervention positively influenced emotional self-efficacy and coping skills. Furthermore, Li's physical activity intervention demonstrated sustained positive impact on self-efficacy compared to BA. In terms of improved coping skills, both Sansom-Daly's Recapture Life CBT programme and Arpaci's psychosocial education intervention had positive results.

Lastly, considering support skills, Haydon's prosocial interventions, particularly peer helping and expressive writing with peer helping, showed a trend towards greater increases in social support. Additionally, Li's physical activity intervention found a positive impact on social support. In summary, various interventions have shown positive influences on different aspects of well-being. Ultimately, prosocial interventions, physical activity, and technology-based psychosocial education programmes demonstrated the highest positive effects across multiple domains.

3. PSYCHOSOCIAL INTERVENTIONS AND NEGATIVE IMPACTS OR 'ADVERSE EVENTS' ON WELL-BEING AND PSYCHOLOGICAL HEALTH OF TYA SURVIVORS

The following interventions were found to be negatively influential on QoL; van Dijk-Lokkart's intervention and Sansom-Daly's Recapture Life online CBT programme did not significantly impact levels of depression and, in some instances, participants showed increased symptoms. In addition, Kazak's SCCIP did not significantly impact anxiety levels in survivors, mothers, or siblings. Grenawalt's BA intervention showed no significant reduction in perceived stress, and again, van Dijk-Lokkart's intervention showed no significant differences in behavioural challenges between intervention and control groups. None of the included interventions were found to have adverse effects on well-being and/or psychological health. However, the absence of this explicit information does not necessarily mean that there were not any adverse effects, as studies tend to prioritise reporting positive or neutral outcomes.

5.13 DISCUSSION

The synthesis of the included studies provides a comprehensive overview of the diverse available psychosocial interventions aiming to improve psychosocial well-being of TYA cancer survivors. Despite this, the heterogeneity in outcome measures and intervention types poses many challenges in drawing definitive conclusions. Future research should strive to use standardised outcome measures and consistent sample sizes, compared by diagnosis and age of cancer experience, to enhance comparability and allow for meta-analyses. Additionally, follow-up studies are needed to assess the sustainability of intervention effects across multiple studies and long-term. This should also include interventions being tested in more than one trial to ascertain reproducibility. The absence of explicit reporting on adverse effects also emphasises the importance of systematically evaluating and reporting both positive and negative outcomes in future research.

QoL

Interventions targeting QoL had mixed effects. While Cheung's music training, Rosenberg's PRISM stress management, and Arpaci's technology-based psychosocial education programme showed positive impact, van Dijk-Lokkart's physical exercise intervention showed limited overall improvement. As with all of the included studies, variability in intervention types and outcomes emphasises the necessity for a nuanced understanding of the factors influencing TYA survivors' QoL. Cheung's music training, for instance, might have positively affected TYA survivors by providing an outlet for emotional expression and fostering a sense of community. Rosenberg's intervention could have addressed specific stressors associated with cancer survivorship, offering coping mechanisms and support. Arpaci's technology-based psychosocial education programme may have targeted information gaps and provided valuable resources for survivors. On the other hand, the limited success of van Dijk-Lokkart's physical exercise intervention prompts considerations about the appropriateness of certain interventions for this demographic. The constraints could stem from physical limitations, preferences, or motivational factors unique to TYA survivors. The variability in intervention types and outcomes across studies signals the heterogeneity within the TYA survivor population. Factors such as age, cancer type, treatment history, and individual preferences can significantly impact effectiveness of interventions. Therefore, a one-size-fits-all approach is unlikely to yield consistent results, highlighting the need for personalised and flexible strategies.

Depression

Studies which measured depression utilised completely different outcomes, making direct comparisons challenging. However, interventions such as Berg's AWAKE app, Cheung's musical training, Grenawalt's internet-based BA, and Haydon's peer helping and expressive writing interventions showed potential in lessening symptoms of depression. This highlights the potential of different interventions in addressing depressive symptoms, where present, in survivors. However, the challenge of comparing studies due to the utilisation of different outcomes to measure depression complicates the synthesis of evidence and the identification of overarching trends. The lack of standardised measures may introduce variability and limit the ability to draw clear conclusions about the effectiveness of these interventions. In addition, comparing single studies of different interventions does not allow for nuanced understanding of impact. Future research should adopt standardised assessments for depression, enabling more robust comparisons and facilitating clearer understanding of the effectiveness of different interventions.

Anxiety

Studies investigating anxiety also had nuanced results. Haydon's prosocial interventions exhibited trends towards increased social support and reduced symptoms of anxiety. Santacroce's HEROS PLUS CST intervention also had positive effects. In contrast, Kazak's family focused intervention showed no significant impact on survivors, mothers, or siblings despite having a moderate impact on fathers' anxiety. The mixed findings across studies could emphasise the heterogeneity across included studies. As with all factors, anxiety may differ based on individual characteristics, family dynamics, and coping mechanisms. As such, a generalised approach to anxiety management may be ineffective. Understanding the nuances of family interactions and their differential impact on anxiety levels among survivors is essential for tailoring interventions to achieve optimal outcomes.

Stress

Interventions targeting stress outcomes demonstrated varying efficacy. Kazak's SCCIP intervention, Santacroce's HEROS PLUS CST, and Rosenberg's PRISM reported positive impacts on stress and post-traumatic stress symptoms. However, Grenawalt's BA intervention showed no significant reduction in perceived stress. Despite this, comparators of results and outcomes across disparate measures and intervention are invalid. The discrepancy in outcomes may refer to the need for greater understanding of specific mechanisms through which interventions impact stress. It is also plausible that these findings may have been due to an underpowered study or used an imprecise reporting measure. However, results suggest the need for targeted stress-reduction strategies in interventions for this population.

Mood

Studies focusing on mood outcomes generally indicated positive effects, particularly with Rabin's physical activity and meditation intervention and Haydon's peer helping and expressive writing interventions. These findings underscore the potential of physical and psychosocial interventions to improve mood among TYA survivors. Yet it is important to note potential limitations and consider broader implications. The positive effects observed could be influenced by various factors such as study design, sample size, and participant characteristics. Replicating these findings in diverse populations and contexts would enhance the generalisability of these interventions. Secondly, the specific mechanisms through which physical and psychosocial interventions impact mood outcomes need further exploration. Understanding the underlying processes can guide the development of more targeted interventions, as well as help in adapting them to individual preferences and needs.

Behaviour

Behavioural outcomes were only assessed in two studies, with mixed results. While van Dijk-Lokkart's physical exercise intervention showed no significant differences in behavioural difficulties, Santacroce's HEROS PLUS CST positively impacted health promotion behaviours. Future studies should place greater emphasis on evaluating behavioural outcomes among survivors. This includes not only assessing the presence of behavioural difficulties but also exploring the promotion of positive health-related behaviours. The multifaceted nature of behavioural changes suggests that interventions should be tailored to address specific aspects of behaviour, acknowledging the specific challenges faced by TYA survivors.

Self-Efficacy

Both Arpaci's technology-based intervention and Li's physical activity intervention demonstrated positive impacts on self-efficacy, emphasising that interventions focusing on enhancing TYA’s beliefs in their emotional and coping abilities may be beneficial across different populations. However, the long-term sustainability of the observed positive impacts on self-efficacy is a critical consideration. It is essential to assess whether these improvements endure over time and whether they contribute to lasting positive changes, given the decades of life TYA survivors have to manage the impact of their experiences.

Coping

Studies focusing on improving coping skills, including Sansom-Daly's Recapture Life CBT programme and Arpaci's psychosocial education intervention, highlighted the benefit of different strategies on improving coping skills, suggesting that this is an important element of support tools for TYA childhood cancer survivors. However, it is crucial to delve deeper into specific components of these interventions that contribute to improved coping skills, and test whether these benefits exist outside the single study. Understanding the active ingredients and mechanisms of change can guide the development of more targeted and effective coping interventions.

Social Support

Interventions targeting social support, such as Haydon's prosocial intervention and Li's physical activity intervention, showed positive impacts. Strengthening social support networks appeared to be a key component in enhancing overall well-being. It is important that the sustainability of the observed positive impacts on social support is considered. Evaluating whether the strengthened social networks endure over time and contribute to ongoing well-being is essential for assessing the long-term effectiveness of these interventions. The implications of these findings highlight the integral role of social support in enhancing overall well-being among survivors. Future research should continue to explore and refine strategies that effectively foster social support, recognising the diverse needs within the TYA survivor community and promoting adaptability for sustained positive outcomes.

Miscellaneous Outcomes

The wide array of miscellaneous outcomes across studies, while not included in specific categories or synthesis, adds value to the understanding of the holistic impact of interventions. Outcomes spanned across factors such as hope, self-esteem, uncertainty, sleep quality, resilience, and other domains, highlighting the complexity of TYA survivors' experiences and the challenge in measuring these. It also emphasises the difficulties with designing a singular intervention to support multiple, complex outcomes.

5.14 METHODOLOGICAL CONSIDERATIONS

The included studies share some common methodological limitations. An issue across several studies was small sample size. For instance, Arpaci (n=62) and Berg (n=56) both faced challenges associated with limited statistical power and the ability to detect significant differences. This highlights the need for interventions to be tested with larger, more diverse samples to ensure findings are robust and clinically meaningful. Furthermore, it is important to note that single studies of an intervention can be problematic in that they are more likely to show positive outcomes. It is important that future studies evaluate interventions across multiple studies and in different populations to assess the generalisability and reliability of the results. Replication across various settings and populations can help establish the true efficacy of an intervention and mitigate the risks of false positive outcomes. Ultimately, rigorous and repeated testing will be crucial for translating these research findings into evidence-based practice that can be confidently applied to delivering these interventions in clinical settings.

Bias, in terms of participant selection and the methods used, was evident in several studies. Grenawalt's study primarily consisting of White, educated males, and the use of a convenience sample strategy introduced further potential biases. Equally, the inclusion of predominantly American literature is not fully transferable to other settings. Additionally, the lack of literature from the UK may reflect the lack of RCT funding for psychosocial interventions. The overrepresentation of one specific demographic may inadvertently perpetuate existing disparities in cancer care and overlook the experiences and perspectives of those from underrepresented backgrounds, who may be in greater need of psychosocial support. Kazak's study demonstrated issues with high dropout rates and biases associated with home-based data collection, compromising the internal validity of findings and introducing confounding variables that may have influenced the findings. Santacroce's intervention relied on telephone-delivery, which may have led to biases related to participant preferences and needs.

Lastly, high dropout rates were a recurring challenge, although statistical methods were used to account for these. Kazak acknowledged the differential dropout rates between the intervention and wait-list control groups, with higher rates in the intervention group. This may suggest that there may have been specific challenges or barriers that affected acceptability of the intervention for some individuals. Similarly, Sansom-Daly's research had an underpowered sample and high attrition rates, impacting the representativeness of the results and suggesting difficulties in maintaining participant commitment throughout the study duration. This may indicate potential issues with acceptability of the intervention or study design, such as the demands of multi-session interventions, or burden of multiple questionnaires over a 12-month period. These consistent issues with retention highlight the importance of addressing factors contributing to attrition and devising strategies to enhance participant adherence in future research.

5.15 LIMITATIONS

In terms of limitations of the review itself, the reported evidence on psychosocial interventions for TYA survivors is derived exclusively from RCTs. Although RCT is considered the 'gold standard' in assessing the efficacy of evidence-based cancer therapies (Thakur, 2023), studies utilising different experimental designs are not captured, and thus some meaningful and effective interventions may be overlooked. Furthermore, the inclusion of studies only published in the English language could potentially have impacted data. Publication bias may be apparent, particularly as non-English studies with different findings may be excluded. The review is therefore not representative to survivors in non-English speaking countries or with different cultures to the origins of the included studies. Excluding these studies reduces the comprehensiveness of the review and may result in a narrower understanding of their efficacy and applicability across diverse populations.

5.16 CLINICAL AND RESEARCH IMPLICATIONS

Various interventions demonstrated positive influences on different aspects of well-being among TYA childhood cancer survivors. Prosocial interventions, physical activity, and technology-based psychosocial education programmes exhibited the most consistent positive effects across multiple domains. Therefore, conducting more in-depth research on these aspects in the future, before implementing such interventions in oncology settings could promote social connections and emotional wellbeing amongst TYA survivors. However, based on this review alone, it is difficult to establish whether there is sufficient evidence to recommend their implementation yet. Further research and later establishing guidelines for professionals to adopt such approaches when designing interventions, as well as integrating comprehensive survivorship psychosocial care plans alongside medical interventions, can help to provide holistic support at all stages. Ultimately, tailored, and multifaceted approaches considering the individual needs of this group of cancer survivors are crucial for optimising intervention efficacy. It is also important for clinicians to be clear about which interventions are effective, and if there is a choice of empirically tested support, providing guidance on which might best suit their patient group. These, however, must be rigorously tested to ensure reliable findings across individual interventions and monitored for bias.

5.17 CONCLUSION

This review highlights the potential of varied psychosocial interventions in positively impacting the mental well-being of TYA childhood cancer survivors, contributing valuable insights to the ongoing efforts to enhance the QoL and psychological health of this population. This is increasingly crucial as more TYA survive their diagnoses and must live with potential late effects that can vastly impact psychological well-being. Despite this, comparing single studies of differing interventions does not provide full understanding of nuanced differences and is subject to reporting bias. Concerted effort is needed to improve understanding of which elements of interventions are helpful, neutral, or detrimental. This may include combining data from different study designs, conducting multiple studies on the same intervention, as well as considering diverse cancer diagnoses and populations of TYA. It is hoped that this review highlights existing support and acts as a guide for considering the development of future interventions.

CHAPTER 6: STUDY 3 – “THAT’S ALL ANYONE NEEDS REALLY, SUPPORT”: A CONTENT ANALYSIS OF RETINOBLASTOMA SURVIVORS’ PSYCHOSOCIAL SUPPORT NEEDS

6.1 CONTEXT

Study three involves a retrospective content analysis on the data collected as part of study one. This method was chosen to fulfil the objectives set out at the beginning of this thesis, to identify potential psychosocial intervention content. This will be combined with the findings from studies one and two to generate robust evidence to inform a future intervention.

6.2 BACKGROUND AND RATIONALE

As this thesis has highlighted, adolescents and young adults are often referred to as the ‘lost tribe’ (Stevens, 2006) in supportive cancer care, due to the barriers they experience in accessing specialist psychological support that is specific to their age and stage of life (Zebrack and Isaacson, 2012; Sender & Zabokrtsky, 2015; Bibby, White, Thompson, & Anazodo, 2017). The third study of my PhD came about from the initial aim to develop a psychoeducation intervention tailored for individuals navigating the challenges of Rb survivorship. However, the trajectory of this PhD was altered when study one yielded an unforeseen amount of rich and novel insights into the experiences of survivors. The unexpected depth of these insights warranted a thorough exploration to ensure that the survivors' voices were adequately represented and that their experiences could inform both clinical practice and future intervention development research. In light of the wealth of data in study one, it became evident that a comprehensive approach was necessary to do justice to this novel information effectively and to clearly identify intervention practices. For these reasons, a retrospective analysis of the qualitative data collected for study one was chosen. This is an established methodology for situations in which to examine questions that differ to those asked in the original research question (Long-Sutehall, Sque and Addington- Hall, 2011). Content analysis is also a widely utilised method for examining secondary data in research studies in health science (Erlingsson and Brysiewicz, 2017). It involves systematically analysing data from an existing study to explore patterns, themes, and trends, allowing the drawing of conclusions about a given topic. The process includes coding data, which involves breaking down the content into manageable categories that can be analysed (Vears and Gillam, 2022). Content analysis is particularly useful for identifying the presence of specific concepts or ideas and how they are communicated within the data. While it shares similarities with other qualitative methods, like reflexive thematic analysis used in study one, it differs in several key aspects (Vaismoradi, Turunen and Bondas, 2013). Reflexive thematic analysis is a more flexible and subjective approach, emphasising the researcher's active role in interpreting and constructing themes based on their engagement with the data. It is less structured, allowing themes to be identified through an iterative process, influenced by the researcher’s perspective and reflexivity. In contrast, content analysis is more systematic and structured, often involving pre-defined categories and quantifying the frequency of themes or patterns within the data. This method is deemed to be more objective, focusing on a consistent coding process that can be replicated by other researchers (de Faria-Schützer *et al.*, 2021).

Using this approach for the current study allowed for a more comprehensive understanding of the long-term psychosocial impacts of Rb, generating more evidence for the development of a psychosocial intervention for this group. By re-examining the rich qualitative data collected for study one, new insights could be uncovered that might inform future intervention, support mechanisms, as well as policies and long-term follow-up care tailored to the unique needs of Rb survivors.

6.3 STUDY AIMS

The aims for this content analysis are:

1. To analyse a sub-section of qualitative data collected during study one, focusing on survivors’ experiences of psychosocial support, any interventions received, interventions they would like in the future, and practical considerations of developing and delivering future interventions.

2. To investigate the perceived support networks and resources utilised by Rb survivors to manage their psychosocial wellbeing, with a focus on understanding the effectiveness and adequacy of available support from survivor perspectives.

3. To identify gaps and unmet needs in existing support services and interventions and propose recommendations for the development of a more targeted and comprehensive psychoeducation intervention.

6.4 METHODOLOGY

The data reported in this chapter were collected within study one, utilising individual interviews and focus groups conducted with TYA Rb survivors. As this is a secondary data analysis, the participant profile, PPI, ethics, recruitment and consent information can be found in chapter 3 outlining study one.

6.5 ANALYSIS

Content analysis was used to explore TYA views on desired content for an Rb-specific psychosocial intervention. This included their experiences of specific support that they have received, that they would have found useful if they had had the opportunity, or that they believe would be important to include in a support package for others living beyond Rb. Content analysis was conducted through the process of identifying and coding themes and patterns within the study one interview and focus group data. An inductive approach was taken, with the categories of interest developing from the data as opposed to pre-conceived theoretical assumptions (Kondracki, Wellman and Amundson, 2002). The analysis involved a systematic examination of the data to identify recurring topics, issues, and perspectives surrounding support expressed by participants. Through this method, key themes were highlighted regarding the types of psychosocial support deemed most beneficial by TYA survivors of Rb, as well as gaps in existing services. This, combined with findings from studies one and two, will form the building blocks of a scientific intervention informed by lived experience partners, which can be developed and test in the post-doctoral phase after this PhD.

Participant quotes were coded using NVIVO data analysis software (QSR International PTY LTD., 2020). With support from my supervisor (Debra Howell; DH), I immersed myself in the baseline data and developed an initial coding frame to organise and categorise the information. I then conducted a systematic coding process where I assigned each quote to the relevant code, based upon the experiences discussed. This involved carefully reading and interpreting the quotes to ensure accurate coding. Throughout this process, I met regularly with DH to discuss and review coding decisions, resolve any discrepancies, ensure consistency and reduce bias. Multiple codes were allocated to each quote if they were relevant to more than one category. Subthemes were then reported descriptively.

6.6 TRUSTWORTHINESS OF DATA

Due to the subjective nature of qualitative data, I wanted to ensure it was trustworthy. I therefore drew upon Lincoln and Guba's criteria (Lincoln & Guba, 1986), which states that data can be analysed to ensure credibility, transferability, dependability, conformability, and authenticity (Kakar *et al.*, 2023). Adhering to this model, I implemented several strategies to meet these criteria. For instance, to enhance credibility, I firstly used peer reflections with another qualitative researcher (DH) to gain a consensus of the initial coding frame, themes and subthemes. This was then checked by two other researchers, and discrepancies resolved with consensus discussion. I also employed member reflections by sharing my findings and interpretations with the study participants and my PPI group. This allowed them to review and validate the accuracy of the data and my interpretation, ensuring that their perspectives were accurately represented and reducing the likelihood of researcher bias. Additionally, I provided detailed contextual descriptions to support transferability. By providing details surrounding the context of the data collected in this study (such as demographic details of participants, and the context of the collection of the data), I aimed to ensure that the study's findings were both trustworthy and reflective of the participants' true experiences. I also maintained an audit trail for dependability, meaning that I documented each stage of the research process to ensure that my method can be followed and/or replicated by others, aiding the consistency and reliability of these finding. Finally, I used reflexive journaling to address conformability and to enhance objectivity. Keeping a reflexive journal throughout the entire PhD process was helpful, enabling me to reflect extensively on my thoughts, biases, and potential influence on the research process, see chapter 2 for more in-depth detail. Through following these steps I hope to allow readers of my work to evaluate the context of the data, ensuring that the findings are relevant and meaningful beyond this study, for example, for Rb survivors and clinicians in different countries and cultures who may be interested in understanding the potential content of a psychosocial intervention.

6.7 RESULTS

Thirty-two TYA who are survivors of Rb took part in interviews and focus groups. For participant demographics, please refer to tables 5, 6, and 7 in chapter three. Due to the topic guides developed for the purpose of this study, all participants commented on psychosocial interventions, including their personal experiences, likes and dislikes, hopes, and recommendations. Responses were only recorded where participants explicitly and organically commented on a particular element. The results are presented according to their category and are accompanied by quotations from transcripts to illustrate participant’s experiences. As per study one, quotes are reported with participant number (P) and by self-reported Rb type (bilateral as B, unilateral as U, heritable as H, non-heritable as NH. E.g., P1, NH, U). In instances where the Rb type reported by the individual was not in line with what we know to be biologically plausible, this is marked with an Asterix (e.g. P2, B, NH*).

Each theme is described below, with the number of TYA discussing each category overall and separated by type of Rb (table 10). The content analysis can be found in table 11 and examples of illustrative quotes can be found in appendix C1. To ascertain this data, I began by gathering quotes from the interviews and focus groups conducted as part of study one. I then identified recurring themes before counting the number of individuals who discussed each theme to quantify the data. This process ensured that I could categorise the frequencies of each experience, including a percentage of individuals from a particular category (i.e. a person can appear in both the bilateral and heritable Rb columns). This allowed for a detailed analysis of the number and distribution of themes within each type of Rb, providing insights into how different forms of the condition affect young people's experiences and desires for intervention content. It was important for me to separate desired intervention content in this way so that the future intervention can be tailored to differing needs and experiences, the importance of which is highlighted in both study one and two.

TABLE 10: NUMBER OF PARTICIPANTS DISCUSSING EACH THEME AND SUBTHEME, OVERALL AND BY RETINOBLASTOMA TYPE

	Overall (n=32) **n (%)**	**Bilateral Rb (n=9)** **n (%)**	**Unilateral Rb (n=23)** **n (%)**	**Heritable Rb (n=10)** **n (%)**	**Non-Heritable Rb (n=21)** **n (%)**	**Unknown heritable status (n=1)** **n (%)**
Psychosocial support provided						
Had therapy	9 (28%)	4 (44%)	5 (21%)	6 (60%)	4 (19%)	0 (0%)
Think therapy beneficial	11 (34%)	5 (55%)	6 (26%)	5 (50%)	6 (28%)	0 (0%)
Received specific Rb psychosocial support	3 (9%)	1 (11%)	2 (8%)	2 (20%)	1 (4%)	0 (0%)
Wishes for interventions						
Therapy/counselling	13 (40%)	2 (22%)	11 (47%)	5 (50%)	7 (33%)	1 (100%)
Talking about feelings	16 (50%)	6 (66%)	10 (43%)	6 (60%)	11 (52%)	0 (0%)
Interact with peers with similar history	24 (75%)	8 (88%)	16 (69%)	9 (90%)	16 (76%)	0 (0%)
Support to deal with:						
• Physical appearance	20 (62%)	6 (66%)	14 (60%)	7 (70%)	13 (62%)	1 (100%)
• Bullying	23 (71%)	7 (77%)	16 (69%)	8 (80%)	15 (71%)	1 (100%)
• Mental health	26 (81%)	7 (77%)	19 (82%)	7 (70%)	17 (81%)	1 (100%)

• Sex and relationships	9 (28%)	3 (33%)	6 (26%)	3 (30%)	6 (28%)	0 (0%)
• Second cancers	18 (56%)	5 (55%)	13 (56%)	8 (80%)	10 (47%)	1 (100%)
• Having children	18 (56%)	5 (55%)	13 (56%)	9 (90%)	10 (47%)	1 (100%)
• Preparing for the future	20 (66%)	8 (88%)	12 (52%)	7 (70%)	13 (61%)	1 (100%)
• Understanding Rb	24 (75%)	5 (55%)	19 (82%)	8 (80%)	15 (71%)	1 (100%)
• Living with visual impairment	18 (56%)	5 (55%)	13 (56%)	6 (60%)	11 (52%)	1 (100%)
• Learning how to communicate with others about Rb	24 (75%)	6 (66%)	18 (78%)	7 (70%)	16 (76%)	1 (100%)
• Health conditions related to Rb (e.g. dry eye)	15 (46%)	6 (66%)	9 (39%)	5 (50%)	10 (47%)	1 (100%)
Barriers to support						
Others have it worse	6 (18%)	2 (22%)	4 (17%)	2 (20%)	5 (23%)	0 (0%)
Not sure what is available	6 (18%)	2 (22%)	4 (17%)	2 (20%)	5 (23%)	0 (0%)
Stigma	4 12%)	1 (11%)	3 (13%)	0 (0%)	4 (19%)	0 (0%)
Practical Intervention considerations						

Adolescence is the time of need	26 (81%)	5 (55%)	21 (91%)	7 (70%)	16 (76%)	1 (100%)
Integrated with existing appointments	5 (15%)	0 (0%)	5 (21%)	0 (0%)	5 (23%)	0 (0%)
Intervention delivery:						
• Online	10 (31%)	2 (22%)	8 (34%)	4 (40%)	7 (33%)	0 (0%)
• In-person	11 (33%)	3 (33%)	8 (34%)	4 (40%)	6 (28%)	1 (100%)
• Modular (on-demand)	17 (53%)	6 (33%	11 (47%)	6 (60%)	8 (38%)	1 (100%)
• Workshops	6 (18%)	2 (22%)	4 (17%)	3 (30%)	3 (14%)	0 (0%)
• Videos	11 (33%)	1 (11%)	10 (43%)	2 (20%)	9 (42%)	0 (0%)
The facilitator needs specific Rb knowledge	15 (46%)	3 (33%)	12 (52%)	5 (50%)	12 (57%)	0 (0%)
Someone who’s not your Mum, or your doctor	15 (46%)	2 (22%)	13 (56%)	5 (50%)	12 (57%)	0 (0%)

PSYCHOSOCIAL SUPPORT PROVIDED

Participants discussed the concept of traditional therapy in detail, considering whether this was something that they had access to, and if so, whether they chose to engage in this and what their experiences were. Of the 32 participants, only 9 had accessed therapy during childhood, six of whom were young adults at time of reporting and three of whom were teenagers. There were not huge differences between individuals impacted bilaterally or unilaterally, nor those impacted heritably or non-heritably. In terms of Rb-specific psychological support, only one third of the nine individuals had had access to this, suggesting that this is not readily available. For those that had, this was seen to be helpful but could be harmful when therapeutic endings were not considered.

"I had this counsellor in year three or year four, I can't remember, but she used to pull me outta lessons and like she, she bought me this book and it was like a worry book, I guess (laughs) and I like draw like just anything really and like she'd get me to make lists of like negative things and positive things about having cancer, like retinoblastoma, so yeah, but then she just sort of stopped coming to see me." (P10, U, NH, focus group)

Eleven participants believed that generic therapy was beneficial to them, again with no differences between diagnostic groups.

"I need therapy to just be a better person or be, be a better, better version of myself not only in relation to retinoblastoma but, you know, every part of me." (P32, B, NH)*

This indicates a broad recognition of the value of psychological support in managing the multifaceted impact of Rb, regardless of the subtype diagnosed. It also suggests that a future therapeutic intervention of this type would be warranted and wanted.

"I think therapy is really good, and I feel as though people should – like everyone should go. I don't feel like you should just be sick." (P25, U, NH)

Experiences like this underscores the holistic benefits of therapy, extending beyond Rb-specific issues to overall personal development and well-being, particularly for young people who have experienced trauma as a result of their diagnosis and treatment, as many of this sample had.

"The psychiatric therapy, that one, was much more targeted to me, my body, how I relate to my body and what Rb, what that … I mean it's Rb in some ways and that's some sort of emotional trauma but I actually think it's the chemo actually that would be my working theory is the, is the, is the real, was the real problem that's, that's how I kind of feel in my emotional life anyway is the, I really kind of think back you know: what would that be like for like a tiny brain to undergo that level of exposure to toxicity?" (P23, U, H)

WISHES FOR INTERVENTIONS

Broadly speaking, participants expressed a desire for various interventions, including therapy/counselling (40% overall; 22% bilateral Rb, 47% unilateral Rb), 'talking about feelings' (50% overall; 66% bilateral Rb, 43% unilateral Rb), and interacting with peers with a similar history (75% overall; 88% bilateral Rb, 69% unilateral Rb). Across all categories, individuals impacted by heritable Rb requested greater needs for support. This might reflect the longer-term challenges that must realistically be considered by this group.

"Maybe having someone to talk to growing up – 'cos, you know, you have down days and you have up days, an if you're having a down day, maybe just talking to someone who isn't your parents might be helpful, who really, really gets it."(P31, U, H)

Thirteen individuals expressed that they did not have access to therapy. The majority of these individuals were impacted unilaterally and non-heritably, with nearly 50% of this group desiring traditional therapeutic support. This highlights a significant gap in the availability of mental health services for these individuals. One participant reflected on the limitations of current support structures:

"I think sometimes like giving solutions or whatever, although it seems like it would be helpful, like sometimes it's just not. I think just support and just a bit more – you're right, like a lot of anxiety had come from it, so just something probably to help with that and support, and talking to other people in the same situation, rather than solutions that are not gonna apply to everyone, but I think most people would probably use that support." (P28, U, NH)

This insight reveals a desire for more personalised and tailored support, rather than generic approaches designed for young people as a whole. Participants highlighted the importance of access to therapeutic services that could address their unique Rb-specific anxieties and help them connect with others facing similar challenges. This need for accessible, tailored psychological support is crucial for improving the mental health and quality of life for these individuals.

"I'm so happy for this awareness around therapy and this platform for like, better health and all that stuff because yeah…it is something and I wish we had that back in the day when I was thirteen, fourteen years old, even earlier." (P32, B, NH)*

Many individuals felt that they did not have the necessary tools to manage difficult emotions, which added to the challenges that they faced in coping with Rb and its aftermath.

"I have learnt by myself and nobody gave me any tools or, you know, how to deal with insecurities, how to deal with feeling different, erm, different or depression or erm, you know, all that kind of stuff that I'm learning now, erm, so yeah, support. That's all, that's all anyone needs really, support." (P32, B, NH)*

Participants expressed a profound need for guidance and support in dealing with insecurities, feelings of being different, depression, and other mental health issues:

"Support from a younger age about the psychological side would have been helpful…I was never really asked about the psychological side, like how it was feeling. I was like maybe if – I was maybe thinking, if you had someone there to talk to you as well as the, erm, psychological side, just like maybe saying, erm, like, I don't know, "my name is so and so, I'm here to help you. how has having one eye affected your school life so far? Like, erm, what do the other kids think of it? What do they say to you?."" (P29, U, NH)

This highlights the gap in early intervention and the need for structured emotional and psychological support from a young age. Another participant emphasised the importance of tools to develop self-confidence as well as wider mental health support:

"for me, I think it probably would be self-confidence…and my mental health, like that's the only things it's really impacted in a major way anyway. I feel like having support to do with them would be good."(P19, U, NH)

The lack of existing tools for managing difficult emotions often left individuals feeling unprepared and overwhelmed. Many had to navigate their emotions alone, without the benefit of professional guidance or peer support.

“obviously I know each situation’s unique to each person and whatnot but I think maybe just some, somewhere that you can go and they’ll point you in the right direction or maybe like, “Oh you won’t need to worry about this until whenever,” or…just something, some information like no matter how little it is I think would definitely help.” (P18, U, U)*

Therefore, participants strongly advocated for interventions that offer practical tools and emotional support, emphasising that such resources could significantly improve their ability to cope with the psychological impacts of Rb.

“probably just knowing the option is there to actually speak to like a professional, like a therapist, a psychologist, whatever, who would be able to help when I was younger, that probably would have been nice.” (P28, U, NH)

Participants in the study frequently emphasised the significance of peer support and the value of sharing experiences with others who have faced similar challenges.

“I think it would have been nice to have other people like in the same age group as me that had also had it [Rb]” (P28, U, NH)

Whether they had accessed this or not, many believed that connecting with peers who understand their struggles on a personal level could provide comfort and a sense of community. This form of support helps to normalise feelings and experiences, making individuals feel less isolated.

“there’s a girl I met through Instagram, and she arranged a meet-up with all people who’ve had RB… and I went to that last year. And I found that amazing, because that – that really made me feel a lot better, just meeting other people who’ve like gone through similar experiences. It made me feel so much better, ‘cos I’ve never actually met anyone who had one eye before until that day, I don’t think. It was just meeting everyone else, and hearing their experiences and how they cope with things. It just made me feel so much better after I left.” (P19, U, NH)

These comments highlight the therapeutic power of peer interactions. For many participants, the opportunity to hear from others who have navigated similar Rb-related challenges provided not only practical insights but also emotional relief. It was felt that peer support would validate negative experiences related to Rb and create a shared understanding that is often hard to find in conversations with those who have not had the same experiences. Practical advice and coping strategies shared among peers are often seen as more relatable and easier to implement compared to advice from professionals who may not have personal experience of the condition. Furthermore, hearing success stories and strategies directly from peers who have overcome similar difficulties can instil hope and motivate individuals to persist through their challenges.

“if they offered that sort of stuff [skills] in the, erm, talk sessions, maybe that would be good, erm, to meet other people who’ve had the same experiences. I would say it helped me, because you listen to these people, erm, and they’ve been through the same stuff as you. You listen to them talk and you realise you’re not as alone as you thought, basically.” (P29, U, NH)

SUPPORT TOPICS

Participants sought support to deal with various challenges. Twenty of the 32 participants discussed their physical appearance, with 66% of individuals with bilateral Rb and 60% with unilateral Rb requesting support to manage this. This was particularly prevalent among young adults, although many were reflecting back to their younger years and the pressure to conform to societal standards of beauty set in adolescence. It is of interest that teenagers discussed this less, and may reflect societal narratives about appearance, such as the body positivity movement. There was little difference between those with heritable and non-heritable Rb (70% vs 62%), but understandably, this was most common amongst those who had experienced facial changes as a result of their cancer or treatment.

“this is just an appearance thing and this is just me being vain…but I’ve got quite kind of noticeable sort of indents on the side of my head, which is where radiotherapy happened...so that makes you a bit more self-conscious.” (P27, B, H).

As explored in study one, experiences of bullying were common, with 23 participants sharing their personal stories of being targeted due to their Rb.

“I did suffer a lot when I was younger with bullying. Erm, it [intervention] probably would have been helpful, ‘cos I never really knew how to respond.” (P19, U, NH)

Requests for support were higher amongst those with bilateral Rb (77%), comparative to unilateral (69%), which may be because impact in both eyes is more noticeable than in one. Additionally, young people with heritable Rb had more concerns about bullying (80%).

"Primary school, you'd get the odd comment, but like I don't think I really took it to heart until I started secondary school… I remember like my first day of school, people being awful about it, and I think that's probably where like the worries and stuff came from. That was when I like started covering it up and – yeah." (P28, U, NH)

The impact of these experiences had implications on their mental health, contributing to the 26 participants who described ongoing struggles with mental health issues (refer to study one for in-depth exploration). Both of these topics were high on participants' priorities for support, stating that they wanted an intervention to offer specific strategies to manage these issues (77% bilateral, 82% unilateral). Those with non-heritable disease expressed higher desire for mental health support (81% vs 70%).

"I can't really feel like peaceful or completely happy, if you know what I mean. Like I always feel like there's something kind of wrong. Like I – erm, I don't know, it's – I'm not – I wouldn't say I'm sad, like in despair, but I can't enjoy myself either. I don't know why that is." (P29, U, NH)

Nine individuals had specific worries about sex and/or relationships, seven of whom were young adults aged between 20 and 29 years.

"I am less inclined to have casual flings, to be quite honest, because there's always the potential for that being an unfair burden for anybody to carry, basically, unless you're in a committed relationship." (P27, B, H)

This is likely to reflect the stage of psychosocial development of these participants, as they navigate independence and finding a romantic partner, which was a priority for many. 28% of individuals requested that support was needed for this issue (33% bilateral, 26% unilateral; 30% heritable, 28% non-heritable).

"that's one thing I did feel. I didn't feel worthy 'cos I was disabled in a way. Like I felt a bit useless, like a burden, if you know what I mean. Erm, obviously, that wasn't true, but it's just something that was like gnawing in the back of my head." (P29, U, NH)

To address these concerns, interventions should encompass tailored support that acknowledges the unique sexual and relationship challenges faced by individuals who have had Rb. Providing education, therapeutic conversations, and resources tailored to their developmental stage and personal circumstances can empower them to navigate these aspects of their lives with confidence and resilience. Moreover, fostering open dialogue and creating safe spaces for discussing sexuality and relationships can help alleviate anxieties and foster healthy interpersonal connections among individuals. Although only discussed by two teenage participants, it may be that for many it was difficult to vocalise concerns worries of this nature. Equally, it is plausible that for younger participants, these concerns had not yet occurred to them.

Having said this, 18 individuals, half of whom had heritable Rb, had worries about having their own children and requested support for this. Seven of these individuals were teenagers between 13 and 19 years old. This suggests that seemingly 'adult' topics are being considered by young people and discussions about having future children may be easier than those about sex and relationships. These worries may stem from uncertainties about the hereditary aspects of Rb reflected by many of this group, as reflected in the below quote from someone who is at population risk of their child developing Rb due to their non-heritable diagnosis.

"if I'm being really honest, it's a big worry. I – you know, I – you're toying – you toy with the idea of whether it's a good – good idea to be pregnant in the first place and start a family, because it's so unknown." (P24, U, NH)

Teenagers, in particular, may be considering their own fertility for the first time, as well as the possibility of passing on the condition to their future children. Whether this is likely for that individual or not is another important consideration, and ties into the need for individuals to understand their own diagnosis and the implications of this. Thus, interventions addressing reproductive concerns should provide age-appropriate information and support tailored to the unique needs and developmental stage of each individual. If an intervention addresses this effectively, individuals can then make informed decisions about their reproductive health and family planning as early as possible, fostering a sense of empowerment and agency in shaping their future. It is hoped that in turn, this will enhance their locus of control and feelings of ownership over the impact of their disease, perhaps for the first time in their life. It is not surprising that individuals impacted by heritable disease were more concerned about this, with 90% expressing a need for support.

“now I’ve got to sort of think about when I have children, like do I want to get all of my embryos screened, and, you know, it was just a – a whole lot of things to think about on top of that.” (P31, U, NH)

Eighteen individuals had concerns about second cancers and wanted an intervention to help with this, with similar worries reflected across all age groups and types of Rb (55% bilateral, 56% unilateral). Rate of concern and support requests were higher amongst those with heritable disease (90%), which is reflective of the reality that second cancers are more likely for this group.

“I used to worry sometimes when I was a bit younger that – am – am I at a higher risk of having cancer again, ‘cos obviously I’ve had it once before. I don’t know whether that – to be honest, I still don’t know whether that’s true, whether – whether I’m at less of a risk or more of a risk, or just the same as everyone else, so that’s sometimes a worry. Like I do panic about my health a lot.” (P19, U, NH)

Among the young people interviewed, 20 expressed anxieties related to preparing for their future (88% bilateral, 52% unilateral; 70% heritable, 47% non-heritable). These anxieties spanned a range of concerns, from uncertainty about career choices to apprehension about everyday tasks. For one participant living with complete blindness, they were less concerned by ‘big’ ambitions and emphasised their stress over smaller and more practical concerns.

“as for worries about the future, erm, for me, it’s less about job worries, erm, because frankly I don’t know what I want to become in the future, so I’m just leaving the door open to be like, whatever happens bloody happens. But for me, what worries me are like very small little things, like, I don’t know, how am I gonna be able to catch a bus, for example.” (P6, B, NH, focus group)*

Twenty-four felt they did not have adequate understanding of Rb; this did not differ by age as 50% of whom were teenagers and 50% who were young adults. This did not also differ wildly amongst Rb type. A psychoeducational component to the intervention that is tailored to different age groups would therefore be appropriate. This could provide comprehensive information about Rb, including the different types of diagnosis, its implications, and long-term management strategies.

“obviously I know each situation’s like unique to each person and whatnot but I think maybe just some, somewhere that you can go and they’ll point you in the right direction or maybe like, “Oh you won’t need to worry about this until whenever,” or what … just something, some information like even if it’s, no matter how little it is I think would definitely help.” (P18, U, U)*

Eighteen expressed needing support for living with visual impairment, the majority of whom were teenagers. This could indicate that support and activities should be delivered at any early age, building coping skills specific to visual impairment.

"I was trying squash the other day with my mate, erm, and we were playing squash, and he was like, erm, winning quite badly, and…he said, "Is it anything to do with your eye?" And I said, "I'm not sure, it could be." And then he tried playing with one eye and he couldn't hit absolutely anything." (P4, U, H, focus group)

This might also include peer support from older survivors, who are more likely to have developed their own strategies.

"my sight is reduced in my – my right eye. So, I have full sight in my left, erm, and just – erm, it's – it's always been quite difficult, erm, kind of – this is something I've always found difficult, explaining, erm, to, er, doctors and people you're going to see, especially from a very young age. They're trying to assess how much sight you've got, and it's always quite difficult to – to kind of impart – impart that on them." (P27, B, H)

Twenty-four desired support about learning to communicate with others about Rb, again, most common amongst the younger participants. Communication support focused on helping individuals articulate their experiences and needs related to Rb would be a solution here.

"It's not even about vocabulary, it's about understanding. This is you giving, you're given this thing that you don't know what to do with it, you don't know what to make of it, you don't know how to deal with it." (P32, B, NH)*

Intervention content could include role-playing exercises and guidance on disclosing health information in social and medical contexts.

"if someone came up to someone and they didn't wanna tell then I can see how that would be quite intimidating, and maybe you – that might make them withdraw from making friends and, you know, talking to other people about it." (P31, U, NH)

This could help alleviate the fear of social withdrawal that many expressed, as well as better facilitate interactions with peers and healthcare providers.

"I feel like it's not, it's not massive what's needed you know…you know as a young person I feel like if I had just got a little, sort of like the direction you go in right, if you just kind of keep going in that direction if you're not steered otherwise. I just feel like just a little adjustment would have probably, I don't know, you never know but I don't feel like it's a massive need, it's probably just a check-in and, and an awareness, you know." (P23, U, NH)

Fifteen wanted an intervention to address the management of health conditions specific to Rb, such as dry eye.

"that was the beginning of, erm, me having severe dry eye and dealing with severe dry eye all my life, erm, and what that causes, you know, what, what that, what that means for you physically." (P32, B, NH)*

Health management sessions led by healthcare professionals or people with lived experience that provide strategies and treatments for conditions could be beneficial.

"I've got nystagmus, which, you know, makes your eyes all wobbly, erm, when you get tired, which is really weird." (P14, B, NH, focus group)*

BARRIERS TO SUPPORT

Participants discussed many different barriers to support, firstly the perception of others having it worse than they have meaning it was difficult to access interventions. Many participants made comparisons to other children undergoing treatment for Rb, but also to children diagnosed with other cancers too.

"it would almost be a bit of a, I guess a joke that I'd say, "Oh, you know, I struggled as well," sitting around with all these people that aren't as fortunate as I was." (P24, U, NH)

This concern was expressed by fewer participants overall (six of 32) and the majority of these were older and impacted bilaterally (22%). This may therefore reflect the level of support available at the time, and does not factor in initiatives which are available for children undergoing Rb treatment currently.

"I never really wanted to do that [go to cancer support groups] because I kind of – I found it a bit sad. It was a bit – as in like it would – it was kind of upsetting to me to be around other kids that also had cancer and had it worse than I did. And I think I was quite aware of that even when I was younger, that there

were people that had it worse, and I felt kind of sad about that, so I didn’t engage with that as much.” (P25, U, NH)

Several young people were uncertain about available support tools, which made them feel isolated and less able to seek out help.

“I feel really lonely and I feel really weird” (P32, B, NH)*

There was little difference between those impacted bilaterally (22%) or unilaterally (17%), heritably (20%) and non-heritably (23%).

“I had that mentality in my head that, “Okay, I’m doing this alone.” I always had my family and everything, but the main part is always dealing with it alone. So, honestly, I’ve never had the opportunity to speak to anyone relating to this circumstance. So, possibly, it could have been beneficial to me, but I’ve never like sought for it, you know.” (P17, U, H)

There were also concerns about the stigma associated with ‘needing therapy’ or psychological help.

“I know, if I ask my mum, “Hey, can I go to therapy?” Like I asked her once and her response was like, “Oh, what’s therapy gonna do to you? Just tell you that everything’s better and – and nonsense?” So, I’ve grown up with this kind of sensation of, eat it, spit it, move it.” (P6, B, NH, focus group)*

Those impacted unilaterally expressed slightly more concern about this (13% vs 11%) but the concern was still relatedly low amongst the majority.

“Now, I think, in this day and age, you will probably get a lot more buy in for what yourselves are trying to do than you would have people in my – I don’t want to say generation, I’m not old enough for that, but when I grew up. I think there’s definitely been a paradigm shift in the openness to mental health and, you know, looking for support psychologically and socially. So, I think there’s some great stuff to be done in that age group with this work, yeah.” (P16, U, NH)

PRACTICAL CONSIDERATIONS FOR INTERVENTION DELIVERY

It was almost unanimous that adolescence was considered the time of need for psychosocial support, with 81% of participants indicating that tools are required as you enter teenagehood.

"I think when, probably younger end of teenage years for me, probably would have been nice to have someone to talk." (P30, U, NH)

This was particularly felt by individuals with unilateral Rb (91%) but almost equally felt across heritability (heritable 70%, non-heritable 76%).

"I think, after the initial period of – of me feeling ashamed, which was when I was just starting to – erm, you know, to hit puberty, which is when I had my fringe and I felt really insecure and really – I think it – it would have felt really bad then." (P25, U, NH)

As discussed in study one, many expressed that it is almost 'too late' to acquire the skills during this period, due to feeling more self-conscious, and less willing to seek out and take advice, even when it is known to be needed.

"I'd say it's just like when you are like, thirteen, and like I used to say thirteen, fourteen is probably the years for me where I was like oh my god, oh my god (laughs) but it's like I think I've got to the age now I'm just like I, like, it's the last thing on my mind, genuinely." (P12, U, H, focus group)

To resolve this, participants felt that an intervention would be most appropriate and acceptable if delivered when a child is between 10 and 12 years old.

"As I perceive it, I think the main benefit would be in the adolescence, the early teens, right through, probably from age 10." (P16, U, NH)

Some felt that support should be integrated with existing appointments, with 15% of all participants expressing a preference for this, all of whom were impacted bilaterally and non-heritably.

"I guess if there was somebody maybe – I think the easiest thing for me would have been if someone was at the appointments that I physically had to go to every year no matter what. I'd be forced there by my mum and dad if I didn't want to go. And then as I got older, I needed to go and I realised that. I guess if there was somebody incorporated into the bits you already had to do, that gave you maybe an option –

everything's quite clinical, which it obviously has to be, I get that, erm, but then maybe that would make it slightly easier to open up if you wanted to, I suppose, and have that option." (P24, U, NH)

INTERVENTION DELIVERY

Participants expressed diverse preferences for how psychosocial interventions should be delivered. A significant proportion (53%) favoured modular or 'on-demand' support, allowing them to access help as needed. This flexibility was seen as beneficial for addressing their needs on their own terms:

"I think that that modular would be a good thing to offer, because then if you have a bad day and you want to, you can, or if you don't and you don't want to and you feel fine, then you don't have to." (P24, U, NH)

10 individuals had a preference for in-person interventions. They described how in-person conversations provided a greater sense of connection and clarity, particularly for those who find comfort in direct communication.

"I'm more of a face-to-face person, like I prefer sitting down talking to someone and that's just like, but obviously I know a lot of people like maybe like have a lot more anxiety doing it like that and would prefer, which I understand, but for me personally I prefer, I'm much more of a like let's sit down, talk, hash it out type of person." (P18, U, U)*

11 expressed a desire for support to be delivered as part of a group workshop. Many felt that such environments provided a more engaging and hands-on learning experience, which contributed to better retention and application of the information presented, as well as combining peer support.

"From past experiences, workshops normally help the best…With a workshop, you're kind of there. It's more hands on. You're more – you remember the stuff they tell you there more and stuff like that." (P1, U, NH, focus group)

31% desired online support, with another 33% specifically stating that videos that could be referred back to would be ideal, reflecting modern methods of seeking support and information:

"I think if like an online thing would be good for like, I don't know, like, you're just like, you're curious about it, like I know I would be 'cos like when I was like, when I got told that it was a 50/50 chance [of my children having Rb), I was like shit and I asked them more about it, erm, but then like it was like if you

did have like an online thing, you were able to like instead of having to wait for like six months or like make an arrangement with your doctor, you’re able to just search it up. So I think an online service would be good, definitely.” (P12, U, H, focus group)

Many liked the idea of having resources that could be interacted with and rewatched when suitable, which aligns with the way in which many of us now consume information.

“Or like a YouTube channel or something like that, where you have like – you have them talking or you have a video of them and stuff like that.” (P3, U, NH, focus group)

Younger participants also requested integration with social media platforms, reflecting their everyday behaviours and during on the sense of connection and community that can often be found when viewing online content created by and for a particular group of people.

“I think having like access to, say, like something on Instagram, say Facebook or TikTok would be quite nice ‘cos you could get loads of people and everything that way. But yeah, maybe things online.” (P5, U, NH, focus group)

Having said this, there was no clear-cut preference across age group or Rb type, and some individuals felt that a mixed-methods approach would be the most appropriate:

“I believe a mix would be a perfect combination, because the digital aspect can be accessed any time, you know. Even for example if there’s like a support line or a support chat, for example, for people to talk with professionals, that would surely be a great addition. If you’re facing a problem now and you feel like you want to talk to someone, sometimes talking not only to a stranger but someone who’s professional in the subject will definitely make you feel better for sure.” (P17, U, H)

FACILITATOR AND SUPPORT PREFERENCES FOR INTERVENTION DELIVERY

It was apparent through many discussions that, when an intervention is offered, the facilitator of it needs to have specific Rb knowledge.

“I think having a counsellor was good, erm, but like obviously, I wouldn’t have told her everything because she wouldn’t of known what I’ve been through and I guess that is kinda hard to explain what’s happened to someone that doesn’t know the, like, about it.” (P10, U, NH, focus group)

46% of participants highlighted this need, given the unique nature of the disease and the long-term implications on health and wellbeing:

"'cos, you know, it's nice to talk about with someone that knows about it, I guess. 'Cos if you go to like a – like a normal counsellor or like a normal doctor, the majority of the time …so it'd be nice to have someone that understands and knows what it's like, perhaps, to talk to you." (P22, U, NH)

It was also highlighted that the support should be offered by someone independent, who is not a family member or a key member of their treating medical team.

"I even found it very difficult to talk about my worries to my parents when I was a kid. I – I just couldn't talk about them. I don't know why." (P29, U, NH)

This independence was seen as vital for creating a safe space where participants could freely express their concerns without fear of judgment or impacting their ongoing relationships.

"It would have been nice to have someone that I'd probably never met before just to be like, "You know what, it's gonna be fine."" (P22, U, NH)

TABLE 11: CONTENT ANALYSIS

Participant	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27	28	29	30	31	32
	🟨	🟩	🟨	🟨	🟨	🟩	🟨	🟨	🟩	🟨	🟨	🟨	🟨	🟩	🟩	🟨	🟨	🟨	🟨	🟩	🟨	🟨	🟨	🟨	🟨	🟩	🟩	🟨	🟨	🟨	🟨	🟩
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Psychosocial support provided																																
Had therapy				X			X		X											X	X		X		X	X						X
Think therapy beneficial				X			X		X		X			X						X	X		X		X				X			X
Didn't have therapy but would have liked it	X	X	X			X									X		X		X					X			X	X	X	X	X	
Received specific Rb psychosocial support									X														X								X	
Wishes for interventions																																
Therapy/counselling							X					X					X	X	X	X	X		X	X	X			X	X			X
Talking about feelings	X	X				X	X										X		X	X	X		X	X	X	X	X	X	X			X
Interact with peers with similar history	X	X		X		X	X	X				X	X	X	X	X	X		X	X	X		X		X	X	X	X	X	X	X	X
Support to deal with:																																
• Physical appearance						X	X		X	X	X					X	X	X	X	X	X	X	X		X	X	X	X	X	X		X
• Bullying	X		X			X	X	X	X	X		X		X	X	X	X	X	X	X	X	X	X			X		X	X	X		X
• Mental health	X	X		X	X	X	X	X		X			X	X	X	X	X	X	X	X	X		X	X	X		X	X	X	X		X
• Sex and relationships			X			X										X							X	X			X	X	X			X
• Second cancers							X	X		X		X	X	X	X	X	X	X	X				X	X	X	X	X		X			X
• Having children	X		X				X		X	X		X			X	X	X	X					X	X		X	X		X	X	X	X
• Preparing for the future	X	X	X	X		X				X		X	X	X	X		X	X		X	X				X	X	X	X	X			X
• Understanding Rb	X		X	X	X	X	X	X	X	X	X	X			X		X	X		X	X	X	X	X				X	X	X	X	X
• Living with visual impairment	X		X	X	X	X	X		X	X		X	X	X	X	X		X				X						X		X		X
• Learning how to communicate with others about Rb	X		X	X	X	X	X	X	X	X		X		X				X	X	X	X	X	X		X		X	X	X	X	X	X
• Health conditions related to Rb (e.g. dry eye)	X		X		X	X	X			X				X	X		X	X	X	X	X					X						X
Barriers to support																																
Others have it worse		X																				X	X	X	X	X						
Not sure what is available						X											X		X							X		X			X	
Stigma						X																X		X					X			

Practical Intervention considerations																																
Adolescence is the time of need			X	X		X			X	X	X	X	X	X		X	X	X	X		X	X	X	X	X	X		X	X	X		X
Integrated with existing appointments																						X		X				X	X		X	
Intervention delivery:																																
• Online							X	X			X	X	X	X			X		X							X				X		
• In-person	X	X	X			X	X									X	X	X					X				X	X				
• Modular (on-demand)						X			X	X		X		X			X	X		X			X	X			X		X	X	X	X
• Workshops	X	X		X																			X	X								X
• Videos			X				X	X		X	X	X	X	X		X					X							X				
The facilitator needs specific Rb knowledge			X						X	X			X	X			X				X	X	X		X	X		X	X	X	X	X
Someone who’s not your Mum, or your doctor			X			X						X	X				X		X			X	X	X	X	X		X	X	X	X	X

Note. Orange=teenager, blue=young adult

Green= bilateral, Yellow=unilateral

Black=heritable, purple=non-heritable, red=unsure

6.8 DISCUSSION

In this chapter, the results of this study will be evaluated and analysed in the context of the research objectives and existing body of literature. I offer my insights and observations on the data, emphasising their importance and potential implications for research in Rb, as well as clinical applications, and policy development.

6.9 MAIN FINDINGS AND THEIR RELATION TO EXISTING LITERATURE

This study aimed to explore TYA Rb survivors' views on psychosocial support, including an understanding of any interventions they were offered, their wishes for future support, and any barriers to their access of this.

PSYCHOSOCIAL SUPPORT PROVIDED

Several participants reflected on their experiences with traditional therapy, like counselling or CBT. However, only nine out of thirty-two participants accessed therapy during childhood. Among these, just a third had received therapy specific to Rb, highlighting the gap in specialised support. The responses indicated that there were no significant differences in therapy access or experiences across different Rb types, highlighting a universal need. Those who had therapy generally found it beneficial, although some faced challenges with discontinuation of services, an issue which is widespread across mental health support systems (Radez *et al.*, 2021). Participants emphasised the overall value of therapy for managing both Rb-specific issues and broader personal development, underscoring the need for accessible and sustained psychological support. Conversations with TYA Rb survivors suggest that, as a group, they have distinct psychosocial support needs which must be addressed at an appropriate age and stage of life. This finding aligns with existing literature, which suggests that TYA's who are undergoing treatment for or are survivors of cancer have unique and under-recognised needs (Jin, Griffith and Rosenthal, 2021). A recent study considering these needs found gaps in information provision (Jones *et al.*, 2020). Results focused on emotional issues and how to cope with normal life stressors as well as cancer-specific difficulties such as worry about recurrence, body image concerns, and identity. These concerns were mirrored in the current study and participants outlined a desire for various support tools, including talking therapy and peer support.

Despite this, in both the current study and wider research, it has been acknowledged that adolescence is a difficult time to seek support (Aguirre Velasco *et al.*, 2020; Jones *et al.*, 2020) and can result in dissatisfaction and miscommunication with healthcare provision (Miedema, Easley and

Robinson, 2013). For this reason, there is a need for improved communication strategies between healthcare providers and survivors. This can, however, be particularly challenging for Rb due to the care pathways often followed. The majority of children diagnosed with Rb are treated whilst they are very young (Chawla, 2020); combined with a high survival rate in the UK (Zhou *et al.*, 2024), this means that many individuals are discharged from their clinical Rb team and not followed up by them until they attend a 'long term follow up' clinic at age 16 or 17 years (Jenkinson, 2015). Whilst it is positive that children are not required to attend multiple hospital visits once in remission, there is a lost opportunity for emotional check-ins. Many participants in the current study highlighted that it might be easier to access support if it was integrated into medical appointments. With the current system, this would be challenging, not only as it would require the provision of additional clinic time, which is deemed medically unnecessary, but also due to the distance that many families have to travel to one of the two national Rb centres. It is plausible that this could be addressed by child's principal treatment centre, the more local, specialist hospital site where care is often shared. Many of these sites offer annual reviews and could therefore play a key role in assessing potential psychosocial difficulties. Additionally, as highlighted by study one, many TYAs outlined experiences of medical trauma, often triggered by sensory stimuli. Therefore subjecting them to additional hospital visits may be re-traumatising and detrimental to their mental health. For these reasons, alternatives need to be developed.

Equally, the timing of the delivery of support is crucial. It was discussed by many in this study that the 'pre-teen' stage (approximately aged 10-12 years) felt the most appropriate time to offer an intervention. This is because it was felt to be significantly more difficult to ask for and accept help once you were older than this, and if you were provided with support tools prior to needing them you would be more able to cope. Considering developmental stages is important and provides an evidence-based grounding for why young people might feel this way. Often deemed as a 'critical period' of psychosocial development, primarily due to uncertainty surrounding identity which is highest between the ages of 12 and 16 years (Meeus, 2016). Although rates of maturity, empathy, and self-awareness often do not peak until older adolescence and young adulthood, if psychosocial support was not offered until then, it would likely be too late, given that a high proportion of challenges occur during the teenage years (Lawford *et al.*, 2020). Despite this, the 'pre-teen' stage could be seen as an already tumultuous time, with children facing a transition to secondary school. Existing research has highlighted that unsuccessful educational transition can lead to decreased motivation, lower academic achievement, lower life satisfaction and long-term negative mental health outcomes (Kiuru *et al.*, 2020). For these reasons, it would be imperative to consider the timing of this support. This further supports the concept of modular and 'on-demand' options for an intervention, meaning that an individual and their support network can have autonomy over accessing this, again enhancing locus of control.

DESIRED CONTENT FOR RB-SPECIFIC PSYCHOSOCIAL INTERVENTION

The current content analysis emphasised several key themes regarding desired content for an Rb-specific intervention. This included physical appearance, bullying, mental health, relationships, concerns about having children in the future, risk of second cancers, and managing health conditions specific to Rb. Each area presents distinct challenges and underscores the necessity for tailored interventions to address these multifaceted issues.

PHYSICAL APPEARANCE CONCERNS

A significant portion of participants expressed concerns about their physical appearance. Participants who experienced facial changes due to their cancer or its treatment were particularly affected, regardless of whether their Rb was heritable or non-heritable. It is well-documented that when an individual experiences changes to their appearance, whether that be due to cancer, other illness or accident, they can experience depression, social anxiety, low self-esteem, and shame (Costa *et al.*, 2014; Vaidya *et al.*, 2019; Kishore *et al.*, 2020). Furthermore, qualitative research highlights that cancer-related facial changes, such as the loss of an eye, can trigger complex thoughts about identity (Thompson, Sewards and Baker, 2020). With one in five people in the UK self-reporting a facial difference, and one in three of them reporting feeling depressed or anxious as a result (Changing Faces, 2024), there are many psychosocial interventions available that address feelings about facial changes (Bessell *et al.*, 2012; Jenkinson *et al.*, 2015; Norman and Moss, 2015; Williamson *et al.*, 2019; Matheson, Lewis-Smith and Diedrichs, 2020; Zelihić *et al.*, 2022). Existing research has highlighted the need for future research with clear theoretical underpinnings, rigor, and clinical relevance to develop effective interventions. It also suggested that future studies should consider the economic impact of interventions and design age-appropriate measures to adequately capture the experiences of living with appearance-altering conditions.

EXPERIENCES OF BULLYING

Bullying was a prevalent issue, with 23 participants recounting personal experiences. Those with bilateral and heritable Rb reported higher rates of bullying. Reports of bullying were centred around physical appearance, meaning that these groups may have had more experiences due to higher likelihood of physical differences through being affected in both eyes. As discussed above, individuals with visible difference can experience higher rates of psychological difficulty than their peers. This can be exacerbated further if they are also subjected to bullying. According to a 2021 survey conducted by ‘Changing Faces’, one in three young people with a visible difference receives hateful comments about their appearance. Additionally, one in four experience prolonged bullying, and individuals are at least three times more likely to received repeated unkind comments about their appearance via social media (Anti-Bullying Alliance, 2021). Participants in the current study stressed the need for interventions to provide strategies for managing and responding to bullying, which for most participants happened during secondary school. As is the case for many of the discussed topics, the evidence for effectiveness of existing psychosocial interventions is limited. However, a 2022 RCT evaluated social anxiety in response to a tailored web intervention to aid with psychosocial adjustment when living with an appearance-affecting condition (Zelihić *et al.*, 2022). The 'Young Person's Face IT' intervention is the only self-guided, eight-week web intervention of its' kind that is delivered in various formats, including written, audio, and video. Strategies used in the intervention include social skills training (SST) and CBT techniques, which help adolescents practice new social interaction skills and learn to challenge negative thoughts and feelings. Findings highlighted that the intervention significantly reduced social anxiety in the intervention group when compared to care as usual but did not show significant improvements in secondary outcomes such as body esteem, perceived stigmatisation, and life disengagement. Authors highlighted several implications for future interventions, importantly indicating that future research should offer interventions to pre-adolescents who are at risk of appearance-related issues in order to mitigate the onset of these and offer coping skills prior to being needed. In combination with the findings of the current study, this highlights a gap in support and offers more evidence into the timing of an Rb-specific intervention.

MENTAL HEALTH CHALLENGES

Mental health emerged as a critical area of concern, with 26 of the 32 participants indicating ongoing struggles. The desire for mental health support was higher among those with non-heritable Rb, potentially due to the greater expectation from others of 'normality' post-remission with no expected late effects and the idea that you should be 'fine', acting as a barrier to support. This poses the question as to how we support communication with 'lower risk' individuals who may not necessarily present to support services. Having said this, those with heritable Rb must consider second cancers and future genetic issues, both of which are known to impact mental health across the lifespan (Gregersen *et al.*, 2021a, 2022; Belson *et al.*, 2022). In contrast, we know that individual differences mean that some TYA do not experience mental health challenges after cancer, and this is also highlighted in literature which suggests that many long-term survivors perceive minimal impact on aspects of their lives, including potential psychological late effects (Willard *et al.*, 2017; Morse, Parris, Qaddoumi, Phipps, Brennan, *et al.*, 2023). The findings in the current study and wider Rb and childhood cancer literature indicate that an individual approach must be provided, offering the tools to all Rb survivors and giving them the opportunity to choose what is useful to them.

SEXUALITY AND RELATIONSHIPS

Concerns about sexuality and relationships were voiced by nine individuals, mostly young adults navigating these aspects of their lives in real time. These individuals suggested that interventions should include psychoeducation, therapeutic conversations, and resources to address the unique sexual and relationship challenges faced by Rb survivors. The relatively low discussion rate among teenagers may indicate a reluctance or embarrassment to vocalise such concerns or participants being at a developmental stage where these issues are less prominent. Either way, it is important that there is the option for individuals to learn about these potential issues, and due to the sensitive topic, it could be that remote provision is most suitable. As an alternative, multiple staged interventions could be provided in a modular fashion, providing access to topics as and when relevant and required, mirroring transition interventions for young people with long-term health conditions (Nagra *et al.*, 2015). Equally, utilising peer support could be equally empowering, with older survivors being able to relate to and reassure individuals who may have concerns about sexuality and/or relationships. This is an approach which has shown to be helpful amongst young cancer survivors (Zamora *et al.*, 2017; Smith-Turchyn *et al.*, 2023), with peer support positively associated with post-traumatic growth (Matsui *et al.*, 2023). Post-traumatic growth is the antithesis of post-traumatic stress and refers to positive psychological changes that are experienced as a result of highly difficult experiences (Henson, Truchot and Canevello, 2021). Signs of such growth include enhanced relationships with others, recognition of opportunities, a sense of resilience, and acceptance and appreciation of life (Jayawickreme *et al.*, 2021). Therefore, integrating a comprehensive support system that includes both professional and peer-led interventions can significantly benefit Rb survivors, promoting their overall well-being and helping them to navigate specific challenges more effectively.

REPRODUCTIVE CONCERNS

Eighteen participants, including a significant number of teenagers, expressed worries about having their own children, reflecting concerns about the hereditary nature of Rb. This anxiety was present amongst those with and without risk of future children having the condition, highlighting the need for information provision. This alone may be enough to reassure those with the non-heritable variant, allowing further time and resources to be allocated to individuals with a genuine genetic risk. In a study considering the information needs of individuals with and with genetic predisposition syndromes, it was uncovered that most found it difficult to access and find relevant information, and that information needs were consistent regardless of risk level (Metcalfe *et al.*, 2009). Although this is an older study to refer to, my research conducted 15 years later reflects the same issues. Therefore, future intervention development should provide information to all based upon perceived rather than actual risk, before targeted specific support to individuals with genuine genetic risk and reproductive concerns. It is imperative that this risk is accurately presented to each individual, and therefore must be delivered by their healthcare team. By sharing the findings of this thesis with the professionals who provide follow-up to individuals affected by Rb, I can highlight the importance of young people’s concerns about fertility, acknowledging that regardless of the variant of disease someone is diagnosed with, anxieties can still be high.

The high proportion of teenagers voicing concern about having their own children was unexpected and may be at odds with sex education provided at school, which does not always encourage open communication about fertility and often focuses on pregnancy prevention (Maslowski *et al.*, 2023, 2024). It could also reflect many other factors, these might include: the provision of information about fertility online (French *et al.*, 2022), a shift towards accessible fertility testing and social egg freezing (Jones *et al.*, 2020; Hussein, Khan and Zhao, 2020; Prior, 2020; O’Neill *et al.*, 2023), a greater awareness of fertility issues in the mainstream media (Dudouet, 2022), and conversations with healthcare professionals about the need to make fertility preservation decisions in the context of cancer (Benedict *et al.*, 2021; Newton *et al.*, 2021; Jones *et al.*, 2022); all of which have the potential to raise anxiety about promote earlier consideration of family planning. Although Rb itself only causes genuine reproductive concerns for those impacted genetically, it is also true that certain cancer treatments can cause sub-or infertility, although this is low risk for most modern Rb treatments (Cassoux *et al.*, 2017; Pawłowski *et al.*, 2023). As above, this underscores the importance of providing age-appropriate reproductive health information and support to all Rb survivors who want it, enabling individuals to make informed decisions about family planning with accurate and relevant information. This could fit into wider education for the general population, acknowledging that fertility issues affect 10-15% of couples unaffected by cancer in their lifetime, highlighting a wider issue and need for general information and support (Nik Hazlina *et al.*, 2022).

RISK OF SECOND CANCERS

Concerns about second cancers were raised by 18 individuals, with higher anxiety levels among those with heritable Rb. The heightened awareness of the increased risk of second cancers among this group highlights the need for interventions that provide clear information and support to manage these fears. Existing research has shown that providing individuals with an estimate of their cancer risk in addition to a behaviour change intervention can help them to make informed decisions about preventative care (e.g. attending cancer screening, exercising regularly, eating well, skin protection in the sun etc) (Usher-Smith *et al.*, 2018). These are all messages which are already echoed by Rb long-term follow-up clinics, but as these are usually a one-off appointment, it may be that principal treatment centres could also provide this information and motivation to survivors from a younger age and over a prolonged period of time. This support could enable individuals to manage the uncertainty that they feel but also feel motivated to take precautionary measures to maximise their chances of cancer prevention. An existing study that specifically considered the impact of second cancer risk to Rb survivors found that individuals want access to this information even if they find it upsetting (Schulz *et al.*, 2003). Whilst treatment centres will discuss risks and prevention with young people when they are seen, the findings of this thesis highlight that this information can be absorbed by parents more often than the individual themselves. Therefore, consideration must be given to locus of control, developmental stage of the individual, the timing and delivery of sensitive information about second cancers (amongst other things) as well as the means in which this information is provided. For example, if information is provided via a digital intervention or by means which are not supported by a trained facilitator, consideration must be given to signposting support in case information induces distress. This is relevant across all aspects of a future intervention and as there are now many online interventions available that cover a range of sensitive topics in the context of cancer, it is recommended that an Rb intervention learns from these (Lally *et al.*, 2019; Marzorati *et al.*, 2021; Cincidda, Pizzoli and Pravettoni, 2022; Lichiello *et al.*, 2022; Micaux *et al.*, 2022).

FUTURE UNCERTAINTIES

Anxieties related to preparing for the future were significant, particularly among those with bilateral and heritable Rb. Participants expressed concerns ranging from career choices to practical everyday tasks, reflecting both the small and often overlooked challenges but also the big life events. This suggests that interventions should offer comprehensive support that addresses a spectrum of topics relevant to those with Rb, fostering independence and confidence e.g. existing support provided by RNIB. There is extensive literature on the topic of cancer-related uncertainty, but much of this is focused on fear of recurrence, as discussed above (Tutelman and Heathcote, 2020). Quality of life (QoL) is a common measure in cancer survivorship literature, with research suggesting that this can be impacted many decades after a diagnosis (van Leeuwen *et al.*, 2018; Liu *et al.*, 2020). Even though there is some QoL literature specific to Rb (Belson *et al.*, 2020, 2022), it does not capture the findings of the current study, which emphasises the nuanced and multifaceted nature of the uncertainties experienced by those living beyond Rb. This again underscore the need for tailored interventions acknowledges common worries about everyday life and the future. There is also role for peer support here, drawing on the topics raised in the current study to enhance survivors' overall well-being.

UNDERSTANDING OF RB

A lack of understanding about Rb and their personal diagnosis was noted by 24 participants, indicating a need for psychoeducational components in interventions. Providing comprehensive, age-appropriate information about Rb can empower individuals with knowledge about their condition and its long-term implications. As discussed above, it is not unusual for individuals diagnosed with health conditions at a young age to feel that they lack knowledge and understanding. This may be due to the need for health care professionals to primarily explain a diagnosis to parents, as well as developmental understanding and a young child's ability to comprehend the extent of their condition. This can be even more likely for children with diagnosis like Rb, who in some instances can be quickly free of treatment and somewhat removed from the hospital environment as they grow up. There is therefore a lack of literature in this area, in comparison to children diagnosed with chronic conditions such as type one diabetes or epilepsy, where they are more likely to be actively engaged in treatment management (e.g. taking medication or administering injections) and therefore gaining knowledge independent to their parents (Lewis and Parsons, 2008; Freeborn *et al.*, 2013; Pauschek *et al.*, 2016; Martin *et al.*, 2017). This can also be true for individuals who live with a prosthetic(s) as a result of Rb, with self-care meaning that they develop extensive experiential experiences in this regard (Moses, Flegg and Dimaras, 2020). Having said this, the current findings emphasise a need for greater psychoeducation around the nature of the condition, including how it might affect them in the future. Developing tailored educational resources and a robust support system could bridge this knowledge gap, ensuring that individuals affected by Rb are well-informed and better prepared to manage their health and well-being throughout their lives, including the lives of any future family members who may also be impacted.

LIVING WITH VISUAL IMPAIRMENT

Eighteen participants, primarily teenagers, expressed the need for support in living with visual impairment. Again, any intervention must provide a variety of information and support tools, as each person will be impacted differently and indeed, some Rb survivors have no difficulties with their vision. Learning can be taken from wider vision literature and the psychosocial interventions already available. A systematic review of trials investigating psychosocial interventions to improve the mental health of those with visual impairment found that there are many negative consequences of having visual difficulties, including loneliness, depression, mental fatigue and anxiety (van der Aa *et al.*, 2016). Having said this, the paper acknowledged a lack of good-quality studies included, meaning that future reviews are needed to make appropriate recommendations for interventions. It also highlighted that many interventions for visual impairment are targeted towards older people, again highlighting a gap for young people like those in the current study. A review focused specifically on children found that there were several interventions that have the potential to positively impact functioning, participation, and quality of life (Elsman *et al.*, 2019). The elements of the effective interventions included group-based programmes focused on locus of control, self-esteem, attitude towards visual impairment, and self-determination. Despite this, the review had mixed results with some interventions showing no significant change across the above outcomes, highlighting that future research must consider using robust methods to provide definitive evidence to inform future interventions. Considering the current findings against existing literature, it can be concluded that interventions, including peer support and visual skills development, can help individuals build effective coping strategies and manage the emotions associated with living with visual impairment.

COMMUNICATION SKILLS

Support for learning to communicate about Rb was a priority for 24 participants, predominantly younger individuals. These participants expressed a strong need for effective strategies to articulate their experiences and challenges related to their diagnosis. The emphasis on this support highlights the importance of equipping young patients with the tools necessary to navigate conversations about their condition confidently and comfortably. Effective communication can significantly impact their social interactions and overall well-being, underscoring the value of targeted interventions.

Interventions designed to enhance communication skills could include role-playing exercises and guidance on disclosing health information in social contexts. Role-playing exercises provide a safe and controlled environment for individuals to practice and refine their communication techniques. By simulating real-life scenarios, these exercises can help participants develop the confidence to discuss their condition openly with friends, family, and healthcare providers. Additionally, providing structured guidance on how to disclose health information can alleviate the anxiety and fear associated with these conversations. This support can reduce the risk of social withdrawal and improve interactions with peers and healthcare providers, fostering a more inclusive and understanding social environment. These are strategies that has been found to be effective in other contexts, including for young people (Rosselet and Stauffer, 2013), individuals with learning disabilities (Lewis *et al.*, 2013) and autism (McCoy *et al.*, 2016). This can be done in person or online, with an emphasis on the efficacy of ‘video modelling’ meaning that young people can learn social and communication skills from the comfort of their own homes (McCoy *et al.*, 2016). This could be beneficial to draw upon in a future Rb intervention, given the wide geographic spread of individuals affected, and the ‘on demand’ nature that young people are now so accustomed to.

It is, however, difficult to consider communication skills without acknowledging the impact of others, as discussed regarding bullying. The social dynamics surrounding young individuals with Rb often include encounters with bullying or insensitive behaviour and comments from others. Such experiences can profoundly affect their willingness and ability to communicate about their condition. Addressing these challenges requires a comprehensive approach that may include educating peers and promoting empathy and acceptance, although this is likely to be outside the scope of one intervention. Ideally, interventions should not only focus on the affected individuals but also aim to create a supportive community that discourages bullying and encourages open, respectful dialogue. Realistically, a future intervention for survivors could focus on assertive communication skills and delivery of health-related information.

Management of Related Health Conditions

Fifteen participants highlighted the need for support in managing health conditions specific to having had Rb, such as dry eye and nystagmus. Health management sessions led by healthcare professionals or individuals with lived experience could provide valuable strategies and treatments for these conditions. Peer-delivered psychoeducation is a concept that is being increasingly seen on social media platforms such as TikTok, both within cancer but in other conditions too (Basch *et al.*, 2022; Chen, Pan and Zuo, 2022; Song *et al.*, 2022; Xue *et al.*, 2022; Morton *et al.*, 2023; Anderer, 2024). Accessing health information in this way can make complex health topics more relatable and understandable for young people, as they receive information in a format and language that is normal for them, and that they may find engaging and accessible. Additionally, peer-shared content can foster a sense of community and support, encouraging young individuals to seek help and share their experiences openly (Kirkpatrick and Lawrie, 2023). Having said this, it is known that accessing health information and social support via these platforms can perpetuate the spread of misinformation, be influenced by commercial bias, and cause harm to the viewer (O’Sullivan *et al.*, 2022; Siegal *et al.*, 2023). It is therefore important that the development of a future Rb intervention learns from the positives of peer and online provision but is also based on evidence-based information that is delivered accurately.

Intervention Delivery

Participants expressed the need for a mixed-methods approach combining digital and in-person support, and there was a preference for flexible, modular support that can be accessed on demand, both online and in person. As discussed in study two of this PhD, which considered psychosocial interventions for young survivors of a variety of childhood cancers, this is feasible, with remote and in person support offered over periods of up to 52 weeks and often using customised modules to target specific issues e.g.

(Berg *et al.*, 2020). A recent evaluation of an in-person psychosocial support program for young adult childhood cancer survivors offered a targeted programme to develop three key ‘life skills’ (Pletschko *et al.*, 2023). Focusing on self-perception, social interaction and conflict management, and self-conscious communication of support needs over a 3-day workshop, participants self-reported positive influences in their behaviour and higher levels of acceptance and self-confidence. Additionally, a study for TYA childhood cancer survivors showed that when offered an online psychoeducation CBT and ACT based intervention, participants showed short-term decrease in distress and improvements in self-efficacy (van Erp *et al.*, 2023). This technique has also been shown to be effective in the management of psychosocial needs of families with a child with a chronic condition (Day *et al.*, 2020; Li, Douglas and Fitzgerald, 2023; Jones *et al.*, 2024). Although these studies individually have relatively small sample sizes, the combined effects highlight the efficacy of many different psychosocial interventions. Considering the positive outcomes demonstrated in these studies, implementing a modular approach for psychosocial support among Rb survivors appears acceptable to survivors whilst also being informed by the evidence base. The flexibility of modular support, accessible both online and in person, allows for tailored interventions that can adapt to individual needs and schedules. The integration of structured, therapeutic elements can provide a comprehensive framework that addresses a range of psychosocial issues, something that is crucial when trying to support so many needs and experiences. Furthermore, the evidence supporting the efficacy of both short-term and sustained interventions underscores the potential for significant, lasting improvements in participants' well-being.

FACILITATOR AND SUPPORT PREFERENCES

The current analysis highlighted participants' preferences for facilitators who have expertise in Rb, due to the unique nature of the disease and its’ long-term impacts on health and wellbeing. Nearly half of participants discussed the necessity for facilitators with specific Rb knowledge to ensure relevant and empathetic support. This finding is replicated across rare disease literature, both in paediatrics and in the adult population. A systematic review of 21 studies found that it was common for adults to feel uncertain about clinician’s knowledge about their diagnosis, and this contributed to the development of psychological distress, loss of confidence, and anxiety (von der Lippe, Diesen and Feragen, 2017). Another study, aptly named ‘common needs in uncommon conditions’ considered individuals with rare diseases across the lifespan highlighted that the patient is often the expert in their own diagnosis, and as such feel frustrated when professionals lack understanding (Smits *et al.*, 2022). The study also emphasised the need for empathic communication and knowledgeable delivery of information, supporting the views in the current research.

Additionally, the need for independence in support provision was evident, as many participants felt uncomfortable discussing their worries with family members or their primary medical team, some of whom may have known them since they were young children. An independent facilitator trained in specific psychosocial support was seen as crucial for providing unbiased and confidential support. This is important for many reasons, firstly, it is common for young people who have undergone cancer treatment to feel dependent on their caregivers for longer than their peers (Brier *et al.*, 2011; Smits-Seemann *et al.*, 2016). Equally, there is often a lack of decision-making for the individual whilst undergoing treatment (Davies, Kelly and Hannigan, 2015), even more so if they are very young like many Rb survivors. Having said this, one study found that Rb survivors rated themselves to have higher autonomy than survivors of other types of cancer, so this may be less of an issue in this population or for childhood cancer survivors generally as they mature (Jervaeus *et al.*, 2014). One European centre has highlighted a role for an 'Rb coordinator' who has knowledge of the healthcare system, was up-to-date with Rb knowledge and emerging evidence, and who was equipped to support survivors holistically (Gregersen *et al.*, 2021b). It could be that the UK adapts this model, with a coordinator providing a key role in psychosocial support.

Identified Gaps and Unmet Needs

The study identified significant gaps in the availability and delivery of psychosocial support services, confirming what was known prior to this study being conducted. Many participants noted the lack of specialised Rb psychological support and the inconsistency in therapy availability. The findings pointed to a broad recognition of the need for comprehensive, ongoing support tailored to the unique experiences of Rb survivors. Recommendations included developing targeted psychoeducation interventions and ensuring consistent access to specialised psychological services to address both immediate and long-term psychosocial needs. As discussed above, this can be particularly difficult to provide to Rb survivors, given the nature of their care provision and a lack of medical need for them to be seen regularly once they are in remission. Although there are now support tools available for children and parents whilst they undergo Rb treatment e.g. (Boujabadi *et al.*, 2021; Gibbs, Reynolds and Shea Yates, 2022; Rich *et al.*, 2024), these were not available when the cohort involved in this study were young. Equally, there is no known, Rb-specific psychosocial support intervention targeted towards survivors as they grow older. The identified gaps emphasised in this PhD provide greater support for the provision of a novel psychosocial intervention, a project which I hope to develop from this evidence post-doctorally.

6.10 STRENGTHS AND LIMITATIONS

This study provides an in-depth analysis of the psychosocial support needs provided to and wanted by Rb survivors, including their hopes for future intervention development. This is the first known study of its' kind and underscores the long-term psychological needs of this population. This is consistent with wider literature that discusses the enduring effects of childhood cancer on survivors' mental health and quality of life, but includes the specific needs of Rb, in addition to wider survivorship needs. This study therefore addresses a gap in the literature concerning a relatively understudied population in cancer survivorship research. As described in the study one discussion (chapter 3; where the data were collected), the sample size of participants was large compared to the number of individuals affected by Rb. It is logical to assume, therefore, that the views reported in this analysis are reflective of many Rb survivors' needs. Although qualitative research does not aim to be generalisable, it is important a comprehensive and nuanced understanding of individual experiences are described. The diversity of the sample enhances the credibility and transferability of the results, meaning insights could inform a future intervention for this group. It is also plausible that these findings could be applied to similar contexts or populations, where young people are affected by other rare cancers, genetic predisposition syndromes, hereditary diseases, or have visual differences, thereby contributing valuable knowledge beyond Rb.

This study used a retrospective content analysis, analysing data originally collected for study one and focusing specifically on Rb survivors' psychosocial support needs to inform future intervention development. Strengths of this method include providing rich insights into personal experiences, drawing on existing data to enhance the credibility and transferability of the findings. As both study one and study three are qualitative, they focus specifically on the lived experience of this population. This group lives with the consequences of their diagnosis and treatment on a daily basis and are therefore considered experts on helpful intervention content. Additionally, the systematic approach of content analysis ensures objectivity and replicability, strengthening the reliability of the original data for informing targeted and effective psychosocial interventions. Content analysis can be deemed 'small q' in the qualitative research paradigm, given its' focus on quantifying themes through coding (Elo *et al.*, 2014). This is positive in that it allows for large quantities of data to be considered efficiently, enabling researchers to identify and analyse widespread patterns and trends. This is appropriate for this research question as I wanted to establish the specific needs of survivors and match these to future intervention content.

However, there are also weaknesses of this method that must be considered. The reliance on existing data means that I was constrained by the scope and quality of my original data collection, which was not specifically tailored to the research questions of the current study. In terms of interpretation of responses, because participants weren't specifically prompted on all elements, a lack of discussion about a particular issue did not necessarily mean it would not have been wanted, rather that it was just not brought up organically.

I did, however, collect the primary data myself for the purpose of an Rb-specific PhD, asking each participant about interventions generally. Additionally, the focus on quantifying themes might overlook the nuanced, context-dependent aspects of the data that are often the focus of more in-depth qualitative analysis. The coding process, while systematic, can also introduce bias if not carefully managed, as the interpretation of data can be influenced by my own perspective. Finally, since content analysis often emphasises patterns and frequencies, it is plausible that it may miss out on the complexities and unique variations within individual experiences.

6.11 IMPLICATIONS

The findings from this study highlight the elements of psychosocial support which are wanted by young Rb survivors, including content of a future intervention and preferences regarding method and mode of delivery. Combined with the wider content of this thesis, these novel findings provide a framework for the development of a psychoeducation intervention. Once developed and tested empirically, insights from this study could be implemented into routine clinical practice, upskilling healthcare providers to recognise and address the psychosocial needs specific to this group. It is hoped that this will have benefits at individual and systemic level, through offering preventative support to reduce future distress, but also offering coping skills to those who may already be managing mental health challenges in the context of Rb. This will, in turn, save money for the NHS by reducing onward referrals to mental health services, easing the burden on already stretched resources. Additionally, by embedding psychosocial support within standard care, there will likely be an improvement in the overall quality of life for Rb survivors. This approach can serve as a model for other rare childhood conditions, further enhancing the scope and impact of integrated mental health interventions across various healthcare settings. Furthermore, this study could be of benefit to policymakers in their consideration of developing guidelines and funding opportunities to support the implementation of an intervention for Rb. By prioritising funding for such interventions, policymakers can ensure that targeted psychosocial care becomes a standardised component of the treatment pathway for Rb survivors, potentially influencing national health policies. Moreover, the evidence generated could also advocate for the expansion of support services within other paediatric oncology and genetic disease frameworks, thereby reinforcing a holistic approach to healthcare that recognises and addresses the long-term psychosocial impact of living with and beyond a life-threatening diagnosis. This is particularly relevant for those who are diagnosed very young and are developing through childhood, adolescence, and beyond. There is a need for these individuals to constantly reassess the long-term impact of their condition, revisiting issues that might be affecting them at different times in their lives. Physical, cognitive, social, and emotional abilities develop extensively across this time (Rutter and Sroufe, 2000) and therefore information needs and support requirements will differ extensively. In considering all of the above, policy efforts can align with the broader goals of public health, ensuring equitable access to comprehensive care that meets the diverse needs of all young people across all developmental stages.

6.12 RECOMMENDATIONS FOR FUTURE RESEARCH

Future research should build on the current findings to co-design an intervention for this group, testing the efficacy of different intervention content and delivery methods. It should also draw on psychological theory and behaviour change methods, as well as be informed by discussions with young Rb survivors. This should be done by conducting a feasibility trial to assess efficacy before conducting a full-scale trial. Additionally, larger scale quantitative or mixed-methods research could be conducted to assess the wider, long-term psychosocial effects of Rb. This could be conducted across different countries and cultures to ascertain the psychological similarities and differences of Rb survivors, and feed into intervention development to avoid Eurocentrism.

In addition to this, further exploration into the longitudinal outcomes of psychosocial interventions for Rb survivors is crucial. Future studies should consider implementing longitudinal designs that follow survivors over extended periods, to evaluate the sustained impact of these interventions on mental health, quality of life, and social functioning. There is also a need for research that investigates the role of family, clinical, and community support systems in the psychosocial well-being of Rb survivors, to determine how these networks can be leveraged within interventions. Furthermore, the integration of emerging technologies, such as digital health tools and telemedicine, offers an innovative role for expanding the reach and accessibility of support services. In line with the NHS Long-Term Plan, research could explore the potential of these technologies in delivering personalised, scalable interventions that can adapt to the evolving needs of survivors as they transition from childhood to adulthood. Lastly, there is an opportunity for collaborative research initiatives that involve stakeholders from diverse sectors, including healthcare providers, educational institutions, and advocacy groups. Such partnerships could facilitate the development of multidisciplinary approaches to care, ensuring that future research and subsequent interventions are holistic, culturally sensitive, and capable of addressing the complex challenges faced by Rb survivors across their lifespan.

6.13 SUMMARY

This study provides novel insights gained from a retrospective data analysis of intervention content specific to teenage and young adult survivors of Rb. Based on the data gathered, several recommendations can be made for a future psychosocial intervention for TYA survivors of Rb:

1. **Early Intervention**: Initiate support programs at a young age (10-12 years) to build coping skills before adolescence.
2. **Tailored Support**: Develop interventions that are specific to the unique needs and experiences of Rb survivors, including those impacted bilaterally, unilaterally, heritably, and non-heritably. Incorporating a range of topics and preferences as identified in this study.
3. **Peer Support and Mentorship**: Incorporate peer mentorship to provide relatable guidance and emotional support from individuals with lived experience. This could be influenced by principal treatment centre's long-term follow-up clinics, who could facilitate groups of young people of similar ages and experiences within a given geographical area.
4. **Professional Training**: Ensure facilitators have specific knowledge of Rb and its impacts to provide empathetic and relevant support.
5. **Flexible Delivery Methods**: Offer modular support to accommodate different preferences and needs.
6. **Practical Skills Training**: Include sessions focused on practical life skills and adaptive strategies to facilitate coping.
7. **Regular Check-Ins**: Provide ongoing, consistent support with flexible and 'on demand' access to address both immediate and long-term needs.

As summarised above, Rb survivorship experiences are complex, and appropriate interventions must be developed to support this. The findings from this study are imperative for informing clinical practice and intervention development and highlight the need for a collaborative effort between researchers, clinicians, and survivors to develop and implement such interventions effectively.

CHAPTER 7: DISCUSSION AND CONCLUSIONS

7.1 INTRODUCTION

This chapter provides an overview of the findings presented in the three studies undertaken as part of this thesis, considering the implications that they have on clinical practice, policy, and future research. The main aim of this thesis was to understand the experiences of young Rb survivors in order to generate evidence for a future psychologically informed, educational intervention to support this group. As stated in chapter one, this was broken down into the following objectives:

1. To understand the views of teenagers and young adult survivors of Rb regarding their psychosocial needs
2. To explore Rb-associated psychosocial challenges arising in adolescence/young adulthood
3. To seek the opinions of teenagers and young adults regarding the support they had/would have liked
4. To identify potential content of a psychosocial intervention through qualitative interviews and existing cancer interventions
5. To draw upon the above to generate robust evidence to inform a novel psychosocial intervention

7.2 SUMMARY OF THESIS AND NOVEL CONTRIBUTIONS

Retinoblastoma (Rb) is an aggressive eye cancer with an incidence of approximately 1 in every 18,000 live births globally (Byroju *et al.*, 2023). In 95% of cases, this rare cancer is diagnosed in children under five years old (Kivelä, 2009) and can occur due to either heritable or non-heritable (spontaneous) factors, with an approximately 50:50 split amongst those diagnosed (Richter *et al.*, 2003). For children with the heritable form, a germline mutation of the RB1 gene means that they are susceptible to second cancers throughout their lifetime, and must consider that every child that they go on to have has a 50% chance of the same diagnosis (Bouchoucha *et al.*, 2023). For individuals who have the non-heritable variant, the disease has occurred due to two random mutations in the RB1 gene, it is unlikely that future family members will be affected, and the individual is at population-level risk of future cancers (Schonfeld *et al.*, 2021). Regardless of type of Rb, a child can be impacted unilaterally (in one eye), bilaterally (both eyes), or rarely, trilaterally, where disease has spread intracranially to the pineal gland at the base of the brain (Pareek *et al.*, 2024). Whilst global survival rate is low at 30% (Global Retinoblastoma Study Group, 2020), advances in screening and effective treatments mean that children in the UK have a near 100% survival rate (Dimaras *et al.*, 2015); this is the population focus of this PhD thesis.

When a child is diagnosed with any form of cancer, the impact on them and their family can be profound (Bakula *et al.*, 2020; Peterson, Chung and Barrera, 2020; Darlington *et al.*, 2021; Iragorri *et al.*, 2021). This can be exacerbated further when a diagnosis has a genetic element to it, meaning that life-long challenges are likely to be present (Frebourg *et al.*, 2020; Barnett *et al.*, 2022). Whilst there have been studies into the psychosocial outcomes for individuals and families affected by Rb (Van Dijk *et al.*, 2007; van Dijk *et al.*, 2009, 2010; Ford *et al.*, 2015; Soliman *et al.*, 2017; Gelkopf *et al.*, 2019; Gregersen *et al.*, 2021a; Belson *et al.*, 2022; Morse, Parris, Qaddoumi, Phipps, Brennan, *et al.*, 2023), there is no routine psychosocial support tailored to this group; this is the gap that this PhD thesis has aimed to fill.

This thesis used multiple methods to undertake three studies to generate evidence to inform a psychoeducation intervention for young people who have had Rb. In study one, I aimed to explore the experiences of teenage and young adult Rb survivors, including their psychosocial needs and challenges. In study two, I systematically examined the literature to explore existing psychosocial interventions designed for teenage and young adult survivors of any childhood cancer. I sought to understand the types of interventions that have been trialed, their content and methods, and how effective (or not) they were at impacting the wellbeing of survivors. I opted to broaden these interventions to childhood cancer survivors generally, given that there are no empirically tested interventions available for Rb. Lastly, in study three I conducted a retrospective content analysis of a subset of data collected as part of study one. The focus of this study was to consider survivors' experiences of psychosocial support, including desired intervention content for the future and practical considerations of this. The full studies can be found in chapters three, five, and six respectively, and are summarised below.

Study 1: Qualitative Exploration of Retinoblastoma Survivors' Experiences

Study one aimed to understand the lived experiences of Rb survivors aged 13-29 years using reflexive thematic analysis. This was a qualitative exploration of the experiences of Rb survivors, focusing on their psychosocial experiences. It involved individual interviews and focus groups with adolescent and young adult survivors respectively. Overall, 32 young people took part and there was a unanimous sense that having had Rb had significantly impacted their life, both positively (e.g. post-traumatic growth and refined coping skills), and negatively (e.g. long-term distress and heightened anxiety).

Through three themes and eight subthemes, findings highlight a significant need for psychosocial support tailored to different life stages, from as early as primary school through to young adulthood. This support should address issues like coping with bullying, exploring identity, and planning for the future.

The implications of these findings are substantial for NHS policy and practice, emphasising the need for integrated psychosocial support in long-term follow-up care, which aligns with the NHS long-term plan. I am aware that it is unusual to conduct a qualitative study prior to a systematic review. However after a scoping search of the literature and finding gaps in terms of young Rb survivors experiences and specific interventions for this population, it was important for me to lead with the voices of lived experience. Once I had a grounding in this, it informed my choice of systematic review question for study two. Additionally, due to the unexpected over-response to recruitment in study one, and the amount of rich data collected, I decided to focus specifically on young people's experiences. It was very important to me to do justice to the narratives and ideas of the people involved, which led me to developing a separate study (study three) to conduct an in-depth analysis of psychosocial support needs.

Study 2: Quantitative Systematic Review of Psychosocial Interventions for Teenage and Young Adult Cancer Survivors

Study two comprised of a systematic review that examined the efficacy of psychosocial interventions in improving the psychosocial wellbeing of teenage and young adult (TYA) cancer survivors. The review included 15 RCTs with a total of 1,109 participants aged 8–39 years, including young people with a range of cancer diagnoses, including Rb. The interventions took places in locations worldwide, including the USA (n=10), China (n=2), and Australia, Turkey, and the Netherlands (n=1 each).

The review aimed to synthesise the main outcomes and trends of these interventions and highlighted varied programmes which positively impacted the mental well-being of teenage and young adult (TYA) cancer survivors. Positive effects on mood were observed, particularly from interventions involving physical activity, meditation, and peer helping with expressive writing. These findings suggest that physical and psychosocial interventions can enhance mood among TYA cancer survivors, though variability in study designs, sample sizes, and participant characteristics may have influenced these outcomes. Mixed results were seen in behavioural interventions, with some studies showing no significant differences in physical exercise outcomes, while others demonstrated improvements in health promotion behaviours. The review also found that interventions positively impacted self-efficacy, particularly technology-based and physical activity interventions, which enhanced TYA's beliefs in their emotional and coping abilities. However, the long-term sustainability of these positive impacts needs to be assessed in future research to determine long-term benefits. Improvements in coping skills were noted across psychoeducation programmes, highlighting the importance of information and support tools for TYA cancer survivors. Identifying the specific components contributing to improved outcomes is essential for developing targeted and effective interventions. Additionally, interventions targeting social support, such as prosocial activities and physical exercise, showed positive impacts, underscoring the role of social support networks in enhancing overall well-being. Various other outcomes, such as hope, self-esteem, uncertainty, sleep quality, and resilience, were also reported, reflecting the complexity of TYA survivors' experiences and the nuances needed to tailor interventions.

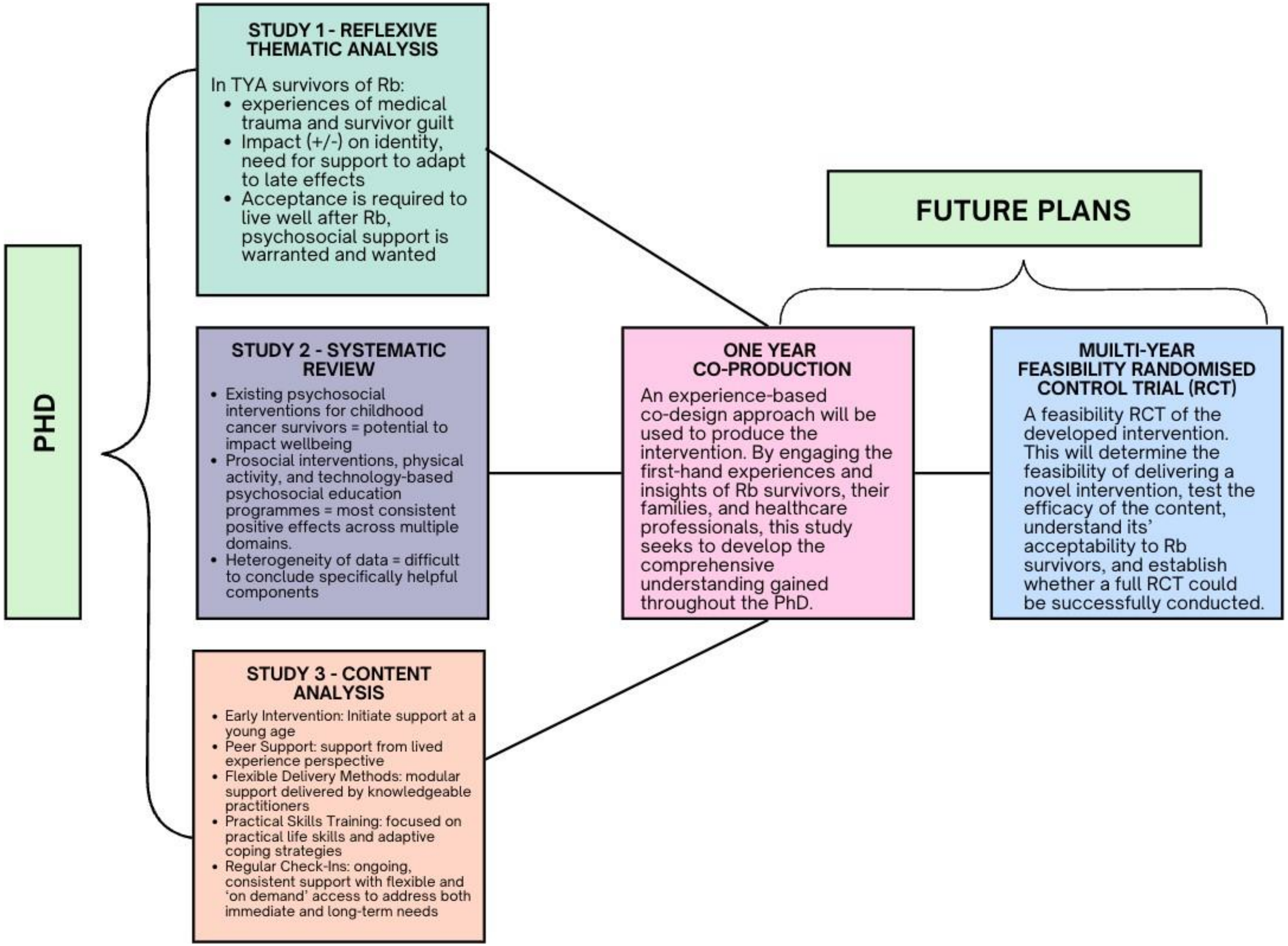
Despite positive findings it is imperative to note that the heterogeneity in outcome measures and intervention types, along with the lack of demonstrated reproducibility, led to an inability to conduct a meta-analysis as well as posing many additional challenges in drawing definitive conclusions to their efficacy. The variability and clinical diversity in participants, interventions and outcomes, means that the true effects and clinical impact of the interventions are difficult to interpret, and this should be held in mind when using these as a basis for a future Rb intervention. Future research should strive to use standardised outcome measures and consistent sample sizes, compared by diagnosis and age of cancer experience, to enhance comparability and allow for meta-analyses. Additionally, follow-up studies are needed to assess the sustainability of intervention effects long-term. The absence of explicit reporting on adverse effects also emphasises the importance of systematically evaluating and reporting both positive and negative outcomes in future research. Lastly, given the British population and treatment context of the rest of this thesis, it is notable that no studies conducted in the UK could be included in this review. After considering why this might be, I believe it could be due to the sole inclusion of interventions evaluated by RCT. I have no doubt that psychosocial support is offered to TYA in the UK, and at conferences have heard about such programmes running very successfully (e.g. ‘MOVE Against Cancer’, an intervention endorsed by the NHS and many charitable organisations (MOVE Against Cancer, 2024)). It is, however, notoriously difficult to secure funding for large scale trials on psychosocial topics, and although this may be changing, it may explain why UK-interventions were not identified within my inclusion criteria. With this in mind, this review intended to provide an overview of psychosocial support and can act as a guide, along with the qualitative findings from studies one and three, to develop an Rb-specific intervention.

Study 3: Content Analysis of Retinoblastoma Survivors' Psychosocial Support Needs

The final element of this thesis, study three, was a retrospective content analysis of a sub-set of qualitative data from study one, focusing specifically on the psychosocial support needs of Rb survivors. It aimed to analyse survivors' experiences with psychosocial support, the interventions they have received or would like in the future, and the practical considerations of developing and delivering such interventions. The study also sought to understand the perceived effectiveness and adequacy of current support systems from the survivors' perspectives and to identify gaps in existing services to inform the development of a targeted psychoeducation intervention. This study highlights several key findings regarding the psychological and emotional support needs of young cancer survivors. Participants emphasised the critical importance of psychological support during both pre-adolescence and adolescence, suggesting that early intervention could mitigate later psychological issues. Integrating this approach into routine clinical practice would ensure continuous support that evolves with the survivor's needs. Participants advocated for ‘modular’ support, accessible at a time that could be tailored to their wants, needs, developmental stage and maturity. Providing an intervention in this way also gives an element of ‘control’ back to the survivor, something which is important in terms personal growth and wellbeing. This is because undergoing a cancer diagnosis and treatment, as well as living with potential life-long late effects and uncertainty, can lead to individuals feeling they lack control and have little choice. In the stereotypical medical model of ‘doing to’, which is often a necessity when it comes to life-saving treatment. It is imperative that this is balanced with choice when considering psychosocial needs and how support is accessed.

Overall, these findings and implications highlight the importance of a comprehensive and inclusive approach to the psychosocial care of Rb survivors and provide a clear roadmap for the future development of an intervention for the UK Rb population. Whilst acknowledging the current limited resources in NHS funding, it is hoped that this can be utilised before later being tested in other countries and cultures, and act as a model for other rare cancers, cancer pre-disposition syndromes, genetic diseases, and conditions that cause facial changes or sight difficulties. Moving forward, future research should explore the scalability and adaptability of such interventions across different healthcare systems and settings. This would involve examining how local resources, cultural perceptions of mental health in the context of physical illness, and healthcare infrastructure impact the implementation and success of the interventions. Further collaboration with international research bodies and healthcare organisations could foster a more robust understanding of these variables and facilitate the sharing of best practices across different countries, cultures, and healthcare systems. An interdisciplinary approach that includes insights from psychology, public health, and medical ethics will be crucial in ensuring that the interventions are not only evidence-based but also ethically sound and culturally sensitive. By establishing a foundation of evidence through rigorous research and fostering global collaboration, future interventions could set a precedent for psychosocial care in the broader context of paediatric oncology and beyond, potentially influencing global standards for the care of young cancer survivors.

In summary, this thesis has outlined the psychosocial needs of young Rb survivors in the UK, evaluated the key components of existing support interventions for wider cancer survivors and highlighted the areas of focus and practical considerations for a future Rb intervention. This work evidences the need and want for an intervention, and how this would fit realistically within existing NHS and wider support structures, particularly given limited public sector resources at present. It was outside of the scope of this thesis to design the intervention, yet this is something I plan to do post-doctorally, with support-in-principal from my current funder and a grant application submitted. Please refer to figure 12 for a diagrammatic overview of the findings of this thesis, and how this links to future plans and implications for the Rb community.



STUDY 1 - REFLEXIVE THEMATIC ANALYSIS

In TYA survivors of Rb:
- experiences of medical trauma and survivor guilt
- Impact (+/-) on identity, need for support to adapt to late effects
- Acceptance is required to live well after Rb, psychosocial support is warranted and wanted

STUDY 2 - SYSTEMATIC REVIEW

- Existing psychosocial interventions for childhood cancer survivors = potential to impact wellbeing
- Prosocial interventions, physical activity, and technology-based psychosocial education programmes = most consistent positive effects across multiple domains.
- Heterogeneity of data = difficult to conclude specifically helpful components

STUDY 3 - CONTENT ANALYSIS

- Early Intervention: Initiate support at a young age
- Peer Support: support from lived experience perspective
- Flexible Delivery Methods: modular support delivered by knowledgeable practitioners
- Practical Skills Training: focused on practical life skills and adaptive coping strategies
- Regular Check-Ins: ongoing, consistent support with flexible and 'on demand' access to address both immediate and long-term needs

ONE YEAR CO-PRODUCTION

An experience-based co-design approach will be used to produce the intervention. By engaging the first-hand experiences and insights of Rb survivors, their families, and healthcare professionals, this study seeks to develop the comprehensive understanding gained throughout the PhD.

MUILTI-YEAR FEASIBILITY RANDOMISED CONTROL TRIAL (RCT)

A feasibility RCT of the developed intervention. This will determine the feasibility of delivering a novel intervention, test the efficacy of the content, understand its' acceptability to Rb survivors, and establish whether a full RCT could be successfully conducted.

FIGURE 12: OVERVIEW OF THESIS FINDINGS, FUTURE PLANS, AND IMPLICATIONS FOR THE FUTURE

CHAPTER 8: IMPLICATIONS OF THIS THESIS

I have conducted this thesis with the ambition of creating impact through generating evidence to support future changes to the psychosocial care of children and young people who have survived Rb. This is based not only on research (my own and the wider literature on this topic) but also on my clinical practice as a psychologist, in which I support individuals living with and beyond cancer to manage complex mental and physical health challenges.

Through three interlinking studies, I have demonstrated that there is a gap in provision for survivors of Rb, a group who have specific support needs that, if left unmanaged, can lead to high levels of distress across the lifetime. The entirety of this thesis has been informed by PPI, and the voices of those with lived experience underpin every implication that I discuss in this section.

8.1 IMPLICATIONS FOR CLINICAL PRACTICE

Both of my qualitative studies provide in-depth insight into the psychosocial experiences of Rb survivorship and the wants and needs for support interventions to manage these. My findings hold significant implications for enhancing NHS policy, particularly in long-term psychosocial care. By providing a comprehensive understanding of the profound psychological and emotional impacts faced by Rb survivors, this work highlights the critical need for integrated, patient-centered psychosocial support in long-term follow-up care. This aligns with the NHS Long-Term Plan's aim to broaden access to preventative support, especially in psychological care (NHS England, 2023) thus advocating for a more robust and structured support system for Rb survivors.

Central to these recommendations is the development of a dedicated psychosocial care pathway, shaped by survivor input and recognising developmental stages and abilities as well as the benefits of peer support, preventative interventions, and targeted psychological therapy. Such approaches are crucial in attempting to prevent mental health crises and reducing the burden on often overstretched mental health services and oncology teams. As discussed earlier in this thesis, a similar model has already been implemented successfully in the fields of genetic counselling, serving as a template for a holistic psychosocial care approach. Although some services do offer psychological support, this is often ad-hoc and request-driven, leaving gaps for individuals who may struggle to ask for help or who live far away from their Rb treatment centre. As a result, this research advocates for a standardised psychosocial intervention that is embedded into routine care and implemented uniformly across services in the UK and potentially beyond. By ensuring that all Rb survivors have access to consistent, evidence-based psychosocial support, this approach aims to reduce disparities in care, improve long-term psychological outcomes, and enhance overall quality of life. A structured intervention would also help integrate psychosocial considerations into the standard oncology care pathway, fostering a more holistic approach that acknowledges the emotional and social dimensions of survivorship alongside medical follow-up.

Whilst this is one of the goals of this work, it is important to consider how this might fit within the current challenges of the NHS system. As of the time of writing, the NHS is often referred to as being in ‘crisis’, facing significant difficulties with regard to funding constraints, workforce shortages, and ongoing attempts to reform and restructure the healthcare system, ultimately threatening its’ long-term viability (Williams, 2024). These systemic pressures raise questions about the feasibility of embedding a new psychosocial intervention into routine care, particularly given the increasing demand on healthcare professionals and limited availability of specialist mental health support. There is also a risk that, without adequate investment and prioritisation, psychosocial care could remain an inconsistent and under-resourced aspect of survivorship support. To address these challenges, it is crucial to consider how this research and a future intervention could be designed for sustainability and scalability within existing NHS structures. This may involve integrating psychosocial care into multidisciplinary team approaches, utilising digital health solutions to extend access, or developing training programs that equip non-specialist staff with the skills to provide basic psychosocial support. A psychological approach which may be suited to this is Acceptance and Commitment Therapy (ACT), which focuses on acceptance of challenging circumstances and supporting individuals to be flexibility with their thoughts and emotions, rather than avoiding or engaging negatively with them. It also encourages consideration of personal values and behaviour which aligns with these, something which is often lost amongst challenging or distressing circumstances. Beyond its application as a direct intervention for Rb survivors, ACT principles could also inform the way psychosocial care is integrated within the NHS. By fostering psychological flexibility at both an individual and systemic level, ACT could support healthcare professionals in navigating the pressures of an overstretched system while maintaining a commitment to delivering high-quality, patient-centred care. For example, embedding ACT-informed approaches within staff training may help equip healthcare workers with strategies to manage stress, adapt to systemic constraints, and maintain their capacity for compassionate care. Additionally, ACT’s emphasis on values-based action could guide the development of policies that prioritise sustainable, embedded psychosocial support, ensuring that interventions are not just introduced but effectively maintained within routine NHS practice. Alongside this, engagement with policymakers, healthcare providers, and patient advocacy groups will be essential in ensuring that any recommendations arising from this research are both realistic and actionable within the broader healthcare landscape. By acknowledging these structural challenges while advocating for improved psychosocial care, this research not only highlights the need for change but also contributes to the ongoing discussion about how best to implement it in a way that is both effective and sustainable.

The qualitative work in this thesis also identifies the specific elements of psychosocial support desired by young Rb survivors, including preferences regarding the mode of delivery and content of interventions. These insights will contribute to the development of a psychoeducation framework tailored to the needs of this unique patient population. By embedding such an intervention into standard clinical practice, healthcare providers will be better equipped to pre-empt, recognise and respond to the psychosocial challenges faced by Rb survivors, leading to both individual and systemic benefits. For survivors, this can provide early preventative support to reduce future distress, whilst also equipping those already dealing with mental health difficulties with effective coping strategies. Systemically, this approach can alleviate strain on NHS mental health services, reducing referrals and conserving resources. Additionally, the integration of psychosocial care within standard healthcare practice will likely improve overall quality of life for Rb survivors and serve as a model for other rare childhood conditions. This can broaden the scope and impact of mental health interventions across various healthcare settings, reinforcing a holistic approach to care that acknowledges the long-term psychosocial impact of surviving a life-threatening condition. It is my intention that policymakers will find the insights of this thesis valuable in advocating for funding opportunities and developing national guidelines for psychosocial interventions, ensuring that this type of care becomes a standardised part of the treatment pathway. As such, this research not only informs future Rb care but could also inspire a broader shift towards comprehensive psychosocial support in paediatric oncology and genetic disease frameworks, promoting equitable access to quality care for all young survivors.

In addition, my systematic review suggested that various interventions, including prosocial activities, physical exercise, and technology-based psychosocial education programs, could positively influence the well-being of TYA childhood cancer survivors. Establishing professional guidelines to adopt such approaches and integrating comprehensive survivorship psychosocial care plans alongside medical interventions can provide holistic support at all stages of their lives. My review highlights that more research is needed, however, to assess which interventions (or aspects of interventions) are effective over time, whilst considering modular support intervention across cancer and genetic disease populations to understand existing tools that could be modified for use within the Rb population. Ultimately, tailored and multifaceted approaches that consider the individual needs of this survivor group are essential for optimising intervention efficacy. Below I will outline the implications for clinical practice in Rb, as this is the focus of this thesis. I will then broaden this out to consider potential implications for other rare paediatric cancers, cancer pre-disposition syndromes, genetic diseases, conditions that cause facial changes, and conditions that cause visual impairment or blindness. This is because I anticipate that there will be overlap between my findings specific to Rb, and for TYA with conditions that have a similar impact. This will require further research that I discuss below but could have wide clinical implications as considered in the following sub-sections.

RETINOBLASTOMA

Findings from studies one and three highlight the specific psychosocial support elements desired by and useful to young Rb survivors, offering a framework for developing a modular psychoeducation intervention. Once empirically tested and implemented, these insights can enhance routine clinical practice, enabling healthcare providers to address the unique psychosocial needs of this group. Over the last few decades, person-centred care has become a key component of NHS and international intervention (Ekman, Ebrahimi and Olaya Contreras, 2021). The principles of this include providing dignity, compassion and respect, coordinated and personalised care, and empowering individuals to make shared decisions about treatment (Moody *et al.*, 2018). The findings from this thesis align with these principles, which emphasises how a future intervention could realistically fit within existing NHS frameworks. As both of the treatment centres for Rb in the UK were involved in this research, they are aware and in support of this work and the clinical implications of it. This is important when considering the realities of potential impact in clinical practice, both in the short and long-term. For example, a future psychoeducation intervention could be offered or implemented within routine follow-up appointments at both Rb treatment centres. These appointments could be enhanced by incorporating intervention modules that address the psychosocial challenges young Rb survivors face, such as managing anxiety, navigating social challenges, or coping with altered self-image. This integrated approach, grounded in the principles of person-centred care, would not only improve survivors' emotional well-being but could also foster a sense of empowerment and internal locus of control. In the long term, this could lead to more sustainable psychological outcomes and better overall quality of life for Rb survivors, reducing the need to access wider mental health services and ensuring the intervention is both clinically impactful and adaptable to the ever-changing context of the NHS.

There has already been impact from study one, and centres have been enthusiastic in hearing the experiences of young survivors and considering how these findings can support the topics addressed in long-term follow-up clinics. In addition, there is clinical impact outside of the UK, with Rb clinics in Denmark and the USA invested in this research. I was recently invited to Denmark to visit their Rb clinic and spent time discussing how their services can learn from the voices of survivors in the UK. After this visit, I was informed that changes had already made to clinical provision and that their psychologist is now a key part of the Rb MDT, providing an easier referral route to support. Alongside this, the Rb geneticist has expressed interest in forming an international collaboration, drawing on the findings of this thesis (and those of a future intervention) to mirror the support that they offer in line with the recommendations of this work. They have also highlighted the application of these insights to other conditions, as discussed below.

It is imperative that this work is as applicable to as many young people as possible, whilst acknowledging that the aim of qualitative research is not to be generalisable to all. Over the past 30 years, Rb treatment has evolved significantly, with advancements in early detection, genetic screening, and more targeted therapies reducing the need for enucleation and systemic chemotherapy (Ancona-Lezama et al., 2020). Modern approaches, such as intra-arterial and intravitreal chemotherapy, have improved ocular preservation and reduced long-term side effects. These developments mean that the experiences of participants who took part in this research, particularly those diagnosed up to three decades ago, may not fully reflect current treatment pathways. Having said this, I would argue that my findings remain relevant and transferable to contemporary practice, as they highlight enduring psychological and social impacts of diagnosis and treatment, the importance of holistic care, and patient experiences that continue to shape long-term outcomes. All of these factors are still important and applicable to children diagnosed with Rb today and in the future, regardless of the treatment modality offered. Understanding historical experiences, such as those presented in this thesis, provides valuable insight into ongoing challenges in survivorship, adaptation, and support needs, ensuring that modern interventions address both medical and psychosocial dimensions of care.

It is crucial to consider the role that gender may have on young people’s experience of engaging with psychosocial research and care. Gender norms, societal expectations, and personal identities can all influence how young people perceive and respond to research participation and psychosocial interventions, meaning that they may be more or less likely to engage and benefit from this . For instance, traditional gender roles may affect help-seeking behaviours, with young men potentially being less likely to engage in psychosocial support due to societal expectations of emotional resilience. Having said this, the uptake from young men in this thesis may be suggestive that this is less problematic in the younger generation. Conversely, young women may face different challenges, such as being more readily expected to express emotional distress, which could shape their interactions with research and care in distinct ways. Non-binary and gender-diverse young people may encounter additional barriers, including a lack of representation in research or care models that do not account for diverse gender identities.

Societal norms often encourage emotional expressiveness in young women while reinforcing the idea that seeking help or discussing mental health struggles is more socially acceptable for them compared to their male or non-binary peers. While this may facilitate engagement with psychosocial support, it can also lead to unintended consequences. For example, young women might feel pressure to conform to expectations of vulnerability, potentially influencing how they report their emotions in research settings.

This could result in either the amplification of distress to align with perceived norms or, conversely, a reluctance to share experiences that do not fit the expected narrative of emotional expressiveness. Additionally, there is a risk that their concerns may be dismissed as ‘typical’ or overly pathologised, leading to gendered biases in the interpretation of research findings and the provision of care. It is important that future interventions developed from this work do not unconsciously cater more to traditionally ‘feminine’ young people, such as designing an intervention that focuses solely on group discussions, potentially making them less accessible or engaging for those who do not align with these norms. As I hope has been the case in this thesis, this underscores the need for research and interventions to adopt a more nuanced, gender-sensitive approach that recognisses the diverse ways young people, regardless of gender, experience and express psychological distress.

It is also important to me that Rb research is not Eurocentric and considers the experiences of children and young people worldwide, particularly in countries where treatment options differ and survival rates are lower than the UK (Wong *et al.*, 2022). In conversations to inform this work, I chose to speak with many individuals in diverse countries and with different experiences of Rb. Although this is outside the scope of this PhD, this helped me to situate myself in the topic and understand the potential impact of the condition across different settings and individuals. One such conversation was with a parent in India, who drew my attention to the issue of how certain cultures ‘shun’ enucleation, and struggle to discuss childhood cancer through fear of religious and social consequences. Whilst conversations around such stigma did not come up from the participants in my studies, wider literature suggests that this can be commonplace when talking to individuals impacted by childhood cancer in other cultures (Graetz *et al.*, 2020; Krishnan *et al.*, 2023). I hope that this work can provide a starting place to considering the psychosocial experiences and support needs of Rb survivors in non-UK cultures, as well as across other conditions.

OTHER CONDITIONS

Although this PhD focuses on one rare paediatric cancer, its methodologies and findings have the potential to be applied to other disease groups. Rare childhood cancers, such as certain brain tumours (e.g. meningioma) (Nangarwal et al., 2021; Young et al., 2023), nasopharyngeal carcinoma (Bonafede et al., 2024), and other head and neck tumours(Covrig et al., 2021), share similarities in terms of medical uncertainty, treatment burden, and potential long-term effects. While late effects vary depending on the specific disease and treatment type, the psychosocial challenges of living with a rare cancer in childhood, including social isolation, altered self-image, and concerns about future health, can overlap across diagnoses. Similarly, individuals with cancer predisposition syndromes, such as Li-Fraumeni syndrome (Barnett et al., 2022; Kiermeier et al., 2025), Lynch syndrome (Campbell-Salome et al., 2021; Kalamo et al., 2021), and Neurofibromatosis types I and II (Aghaei et al., 2024), may experience similar psychological and social concerns as those with Rb, particularly due to the hereditary nature of their conditions and the associated risks of multiple malignancies. The insights gained from this thesis, particularly regarding identity, coping, and long-term well-being, can inform psychosocial research and interventions for these populations.

Beyond cancer, the findings of this research may also be relevant to individuals with other hereditary and genetic conditions, such as cystic fibrosis (Dobra et al., 2024; Li et al., 2023), sickle cell anaemia (Coco et al., 2024; Dyson et al., 2010; Essien et al., 2023), and Duchenne muscular dystrophy (Bever et al., 2024; Huang et al., 2023), all of which present lifelong medical challenges that can impact self-identity, social relationships, and mental health. Similarly, those with acquired or congenital facial differences, including conditions like Cushing’s syndrome (Santos et al., 2021), facial nerve palsy (Hotton et al., 2022; Rasing et al., 2024), and craniofacial injuries from burns or trauma (Sarwer et al., 2022; Woolard et al., 2021), may experience comparable psychosocial difficulties, particularly in relation to societal perceptions, managing challenging interactions with others, and self-esteem. Additionally, individuals with visual impairments or progressive sight loss (Xie et al., 2022), such as those with juvenile macular degeneration or congenital blindness (Ibrahim et al., 2022; Roxana et al., 2023), often need to navigate challenges related to independence, accessibility, and social inclusion, themes that are also central to the lived experiences of Rb survivors. The understanding gained from Rb survivors throughout this thesis provides a valuable foundation for exploring these shared experiences, identifying common psychosocial support needs, and tailoring future interventions accordingly.

Furthermore, there is potential for these findings to inform psychosocial research and interventions for individuals with childhood-onset conditions beyond those that are genetic or rare. For example, young people diagnosed with chronic illnesses such as type one diabetes (Jones et al., 2021 ; O’Donnell et al., 2022), epilepsy (Yeni, 2023), or inflammatory conditions (e.g. juvenile idiopathic arthritis) (Rongo et al., 2024) often experience disruptions to their daily lives, education, and future aspirations, much like childhood cancer survivors. While the medical trajectories of these conditions differ, they similarly require long-term management and can lead to concerns about stigma, social belonging, and identity formation. The methodologies used in this thesis could therefore be adapted to explore these issues in other paediatric populations such as those described, helping to develop targeted interventions that address the wider psychosocial challenges of growing up with a medical condition. It is therefore plausible that, once the current research informs an Rb-specific psychosocial intervention, this model could be adapted for other conditions with overlapping challenges. By applying the same methodological approaches to these populations, future research can explore both the universal and condition-specific aspects of psychosocial adjustment, enabling the development of tailored support strategies that extend beyond Rb. This broader application would enhance the impact of this research, contributing to a more comprehensive understanding of how life-altering medical diagnoses affect young people’s psychological and social well-being. Ultimately, recognising the transferable nature of these findings underscores the importance of a more integrated, cross-condition approach to psychosocial support in paediatric and young adult healthcare.

8.2 IMPLICATIONS FOR POLICY

This thesis underscores the profound psychological and emotional of Rb, highlighting the necessity for integrated and specific psychosocial support in long-term follow-up. Incorporating these insights can significantly contribute to the NHS Long-Term Plan, which aims to offer patients more support options, and particularly preventative and psychological aid (NHS England, 2023). This aligns with the NHS post-COVID-19 adaptations, which emphasise planning for non-hospital-based follow-up, including community care and digital or remote support. By recognising the emotional and behavioural impacts of Rb, this research can inform the creation of a dedicated care pathway, shaped by survivor experiences. It could also help policymakers develop guidelines and allocate resources for implementing comprehensive psychosocial support, mirroring the success of other aspects of Rb long-term follow-up care, like genetic screening and routine eye examinations. This could eventually inform National Institute for Health and Care Excellence (NICE) guidance for the psychological management of individuals living with and beyond childhood cancer.

8.3 IMPLICATIONS FOR RESEARCH

This thesis provides comprehensive understanding into the psychological and emotional impact of Rb on survivors, suggesting the need for dedicated psychosocial support pathways. It is important to disseminate this research widely and in a timely manner, particularly as this is a growing research area. The first qualitative study has already been published, the systematic review has been submitted for publication, and study three is currently being finalised for publication. After the publication of study one, I received interest from the media and from journals to offer further insights into the psychosocial needs of Rb survivors. This has led to potential opportunities within the mainstream media and submission of a second paper using the data collected in study one, but specifically focused on experiences of bullying. This paper is an inter-disciplinary collaboration with the Department of Education at the University of York and will mean that this work is disseminated wider than the field of health. In the mid-and longer-term, further research will focus on designing and empirically testing the aforementioned intervention to feed into such a pathway, with an emphasis on survivor-led recommendations, co-design, and co-production.

Co-design refers to the active collaboration between different stakeholders to devise a solution to a particular issue (Vargas *et al.*, 2022). In this thesis, I engaged individuals with lived experience, parents of children who have/have had Rb, and health care professionals at each stage, including collaborating on research aims, documentation, and results. Following on from this, co-production is a process which aims for projects to collaborative and integrative between researchers and participants, or clinicians and service users. Described as ‘aspirational and methodological’ (Turnhout *et al.*, 2020), these approaches are often deemed to reduce power dynamics between ‘patient and professional’. This is a concept I continue to be passionate about, particularly as I have not experienced what it is like to have Rb, so am not a member of the population I am researching, meaning it has been imperative that I consider the power dynamic between myself and the research participants (Cousin, 2010). It was important for me to own this position, openly acknowledging that I do not know what it is like to have had cancer, and thus elevating the participants to the position of ‘expert’ and emphasising that I am not conducting research ‘on’ these individuals but co-designing and producing it ‘with’ them. It would be remiss to consider that the relationship between the researcher and research participants does not influence the research itself (Orr and Bennett, 2019). Additionally, generating evidence to design an intervention for teenagers means that co-design, and key consideration of their needs, is particularly crucial. This is because this age group’s specific needs are often omitted from targeted support and based on support designed for children or for adults, which is modified to try and suit teenagers. Once tested and refined, this intervention can be used to establish evidence-based practices that can be integrated into routine care. This should build on elements of existing effective interventions highlighted in study two, acknowledging that further research is needed into the specific mechanisms through which these interventions exert their benefits and how they can be tailored to individual needs.

8.4 PPI ENGAGEMENT

Throughout this thesis, PPI has been at the forefront. I am passionate about highlighting voices of lived experience, and place equal value on this as I do with academic skills and expertise. Due to the rare nature of Rb and the relatively small number of individuals affected, there was a small pool of individuals to work with, whilst making sure that my PPI group were separate to those who were research participants. Thankfully there was no point during the PhD process that I could not access PPI, and I have even had people reach out to me since my studies have ended, asking when they can get involved in future projects. It is plausible that involvement was advanced by the COVID-19 pandemic, meaning that contributing to research became more accessible and removed geographical barriers. This enthusiasm could also be due to CHECT’s loyal support network and the recognition that there is currently a lack of formalised psychosocial support available for Rb. Furthermore, I hypothesise that the experience of young people’s involvement was in itself therapeutic, and I have received extensive feedback about how valuable it has been to meet others affected by Rb and who face similar challenges (e.g. visual impairment, living with a prosthetic eye, living with a genetic condition). Reflecting back, I hope that the PPI shines through this thesis and goes on to inform the psychosocial intervention from this work. Disseminating the findings from this PhD in places that are accessible to members of the public and individuals outside of academia is also important. PPI has been and will continue to be crucial in this process, and the production of our podcast mini-series and video accompaniment to study one are examples of how I have done this (please refer back to chapter 4 for access). I hope that by working with young people affected by Rb and making a concerted effort to make my research accessible, that the implications of this work will be wider reaching and thus more impactful for young survivors of Rb.

8.5 CONCLUSION

Retinoblastoma is a rare childhood cancer with a high survival rate in the UK. Whilst this is a testament to medical advancements over the years, there are improvements to be made with regard to psychosocial care. This thesis examined the experiences of teenage and young adult survivors, and is, as far as I know, the only body of work which considers Rb psychosocial survivorship needs as a whole, including for those impacted by both heritable and non-heritable disease. I used qualitative methods to highlight individual narratives across the age range in which cancer-related distress is often prominent. In addition I conducted a systematic review which analysed existing psychosocial interventions for survivors of any form of childhood cancer, providing empirical evidence of support which has been shown to be impactful, as well as emphasising the need for robust consideration of how we measure efficacy. Lastly, I conducted a content analysis to provide a framework of practical suggestions regarding the design and implication of a tailored Rb psychoeducation intervention that I plan to develop and test post-doctorally in collaboration with experts by experience. This will consider the developmental stages of the individuals accessing it and aims to foster a sense of internal locus of control and empowerment amongst all survivors.

These three interlinked studies clearly demonstrate the psychosocial needs of young people living beyond Rb and proposes a comprehensive overview, underpinned by evidence, of how to support them, considering the realities of the NHS and wider societal context and potential challenges with implementation into long-term follow-up care. This thesis represents an exciting shift towards realistic change for the Rb community, and with the continued support of the UK clinical care teams and other key stakeholders (both nationally and internationally), I believe that the evidence generated will develop a significantly high standard for the future psychosocial care provided to this group of young people.

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Zelihić, D. *et al.* (2022) ‘Reducing social anxiety in adolescents distressed by a visible difference: Results from a randomised control trial of a web-based intervention’, *Body Image*, 40, pp. 295–309. doi: https://doi.org/10.1016/j.bodyim.2022.01.008.

Zhou, M. *et al.* (2024) ‘Recent progress in retinoblastoma: Pathogenesis, presentation, diagnosis and management’, *Asia-Pacific Journal of Ophthalmology*, 13(2), p. 100058. doi: 10.1016/j.apjo.2024.100058.

APPENDICES

APPENDIX A

This appendix includes all appendices related to chapter three of this thesis.

APPENDIX A1: NICOLA O'DONNELL GCP REFRESHER TRAINING CERTIFICATE

NIHR | National Institute for Health and Care Research

Issued by:
Clinical Research Network
Coordinating Centre

CERTIFICATE OF ACHIEVEMENT

Nicola O'Donnell

has completed the course:

Good Clinical Practice (GCP) Refresher ELearning

August 2, 2023

Modules Completed

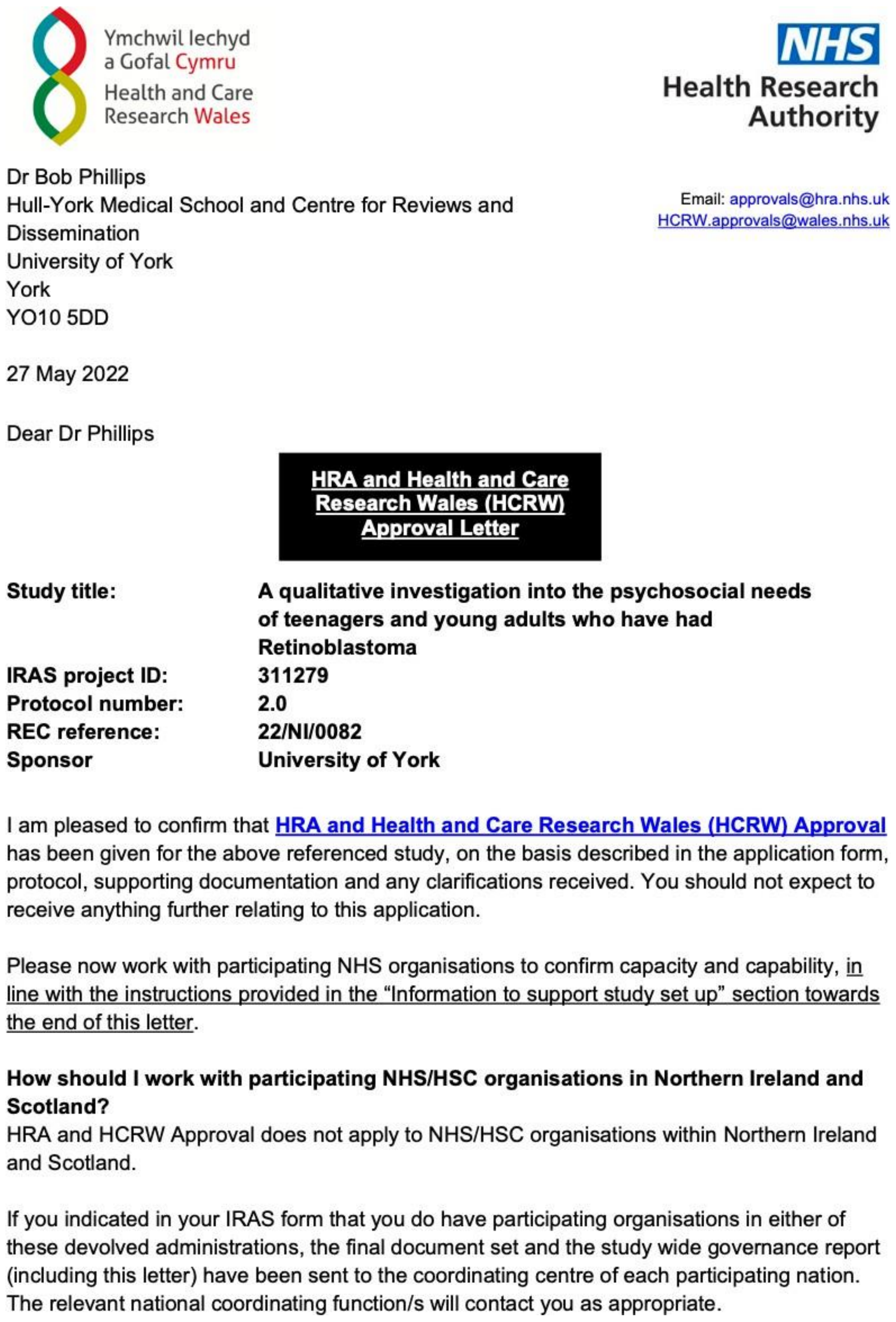
- Good Clinical Practice (GCP) Refresher: Revisiting Key Concepts
- GCP Refresher Hot Topics
- Good Clinical Practice (GCP) Refresher: Reflecting on your own practice and experience

This course is worth 3 CPD Credits

CPD
The CPD Certification Service

Version: May 2022

Appendix A2: HRA and HCRW Approval, 27.05.22



Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales

NHS
Health Research
Authority

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

Dr Bob Phillips
Hull-York Medical School and Centre for Reviews and
Dissemination
University of York
York
YO10 5DD

27 May 2022

Dear Dr Phillips

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma**
IRAS project ID:	**311279**
Protocol number:	**2.0**
REC reference:	**22/NI/0082**
Sponsor	**University of York**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **311279**. Please quote this on all correspondence.

Yours sincerely,
Rachel Katzenellenbogen
Approvals Specialist

Email: approvals@hra.nhs.uk

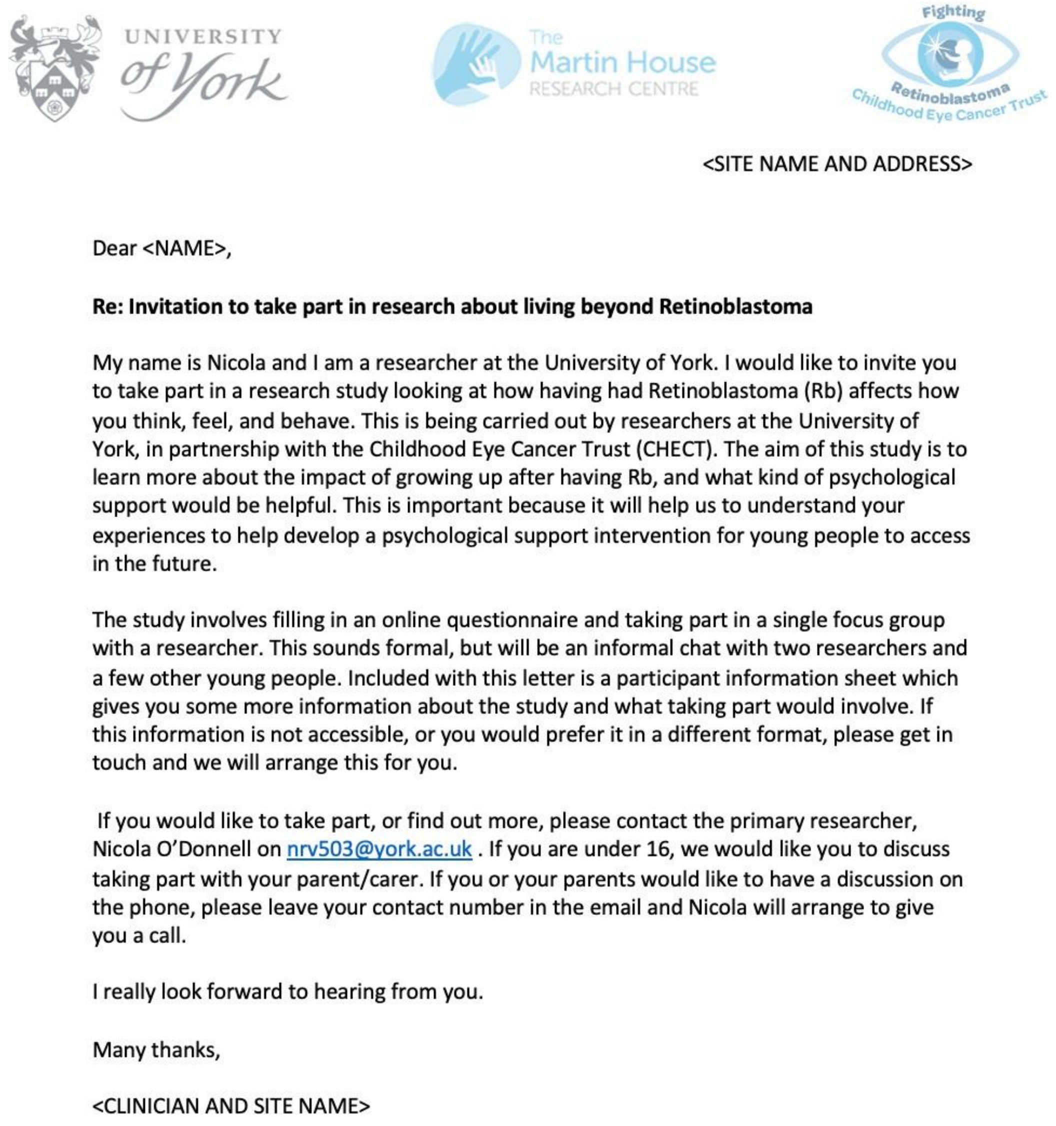
Copy to: Mr Michael Barber

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Covering letter on headed paper [Response to REC provisional opinion]		16 May 2022
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [University of York sponsor insurance evidence]	1	27 July 2021
Interview schedules or topic guides for participants	1	20 January 2022
Interview schedules or topic guides for participants [Interview topic guide]	1	20 January 2022
IRAS Application Form [IRAS_Form_11042022]		11 April 2022
Letter from sponsor [Sponsor letter]	1	30 March 2022
Letters of invitation to participant [Invite focus groups]	2	10 May 2022
Letters of invitation to participant [Invite interviews]	2	10 May 2022
Non-validated questionnaire [Questionnaire teenagers]	2	10 May 2022
Organisation Information Document [Organisation Information Document]	1	31 March 2022
Other [Questionnaire Young adults]	2	10 May 2022
Other [PIS focus groups]	3	10 May 2022
Other [Distress Protocol]	1	11 May 2022
Other [Parents PIS for teenage participants]	1	10 May 2022
Other [Alternative PIS Video Links]	1	18 May 2022
Other [Interview consent]	1	12 December 2021
Other [PPI recruitment poster]	1	18 January 2022
Participant consent form [Focus group consent form]	1	12 December 2021
Participant consent form [Interview consent form]	1	12 December 2021
Participant information sheet (PIS) [Focus groups]	3	10 May 2022
Participant information sheet (PIS) [Interviews]	3	10 May 2022
Participant information sheet (PIS) [Focus groups - parents]	1	10 May 2022
Participant information sheet (PIS) [Participant Information sheet Interviews]	3	10 May 2022
Protocol	3	11 May 2022
Research protocol or project proposal [Protocol]	3	11 May 2022
Schedule of Events or SoECAT [SoE]		
Summary CV for Chief Investigator (CI) [CV CI]		18 June 2021
Summary CV for student [CV CI]		22 March 2022
Summary CV for supervisor (student research) [Supervisor CV]		18 June 2021

Appendix A3: Invitation letter for teenager focus groups, version 2, 10.05.22



<SITE NAME AND ADDRESS>

Dear <NAME>,

Re: Invitation to take part in research about living beyond Retinoblastoma

My name is Nicola and I am a researcher at the University of York. I would like to invite you to take part in a research study looking at how having had Retinoblastoma (Rb) affects how you think, feel, and behave. This is being carried out by researchers at the University of York, in partnership with the Childhood Eye Cancer Trust (CHECT). The aim of this study is to learn more about the impact of growing up after having Rb, and what kind of psychological support would be helpful. This is important because it will help us to understand your experiences to help develop a psychological support intervention for young people to access in the future.

The study involves filling in an online questionnaire and taking part in a single focus group with a researcher. This sounds formal, but will be an informal chat with two researchers and a few other young people. Included with this letter is a participant information sheet which gives you some more information about the study and what taking part would involve. If this information is not accessible, or you would prefer it in a different format, please get in touch and we will arrange this for you.

If you would like to take part, or find out more, please contact the primary researcher, Nicola O'Donnell on nrv503@york.ac.uk . If you are under 16, we would like you to discuss taking part with your parent/carer. If you or your parents would like to have a discussion on the phone, please leave your contact number in the email and Nicola will arrange to give you a call.

I really look forward to hearing from you.

Many thanks,

<CLINICIAN AND SITE NAME>

Appendix A4: Invitation letter for young adult individual interview, version 2, 10.05.22

<SITE NAME AND ADDRESS>

Dear <NAME>,

Re: Invitation to take part in research about living beyond Retinoblastoma

My name is Nicola and I am a researcher at the University of York. I would like to invite you to take part in a research study looking at the psychological and social impact of having had Retinoblastoma (Rb). This is being carried out by researchers at the University of York, in partnership with the Childhood Eye Cancer Trust (CHECT). The aim of this study is to learn more about the impact of growing up after having had Rb, and what kind of psychological support would be helpful. This is important because it will help us to understand your experiences to inform the development of a psychological support intervention for individuals to access in the future.

The study involves filling in an online questionnaire and taking part in a single individual interview with a researcher. Included with this letter is a participant information sheet which gives you some more information about the study and what taking part would involve. If this information is not accessible, or you would prefer it in a different format, please get in touch and we will arrange this for you.

If you would like to take part, or find out more, please contact the primary researcher, Nicola O'Donnell on nrv503@york.ac.uk . If you would like to have a discussion on the phone, please leave your contact number in the email and Nicola will arrange to give you a call.

I really look forward to hearing from you.

Many thanks,

<CLINICIAN AND SITE NAME>

APPENDIX A5: ALTERNATIVE (VIDEO) PARTICIPANT INFORMATION SHEET, VERSION 1, 18.05.22

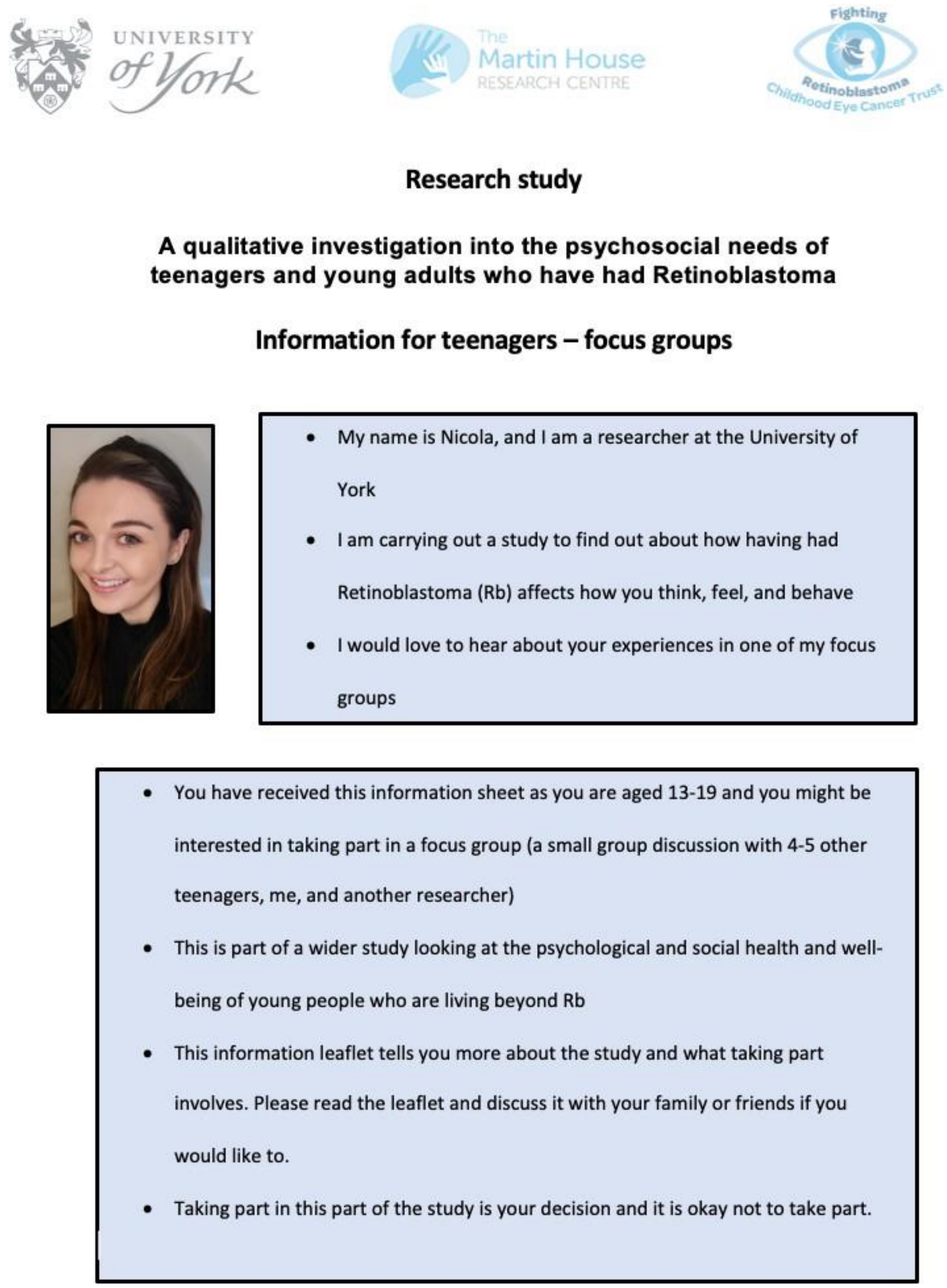
Below are the links for the alternative PIS – these have been recorded to increase accessibility for young people and for young people with visual impairment who may prefer to listen to the study information. These have been approved by our young people's PPI. There is no new information in these videos that is not in the PIS.

Interview information video:
https://youtu.be/oXRtJJkvfrg

Focus group information video:

https://youtu.be/0oNhs0oJhc4

Appendix A6: Participant Information Sheet for teenagers completed focused groups, version 3, 10.05.22



Research study

A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

Information for teenagers – focus groups

- My name is Nicola, and I am a researcher at the University of York
- I am carrying out a study to find out about how having had Retinoblastoma (Rb) affects how you think, feel, and behave
- I would love to hear about your experiences in one of my focus groups

- You have received this information sheet as you are aged 13-19 and you might be interested in taking part in a focus group (a small group discussion with 4-5 other teenagers, me, and another researcher)
- This is part of a wider study looking at the psychological and social health and well-being of young people who are living beyond Rb
- This information leaflet tells you more about the study and what taking part involves. Please read the leaflet and discuss it with your family or friends if you would like to.
- Taking part in this part of the study is your decision and it is okay not to take part.

Why is this research being carried out?

Rb is a rare eye cancer affecting 40-50 children a year. Although it is highly curable, this experience can leave some people feeling worried, angry, or sad. At the moment there isn't much support to help young people with these feelings, and we want to do something about this.

This research study is important because it will help us to understand the experiences of young people and some of the challenges that they face. It will also help us to understand what support they think would be helpful. The findings from this study will be used to design support for teenagers in the future.

Why have I been chosen, and do I have to take part?

You may have provided your details to a researcher to learn more about this study, or someone (a member of your clinical team, or a family member) may have thought you may be interested in taking part. Taking part in a focus group is entirely your decision and whatever you decide will **not** affect the care and support you receive.

What will happen if I decide to take part?

We will use these focus groups to understand your experiences. If you decide to take part, we would like you to talk to your parents so that they are aware of the study and what it will involve – feel free to share this information sheet with them!

We will try to organise the group for a time and date that is convenient for everyone taking part. It is possible for this to happen in an evening, a weekend, or during school holidays if this is preferred by the group.

When you sign up, you will be asked to complete a quick questionnaire about your details. If you would like support to fill this in, a researcher can go through this with you. If you do not know the answer to one of the questions, please do not worry.

The focus group will last between one and two hours with a break in the middle. During the group you will be asked to talk about your experiences as a teenager who has had Rb, your feelings and how you think your experiences have affected you. You will also be asked about ways in which you wish you had been supported through these experiences.

The focus groups can be face-to-face (depending on COVID-19 restrictions), or via video call e.g., Zoom. With your permission, the group will be audio and video recorded so that we don't miss anything that you tell us. These recordings will be deleted once we have made full notes of the conversations that we have.

What topics might we talk about?

It is your decision to share whatever you feel comfortable talking about. You will never be asked to take part in a conversation that you do not want to be involved in.

Some topics that teenagers have told us that they worry about:

- Talking to friends about their cancer experience
- Changes to their appearance after Rb
- Late effects caused by cancer treatment
- Cancer coming back
- Having relationships
- Having children in the future
- Lacking confidence
- Feeling different

Will taking part be kept private?

Yes, if you take part, you will be given a unique ID number which will mean that your data is pseudonymised, i.e. unable to be identified as you. The topics that we discuss in the group and any conversations between you and the research team are completely confidential, unless you tell us something that makes us seriously worried for yours or someone else's safety. If this happens, we would talk to you first about the best thing to do. If you are under 16, this may involve speaking with your parents. If you are under an Rb team, it may involve sharing our worries with them. We will only share information if we are really worried.

We will follow strict rules about confidentiality and all information will be stored in locked filing cabinets and in password-protected folders on computers.

What are the pros and cons of taking part?

Pros:

- Help other teenagers in the future
- Help researchers understand what young people who have had Rb need to support them
- Meet other young people who have experienced having Rb
- Share your experiences

Cons:

- You might find it difficult to talk about or answer questions about your experience – but you can stop at any time and researchers will provide details of support. We would always encourage you to share any worries with your parents/carers. If it is helpful, you can choose for the research team to speak to your parent/carers to provide post-study support.
- You can also contact CHECT for support at any point during or after the study. They can be contacted via email (support@chect.org.uk) or phone (0207 3775578).

Will it cost me anything to take part?

If you have to travel in order to take part in the group, we will pay for your travel costs.

Will I be paid to take part in this study?

We really appreciate you giving your time to help with our research, so we would like to offer you a £20 retail voucher for taking part.

If I want to take part, what do I need to do next?

Great news! Please contact the research team to let us know. You will be given a consent form to sign before the group. If you are under 16, we would like you to discuss taking part with your parent/carer, as it's really important that they are aware of your involvement. If you have any questions please contact Nicola O'Donnell, nrv503@york.ac.uk

What if I change my mind during the study?

You can withdraw at any time and you do not need to give us a reason. We will keep the information we have already taken from you. This will include your personal details so that if you contact us again, we know that you have taken part. We will not use this information to contact you though. Your part of the focus group will not be included in the study as long as they have not already been used for analysis.

Part Two

Who has reviewed this study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Health Research Authority (IRAS: 311279)

How will you use information about me?

We will need to use information from you for this research project. To safeguard your rights, we will use the minimum personally identifiable information possible.

This information that we collect for the research will include:

- The information from our focus group will be audio recorded and transcribed by a third party
- The information from your completed questionnaire
- Your name and contact details

We will use your personal information only to do the research. Other authorised individuals may check your records to make sure that the research is being done properly.

Your name and contact details will be kept separate from the other information that we obtain from you for this research. Your research records will contain a unique code number instead, so you are not directly identifiable when we use your data in our research.

Your name and contact details and research records will be stored electronically on a secure server at the University of York. Access to this information will be restricted to authorised persons only.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data for at least ten years. Personal data including consent forms will be kept for 3 years and then destroyed. This will enable us to complete our publications and reports. We will write our reports in a way that no-one can work out that you took part in the study. As soon as the focus groups have been transcribed, we will destroy all recordings of these.

What are my choices about how my information is used?

The University of York is a publicly funded organisation that conducts research to improve health and healthcare services. In legal terms, we are using your information for this research as part of 'a task in the public interest'. The ability to change the data that we have collected, however, is limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate.

You can stop being part of the study at any time, without giving a reason. You may request that your data is not used in the study up to 14 days after the interview. We will destroy your data upon this request. After this time, data gathered may have started to be analysed and used in the study findings. Any information that has been used in the study findings cannot be withdrawn.

By taking part, you understand that in the unlikely event of a loss of capacity, the research team would retain the information that you have provided and will continue to use it confidentially in connection with the purposes of the study.

Where can I find out more about how my information is used?

You can find out more about how we use your information

- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/
- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/research-partcipants/
- by sending an email to nrv503@york.ac.uk
- by emailing the University of York's data protection officer on dataprotection@york.ac.uk

Will my taking part in this study be kept confidential?

If you decide to take part in the study, what you tell us will be kept confidential and not shared with those involved in your care and treatment. No one outside the research team will know that you have taken part in the study.

We will write our reports in a way that no-one can work out that you took part in the study.

Data collected for the study may be looked at by authorised persons who are organising the research. Data may also be looked at by other authorised people to check that the study is being carried out correctly. All have a duty of confidentiality to you as a research participant.

The only time we would break our duty of confidentiality is if we are worried that you – or someone else – was being, or was likely to be, harmed. **If that happens, we will talk with you about it.**

What will happen to the results of the research study?

Researchers from the University of York will analyse the material collected.

The results of the study will be published in academic journals.

This may include direct quotes from you, but these will be anonymous and unable to be linked to you.

What if there is a problem?

Complaints

If you have a concern about any aspect of this study, you should ask to speak to the research team who will do their best to answer your questions (contact details below). You can also contact the Chief Investigator, Nicola O'Donnell. She will be happy to discuss your concerns. If you are unhappy with Nicola's response, you can contact Professor Lorna Fraser, who is Head of Department (Health Sciences).

If you remain unhappy following this and you wish to complain formally, you can do this by contacting the Parliamentary and Health Service Ombudsman, who is independent of the NHS and government, at 0345 015 4033.

Data Protection

The University of York is the data controller for the information collected for this research, which means we are responsible for looking after your personal information and using it properly. If you are unhappy with the way your personal information has been handled, please contact the University's Data Protection Officer at dataprotection@york.ac.uk. If you are not satisfied with our response, you have a right to complain to the Information Commissioner's Office. For information on reporting a concern to the Information Commissioner's Office, see www.ico.org.uk/concerns.

What will happen if the study stops earlier than expected?

Should the research stop earlier than planned and you are affected in any way, we will tell you and explain why.

Thank you so much for reading this information sheet. Please keep this copy.

Get in touch with the research team:

Nicola O'Donnell

Nrv503@york.ac.uk

Study funding and approvals:

The University of York is the sponsor for this study.

The study is funded by the Childhood Eye Cancer Trust (Study Reference: 311279).

Appendix A7: Participant Information Sheet for parents of teenagers who completed focused groups, version 1, 10.05.22

Research study

A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

Information for parents of teenagers – focus groups

- My name is Nicola, and I am a researcher at the University of York
- I am carrying out a study to find out about how having had Retinoblastoma (Rb) affects how you think, feel, and behave
- I would love to hear about young people's experiences in one of my focus groups

- You have received this information sheet as you are a parent of a young person aged 13-19 who you might be interested in taking part in a focus group
- This is part of a wider study looking at the psychological and social health and well-being of young people who are living beyond Rb
- This information leaflet tells you more about the study and what taking part involves. Please read the leaflet and discuss it with your child, helping them to decide about taking part
- Taking part in this study is your child's decision and it is okay not to take part.

Why is this research being carried out?

Rb is a rare eye cancer affecting 40-50 children a year. Although it is highly curable, this experience can leave some people feeling worried, angry, or sad. At the moment there isn't much support to help young people with these feelings, and we want to do something about this.

This research study is important because it will help us to understand the experiences of young people and some of the challenges that they face. It will also help us to understand what support they think would be helpful. The findings from this study will be used to design support for teenagers in the future.

Why has my child been chosen, and do they have to take part?

Your child may have provided your details to a researcher to learn more about this study, or someone (e.g., a member of their clinical team) may have thought that they may be interested in taking part. Taking part in a focus group is entirely their decision and whatever they decide will **not** affect the care and support that they receive.

What will happen if they decide to take part?

We will use these focus groups to understand young people's experiences. If your child decides to take part, we ask them to talk to you first so that you are aware of their involvement.

We will try to organise the group for a time and date that is convenient for everyone taking part. It is possible for this to happen in an evening, a weekend, or during school holidays if this is preferred by the group. When your child signs up, they will be asked to complete a quick questionnaire about their details.

The focus group will last between one and two hours with a break in the middle. During the group your child will be asked to talk about their experiences as a teenager who has had Rb, their feelings and how they think your experiences have affected them. They will also be asked about ways in which they wish they had been supported through these experiences.

The focus groups can be face-to-face (depending on COVID-19 restrictions), or via video call e.g., Zoom. With your child's permission, the group will be audio and video recorded so that we don't miss anything that is spoken about. These recordings will be deleted once we have made full notes of the conversations that we have.

What topics might we talk about?

It is your child's decision to share whatever they feel comfortable talking about. They will never be asked to take part in a conversation that they do not want to be involved in.

Some topics that teenagers have told us that they worry about:

- Talking to friends about their cancer experience
- Changes to their appearance after Rb
- Late effects caused by cancer treatment
- Cancer coming back
- Having relationships
- Having children in the future
- Lacking confidence
- Feeling different

Will taking part be kept private?

Yes, if your child takes part, they will be given a unique ID number which will mean that their data is pseudonymised, i.e. unable to be identified as them. The topics that we discuss in the group and any conversations between your child and the research team are completely confidential, unless they tell us something that makes us seriously worried for theirs or someone else's safety. If this happens, we would talk to them first about the best thing to do. If they are under 16, this would likely involve speaking with you. If your child is currently under an Rb team, it may involve sharing our worries with them. We will only share information if we are really worried.

We will follow strict rules about confidentiality and all information will be stored in locked filing cabinets and in password-protected folders on computers.

What are the pros and cons of taking part?

Pros:

- Help other teenagers in the future
- Help researchers understand what young people who have had Rb need to support them
- Meet other young people who have experienced having Rb
- Share their experiences

Cons:

- Your child might find it difficult to talk about or answer questions about their experience – but they will be informed that they can stop at any time and researchers will provide details of support. We would always encourage them to share any worries with you as their parents/carers. The research team are very happy to speak to you as parent/carers at any stage to provide post-study support.
- You and your child can also contact CHECT for support at any point during or after the study. They can be contacted via email (support@chect.org.uk) or phone (0207 3775578).

Will it cost my child anything to take part?

If your child has to travel in order to take part in the group, we will pay for their travel costs.

Will my child be paid to take part in this study?

We really appreciate your child giving their time to help with our research, so we would like to offer them a £20 retail voucher for taking part.

If they want to take part, what do they need to do next?

Great news! Please ask them to contact the research team to let us know. Your child will be given a consent form to sign before the group. If they are under 16, we will ask them to discuss taking part with you, as it's really important that you are aware of their involvement in our study. If you or they have any questions at any point, please contact Nicola O'Donnell, nrv503@york.ac.uk

What if my child changes their mind during the study?

Your child can withdraw at any time, and they do not need to give us a reason. We will keep the information we have already taken from them. This will include their personal details so that if they contact us again, we know that they have taken part. We will not use this information to contact them though. Your child's part of the focus group will not be included in the study as long as it has not already been used for analysis.

Part Two

Who has reviewed this study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Health Research Authority (IRAS: 311279)

How will you use information about my child?

We will need to use information from your child for this research project. To safeguard your child's rights, we will use the minimum personally identifiable information possible.

This information that we collect for the research will include:
- The information from our focus group will be audio recorded and transcribed by a third party
- The information from your child's completed questionnaire
- Your child's name and contact details

We will use your child's personal information only to do the research. Other authorised individuals may check your child's records to make sure that the research is being done properly.

Your child's name and contact details will be kept separate from the other information that we obtain from your child for this research. Your child's research records will contain a unique code number instead, so they are not directly identifiable when we use your data in our research.

Your child's name and contact details and research records will be stored electronically on a secure server at the University of York. Access to this information will be restricted to authorised persons only.

We will keep all information about your child safe and secure.

Once we have finished the study, we will keep some of the data for at least ten years. Personal data including consent forms will be kept for 3 years and then destroyed. This will enable us to complete our publications and reports. We will write our reports in a way that no-one can work out that your child took part in the study. As soon as the focus groups have been transcribed, we will destroy all recordings of these.

What are my child's choices about how their information is used?

The University of York is a publicly funded organisation that conducts research to improve health and healthcare services. In legal terms, we are using your child's information for this research as part of 'a task in the public interest'. The ability to change the data that we have collected, however, is limited, as we need to manage your child's information in specific ways in order for the research to be reliable and accurate.

Your child can stop being part of the study at any time, without giving a reason. Your child may request that their data is not used in the study up to 14 days after the interview. We will destroy their data upon this request. After this time, data gathered may have started to be analysed and used in the study findings. Any information that has been used in the study findings cannot be withdrawn.

By taking part, you understand that in the unlikely event of a loss of capacity, the research team would retain the information that your child has provided and will continue to use it confidentially in connection with the purposes of the study.

Where can I find out more about how my child's information is used?

You can find out more about how we use your information

- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/
- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/research-partcipants/
- by sending an email to nrv503@york.ac.uk
- by emailing the University of York's data protection officer on dataprotection@york.ac.uk

Will my child's taking part in this study be kept confidential?

If your child decides to take part in the study, what they tell us will be kept confidential and not shared with those involved in their care and treatment. No one outside the research team will know that your child has taken part in the study.

We will write our reports in a way that no-one can work out that your child took part in the study.

Data collected for the study may be looked at by authorised persons who are organising the research. Data may also be looked at by other authorised people to check that the study is being carried out correctly. All have a duty of confidentiality to your child as a research participant.

The only time we would break our duty of confidentiality is if we are worried that your child – or someone else – was being, or was likely to be, harmed. **If that happens, we will talk with your child about it.**

What will happen to the results of the research study?

Researchers from the University of York will analyse the material collected.

The results of the study will be published in academic journals.

This may include direct quotes from you, but these will be anonymous and unable to be linked to you.

What if there is a problem?

Complaints

If you have a concern about any aspect of this study, you should ask to speak to the research team who will do their best to answer your questions (contact details below). You can also contact the Chief Investigator, Nicola O'Donnell. She will be happy to discuss your concerns. If you are unhappy with Nicola's response, you can contact Professor Lorna Fraser, who is Head of Department (Health Sciences).

If you remain unhappy following this and you wish to complain formally, you can do this by contacting the Parliamentary and Health Service Ombudsman, who is independent of the NHS and government, at 0345 015 4033.

Data Protection

The University of York is the data controller for the information collected for this research, which means we are responsible for looking after your child's personal information and using it properly. If you are unhappy with the way your child's personal information has been handled, please contact the University's Data Protection Officer at dataprotection@york.ac.uk. If you are not satisfied with our response, you have a right to complain to the Information Commissioner's Office. For information on reporting a concern to the Information Commissioner's Office, see www.ico.org.uk/concerns.

What will happen if the study stops earlier than expected?

Should the research stop earlier than planned and your child is affected in any way, we will tell you and explain why.

Thank you so much for reading this information sheet. Please keep this copy.

Get in touch with the research team:

Nicola O'Donnell

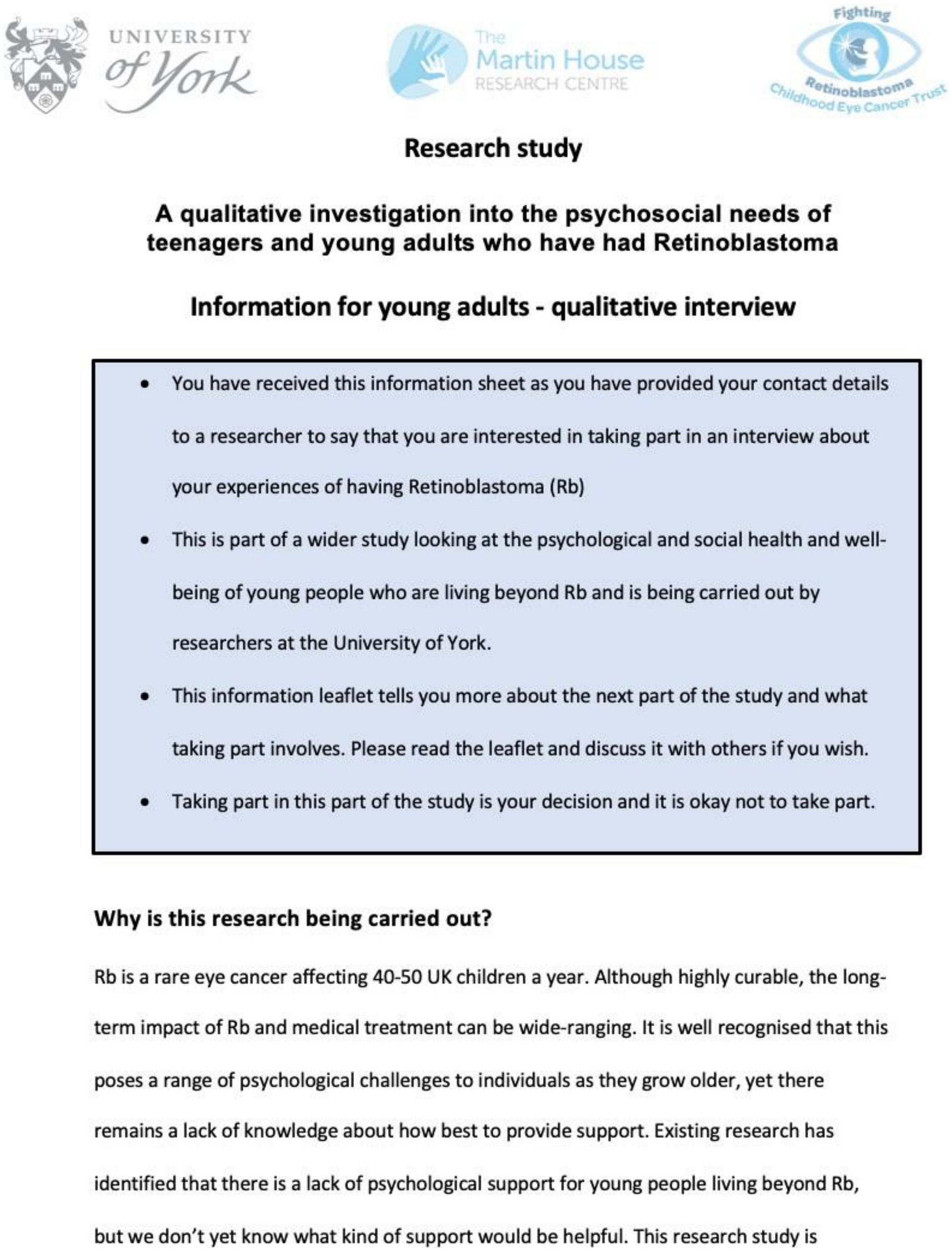
Nrv503@york.ac.uk

Study funding and approvals:

The University of York is the sponsor for this study.

The study is funded by the National Institute for Health Research (NIHR) (Academy) (Study Reference: 311279

APPENDIX A8: PARTICIPANT INFORMATION SHEET FOR YOUNG ADULTS COMPLETING INDIVIDUAL INTERVIEWS, VERSION 3, 10.05.22



Research study

A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

Information for young adults - qualitative interview

- You have received this information sheet as you have provided your contact details to a researcher to say that you are interested in taking part in an interview about your experiences of having Retinoblastoma (Rb)
- This is part of a wider study looking at the psychological and social health and well-being of young people who are living beyond Rb and is being carried out by researchers at the University of York.
- This information leaflet tells you more about the next part of the study and what taking part involves. Please read the leaflet and discuss it with others if you wish.
- Taking part in this part of the study is your decision and it is okay not to take part.

Why is this research being carried out?

Rb is a rare eye cancer affecting 40-50 UK children a year. Although highly curable, the long-term impact of Rb and medical treatment can be wide-ranging. It is well recognised that this poses a range of psychological challenges to individuals as they grow older, yet there remains a lack of knowledge about how best to provide support. Existing research has identified that there is a lack of psychological support for young people living beyond Rb, but we don't yet know what kind of support would be helpful. This research study is important because it will help us to understand the experiences of young people and some of the challenges that they have faced. It will also help us to understand what support they wish they could have had access to. The findings from this study will be used to design psychological support for individuals to access in the future.

Why have I been chosen, and do I have to take part?

You may have provided your details to a researcher to learn more about this study, or someone (one of your clinicians, or a family member etc.) may have thought you may be interested in taking part. Taking part in an interview is entirely your decision and whatever you decide will **not** affect the standard of care and support you receive.

What will happen if I decide to take part?

We will use this interview to gain more of an in-depth understanding of your experiences and psychological wellbeing. If you decide to take part in an interview, we will organise it for a time and date that is convenient for you. When you sign up, you will also be asked to complete a quick questionnaire about your details. If you would like support to fill this in, a researcher can go through this with you. If you do not know the answer to one of the questions, please do not worry.

The interview will last between 45 and 90 minutes. During the interview you will be asked to talk about your experiences as a young adult who has had Rb, your psychological health and how you think your experiences have affected your wellbeing. You will also be asked about ways in which you wish you had been supported through these experiences.

You can choose where the interview takes place and you can have someone with you if you would like. It can be face-to-face (depending on COVID-19 restrictions), over the phone or via video call e.g., Zoom. With your consent, the interview will be audio and video recorded so that we don't miss anything that you tell us. This recordings will be deleted once we have made full notes of the conversation that we have.

Will my taking part be kept confidential?

Yes, if you take part you will be given a unique participant ID which will mean that your data is pseudonymised, i.e. unable to be identified as you. The topics that we discuss in the interview and any conversations arising during contact with the research team completely confidential, unless you tell us something that raises concerns that you or someone else is at serious risk of harm. If this happens, we would talk to you first about the best thing to do. If you are under an Rb team, it may involve sharing our worries with them. We will only share information if we are really worried.

We will follow strict rules about confidentiality and all information will be stored in locked filing cabinets and in password-protected folders on computers.

What are the possible benefits and risks to me of taking part?

By taking part in this study you could help young people in the future. Information collected from you and from others could lead to a better understanding of the experiences and psychological and social wellbeing of young people living beyond Rb. It is possible that you may find it difficult to talk about or answer questions relating to your experiences. If you find the interview distressing, please contact the research team and we can provide details of local organisations who can help. You can also contact CHECT for support at any point during or after the study. They can be contacted via email (support@chect.org.uk) or phone (0207 3775578). Taking part is voluntary and you can decide to withdraw from the study at any time.

Are there any expenses or payment for taking part in this study?

We really appreciate you giving your time to help with our research, so we would like to offer you a £20 retail voucher for taking part. If you have to travel in order to participate in the study, we will reimburse your travel costs.

If I want to take part, what do I need to do next?

If you decide that you want to take part in an interview, please contact the research team to organise an interview. You will be provided with a consent form to sign before the interview.

If you have any questions about the study or about any of the statements on this form please contact Nicola O'Donnell, nrv503@york.ac.uk

What if I change my mind during the study?

You can withdraw at any time and you do not need to give us a reason. We will keep the information we have already obtained from you. This will include your personal details so that if you contact us again, we know that you have taken part. We will not use this information to contact you though. Your interview responses will not be included in the study as long as they have not already been used for analysis.

Part Two

Who has reviewed this study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Health Research Authority (IRAS: 311279)

How will you use information about me?

We will need to use information from you for this research project. To safeguard your rights, we will use the minimum personally identifiable information possible.

This information that we collect for the research will include:
- The information from our focus group will be audio recorded and transcribed by a third party
- The information from your completed questionnaire
- Your name and contact details

We will use your personal information only to do the research. Other authorised individuals may check your records to make sure that the research is being done properly.

Your name and contact details will be kept separate from the other information that we obtain from you for this research. Your research records will contain a unique code number instead, so you are not directly identifiable when we use your data in our research.

Your name and contact details and research records will be stored electronically on a secure server at the University of York. Access to this information will be restricted to authorised persons only.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data for at least ten years. Personal data including consent forms will be kept for 3 years and then destroyed. This will enable us to complete our publications and reports. We will write our reports in a way that no-one can work out that you took part in the study. As soon as the interviews have been transcribed, we will destroy all recordings of these.

What are my choices about how my information is used?

The University of York is a publicly funded organisation that conducts research to improve health and healthcare services. In legal terms, we are using your information for this research as part of 'a task in the public interest'. The ability to change the data that we have collected, however, is limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate.

You can stop being part of the study at any time, without giving a reason. You may request that your data is not used in the study up to 14 days after the interview. We will destroy your data upon this request. After this time, data gathered may have started to be analysed and used in the study findings. Any information that has been used in the study findings cannot be withdrawn.

By taking part, you understand that in the unlikely event of a loss of capacity, the research team would retain the information that you have provided and will continue to use it confidentially in connection with the purposes of the study.

Where can I find out more about how my information is used?

You can find out more about how we use your information

- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/
- At https://www.york.ac.uk/healthsciences/research/trials/trials-gdpr/research-partcipants/
- by sending an email to nrv503@york.ac.uk
  - by emailing the University of York's data protection officer on dataprotection@york.ac.uk

Will my taking part in this study be kept confidential?

If you decide to take part in the study, what you tell us will be kept confidential and not shared with those involved in your care and treatment. No one outside the research team will know that you have taken part in the study.

We will write our reports in a way that no-one can work out that you took part in the study. Data collected for the study may be looked at by authorised persons who are organising the research. Data may also be looked at by other authorised people to check that the study is being carried out correctly. All have a duty of confidentiality to you as a research participant.

The only time we would break our duty of confidentiality is if we are worried that you – or someone else – was being, or was likely to be, harmed. **If that happens, we will talk with you about it.**

What will happen to the results of the research study?

Researchers from the University of York will analyse the material collected.

The results of the study will be published in academic journals. This may include direct quotes from you, but these will be anonymous and unable to be linked to you.

What if there is a problem?

Complaints
If you have a concern about any aspect of this study, you should ask to speak to the research team who will do their best to answer your questions (contact details below). You can also contact the Chief Investigator, Nicola O'Donnell. She will be happy to discuss your concerns.

If you are unhappy with Nicola's response, you can contact Professor Lorna Fraser, who is Head of Department (Health Sciences).

If you remain unhappy following this and you wish to complain formally, you can do this by contacting the Parliamentary and Health Service Ombudsman, who is independent of the NHS and government, at 0345 015 4033.

Data Protection
The University of York is the data controller for the information collected for this research, which means we are responsible for looking after your personal information and using it properly. If you are unhappy with the way your personal information has been handled, please contact the University's Data Protection Officer at dataprotection@york.ac.uk. If you are not satisfied with our response, you have a right to complain to the Information Commissioner's Office. For information on reporting a concern to the Information Commissioner's Office, see www.ico.org.uk/concerns.

What will happen if the study stops earlier than expected?

Should the research stop earlier than planned and you are affected in any way, we will tell you and explain why.

Thank you so much for reading this information sheet. Please keep this copy.

Get in touch with the research team:

Nicola O'Donnell

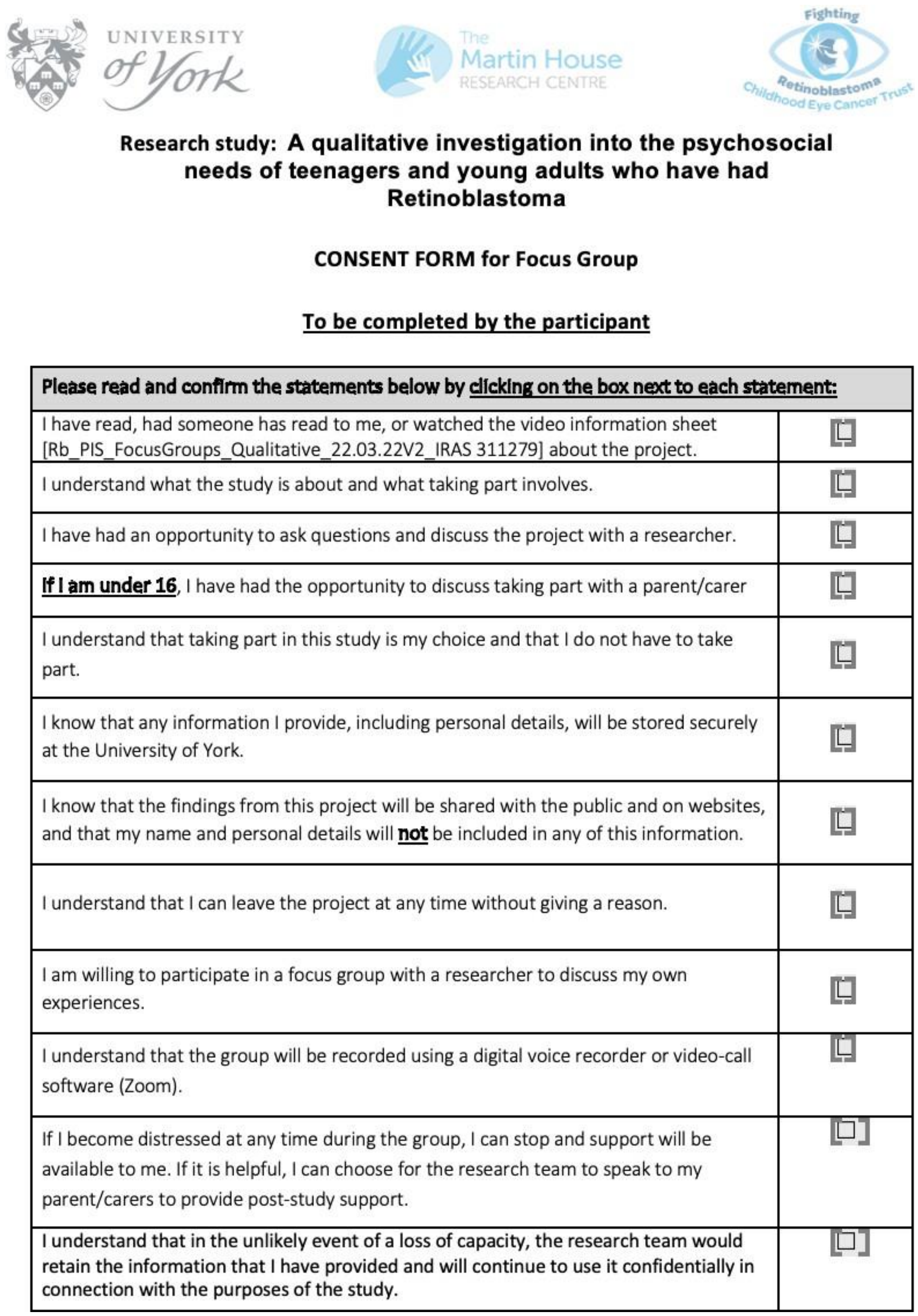
Nrv503@york.ac.uk

Study funding and approvals:

The University of York is the sponsor for this study.

The study is funded by the Childhood Eye Cancer Trust (Study Reference: 311279).

Appendix A9: Consent form for teenagers completing focus groups, version 3, 10.05.22



UNIVERSITY of York

The Martin House RESEARCH CENTRE

Fighting Retinoblastoma Childhood Eye Cancer Trust

Research study: A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

CONSENT FORM for Focus Group

To be completed by the participant

Please read and confirm the statements below by clicking on the box next to each statement:	
I have read, had someone has read to me, or watched the video information sheet [Rb_PIS_FocusGroups_Qualitative_22.03.22V2_IRAS 311279] about the project.	☐
I understand what the study is about and what taking part involves.	☐
I have had an opportunity to ask questions and discuss the project with a researcher.	☐
If I am under 16, I have had the opportunity to discuss taking part with a parent/carer	☐
I understand that taking part in this study is my choice and that I do not have to take part.	☐
I know that any information I provide, including personal details, will be stored securely at the University of York.	☐
I know that the findings from this project will be shared with the public and on websites, and that my name and personal details will **not** be included in any of this information.	☐
I understand that I can leave the project at any time without giving a reason.	☐
I am willing to participate in a focus group with a researcher to discuss my own experiences.	☐
I understand that the group will be recorded using a digital voice recorder or video-call software (Zoom).	☐
If I become distressed at any time during the group, I can stop and support will be available to me. If it is helpful, I can choose for the research team to speak to my parent/carers to provide post-study support.	☐
I understand that in the unlikely event of a loss of capacity, the research team would retain the information that I have provided and will continue to use it confidentially in connection with the purposes of the study.	☐

I agree to take part in the above study.	☐
I would like to be sent a summary of the study findings (optional – if yes, please provide an email address here [Click or tap here to enter text]	☐

Please complete, sign and date below:

Your name: Click or tap here to enter text

Your electronic signature:

Click or tap here to enter text

Date: Click or tap here to enter text.

For the research team to complete:

Name of researcher taking or checking consent:

....

Researcher signature: .... Date: ....

Thank you for agreeing to take part in this study.

You will receive a copy of your signed consent form by email.

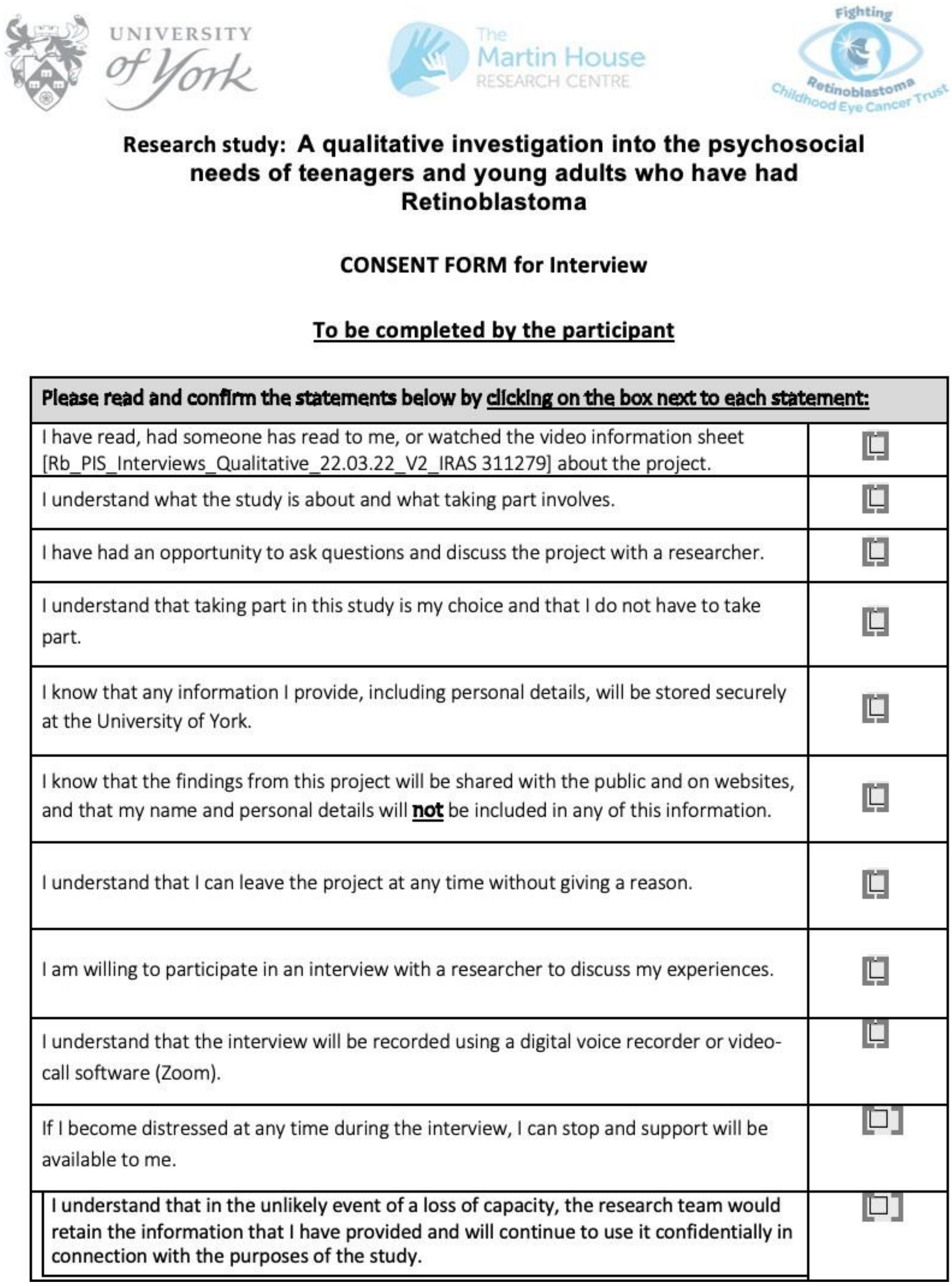
If you have any questions, please contact:

Research Team

Nicola O'Donnell

Nrv503@york.ac.uk

Appendix A10: Consent form for young adults completing individual interviews, version 1, 20.12.21



UNIVERSITY of York

The Martin House RESEARCH CENTRE

Fighting Retinoblastoma Childhood Eye Cancer Trust

Research study: A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma

CONSENT FORM for Interview

<u>To be completed by the participant</u>

Please read and confirm the statements below by <u>clicking on the box next to each statement:</u>	
I have read, had someone has read to me, or watched the video information sheet [Rb_PIS_Interviews_Qualitative_22.03.22_V2_IRAS 311279] about the project.	☐
I understand what the study is about and what taking part involves.	☐
I have had an opportunity to ask questions and discuss the project with a researcher.	☐
I understand that taking part in this study is my choice and that I do not have to take part.	☐
I know that any information I provide, including personal details, will be stored securely at the University of York.	☐
I know that the findings from this project will be shared with the public and on websites, and that my name and personal details will **<u>not</u>** be included in any of this information.	☐
I understand that I can leave the project at any time without giving a reason.	☐
I am willing to participate in an interview with a researcher to discuss my experiences.	☐
I understand that the interview will be recorded using a digital voice recorder or video-call software (Zoom).	☐
If I become distressed at any time during the interview, I can stop and support will be available to me.	☐
I understand that in the unlikely event of a loss of capacity, the research team would retain the information that I have provided and will continue to use it confidentially in connection with the purposes of the study.	☐

I agree to take part in the above study.	☐
I would like to be sent a summary of the study findings (optional – if yes, please provide an email address here [Click or tap here to enter text]	☐

Please complete, sign and date below:

Your name: [Click or tap here to enter text]

Your electronic signature:

[Click or tap here to enter text]

Date: Click or tap here to enter text.

For the research team to complete:

Name of researcher taking or checking consent:

……

Researcher signature: …………………………………………………………… Date: …………………………………

Thank you for agreeing to take part in this study.

You will receive a copy of your signed consent form by email.

If you have any questions, please contact:

Research Team

Nicola O'Donnell

Nrv503@york.ac.uk

Appendix A11: Original recruitment poster for social media, version 1, 18.01.22

Fighting Retinoblastoma Childhood Eye Cancer Trust

University of York

The Martin House Research Centre

ARE YOU LIVING BEYOND RETINOBLASTOMA?

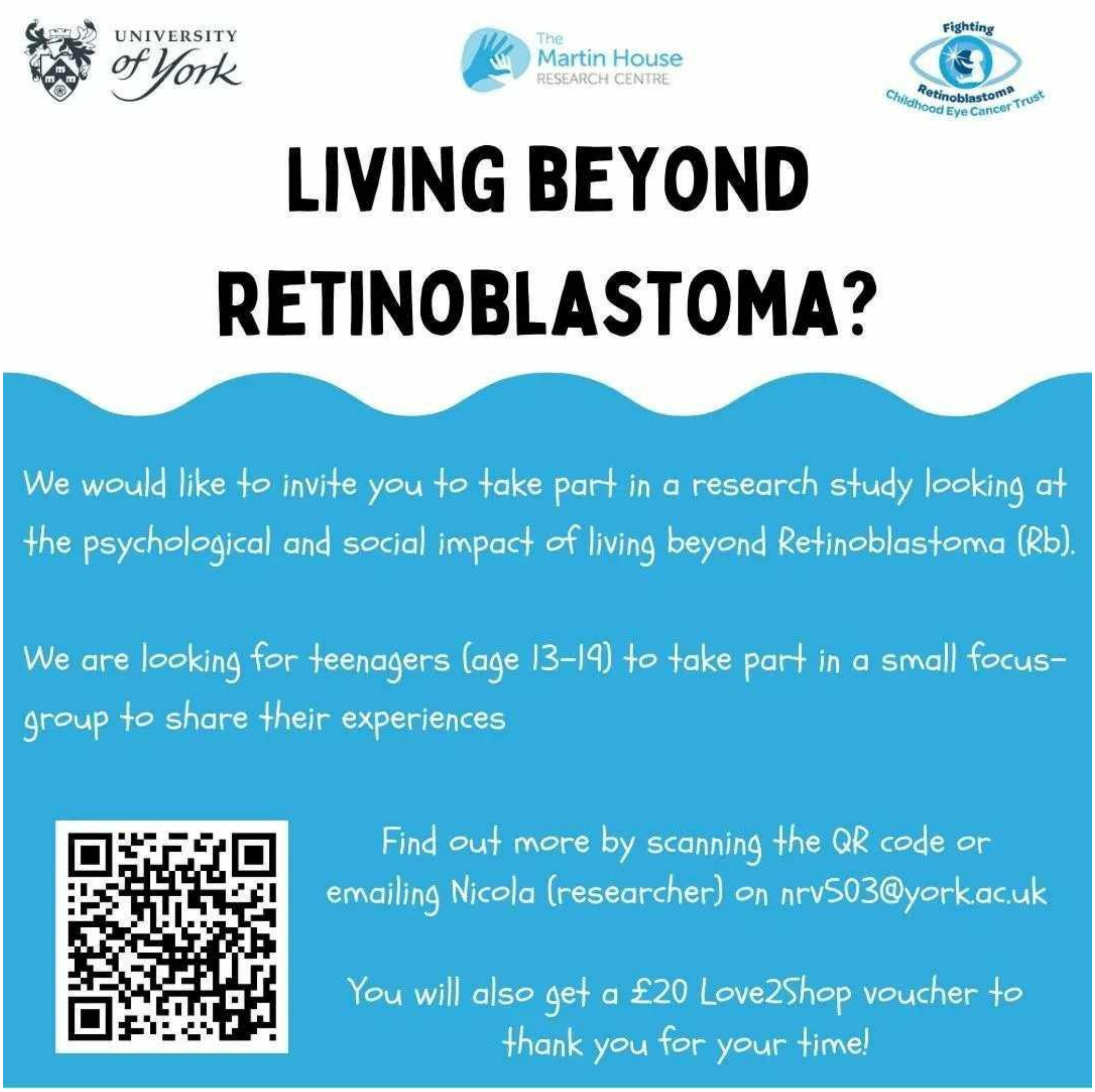
We would like to invite you to take part in a research study looking at the psychological and social impact of living beyond Retinoblastoma (Rb). There is limited research in this area, partly because of the small number of children diagnosed each year.

We are looking for **teenagers (age 13-19)** to take part in a small focus-group to share their experiences

and **young adults (age 20-29)** to take part in individual interviews focused on the psychological support that they wish they had had access to.

To find out more, please contact Nicola O'Donnell on nrv503@york.ac.uk

Appendix A12: Second recruitment poster for social media, version 1, 01.11.22



UNIVERSITY of York

The Martin House RESEARCH CENTRE

Fighting Retinoblastoma Childhood Eye Cancer Trust

LIVING BEYOND RETINOBLASTOMA?

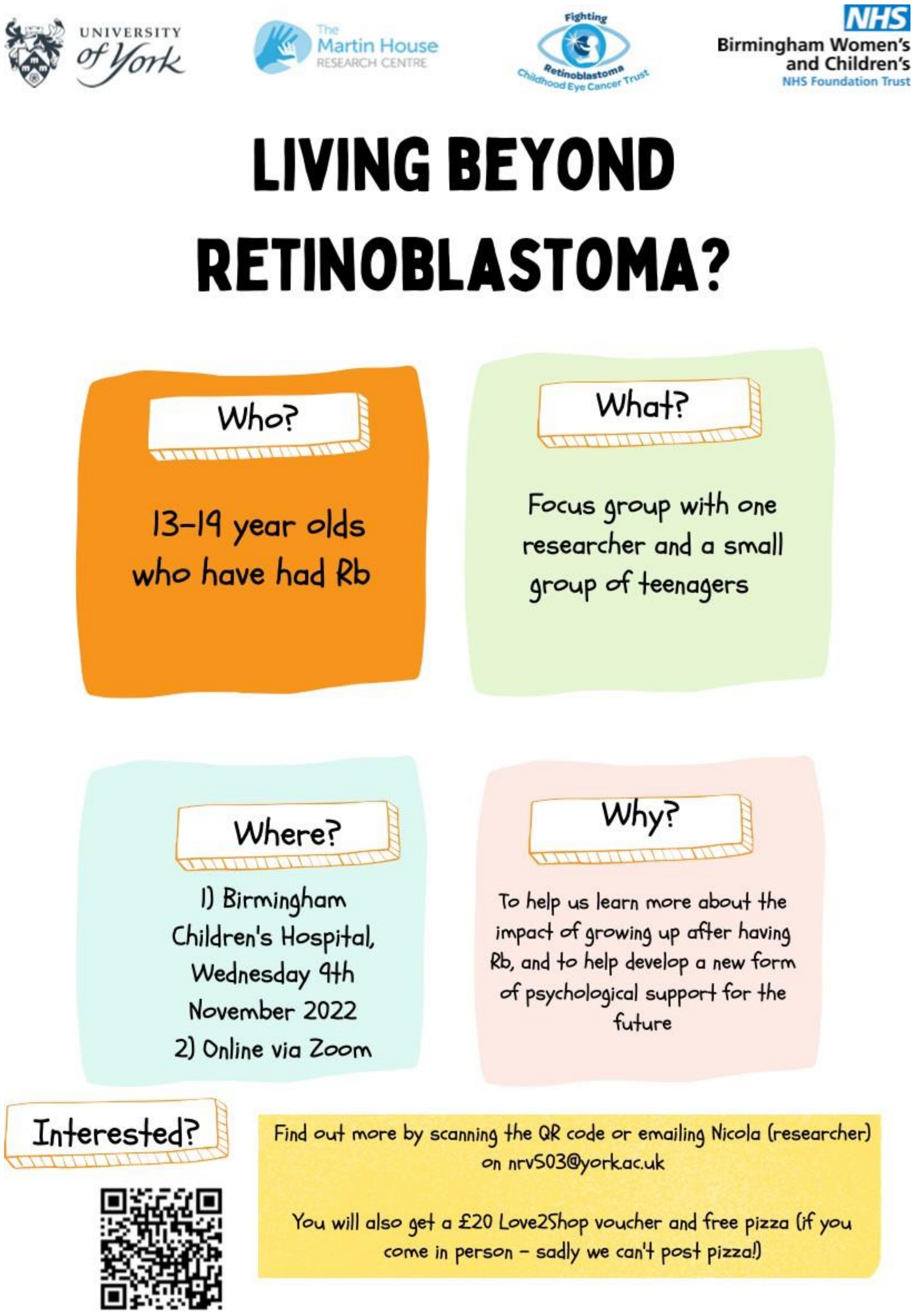
We would like to invite you to take part in a research study looking at the psychological and social impact of living beyond Retinoblastoma (Rb).

We are looking for teenagers (age 13-19) to take part in a small focus-group to share their experiences

Find out more by scanning the QR code or emailing Nicola (researcher) on nrv503@york.ac.uk

You will also get a £20 Love2Shop voucher to thank you for your time!

Appendix A13: Recruitment poster for Birmingham Children's Hospital Rb clinic waiting room, version 1, 07.09.22



University of York

The Martin House Research Centre

Fighting Retinoblastoma Childhood Eye Cancer Trust

NHS Birmingham Women's and Children's NHS Foundation Trust

LIVING BEYOND RETINOBLASTOMA?

Who?

13-19 year olds who have had Rb

What?

Focus group with one researcher and a small group of teenagers

Where?

1) Birmingham Children's Hospital, Wednesday 9th November 2022
2) Online via Zoom

Why?

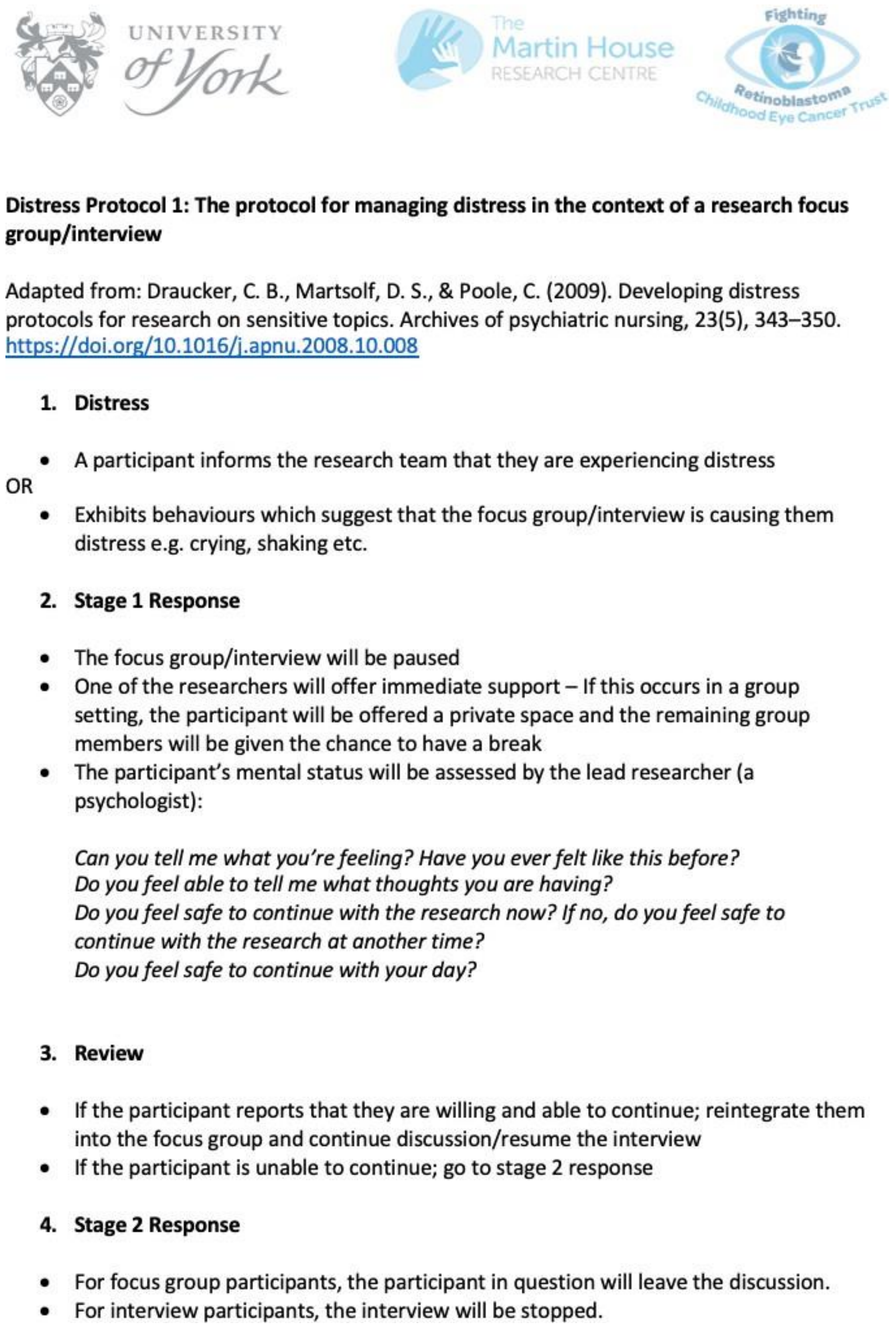
To help us learn more about the impact of growing up after having Rb, and to help develop a new form of psychological support for the future

Interested?

Find out more by scanning the QR code or emailing Nicola (researcher) on nrv503@york.ac.uk

You will also get a £20 Love2Shop voucher and free pizza (if you come in person – sadly we can't post pizza!)

APPENDIX A14: DISTRESS PROTOCOL, VERSION 1, 11.05.22



Distress Protocol 1: The protocol for managing distress in the context of a research focus group/interview

Adapted from: Draucker, C. B., Martsolf, D. S., & Poole, C. (2009). Developing distress protocols for research on sensitive topics. Archives of psychiatric nursing, 23(5), 343–350. https://doi.org/10.1016/j.apnu.2008.10.008

1. **Distress**

- A participant informs the research team that they are experiencing distress

OR

- Exhibits behaviours which suggest that the focus group/interview is causing them distress e.g. crying, shaking etc.

2. **Stage 1 Response**

- The focus group/interview will be paused
- One of the researchers will offer immediate support – If this occurs in a group setting, the participant will be offered a private space and the remaining group members will be given the chance to have a break
- The participant's mental status will be assessed by the lead researcher (a psychologist):

Can you tell me what you're feeling? Have you ever felt like this before?
Do you feel able to tell me what thoughts you are having?
Do you feel safe to continue with the research now? If no, do you feel safe to continue with the research at another time?
Do you feel safe to continue with your day?

3. **Review**

- If the participant reports that they are willing and able to continue; reintegrate them into the focus group and continue discussion/resume the interview
- If the participant is unable to continue; go to stage 2 response

4. **Stage 2 Response**

- For focus group participants, the participant in question will leave the discussion.
- For interview participants, the interview will be stopped.

- All participants will be supported to reschedule participation at a later date if they choose to and are safe to do so.
- All participants will be provided with sources of support including CHECT's designated post-study support. Details of CHECT's email (support@chect.org.uk) and phone number (0207 3775578) will be provided to all participants via the participant information sheet given prior to the study.
- All participants will be encouraged to contact their GP and Rb clinical team (if applicable)
- All participants under the age of 18 will be encouraged to discuss their distress with their parents/carers
- All participants under 16 will be informed that their parents will be contacted to share any risk issues
- If risk issues are identified for participants (of any age) recruited via hospital sites, they will be informed that these will be shared with their clinical team.

5. Follow up

- All participants will be offered a follow-up call by the lead researcher 48 hours after participation to check in
- All participants will be encouraged to contact the research team/CHECT if they are experiencing increased distress in the hours and days following the focus group/interview
- Participants under 18 will be informed that we can arrange a phone call with their parents if this is wanted by the participant

UNIVERSITY of York

The Martin House RESEARCH CENTRE

Fighting Retinoblastoma Childhood Eye Cancer Trust

Distress Protocol 2: The protocol for managing researcher distress in the context of delivering focus groups/interviews and analysis of content

1. **Pre-Data Collection**

- The research team will consider the potential psychological impact on themselves when listening to participant's experiences of childhood cancer and/or their mental health challenges
- The research team will consider how many focus groups/interviews could be undertaken in a short period of time (i.e. one week) and will space these accordingly to prevent emotional burn out

2. **Data Collection Stage**

- Two members of the research team will be present for focus groups to manage group dynamics and to provide support should the topics discussed become distressing
- De-briefing sessions will be offered regularly within the research and supervisory teams
- The research team will keep a reflective journal of their thoughts and feelings which will also become field notes

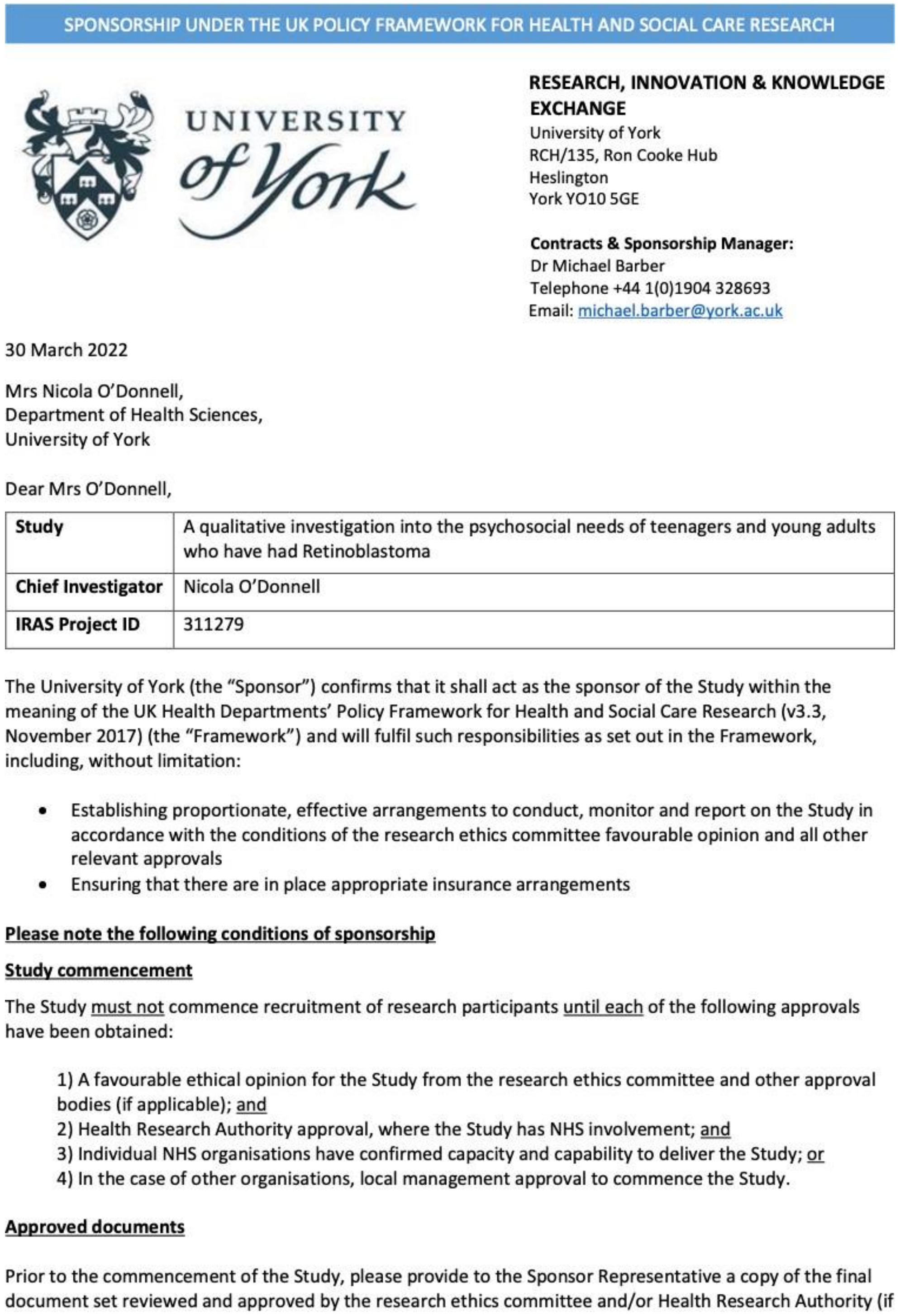
3. **Analysis**

- Focus groups/interviews with distressing content will be highlighted prior to transcription so that the analyst can prepare to hear potentially upsetting information
- De-briefing sessions will be offered regularly within the research and supervisory teams

4. **Follow-up**

- The research team will be encouraged to seek external support e.g. University counselling if they experience increased distress following the focus group/interview delivery, analysis, or transcription

APPENDIX A15: SPONSOR LETTER, VERSION 1, 30.03.22



SPONSORSHIP UNDER THE UK POLICY FRAMEWORK FOR HEALTH AND SOCIAL CARE RESEARCH

UNIVERSITY of York

RESEARCH, INNOVATION & KNOWLEDGE EXCHANGE
University of York
RCH/135, Ron Cooke Hub
Heslington
York YO10 5GE

Contracts & Sponsorship Manager:
Dr Michael Barber
Telephone +44 1(0)1904 328693
Email: michael.barber@york.ac.uk

30 March 2022

Mrs Nicola O'Donnell,
Department of Health Sciences,
University of York

Dear Mrs O'Donnell,

Study	A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma
Chief Investigator	Nicola O'Donnell
IRAS Project ID	311279

The University of York (the "Sponsor") confirms that it shall act as the sponsor of the Study within the meaning of the UK Health Departments' Policy Framework for Health and Social Care Research (v3.3, November 2017) (the "Framework") and will fulfil such responsibilities as set out in the Framework, including, without limitation:

- Establishing proportionate, effective arrangements to conduct, monitor and report on the Study in accordance with the conditions of the research ethics committee favourable opinion and all other relevant approvals
- Ensuring that there are in place appropriate insurance arrangements

Please note the following conditions of sponsorship

Study commencement

The Study must not commence recruitment of research participants until each of the following approvals have been obtained:

1) A favourable ethical opinion for the Study from the research ethics committee and other approval bodies (if applicable); and
2) Health Research Authority approval, where the Study has NHS involvement; and
3) Individual NHS organisations have confirmed capacity and capability to deliver the Study; or
4) In the case of other organisations, local management approval to commence the Study.

Approved documents

Prior to the commencement of the Study, please provide to the Sponsor Representative a copy of the final document set reviewed and approved by the research ethics committee and/or Health Research Authority (if applicable). The final document set is recorded in the letter of favourable ethical opinion and the Health Research Authority's letter of approval (if applicable).

Notifications

Throughout the course of the Study you are required to make the following notifications:

- Notify the Sponsor Representative of amendments to the Study prior to submission for research ethics committee or other relevant approval body review
- Notify the Sponsor Representative of suspected serious breaches of the research protocol or breaches of confidentiality or data security
- Notify the Sponsor Representative of adverse events exhibited by participants directly arising from the conduct of the Study in accordance with any procedures described in the research protocol
- Within 90 days of the end of study (as defined in the research protocol), notify the research ethics committee, other relevant approval bodies and the Sponsor Representative of the end of the Study
- Submit annual and end of study reports (within 12 months of the end of Study) to the research ethics committee and other relevant approval bodies, notifying the Sponsor Representative of the submission of such reports and providing copies

Yours sincerely,

Dr Michael Barber
Sponsor Representative

APPENDIX A16: FAVOURABLE ETHICAL OPINION, 19.05.22

Business Services
Organisation

Office for Research Ethics Committees
Northern Ireland (ORECNI)

Customer Care & Performance Directorate
Unit 5, Lissue Industrial Estate West
Rathdown Walk
Moira Road
Lisburn
BT28 2RF
Tel: 028 95361400
www.hscbusiness.hscni.net/services/orecni.htm

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

19 May 2022

Dr Bob Phillips
Hull-York Medical School and Centre for Reviews and Dissemination
University of York
York
YO10 5DD

Dear Dr Phillips

Study title:	**A qualitative investigation into the psychosocial needs of teenagers and young adults who have had Retinoblastoma**
REC reference:	**22/NI/0082**
Protocol number:	**2.0**
IRAS project ID:	**311279**

Thank you for your letter of 17th May 2022, responding to the Research Ethics Committee's (REC) request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm **a favourable ethical opinion** for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Providing Support to Health and Social Care

Good practice principles and responsibilities

The UK Policy Framework for Health and Social Care Research sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of research transparency:

1. registering research studies
2. reporting results
3. informing participants
4. sharing study data and tissue

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as:

- clinical trial of an investigational medicinal product
- clinical investigation or other study of a medical device
- combined trial of an investigational medicinal product and an investigational medical device
- other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice.

Failure to register a clinical trial is a breach of these approval conditions, unless a deferral has been agreed by the HRA (for more information on registration and requesting a deferral see: Research registration and research project identifiers).

If you have not already included registration details in your IRAS application form you should notify the REC of the registration details as soon as possible.

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit: https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-summaries/

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease (COVID-19) in the full title of your study. Approved COVID-19 studies can be found at: https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report
- Reporting results

The latest guidance on these topics can be found at https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites taking part in the study, subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Covering letter on headed paper [Response to REC provisional opinion]		16 May 2022
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [University of York sponsor insurance evidence]	1	27 July 2021
Interview schedules or topic guides for participants	1	20 January 2022
Interview schedules or topic guides for participants [Interview topic guide]	1	20 January 2022
IRAS Application Form [IRAS_Form_11042022]		11 April 2022
Letter from sponsor [Sponsor letter]	1	30 March 2022
Letters of invitation to participant [Invite focus groups]	2	10 May 2022
Letters of invitation to participant [Invite interviews]	2	10 May 2022
Non-validated questionnaire [Questionnaire teenagers]	2	10 May 2022
Other [Interview consent]	1	12 December 2021
Other [PPI recruitment poster]	1	18 January 2022
Other [Questionnaire Young adults]	2	10 May 2022
Other [PIS focus groups]	3	10 May 2022
Other [Distress Protocol]	1	11 May 2022
Other [Parents PIS for teenage participants]	1	10 May 2022
Other [Alternative PIS Video Links]	1	18 May 2022
Participant consent form [Focus group consent form]	1	12 December 2021
Participant consent form [Interview consent form]	1	12 December 2021
Participant information sheet (PIS) [Participant Information sheet Interviews]	3	10 May 2022
Research protocol or project proposal [Protocol]	3	11 May 2022
Summary CV for Chief Investigator (CI) [CV CI]		18 June 2021
Summary CV for student [CV CI]		22 March 2022
Summary CV for supervisor (student research) [Supervisor CV]		18 June 2021

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at: https://www.hra.nhs.uk/planning-and-improving-research/learning/

IRAS project ID: 311279   Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

pp Professor Patrick Murphy
HSC REC B Chair

Email:recb@hscni.net

Enclosures: "After ethical review – guidance for researchers" *[SL-AR2]*

Copy to: Mr Michael Barber

Appendix A17: Focus group topic guide for teenagers, version 1, 20.01.22

Introductory Questions

Researchers will introduce themselves and ask the group to share their names. Participants will then be reminded about the study aims, confirming their consent to take part, and reiterating the right to withdraw from the study. Participants will also be told how long the group will last and given the opportunity to ask questions. After this, the audio recording will begin.

My name is Nicola and I am the PhD student running this project. We will chat for between 1-2 hours today, depending on how much you want to chat about.

I just want to double check again that you consent to taking part? This is voluntary and you can withdraw at any time during the interview and afterwards.

Do you agree that anonymised quotes can be used?

And do you consent for this interview to be audio recorded?

There is a risk that asking questions about cancer experiences and psychological wellbeing may be upsetting; if you need a break or want to stop, just let me know.

Any questions?

1. **Understanding of Rb**

   What do you understand about Rb?
   - Do you know about the two different types (heritable and non-heritable)?
   - Do you know which type you have?

2. **Finding out about individual experiences**

   How old were you when your Rb was diagnosed? Can you tell me what you remember about this time and your treatment?

   Are you living with any other health conditions as a result of Rb?

3. **The impact of Rb on what individuals think, feel, and do**

   Some teenagers feel that having had Rb makes them feel different. Can you tell me, if at all, about how Rb has impacted how you feel about yourself?
   - If not mentioned, ask about: image, worrying about what others think, visual impairment.

4. Some teenagers feel that Rb has influenced what they do. Can you tell me about how Rb has impacted how you live your life?
    - If not mentioned, ask about: impact of worry on behaviour, having to attend follow-up appointments, thoughts about the future e.g. having a relationship and/or children, second cancers

5. **Psychological support**

   Has there ever been a time when you felt you would like some support for how your mental health, or how you are thinking or feeling?
    - If yes, did you receive any?
    - If yes, can you tell me about that? Was it helpful?

   If you have not accessed any psychological support, but think it would be helpful, can you tell me what you think this should look like?

6. **Intervention Planning**

   We are planning to design a psychoeducation intervention to provide support to young people living beyond Rb (explain what psychoeducation interventions mean). How do you think researchers could design psychological support resources to help young people manage how they feel?
    - If not mentioned, talk about: group work vs. individual work, technological interventions, accessibility

7. **Closing questions**

   Do you want to say more about the topics we have discussed? Is there anything that we've missed that you would like to talk about?

Debriefing will be offered to participants immediately after the focus group discussions and a telephone number of Nicola O'Donnell will be provided in case further discussions are wanted. All medical queries raised by the participants during the focus group discussions will be redirected to their clinical care team.

- I will now give you a £20 retail voucher to thank you for your time

- All participants will be provided with sources of support including CHECT's designated post-study support. Details of CHECT's email (support@chect.org.uk) and phone number (0207 3775578) will be provided to all participants via the participant information sheet given prior to the study.
- All participants will be encouraged to contact their GP and Rb clinical team (if applicable)

- All participants under the age of 18 will be encourage to discuss their distress with their parents/carers
- All participants under 16 will be informed that their parents will be contacted to share any risk issues
- If risk issues are identified for participants (of any age) recruited via hospital sites, they will be informed that these will be shared with their clinical team.

Can I take your best contact number to conduct a follow-up call in a couple of days?/your parents are also welcome to speak to me too

- All participants will be encouraged to contact the research team/CHECT if they are experiencing increased distress in the hours and days following the focus group/interview

APPENDIX A18: INTERVIEW TOPIC GUIDE FOR YOUNG ADULTS, VERSION 1, 20.01.22

Introductory Questions

Participants will be reminded about the study aims, confirming their consent to take part, and reiterating the right to withdraw from the study. Participants will also be told how long the interview will last and given the opportunity to ask questions. After this, the audio recording will begin.

My name is Nicola and I am the PhD student running this project. We will chat for about an hour today, depending on how much you want to chat about.

I just want to double check again that you consent to taking part? This is voluntary and you can withdraw at any time during the interview and afterwards.

Do you agree that anonymised quotes can be used?

And do you consent for this interview to be recorded?

There is a risk that asking questions about cancer experiences and psychological wellbeing may be upsetting; if you need a break or want to stop, just let me know.

Any questions?

1. **Finding out about the individual and their experiences**

   Example opening script "To begin, would you mind telling me a little bit about yourself and your experience with Rb?"

   Probes:

   - What do you understand about Rb?
   - Do you know about the two different types (heritable and non-heritable)?
   - Do you know which type you have?
   - How old were you when your Rb was diagnosed?
   - Can you tell me what you remember about this time and your treatment?
   - Are you living with any health conditions as a result of Rb?

2. The impact of Rb on what individuals think, feel, and do

Exploration of individual's experience of Rb on their wellbeing. Example opening script "Now we are going to move on to talk a bit about the impact of Rb on your mental health, is that ok?"

Probes:

- Do you, or have you ever, have any worries about the way you feel/ your mental health?
- Has Rb impacted how you feel about yourself? Is this the same as when you were a teenager?

If no, how is this different?

- Has Rb impacted how you live your life? Is this the same as when you were a teenager?

If no, how is this different?

- Thoughts/feelings associated with Rb, both now and during adolescence

3. Psychological Support

Probes:

Has there ever been a time when you felt you would like some support for how your mental health, or how you are thinking or feeling?

  - If yes, did you receive any? (Particularly during adolescence)
  - If yes, can you tell me about that? Was it helpful?

If you have not accessed any psychological support, but think it would have been helpful, can you tell me what you think this should look like?

- Do you have any worries about Rb?
- Recurrence?
- Fertility?

4. Follow-up questions on target topic areas

- Views on mental health impact during adolescence vs. now
- Views on appropriate psychological and social support for people living beyond Rb
- Anything else that would be helpful if you had a dream support package?
- Content?
- How to access this?
- What would you say to your younger self if you could?

5. Close

- Ask if there is anything else that they would like to discuss or go back to
- Ask if they have any questions
- Express thanks and appreciation for time

Debriefing will be offered to participants immediately after the interview. All medical queries raised by the participants during the focus group discussions will be redirected to their clinical care team.

- I will now send you a £20 retail voucher to thank you for your time – please provide me with your address to send this to

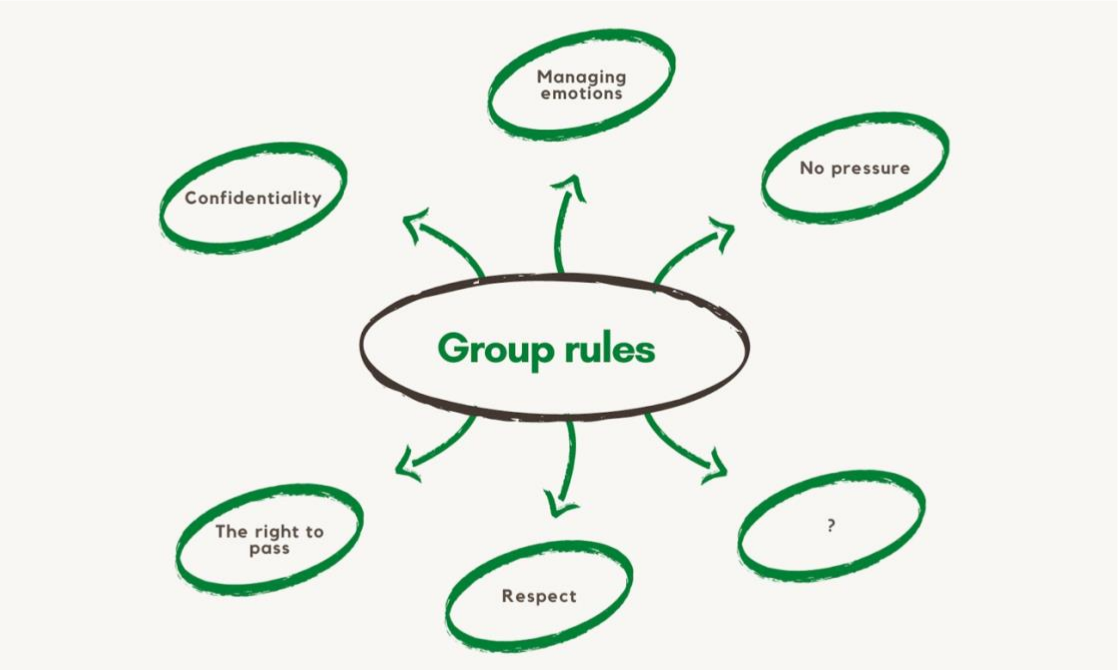
- All participants will be provided with sources of support including CHECT's designated post-study support. Details of CHECT's email (support@chect.org.uk) and phone number (0207 3775578) will be provided to all participants via the participant information sheet given prior to the study.
- All participants will be encouraged to contact their GP and Rb clinical team (if applicable)
- All participants under the age of 18 will be encourage to discuss their distress with their parents/carers
- All participants under 16 will be informed that their parents will be contacted to share any risk issues
- If risk issues are identified for participants (of any age) recruited via hospital sites, they will be informed that these will be shared with their clinical team.

- All participants will be offered a follow-up call by the lead researcher 48 hours after participation to check in
- All participants will be encouraged to contact the research team/CHECT if they are experiencing increased distress in the hours and days following the focus group/interview
- Participants under 18 will be informed that we can arrange a phone call with their parents if this is wanted by the participant

Can I take your best contact number to conduct a follow-up call in a couple of days?/your parents are also welcome to speak to me too

- All participants will be encouraged to contact the research team/CHECT if they are experiencing increased distress in the hours and days following the focus group/interview

APPENDIX A19: GROUP RULES USED IN FOCUS GROUP DISCUSSIONS



Appendix A20: Email sent to participants following ethical concern, 27.03.23

Dear [NAME],

I am getting in touch as you took part in my research study, investigating the psychosocial impact of having had Retinoblastoma (Rb).

As part of this you kindly filled in some questionnaires for me, and one question asked you which type of Rb you have (genetic/non genetic).

Now that we have closed the study, I have reviewed all of the data and our team have become aware that the phrase 'genetic' might have been unclear, and led to potentially some misunderstanding.

We intended 'genetic' to mean 'retinoblastoma with mutations in the Rb gene', which is important to understand for healthy lives in the future. We have realised that you may be unclear on the type of Rb that you have, either through writing ‘unknown’ or that you have told us that you have bilateral disease which is non-genetic, which requires clarification. In some instances, we understand that you may have been mistaken in what was said or written.

There is no cause to be concerned, but we would encourage you to get in touch with your Rb team at London/Birmingham to clarify the type of Rb that you have. It is important that you have accurate information about your condition, which differs depending on the type of Rb that you have had.

London:

The Judith Kingston Retinoblastoma Service
The Royal London Hospital
E1 1FR

Tel: 020 359 41419

Birmingham:

Retinoblastoma Service
Birmingham Children’s Hospital
B4 6NH

Tel: 0121 333 8411

I would also like to remind you of the following sources of support, should you need them now or in the future:

Childhood Eye Cancer Trust (CHECT): support@chect.org.uk, 0207 3775578
Young Minds (for teenagers): text line, text ‘YM’ to 85258 for free 24/7 support
Shout (for adults): text line, text ‘SHOUT’ to 85258 for free 24/7 support
NHS psychological therapies: https://www.nhs.uk/service-search/mental-health/find-a-psychological-therapies-service/
IAM (Teenage Cancer Trust): emotional and clinical support tool for young people who’ve had cancer: https://www.teenagecancertrust.org/help-and-support/apps-and-tools/iam-emotional-and-clinical-support-tool

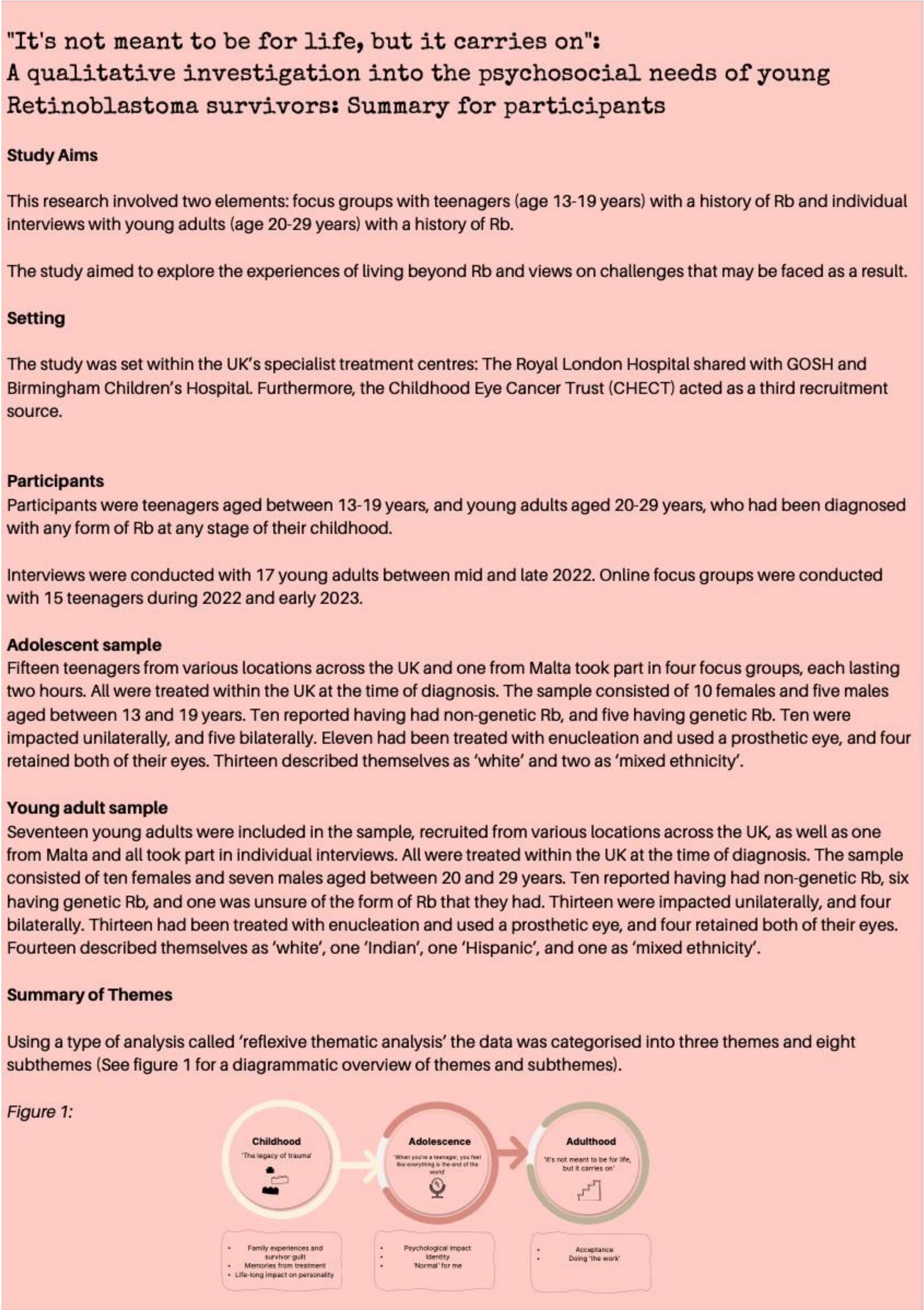
Best wishes,

Nicola

Nicola O'Donnell
Second year doctoral researcher

Epidemiology and Cancer Statistics Group
Department of Health Sciences
Faculty of Sciences
University of York
Heslington
YO10 5DD

Appendix A21: Results summary sent to PPI group



"It's not meant to be for life, but it carries on": A qualitative investigation into the psychosocial needs of young Retinoblastoma survivors: Summary for participants

Study Aims

This research involved two elements: focus groups with teenagers (age 13-19 years) with a history of Rb and individual interviews with young adults (age 20-29 years) with a history of Rb.

The study aimed to explore the experiences of living beyond Rb and views on challenges that may be faced as a result.

Setting

The study was set within the UK's specialist treatment centres: The Royal London Hospital shared with GOSH and Birmingham Children's Hospital. Furthermore, the Childhood Eye Cancer Trust (CHECT) acted as a third recruitment source.

Participants

Participants were teenagers aged between 13-19 years, and young adults aged 20-29 years, who had been diagnosed with any form of Rb at any stage of their childhood.

Interviews were conducted with 17 young adults between mid and late 2022. Online focus groups were conducted with 15 teenagers during 2022 and early 2023.

Adolescent sample

Fifteen teenagers from various locations across the UK and one from Malta took part in four focus groups, each lasting two hours. All were treated within the UK at the time of diagnosis. The sample consisted of 10 females and five males aged between 13 and 19 years. Ten reported having had non-genetic Rb, and five having genetic Rb. Ten were impacted unilaterally, and five bilaterally. Eleven had been treated with enucleation and used a prosthetic eye, and four retained both of their eyes. Thirteen described themselves as 'white' and two as 'mixed ethnicity'.

Young adult sample

Seventeen young adults were included in the sample, recruited from various locations across the UK, as well as one from Malta and all took part in individual interviews. All were treated within the UK at the time of diagnosis. The sample consisted of ten females and seven males aged between 20 and 29 years. Ten reported having had non-genetic Rb, six having genetic Rb, and one was unsure of the form of Rb that they had. Thirteen were impacted unilaterally, and four bilaterally. Thirteen had been treated with enucleation and used a prosthetic eye, and four retained both of their eyes. Fourteen described themselves as 'white', one 'Indian', one 'Hispanic', and one as 'mixed ethnicity'.

Summary of Themes

Using a type of analysis called 'reflexive thematic analysis' the data was categorised into three themes and eight subthemes (See figure 1 for a diagrammatic overview of themes and subthemes).

Figure 1:

Theme 1: Childhood – ***'the legacy of trauma' (P27, B, H)***

One potential impact arising from Rb diagnosis is life-long trauma, which is an issue that is clearly highlighted within almost all participant narratives. Variations of this legacy appear to be influenced by the age a child is diagnosed, the genetic nature of the diagnosis, wider family history of the condition, the severity of visual and facial impact, and late affects from the treatment received. The legacy of trauma theme encapsulates 'family experiences and survivor guilt', which considers the role of parents and siblings in the trauma experience. It also considers 'memories from treatment', and how these link to the individuals' emotional response and understanding of the world and others around them. Lastly, the combination of these trauma experiences and 'the long-lasting impact on personality' are considered. This is because individuals commonly spoke about continuing to carry the legacy of their experiences into adolescence and young adulthood. This understanding helps us to best conceptualise the level of psychosocial support that may be most beneficial.

Theme 2: Adolescence – ***'when you're a teenager, you feel like everything is the end of the world' (P28, U, NH)***

Adolescence was widely considered the most difficult period of life and a time when support skills are required, and this theme encapsulates three subthemes. The first, 'psychological, social, and behavioural impact', reflects on participants' feelings of unfairness and powerlessness at both the perceived and actual restrictions caused by the Rb. It also reveals how being the survivor of a rare cancer can make it hard to express yourself, as many anxieties and difficulties are so unique that it is hard to find someone who has experienced the same or a similar trauma, who can relate to this. This ability to share feelings and be understood is a vital part of upholding good self-esteem, and without it left many feeling lonely.

The subsequent influence of these thoughts and feelings on personal behaviour are also considered. Finally, the conflict of feeling lucky to be alive and of downward comparison to those less fortunate are discussed. The second subtheme, 'identity', explores narratives common amongst teenagers, including discovery of who you are and how that fits with how others perceive you. Lastly, the subtheme, 'normal for me', considers how limited vision and/or wearing a prosthetic eye shapes life, the expectation that others openly and freely comment on what you look like without invitation, as well as asking for information about you, and what it is like to not know any different.

Theme 3: Adulthood – ***'it's not meant to be for life, but it carries on' (P5, U, NH)***

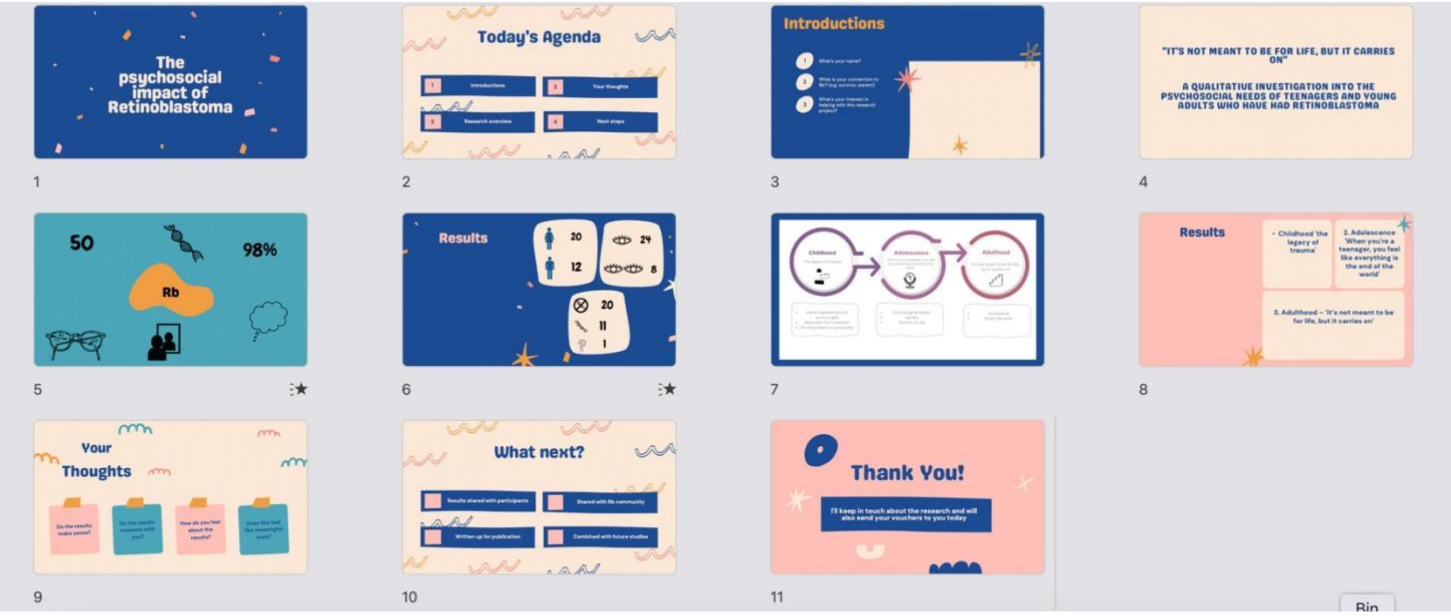
Adulthood was both a time for reflection and projection, dependent on the life stage of the individual participant. This theme is divided into two subthemes, the first being 'acceptance', a state of being that was universally considered to be unachievable whilst still young. Reasons why are discussed, and include a lack of choice, being able to validate yourself, and the power of seeing others who are like you. This led into the second subtheme, 'doing the work', encapsulating the need to seek out information to answer unknown questions, as well as the role of strategies such as therapy and peer support in improving psychosocial wellbeing.

Nicola O'Donnell, Bob Phillips, Jess Morgan, & Debra Howell

Epidemiology and Cancer Statistics Group, University of York
Centre for Reviews and Dissemination, University of York
The Hull York Medical School, University of York

✉ nicola.odonnell@york.ac.uk
𝕏 @nicolaod_
landofnod__

Appendix A22: PPI group meeting PowerPoint slides, 25th July 2023



The psychosocial impact of Retinoblastoma

Today's Agenda

1 Introductions

2 Research overview

3 Your thoughts

4 Next steps

Introductions

1 What's your name?

2 What is your connection to Rb? (e.g. survivor, parent)

3 What's your interest in helping with this research project?

"IT'S NOT MEANT TO BE FOR LIFE, BUT IT CARRIES ON"

A QUALITATIVE INVESTIGATION INTO THE PSYCHOSOCIAL NEEDS OF TEENAGERS AND YOUNG ADULTS WHO HAVE HAD RETINOBLASTOMA

Results

Results

- Childhood 'the legacy of trauma'

2. Adolescence 'When you're a teenager, you feel like everything is the end of the world'

3. Adulthood – 'It's not meant to be for life, but it carries on'

Your Thoughts

Do the results make sense?

Do the results resonate with you?

How do you feel about the results?

Does this feel like meaningful work?

What next?

Results shared with participants

Shared with Rb community

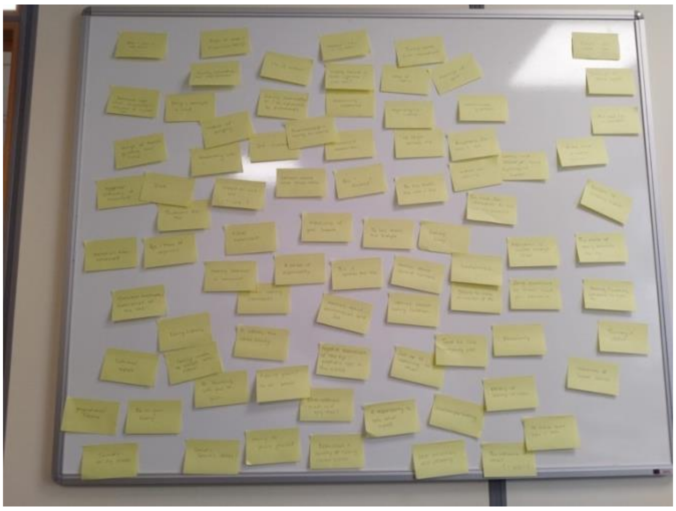
Written up for publication

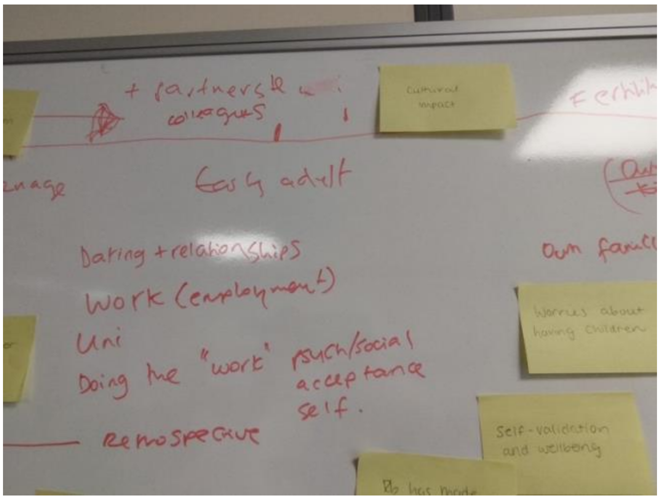
Combined with future studies

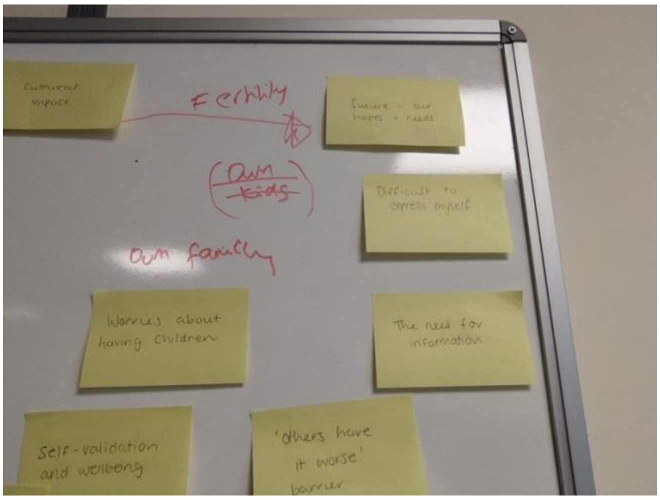
Thank You!

I'll keep in touch about the research and will also send your vouchers to you today

Appendix A23: Mapping of Themes with wider research team, 9th May 2023







Appendix A24: Letter to editor published in BMJ ADC, May 2023

Letters

Fraudulent participants in qualitative child health research: identifying and reducing bot activity

We are researchers working within paediatric health sciences across the UK. We would like to raise our concerns about a novel, seemingly growing issue within participant recruitment for qualitative research in child health; fraudulent participants, 'bots', also known as malicious automated software, and 'human bots', people paid by third parties to undermine studies.[1] We want to raise awareness of the impact that this is having at all levels; to young people who are genuinely impacted by a condition, public understanding of health, integrity of scientific research, development of evidence-based child health practice and policy, and ourselves as researchers.

Although there is literature on this topic, this has focused on questionnaire studies, where ineligible individuals complete online research to profit from incentives.[2] Individually, we have all experienced something different; fraudulent individuals posing as young people and caregivers completing screening questionnaires and committing to joining online focus groups or interviews. Said participants have attempted to bypass screening measures designed to prevent such activity (eg, asking specific questions to check for genuine, consistent responses; a known method of detecting fraudulent participants or 'bots'). It must be noted that in all studies, recruitment occurred across numerous social media platforms (including Twitter, Facebook and Instagram), with voucher payments offered as reimbursement in most studies (ranging from £20 to £40).

What we are experiencing is bizarre and concerning; the sophistication of the messages received goes beyond recognised language of automated software, mirroring human communication and referencing study details. This has the potential to undermine the integrity of key data and compromises researcher and genuine participant safety, should these fraudulent individuals end up in research settings. One of us had over 150 fraudulent expressions of interest in less than 24 hours. This wastes time and takes up valuable, often scarce resources. Furthermore, the fabrication of serious health conditions (in our experience cancer, gastrointestinal conditions and eating disorders) is worrying. This is not a new phenomenon, but using this to influence major research studies may be. We should also highlight that all of the affected researchers are young women, another factor which may influence the type of research targeted in this way.

Below, we highlight a series of 'red flags' to support identification of fraudulent participants in child health research (table 1). These are based on our experiences and are not all-encompassing. We propose that a single 'red flag' is not enough to identify a fraudulent participant, but instead, may raise suspicion, prompting further follow-up. Our attempts to address these have included further screening with participants via questionnaires, video or phone calls. Out of 483 expressions of interest, we identified at least 385 fraudulent participants using enhanced screening (80%). Our experiences highlight the importance of rigorous screening, prior to informed consent, to support the rigour of qualitative child health research. Financial incentives can increase the rate of response from participants[3] but may also motivate ineligible participants to deceive about their eligibility for study enrolment to secure payment.[4] To mitigate the risk of fraudulent activity, we recommend that any financial incentives are omitted from advertisements shared on social media.

Nicola O'Donnell ,[1,2] **Rose-Marie Satherley,**[2] **Emily Davey,**[3] **Gemma Bryan**[4]

[1]Epidemiology and Cancer Statistics Group, University of York, York, UK
[2]School of Psychology, University of Surrey, Guildford, UK
[3]Great Ormond Street Institute of Child Health, University College London, London, UK
[4]School of Health Science, University of Surrey, Guildford, UK

Correspondence to Nicola O'Donnell, Epidemiology and Cancer Statistics Group, University of York, York, UK; nrv503@york.ac.uk

Twitter Nicola O'Donnell @nicolaod_, Emily Davey @emilysarahdavey and Gemma Bryan @ drgemmabryan

Contributors NO'D raised the issue, conceived the letter and cowrote the letter. R-MS cowrote the letter and devised the table. ED cowrote the letter. GB cowrote the letter.

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Table 1

Red flag	Example
Prestudy communications with research team	Emails/contact from potential participants which do not include a preamble. Participants typically describe their reasons for interest in the study (eg, '*I/My child was diagnosed with [condition] in 2016, and I saw information about this research project…*' When a preamble is not provided, a red flag may be raised (eg, simply stating '*I want to take part in the study.*'). Correspondence that are lacking in pertinent details about the participant (eg, '*I am a carer and would like to take part in the survey*'). Correspondence that appears overly formal and generic, with no mention of the specific study (eg, '*Good morning researcher, I am emailing you to express my interest and eligibility in a focus group you are conducting.*'). Correspondence that does not include salutations and or valedictions, or that does not address the researcher by name (eg, '*I am indicating my interest in your study*' or '*I am eligible for your study*').
Details around health conditions that appear implausible	Being a carer for multiple children with a rare disease or being diagnosed with a disease far outside the usual age-range for that condition. Reporting the diagnosis of a condition in later years, that is typically diagnosed in early childhood (eg, in a paediatric cancer that is almost always diagnosed at age <5, reporting diagnosis at age 17).
Details around medical support appear vague, incorrect or are falsified	When asked for the hospital that a young person received treatment at, responding with '*private clinic*', a hospital that does not exist (in the UK or worldwide) or in the case of rare diseases, a hospital that is not known for treating the condition.
Email addresses and communications appear to follow a generic format across multiple enquiries	There may be [no title] in the description of the email/messaging, a direct copy of the entire study advert in the title of the email or email addresses that appear unlikely or are very similar/the same as the name of another 'participant'.
Provision of a phone number that is unobtainable	Contact numbers that do not exist, redirect to an incorrect number or are unanswerable.

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ORCID iD

Nicola O'Donnell http://orcid.org/0000-0003-0661-4129

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APPENDIX A25: ARTICLE FROM STUDY ONE PUBLISHED IN BMJ OPEN, APRIL 2024

Open access **Original research**

BMJ Open

'It's not meant to be for life, but it carries on': a qualitative investigation into the psychosocial needs of young retinoblastoma survivors

Nicola O'Donnell ,[1] Bob Phillips ,[2] Jessica Elizabeth Morgan ,[2]
Debra Howell [1]

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[1]Department of Health Sciences, University of York, York, UK
[2]Centre for Reviews and Dissemination, University of York, York, UK

Correspondence to
Nicola O'Donnell;
nicola.odonnell@york.ac.uk

ABSTRACT

Objective and design Retinoblastoma (Rb) is a rare childhood eye cancer, with 45% of individuals impacted by heritable disease and the remainder impacted non-heritably. The condition can leave survivors with life-long psychological and social challenges. This qualitative study examined the psychosocial needs of teenagers and young adults living beyond Rb.

Setting A qualitative, exploratory study was conducted using focus groups with teenagers and interviews with young adults. Participants were recruited via the Childhood Eye Cancer Trust and the two national Rb treatment centres in the UK. Reflexive thematic analysis was used to analyse data using exploratory and inductive methods.

Participants 32 young survivors of Rb (10 heritable, 21 non-heritable, 1 unknown; 23 unilateral, 9 bilateral) aged between 13 and 29 years (12 male, 20 female).

Results Data were rich and spanned the life course: three key themes were generated, containing eight subthemes. Theme 1 describes participants' experiences of childhood and trauma, including survivor guilt, memories from treatment and impact on personality. Theme 2 focuses on the challenges of adolescence, including the psychological impact of Rb, the impact on identity, and the sense of normality and adaptation to late effects. The third theme considered adulthood and the development of acceptance, a state of being widely considered unachievable during childhood, as well as the 'work' needed to feel supported, including seeking out information, peer support and therapeutic strategies.

Conclusions This study provides in-depth insight into the experiences of life beyond Rb. Findings highlight the need for specific psychosocial interventions informed by codesign.

STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ This is one of the first qualitative studies to seek an in-depth understanding of the range of psychosocial experiences of retinoblastoma (Rb) survivors, who are now teenagers and young adults.
- ⇒ It includes a relatively large sample (given the small population impacted by Rb), with heritable, non-heritable, unilateral or bilateral diagnoses.
- ⇒ It uses reflexive thematic analysis, which is flexible enough to accommodate different data collection methods (interviews and focus groups), to suit varied ages.
- ⇒ The majority of participants were recruited through a single charitable organisation, which may have a minor impact on the transferability of findings.

INTRODUCTION

Retinoblastoma (Rb) is a rare childhood cancer affecting the eye(s), which is diagnosed in 40–50 children in the UK a year. Around 45% are heritable,[1 2] the remainder occurring spontaneously. Although highly curable (~80% survival 10 years postdiagnosis),[3] Rb can greatly impact psychological well-being long after treatment ends.[4] For many, vision is impaired, and they may experience eye loss or facial changes.[5 6] Others may show no outward indication of their experience, but experience internal lifelong anxiety about the development of second cancers and the possibility of their own children developing Rb.[7]

Most instances of Rb are diagnosed before age 5, during a time of rapid development, when attachments with caregivers, cognitive and linguistic abilities, and emotional regulation are evolving in line with the environment.[8] As Rb impacts very young children, treatment is mostly complete before the individual is able to process the experience with meaningful language; their developmental stage hindering the ability to verbalise their experiences. This can distort memories and complicate psychosocial outcomes as those affected become teenagers and young adults.[9]

Unprocessed trauma is generally considered an integrated mind-body response, and the adaptive information processing (AIP) model describes the dysfunctional way the human brain stores such experiences,[10] potentially resulting in psychological distress, maladaptive thoughts or behaviours, and even physical pain. This has been verified in children experiencing medical trauma, with procedures found to be linked to a high degree of threat and frightening associations with pain, along with an immature understanding of events, including confusing feelings of betrayal towards their parents, whom they may associate with their treatment.[11] Consequently, parents often experience strong psychological reactions to their child's diagnosis,[12] including feelings of powerlessness, guilt and trauma.[13]

Despite the challenging circumstances associated with Rb, there is currently no known specific, evidence-based psychological support offered to young people in the UK, as they transition to adulthood and begin to navigate life beyond Rb. Existing research has highlighted the need for psychosocial support, yet what is required is under-researched and largely unavailable.[4,12,14,15] To address this, the current study aimed to explore, understand and describe the psychosocial needs of young people living beyond Rb, to inform future psychoeducation interventions.

METHODS

This study is part of a broader portfolio of work to understand the psychosocial needs of Rb survivors and develop a novel psychoeducation resource. Methods and results used the Consolidated criteria for Reporting Qualitative research to ensure appropriate empirical reporting.[16]

Study design

A qualitative study was conducted, with in-depth, semi-structured focus groups for teenage participants (aged 13–19 years) and individual interviews for young adults (aged 20–29). The rationale for the different approaches was based on evidence highlighting the distinct psychosocial needs of the two groups, in terms of identity, autonomy, emotional and sexual intimacy, cognitive ability, family planning, financial responsibility, education, and employment.[17]

Setting

All participants were treated in the UK at one of the two national Rb treatment centres: The Royal London Hospital shared with Great Ormond Street Hospital and Birmingham Children's Hospital. The third organisation involved was the Childhood Eye Cancer Trust (CHECT), a UK charity dedicated to supporting people impacted by Rb.

Sample

Sampling was purposive to include participants diagnosed with any form of Rb at any stage of childhood, receiving any type of treatment. Information power was used to guide the sample size.[18] Data quality and participant diversity were monitored during data collection to determine the final sample size.

Patient and public involvement

Patient and public involvement (PPI) involvement was reported according to GRIPP2 guidance on effectively involving experts by experience in health research[19] and conducted according to National Institute for Health Research guidelines.[20] Continuous involvement of CHECT members maintained the personal experience perspective throughout, including identifying the need for the study, piloting measures and commenting on initial findings.

Recruitment

Recruitment commenced on 9 June 2022 and ceased on 15 January 2023 and used online posters circulated via social media, as well as physical posters in hospital clinic waiting rooms.

Interested individuals contacted researchers via email and if eligible (see figure 1) were invited to take part. All participants completed brief questionnaires about their demographic and clinical characteristics (see online supplemental materials). Data collection techniques are discussed separately, for each method.

Data collection

Written consent was obtained from all participants, including for the use of direct quotations. Topic guides were prepared and used to structure interviews and focus groups (box 1) and were informed by the PPI group associated with the study (described in box 1). Using a semistructured approach, questions were open-ended to allow participants to discuss topics that felt meaningful to them. Interviews and focus groups were audio recorded, transcribed verbatim, anonymised and rechecked.

Teenage focus groups

Focus groups were conducted online via video call, between August 2022 and January 2023. Each was facilitated by the lead researcher (NO'D) and a second researcher (BP) to ensure consistency and reduce bias. Alongside conversational content, group dynamics and reflexive notes were kept in a field diary (NO'D).

Young adult interviews

One-to-one, in-depth, semi-structured interviews were conducted by video call, between June 2022 and December 2022 (NO'D). To enable choice and maximise accessibility, participants were offered the opportunity to undertake these interviews over the phone. In this event, all participants opted for video call interviews.

All participants were debriefed at the end and contacted 2 days later to check-in on well-being and provide signposting to support services if required. Participants received a £20 retail voucher in recognition of their participation.

Analysis

Data were analysed using reflexive thematic analysis (RTA)[21] and coded in NVivo.[22] To become familiar with the data, NO'D read all transcripts and DH read 10%.

Table 1 Demographic characteristics of participants

Participants N (%)		Adolescents (13–19)	Young adult (20–29)
Total	32 (100)	15 (47)	17 (53)
Type of retinoblastoma			
Heritable	10 (31)	5 (33)	5 (29)
Non-heritable	21 (66)	10 (66)	11 (65)
Unknown	1 (3)	0 (0)	1 (6)
Eye(s) impacted			
Unilateral	23 (72)	10 (66)	13 (76)
Bilateral	9 (28)	5 (33)	4 (24)
Median age in years (range)	20 (13–29)	15 (13–19)	25 (20–29)
Age at diagnosis			
<6 months	8 (25)	3 (20)	5 (29.4)
7–11 months	5 (15.6)	2 (13.3)	3 (17.6)
1 year to 1 year 11 months	5 (15.6)	2 (13.3)	3 (17.6)
2 years to 2 years 11 months	8 (25)	5 (33.3)	3 (17.6)
3 years to 3 years 11 months	4 (12.5)	1 (6.6)	3 (17.6)
4 years to 4 years 11 months	1 (3.1)	1 (6.6)	0 (0)
>5 years	1 (3.1)	1 (6.6)	0 (0)
Treating hospital			
Birmingham Children's	13 (41)	8 (53.3)	5 (29.4)
Royal London/Great Ormond Street	19 (59)	7 (46.6)	12 (70.6)
Treatment (in isolation or combination)			
Enucleation	24 (75)	10 (66.6)	14 (82.3)
Cryotherapy	7 (22)	4 (26.6)	3 (17.6)
Systemic chemotherapy	17 (53)	6 (40)	11 (64.7)
Intra-arterial chemotherapy	6 (19)	1 (6.6)	5 (29.4)
Laser therapy	5 (16)	4 (26.6)	1 (5.8)
Radioactive plaque	7 (22)	0 (0)	7 (41.2)
Gender			
Male	12 (38)	5 (33)	7 (41)
Female	20 (63)	10 (67)	10 (59)
Religion			
None	19 (59.4)	11 (73.3)	8 (47)
Christian	10 (31.2)	2 (13.3)	8 (47)
Other (unspecified)	3 (9.4)	2 (13.3)	1 (6)
Ethnicity			
White British	24 (75)	11 (73.3)	13 (76.5)
White Irish	1 (3)	0 (0)	1 (6)
Other white	2 (6)	1 (6.6)	1 (6)
Indian	1 (3)	1 (6.6)	0 (0)
Mixed/multiple	3 (9)	1 (6.6)	2 (11.8)
Hispanic	1 (3)	0 (0)	1 (6)
Relationship status*			
Single	22 (69)	15 (100)	7 (41.2)
In a relationship	4 (13)	0 (0)	4 (23.5)

Continued

Table 1 Continued

Participants N (%)		Adolescents (13–19)	Young adult (20–29)
Cohabiting	6 (19)	0 (0)	6 (35.3)
Highest level of educational			
Pre-GCSE	9 (28.1)	9 (60)	0 (0)
GCSE	4 (12.5)	4 (26.6)	0 (0)
BTEC	2 (6)	0 (0)	2 (11.8)
A-level or equivalent	3 (16)	2 (13.3)	1 (6.6)
Degree or equivalent	14 (43.75)	0 (0)	14 (82.3)
Employment status*			
Student	18 (56)	14 (93.3)	4 (23.5)
Employed full time	13 (41)	1 (6.6)	12 (70.6)
Unemployed	1 (3)	0 (0)	1 (6)

*At time of study.
A-level, advance level; BTEC, Business and Technology Education Council; GCSE, General Certificate of Secondary Education.

Family experiences and survivor guilt

The concept that Rb *'started with you' (P17:U-H)* was often brought up, in the context of survivor guilt: *"I started this gene. So, my mother and father didn't have it. My brother doesn't have it…I'm the 'lucky' one who started with this gene." (P17:U-H)*

Many acknowledged empathy for their parents and the difficult experiences they endured at this time, and the decisions they had had to make:

> my parents told me at a certain age and described the pressure there had been … the stress that had been there. My mum had told me that she'd kind of gone through this really, really kind of distressing time (P23:U-H)

The wider impact was feeling unable to talk to parents about their diagnosis and feelings as teenagers and young adults. *"My Mum doesn't really like reliving it.… She said that the few weeks between me getting diagnosed and my eye being removed, it's just a blur for her.… she doesn't really remember any of it, 'cos she was just going through the motions and stuff, so I don't really ask her about it." (P19:U-NH)*

Memories from treatment

Due to the age of Rb diagnosis, many participants spoke about others' expectations, and sometimes the reality of not having fully formed memories of diagnosis and treatment themselves. This fed into the narrative of guilt and difficulties discussing what had happened with parents, because of perception that their parents *'had it worse' (P24:U-NH)* as they had adult recollection of events.

> [parents'] memory is vivid and it is that specific that they can remember every blood transfusion, every

Figure 2 Diagrammatic overview of themes and subthemes.

> chemo…everything that went wrong, what day it was, what month it was, where they were at the time. So, I always just felt very guilty speaking about how I felt with them, because in my opinion, they'd had it worse than I had. (P24:U-NH)

Various memories about treatment were discussed, countering the myth that these are forgotten when this occurs at a young age. These ranged from distressing and traumatic to neutral or even fond recollections, particularly of the '*fun' (P25:U-NH)* and '*kindness' (P13:U-H)* clinical staff instilled into inherently traumatic procedures, such as enucleation. One individual remembered particularly distinctively behaviour post-treatment, when people got close to her face.

> I kind of remember kicking and screaming at the doctors… I just remember absolutely throwing a fit because of it. (P5:U-NH)

Although many participants received treatment while very young; key memories could be vividly recalled during adolescence and young adulthood. For some, these remained dormant until triggered, often by external sensory experiences, primarily focused on distinctive smells, which could elicit strong emotional reactions. This was often combined with attending routine health appointments, as may be expected.

> certain smells set me off as well, like when you go to the dentist and you've got that anaesthetist kind of smell (P24:U-NH)

For others, this was more shocking, with memories seeming to come from nowhere, with some describing feeling *'overwhelming sadness' (P7:U-H)* reminiscent of clinical trauma responses.

Long-lasting impact on personality

Many felt that their experiences of Rb had shaped them as a person. *"If I didn't have Rb when I was young, I would have been a totally different person." (P17:U-H)*

For some, this focused on feeling '*special' (P23:U-H)*, prominent during younger childhood, before this tapered off as they got older. For a few, this positive sense of self lasted throughout their lifetime, often due to consistent reassurance from their parents about being themselves:

> My mum just drilled it into me that, Do you know what, you're unique, just go along with it (P5:U-NH)

The idea that it is easier to '*be me' (P18:U-U*)* when young, while it is '*worse later on' (P25:U-NH)* reflects typical childhood experiences and the turbulence of adolescence, including transition to secondary school, hormonal changes, complex social dynamics, seeking independence and a focus on appearance.

> up until the age of year six, I was fine, completely fine. In fact, I used to talk about it [Rb] all the time and bring it [prosthetic] into school—you know, bring my spare eyes into school and sort of show off a bit about it…it gets worse later on (P25:U-NH)

Despite the best efforts of parents to instil a sense of pride and individuality, for some the '*realisation' (P18:U-U*)* that you are not '*like everyone else' (P18:U-U*)* still leads to a sense of insecurity, suggesting that the onus of support should not be on families, but on a wider network.

Some thought that trauma had '*made me stronger' (P23:U-H)*, suggesting that personality can be positively shaped from negative formative experiences. This was sometimes linked to ambition and a desire to achieve, with goal setting that may not otherwise have been aspired to.

> If I didn't have Rb when I was young… I would have different goals and aspirations in life. (P17:U-H)

Theme 2: adolescence: 'when you're a teenager, you feel like everything is the end of the world' (P28:U-NH)

Adolescence was widely considered the most difficult period and when support skills are required, with survivors of Rb managing late effects alongside the usual challenges faced by peers: *"I feel really lonely and I feel really weird" (P32:B-NH*)*

Psychological, social and behavioural impact

Adolescence was considered the most difficult life stage across participants, due to them often being in '*sustained periods of stress mode' (P23:U-H),* managing all the '*normal*' challenges of adolescence, alongside being a childhood cancer survivor. Many felt this was when they were most aware of how they looked, and experienced their peers as more critical, thus adding to the sense of feeling '*abnormal' (P16:U-NH)*.

For some, these feelings led to changed behaviour, with many overcompensating for their eye(s) by trying to improve other areas of their image.

> I think there was a time where I was more worried about my appearance and whatnot and I was trying to compensate by getting in the gym and making myself look better (P18:U-U*)

Ironically, adolescence was said to be the time when help was most needed, but also most difficult to acquire. This appeared particularly problematic for males, who seemed more isolated, perhaps reflecting wider cultural norms and expectations *"(I) just grit my teeth together and get through it" (P29:U-NH),* and stigma for expressing emotions, which may be perceived as weak:

> I feel like there's definitely a stigma attached to men nowadays, because there's like this whole thing of "Oh, you've got to be a man, you've got to be tough…a strong, tough, alpha male, I guess." (P29:U-NH).

For a few this led to maladaptive coping strategies, such as drug use and joining cult internet forums. This did not differ by heritable and non-heritable Rb status, with every male involved in this research describing it as the first time they had felt able to speak about the impact of Rb on how they think and feel.

> I do think I could have got help if I actually asked for it. I think my problem was that I probably just suffered a bit in silence. (P28:U-NH)

Developing a culture of open discussion around mental health may therefore be particularly crucial for pre-adolescent boys.

> I found a lot of… relatability, online. And there were all these other angry young men who I thought, "Oh, these guys are the same as me," which they weren't… people who've had bad experiences, when they feel inadequate, they'll eventually fall into this like circle or this group of less than pleasant people online… I was sort of like teetering on the edge of it. (P29:U-NH)

As these individuals reached older adolescence, they often behaved contrary to expected norms, with many taking more responsibility for living a healthy lifestyle compared with their peers. Such behaviour change and self-care can be positive but needs to be balanced to prevent the anxiety and constant body surveillance described by some.

> One of the hangover effects of Rb is that I felt this sense of responsibility to look after myself, so I always felt quite strongly against things like smoking or excessive alcohol consumption, starting about 17 [years old] I suppose (P23:U-NH)

High levels of anxiety were prominent across the sample, beginning in adolescence and often extending into adulthood. These were commonly linked to three areas: sex and relationships, having your own children and developing second cancers.

> It was really panic inducing… I just thought I look ugly… you know when you're growing up, you're reading like Roald Dahl and watching pirate films, and it's all kind of this really ugly thing [one eye] that is attributed to people that are weird or mean, and so I think I didn't yet have the kind of separation from that. (P25:U-NH)
>
> I am less inclined to have casual flings, to be quite honest, because there's always the potential for that being an unfair burden for anybody to carry, basically, unless you're in a committed relationship (P27:B-H)
>
> The side effects of like, catching cancer again is quite scary (P12:U-NH)

Identity

For many young adults looking back to adolescence, there was a recognition that they had numerous questions about themselves, and how Rb impacted their sense of self.

> it's just a time where you don't feel, you don't even know who you are, you're trying to figure out, you know, what kind of clothes am I supposed to wear, what's my style, to what do I want to do. Like, who am I? (P32:B-NH*)

Many kept coming back to the idea that life was unfair, in terms of their Rb diagnosis and experience of childhood cancer, but also the impact this had had on their perceptions of self, others and the world around them.

> I always did ask why did it happen—this is a very rare disease, like why did it have to happen to me. I kind of still ask that now, like why me (P29:U-NH)

Dealing with identity as a teenager was complicated by the nature of Rb, with many doubting the legitimacy of their diagnosis, particularly after hearing it described as a '*good cancer*' due to its high survival rates. This was particularly prominent among people not treated with enucleation, who had no visible '*signs*' of anything having been '*wrong*', leading to them making downward comparisons to others.

> it would almost be a bit of a, I guess a joke that I'd say, "Oh, you know, I struggled as well," sitting around with all these people that aren't as fortunate as I was. (P24:U-NH)

Such comparisons were also noted as barriers to accessing support, particularly from generic cancer support groups with children who had cancers that were perceived '*worse*' than Rb.

> It was kind of upsetting to me to be around other kids that also had cancer and had it worse than I did. And I think I was quite aware of that even when I was younger, that there were people that had it worse, and I felt kind of sad about that, so I didn't engage with that [support] as much. (P25:U-NH)

'Normal' for me

As Rb largely affects very young children, many individuals could not remember a life any different to what they knew now. This was the same across individuals with and without prosthetics, as well as those whose treatment had resulted in facial changes.

> It's not really a big part of my life, to be honest, 'cos I was so young when I lost it [eye], I don't think about it much on a daily basis. It's just something that I kind of exist with (P25:U-NH)

It was often other people and their comments that reminded people of their differences; behaviour that was experienced throughout childhood into young adulthood, from peers, friends and strangers.

> I came home one day and I was like, "Oh, what's a Cyclops?" And they [parents] were like, "Oh, it's a mythical creature with one eye." And I was like, "Oh, that's what this boy calls me at school." (P30:U-NH)

Some individuals choose to embrace and make a feature of their difference through wearing a coloured or uniquely designed prosthetic, which could fuel hateful comments and encourage self-blame.

> I still get horrible comments … people tend to go, "Oh well, you're asking for it by wearing something like a coloured prosthetic" (P19:U-NH)

Theme 3: adulthood: 'it's not meant to be for life, but it carries on' (P5:U-NH)

Adulthood was both a time for reflection and projection, dependent on the life stage of the participant at the time of interview.

Acceptance

Many young adults developed the ability to accept themselves and their identity, which involved acknowledging their experience of Rb, without making it the only thing about them.

> Don't compare yourself to anybody… You need to make your reality your possibility… figure out, write it down on a piece of paper what are you able to do… I started to accept, not appreciate, not be grateful, because I'm not grateful for what I've been through, I'm grateful for the strength that I developed to get out of it (P32:B-NH*)

Acknowledging that the impact of Rb does not go away seemed to come with time, with one individual clearly highlighting the potential lifelong impact of their childhood cancer.

> It's not meant to be life but it does carry on throughout your years (P5:U-NH)

The ability to validate experience and provide self-reassurance was common, and for some this involved efforts to '*make friends*' with their Rb and accept how it had shaped their identity and life, rather than fighting it and denying its' existence.

Doing 'the work'

'The work' (P32:B-NH)* is an overarching term used by a participant and developed within this study to refer to the different coping strategies used by participants. A key feature of this was the need to seek accurate information, which was prominent for developing personal understanding, and for preventing childhood beliefs and anxieties being carried into adulthood.

> I'd internalised that, that there's these tumours that could suddenly…go off like a volcano (P27:B-H)

Contrasting with the diagnostic and treatment periods when it was parents who were given information, participants focused on the need for '*information for me, not my parents' (P4:U-H*). Many however, said it could be hard to know what information was needed, and who to ask for for advice:

> it's small stuff like that that concerns me personally. Like basically, to sum it up in a word, how can I live my life without relying on other people to take my hand and—and gently guide my way through it, you know. (P1:U-NH)

It was apparent across the sample that a sense of being proactive and having someone independent to talk to was important for preventing later difficulties.

> I didn't really understand it growing up. I think you need to be told early, then it prepares for the future if that makes sense, rather than just seek out the information when like you get curious or you need it. (P8:U-U*)

Many young adult participants stated that '*the work*' included accessing and integrating therapy into their lives, which was said to bring substantial benefits, and may be useful in earlier life alongside peer support.

> It sort of feels like there could have been a few things nipped in the bud just by talking… psychological support in the sense of therapy, talking about my experience, talking about how I view the world; talking about how the impact of it on my family has impacted me. (P31:U-NH)

Others expressed a desire to have someone to talk to who was independent of doctors and parents but had not accessed this.

> Maybe having someone to talk to growing up—'cos, you know, you have down days and you have up days, and if you're having a down day, maybe just talking to someone who isn't your parents might be helpful, who really, really gets it. (P31:U-H)

This was summarised by one adult who stated you '*just need someone to steer you in the right direction' (P23:U-H*).

DISCUSSION

This study explored experiences of living beyond Rb, with a focus on childhood (family experiences and survivor guilt, memories from treatment, lifelong impact on personality), adolescence (psychological impact, identity, 'normal' for me') and adulthood (acceptance and doing 'the work').

For most, Rb was said to cause long-term distress, heightened feelings of anxiety and lowered self-esteem. Despite this, many found that this experience enabled them to develop resilience and enhanced coping strategies that may not otherwise have developed. We identified several themes and subthemes that were common across Rb types, providing a unique contribution to the growing evidence on psychosocial experiences among young survivors of Rb.

A key theme identified in this study was the lifelong trauma impacting the survivor and their family following Rb diagnosis. This is common among childhood cancer generally[23–26] and is well understood in more frequently diagnosed cancer types. The experiences of family members, particularly parents, is better understood than that of affected individuals.[27–29] This may be because most Rb diagnoses are made in early childhood, so it is parents or carers who take on information and make key decisions. Many participants also shared complex thoughts about 'putting' their parents through difficult experiences, reporting emotional difficulties, stress and anxiety as they adapted to the diagnosis and authorised medics to treat their child, which in some cases involved enucleation and use of a prosthetic eye.[27 29] Many participants acknowledged empathy for their parents and the treatment decisions they had to make, as well as the wider impact on their own behaviour, and their inability to talk about how they feel. This finding is reflected in wider childhood cancer research[30] and emphasises the need for young people to access information and support independently from parents and family members, and at an age and developmentally appropriate time.

'Memories from treatment' was a key finding and was linked to the individuals' emotional response and understanding of the world and others around them, as indicated by the verbatim quotations. Although reported in previous childhood cancer survivor literature,[31 32] this has not been noted in Rb before or cancers affecting very young children, who may be preverbal, yet still have sensory, salient recollections of this period. This finding can be described by the AIP model, which states that early life trauma can have lifelong impact.[33 34] For individuals who experience cancer as teenagers or young adults, it has been noted that cancer diagnosed earlier in life disrupts key developmental stages and shapes cognitive processes.[35] This fits with broader research on trauma experiences and the impact this can have on personality development throughout the lifespan, both in terms of post-traumatic growth and influencing positive coping mechanisms like resilience, and more negatively with risks for later mental health difficulties.[36–39]

Correlating with existing childhood cancer survivorship literature, some participants thought that their traumatic experiences had 'made them stronger'. Such positivity is common with respect to cancer diagnoses and negative formative experiences,[40–42] which has been termed 'post-traumatic growth'.[43 44] This can be particularly relevant to children diagnosed at a very young age, who often adapt well to their situation, as they do not recall ever living differently; a position shared by many in the current study.

Adolescence was widely considered the most difficult phase, with a particular need for support and coping mechanisms. We highlighted the psychological impact of Rb at this time, including feelings of unfairness and powerlessness in the face of restrictions (perceived and actual), and demonstrating how survivors from a rare cancer may struggle to identify others with whom they can relate, express themselves and share anxieties and difficulties. Previous literature has referred to this as 'negotiating the psychosocial challenges from two different worlds—'the healthy' and 'the ill'.[45]

The ability to share feelings and be understood is vital for self-esteem, which if absent may result in loneliness. This contradicts work suggesting survivors of Rb report lower levels of depressive, anxious and somatic symptoms than non-Rb childhood cancer survivors.[14] However, these findings may not reflect survivors' true feelings, but instead, reluctance to disclose complex emotional responses and distress through fear of judgement. Existing research[4] has found that adult Rb survivors have 20% more psychological difficulties, such as anxiety and depression, than a healthy comparison group. These findings are echoed by Morse *et al*,[46] who found that although survivors of Rb report typical emotional health and quality of life, they are more anxious than a comparison group of peers who have not experienced cancer.

Regarding the concept of Rb being a 'good cancer', this sentiment has been shared by other survivors who are told they should be 'grateful' as they have a 'good' cancer that they are unlikely to die from or may live 'with' for many years.[47–50] These studies highlight how some cancer experiences can be downplayed, potentially leading to downward comparisons that create a barrier to help-seeking and emotional expression, meaning individuals do not feel they are 'genuine' survivors or 'entitled' to support.

Uncertainty was expressed about many aspects of adult life, notably sex and relationships, having children and developing a second cancer. Such feelings are well documented in cancer survivors of all ages, often leading to over-appraisal of the likelihood of something 'bad' happening again.[51–55] This can be amplified when access to information and social support is limited, and ambiguous healthcare systems are encountered.[56] As may be expected of adolescence, identity was a key theme, with individuals having many questions about themselves, and how Rb had impacted their sense of self. Rb can differ to other childhood cancers as it can leave a physical reminder, commonly via enucleation and use of a prosthetic eye. Existing literature, however, mainly focuses on temporary appearance changes during treatment, for example, loss of hair from chemotherapy,[57 58] or weight gain from steroids[59–61]; or permanent changes that can be hidden, for example, implant surgery or prothesis use in breast cancer survivors.[62 63] Other work addresses permanent non-cancer-related illnesses or accidents, for example, burns,[64 65] cleft palate,[66 67] scarring[68–70]; or inherited conditions like neurofibromatosis,[71–73]which may have different psychosocial impacts to cancer. All these conditions can have a deeply distressing psychosocial impact,[58 74 75] although Rb, particularly when heritable[15] or treated with enucleation[76] is distinct, and must be supported as such.

Adulthood was a time for reflection and projection, dependent on life stage at the time of interview, with Rb acceptance universally considered unachievable while still young. 'Doing the work' encapsulates the need to answer questions, and the role of strategies such as psychotherapy and peer support to improve well-being. In comparison to adolescence, many young adults had developed the ability to accept themselves and their identity, acknowledging the experience of Rb without making it the only thing about them. This corroborates wider cancer survivorship literature, which emphasises the psychosocial challenges unique to adolescent cancer survivors who must manage 'normal' life while working through questions about identity,[77] interpersonal relationships,[78] education,[79] and cancer-specific anxieties.[80 81] These appear to ease as individuals transition to young adulthood, particularly if receiving psychological support with a specific acceptance component.[82–89] Concurring with the current study, these findings highlight how structured psychological therapy may be useful earlier in life alongside peer support, another factor considered useful to survivors of Rb.

Strengths and limitations

To our knowledge, this is one of the first qualitative studies seeking to understand the psychosocial experiences of survivors of Rb, regardless of their genetic status or the unilateral or bilateral impact. It presents a novel exploration of the interplay between Rb, self-perception, interpersonal dynamics, and wider societal and healthcare contexts. Furthermore, it provides insight into the psychological complexity of rare childhood cancer survivorship during the transition from adolescence to young adulthood and beyond. Despite the rarity of Rb, a relatively large sample was recruited, mostly via CHECT's social media platforms, reflecting a desire to 'give back' to the charity and community; this may, however, have risked self-selection bias. Interviews were said to offer a cathartic experience from which participants could view and process their journey. As is increasingly common in qualitative health research,[90 91] we experienced a problem with 'bot' or fraud participants, disrupting our recruitment timescale.[92] All recruitment material was designed for people from 13 years old to promote inclusivity, with video information for visually impaired candidates, or those who preferred to hear/see the research team discuss the study aims and processes. The methodology used, RTA, is a flexible approach which allowed the opportunity to combine different methods of data collection through conducting both interviews and focus groups. At the outset of this research we did not intend to analyse the data as one, but it made sense to do so due to the heterogeneity of themes across adolescent and young adult participants. As an iterative process, this approach allowed us to continually revisit and refine the themes over a long period, holding discussions with the wider research team to delve into the nuances of participant meaning.

Implications for practice and policy

We have clearly demonstrated the profound psychological and emotional impact of Rb on survivors and highlighted the need for integrated, specific psychosocial support, delivered within long-term follow-up care. Regarding policy, our findings align with the National Health Service Long-Term Plan, which aims to provide patients with more support options, particularly for psychological needs and preventative care.[93] It also fits with healthcare changes post-COVID-19, whereby non-hospital-based follow-up in community settings, with digital and remote support options is emphasised.

X Nicola O'Donnell @nicolaod_, Bob Phillips @drbobphillips and Jessica Elizabeth Morgan @drjessmorgan

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Contributors NO'D (female): conceptualisation, data curation, facilitation of focus groups and interviews, formal analysis, project administration, writing–original draft, writing–review and editing, responsible for data ownership and guarantor. JEM (female): Supervision, writing–review and editing. DH (female): supervision, formal analysis, writing–review and editing. BP (male): conceptualisation, co-facilitation of focus groups, supervision, writing–review and editing.

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ORCID iDs

Nicola O'Donnell http://orcid.org/0000-0003-0661-4129
Bob Phillips http://orcid.org/0000-0002-4938-9673
Jessica Elizabeth Morgan http://orcid.org/0000-0001-8087-8638
Debra Howell http://orcid.org/0000-0002-7521-7402

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Appendix A26: Certificate of attendance at ISOO, August 2023

ISOO 2023 AFRICA
AUGUST 21-23, 2023 MOMBASA, KENYA

International Society of Ocular Oncology

CERTIFICATE OF ATTENDANCE VIRTUAL MEETING

This Certificate is Presented to

NICOLA ODONNELL

21-23 August 2023

Dan S Gombos, M.D.
President, ISOO

Jesse L. Berry, M.D.
Vice President, ISOO

Ido Didi Fabian

Ido Didi Fabian, M.D.
Conference President

Appendix A27: Certificate of Good Clinical Practice Refresher, August 2023

NIHR | National Institute for Health and Care Research

Issued by:
Clinical Research Network
Coordinating Centre

CERTIFICATE OF ACHIEVEMENT

Nicola O'Donnell

has completed the course:

Good Clinical Practice (GCP) Refresher ELearning

August 2, 2023

Modules Completed

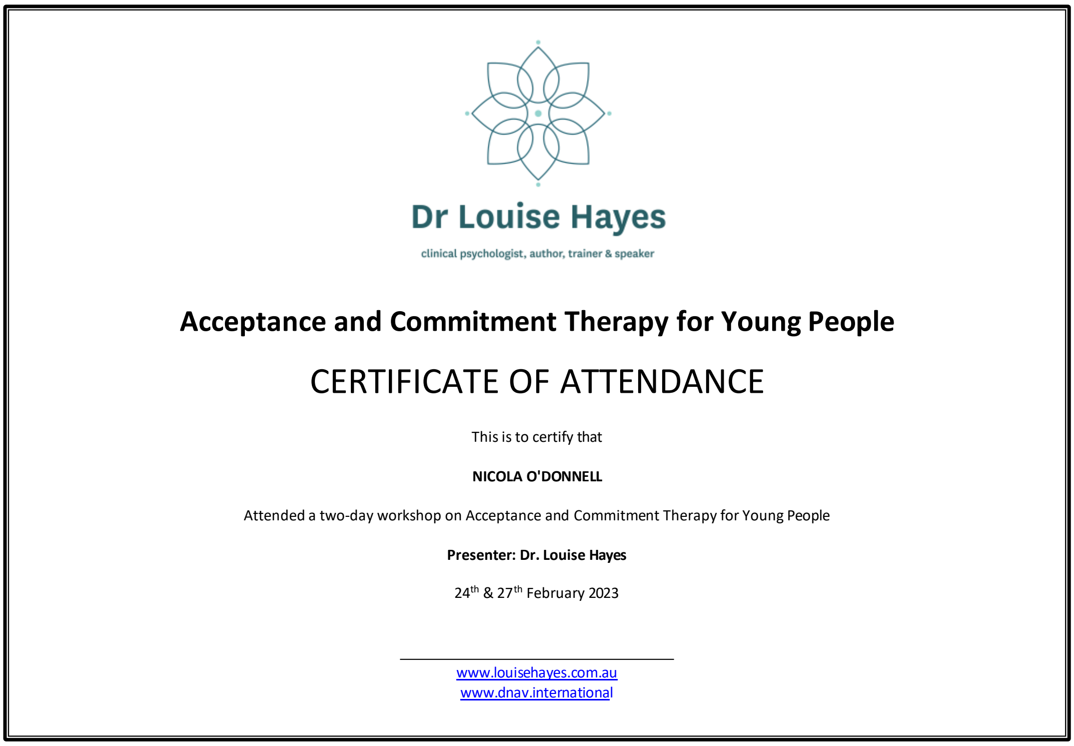
- Good Clinical Practice (GCP) Refresher: Revisiting Key Concepts
- GCP Refresher Hot Topics
- Good Clinical Practice (GCP) Refresher: Reflecting on your own practice and experience

This course is worth 3 CPD Credits

CPD®
The CPD Certification Service

Version: May 2022

Appendix A28: Certificate of attendance at ACT for Young People, February 2023



Dr Louise Hayes
clinical psychologist, author, trainer & speaker

Acceptance and Commitment Therapy for Young People

CERTIFICATE OF ATTENDANCE

This is to certify that

NICOLA O'DONNELL

Attended a two-day workshop on Acceptance and Commitment Therapy for Young People

Presenter: Dr. Louise Hayes

24th & 27th February 2023

www.louisehayes.com.au
www.dnav.international

APPENDIX A29: CERTIFICATE OF ATTENDANCE AT BPOS CONFERENCE, MARCH 2023

BPOS

Certificate of Attendance

This is to certify that

Mrs Nicola O'Donnell

has attended the

British Psychosocial Oncology Society Annual Conference 2023

Cambridge, UK

on

Thursday 23rd March 2023

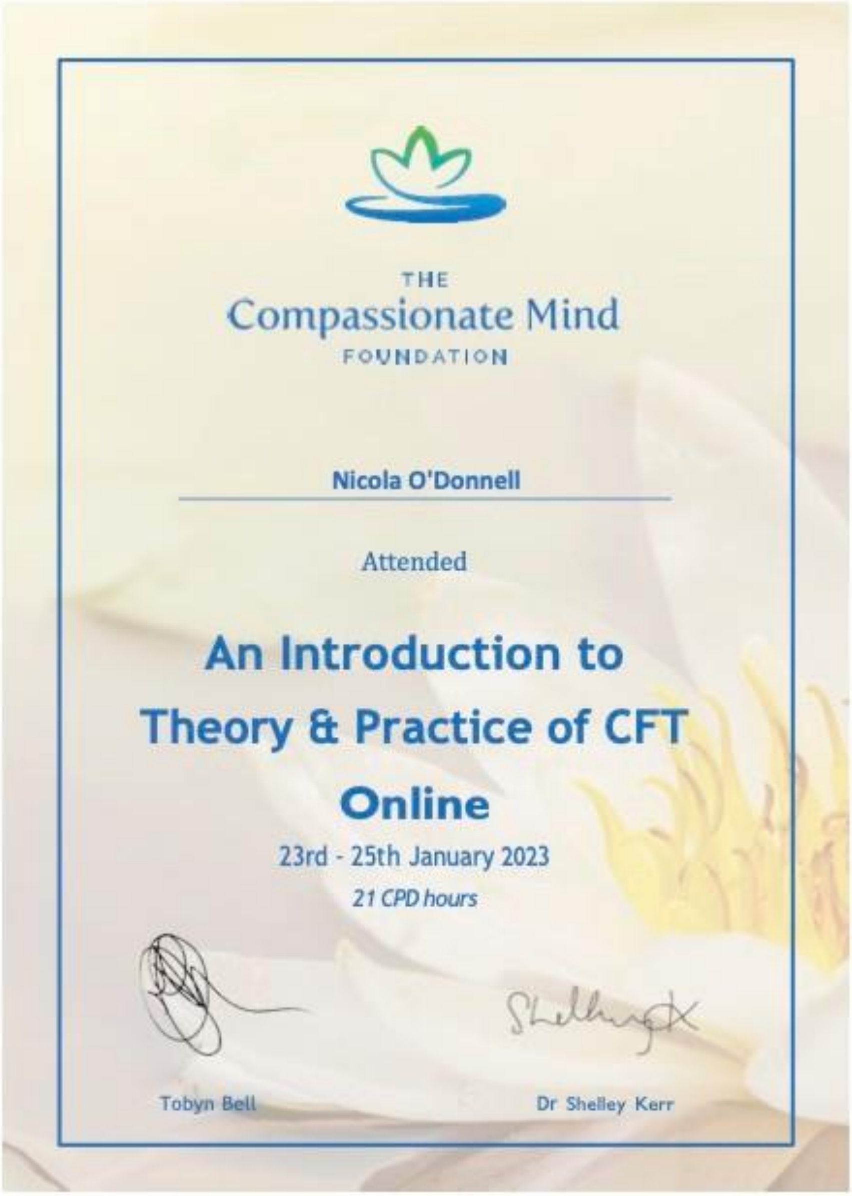
S. Archer.

Dr Stephanie Archer
Chair, BPOS

British Psychosocial Oncology Society
[Registered Charity 294303]

Website: www.bpos.org

Appendix A30: Certificate of attendance at CFT training, January 2023



THE
Compassionate Mind
FOUNDATION

Nicola O'Donnell

Attended

An Introduction to
Theory & Practice of CFT
Online

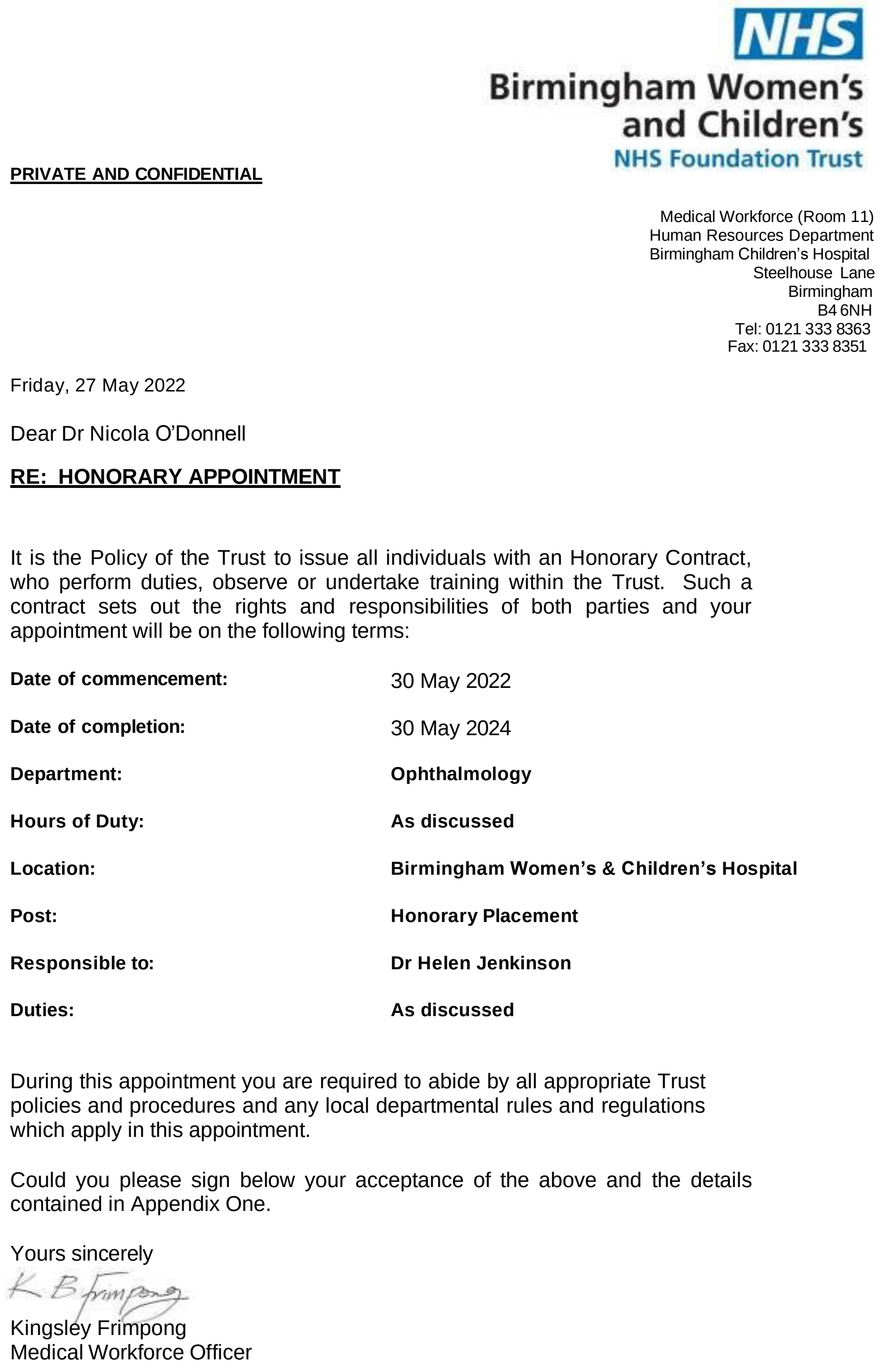
23rd - 25th January 2023
21 CPD hours

Shelley Kerr

Tobyn Bell

Dr Shelley Kerr

APPENDIX A31: HONORARY CONTRACT AT BIRMINGHAM WOMEN'S AND CHILDREN'S, MAY 2022



NHS
Birmingham Women's
and Children's
NHS Foundation Trust

PRIVATE AND CONFIDENTIAL

Medical Workforce (Room 11)
Human Resources Department
Birmingham Children's Hospital
Steelhouse Lane
Birmingham
B4 6NH
Tel: 0121 333 8363
Fax: 0121 333 8351

Friday, 27 May 2022

Dear Dr Nicola O'Donnell

RE: HONORARY APPOINTMENT

It is the Policy of the Trust to issue all individuals with an Honorary Contract, who perform duties, observe or undertake training within the Trust. Such a contract sets out the rights and responsibilities of both parties and your appointment will be on the following terms:

Date of commencement:	30 May 2022
Date of completion:	30 May 2024
Department:	**Ophthalmology**
Hours of Duty:	**As discussed**
Location:	**Birmingham Women's & Children's Hospital**
Post:	**Honorary Placement**
Responsible to:	**Dr Helen Jenkinson**
Duties:	**As discussed**

During this appointment you are required to abide by all appropriate Trust policies and procedures and any local departmental rules and regulations which apply in this appointment.

Could you please sign below your acceptance of the above and the details contained in Appendix One.

Yours sincerely

K B Frimpong

Kingsley Frimpong
Medical Workforce Officer

Appendix A32 and A33: Certificates of attendance at Social Research Association Courses

SRA SOCIAL RESEARCH ASSOCIATION

CERTIFICATE OF ATTENDANCE

AWARDED TO

Nicola O'Donnell

FOR ATTENDANCE AT

INTRODUCTION TO PARTICIPATORY ACTION RESEARCH - ONLINE
30 MARCH 2022

GRAHAM FARRANT
CHIEF EXECUTIVE

SOCIAL
RESEARCH
ASSOCIATION

CERTIFICATE OF ATTENDANCE

AWARDED TO

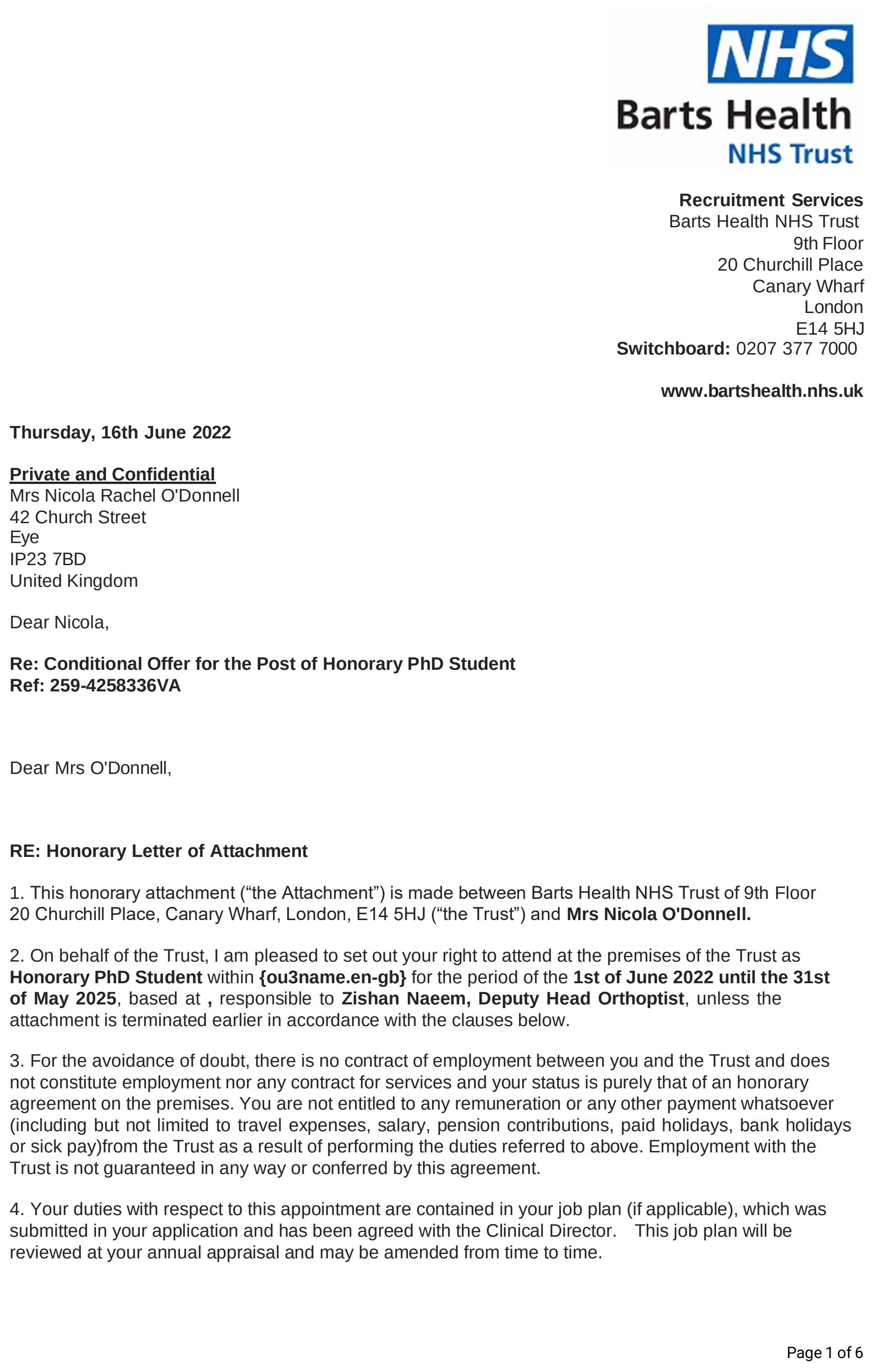
Nicola O'Donnell

FOR ATTENDANCE AT

POSITIONALITY AND REFLEXIVITY IN QUALITATIVE RESEARCH - ONLINE
24 & 25 FEBRUARY 2022 (2 CONSECUTIVE MORNINGS)

GRAHAM FARRANT
CHIEF EXECUTIVE

Appendix A34: honorary contract at Barts Health, June 2022



Recruitment Services
Barts Health NHS Trust
9th Floor
20 Churchill Place
Canary Wharf
London
E14 5HJ
Switchboard: 0207 377 7000

www.bartshealth.nhs.uk

Thursday, 16th June 2022

Private and Confidential
Mrs Nicola Rachel O'Donnell
42 Church Street
Eye
IP23 7BD
United Kingdom

Dear Nicola,

Re: Conditional Offer for the Post of Honorary PhD Student
Ref: 259-4258336VA

Dear Mrs O'Donnell,

RE: Honorary Letter of Attachment

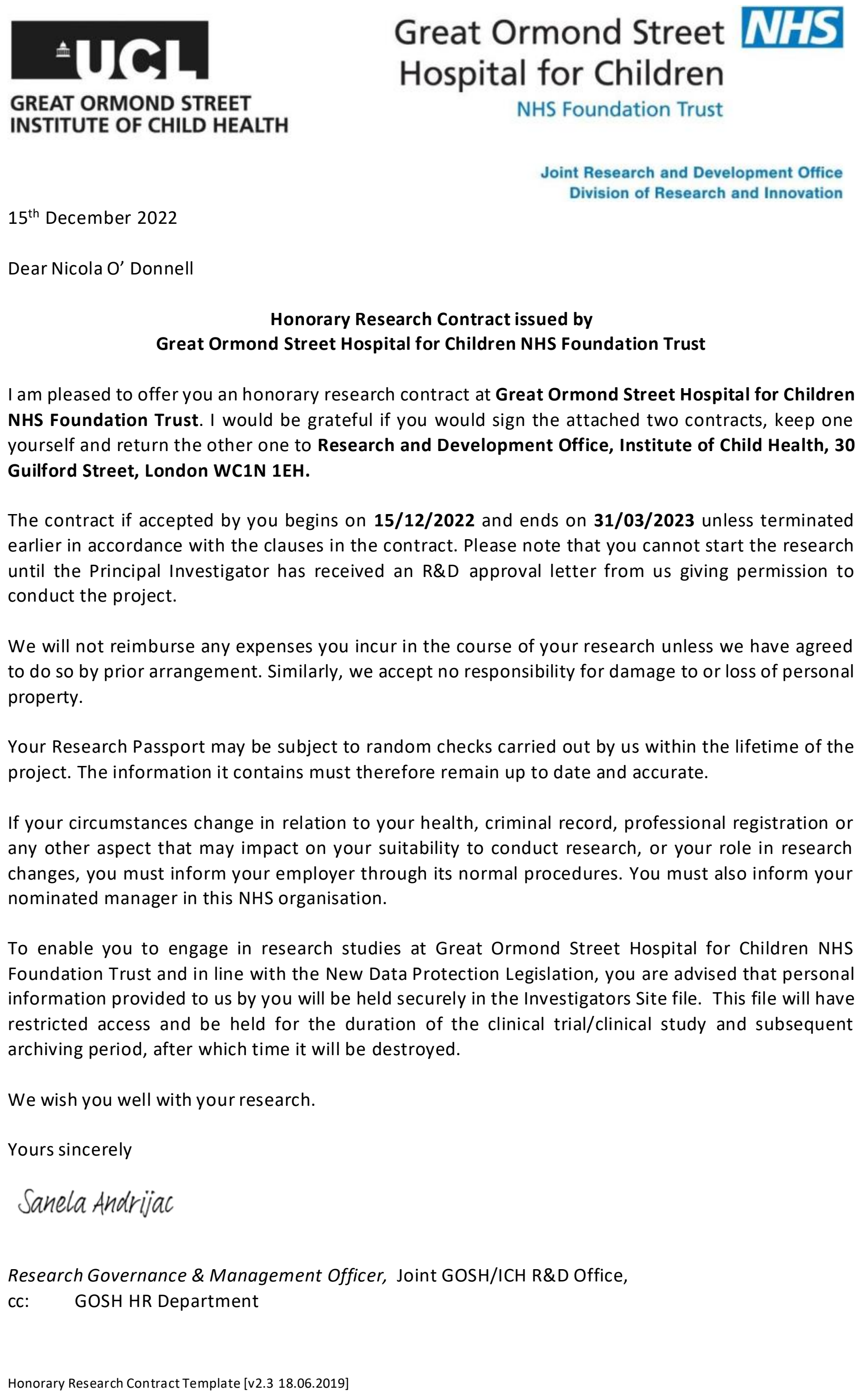
1. This honorary attachment (“the Attachment”) is made between Barts Health NHS Trust of 9th Floor 20 Churchill Place, Canary Wharf, London, E14 5HJ (“the Trust”) and **Mrs Nicola O'Donnell.**

2. On behalf of the Trust, I am pleased to set out your right to attend at the premises of the Trust as **Honorary PhD Student** within **{ou3name.en-gb}** for the period of the **1st of June 2022 until the 31st of May 2025**, based at **,** responsible to **Zishan Naeem, Deputy Head Orthoptist**, unless the attachment is terminated earlier in accordance with the clauses below.

3. For the avoidance of doubt, there is no contract of employment between you and the Trust and does not constitute employment nor any contract for services and your status is purely that of an honorary agreement on the premises. You are not entitled to any remuneration or any other payment whatsoever (including but not limited to travel expenses, salary, pension contributions, paid holidays, bank holidays or sick pay)from the Trust as a result of performing the duties referred to above. Employment with the Trust is not guaranteed in any way or conferred by this agreement.

4. Your duties with respect to this appointment are contained in your job plan (if applicable), which was submitted in your application and has been agreed with the Clinical Director. This job plan will be reviewed at your annual appraisal and may be amended from time to time.

APPENDIX A35: HONORARY CONTRACT AT GOSH, DECEMBER 2022



UCL
GREAT ORMOND STREET
INSTITUTE OF CHILD HEALTH

Great Ormond Street Hospital for Children NHS
NHS Foundation Trust

Joint Research and Development Office
Division of Research and Innovation

15th December 2022

Dear Nicola O' Donnell

Honorary Research Contract issued by
Great Ormond Street Hospital for Children NHS Foundation Trust

I am pleased to offer you an honorary research contract at **Great Ormond Street Hospital for Children NHS Foundation Trust**. I would be grateful if you would sign the attached two contracts, keep one yourself and return the other one to **Research and Development Office, Institute of Child Health, 30 Guilford Street, London WC1N 1EH.**

The contract if accepted by you begins on **15/12/2022** and ends on **31/03/2023** unless terminated earlier in accordance with the clauses in the contract. Please note that you cannot start the research until the Principal Investigator has received an R&D approval letter from us giving permission to conduct the project.

We will not reimburse any expenses you incur in the course of your research unless we have agreed to do so by prior arrangement. Similarly, we accept no responsibility for damage to or loss of personal property.

Your Research Passport may be subject to random checks carried out by us within the lifetime of the project. The information it contains must therefore remain up to date and accurate.

If your circumstances change in relation to your health, criminal record, professional registration or any other aspect that may impact on your suitability to conduct research, or your role in research changes, you must inform your employer through its normal procedures. You must also inform your nominated manager in this NHS organisation.

To enable you to engage in research studies at Great Ormond Street Hospital for Children NHS Foundation Trust and in line with the New Data Protection Legislation, you are advised that personal information provided to us by you will be held securely in the Investigators Site file. This file will have restricted access and be held for the duration of the clinical trial/clinical study and subsequent archiving period, after which time it will be destroyed.

We wish you well with your research.

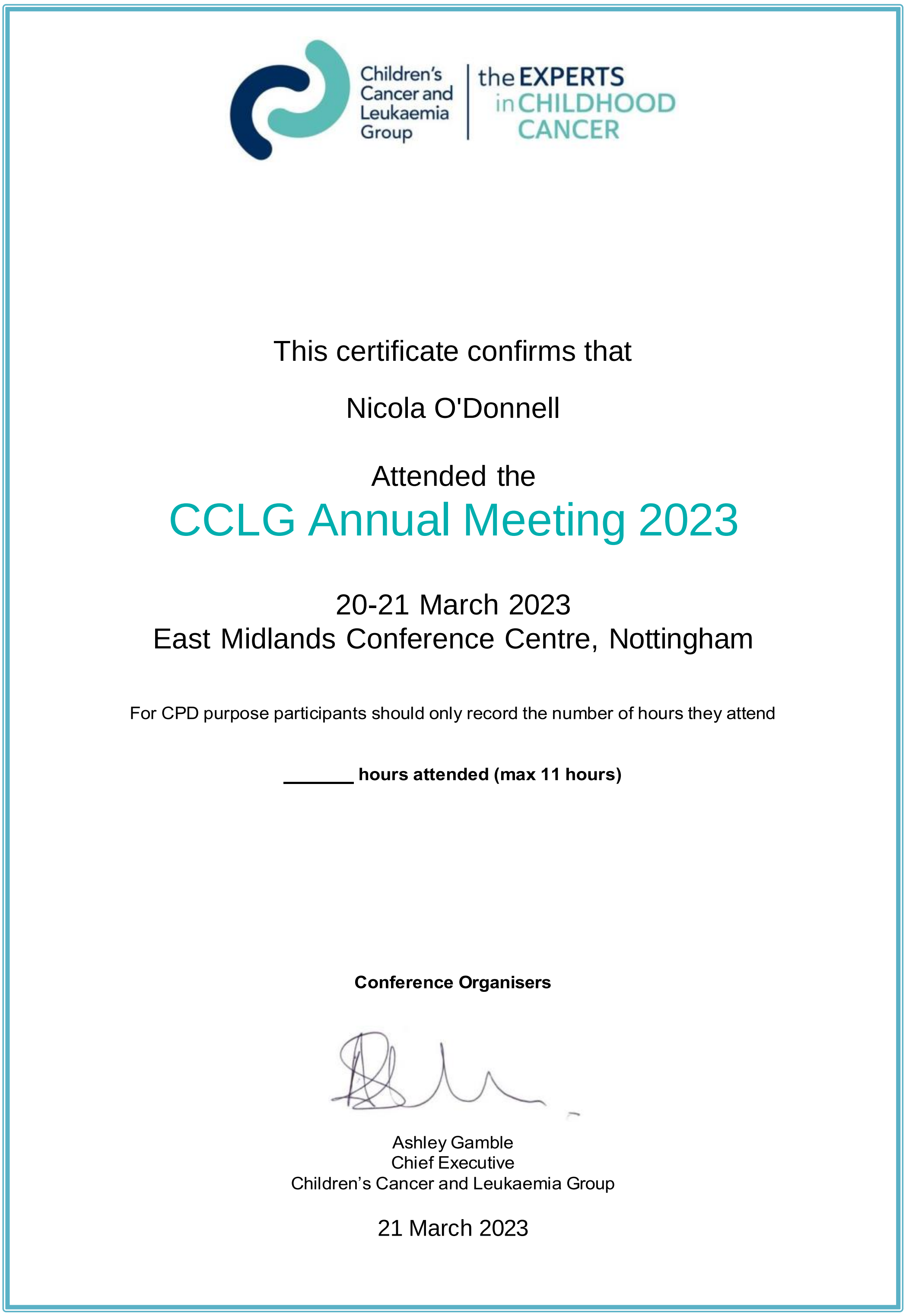
Yours sincerely

Sanela Andrijac

Research Governance & Management Officer, Joint GOSH/ICH R&D Office,
cc: GOSH HR Department

Honorary Research Contract Template [v2.3 18.06.2019]

Appendix A36: Certificate of attendance at CCLG, March 2023



Children's Cancer and Leukaemia Group

the EXPERTS in CHILDHOOD CANCER

This certificate confirms that

Nicola O'Donnell

Attended the

CCLG Annual Meeting 2023

20-21 March 2023
East Midlands Conference Centre, Nottingham

For CPD purpose participants should only record the number of hours they attend

_______ **hours attended (max 11 hours)**

Conference Organisers

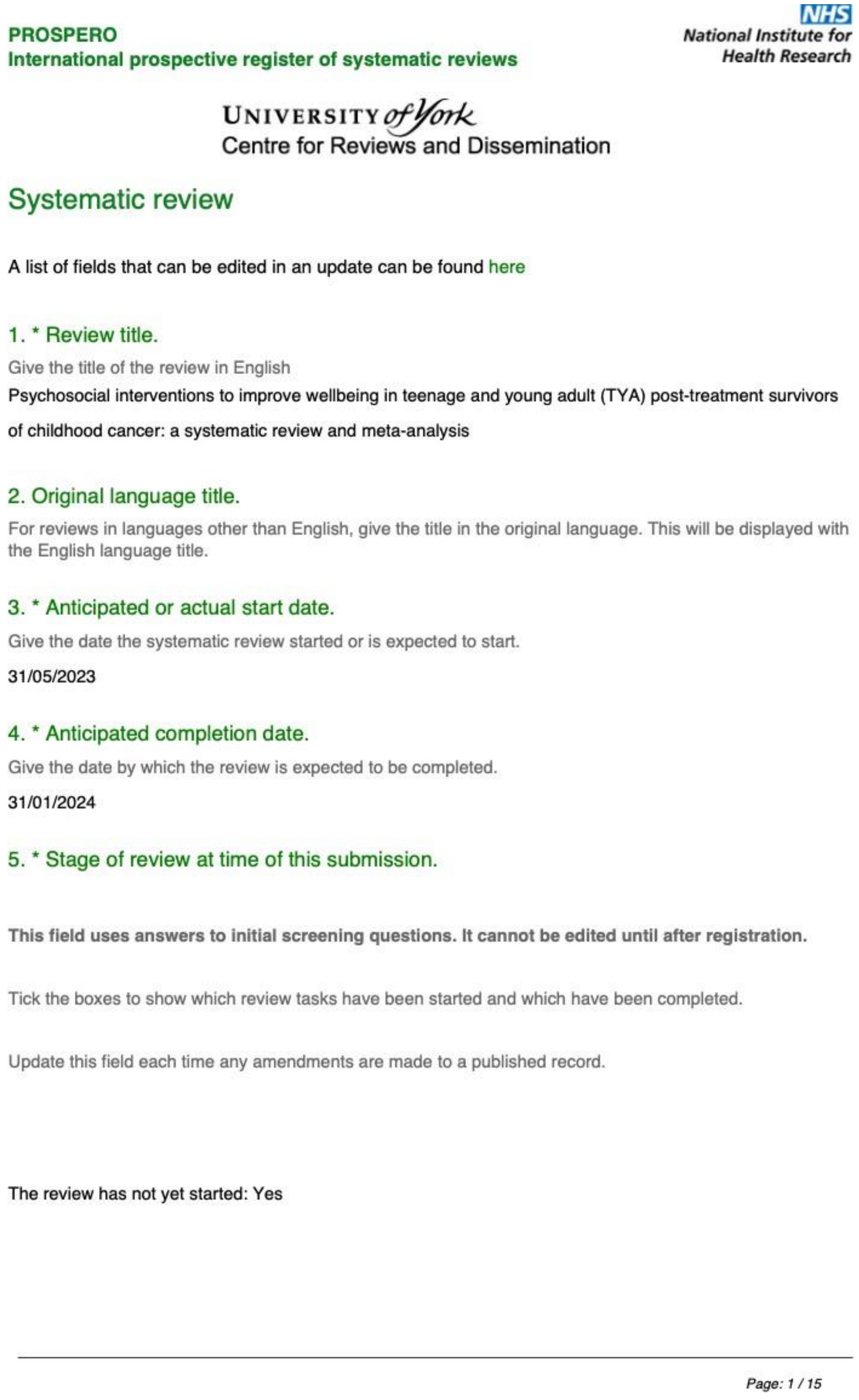
Ashley Gamble
Chief Executive
Children's Cancer and Leukaemia Group

21 March 2023

APPENDIX B

This appendix includes all appendices related to chapter five of this thesis.

APPENDIX B1: PROSPERO REGISTRATION



PROSPERO
International prospective register of systematic reviews

NHS
National Institute for Health Research

UNIVERSITY of York
Centre for Reviews and Dissemination

Systematic review

A list of fields that can be edited in an update can be found here

1. * Review title.

Give the title of the review in English

Psychosocial interventions to improve wellbeing in teenage and young adult (TYA) post-treatment survivors of childhood cancer: a systematic review and meta-analysis

2. Original language title.

For reviews in languages other than English, give the title in the original language. This will be displayed with the English language title.

3. * Anticipated or actual start date.

Give the date the systematic review started or is expected to start.

31/05/2023

4. * Anticipated completion date.

Give the date by which the review is expected to be completed.

31/01/2024

5. * Stage of review at time of this submission.

This field uses answers to initial screening questions. It cannot be edited until after registration.

Tick the boxes to show which review tasks have been started and which have been completed.

Update this field each time any amendments are made to a published record.

The review has not yet started: Yes

Page: 1 / 15

Review stage	**Started**	**Completed**
Preliminary searches	No	No
Piloting of the study selection process	No	No
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

Provide any other relevant information about the stage of the review here.

6. * Named contact.

The named contact is the guarantor for the accuracy of the information in the register record. This may be any member of the review team.

Nicola O'Donnell

Email salutation (e.g. "Dr Smith" or "Joanne") for correspondence:

Nicola O'Donnell

7. * Named contact email.

Give the electronic email address of the named contact.

nicola.odonnell@york.ac.uk

8. Named contact address

Give the full institutional/organisational postal address for the named contact.

Epidemiology and Cancer Statistics Group, Department of Health Sciences, Faculty of Sciences, University of York, Heslington, YO10 5DD

9. Named contact phone number.

Give the telephone number for the named contact, including international dialling code.

01904 320 000

10. * Organisational affiliation of the review.

Full title of the organisational affiliations for this review and website address if available. This field may be completed as 'None' if the review is not affiliated to any organisation.

Epidemiology and Cancer Statistics Group, Department of Health Sciences, Faculty of Sciences, University of York, Heslington, YO10 5DD, United Kingdom

Hull-York Medical School, University of York, York, YO10 5DD, United Kingdom

Centre for Reviews and Dissemination, University of York, York, YO10 5DD, United Kingdom

Population Health Science, Bristol Medical School, University of Bristol, Bristol, BS8 2PS, United Kingdom

University Hospitals Bristol and Weston NHS Foundation Trust, United Kingdom

Department of Psychology, St. Jude Children's Research Hospital, 262 Danny Thomas Place, Memphis, Tennessee, 38105, United States of America

Cancer Survivorship, St. Jude Children's Research Hospital, 262 Danny Thomas Place, Memphis, Tennessee, 38105, United States of America

Organisation web address:

11. * Review team members and their organisational affiliations.

Give the personal details and the organisational affiliations of each member of the review team. Affiliation refers to groups or organisations to which review team members belong. **NOTE: email and country now MUST be entered for each person, unless you are amending a published record.**

Mrs Nicola O'Donnell. Epidemiology and Cancer Statistics Group, University of York
Dr Bob Phillips. Hull-York Medical School/Centre for Reviews and Dissemination, University of York
Dr Jessica Morgan. Centre for Reviews and Dissemination, University of York
Dr Debra Howell. Epidemiology and Cancer Statistics Group, University of York
Dr Leila Ellis. Population Health Science, Bristol Medical School/University Hospitals Bristol and Weston

NHS Foundation Trust
Dr Victoria Willard. Department of Psychology, St. Jude Children's Research Hospital
Dr Daniel Mulrooney. Cancer Survivorship, St. Jude Children's Research Hospital

12. * Funding sources/sponsors.

Details of the individuals, organizations, groups, companies or other legal entities who have funded or sponsored the review.

This review is funded by the Childhood Eye Cancer Trust (CHECT) as part of an award given to Bob Phillips to supervise Nicola O'Donnell to undertake her PhD.

Sponsor: University of York

Grant number(s)

State the funder, grant or award number and the date of award

19/03/Phillips3 September 2021

13. * Conflicts of interest.

List actual or perceived conflicts of interest (financial or academic).

None

14. Collaborators.

Give the name and affiliation of any individuals or organisations who are working on the review but who are not listed as review team members. **NOTE: email and country must be completed for each person, unless you are amending a published record.**

15. * Review question.

State the review question(s) clearly and precisely. It may be appropriate to break very broad questions down into a series of related more specific questions. Questions may be framed or refined using PI(E)COS or similar where relevant.

This systematic review and meta-analysis aims to explore psychosocial interventions designed for Teenage and Young Adult (TYA) survivors of childhood cancer.

Specifically, it will address:

What types of psychosocial interventions exist for TYA survivors of childhood cancer?

Is there a type of psychosocial intervention that provides higher efficacy in improving survivors' mental wellbeing?

Do psychosocial interventions positively influence the wellbeing and psychological health of TYA survivors' and are there any possible negative impacts or 'adverse events'?

16. * Searches.

State the sources that will be searched (e.g. Medline). Give the search dates, and any restrictions (e.g. language or publication date). Do NOT enter the full search strategy (it may be provided as a link or attachment below.)

The literature will be searched extensively and systematically for studies which evaluate any intervention which target TYA aged 13-39 post-treatment survivors of any type of cancer. This age range is determined to incorporate both the UK and USA definitions of 'TYA', as per National Cancer Institute, Cancer Research UK, and James Lind Alliance. We will search for studies published from the year 2000 onwards in the databases of MEDLINE ALL, PsycINFO, Scopus, the Cochrane Library, CINAHL (EBSCO), BNI, PsycARTICLES, and EMBASE. PROSPERO searches and clinical trial registries will be searched to identify any unpublished or ongoing reviews and studies on a similar topic. Forward and backward citation searches will also be utilised. Only articles written in the English language will be included.

Search terms will be exploded and chosen a priori through break down of the research question. Terms that relate to the same concept will be combined using the Boolean operator 'OR'. Concepts will be separated using the operator 'AND'. Terms will be searched in titles, abstracts, and keywords. Search terms will first be devised for MEDLINE ALL and adapted for subsequent databases.

There will be key words that look for:

Child*.tw OR adolesc*.tw OR teenage*.tw OR young adult*.tw

AND

Cancer*.tw OR neoplasm OR survivor* OR oncolog* OR medical oncolog*

AND

Interven* OR educa* OR psychosocial*.tw OR behav* OR mental health OR wellbeing OR group therapy OR support groups OR skills training OR CBT OR psychotherapy OR family therapy OR counselling

17. URL to search strategy.

Upload a file with your search strategy, or an example of a search strategy for a specific database, (including the keywords) in pdf or word format. In doing so you are consenting to the file being made publicly accessible. Or provide a URL or link to the strategy. Do NOT provide links to your search **results**.

https://www.crd.york.ac.uk/PROSPEROFILES/422933_STRATEGY_20230501.pdf

Alternatively, upload your search strategy to CRD in pdf format. Please note that by doing so you are consenting to the file being made publicly accessible.

Do not make this file publicly available until the review is complete

18. * Condition or domain being studied.

Give a short description of the disease, condition or healthcare domain being studied in your systematic review.

Although survival rates are improving, the impact of the cancer and/or treatment can have both long- and short-term effects on psychosocial wellbeing throughout the lifespan (Martiniuk et al., 2014). For the purpose of this review, psychosocial wellbeing and mental health will be considered as unique to each individual and may include quality of life, resilience, social functioning etc. To reduce the impact on wellbeing, psychosocial interventions are increasingly used in clinical oncology practice. Despite this, little is known about the feasibility and efficacy of these, particularly for childhood cancer survivors who are now teenagers and young adults (TYA) (van Dijk-Lokkart et al., 2015; Campo et al., 2017). TYA in cancer settings are classed as a unique group, situated in the middle of healthcare systems aimed at either children or adults (James Lind Alliance, 2018). The psychosocial impact of childhood cancer on this group can be vast, as many experience interrupted development, impacting their cognitive and social outcomes (Patterson et al., 2015). Therefore tailored psychosocial care and interventions must be offered, flexibly responding to the needs of individuals at this life stage (D'Agostino et al., 2011).

19. * Participants/population.

Specify the participants or populations being studied in the review. The preferred format includes details of both inclusion and exclusion criteria.

Survivors of any type of childhood cancer who are currently aged 13-39. This will include survivors of central nervous system/brain tumours, of which we anticipate specific interventions will have been conducted. Post-treatment survivors refer to young people who are in complete remission (including those in remission post-treatment for relapse). This is defined as all signs and symptoms of cancer being completely absent (National Cancer Institute, 2019). In the UK, TYA are referred to as anyone aged between 13-24 years (James Lind Alliance, 2018; Cancer Research UK, 2023) and in the USA is defined as individuals aged between 15-39 (National Cancer Institute, 2023).

20. * Intervention(s), exposure(s).

Give full and clear descriptions or definitions of the interventions or the exposures to be reviewed. The preferred format includes details of both inclusion and exclusion criteria.

All interventions which aim to improve the wellbeing of childhood cancer survivors who are now teenagers or young adults. All intervention types e.g. psychological, social, behavioural, educational if aimed at childhood cancer survivors (e.g. CBT, family systems, counselling, psychoeducation etc). The type of intervention may be considered in sub-group analysis. The intervention may include survivor-only interventions or whole family interventions, but survivor wellbeing must a primary outcome.

21. * Comparator(s)/control.

Where relevant, give details of the alternatives against which the intervention/exposure will be compared (e.g. another intervention or a non-exposed control group). The preferred format includes details of both inclusion and exclusion criteria.

Any, including usual clinical care.

22. * Types of study to be included.

Give details of the study designs (e.g. RCT) that are eligible for inclusion in the review. The preferred format includes both inclusion and exclusion criteria. If there are no restrictions on the types of study, this should be stated.

Inclusion criteria:

Studies that evaluate any psychosocial intervention targeting adolescents aged 13-39 post-treatment of any type of cancer (from day after completion of treatment onwards)

- Interventions that target the family as a whole if survivor wellbeing is a primary outcome

- All intervention types e.g. psychological, social, behavioural, educational if aimed at childhood cancer survivors

- Studies that consider long-term conditions as a whole if they meet other criteria and that oncology data can be analysed separately

- Studies with RCT design including mixed-methods studies

Exclusion criteria:

- Studies that are not published in English

- Interventions focused on individuals receiving palliative care

23. Context.

Give summary details of the setting or other relevant characteristics, which help define the inclusion or exclusion criteria.

24. * Main outcome(s).

Give the pre-specified main (most important) outcomes of the review, including details of how the outcome is defined and measured and when these measurement are made, if these are part of the review inclusion criteria.

Childhood cancer survivor wellbeing as measured by quality of life questionnaires.

Measures of effect

Please specify the effect measure(s) for you main outcome(s) e.g. relative risks, odds ratios, risk difference, and/or 'number needed to treat.

Standardised mean difference. Odds ratio and relative risk may also be considered.

25. * Additional outcome(s).

List the pre-specified additional outcomes of the review, with a similar level of detail to that required for main outcomes. Where there are no additional outcomes please state 'None' or 'Not applicable' as appropriate to the review

Other validated measures of psychological wellbeing e.g. Warwick-Edinburgh Mental Well-being Scale (WEMWBS).

Measures of effect

Please specify the effect measure(s) for you additional outcome(s) e.g. relative risks, odds ratios, risk difference, and/or 'number needed to treat.

26. * Data extraction (selection and coding).

Describe how studies will be selected for inclusion. State what data will be extracted or obtained. State how this will be done and recorded.

The lead reviewer Nicola O'Donnell will conduct database searches and import these into Covidence reference management software. Any duplicate or irrelevant articles will be identified, noted, and removed. Remaining texts will be screened by title and abstract for inclusion suitability. The full text of all records deemed appropriate will be retrieved and reviewed against inclusion criteria. Reference lists of all relevant articles will also be searched. All above searches will be completed by the lead reviewer due to resource and time limitations. A proportion will, however, be checked="checked" value="1" by a second reviewer, Dr Leila Ellis. Any disagreements over inclusion will be managed using consensus discussion and through a third reviewer. Information will be extracted from the selected studies and summarised in a data extraction table. This will include author, publication year, study setting, characteristics of survivors (age, sex, ethnicity, sociodemographic factors, type of cancer, length of remission), inclusion/exclusion criteria, details of the intervention and comparator (including mode of delivery, setting of intervention, individuals delivering intervention, duration, theoretical basis, post-intervention follow-up), outcome measures, effect sizes, study limitations. If studies include a mixed population (e.g. survivors vs not), survivor data will be extracted.

If it is found that any necessary eligibility data is missing, the authors will be contacted. Should this information remain inaccessible it will be removed from the study and noted.

27. * Risk of bias (quality) assessment.

State which characteristics of the studies will be assessed and/or any formal risk of bias/quality assessment tools that will be used.

Version 2 of the Cochrane risk-of-bias tool for randomised trials (RoB 2) will be used. This tool provides a judgement of 'low risk' to 'high risk' and determines whether there are 'some concerns' about specific bias.

28. * Strategy for data synthesis.

Describe the methods you plan to use to synthesise data. This **must not be generic text** but should be **specific to your review** and describe how the proposed approach will be applied to your data. If meta-analysis is planned, describe the models to be used, methods to explore statistical heterogeneity, and software package to be used.

A preliminary synthesis of the data will take place focusing on the type of intervention, type of outcome, intervention content, duration of intervention/follow-up and population characteristics. Depending upon the clinical heterogeneity of the studies, a fixed-effect or random-effect meta-analysis will be performed. Visual inspection of forest plots and calculation of the I^2 statistic will be used with a 50% limit as an indicator of substantial heterogeneity (Higgins, 2011). End point scores will be expressed as standardised mean differences (SMDs) with associated 95% confidence intervals (CIs). Random or fixed effect meta-analysis will be performed using RevMan 5 software on the basis of adequate similarity of outcomes assessed.

29. * Analysis of subgroups or subsets.

State any planned investigation of 'subgroups'. Be clear and specific about which type of study or participant will be included in each group or covariate investigated. State the planned analytic approach.

Sub-group analysis may be conducted based upon the age of participants in relation to the impact of the intervention, due to interest in any difference in intervention efficacy for teenagers vs young adults. Additional sub-group analysis may be conducted when participants have undergone treatment for brain tumours, such as cranial irradiation.

30. * Type and method of review.

Select the type of review, review method and health area from the lists below.

Type of review

Cost effectiveness
No

Diagnostic
No

Epidemiologic
No

Individual patient data (IPD) meta-analysis
No

Intervention
Yes

Living systematic review
No

Meta-analysis
Yes

Methodology
No

Narrative synthesis
No

Network meta-analysis
No

Pre-clinical
No

Prevention
No

Prognostic
No

Prospective meta-analysis (PMA)
No

Review of reviews
No

Service delivery
No

Synthesis of qualitative studies
No

Systematic review
Yes

Other
No

Health area of the review

Alcohol/substance misuse/abuse
No

Blood and immune system
No

Cancer
Yes

Cardiovascular
No

Care of the elderly
No

Child health
No

Complementary therapies
No

COVID-19
No

Crime and justice
No

Dental
No

Digestive system
No

Ear, nose and throat
No

Education
No

Endocrine and metabolic disorders
No

Eye disorders
No

General interest

No

Genetics

No

Health inequalities/health equity

No

Infections and infestations

No

International development

No

Mental health and behavioural conditions

Yes

Musculoskeletal

No

Neurological

No

Nursing

No

Obstetrics and gynaecology

No

Oral health

No

Palliative care

No

Perioperative care

No

Physiotherapy

No

Pregnancy and childbirth

No

Public health (including social determinants of health)

No

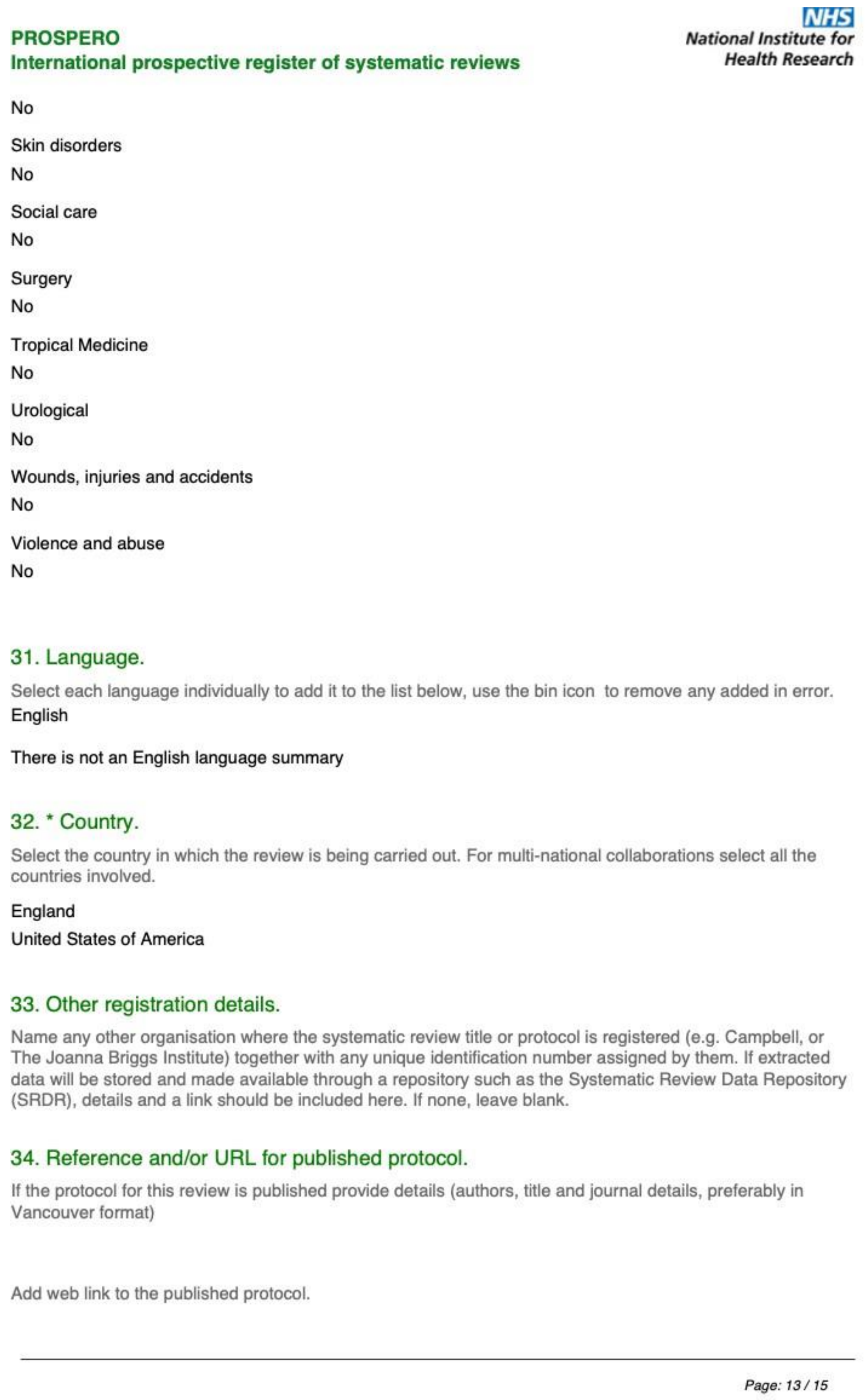
Rehabilitation

No

Respiratory disorders

No

Service delivery



No

Skin disorders

No

Social care

No

Surgery

No

Tropical Medicine

No

Urological

No

Wounds, injuries and accidents

No

Violence and abuse

No

31. Language.

Select each language individually to add it to the list below, use the bin icon to remove any added in error.
English

There is not an English language summary

32. * Country.

Select the country in which the review is being carried out. For multi-national collaborations select all the countries involved.

England
United States of America

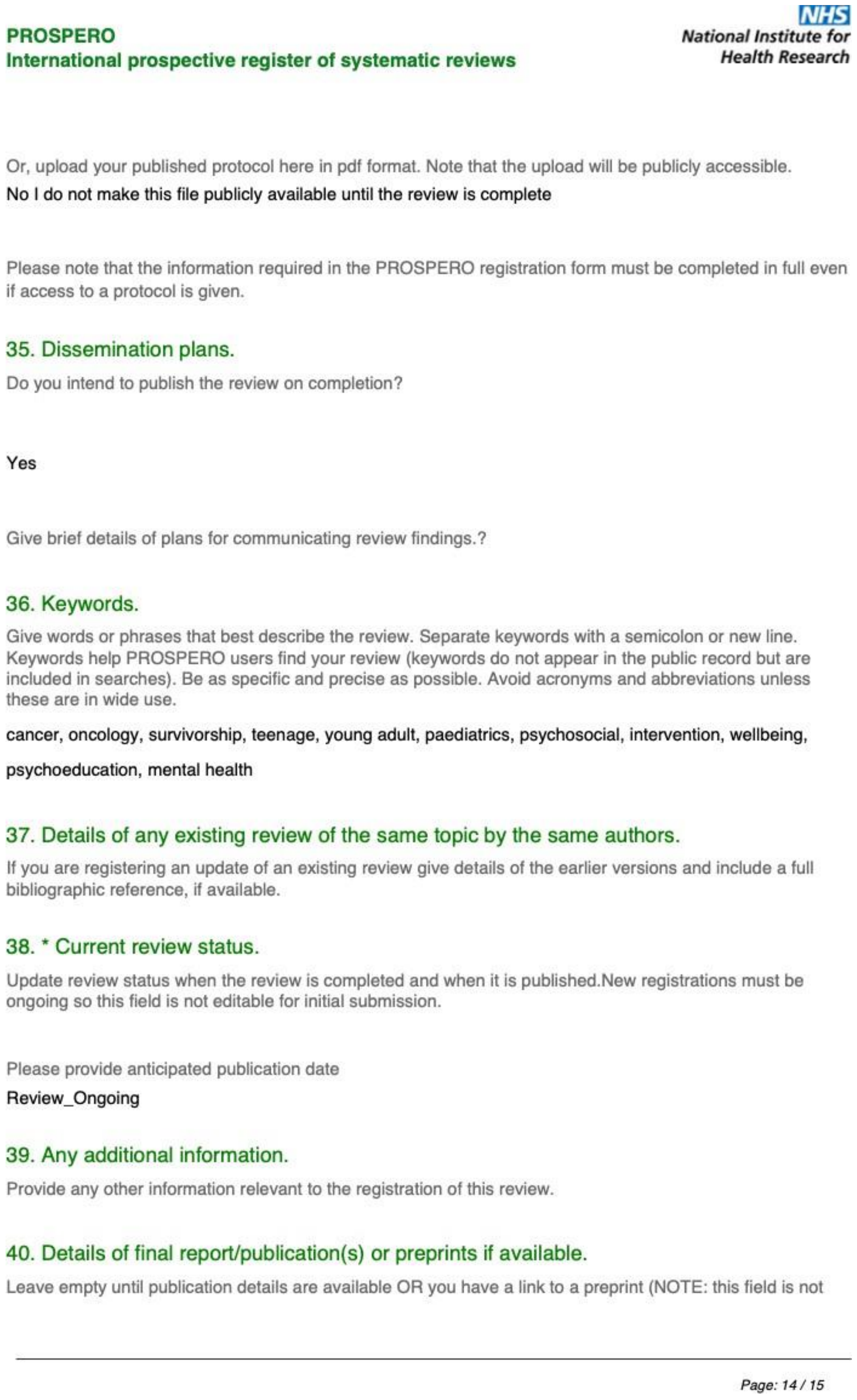
33. Other registration details.

Name any other organisation where the systematic review title or protocol is registered (e.g. Campbell, or The Joanna Briggs Institute) together with any unique identification number assigned by them. If extracted data will be stored and made available through a repository such as the Systematic Review Data Repository (SRDR), details and a link should be included here. If none, leave blank.

34. Reference and/or URL for published protocol.

If the protocol for this review is published provide details (authors, title and journal details, preferably in Vancouver format)

Add web link to the published protocol.



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Please note that the information required in the PROSPERO registration form must be completed in full even if access to a protocol is given.

35. Dissemination plans.

Do you intend to publish the review on completion?

Yes

Give brief details of plans for communicating review findings.?

36. Keywords.

Give words or phrases that best describe the review. Separate keywords with a semicolon or new line. Keywords help PROSPERO users find your review (keywords do not appear in the public record but are included in searches). Be as specific and precise as possible. Avoid acronyms and abbreviations unless these are in wide use.

cancer, oncology, survivorship, teenage, young adult, paediatrics, psychosocial, intervention, wellbeing,

psychoeducation, mental health

37. Details of any existing review of the same topic by the same authors.

If you are registering an update of an existing review give details of the earlier versions and include a full bibliographic reference, if available.

38. * Current review status.

Update review status when the review is completed and when it is published.New registrations must be ongoing so this field is not editable for initial submission.

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39. Any additional information.

Provide any other information relevant to the registration of this review.

40. Details of final report/publication(s) or preprints if available.

Leave empty until publication details are available OR you have a link to a preprint (NOTE: this field is not editable for initial submission). List authors, title and journal details preferably in Vancouver format.

Give the link to the published review or preprint.

APPENDIX B2: PRISMA CHECKLIST

../3. PRISMA checklist.pdf

APPENDIX B3: SEARCHES

4. Searches.xlsx

Appendix B4: Characteristics of Included Studies

Author, publication date, study design, *doi*	**Study setting**	**Participant characteristics** *(age, sex, ethnicity, sociodemographic factors, type of cancer, length of remission)*	**Inclusion/exclusion criteria**	**Intervention/comparator** *(including mode of delivery, setting of intervention, individuals delivering intervention, duration, theoretical basis, post-intervention follow-up)*	**Outcome Measures***
Arpaci 2023 RCT *10.1097/NCC.0000000000001091*	Turkey	122 were assessed for eligibility, 60 were excluded. 62 were randomised; 30 to intervention, 32 to control. **Intervention n=24** **Control=31** **Mean age:** 14.87 **Sex:** 13 female, 11 male **Ethnicity:** not reported **Sociodemographic:** *Education:* secondary school 12, high school 12 *Family income:* expenses more than	**Inclusion criteria:** 1) patients who received chemotherapy and/or radiotherapy for childhood leukaemia, 2) aged 12 to 18 years, 3) at least 2 or more years since the completion of treatment, 4) able to use a computer and/or smartphone, 5) able to access the internet, 6) able to speak Turkish, and 7) their parents agreed to participate in the study. **Exclusion criteria:** 1) were treated with hematopoietic stem cell transplantation, 2) have relapsed, 3) are	**Technology-Based Psychosocial Education and Counseling Programme** **Intervention Group:** **Intervention Type:** Telephonic education and counselling programme **Duration:** 10 weeks **Delivery:** Telephonic communication, website interaction, and messages **Details of the Intervention:** 1. Participants informed about registration, website usage, and programme execution via telephone. 2. Participants created personal login credentials after registration. 3. 1-week review period for website access with specific menus.	The Pediatric Quality of Life Inventory (The PedsQL 4.0) The Self-Efficacy Questionnaire for Children (SEQ-C) The KIDCOPE

		income 5, income equal to expenses 14, income more than expenses 5 **Cancer type:** ALL 22, AML 2 **Mean years since treatment completed:** 6.52 **Control n=31** **Mean age:** 15.50 **Sex:** 15 female, 16 male **Ethnicity:** not reported **Sociodemographic:** *Education:* secondary school 11, high school 20 *Family income:* expenses more than income 5, income equal to expenses 21, income more than expenses 5 **Cancer type:** ALL 30, AML 1 **Mean years since**	physically or mentally disabled, or 4) have severe psychiatric problems. Adolescents who developed any of these exclusion criteria during the study and could not complete the programme were withdrawn.	4. 10-week education and counselling programme with modular structure. 5. Each module divided into a 2-week period (1 week for education, 1 week for counselling). 6. Modules conducted by the first author, including survivor interaction, feedback, and discussion. 7. Messages sent to adolescents for reminders, motivation, and reinforcement. 8. Motivational messages sent during the second week of each module. 9. Telephone support and counselling available during the process. 10. General module evaluation and individual positive feedback provided after completion. **Control Group:** **Intervention Type:** Routine LTFU care **Duration:** Continued throughout the programme and later included in the 10-week education and counselling programme for the intervention group. **Delivery:** Standard LTFU care with measurements at follow-up times.	

		treatment completed: 6.89			
Berg 2020 RCT *10.1007/s13187-019-01574-7*	USA	63 participants were originally enrolled: 44 in the intervention group and 19 in the control. 7 withdrew leaving a final sample size of 56. **Intervention n=38** **Control n= 18** **Intervention group:** **Mean age:** 32.63 **Sex:** 28 female, 10 male **Ethnicity:** White 32, Black 3, other 3 **Sociodemographic:** *Marital status;* married (22), other (16) *Education;* <bachelors (19), >bachelors (19) *Employment status;* employed (24), student (5), other (9) Income; <$2,400 per month (17),	**Inclusion Criteria:** (1) 18–40 years old; (2) speak English; (3) within 2 years of cancer treatment completion; (4) functioning smartphone; and (5) willing to complete study activities (including scheduling an in-person appointment with survivorship clinic). **Exclusion criteria:** (1) cancer recurrence since treatment completion; (2) diagnosis of a central nervous system cancer (to ensure requisite mental/emotional functioning to engage in the programme); (3) prior diagnosis of alcohol or drug dependency, psychosis, bipolar disorder, or major	**The AWAKE intervention** **Intervention Group (AWAKE):** **Intervention Type:** Empirically supported protocol augmented with evidence-based strategies, specifically targeting Young Adult Cancer Survivors (YACS) **Duration:** Eight weekly modules **Delivery:** App-based platform with customised daily mood and health behaviour monitoring, accompanied by weekly calls from trained Masters-level female staff following an empirically supported protocol **Attention Control Group (AC):** **Intervention Type:** Educational materials on personal finance delivered via text **Duration:** Eight weeks **Delivery:** Weekly opportunities for coaching calls, no homework assignments, limited ability to track health behaviours and mood	Primary efficacy outcome was hope as measured by the Adult Trait Hope Scale. QOL: (1) the RAND Medical Outcome Study 36-Item Short Form Health Survey (SF-36) and (2) the Functional Assessment of Cancer Therapy-General (FACT-G). Depressive symptoms were assessed using

		>$2,400 per month (21) *Insurance;* other (25), Medicare/Medicaid (9), none (3) **Cancer type:** breast (10), melanoma (6), leukaemia/lymphoma (5), sarcoma (5), colorectal (1), testicular (1), cervical (2), other (8) **Mean years since diagnosis**: 2.11 years **Control group:** **Control n=18 Mean age:** 32.39 **Sex:** 14 female, 4 male **Ethnicity:** White 13, Black 4, other 1 **Sociodemographic:** *Marital status;* married (16), other (2) *Education;* <bachelors (10), >bachelors (8) *Employment status;* employed (12), student	depressive disorder; and (4) in hospice.		the Patient Health Questionnaire-9 item (PHQ-9).

		(3), other (3) Income; <$2,400 per month (6), >$2,400 per month (12) *Insurance;* other (15), Medicare/Medicaid (3), none (0) **Cancer type:** breast (6), melanoma (3), leukaemia/lymphoma (2), sarcoma (0), colorectal (2), testicular (2), cervical (0), other (3) **Mean years since diagnosis:** 2.00 years			
Cheung 2019 RCT *10.1002/pon.4929*	Hong Kong, China	78 expressed an interest, 15 were excluded. 60 were randomly assigned to experimental or control group. 1 experimental group participant discontinued, 3 in the control group dropped out.	**Inclusion:** Hong Kong Chinese paediatric brain tumour survivors who had a baseline Centre for Epidemiological Studies Depression Scale for Children(CES-DC) score of 16 or above, and a Modified Mini-Mental Scale (MMSE) score of 18 or above (to ensure that participants had	**Musical Training** **Intervention Group:** **Intervention Type:** Weekly 45-minute lessons on musical training **Duration:** 52 weeks **Delivery:** Weekly home visits by research assistants focusing on musical training **Control Group:**	The primary outcome was depressive symptoms at 12 months, which was measured by using the Chinese version of the CES-DC.

		Intervention n=30 **Mean age:** 12.5 **Sex:** 13 female, 17 male Ethnicity: none reported **Sociodemographic:** none reported *except parental educational attainment;* 8 with tertiary education, 13 upper secondary, 6 lower secondary, 3 primary or lower **Cancer type:** brain tumours **Months since treatment completed:** 6-11 (11), 13-24 (5), 25-36 (3), 37-48 (3), 49-60 (1), >60 (7) **Control n=30 Mean age:** 13.9 **Sex:** 12 female, 18 male **Ethnicity:** none reported **Sociodemographic:** *none reported except*	the capability to receive musical training) - survivors who have completed cancer treatment at least 2 months previously -aged between 7 and 16 years -able to speak Cantonese and read Chinese -those who did not undertake any musical training before. **Exclusion:** survivors with evidence of cancer recurrence or second malignancy in their medical records.	**Intervention Type:** Placebo control intervention **Duration:** 52 weeks **Delivery:** Weekly 45-minute home visits by research assistants involving unstructured leisure activities like card games, chess, and watching online content; designed to mimic time and attention given to the experimental group without specific effects on dependent variables.	Rosenberg Self-Esteem Scale (RSES) is a tool designed to measure the global self-esteem of children and adolescents. Chinese version of the Pediatric Quality of Life Inventory 4.0 Generic Core Scale (PedsQL 4.0),

		parental educational attainment; 5 with tertiary education, 15 upper secondary, 7 lower secondary, 3 primary or lower **Cancer type:** brain tumours **Months since treatment completed:** 6-11 (10), 13-24 (4), 25-36 (5), 37-48 (4), 49-60 (1), >60 (6)			
van Dijk-Lokkart 2016 RCT *10.1002/pon.4016*	The Netherlands	Of the 174 patients eligible for the QLIM study, 68 participated. **Intervention n= 30** **Mean age:** 13.0 **Sex:** 14 female, 16 male **Ethnicity:** None reported **Sociodemographic:** None reported **Cancer type:** Leukaemia/lymphoma	**Inclusion:** diagnosed with any type of childhood malignancy, were ≤12 months off treatment, and were treated with chemotherapy and/or radiotherapy. **Exclusion:** patients requiring stem cell transplantation and/or growth hormone therapy, as well as patients who were wheelchair-	**FitSurvivor** **Physical Exercise Training:** **Intervention Type:** Highly intensive combination of cardiorespiratory and muscle strength training **Duration:** Two sessions per week (45 min each) for 12 weeks **Delivery:** Conducted in a local physiotherapy practice with sessions progressively increasing in duration and intensity; sessions guided by a local physiotherapist using a detailed instruction manual.	Health-related QoL Both self-report and parent-proxy report of HrQoL were assessed using three versions of the Dutch PedsQL. To assess parental perception of

		(20), brain tumours/CNS tumours (2), solid tumours (8) **Mean years since treatment completed:** Not reported **Control n= 38 Mean age:** 12.6 **Sex:** 18 female, 20 male **Ethnicity:** None reported **Sociodemographic:** None reported **Cancer type:** Leukaemia/lymphoma (26), brain tumours/CNS tumours (5), solid tumours (7) **Mean years since treatment** completed: Not reported	dependent, not able to 'ride a bike', and not able to read, write, self-reflect and/or follow instructions because of learning difficulties.	**Psychosocial Training:** **Intervention Type:** Individualized structured programme focusing on socio-emotional functioning and coping with disease-related effects **Duration:** Six child sessions of 60 min each (once every 2 weeks), and two parent sessions (at the start and end of the programme) over 12 weeks **Delivery:** Child sessions held in the treating hospital, covering psychoeducation and cognitive-behavioural techniques on various topics; positive evaluations and good adherence rates reported. **Control Group:** **Intervention Type:** 'Care as usual' with variations based on local guidelines and preferences **Duration:** Varied based on local circumstances **Delivery:** No routine exercise or psychosocial training; psychological care and/or physiotherapy available on demand.	behavioural problems in children aged 6–18 years, the Child Behaviour Checklist was used. All participants aged ≥11 years also completed the Youth Self-Report to assess self-reported behavioural problems. Depressive symptoms Patients completed the Children's Depression Inventory The Dutch versions of the

					Self Perception Profile for children and adolescents were used to measure self-perception of the patients.
Grenawalt 2023 RCT *https://doi.org/10.1080/09638288.2022.2094478*	USA	142 participants were recruited, 139 were eligible. 127 of these were retained and included in the present analysis; 63 control participants and 64 intervention group participants. **Intervention n= 64** **Mean age:** 23.8 **Sex:** 21 female, 42 male **Ethnicity:** 49 White, 6 Black or African American, 5 Hispanic or Latino/a, 1 Asian, 2 Native Hawaiian or	**Inclusion:** young adults between the ages of 18 and 30 with a diagnosis of brain tumour before the age of 18. **Exclusion:** individuals that did not have capacity to consent to research as determined by CBTF personnel (e.g., neuropsychological records).	**Internet-based BA Intervention (two parallel groups)** **Intervention Type:** BA Intervention **Duration:** Four modules, each expected to take approximately 30 minutes Participants were asked to complete one module per week with a minimum of three days between modules, lasting for a total of four weeks **Delivery:** Internet-based intervention with a focus on promoting QOL • Modules included values assessment, mindfulness exercises, and social skills education based on Kanter et al.'s core	The Life Satisfaction Questionnaire (LiSat-9) Perceived Stress Scale (PSS-10). BA for Depression Scale – Short Form (BADS-SF)

		Pacific Islander, 1 Native American or Alaskan Native, 0 preferred not to answer **Sociodemographics:** *Living situation:* with parents/family: 41, on own: 5, with spouse: 6, with roommates: 11. *Education status:* did not complete high school: 8, currently in high school: 7, completed high school: 20, currently in college: 8, completed associate degree: 13, completed bachelor's degree: 4, completed graduate degree:2, other: 2 *Employment status:* FT 27, PT 13, not employed 13, student 8, volunteer 2 **Cancer type:** brain tumour **Mean years since**		components of Behavioural Activation (BA) • Permission obtained to adapt a brief; Internet-based BA intervention developed by Bunge et al. • Modules guided participants through activities related to values, goal setting, identifying barriers, mindfulness practices, and positive social relationships • Emphasis on activities leading to happiness, a sense of accomplishment, or meaningfulness • Encouraged commitment through the creation of a written contract as a certificate of completion.	

		treatment completed: Not reported **Control n= 63 Mean age:** 23.9 **Sex:** 25 female, 38 male **Ethnicity:** 47 White, 6 Black or African American, 4 Hispanic or Latino/a, 3 Asian, 2 Native Hawaiian or Pacific Islander, 0 Native American or Alaskan Native, 1 preferred not to answer **Sociodemographics:** *Living situation:* with parents/family: 49, on own: 6, with spouse: 3, with roommates: 4. *Education status:* did not complete high school: 7, currently in high school: 6, completed high school: 20, currently in college: 5, completed			

		associate degree: 14, completed bachelor's degree: 11 completed graduate degree:0, other: 0 *Employment status:* FT 21, PT 18, not employed 11, student 11, volunteer 2 **Cancer type:** brain tumour **Mean years since treatment completed:** Not reported			
Haydon 2021 RCT (three arm) *(* *– no DOI)*	USA	2,000 survivors were contacted, 2 more got in contact via an invited friend. 284 responded and 217 were screened; 8 declined, 6 were ineligible. The final sample was 203. **Intervention n= 67** **Mean age:** 31.99 **Sex:** 32 female, 35 male	**Inclusion:** (1) a cancer diagnosis between the ages of 15 and 39 years; (2) completion of primary treatment and currently in partial or complete remission; (3) current age between 18 and 39 years old; (4) fluent in English; and (5) access to the internet and email.	**Online prosocial intervention** **Intervention Type:** • Peer Helping Condition • Expressive Writing + Peer Helping Condition • Fact-Writing Control Condition **Duration:** • Four weeks **Delivery:**	14-item Mental Health Continuum-Short Form (the MHC-SF) 20-item Centre for Epidemiologic Studies Depression Scale.

		Ethnicity: Black 3%, Asian 7%, White 44%, Other 9%, Latina 14% **Sociodemographics:** *Education:* <College 16%, college graduate 33%, >college 18% *Employment:* Employed FT/PT: 44% *Income:* >$100,000: 37% *Relationship:* Married: 29% **Cancer type:** Brain and CNS tumours 2%, Breast 10%, Leukaemia 11%, Lymphoma 9%, Melanoma 5%, Sarcoma 4%, Testicular 2%, Thyroid 13%, Other 11% **Mean years since treatment completed:** Not reported **Intervention 2 n= 66**	**Exclusion:** (1) currently receiving primary treatment for a cancer recurrence and (2) inability to commit to the intervention for 4 consecutive weeks.	**Peer Helping Condition:** Participants write about their cancer experience with a focus on benefiting a newly diagnosed AYA cancer patient. **Expressive Writing + Peer Helping Condition:** Participants write about their deepest thoughts and feelings regarding their cancer experience in the first three activities and provide advice or support for a newly diagnosed AYA cancer patient in the fourth activity. **Fact-Writing Control Condition:** Participants provide factual details about their diagnosis, treatment, and daily lives, focusing on creating a personal resource without delving into their thoughts or feelings at the time.	7-item generalized anxiety disorder scale (GAD-7) Cognitive function, sleep disturbance, fatigue, and pain were assessed via brief measures from the National Institute of Health funded Patient Reported Outcomes Measurement Information System (PROMIS). Pain intensity over the past week was

		Mean age: 32.17 **Sex:** 32 female, 35 male **Ethnicity:** Black 3%, Asian 6%, White 47%, Other 10%, Latina 18% **Sociodemographics:** *Education:* <College 19%, college graduate 24%, >college 23% *Employment:* Employed FT/PT: 42% Income >$100,000: 24% *Relationship:* Married: 30% **Cancer type:** Brain and CNS tumours 1%, Breast 10%, Leukaemia 10%, Lymphoma 7%, Melanoma 1%, Sarcoma 6%, Testicular 2%, Thyroid 12%, Other 17% **Mean years since treatment completed:** Not reported			measured using the 1-item PROMIS Numeric Rating Scale, Pain Intensity 1a. Social support was measured using the 21-item 2-way Social Support Scale (2-Way SSS) Affect was measured using subscales from the Positive and Negative Affect Schedule (PANAS-X) The 9-item Balanced Measure of

		Control n = 68 **Mean age:** 32.87 **Sex:** 37 female, 31 male **Ethnicity:** Black 0%, Asian 10%, White 50%, Other 7%, Latina 11% **Sociodemographics:** *Education:* <College 16%, college graduate 23%, >college 29% *Employment:* Employed FT/PT: 47% Income >$100,000: 34% *Relationship:* Married: 30% **Cancer type:** Brain and CNS tumours 5%, Breast 10%, Leukaemia 3%, Lymphoma 10%, Melanoma 4%, Sarcoma 7%, Testicular 3%, Thyroid 9%, Other 17% **Mean years since treatment completed:** Not reported			Psychological Needs. The 20-item Self-Report Altruism Scale.

Howell 2018 RCT *10.1002/pbc.27216*	USA	189 eligible, 97 enrolled, 78 completed. Aged >11 and <15 and in active follow up, and did not meet the centres for Disease Control and Prevention physical activity guidelines at enrolment (>60 min of activity a day, 7 days a week) **Intervention n=53** **Mean age:** 12.8 **Sex:** 29 female, 24 male **Ethnicity:** 7 Black, 44 White, 2 'other' **Sociodemographics**: None reported **Cancer type:** 12 had ALL, 0 AML, 14 CNS tumours, 1 Ewing Sarcoma, 1 Germ Cell tumour, 1 Hodgkin lymphoma, 3	Not reported	**Web-based physical activity intervention** **Intervention Type:** Web-Based Physical Activity Intervention **Duration:** 24 weeks **Delivery:** • Participants in the intervention group received educational materials, an activity monitor, and access to an interactive website. • The website encouraged physical activity through a reward system where participants earned points based on daily activity levels. • Participants created avatars to represent themselves on the website. • The goal was to progress the avatar through various levels using earned points. • Points could be redeemed for small prizes (e.g., t-shirts, stickers) and/or gift cards. • Participants in the control group received only an activity monitor and educational materials.	**Pediatric Quality of Life Inventory (PedsQL) v4.0**

		Neuroblastoma, 3 Non-Hodgkin lymphoma, 1 Retinoblastoma, 3 Rhabdomyosarcoma, 1 Soft tissue sarcoma, 4 Wilms tumour, 1 ‘other’ **Mean years since treatment completed:** not reported **Control n= 25 Mean age:** 12.4 **Sex:** 14 female, 11 male **Ethnicity:** 6 Black, 18 White, 1 ‘other’ **Sociodemographics:** None reported **Cancer type:** 6 had ALL, 1 AML, 3 CNS tumours, 0 Ewing Sarcoma, 0 Germ Cell tumour, 1 Hodgkin lymphoma, 3 Neuroblastoma, 3 Non-Hodgkin lymphoma, 2 Retinoblastoma, 1			

		Rhabdomyosarcoma, 0 Soft tissue sarcoma, 2 Wilms tumour, 3 ‘other’ **Mean years since treatment completed:** not reported			
Judge Santacroce 2010 RCT *10.1177/1043454209340325*	USA	126 potential participants, 51 eligible, 45 approached. 21 AYA enrolled, 19 retained. Intervention and control group demographics not separated. **Total n= 19** **Mean age:** 21 **Sex:** 9 female, 15 male **Ethnicity:** White 85%, Hispanic 5%, Asian 5%, Black 0%, mixed race 5%. **Sociodemographics :** None reported **Cancer type:**	**Inclusion:** (a) diagnosis of childhood cancer at least 5 years ago, (b) treatment completed at least 2 years ago, (c) no evidence of active cancer, (d) current age between 15 and 25 years, (e) English-speaking, and (f) available to receive the intervention by telephone within the state of Connecticut.	**Telephone-Delivered Craniosacral Therapy Technique (CTT)** **Intervention Type:** Telephone-Delivered Craniosacral Therapy Technique (CTT) **Duration:** Seven sessions of 30 minutes each for both the control (HEROS) and experimental (HEROS PLUS) conditions **Delivery:** • Participants randomized into two treatment conditions after T1 data collection: a. Control condition (HEROS) involving usual Long-Term Follow-Up (LTFU) clinical care b. Experimental condition (HEROS PLUS) involving usual LTFU clinical care plus telephone-	Mishel Uncertainty in Illness Scale–Community (MUIS-C) 20-item StateTrait Anxiety Inventory–State subscale **Post trauma symptoms** measured by the 20-item Posttraumatic Stress Disorder Reaction Index

		Leukaemia 29%, Lymphoma 19%, CNS tumour 10%, other solid tumour 42%. **Mean time since treatment completed:** 9.9 years		delivered Craniosacral Therapy Technique (CTT) - HEROS PLUS sessions were based on Forman's outline for CST, adapted for AYA childhood cancer survivors and parents. - Telephone-delivered one-to-one sessions following regular clinical visits. - HEROS PLUS CST was outlined in a manual and delivered by an interventionist with training and ongoing supervision. - Sessions 2 through 6 included instruction, practice, and out-of-session assignments at the end of each session.	**Benefit finding** Benefit finding was measured by 2 indicators: the Posttraumatic Growth Inventory (PTGI) and the Growth Through Uncertainty Scale Health promotion behaviour was measured by the Health Promoting Lifestyle II

Kazak 2004 RCT (wait list control) *10.1037/0893-3200.18.3.493*	USA	530 presumed eligible families were contacted, 330 were confirmed eligible. 150 were enrolled in the study. Survivors as a whole were 85% White, 9% Black, 5% Hispanic, and 1% Asian. Median household income was between $50,000-$70,000. **Intervention n= 76** **Mean age:** 14.62 **Sex:** 39 female, 37 male **Ethnicity:** 88% White, no other ethnicities reported **Sociodemographics:** None reported **Cancer type:** 33% had Leukaemia, 25% solid tumours, 17%	**Exclusion:** relapse, ‘mental retardation’, lack of fluency in English, and residence greater than 150 miles from the hospital.	**The Surviving Cancer Competently Intervention Programme (SCCIP)** **Intervention Type:** • Surviving Cancer Competently Intervention Programme (SCCIP) • Family Group Treatment Model **Duration:** • Four sessions conducted in a single day • Sessions 1 and 2 (morning) focused on cognitive-behavioural principles to reduce distress • Sessions 3 and 4 (afternoon) utilized family therapy approaches • SCCIP provided 5 hours of direct therapeutic contact and an additional 2 hours of informal contact during breaks **Delivery:** • Manualized intervention designed to reduce Posttraumatic Stress Symptoms (PTSS) in adolescent survivors of childhood cancer and their families • Separate sessions for survivors, mothers, fathers, and siblings in the morning	**Impact of Events Scale—Revised (IES-R)** **Post-Traumatic Stress Disorder Reaction Index (PTSD-RI)** **State–Trait Anxiety Inventory (STAI)** **Revised Children’s Manifest Anxiety Scale (RCMAS)**

		Lymphoma, 8% bone tumours, 17% ‘other’ **Mean years since treatment completed:** 5.49 **Control n= 74** **Mean age:** 14.60 **Sex:** 38 female,36 male **Ethnicity:** 81% White, no other ethnicities reported **Sociodemographics:** None reported **Cancer type:** 28% had Leukaemia, 20% solid tumours, 28% Lymphoma, 8% bone tumours, 15% ‘other’ **Mean years since treatment completed:** 5.06		- Afternoon sessions involved all members of participating families together - Family group treatment model implemented in both morning and afternoon sessions - All families completed all four sessions.	
Kunin-Batson 2016	USA	Invitations were sent to 88 participants, 52	**Inclusion:** participants were between 15–29 years of age at	**Web-based resource to improve cancer knowledge**	State Trait Anxiety

RCT (standard of care control) *10.1002/pon.3956*		responded and provided informed consent and assent for those <18. **Intervention n=26:** **Mean age:** 21.5 **Sex:** 13 female, 13 male **Ethnicity:** 89% White/non-Hispanic **Sociodemographics:** N one reported **Cancer type:** ALL 27, AML/CML 7.7, Hodgkin/other Lymphoma 19.2, Soft tissue Sarcoma 7.7, Wilms Tumour 3.8, CNS Tumour 7.7, Bone Tumour 15.4, other* 11.5 **Mean years since treatment completed:** not reported **Control n=26** **Mean age:** 21.5	recruitment, with a history of hematologic malignancy or malignant neoplasm, and were off treatment and in first remission. Eligible participants were also English speaking and reported having access to a computer with Internet access and were without significant visual or neurologic/cognitive impairments which in the judgment of their oncologist would restrict their ability to see and understand website content and complete survey measures.	**Intervention Type:** Web-Based Health Information and Support Intervention **Duration:** One year **Delivery:** • Participants in the intervention group received access to a HIPAA-compliant, password-protected website. • Components of the website included the participant's treatment summary, hyperlinks to user-friendly information about health and treatment-related late effects, optional fields for other aspects of health history, tips for communication with healthcare providers, an e-journal, a "contact us" messaging system to connect with the medical team, and links to relevant local and national resources. • Continuous access to the personalized website was provided throughout the duration of the study for individuals randomized to the intervention condition.	Inventory (STAI) Multidimensional Health Locus of Control (HLC)

		Sex: 11 female, 15 male **Ethnicity:** 92% White/non-Hispanic **Sociodemographics:** None **Cancer type:** ALL 35, AML/CML 3.8, Hodgkin/other Lymphoma 11.5, Soft tissue Sarcoma 7.7, Wilms Tumour 3.8, CNS Tumour 3.8, Bone Tumour 26.9, other* 7.7 *Neuroblastoma, colon, testicular, uterine, liver **Mean years since treatment completed:** none			
Li 2022 RCT	China	268 assessed for eligibility. 153 surveyed at baseline and randomly assigned; 48 control	**Inclusion:** (1) 15 to 39 years of age, (2) diagnosed with cancer, (3) voluntary participation in research, and (4) no history of mental	**Intervention Type:** **PA Group:** Physical Activity Intervention **BA Group:** BA Intervention **Control Group:** Usual Treatment and Follow-Up	The Pittsburgh Sleep Quality Index (PSQI) was developed by

10.1097/NCC.0000000000000932		group, 47 PA group, 48 BA group. **PA intervention n=47:** **Mean age:** 28.4 **Sex:** 38 female, 9 male **Ethnicity:** None reported **Sociodemographics:** *Education:* elementary school or lower (10), middle school (15), high school (8), junior college or university (14), master and above (0) *Marital status:* single (8), married (36), divorced/widowed (3) *Monthly income:* <500 (14), 500-1000 (6), 1000-3000 (13), 3000-5000 (9), >5000 (5) **Cancer type:** Lung (10), gynaecological (10), breast (14), others*	illness before the diagnosis of cancer. **Exclusion:** (1) had an existing mental illness or other system diseases, (2) had a communication disorder, (3) exhibited drug or alcohol dependency, and (4) the DT was less than 4 points (no psychological distress) or the PAR-Q was more than 1 "yes" (need medical examination and no exercise intervention).	**Duration:** **PA Group:** 8-week **BA Group:** 8-week progressive content with a weekly 120- to 180-minute online video seminar. **Control Group:** Usual treatment and follow-up, duration not specified. **Delivery:** **PA Group:** Participants received an intelligent sports bracelet and an exercise instruction manual detailing the method, intensity, frequency, precautions, and overall intervention schedule. **BA Group:** Participants attended weekly online video seminars in groups of 6 to 10, following an 8-week progressive content inspired by modified BA treatment. **Control Group:** Received usual treatment and follow-up, including creating a comfortable living environment, disease-related knowledge, training of self-care skills, and routine nursing psychological care by clinical nurses.	Quality of life was evaluated by the Functional Assessment of Cancer Therapy Scale–General (FACT-G). The Social Support Rating Scale (SSRS) The General Self-Efficacy Scale (GSES)

		nasopharyngeal or colorectal (13) **Mean years since treatment completed:** Not reported **BA intervention n=48** **Mean age:** 29.21 **Sex:** 35 female, 13 male **Ethnicity:** none reported **Sociodemographics:** *Education:* elementary school or lower (8), middle school (18), high school 11), junior college or university (11), master and above (0) *Marital status:* single (9), married (38), divorced/widowed (1) *Monthly income:* <500 (6), 500-1000 (13),			

		1000-3000 (15), 3000-5000 (8), >5000 (6) **Cancer type**: Lung (16), gynaecological (12), breast (12), others* nasopharyngeal or colorectal (8) **Mean years since treatment completed:** Not reported **Control (n=48):** **Mean age:** 31.21 **Sex:** 36 female, 12 male **Ethnicity:** None reported **Sociodemographics:** *Education:* elementary school or lower (13), middle school (19), high school (8), junior college or university (7), master and above (1) *Marital status:* single (5), married (42), divorced/widowed (1)			

		Monthly income: <500 (11), 500-1000 (15), 1000-3000 (16), 3000-5000 (5), >5000 **Cancer type:** Lung (17), gynaecological (10), breast (14), others* nasopharyngeal or colorectal (7) **Mean years since treatment completed:** Not reported			
Psihogios 2021 RCT *10.1093/abm/kaab008*	Philadelphia, USA	323 participants were contacted, 253 enrolled, and 226 completed baseline measures. Two withdraw shortly after and left 224 for randomisation. 110 were randomised to receive	**Inclusion:** (a) English-speaking AYA between the ages 15 and 29, (b) completed curative treatment for cancer at a paediatric cancer centre, and now receiving follow-up care at mid-Atlantic children's hospital or	**AYA STEP (self-management via Texting, Education, and Plans for Survivorship)** **Intervention Type:** AYA STEPS: Disease Self-Management App **Duration:** 16 weeks **Delivery:**	Quality of Life (QOL) was assessed using the PROMIS Profiles (Pediatric and Adult Versions—

		the intervention and 114 to the control group. **Total n=110:** **Mean age:** 20.51 **Sex:** 47 female, 63 male **Ethnicity:** 'Racial or ethnic minority' 33 **Sociodemographics:** None reported **Cancer type:** Leukaemia/lymphoma 57, solid tumour 43, brain tumour 10, had relapse or second cancer, 23 **Mean years since treatment completed:** 92/110 were >2 years off therapy	geographically adjacent adult hospital, and (c) cognitively capable to complete study procedures (as determined by the medical team and/or medical chart review).	• Participants use the multifunctional AYA STEPS app for disease self-management. • The app stores an electronic Survivorship Care Plan (SCP) containing personalized information about cancer treatment history, risks for late effects, and health promotion resources. • Two-way, tailored mobile messages are provided to reinforce and enhance the uptake of SCP recommendations. • Additional features of the app include a step counter, nutrition journal, medication list, and notifications about upcoming medical appointments. • Features and messages were developed iteratively with input from experts and AYA stakeholders. • Participants receive 1–2 tailored mobile messages per day for the 16-week duration of the intervention.	Profiles 29 and 25) profiles),
Rabin 2016 RCT *10.1089/jayao.2011.0040*	USA	A total of 119 cancer survivors were screened for the study: 60 were ineligible, 21 were eligible but not	**Inclusion:** currently between 18 and 39 years of age; diagnosed with cancer between 18 and 39 years of age; diagnosed with cancer in	**Intervention Type:** • RENEW (Reach out to ENhancE Wellness): Behavioural Intervention incorporating Physical Activity and Mindfulness Meditation	**Profile of Mood States (POMS).**

		interested, and 38 were eligible and interested. The most common reasons for ineligibility were medical or psychiatry contraindication (22%) and not sedentary (25%). Of the 38 eligible survivors, three signed the consent form but withdrew prior to randomization. Thus, 35 YACS were enrolled and randomized to the intervention (n = 19) or control (n = 16) groups. **Intervention group n=19:** **Mean age:** 33.3 **Sex:** 15 female, 4 male **Ethnicity:** 2 Hispanic/Latino, 17 not, White 15, non-White 4	the past 10 years; completed all cancer treatment (with the possible exception of hormone treatment); in cancer remission; able to read, write, and speak fluently in English; not regularly participating in a form of relaxation (e.g., yoga); and sedentary	**Duration:** • **Active Intervention Period:** 12 weeks of weekly calls, followed by 3 months of monthly calls • **Control Group Intervention:** Initiated after the 12-week assessment, identical to the active intervention but without monthly booster calls **Delivery:** • Participants received guidance on gradually increasing moderate-intensity aerobic activity to reach a goal of 30 minutes per day on at least 5 days a week. • Provided with a pedometer to track planned physical activity (PA). • Given an introduction to mindfulness meditation, including a mindfulness CD with sitting meditation, body scan, and yoga stretches. • Weekly calls for 12 weeks with a Personal Developmental (PD) coach to review progress, address barriers, and set goals. • Behavioural coaching tailored to participants' readiness for lifestyle changes.	

		Sociodemographics: *Relationship:* Married or living with partner 10, single/separated/divorced 9. *Employment:* FT/PT 14, unemployed 5 **Cancer type:** ? **Mean years since treatment completed:** not reported **Control n=16:** **Mean age:** 33.9 **Sex:** 14 female, 2 male **Ethnicity:** 3 Hispanic/Latino, 13 not. White 11, non-White 5 **Sociodemographics:** *Relationship:* Married or living with partner 8, single/separated/divorced 8. *Employment:* FT/PT 11, unemployed 5 **Cancer type: ?**		• Participants had access to an online forum for peer support. • Control group received the same intervention as the active group but without the three-monthly booster calls after the 12-week assessment.	

		Mean years since treatment completed: Not reported			
Rosenberg 2021 RCT (2-year trajectories) *10.1001/jamanetworkopen.2021.36039*	USA	100 AYA enrolled at baseline and randomised to intervention group or control group (TAU). 1 was ineligible and excluded and 7 discontinued prior to baseline surveys = 92 for analysis (44 TAU, 48 intervention). **Intervention n=48:** **Mean age:** 17, 35 aged 12-17, 13 aged 18-25 at enrolment. **Sex:** 16 female, 32 male **Ethnicity:** 6 were Asian, 2 Black or African American, 5 'other', 4 mixed race, 34	**Inclusion:** (1) between ages 13 and 25 years; (2) fluent in spoken and written English, including as a second language; (3) diagnosed with either new or progressive malignant neoplasm treated with systemic chemotherapy; and (4) deemed by clinical staff and/or caregivers to be cognitively able to participate in the intervention. All participants were treated at a large quaternary children's hospital (Seattle Children's Hospital).	**PRISM (Promoting Resilience in Stress Management)** **Intervention Type:** PRISM (Promoting Resilience In Stress Management): Individualized Stress Management Programme **Duration:** Four sessions delivered approximately every other week Each session lasted between 20 and 50 minutes **Delivery:** • Trained, bachelors-level nonclinical coaches administered PRISM. • Each AYA in the intervention group received 1-on-1 sessions covering stress management, SMART goal setting, positive reframing, and benefit finding. • Sessions took place during inpatient hospital stays or before/after outpatient clinic visits.	**Pediatric Quality of Life (PedsQL)** **The hope scale** **Connor Davidson Resilience Scale (CDRISC-10)** **Kessler-6 psychological distress scale**

		White, 5 Hispanic or Latino. **Sociodemographics:** None reported **Cancer type:** 31 had Leukaemia/lymphoma, 3 had CNS, 14 had non-CNS solid tumours **Mean years since treatment completed:** Not reported **Control n=44:** **Mean age:** 16, 32 aged 12-17, 12 aged 18-25 at enrolment. **Sex:** 24 female, 20 male **Ethnicity:** 3 were Asian, 0 Black or African American, 12 'other', 4 mixed race, 25 White, 17 Hispanic or Latino. **Sociodemographics:** None reported		• Coaches received at least 8 hours of standardized training, including roleplaying and mock sessions. • Participants received worksheets between sessions to further develop skills. **Control Group:** • Usual psychosocial care provided by an assigned social worker. • Comprehensive psychosocial assessments were conducted upon initiation of care. • Ad hoc support included financial, housing, and concrete supportive care for families, as well as intermittent mental health support for AYAs.	

		Cancer type: 29 had Leukaemia/lymphoma, 4 had CNS, 11 had non-CNS solid tumours **Mean years since treatment completed:** Not reported			
Sansom-Daly 2021 RCT (three-arm, phase two) *10.3390/cancera13102460*	Australia	148 AYA were approached, 88 were lost to follow up, 11 did not opt in 49 took part in an intake interview, 2 were ineligible, 4 had relapse, 3 others opted in but were not randomised. 40 were randomised, 19 to Recapture Life, 10 to active control, 11 to waitlist.	Not reported	**Intervention Type:** 1. Recapture Life Intervention: Cognitive-Behavioural Therapy (CBT) Online Programme 2. Non-Directive, Peer-Support Group Control (Active Control) 3. Waitlist Control **Duration:** - Recapture Life Intervention: Six once-weekly 90-minute small-group sessions - Non-Directive, Peer-Support Group Control: Matched Recapture Life in terms of frequency and contact type, with a focus	Quality of life. Five subscales from the Impact of Cancer Scale AYA module assessed positive and negative impacts of cancer. Psychological Outcomes Depression and anxiety

		Intervention n=19 **Mean age:** 19.4 **Sex:** 11 female, 8 male **Ethnicity:** Aboriginal, 1 **Sociodemographics:** *Education:* year 10 or below (4), year 12 (10), apprenticeship (2), TAFE or certificate/diploma, college (1), university degree (1) *Employment:* FT/PT (8), unemployed student (7), unemployed (3) **Cancer type:** Cancer type; Blood 10, solid tumour 9, brain 0. **Mean years since treatment completed:** Not reported **Active control n=10** **Mean age:** 22.5		on different cancer-survivorship themes each week • Waitlist Control: Waitlisted for six weeks, completing baseline assessments before and after the waitlist period, then re-randomized to receive either Recapture Life or peer-support group. **Delivery:** • Recapture Life Intervention: Facilitated by a psychologist through online videoconferencing with 3–5 AYAs per group. Participants received a Recapture Life workbook and home-practice activities. • Non-Directive, Peer-Support Group Control: Focus on non-directive, supportive group discussion on cancer-survivorship themes with the same frequency and type of contact as Recapture Life. • Waitlist Control: Completed baseline assessments, waitlisted for six weeks, completed repeat baseline assessments, and then re-randomized to receive either Recapture Life or peer-support group.	symptoms. Included the Depression, Anxiety and Stress Scale-Short Form depression and anxiety subscales. Psychological Mechanisms Identity changes. Used the Centrality of Events Scale-Short Form, Unmet cancer-related needs. Used a subset of 17 items from the Cancer Needs Questionnaire for

		Sex: 5 female, 5 male Ethnicity: Aboriginal, 0 **Sociodemographics:** *Education:* year 10 or below (0), year 12 (5), apprenticeship (1), TAFE or certificate/diploma, college (1), university degree (3) *Employment:* FT/PT (7), unemployed student (1), unemployed (2) **Cancer type:** Blood 4, solid tumour 4, brain 2. **Mean years since treatment completed:** Not reported **Waitlist control n=11:** **Mean age:** 20.9 **Sex:** 5 female, 6 male **Ethnicity:** Aboriginal, 0			Parents/Carers of Adolescents and Young Adults with Cancer to gauge support person unmet needs over time. Coping Strategies Positive and negative coping approaches. The KIDCOPE (Older version) measured positive and negative adolescent coping approaches. Both AYAs and support person participants

		Sociodemographics: *Education:* year 10 or below (2), year 12 (3), apprenticeship (0), TAFE or certificate/diploma, college (2), university degree (4) *Employment:* FT/PT (6), unemployed student (2), unemployed (3) **Cancer type:** Blood 6, solid tumour 4, brain 1. **Mean years since treatment completed:** Not reported			completed three subscales of the McMaster Family Assessment Device.

Notes.

QoL measures: The Pediatric Quality of Life Inventory (The PedsQL 4.0) (Upton *et al.*, 2005)

Depression measures: Patient Health Questionnaire-9 item (PHQ-9) (Kurt Kroenke, Spitzer and Williams, 2001), The Centre for Epidemiological Studies Depression Scale for Children (CES-DC) (Shahid *et al.*, 2012), Children's Depression Inventory (Saylor *et al.*, 1984), BA for Depression Scale – Short Form (BADS-SF) (Manos, Kanter and Luo, 2011), 20-item Centre for Epidemiologic Studies Depression Scale (Santor, 2004), and Depression, Anxiety, and Stress Scale-Short Form depression and anxiety subscales (Shahid *et al.*, 2012).

Anxiety measures: 7-item generalized anxiety disorder scale (GAD-7) (Spitzer *et al.*, 2006), State–Trait Anxiety Inventory (STAI) (Marteau and Bekker, 1992), Revised Children's Manifest Anxiety Scale (RCMAS) (Reynolds and Paget, 1983).

Stress measures: 20-item Posttraumatic Stress Disorder Reaction Index (Frederick, 1985), Kessler-6 psychological distress scale (Andrews et al., 2002), Perceived Stress Scale (PSS-10) (Cohen, Kamarck and Mermelstein, 1983).

Mood scales: Profile of Mood States (POMS) (Curran, Andrykowski and Studts, 1995) and Positive and Negative Affect Schedule (PANAS-X) (Watson and Clark, 1999).

Behaviour measures: Child Behaviour Checklist (Nolan et al., 1996), Health Promoting Lifestyle II (Walker et al., 1987).

Self-efficacy measures: The Self-Efficacy Questionnaire for Children (SEQ-C) (Muris, 2001), The General Self-efficacy Scale (GSES) (Schwarzer and Jerusalem, 1995).

Coping measures: The KIDCOPE (Spirito, 1996).

Support measures: 21-item 2-way Social Support Scale (2-Way SSS), (Shakespeare-Finch and Obst, 2011).

Misc. measures: Adult Trait Hope Scale (Snyder, 2002), RAND Medical Outcome Study 36-Item Short Form Health Survey (SF-36) (Hays, Sherbourne and Mazel, 1993), Functional Assessment of Cancer Therapy-General (FACT-G) (Yost *et al.*, 2013), Rosenberg Self-Esteem Scale (RSES) (Rosenberg, 1989), Self-Perception Profile for children and adolescents (Muris, Meesters and Fijen, 2003), The Life Satisfaction Questionnaire (LiSat-9) (Melin, Fugl-Meyer and Fugl-Meyer, 2003), 14-item Mental Health Continuum-Short Form (the MHC-SF) (Keyes *et al.*, 2008), Patient Reported Outcomes Measurement Information System (PROMIS) (Kroenke *et al.*, 2019), 1-item PROMIS Numeric Rating Scale (Kroenke *et al.*, 2019), The 9-item Balanced Measure of Psychological Needs (Sheldon and Hilpert, 2012), The 20-item Self-Report Altruism Scale (Philippe Rushton, Chrisjohn and Cynthia Fekken, 1981), Mishel Uncertainty in Illness Scale–Community (MUIS-C) (Mullins *et al.*, 1997), Posttraumatic Growth Inventory (PTGI) and the Growth Through Uncertainty Scale (Richard G. Tedeschi and Calhoun, 1996), Impact of Events Scale—Revised (IES-R) (Creamer, Bell and Failla, 2003), Multidimensional Health Locus of Control (HLC)(Norman and Bennett, 1996),The Pittsburgh Sleep Quality Index (PSQI) (Carole, 1999), Functional Assessment of Cancer Therapy Scale–General (FACT-G) (Wilailak *et al.*, 2011), Connor Davidson Resilience Scale (CDRISC-10) (Burns and Anstey, 2010), Impact of Cancer Scale (Zebrack *et al.*, 2006), Centrality of Events Scale-Short Form (Clauss *et al.*, 2021), Cancer Needs Questionnaire for Parents/Carers of Adolescents and Young Adults with Cancer (Carey *et al.*, 2012), McMaster Family Assessment Device (Epstein, Baldwin and Bishop, 1983).

Appendix B5: Effect size table

	Outcome Measures	Effect size or Mean (SD/CI) for Intervention vs Control for post-intervention and longest follow up time period	
Arpaci 2023	The Pediatric Quality of Life Inventory (The PedsQL 4.0)	Friedman test experimental = F χ2 30.66, P 0.000 Friedman test control = F χ2 2.92, P 0.404 Wilcoxon test experimental difference between baseline and 10 weeks postintervention = Z −3.42, P 0.001 Wilcoxon test experimental difference between baseline and 3 months after intervention completion = Z −3.68, P 0.000 Wilcoxon test experimental difference between <1 month after intervention completion and 3 months after intervention completion = Z −3.64, P 0.000 Mann-Whitney U test baseline = Z −0.110, P 0.912 Mann-Whitney U test 10 weeks postintervention = Z −0.408, P 0.684 Mann-Whitney U test <1 month after intervention completion = Z −0.790, P 0.429 Mann-Whitney U test 3 months after intervention completion = Z −2.404, P 0.016	
	The Self-Efficacy Questionnaire for Children (SEQ-C)	Friedman test experimental = Fχ2 11.14, P 0.011 Friedman test control = Fχ2 10.76, P 0.013 Wilcoxon test experimental difference between <1 month after intervention completion and 3 months after intervention completion = Z −3.368, P 0.001 Wilcoxon test control difference between <1 month after intervention completion and 3 months after intervention completion = Z −3.188, P 0.001 Mann-Whitney U test baseline = Z −0.544, P 0.587 Mann-Whitney U test 10 weeks postintervention = Z −0.382, P 0.702 Mann-Whitney U test <1 month after intervention completion = Z −0.314, P 0.753 Mann-Whitney U test 3 months after intervention completion = Z −0.756, P 0.450	
	The KIDCOPE	Friedman test experimental = Fχ2 11.56, P 0.009 Friedman test control = Fχ2 2.620, P 0.454 Wilcoxon test experimental difference between baseline and 3 months after intervention completion = Z −3.074, P .002 Wilcoxon test experimental difference between <1 month after intervention completion and 3 months after intervention completion = Z −3.151, P 0.002 Mann-Whitney U test baseline = Z −1.275, P 0.202 Mann-Whitney U test 10 weeks postintervention = Z −0.181, P 0.857 Mann-Whitney U test <1 month after intervention completion = Z −0.994, P 0.320 Mann-Whitney U test 3 months after intervention completion = Z −2.745, P 0.006	
Berg 2020	Adult Trait Hope Scale	POST-INTERVENTION AT 8 WEEKS Hope = 0.69 (13.46) vs − 2.65 (7.40), P 0.902 Agency = 0.00 (6.97) vs − 1.00 (3.71), P 0.584 Pathways = 0.70 (7.03) vs − 1.65 (4.11), P 0.212	FOLLOW UP AT 6 MONTHS Hope = 3.73 (9.88) vs − 1.18 (7.88), P 0.083 Agency = 1.61 (5.04) vs − 0.24 (3.88), P 0.195 Pathways = 2.12 (5.45) vs − 0.94 (4.49), P 0.052
	RAND Medical Outcome Study 36-Item Short Form Health Survey (SF-36)	POST-INTERVENTION AT 8 WEEKS Physical functioning = − 1.21 (23.05) vs 5.88 (14.17), P 0.253 Role limitations due to physical health = 28.79 (15.14) vs 22.79 (20.37), P 0.245 Role limitations due to emotional problems = 0.00 (41.67) vs − 3.92 (30.92), P 0.734 Energy/fatigue = 3.64 (17.60) vs 0.88 (17.76), P 0.564 Emotional well-being = 1.33 (13.40) vs 2.59 (13.34), P 0.755 Social functioning = 28.79 (15.14) vs 22.79 (20.37), P 0.245 Pain = 43.26 (24.94) vs 45.74 (23.89), P 0.737 General health = 0.61 (10.21) vs 2.94 (16.62), P 0.527 Health change = − 2.27 (23.69) vs 0.00 (21.65), P 0.742	FOLLOW UP AT 6 MONTHS Physical functioning = 3.79 (20.69) vs 6.18 (11.80), P 0.662 Role limitations due to physical health = 31.44 (20.99) vs 30.88 (25.43), P 0.934 Role limitations due to emotional problems = 5.05 (43.40) vs 0.00 (44.10), P 0.700 Energy/fatigue = 2.73 (4.41) vs 17.59 (19.68), P 0.759 Emotional well-being = 2.67 (12.95) vs 4.71 (14.44), P 0.614 Social functioning = 31.44 (20.99) vs 30.88 (25.43), P 0.934 Pain = 43.41 (23.28) vs 41.62 (18.18), P 0.783 General health = 7.12 (16.30) vs − 3.53 (17.66), P 0.038 Health change = 6.06 (30.64) vs 1.47 (25.72), P 0.390
	Functional Assessment of Cancer Therapy-General (FACT-G)	POST-INTERVENTION AT 8 WEEKS Physical well-being = 1.24 (4.04) vs 1.41 (4.03), P 0.889 Social/family well-being = 0.71 (3.35) vs − 2.15 (5.87), P 0.078 Emotional well-being = − 0.64 (2.93) vs − 0.47 (4.08), P 0.869 Functional well-being = 0.91 (4.20) vs − 0.41 (3.50), P 0.272	FOLLOW UP AT 6 MONTHS Physical well-being = 1.79 (3.92) vs 0.88 (4.46), P 0.464 Social/family well-being = 0.38 (4.18) vs 1.00 (5.76), P 0.719 Emotional well-being = − 0.03 (3.40) vs − 0.41 (3.52), P 0.712 Functional well-being = 0.76 (4.74) vs 0.76 (4.53), P 0.996
	Patient Health Questionnaire-9 item (PHQ-9)	POST-INTERVENTION AT 8 WEEKS Depressive symptoms = − 1.15 (3.24) vs − 0.88 (4.36), P 0.061 Days of alcohol use past 30 days = 5.92 (22.36) vs 1.17 (3.00), P 0.375 Days of marijuana use past 30 days = 0.11 (0.367) vs − 0.33 (0.97), P 0.053	FOLLOW UP AT 6 MONTHS Depressive symptoms = − 0.58 (3.98) vs − 1.18 (4.22), P 0.623 Days of alcohol use past 30 days = 13.00 (33.98) vs 6.44 (23.26), P 0.463 Days of marijuana use past 30 days = 0.34 (3.48) vs 0.29 (1.26), P 0.293
Cheung 2019	Center for Epidemiological Studies Depression Scale for Children	Time effect = F 12.879, P 0.000, Eta squared 0.311, Power 0.996 Intervention effect = F 4.659, P 0.035, Eta squared 0.074, Power 0.565 Interaction effect (time and intervention) = F 26.854, P 0.000, Eta squared 0.485, Power 1.00	

	(CES-DC)		
	Rosenberg Self-Esteem Scale (RSES)	Time effect = F 11.129, P 0.000, Eta squared 0.281, Power 0.989 Intervention effect = F 6.162, P 0.016, Eta squared 0.096, Power 0.685 Interaction effect (time and intervention) = F 14.298, P 0.000, Eta squared 0.334, Power 0.989	
	Pediatric Quality of Life Inventory 4.0 Generic Core Scale	Time effect = F 28.745, P 0.000, Eta squared 0.502, Power 1.00 Intervention effect = F 4.036, P 0.049, Eta squared 0.065, Power 0.506 Interaction effect (time and intervention) = F 49.663, P 0.000, Eta squared 0.635, Power 1.00	
van Dijk-Lokkart 2016	Dutch PedsQL	POST-INTERVENTION Generic self-report = 2.0 (-4.6; 8.6) Cancer self-report = -1.1 (-5.0; 2.8) Multidimensional fatigue self-report = -1.2 (-7.2; 4.7)	FOLLOW UP AT 12 MONTHS Generic self-report = -2.3 (-9.0; 4.2) Cancer self-report = -1.2 (-6.9; 4.5) Multidimensional fatigue self-report = -1.6 (-9.6; 6.4) Parents in the intervention group reported a significant improvement in levels on the Pain and Hurt subscale indicating less pain, in contrast to the parents in the control group, both shortly after the intervention (β 13.4; 95% CI: 3.0; 23.8) and at long-term follow-up (β 13.0; 95% CI: 1.6; 24.4). At short-term follow-up parents in the intervention group reported significantly less perception of Procedural Anxiety in their children than parents in the control group (β 12.6; 95% CI: 1.9; 23.3). However, this effect did not persist into long-term follow-up. At long-term follow-up, parents in the control group reported significantly more improvement in scores on the Nausea subscale indicating less nausea (β -8.8; 95% CI: -16.3; -1.4), compared with parents in the intervention group.
	Child Behaviour Checklist	At baseline 17.2% of the parents reported clinical significant total problem behavior, 28.1% clinical significant internalizing behavior and 5.9% clinical significant externalizing behavior. The children aged ≥11 years reported less clinical significant problem behavior: respectively, 2.6%, 12.8%, and 0% on total problem behavior, internalizing behavior and externalizing behavior.	
	Children's Depression Inventory	Of all participants 6.8% reported depressive symptoms in the clinical range. After the intervention period, at both measurement points (corrected for baseline scores), no significant differences were found in behavioral problems and depressive symptoms between the two groups, as reported by both the parents and the children.	
	Self Perception Profile for children and adolescents	On most domains of the Self-Perception Profile, 7–15% of the total study group scored below average, with an exception of 32.2% of the participants rating their athletic competence below average. Both at short-term and long-term follow-up (after correction for baseline scores), no differences were found in improvement on all aspects of self-perception between the intervention and control group.	
Grenawalt 2023	The Life Satisfaction Questionnaire (LiSat-9)	There was a statistically significant interaction between the intervention and time on life satisfaction, F(1, 125) 4.793, P 0.03, partial n2 0.037. There was a statistically significant effect of time on life satisfaction for the experimental group, F(1, 63) 6.143, P 0.016, partial n2 0.089.	
	Perceived Stress Scale (PSS-10)	There was no statistically significant interaction between the intervention and time on perceived stress, F(1, 125) 0.039, P 0.843, partial n2 0.000. The main effect of time did not show a statistically significant difference in mean perceived stress at the pre- and post-test time points, F(1, 125) 2.783, P 0.098, partial n2 0.022. The main effect of condition showed that there was not a statistically significant difference in mean perceived stress between intervention groups F(1, 125) 0.792, P 0.375, partial n2 0.006.	
	Behavioral Activation for Depression Scale – Short Form (BADS-SF)	There was no statistically significant interaction between the intervention and time on activation, F(1, 125) 0.001, P 0.974, partial n2 0.000. The main effect of time did not show a statistically significant difference in mean activation at the pre- and post-test time points, F(1, 125) 0.685, P 0.409, partial n2 0.006. The main effect of condition showed that there was a statistically significant difference in mean activation between intervention groups F(1, 125) 5.53, P 0.02, partial n2 0.044. There was not a statistically significant effect of time on life satisfaction for the control group, F(1, 62) 0.190, P 0.665, partial n2 0.003.	
Haydon 2022	14-item Mental Health Continuum-Short Form (the MHC-SF)	POST-INTERVENTION Wellbeing Total PH vs. Control = 2.70 1.46 .065 -0.17, 5.57 Wellbeing EPH vs. Control = 1.99 1.59 .21 -1.13, 5.11	FOLLOW UP AT 1 MONTH Wellbeing Total PH vs. Control = 0.25 1.54 .87 -2.78, 3.27 Wellbeing EPH vs. Control = 1.37 1.69 .42 -1.94, 4.69
	20-item Center for Epidemiologic Studies Depression Scale	POST-INTERVENTION Depressive symptoms PH vs. Control = -1.26 1.52 .41 -4.23, 1.72 Depressive symptoms EPH vs. Control = -0.34 1.40 .81 -3.09, 2.41	FOLLOW UP AT 1 MONTH Depressive symptoms PH vs. Control = -0.33 1.59 .84 -3.43, 2.78 Depressive symptoms PH vs. Control = -0.68 1.43 .64 -3.47, 2.12
	7-item generalized anxiety disorder scale (GAD-7)	POST-INTERVENTION PH vs. Control = -0.27 0.71 .71 -1.66, 1.13 EPH vs. Control = -0.01 0.74 .99 -1.46, 1.44	FOLLOW UP AT 1 MONTH PH vs. Control = -0.63 0.74 .40 -2.08, 0.83 EPH vs. Control = -0.54 0.76 .48 -2.04, 0.95

	Patient Reported Outcomes Measurement Information System (PROMIS)	POST-INTERVENTION Cognitive function PH vs. Control = -1.31 1.39 .35 -4.04, 1.41 Cognitive function EPH vs. Control = 0.36 1.45 .81 -2.48, 3.20 Sleep disturbance PH vs. Control = 0.16 1.50 .92 -2.78, 3.10 Sleep disturbance EPH vs. Control = -0.27 1.44 .85 -3.10, 2.56 Fatigue PH vs. Control = 0.58 1.49 .70 -2.34, 3.51 Fatigue EPH vs. Control = 1.37 1.64 .41 -1.86, 4.59 Pain intensity PH vs. Control = -0.08 0.25 .74 -0.58, 0.41 Pain intensity EPH vs. Control = -0.32 0.25 .21 -0.81, 0.18 Pain inference PH vs. Control = -0.46 1.10 .68 -2.63, 1.70 Pain inference EPH vs. Control = -0.81 1.12 .47 -3.00, 1.38	FOLLOW UP AT 1 MONTH Cognitive function PH vs. Control = -2.16 1.45 .14 -5.01, 0.69 Cognitive function EPH vs. Control = 0.21 1.49 .89 -2.70, 3.13 Sleep disturbance PH vs. Control = 1.09 1.53 .48 -1.91, 4.09 Sleep disturbance EPH vs. Control = 0.35 1.46 .81 -2.51, 3.22 Fatigue PH vs. Control = 2.67 1.52 .080 -0.32, 5.66 Fatigue EPH vs. Control = 2.06 1.70 .23 -1.28, 5.39 Pain intensity PH vs. Control = 0.00 0.27 .99 -0.52, 0.53 Pain intensity EPH vs. Control = -0.32 0.27 .22 -0.84, 0.20 Pain inference PH vs. Control = 0.19 1.12 .86 -2.00. 2.39 Pain inference EPH vs. Control = -0.99 1.15 .39 -3.24, 1.26
	21-item 2-way Social Support Scale (2-Way SSS)	POST-INTERVENTION PH vs. Control = 3.47 1.77 .050* -0.00, 6.94 EPH vs. Control = 2.64 1.49 .077 -0.29, 5.56	FOLLOW UP AT 1 MONTH PH vs. Control = 1.04 1.85 .057 -2.58, 4.66 EPH vs. Control = 1.29 1.61 .42 -1.87, 4.45
	Positive and Negative Affect Schedule (PANAS-X)	POST-INTERVENTION Positive affect = PH 33.21 (8.56) EPH 33.95 (8.62) control 32.83 (8.27) Negative affect = PH 21.46 (8.37) EPH 18.86 (8.21) control 20.38 (9.43)	FOLLOW UP AT 1 MONTH Positive affect = PH 32.47 (8.41) EPH 34.58 (8.39) control 34.38 (7.71) Negative affect = PH 21.24 (9.07) EPH 17.63 (6.75) control 18.83 (8.57)
	9-item Balanced Measure of Psychological Needs	POST-INTERVENTION Relatedness = PH 4.27 (0.86) EPH 4.29 (0.79) control 4.16 (0.86) Competence = PH 3.64 (1.08) EPH 3.76 (0.97) control 3.64 (1.02) Autonomy = PH 4.08 (0.98) EPH 4.19 (0.83) control 3.84 (0.93)	FOLLOW UP AT 1 MONTH Relatedness = PH 4.16 (0.89) EPH 4.33 (0.84) control 4.39 (0.82) Competence = PH 3.57 (1.12) EPH 3.94 (0.96) control 3.80 (1.02) Autonomy = PH 3.92 (1.12) EPH 4.28 (0.80) control 4.06 (0.71)
	20-item Self-Report Altruism Scale	Not reported	
Howell 2018	Pediatric Quality of Life Inventory (PedsQL)	Baseline to 24-week follow-up intervention = 74.19 (16.02) 3.83 2.63 0.01 24 75.32 (17.98) Baseline to 24-week follow-up intervention = 1.71 0.70 0.49 Comparison of change = P 0.44	
Judge Santacroce 2010	Mishel Uncertainty in Illness Scale–Community (MUIS-C) 20-item StateTrait Anxiety Inventory–State subscale (STAI-S) 20-item Posttraumatic Stress Disorder Reaction Index (RI) Posttraumatic Growth Inventory (PTGI) Growth Through Uncertainty Scale (GTUS) Health Promoting Lifestyle II (HPLP II)	ADOLESCENT–YOUNG ADULT CORRELATION MATRIX MUIS-C vs STAI-S = 0.36, P 0.12 MUIS-C vs RI = 0.46, P 0.04 MUIS-C vs HPLP II = −0.23, P 0.33 MUIS-C vs GTUS = −0.02, P 0.93 MUIS-C vs PTGI = −0.27, P 0.26 STAI-S vs RI = 0.68, P < 0.001 STAI-S vs HPLP II = −0.56, P 0.01 STAI-S vs GTUS = −0.47, P 0.04 STAI-S vs PTGI = −0.48, P 0.03 RI vs HPLP II = −0.31, P 0.19 RI vs GTUS = −0.28, P 0.24 RI vs PTGI = −0.46, P 0.04 HPLP II vs GTUS = 0.74, P < 0.001 HPLP II vs PTGI = 0.67, P < 0.001 GTUS vs PTGI = 0.51, P 0.02	
Kazak 2004	Impact of Events Scale—Revised (IES-R)	BASELINE TO FOLLOW UP AT 4-6 MONTHS Intrusion = t(140) 1.32, P 0.19 Avoidance = t(145) -0.66, P 0.51 Arousal = t(143) 2.77, P 0.01	
	Post-Traumatic Stress Disorder Reaction Index (PTSD-RI)	BASELINE TO FOLLOW UP AT 4-6 MONTHS Total = t(143) 1.21, P 0.23	
	State–Trait Anxiety Inventory (STAI)	No group differences were evident for the teens (treatment Mdifference 2.38, SD 9.60, vs. control Mdifference 1.57, SD 9.10), t(110) 0.46, p .65)	

	Revised Children's Manifest Anxiety Scale (RCMAS)		
Kunin-Batson 2016	State Trait Anxiety Inventory (STAI)	State = F(1,36) 1.36, P 0.26, η2 0.04 Trait = F(1,36) 0.72, P 0.40, η2 0.02	
	Multidimensional Health Locus of Control (HLC)	Internal = F(1,36) 0.18, P 0.68, η2 0.005 Chance = F(1,36) 0.38, P 0.54, η2 0.01 Powerful = F(1,36) 0.04, P 0.84, η2 0.001	
Li 2022	Pittsburgh Sleep Quality Index (PSQI)	POST-INTERVENTION AT 1 WEEK Control vs physical activity group = 0.955 (0.094, 1.816), P 0.030 Control vs behavior activity group = 0.583 (−0.274, 1.440), P 0.181 Physical activity group vs behavior activity group = −0.372 (−1.233, 0.489), P 0.394	FOLLOW UP AT 3 MONTHS Control vs physical activity group = 1.020 (0.166, 1.873), P 0.020 Control vs behavior activity group = 0.836 (−0.013, 1.685), P 0.054 Physical activity group vs behavior activity group = −0.184 (−1.037, 0.670), P 0.671
	Functional Assessment of Cancer Therapy Scale–General (FACT-G)	POST-INTERVENTION AT 1 WEEK Control vs physical activity group = −11.791 (−16.187, −7.395), P 0.000 Control vs behavior activity group = −10.025 (−14.599, −5.451), P 0.000 Physical activity group vs behavior activity group = 1.766 (−2.782, 6.314), P 0.443	FOLLOW UP AT 3 MONTHS Control vs physical activity group = −11.791 (−14.658, −7.695), P 0.000 Control vs behavior activity group = −10.897 (−14.609, −7.184), P 0.000 Physical activity group vs behavior activity group = 0.280 (−3.392, 3.951), P 0.880
	The Social Support Rating Scale (SSRS)	POST-INTERVENTION AT 1 WEEK Control vs physical activity group = −7.431 (−9.467, −5.216), P 0.000 Control vs behavior activity group = −7.316 (−9.520, −5.112), P 0.000 Physical activity group vs behavior activity group = 0.115 (−2.100. 2.331), P 0.918	FOLLOW UP AT 3 MONTHS Control vs physical activity group = −6.631 (−9.146, −4.116), P 0.000 Control vs behavior activity group = −6.336 (−8.838, −3.835), P 0.000 Physical activity group vs behavior activity group = 0.295 (−2.220, 2.810), P 0.880
	The General Self-efficacy Scale (GSES)	POST-INTERVENTION AT 1 WEEK Control vs physical activity group = 0.108 (−0.112, 0.329), P 0.334 Control vs behavior activity group = 0.351 (0.132, 0.571), P 0.002 Physical activity group vs behavior activity group = 0.243 (0.023, 0.464), P 0.031	FOLLOW UP AT 3 MONTHS Control vs physical activity group = 0.271 (0.098, 0.444), P 0.002 Control vs behavior activity group = 0.275 (0.103, 0.447), P 0.002 Physical activity group vs behavior activity group = 0.004 (−0.169, 0.176), P 0.966
Psihogios 2021	PROMIS Profiles (Pediatric and Adult Versions—Profiles 29 and 25)	Physical function = α's 0.82–0.89 Anxiety = α's 0.91–0.93 Depression = α's 0.88–0.95 Fatigue = α's 0.91–92 Pain Interference = α's 0.86–0.97	
	26 multiple-choice items Health Knowledge Questionnaire	Total number of correct responses out of 26 = α 0.63	
Rabin 2016	Profile of Mood States (POMS)	POMS total score = intervention 44.4 (28.5) vs control 46.8 (37.4), F(1, 33) = 0.05, P 0.83	
Rosenberg 2021	Pediatric Quality of Life (PedsQL)	PRISM was associated with an improvement from baseline to 6 months (β 9.1; 95% CI, 2.8 to 15.4; P 0.01) and 12 months (β 7.4; 95% CI, 0.8 to 14; P 0.03) but not at 2 years (β 5.9; 95% CI, −1.1 to 12.9; P 0.10)	
	The hope scale	PRISM-participants reported significant improvements between baseline and 6-month (β 3.5; 95% CI, 0.3 to 6.8; P 0.04) and their scores remained higher than baseline at 12 months (β 6.2; 95% CI, 2.7 to 9.6; P < 0.001), and at 2 years (β 4.6; 95% CI, 1.0 to 8.3; P 0.01)	
	Connor Davidson Resilience Scale (CDRISC-10)	PRISM-participants did not report significantly more improvement from baseline at 6 months (β 1.8; 95% CI, −0.4 to 4.0; P 0.10). Their scores were not statistically significantly higher than usual care–recipient improvements from baseline at 12 months (β 1.8, 95% CI −0.5-4, P 0.12) or 24 months (β 1.2, 95% CI −1.2 to 3.6, P 0.34)	
	Kessler-6 psychological distress scale	There were no differences between groups with respect to sustained differences in scores from baseline to 6 months (β −0.7; 95% CI, −2.7 to 1.4; P 0.54), 12 months (β 0.4; 95% CI, −1.8 to 2.6; P 0.72), or 24 months (β 0.1; 95% CI, −2.2 to 2.4; P 0.94)	
Sansom-Daly 2021	5 subscales from the Impact of Cancer Scale AYA module assessed positive and negative impacts of cancer	POST-INTERVENTION AT 6 WEEKS Not reported	FOLLOW UP AT 12 MONTHS High perceived benefit = peer-support 7 (64.0) vs recapture life 9 (75.0) Low burden = peer-support 10 (91.0) vs recapture life 10 (83.3)
	Depression, Anxiety and Stress Scale - 21 Items (DASS-21)	POST-INTERVENTION AT 6 WEEKS Depression = peer-support 4.4 (−0.7, +9.5) vs recapture life 4.0 (0.7, 7.4) Anxiety = peer-support 4.3 (0.3, 8.2) vs recapture life 5.3 (2.6, 8.0)	FOLLOW UP AT 12 MONTHS Depression = peer-support 1.7 (−3.5, 6.8 vs recapture life 5.8 (1.8, 9.7) Anxiety = peer-support 2.8 (−1.3, 7.0) vs recapture life 5.4 (2.2, 8.6)

	Centrality of Events Scale-Short Form	POST-INTERVENTION AT 6 WEEKS Centrality of events = peer-support 26.09 (23.41, 28.78) vs recapture life 26.28 (24.44, 28.12) Survivor label = peer-support 6.4 (5.0, 7.9) vs recapture life 7.5 (6.6, 8.4)	FOLLOW UP AT 12 MONTHS Centrality of events = peer-support 27.99 (25.22, 30.77) vs recapture life 27.48 (25.37, 29.60) Survivor label = peer-support 6.9 (5.4, 8.3) vs recapture life 8.0 (7.0, 9.0)
	17 items from the Cancer Needs Questionnaire for Parents/Carers of Adolescents and Young Adults with Cancer	POST-INTERVENTION AT 6 WEEKS Number of high/very high needs = peer-support 0.6 (0.1, 4.8) vs recapture life 2.4 (0.6, 7.0)	FOLLOW UP AT 12 MONTHS Number of high/very high needs = peer-support 0.7 (0.1, 4.0) vs recapture life 1.0 (0.2, 3.7)
	Positive and negative coping approaches	POST-INTERVENTION AT 6 WEEKS CBT skills confidence = peer-support 8.8 (7.1 9.5) vs recapture life 9.7 (9.0, 9.9) CBT skills actual use = peer-support 7.2 (5.5, 8.4) vs recapture life 9.3 (8.7, 9.7)	FOLLOW UP AT 12 MONTHS CBT skills confidence = peer-support 8.6 (6.8, 9.5) vs recapture life 9.7 (8.9, 9.9) CBT skills actual use = peer-support 8.0 (6.4, 9.0) vs recapture life 9.1 (8.2, 9.6)
	KIDCOPE	POST-INTERVENTION AT 6 WEEKS Strategies used = peer-support 5.8 (4.6, 6.6) vs recapture life 6.3 (5.7, 6.8) Strategies that helped = peer-support 5.0 (3.9, 6.0) vs recapture life 5.7 (5.0, 6.3)	FOLLOW UP AT 12 MONTHS Strategies used = peer-support 4.5 (3.2, 5.6) vs recapture life 6.0 (5.1, 6.7) Strategies that helped = peer-support 4.1 (2.9, 5.3) vs recapture life 5.2 (4.3, 5.9)
	McMaster Family Assessment Device	POST-INTERVENTION AT 6 WEEKS General functioning = peer-support 1.81 (1.50, 2.12) vs recapture life 1.97 (1.76, 2.19) Communication = peer-support 2.32 (2.04, 2.60) vs recapture life 2.24 (2.05, 2.43) 2 Problem-solving = peer-support 2.26 (1.95, 2.58) vs recapture life 2.25 (2.03, 2.48)	FOLLOW UP AT 12 MONTHS General functioning = peer-support 1.92 (1.60, 2.23) vs recapture life 2.00 (1.77, 2.24) Communication = peer-support 2.15 (1.87, 2.42) vs recapture life 2.19 (1.98, 2.40) Problem-solving = peer-support 2.31 (1.96, 2.66) vs recapture life 2.07 (1.81, 2.34)

APPENDIX B6: STUDY OUTCOME MEASURES

5. Study outcome measures.xlsx

Appendix C

This appendix includes all appendices related to chapter six of this thesis.

Appendix C1: Additional illustrative quotes from the content analysis

Psychosocial support provided	
Had therapy	I felt like I'd lost control of my life and I felt that that therapy was like: oh, okay, yeah, you can, you can start living again. *(P23, U, H)* I was just in – in counselling for six years *(P14, B, NH*, focus group)*
Think therapy beneficial	when I go to therapy and we speak about, you know, me, me and what I'm going through, it all relates back to one thing and it's my childhood and how because you didn't feel good enough, you always try to prove yourself and be better than the rest so that by you being better and the average they would look at that and they wouldn't look at how you're different. *(P23, U, H)*
Received specific Rb psychosocial support	I went to this kids' club about two times, and, erm, there was one boy who kept making comments about my eye and my visual impairment and stuff. And I think my – my parents were always very aware of – aware of it, and so I came home and I said to them, "Oh look, this boy's not been very nice to me about it," and they did actually give me – they – they took me to this counsellor and, you know, they got me to draw what happened, and sort of process what was going on, erm, and after that, I was absolutely fine. *(P31, U, NH).*
Wishes for interventions	
Therapy/counselling	I think that for me the – the kind of like one on – one on one, speaking to someone, like I said, that would maybe have some understanding, erm, of – of it as a – as an illness is really helpful. I always kind of shied away from the peer support stuff. *(P25, U, NH)* I think they should ask only like the person if they want it like privately, because, erm, there are maybe some parents out there, they'd be like, "Oh, you don't need that, you're fine." Like they should just ask the – erm, the child themselves what they think. *(P29, U, NH)*

Talking about feelings	I've still got like loads of check-ups and stuff, erm, but erm, yeah, every time they ask me how I'm doing, like, if anything's wrong, like even at the opticians they ask me. Erm, so that's, that's really good *(P13, U, H, focus group)* I think that might help, because then – just maybe talking to someone would help. *(P29, U, NH)* for me, I think it probably would be self-confidence, but that's pretty much – and my mental health, like that – that's the only things it's really impacted in a major way anyway. I feel like having support to do with them would be good. *(P19, U, NH)* It's just me, it's me, it's within myself, no, noth…, no sweet words, no having, erm, a wonderful family that loves you and supports you, no having good friends, I do have that but that would mean nothing if I didn't do the work inside *(P32, B, NH*)*
Interact with peers with similar history	I guess maybe just talking to people of a similar age who'd obviously been through similar experiences. Obviously, you're never gonna find someone with exactly the same experience, but, erm, a similar experience. *(P26, B, H)* at that time, meeting people would have been more beneficial for me. Obviously, children that are going through RB right now are involved in the digital era more and they have more knowledge of technology and everything, so a mix would certainly be more beneficial. I believe that meeting people face to face could also help these children, because like the interaction helps them socialise with the public as well. So, even something like growing up, going to interviews and everything, it surely affects them for sure, and having that interaction when they were young will surely, you know, build a stronger character to be able to talk about these problems in the future, for sure. *(P17, U, H)* Something I'm also seeing a lot recently is the Childhood Eye Cancer Trust, their Facebook page and Instagram page always providing a lot of information and like experience from other people, so that definitely gives more, you know, recognition about the subject. I also have done an article for them a couple of years back. So, even seeing their posts pop up on Facebook and Instagram, it makes me feel like a sort of, you know, acknowledgement in a way. So, definitely, having these experiences shared with other people would definitely have played a better role in my life, for sure. *(P17, U, H)*

	if they offered that sort of stuff in the, erm, talk sessions, maybe that would be good, erm, to meet other people who've had the same experiences. I would say it helped me, because you listen to these people, erm, and they've been through the same stuff as you. You listen to them talk and you realise you're not as alone as you thought, basically. *(P29, U, NH)*
Support to deal with:	
• Physical appearance	I think, after the initial period of – of me feeling ashamed, which was when I was just starting to – erm, you know, to hit puberty, which is when I had my fringe and I felt really insecure and really – I think it – it would have felt really bad then…. I felt kind of like it was ugly or disgusting, like it was a – it was a kind of gross thing. It wasn't just like a – you know when – I've always said this to people, like when I first tell people about it, I tell them that I had cancer. I don't say that my eye is prosthetic, because I feel like, when you start by telling somebody that you have cancer, you have their sympathy (laughter), which is really kind of twisted. *(P25, U, NH)*
• Bullying	'cos I've been through a lot, I've been through like the bullying, erm, the self-depreciation, the intrusive thoughts, the anxiety, like I just feel kind of numb now, like nothing really affects me that much. *(P29, U, NH)* my dad wanted me to move schools anyway because I came home one day and umm, I was like, "Oh, what's a Cyclops?" And they were like, "Oh, it's a mythical creature with one eye." And I was like, "Oh, that's what this boy calls me at school." *(P30, U, NH)*
• Mental health	there's something about looking at the individual and wondering about, I suppose it's yeah, just focusing on the not biological stuff, hopefully with future individuals that stuff all gets taken care of but it's like I, I believe that you know in the face of trauma, in the face of difficulty that you can not only heal but then thrive. And the constant 'thriving' of your life is what protects you or umm, you know, keeps up your immunity against umm, you know, negative things. *(P23, U, NH)* But as I hit 16 and I was acting, and I felt confident in who I was, and I felt, you know, confident that I had good friends that I loved and that would support me and back me up, and I had a boyfriend and he didn't care, you know, that – all of those parts of my life kind of I guess, erm, confirmed in me that it was okay, and so I stopped feeling worried about it and I started finding it to be something that I was able to talk about. *(P25, U, NH)*

• Sex and relationships	But my, erm, partner now who I live with, I didn’t tell him until about a year into our relationship ‘cos I was so worried like about how he’d react, and he didn’t give a crap, do you know what I mean? But I think it was just that worry, like, “Oh god.” *(P28, U, NH)*
• Second cancers	I used to worry sometimes when I was a bit younger that – am – am I at a higher risk of having cancer again, ‘cos obviously I’ve had it once before. I don’t know whether that – to be honest, I still don’t know whether that’s true, whether – whether I’m at less of a risk or more of a risk, or just the same as everyone else, so that’s sometimes a worry. Like I do panic about my health a lot. *(P19, U, NH)* it does kind of pop into your mind every now and then. Like I got some headaches whilst I was at uni and that was like, “Must go to the doctor.” So yeah, and then when you tell them your history, they’re like, “Oh god.” So yeah, and then like you go for a scan, and everything’s fine, luckily it was, but yeah, it does kind of, I don’t know, make you – not paranoid in case it’s – but every now and then, you think, “Oh dear” *(P26, B, H)*
• Having children	I think the more pressing worry, especially I’ve sort of been with my partner for a number of years now, umm, and although we’re not planning to have any children sort of in the immediate future, that is something that kind of being raised in a family I think oh that might be something that I want to do later on. Umm, and that worries me more. Umm, I think there’s almost this feeling of like, err, placing a bet because it is, it is 50/50 whether that gene’s inherited and that does weigh on my mind, it weighs on my mind about obviously that child but also the impact that that might have on my partner. *(P23, U, H)*
• Preparing for the future	it’s really strange that they – like you just went from being in hospital, like completely cold, just to having to live. Like you – there definitely should have been, erm, like what you were saying, skills for independence. *(P1, U, NH, focus group)* I think then it prepares for the future if that makes sense rather than just seek out the information when like you get curious or you need it. *(P18, U, U*)*

	as for worries about the future, erm, for me, it's less about job worries, erm, because frankly I don't know what I want to become in the future, so I'm just leaving the door open to be like, whatever happens bloody happens. But for me, what worries me are like very small little things, like, I don't know, how am I gonna be able to catch a bus, for example. *(P6, B, NH*, focus group)*
• Understanding Rb	if I should be worried about it for the future and whatnot, and like nobody's told me or I've got any like information about stuff so I sort of need to seek it out myself. *(P18, U, U*)* For some reason, they told my parents the information that they had to know and they didn't tell me *(P4, U, H, focus group)*
• Living with visual impairment	I'm right-handed, with a right eye that can't see, so then it was difficult. And being taught the way that a normal person would do it didn't work, because I couldn't see. So, it was things like that that were then frustrating for me, but I wouldn't – I didn't like to be not a winner*(P24, U, NH)* it usually comes up where someone will go to hand me something and they'll go, "Did you not see me?" or something. And I'll say, "Ah well, maybe not, you're on my blind side." *(P30, U, NH)* *I was trying squash the other day with my mate, erm, and we were playing squash, and he was like, erm, winning quite badly, and I – I was there – erm, he said, "Is it anything to do with your eye?" And I said, "I'm not sure, it could be." And then he tried playing with one eye and he couldn't hit absolutely anything. (P4, U, H, focus group)*
• Learning how to communicate with others about Rb	I was so sensitive as a kid, so if like a teacher – like they would put me – sit me somewhere in a class, and then they would like say in front of everyone, "Are you alright there? Can you like see properly there?" And I'd be like, "Oh god, don't draw attention to it" (laughter). "Oh yes, I can see fine." So, they were good, like I think they were probably doing what they thought was best, but, you know, it doesn't – it didn't – it didn't like make me feel good or like they actually were, I don't know, being more sensitive to it, if that makes sense...and I was like, "Oh god, yeah, I can see fine" (laughter). You wouldn't say, "No, I can't," and then have a whole big thing where you have to like move about and stuff. *(P28, U, NH)*
• Health conditions related to Rb (e.g. dry eye)	I get, erm, quite often like these infections and have to have chloramphenicol drops in my socket. *(P1, U, NH, focus group)*

	so firstly, because both eyes are enucleated, what I have here are prosthetics, erm, they get infected so goddamn easily, it's not even funny. Erm, and then it's just incredibly irritating when it happens. *(P6, B, NH*, focus group)*
Barriers to support	
Others have it worse	a personal preference, in, erm, the magazine, there were – I can remember there being, erm, like things written in there saying, you know, that there's a group for young people that are missing eyes or lost eyes and, you know, they can all come and chat, and I just really didn't – that sounded like my worst nightmare (laughter), so - erm, partially because I didn't want to deal with that on any other day other than my appointment day if I didn't have to, and partially because I – again, my situation could have been 100 times worse, and I can remember meeting children that had glass eyes or, you know, lost their eye and, you know, to me, my situation wasn't as bad as theirs*(P24, U, NH)* I'd say sometimes it's like a certain sense of like guilt that I had, 'cos I'd be like, "Oh, even though I'm feeling bad…" Erm, I wouldn't say I was – I was depressed as such. Maybe I was, I don't know. Maybe I am, I don't know. But, erm, it's not – I – I always thought like there was a certain sense of guilt, like, oh, there's people out there who have it a lot worse and, I don't know, I'm just – I'm just moping about here. *(P29, U, NH)* sometimes there'd be like the CHECT meet-ups as well. Obvious- - like most of the other – I say children, we were all children at the time, but they'd often have had it worse as well, and, I don't know, you just feel a bit – not bad, but you just feel like, "Oh no, I don't feel…" Not as though you shouldn't be there, but you don't feel as though you've been impacted as heavily, so you don't feel as kind of… I don't know, not worthy, but, yeah, you just think, oh, I've had it pretty lucky, so I don't – I don't know if I should – yeah, should be able to, er, kind of – when you share experiences, obviously, you don't feel as though you've had it as bad, so you feel a bit – not guilty, but I guess a little bit. I don't know. It's hard, yeah. *(P26, B, H)*
Not sure what is available	I think there are services around, and I think obviously a lot of it has come online now as well due to the pandemic, so I think it is slightly more accessible. *(P26, B, H)*

	They exist out in the wild, but they don't do – like if you don't go actively hunting them down and basically run after them, you're never gonna know about them, which is – you know, personally speaking, what is the point in a goddamn organisation for the blind and visually impaired if they don't make themselves known. *(P6, B, NH*, focus group)* like when you get to be an adult, you know, like if I need someone to speak to, like I could go to like a therapist. Like I could seek that resource. But when you're a kid, you don't know what's available, do you? You just have no idea. *(P28, U, NH)* I didn't go searching for a lot, to be fair, so I don't know whether there was much out there. Erm, but yeah, I think sometimes – yeah, especially when you get to that age as well, you – you think, "Oh well, I'm not – I can't really go and talk to like certain child services anymore because you're not really a child." But then also at the same time, yeah, you don't really feel grown up (laughter). So yeah, it was – was a bit of a – yeah, kind of at a limbo kind of time. *(P26, B, H)*
Stigma	"when I was growing up, like mental health was unheard of, you know. It's now that everyone is speaking about mental health, but we still are a very, very, very long way away from actually, you know, discussing mental health. So, possibly that has been the reason why I've never found the use for talking to anyone, you know." (*(P17, U, H)* "psychologists and therapists were not looked upon as in oh, you're going to the psychologist because you have an issue and you're, you're sad or you're, I wish I had that support, erm, because my parents, they didn't have the tools to and they didn't even know like there's a lot of things that my parents don't know" *(P32, B, NH*)* My university offers like a, erm, counselling thing, so I just – I mean, they're just a – I can just call them up if I wanted. But I usually go through the stages of like, "Oh, I'm feeling really bad now, I'd better call them up." Then like I'd be feeling better, like, "I'll be fine." Then I feel bad again, yeah. *(P29, U, NH)*
Practical intervention considerations	
Adolescence is the time of need	You just have no idea, and you kind of feel like there is like – I guess when you're a teenager, you do just feel like everything's the end of the world, and you could talk to people but they actually probably wouldn't understand, or they would give you solutions for things that weren't the solution that you wanted, do you see what I mean? *(P28, U, NH)*

	I think when, probably younger end of teenage years for me, probably would have been nice to have someone to talk *(P30, U, NH)* horrible, it was literally the worst time of my life, erm, teenage years that I would not go back to that (*P32, B, NH*)* Like I think this is probably the best I've felt about it like ever, the older I get, like, you know, completely superficial-wise. But no to like the…lead up to secondary [school]. (P28, U, NH)
Integrated with existing appointments	obviously it's much harder to kind of give a resource that's available to everyone in person all the time, that's really difficult to kind of orchestrate like for, not only for, you know, appointment-wise but also like financially not everyone has the means to kind of travel for an appointment like that. *(P30, U, NH)* I guess if there was somebody maybe – I think the easiest thing for me would have been if someone was at the appointments that I physically had to go to every year no matter what. I'd be forced there by my mum and dad if I didn't want to go. And then as I got older, I needed to go and I realised that. I guess if there was somebody incorporated into the bits you already had to do, that gave you maybe an option – everything's quite clinical, which it obviously has to be, I get that, erm, but then maybe that would make it slightly easier to open up if you wanted to, I suppose, and have that option. *(P24, U, NH)* I think actually like with, erm, like my six months check-up, they always said like if anything was wrong then they'd like, they'd say like places where I could go if I wanted any help. So I think actually like the hospital itself helped *(P12, U, H, focus group)* support from a younger age about the psychological side. Erm, just like if you had – 'cos I went up to Birmingham from (place) every few months, which is exhausting, to have a – erm, a check-up on, er, my eye. They'd like do the – they'd shine into it, see if the cancer had returned. They'd do the eyedrops to make the eye bigger and everything. Erm, but it was just the medical side of it. I was never really asked about the psychological side, like how it was feeling. I was like maybe if – I was maybe thinking, if you had someone there to talk to you as well as the, erm, psychological side, just like maybe saying, erm, like, I don't know, "I'm – I'm – my name is so and so, I'm here to help you. Erm, how has having one eye affected your school life so far? Like, erm, what do the other kids think of it? What do they say to you?" *(P29 ,U, NH)*

	Other aspects that might have, for example, helped me as well, is possibly like professionals always keeping contact with these people. So, from my experience, the only professional advice I've got was when giving the check-ups and like that's it, you know. So, if there were regular follow-ups, I believe that that would have been more – you know, not knowledge, but like recognition about the situation *(P17, U, H)* 'Cos I know like, erm… Erm, I wouldn't have been someone who would seek that [support] out, erm, so I think, if you integrate it, that would really help. And… I think they should ask only like the person if they want it like privately, because, erm, there are maybe some parents out there, they'd be like, "Oh, you don't need that, you're fine." Like they should just ask the child themselves what they think… *(P29 ,U, NH)*
Intervention delivery:	
• Online	I believe a mix would be a perfect combination, because the digital aspect can be accessed any time, you know. Even for example if there's like a support line or a support chat, for example, for people to talk with professionals, that would surely be a great addition. If you're facing a problem now and you feel like you want to talk to someone, sometimes talking not only to a stranger but someone who's professional in the subject will definitely make you feel better for sure. *(P17, U, H)* obviously it's much harder to kind of give a resource that's available to everyone in person all the time, that's really difficult to kind of orchestrate like for, not only for, you know, appointment-wise but also like financially not everyone has the means to kind of travel for an appointment like that. *(P30, U, NH)* an online talk service maybe, you know, when you get like those, erm, messaging things, like, erm, when you can just talk to someone online through messaging, 'cos that would – that would help I think – 'cos a lot of people are anxious, they don't want to do it face to face. Like I think it would be helpful to have it like that. *(P29 ,U, NH)* But then the thing with a workshop is, if it's a face to face workshop, erm, the – the – the issue then is – maybe you don't have a time – maybe you don't have time to go to the workshop. You have other stuff. Maybe you can't get to the workshop full stop. Erm, which – in situations like that, I feel like, at least they have a website, not – not a dedicated app or anything, not that extravagant, but even like a dedicated website would

	be a step in the right direction, so, “Hey, if you can’t go to the workshop, you can still get something, you know. You can still get this list of resources.” *(P4, U, H, focus group)*
• In-person	I think there’s something about the person-to-person that’s vitally important, but I think it could be good this idea of like I had eye check-ups right, but I never had like mind check-ups or heart check-ups right, so there’s something about that. And I think definitely the in-person I think would be the most powerful because both those counselling and the psychotherapy ones were all in-person and the power of that was so present and kind of really being in somebody else’s room and really being with them and being with their energy and being in their space was so umm, powerful and sort of being held by them. *(P23, U, H)*
• Modular (on-demand)	but just say like at the end, maybe like offer it – offer like a support service, if – if you want to come back, if you’re still feeling like there’s a – a reason to come back, if you want to talk to someone, I think that would be really good. *(P29 ,U, NH)* I think that modular would be a good thing to offer, because then if you have a bad day and you want to, you can, or if you don’t and you don’t want to and you feel fine, then you don’t have to. *(P24, U, NH)* for young adults per se, I think, yeah, whether – say when you’re at school or going to college or university, whether there’s kind of – not a pack, but something like, you know, “During this time, this is what you might experience. Here’s how to navigate it, and here’s who to turn to if you want.” *(P26, B, H)*
• Workshops	I agree with everybody else, workshops – I mean, frankly, you know, it’s – it’s better talking to an actual person and have an actual person guide you, and teach you about stuff, than reading a webpage. It’s more intuitive. *(P6, B, NH*, focus group)* I think this is much better than having a website, because like you – if you have a question now, you can ask that rather than like you having to find someone to email from a website, and then having to wait like a month or something for the – for – for them to respond. *(P3, U, NH, focus group)*

- Videos	Or like a YouTube channel or something like that, where you have like – you have them talking or you have a video of them and stuff like that. *(P3, U, NH, focus group)* it's amazing to see videos recently of multiple young people from mid-teenagers to mid-twenties – and I've seen various videos of people and they're, you know, posing for pictures and videos with the prosthesis out and things like that *(P16, U, NH)* I think having like access to, say, like something on Instagram, say Facebook or TikTok would be quite nice 'cos you could get loads of people and everything that way. But yeah, maybe things online. *(P5, U, NH, focus group)*
The facilitator needs specific Rb knowledge	I guess if there was somebody maybe – I think the easiest thing for me would have been if someone was at the appointments that I physically had to go to every year no matter what. I'd be forced there by my mum and dad if I didn't want to go. And then as I got older, I needed to go and I realised that. I guess if there was somebody incorporated into the bits you already had to do, that gave you maybe an option – everything's quite clinical, which it obviously has to be, I get that, erm, but then maybe that would make it slightly easier to open up if you wanted to, I suppose, and have that option. *(P24, U, NH)* If only I had someone just telling me this, just telling me oh no, you know what, let's sit down and write down what are the things that you were able to do and that you like to do and let's focus on that instead of focusing on what you're not able to do and the rest is able to do and that makes you different. *(P32, U, NH*)* obviously I always knew just to go to the doctor if I had any concerns, erm, sometimes it is good to speak to people that are specialists, because it is quite, you know, a rare condition. And even when you go to the opticians and you say about it, they're like, "I've never heard of that," or, "Oh, I've never seen anyone with that" (laughter), so you go, "Oh great." 'Cos when they do the eye test and there's just nothing there, they're like, "What?" *(P26, B, H)*
Someone who's not your Mum, or your doctor	

	I’m sure I got wrote off when I was either 10, 11 or 12, like before I started hitting that age where people started making really cruel comments and obviously all of them. Erm, but I feel like, if I spoke to someone during that time, that probably would have been better for me, ‘cos I would have been able to ask them everything that was on my mind. *(P19, U, NH)* it sort of feels like there could have been a few things like nipped in the bud like just sort of talking about some of my … so there’s something about err, psychological support in the sense of err, therapy, talking about my experience, talking about how I view the world; talking about how the impact of it on my family has impacted me. *(P23, U, H)*