

**Living with and beyond a childhood brain  
tumour diagnosis:  
The supportive care needs of teenagers and  
young adults and their caregivers**

**Emma Nicklin**

**Submitted in accordance with the requirements for the degree of  
Doctor of Philosophy**

**The University of Leeds  
School of Medicine**

**February 2021**

The candidate confirms that the work submitted is his/her/their own, except where work which has formed part of jointly authored publications has been included. The contribution of the candidate and the other authors to this work has been explicitly indicated below. The candidate confirms that appropriate credit has been given within the thesis where reference has been made to the work of others.

Nicklin E, Velikova G, Hulme C, Rodriguez Lopez R, Glaser A, Kwok-Williams M, et al. 2019. Long-term issues and supportive care needs of adolescent and young adult childhood brain tumour survivors and their caregivers: A systematic review. *Psycho-oncology*. 28 (3):477-87.

*This publication reports on the systematic review described in Chapter 2 of this thesis. I was responsible for all aspects of planning and executing of the review, including: review design, management of searches, data extraction, critical appraisal, analysis and the writing of the manuscript. Florian Boele, Claire Hulme and Galina Velikova supervised the overall systematic review. Florian Boele also screened a random sample of eligible articles (20%). Rocio Rodriguez Lopez helped to guide the literature search. Adam Glaser and Michelle Kwok-Williams helped to guide the focus of the systematic review. All authors provided critical feedback and contributed to the final version of the manuscript.*

Nicklin E, Velikova G, Boele F. 2020. Technology is the future, but who are we leaving behind? *The Lancet Oncology*. 21(1): 29-29

*This commentary piece shares some reflections from the PhD research around technology use in the study population. Chapter 4 references the article and it is included in the appendices. I carried out the descriptive analysis presented in the manuscript and prepared the manuscript for publication. Other authors provided critical feedback and contributed to the final version of the manuscript.*

Nicklin E, Pointon L, Glaser A, Sarwar N, Kwok-Williams M, Debono M, Velikova G and Boele F. 2021. "Unmet support needs in teenage and young adult childhood brain tumour survivors and their caregivers: "It's all the aftermath, and then you're forgotten about". [Manuscript accepted for publication] *Journal of Cancer Survivorship*.

*This publication focuses on the qualitative interview results reported in Chapter 6 of this thesis. I carried out the interview design, topic guides, undertook the interviews and analysis. The supervision team (Florien Boele and Galina Velikova) oversaw all aspects of the qualitative work. Florian Boele also double-coded a number of interviews. Lucy Pointon and Florian Boele wrote the manuscript. Other authors provided critical feedback and contributed to the final version of the manuscript.*

This copy has been supplied on the understanding that it is copyright material and that no quotation from the thesis may be published without proper acknowledgement.

The right of Emma Nicklin to be identified as Author of this work has been asserted by her in accordance with the Copyright, Designs and Patents Act 1988.

© 2021 The University of Leeds and Emma Nicklin

## Acknowledgements

Firstly, I would like to acknowledge and give particular thanks to the participants of this study who gave their time. It was a pleasure to meet you all. I am especially grateful to those who agreed to be interviewed. I am conscious of the courage needed to share personal experiences and discuss some of the most painful times of your lives; your strength and resilience never failed to amaze me.

I am very grateful to Ellie's Fund and Yorkshire Cancer Research for the financial support for this PhD. Ellie's story is heart-breaking but inspiring and was one of the reasons I wanted to do this PhD in the first place.

I would like to give special thanks to my supervisors. A huge thank you to Dr Florian Boele for being consistently supportive from day one of my PhD journey. Your knowledge, advice and enthusiasm has been invaluable. I would like to give particular thanks to Florian for the final year of my PhD, especially the quick and detailed feedback and encouragement during this time. A big thank you to Professor Galina Velikova who has given expert clinical guidance, vital feedback, and support over the past three plus years. I believe I have learnt so much from you in this time. I also wish to thank Claire Hulme, who was my third supervisor in the first year of my PhD, for her support.

I would like to thank all the clinical staff who have been part of this study. Especially Professor Adam Glaser, Dr Michelle Kwok-Williams, Dr Miguel Debono, Naseem Sarwar, Angela Scotland and Emily West. I really appreciate your interest towards the study and all your advice that has undoubtedly improved the study.

I want to acknowledge the support from the Patient Centred Outcomes Research (PCOR) team in Leeds where I have been based. Not only are you the friendliest people I have ever worked with, you have also provided amazing advice, encouragement and most importantly cake(tiffin)! I have been very lucky to have such fantastic colleagues and to have been able to do my PhD in such a supportive environment.

Finally, I want to thank my fabulous family and friends. Especially my parents for supporting me always. And my husband for always believing in me, keeping me (relatively) sane and for backing me to apply for this PhD in the first place – it's not been an easy few years!

I dedicate this PhD to my daughter, Olivia. You were born in such uncertain times, but we've muddled through. You have been my ray of sunshine in the dark. I hope you are proud, and this inspires you to have the courage to do whatever you wish in the future.

## Abstract

The aim of this PhD was to understand the long-term issues and supportive care needs experienced by teenage and young adult (TYA) childhood brain tumour survivors and their caregivers. A systematic review of the literature identified social issues (e.g. isolation) were most reported by survivors, followed by cognitive (e.g. impaired memory) and physical issues (e.g. endocrine dysfunctions). Caregivers reported uncertainty, increased responsibilities, and problems maintaining their own self-well-being and relationships. Few studies in the review addressed unmet supportive care needs.

Following the review findings, a convergent mixed methods study was designed to understand the supportive care needs experienced by TYA childhood brain tumour survivors and their caregivers. A quantitative phase using a cross-sectional survey and a qualitative phase using semi-structured interviews was conducted, and the results of these two phases integrated. Participants included childhood brain tumour survivors, at least five years from diagnosis, currently aged 13-30 and their primary caregivers.

112 participants (69 survivors/43 caregivers) completed the survey and 22 face-to-face semi-structured interviews were conducted (11 survivors/11 caregivers).

The integrated findings indicate that both survivors and caregivers have unmet needs many years after diagnosis. Survivors were faced with wanting to achieve key milestones as they move into young adulthood, but late effects of treatment often made this difficult. Survivors specifically had high unmet needs in relation to their psychological health, social lives (including romantic relationships), employment, and independence. Whilst caregivers had unmet needs regarding their own psychological well-being and survivor financial issues. Survivors further from diagnosis, unemployed survivors and single caregivers were more likely to report unmet needs. Currently there are barriers preventing survivors and caregivers accessing supportive services. This thesis provides leads to improving supportive care and long-term follow-up services. Understanding unmet needs and recognising what services are required is critical to improving survivor and caregiver quality of long-term survival.

## Table of contents

Acknowledgements .....	3
Abstract .....	4
Table of contents .....	5
List of tables .....	10
List of figures .....	12
List of abbreviations .....	13
<b>Chapter 1: Introduction .....</b>	<b>14</b>
1.1 Childhood brain tumours .....	14
1.1.1 Descriptive epidemiology .....	14
1.1.2 Classification of tumours .....	15
1.1.3 Incidence rates .....	16
1.1.4 Treatment .....	18
1.1.5 Survival rates .....	19
1.2 Teenage and young adult survivors .....	20
1.2.1 Definition of teenagers and young adults .....	20
1.2.2 Developments during adolescence .....	21
1.2.3 The involvement of TYAs in research .....	21
1.3 Primary informal caregivers .....	22
1.4 Defining long-term survivorship .....	23
1.4.1 Cancer survivorship .....	23
1.4.2 Long-term survivorship care for TYA childhood brain tumour survivors .....	24
1.4.4 Long-term issues/Late effects .....	25
1.4.5 Supportive care/Supportive care needs .....	25
1.5 Thesis objectives .....	25
<b>Chapter 2: Systematic review .....</b>	<b>28</b>
2.2 Aim .....	28
2.2.1 Rationale and previous systematic reviews .....	28
2.3 Methods .....	29
2.3.1 Search strategy .....	29
2.3.2 Selection criteria .....	29
2.3.3 Data extraction and quality assessment .....	30

2.3.4	Narrative Synthesis.....	31
2.4	Results .....	33
2.4.1	Search Results.....	33
2.4.2	Study characteristics.....	34
2.4.3	Critical appraisal of included studies .....	34
2.4.4	Section 1: Survivor issues .....	38
2.4.5	Section 2: Caregiver Issues.....	44
	Survivor well-being.....	45
2.4.6	Section 3: Supportive care needs .....	46
2.5	Discussion.....	48
2.5.1	Strengths and limitations .....	49
2.5.2	Clinical implications .....	51
2.6	Conclusions and implications for research and practice.....	51
2.7	Planning the mixed methods study .....	52
<b>Chapter 3:</b>	<b>Methodology.....</b>	<b>54</b>
3.1	Mixed methods research methodology .....	54
3.1.1	Definition .....	54
3.1.2	History of mixed methods in health research.....	55
3.2	Rationale for the choice of a mixed methods approach.....	58
3.2.1	Challenges in choosing mixed methods research .....	60
3.3	Philosophical worldview in mixed methods .....	62
3.3.1	A pragmatic approach.....	62
3.4	Mixed methods design.....	64
<b>Chapter 4:</b>	<b>Mixed methods study - design, recruitment and analysis.....</b>	<b>67</b>
4.1	Brief overview of research design .....	67
4.2	Ethics .....	68
4.2.1	Ethical considerations .....	68
4.2.2	Ethical approvals .....	71
4.3	Quantitative data: survey .....	72
4.3.1	Research setting.....	74
4.3.2	Participant Sample.....	75
4.3.3	Recruitment and data collection .....	78
4.3.4	Outcome measure selection.....	80
4.3.5	Data analysis .....	85
4.4	Qualitative data: Semi-structured interviews.....	92

4.4.1	Participant Sample.....	93
4.4.2	Data collection .....	96
4.4.3	Data management .....	97
4.4.4	Data Analysis .....	98
4.4.5	Quality of qualitative research .....	103
4.5	Integration of quantitative and qualitative data .....	104
<b>Chapter 5:</b>	<b>Quantitative Results.....</b>	<b>105</b>
5.1	Survivor recruitment and response rate .....	105
5.2	Survivor participant characteristics.....	106
5.3	Objective 1: Survivor unmet supportive care needs .....	109
5.3.1	Psychological needs .....	112
5.3.2	Physical and Daily Living needs .....	113
5.3.3	Patient Care and Support needs .....	113
5.3.4	Health System and Information needs .....	113
5.2.5	Sexuality needs.....	113
5.4	Objective 2: Survivor unmet supportive care needs and sociodemographic/clinical data.....	114
5.4.1	Total level of unmet needs .....	114
5.4.2	Psychological needs .....	115
5.4.3	Physical and Daily Living needs .....	116
5.4.4	Patient Care and Support needs .....	117
5.4.5	Health system and Information needs .....	118
5.4.6	Sexuality needs.....	119
5.5	Objective 3: Survivor unmet supportive care needs and quality of life .	119
5.6	Objective 4: Survivor service and information use.....	121
5.7	Caregiver recruitment and response rate.....	124
5.8	Caregiver participant characteristics .....	125
5.9	Objective 1: Caregiver unmet supportive care needs .....	127
5.9.1	Psychological and emotional needs .....	131
5.9.2	Information needs .....	131
5.9.3	Health Care Service needs.....	131
5.9.4	Work and Social needs .....	132
5.10	Objective 2: Caregiver unmet supportive care needs and sociodemographic/clinical data.....	132
5.10.1	Total level of unmet needs .....	133



5.10.2	Psychological and emotional needs .....	133
5.10.3	Informational needs .....	134
5.10.3	Health Care Service needs .....	135
5.10.4	Work and social needs .....	136
5.11	Objective 3: Caregiver unmet supportive care needs and caregiver QoL .....	136
5.12	Objective 4: Caregiver service and information use .....	138
5.13	Discussion.....	141
5.13.1	Strengths and limitations .....	145
<b>Chapter 6:</b>	<b>Qualitative results.....</b>	<b>148</b>
6.1	Interview recruitment and procedures .....	148
6.2	Participant characteristics.....	149
6.3	Themes .....	152
6.3.1	Theme 1: Preferences for support and services .....	154
6.3.2	Theme 2: Decline in support.....	166
6.3.3	Theme 3: Barriers to not obtaining adequate support .....	170
6.3.4	Theme 4: The role of long-term follow-up care .....	178
6.4	Summary of findings .....	185
6.4.1	Strengths and limitations .....	187
6.4.2	<b>Reflective thinking.....</b>	<b>189</b>
<b>Chapter 7:</b>	<b>Integration.....</b>	<b>192</b>
7.1	Approach to mixed methods integration.....	192
7.2	Mixed methods integration: Joint Display Table .....	194
7.3	Summary of integration.....	204
7.3.1	Strengths and limitations .....	204
<b>Chapter 8:</b>	<b>Discussion .....</b>	<b>206</b>
8.1	Key findings .....	206
8.2	Strengths and weaknesses of the thesis.....	207
8.3	Implications .....	210
8.2.1	Clinical care .....	210
8.2.2	Support services .....	211
8.2.3	Future research.....	212
8.4	Planned future work and dissemination .....	213
8.5	A reflective account: learning experiences during PhD .....	214
8.6	Conclusion .....	216

<b>References.....</b>	<b>217</b>
<b>Appendices.....</b>	<b>231</b>
Appendix 1 – Systematic review search strategy.....	231
Appendix 2 - Study details .....	234
Appendix 3 – Ethical approval letter .....	244
Appendix 4 – Ethical approval (substantial amendment) : online recruitment .....	245
Appendix 5 - Survivor Participant Information Sheet (over 16's).....	246
Appendix 6 – Survivor and caregiver interview guide .....	248
Appendix 7 – Accuracy of self-report vs. medical data (Poster BPOS 2020) .....	253
Appendix 8 – Comparison between online responders and participants recruited in LTFU clinics. ....	254
Appendix 9 – Example of multivariable linear regression .....	255
Appendix 10 – Assumption checks -multivariable linear regression analyses .....	257
Appendix 11 – Dissemination .....	259

## List of tables

<b>Table 1- Age adjusted incidence rates per 100,000 persons, by region, age group and sex.....</b>	<b>17</b>
<b>Table 2 - Critical appraisal scores using the MMAT Tool. See Pluye (2011) for MMAT guidelines and details of screening questions. <sup>81</sup> .....</b>	<b>35</b>
<b>Table 3 - Good Reporting of A Mixed Methods Study (GRAMMS) as proposed by O'Cathain (2008) <sup>147</sup> .....</b>	<b>54</b>
<b>Table 4 - Comparing quantitative and qualitative approaches .....</b>	<b>56</b>
<b>Table 5- Reasons for using mixed methods, Source: Bryman (2006)<sup>167</sup> .....</b>	<b>58</b>
<b>Table 6 - A pragmatic approach, adapted from Morgan (2007)<sup>146</sup> .....</b>	<b>63</b>
<b>Table 7 - Participant inclusion and exclusion criteria.....</b>	<b>77</b>
<b>Table 8 - Percentage of missing survey data by individual questionnaire tools.....</b>	<b>86</b>
<b>Table 9 - Guidelines for interpreting the Pearson correlation coefficient .....</b>	<b>91</b>
<b>Table 10 – Survivor sociodemographic, social and clinical characteristics .....</b>	<b>107</b>
<b>Table 11 - Frequency of survivor unmet needs.....</b>	<b>110</b>
<b>Table 12 - Number of survivors who identified an unmet need, by each item of the SCNS-SF34 by domain. Table also shows the top ten most frequent supportive care needs in survivors.....</b>	<b>111</b>
<b>Table 13 - Univariate regression analysis: associations between total number of unmet needs and clinical/social characteristics.....</b>	<b>115</b>
<b>Table 14 - Multivariable model: total number of unmet needs.....</b>	<b>115</b>
<b>Table 15- Univariate regression analysis: associations between Psychological needs and clinical/social characteristics .....</b>	<b>116</b>
<b>Table 16- Multivariable model: Psychological needs.....</b>	<b>116</b>
<b>Table 17- Univariate regression analysis: associations between Psychical and Daily Living needs and clinical/social characteristics .....</b>	<b>117</b>
<b>Table 18- Univariate regression analysis: associations between Patient Care Support needs and clinical/social characteristics.....</b>	<b>117</b>
<b>Table 19- Multivariable model: Patient Care and Support needs.....</b>	<b>118</b>
<b>Table 20- Univariate regression analysis: associations between Health system and Information needs and clinical/social characteristics .....</b>	<b>118</b>
<b>Table 21 - Multivariable model: Health system and Information needs.....</b>	<b>119</b>
<b>Table 22- Univariate regression analysis: associations between Sexuality needs and social and clinical characteristics .....</b>	<b>119</b>
<b>Table 23 - Quality of life scores in correlation to the total number of unmet needs and individual SCNS-SF34 domains .....</b>	<b>121</b>

<b>Table 24 - Survivor support service use</b> .....	122
<b>Table 25 - Survivor information resource use</b> .....	123
<b>Table 26 - Caregiver social characteristics and survivor clinical characteristics</b> .....	125
<b>Table 27 - Frequency of caregiver unmet needs</b> .....	128
<b>Table 28 - Number of caregivers who identified an unmet need, by each item of the SCNS-P&amp;C by domain. Table also shows the top ten most prevalent supportive care needs in caregivers</b> .....	129
<b>Table 29 – Univariate regression analysis: associations between total number of unmet needs and caregiver characteristics/survivor clinical factors</b> .....	133
<b>Table 30- Univariate regression analysis: associations between Psychological and emotional needs and caregiver characteristics/survivor clinical factors</b> .....	134
<b>Table 31 - Multivariable model: psychological needs</b> .....	134
<b>Table 32 – Univariate regression analysis: associations between Informational needs and caregiver characteristics/survivor clinical factors</b> .....	135
<b>Table 33 – Univariate regression analysis: association between Patient care and Support needs and caregiver characteristics/ survivor clinical factors</b> .....	135
<b>Table 34- Univariate regression analysis: associations between Work and Social needs and caregiver characteristics/social clinical factors</b> .....	136
<b>Table 35 - Caregiver quality of life score in correlation to total number of unmet needs</b> .....	137
<b>Table 36- Caregiver support service use</b> .....	138
<b>Table 37- Caregiver information resource use</b> .....	139
<b>Table 38 - Survivor characteristics</b> .....	150
<b>Table 39 - Caregiver characteristics</b> .....	151
<b>Table 40 - Summary of themes and subthemes</b> .....	153
<b>Table 41 - Mixed methods Joint Display Table</b> .....	195

## List of figures

<b>Figure 1 - Narrative synthesis process</b> .....	32
<b>Figure 2- Flow chart showing article selection</b> .....	33
<b>Figure 3 - Adapted Quality of Life Cancer Survivorship model</b> .....	38
<b>Figure 4 - Frequency of articles that reported survivor issues by theoretical framework theme</b> .....	39
<b>Figure 5 - Frequency of articles that reported caregiver issue by theoretical framework theme</b> .....	44
<b>Figure 6- Convergent mixed methods design (adapted from Creswell and Plano Clark, 2011)</b> .....	65
<b>Figure 7 - How unmet need was dichotomised (SCNS-SF34 and SCNS-P&amp;C)</b> .....	87
<b>Figure 8 - Questions to aid developing themes - adapted from Braun and Clark's (2013)</b> .....	102
<b>Figure 9 - A flow diagram to represent survivor response rate</b> .....	106
<b>Figure 10 - Histogram to show total number of unmet needs per survivor</b> .....	109
<b>Figure 11- Histogram to show survivor Quality of Life scores</b> .....	120
<b>Figure 12- A flow diagram to represent caregiver response rate</b> .....	124
<b>Figure 13- Histogram to show total number of unmet needs per caregiver</b> .....	128
<b>Figure 14 - Histogram to show caregiver Quality of life scores</b> .....	137
<b>Figure 15 - Figure to show the steps taken to build a mixed methods joint display table</b> .....	194

## **List of abbreviations**

BSFC-S - Burden Scale for Family Caregivers – short version

CQOLC - Caregiver Quality of Life Index–Cancer

IPA - Interpretative phenomenological analysis

JLA – James Lind Alliance

LTFU – Long-term follow-up

MMAT – Mixed Methods Appraisal Tool

NCCS – National Coalition for Cancer Survivorship

NCI – National Cancer Institute

NCRI – National Cancer Research Institute

NHS - National Health Service

QoL – Quality of Life

Peds-FACT-Br – Paediatric Functional Assessment of Cancer Therapy – Brain

PNET - Primitive Neuro-Ectodermal Tumour

PPI - Patient and Public Involvement

SCNS-SF34 - Supportive Care Needs Survey - Short Form 34

SCNS-P&C - The Supportive Care Needs Survey - Partners and Caregivers

SD – Standard deviation

SPSS - Statistical Package for the Social Sciences

TYA – Teenage and young adult

WHO – World Health Organisation

# Chapter 1: Introduction

In this thesis the patient group studied is teenage and young adults (TYAs) who have had a primary childhood brain tumour and their caregivers. This chapter will give a summary of the epidemiological and clinical factors which underpin the research. The Chapter begins with describing childhood brain tumours in detail including information about classification, incidence, treatment and survival rates (1.1). Then, TYA survivors are defined, as well as the developments during adolescence and their involvement in research (1.2). Next, the role of the primary informal caregivers is described (1.3). The key concepts used within this thesis are then explored including cancer survivorship, long-term survivorship care, late effects/long-term issues and supportive care needs (1.4). The chapter concludes by providing a summary of the content that is presented in each chapter of the thesis (1.5).

## 1.1 Childhood brain tumours

### 1.1.1 Descriptive epidemiology

Brain tumours are masses or growths of abnormal cells that occur in a child's brain or the tissue and structures that are near it. There are many types of childhood brain tumours, each type of tumour has its own biology, treatment, and prognosis. The tumours may be benign or malignant. Benign brain tumours generally grow slowly, press on nearby areas of the brain, but rarely spread into other tissues. Benign tumours can be life-threatening because of their space-occupying effects within the cranium, local infiltration, and some become malignant over time.<sup>1</sup> Malignant brain tumours are likely to grow more quickly and spread into other brain tissue and elsewhere in the central nervous system. When a tumour grows into or presses on an area of the brain, it may stop that part of the brain from working the way it should.<sup>1</sup> Both benign and malignant brain tumours can cause signs or symptoms and need treatment. The extent of tumour malignancy is classified by grading the tumour in terms of its aggressiveness. Tumours are assigned World Health Organisation (WHO) grades from I to IV, Grade I indicating tumours that are slow growing and Grade IV representing the fastest growing and most aggressive tumours. In most cases, the exact cause of a paediatric brain tumour is not known.

### 1.1.2 Classification of tumours

In children there are more than 100 histological entities of brain tumours, as defined by the World Health Organisation.<sup>2</sup> The classification of childhood brain tumours is based on histology and location.<sup>2</sup>

The most common types of brain tumours in children are briefly described below.

#### a) Gliomas

Most childhood brain tumours start in glial cells – the supporting cells of the brain. These tumours are known as gliomas.<sup>2</sup> Around half of all brain and spinal cord tumours in children are gliomas.<sup>3</sup> A number of tumours can be considered gliomas, including:

- Astrocytomas (including Pilocytic astrocytomas, Low-grade astrocytoma, Anaplastic astrocytomas and Glioblastomas)
- Oligodendrogliomas
- Ependymomas
- Brain stem gliomas
- Optic gliomas
- Mixed Glioma

Pilocytic and Low-grade (grade I or II) astrocytomas are the most common type in children.<sup>3</sup>

#### b) Embryonal tumours

Another group of tumours arise from embryonal cells, cells that line the passageways in the brain where cerebrospinal fluid is produced and stored. Approximately 10% - 20% of brain tumours in children are embryonal tumours.<sup>3</sup> These tumours are more common in younger children and tend to grow quickly.<sup>2</sup> Medulloblastomas are the most common type of embryonal tumour, these tumours are malignant, grade IV tumours that start in the cerebellum.<sup>2, 4</sup>

Other less common tumours in this group include:

- Embryonal tumours with multi-layered rosettes (previously Primitive Neuro-Ectodermal Tumours i.e. PNET)
- Atypical Teratoid Rhabdoid Tumours (ATRT)
- Neuroblastomas
- Ganglioneuroblastomas
- Medulloepitheliomas

#### c) Craniopharyngiomas



Craniopharyngiomas develop in the pituitary gland (hormone-producing gland) and can extend to other areas of the brain. They account for around 4% of brain tumours in children.<sup>3</sup>

d) Germ cell tumour

Germ cell tumours are fast growing, malignant tumours. These tumours develop from germ cells, which normally form egg cells in women and sperm cells in men. Most germ cell tumours occur outside the brain, but those that do grow in the brain are most often in the area close to the pituitary gland. A germinoma is a type of germ cell tumour that is most commonly found in the brain and usually respond well to treatment.<sup>2</sup> They represent around 3% of all brain tumours.<sup>3</sup>

e) Pineal tumours

Some types of tumours occur in the pineal gland, a small gland in the middle of the brain that makes the hormone melatonin that controls sleep. Pineal gland tumours as a group are rare, accounting for less than 1% of all childhood primary brain tumours.<sup>3</sup> The most common and fastest growing of these are called pineoblastomas.<sup>2</sup>

### 1.1.3 Incidence rates

For children in the UK aged 0 - 14 years, brain tumours are the second most common cancer registrations after leukaemia and the most common solid tumour.<sup>5</sup> In the UK each year approximately 500 children are diagnosed with a brain tumour.<sup>5</sup> Incidence varies by country, with the highest incidence in the United States (Table 1), reporting 5.65 cases per 100,000 persons.<sup>3</sup> Recent US statistics have even highlighted that primary brain and central nervous system tumours are now the most common cancer diagnoses in children.<sup>3</sup> The majority of the incidences are malignant.<sup>3</sup> Additionally, the incident rate is generally higher in males than females.<sup>3, 6-8</sup>

The most commonly specified tumours registered in children aged 0 - 14 year age group are pilocytic astrocytomas and medulloblastomas (as described in 1.1.2).<sup>3, 8</sup> In children the largest percentage of tumours are located in cerebellum and brain stem.<sup>3, 8</sup>

In recent decades, there have been reports of increasing incidence of primary brain tumours. However, these claims need to be interpreted with caution. Improved diagnostic imaging, following the introduction of radio isotope imaging, computed tomography, and magnetic resonance imaging in the 1970s and 1980s, has led to higher detection rates and better

differential diagnosis of brain tumours.<sup>14, 15</sup>

**Table 1- Age adjusted incidence rates per 100,000 persons, by region, age group and sex**

Region	Reference	Years	Child age group	Incidence in males per 100,000	Incidence in females per 100,000	Overall age specific incident rates per 100,000 person
Canada	Walker et al. (2018) <sup>9</sup>	2009-2013	0-14	-	-	3.69
Denmark, Finland, Norway, and Sweden	Schmidt et al (2011) <sup>10</sup>	1985–2006	0-14	-	-	4.20
England	Arora et al (2009) <sup>8</sup>	1995-2003	0-14	3.72	3.39	3.56
France	Desandes et al (2014) <sup>11</sup>	2000-2008	0-14	-	-	3.9
Germany	Kaatsch et al (2001) <sup>12</sup>	1990–1999	0-15	-	-	2.6
Japan	Makino et al (2010) <sup>13</sup>	1989-2008	0-14	-	-	3.61
Korea	Yun-Sik Dho et al (2017) <sup>7</sup>	2013	0-19	5.17	5.03	5.27
United States	Ostrom et al. (2018) <sup>3</sup>	2011-2015	0-14	5.84	5.45	5.65

#### 1.1.4 Treatment

Treatments for childhood cancer have evolved over the past 50 years, with the goal of maximising the proportion of patients who achieve long-term survival, while minimising the adverse effects of therapy.<sup>16 17</sup>

Treatment of young children can be particularly challenging because brain directed treatment can have a serious impact on the child's subsequent development. Treatment choices are based on tumour histology and location, but also on the child's age and the effects therapy may have on the developing brain. The immaturity of the child's brain can make treatment decisions difficult, particularly in infants.

The management of childhood brain tumours often incorporates multimodal approaches involving surgery, irradiation, chemotherapy and medication.<sup>18</sup>

Neurosurgical intervention is the initial treatment modality for the majority of paediatric brain tumours. The main aim being to remove as much abnormal tissue as is safely possible.<sup>19</sup> Neurosurgical procedures carry many intraoperative and post-operative risks. Complications can include infection, blood loss, and neurological morbidity.<sup>19</sup> Furthermore, cerebellar mutism (also known as posterior fossa syndrome) is also a potential complication of posterior fossa surgery. Posterior fossa syndrome can mean that the child can develop mutism, emotional lability, and neurocognitive impairment.<sup>20</sup>

It is not always possible to remove all of the tumour through surgery, in many cases further treatment with radiotherapy or chemotherapy may be needed to treat any abnormal cells left behind.<sup>21</sup> Radiotherapy is generally an important component of the multimodal plan for the management of most brain tumours in children.<sup>22</sup> Radiotherapy uses beams of high energy waves or particle streams to destroy cancerous cells and is typically recommended after surgery or if the tumour is inoperable. The pathology, location of the tumour and age of the child will determine the dosage and type of radiation. Radiotherapy treatment cannot distinguish between cancerous and healthy cells, which often means normal tissue is also affected. The frequency and dosage of treatment varies between patients, but typically radiotherapy will be administered daily for approximately 6 weeks.<sup>23</sup> Common side effects include: fatigue, headaches and skin irritation.

The aim of chemotherapy is to destroy cancerous cells using chemical agents.

Chemotherapy can be used prior to surgery or in combination with radiotherapy as adjuvant therapy. Again treatment differs between patients - guided by the tumour's pathology and

age of the child, but almost always results in a wide range of side effects (i.e. fatigue, hair loss, nausea).<sup>24</sup>

Finally, Proton Beam Therapy is novel treatment starting to be available to some UK childhood brain tumour patients. Proton Beam Therapy is a specialist form of radiotherapy that enables a dose of high energy protons to be precisely targeted at a tumour, reducing the damage to surrounding healthy tissues and vital organs. It is worth noting that Proton Beam Therapy is not suitable for all types of brain tumour. The side effects are similar to those experienced from other forms of radiotherapy.<sup>25</sup> However, as it is a relatively new therapy there is little evidence about its effectiveness compared to conventional radiotherapy, particularly about long-term side-effects.

### **1.1.5 Survival rates**

Children generally have better survival outcomes than those diagnosed with a brain tumour at an older age.<sup>1, 3</sup> Over the past several decades, advances in neurosurgery, radiotherapy and chemotherapy, have meant that the survival of children treated for brain tumours has improved significantly.<sup>17, 26-29</sup> However, survival rates vary greatly between countries, tumour types and locations. Recent statistics report that the five-year survival rates for all childhood brain tumours (aged 0-14 years) is 73.3% and ten year survival is 69.9%.<sup>3</sup> Brain tumours with the highest ten-year survival rates include: Pilocytic astrocytoma's (95.8%), Oligodendroglioma's (88.6%) and Diffuse astrocytoma's (81.1%). Brain tumours with the lowest ten-year survival rates include: glioblastomas (14.8%); ATRT- Atypical Teratoid Rhabdoid Tumour's (28.7%) and - Embryonal tumours with multi-layered rosettes (previously PNET) (49.0%).<sup>3</sup>

Even after successful treatment of the tumour, many survivors experience significant long-term effects. Outcomes not only include physical or medical effects, but also may include social, emotional, behavioural, and neurocognitive domains. Medically, survivors may experience complications including but not exclusive to endocrine, renal, or gastrointestinal functioning. Problems with hearing and vision are also common. Neurocognitive outcomes may include epilepsy/seizures and motor functioning symptoms. More details about long-term issues and outcomes are discussed in the results of chapter two.

## 1.2 Teenage and young adult survivors

Teenage and young adult survivors of childhood brain tumours are an ever-growing population of patients, many of whom remain at lifelong risk for potential late effects as a consequence of their cancer treatment.<sup>30, 31</sup> As new patients continue to be diagnosed with childhood brain tumours and survival continues to improve, these numbers will undoubtedly increase.<sup>32</sup>

Teenagers and young adults (TYAs) are a unique patient cohort with specific challenges and vulnerabilities differing from those of children or older adults.

### 1.2.1 Definition of teenagers and young adults

Various terms, including adolescents, youth, teenagers, young adults and young people, are used to describe people who are neither children nor adults. In this thesis, I will be referring to this group as ‘teenagers and young adults (TYAs)’ or ‘young survivors’. There is great disparity across the oncology literature regarding how the TYA age group are defined.<sup>33, 34</sup> Definitions differ greatly amongst organisations, countries and oncology literature. These discrepancies are understandable given the realities of the complex biological and psychosocial developmental processes experienced in the lengthy transition from childhood to adulthood in Western cultures.

Commonly, the lower age boundary ranges between 10-15 years old.<sup>35, 36</sup> In the United Kingdom, the lower limit for TYA care is generally defined as 13 years of age.<sup>34, 37</sup> The upper age limit classification for young adults remains highly variable. In the United Kingdom, the upper limit is normally defined as 24 years of age.<sup>34, 37</sup> Yet, in the United States the upper age limit is commonly 39 years,<sup>36</sup> this is because cancer is a disease more common in old age, hence they classify cancer patients as ‘young’ when they are below 40.

The notion of *adolescence* was introduced only a century ago, described in 1904 by psychologist G. Stanley Hall as a distinct stage of development that extended dependency beyond childhood and delayed entry into adult roles. Hall called adolescence a time of “*storm and stress*”.<sup>38</sup> Since then adolescence has been firmly recognised as a distinct and important developmental period.<sup>39</sup> This period of change is dramatically characterised by sexual maturity, hormonal activity, rapid physiological development, and complex psychological and emotional change. During this time young people are also acquiring autonomy and building a sense of self. For most young people in Western countries, these developmental years are profound and important, a time of various possibilities in love, work, and worldviews.<sup>39</sup>

## **1.2.2 Developments during adolescence**

There have been several theoretical contributions to the understanding of adolescence. One early contribution and arguably the most known was made by psychologist Erik Erikson. Erikson's Psychosocial Developmental Theory was an enormously influential force during the first half of the twentieth century. According to Erikson, the adolescent mind is fundamentally a pause, a psychosocial stage between childhood and adulthood.<sup>39</sup> Children are becoming more independent, and begin to look at the future in terms of career, relationships, families, housing, etc. Most importantly young people want to belong to a society and fit in.<sup>39</sup>

Adolescence is also a critical period for brain development and the experiences of each adolescent during this time helps to shape their adult brain.<sup>40</sup> Through the period between childhood and adulthood there is a remodelling of the brain that starts often just before the teenage years begin and continues into the mid-twenties. Brain development in adolescence is a mix of expansion and regression. The regression refers to the pruning down of existing neurons focused on processing and communicating information.<sup>40</sup>

Not all brain changes during adolescence are regressive, with some neurons remaining to grow and establish new synaptic connections. The laying down of myelin sheaths connecting the remaining linked neurons continues years after adolescence. Myelin enables the remaining and connected neurons to communicate with each other with more coordination and speed. Although myelination begins early in life and continues into adulthood, its production escalates notably during adolescence, thereby speeding information flow across distant regions and magnifying its impact.<sup>40</sup>

## **1.2.3 The involvement of TYAs in research**

It is critical to consult young people about their experiences. The importance of consulting children and young people is a principle enshrined in the United Nations Convention on the Rights of the Child passed in 1989 and signed by the UK in 1991. Since then there has been a growing commitment from health and social researchers to listen to young peoples' voices and their experiences and needs.<sup>41 42</sup>

Traditional perspectives conceive young people as still developing and not yet able to speak in their own voices.<sup>43</sup> This perspective has led to data being collected either by proxy, from adults close to the young person, or via objective measures where the young person remains passive.<sup>43</sup> This approach neglects the competence of the young person to contribute. In the last twenty years it has been recognised that children and TYAs need to be

given the opportunity to share their feelings and wishes about issues affecting them.<sup>44 45</sup> A clear trend within literature, service provision and legislation recognises young people as ‘active beings’ in all aspects of their lives.<sup>45</sup> Since this time there has been increasing obligation to hear the voices of young people in research that is about them and will affect the lives of others like them in the future.<sup>42</sup>

### **1.3 Primary informal caregivers**

A brain tumour affects not only the survivor but also those around them. Responsibility for caring and supporting childhood brain tumour survivors is often met by their immediate family. This caring role often continues into long-term survivorship. Parents, family members or close friends take on a new role as the informal caregiver, providing daily support. In this thesis the term ‘caregiver’ is used to describe a person who provides regular help and support (mentally and physically) to the brain tumour survivor, on an informal basis. While many caregivers experience this role as rewarding, it also can have its downsides.

In caring for someone with a brain tumour, caregivers face unique challenges posed by the long-term effects of the diagnosis and treatment. Treatment-related outcomes leave many childhood brain tumour survivors with a variety of challenges that may require care and management over the survivor’s lifetime. It is important to understand the demands of caring for these TYA survivors because of the implications for their ongoing care, as well as for the issues caregivers can experience themselves.

Previous research has suggested that caregivers of brain tumour survivors may have: lower quality of life than caregivers of other cancer groups,<sup>46</sup> greater stress and worse parental mental health than parents with children without health problems.<sup>47, 48</sup> Unsurprisingly being a caregiver also means increased burden, and less leisure time.<sup>49</sup> The burden of caregiving has also been linked to lower self-esteem<sup>50</sup> and depressive symptoms.<sup>51</sup> Caregivers of brain tumour survivors can experience high levels of stress related to the unique care demands associated with both cancer and brain injury.

Previous studies in other cancer groups have also found associations between aspects of caregiver wellbeing and patient survival,<sup>52</sup> outcomes and well-being.<sup>53, 54</sup> Poor physical and emotional health in caregivers may have a negative impact on the brain tumour survivor but also may put more burden on the healthcare system in general. Therefore, supporting caregivers to perform their responsibilities, while keeping their own physical and emotional health, is essential.

However, the majority of preceding research has primarily focused on the survivor alone. There is a paucity of research on caregiver health and the ability to cope with long-term caring for a TYA after a childhood brain tumour.<sup>55</sup> When the survivor transitions into adulthood, and tries to find their way to independent living, the role of the informal caregiver changes. Currently, there is little published data about how the issues experienced during this transition period influence caregivers' emotional health, employment status and economic burden.<sup>55</sup>

## **1.4 Defining long-term survivorship**

Traditionally brain tumour research, has focused on increasing overall survival and stopping tumour growth, with less focus on the long-term side effects after treatment. However, survival alone is an inadequate outcome. With improved treatment and duration of survival, more attention is needed to address the late effects of treatment in survivorship.

### **1.4.1 Cancer survivorship**

Since the National Coalition for Cancer Survivorship (NCCS) was founded in 1986, the field of cancer survivorship has become a significant topic within oncology. The terms cancer survivor and cancer survivorship were novel and were defined by the NCCS.<sup>56</sup> The scope of the population included as cancer survivors and those experiencing cancer survivorship has implications for who should be included in cancer survivorship care, and, by extension, who should be targeted for cancer survivorship research. The NCCS definition states that an individual is considered a survivor from the time of treatment onward and that the term survivor includes the individual's community of friends and family, in recognition of the effects that cancer has on them.

In 1996, the National Cancer Institute (NCI) established the Office of Cancer Survivorship dedicated to developing evidence-based research defining the impact of cancer and its treatment, including the long-term effects, with the aim to improve survivors' quality of life and identify their unique needs.<sup>57</sup> Therefore, a survivorship concept focusing on life after treatment slowly infiltrated policy measures and academic fields.

The National Cancer Institute (NCI) definition states:

*"In cancer, survivorship focuses on the health and life of a person with cancer post treatment until the end of life. It covers the physical, psychosocial, and economic issues of cancer, beyond the diagnosis and treatment phases. Survivorship includes issues related to the ability to get health care and follow-up treatment, late effects of treatment, second cancers,*



*and quality of life. Family members, friends, and caregivers are also considered part of the survivorship experience.”*<sup>58</sup>

Subsequent to the development of these groups, and medical reports published in the 2000s<sup>59</sup>, movement to focus on life after cancer has grown worldwide.<sup>60</sup> More recently in England the National Cancer Research Institute (NCRI) with the James Lind Alliance (JLA) have identified 26 top research priorities that will help people live better with and beyond cancer.<sup>61</sup> The JLA brings together patients/survivors, caregivers and clinicians to identify and prioritise the unanswered questions they want health research to address. This is the first time that clear research priorities have been identified in this area.

The term ‘cancer survivorship’ has become a keyword in the field of cancer care. It is now recognised that the time after primary treatment for cancer is a distinct phase in the cancer continuum, a time when individuals make a transition from “*patient*” to “*survivor*”.<sup>56</sup>

#### **1.4.2 Long-term survivorship care for TYA childhood brain tumour survivors**

As highlighted by D'Angio (1975) more than 40 years ago, lifelong survivorship care is needed for the majority of paediatric cancer survivors:

*“It is clear that the child cured of cancer must be followed for life, not so much because late recurrence of disease is feared as to permit early detection of the delayed consequences of radio- and chemotherapy.”* (p.868)<sup>62</sup>

Long-term care is a general term that describes the care that people need over an extended period of time. Despite general agreement on the need of follow-up care, there still is great discrepancy in the delivery of long-term follow-up care for paediatric survivors. Furthermore, due to the unique issues and needs faced by neuro-oncology patients, evidence from survivorship research conducted within other cancer populations may not be valid or meaningful to the survivorship experiences of those with neuro-oncologic diagnoses.

The 2006 National Institute for Health and Clinical Excellence (NICE) guidelines promote that continuous aftercare should meet the needs of brain tumour survivors allowing for age, sex, and cultural differences. The guidelines state:

*“Younger patients with continuing care needs should also be carefully considered ... procedures should be in place to ensure the continuing care needs of younger patients with CNS tumours are appropriately met.”*<sup>63</sup>

However, there is little information in the guidelines on how follow-up care should address and meet young survivors and caregivers supportive care needs.

#### **1.4.4 Long-term issues/Late effects**

Improvements in survival rates are encouraging, yet the quality of extended survival is equally important. Late effects are problems that survivors can develop after cancer treatments have ended. Even after successful treatment of the tumour, many survivors experience significant late effects. The systematic review in Chapter 2 details the late effects/issues TYA survivors' and their caregivers encounter that may impact their overall quality of life (QoL).

#### **1.4.5 Supportive care/Supportive care needs**

Several terms have been used to describe care for patients that is additional to primary treatment, including survivorship care, after care, rehabilitation, follow-up care and supportive care. There are numerous definitions for these terms, but many overlap and the distinction between them is often blurred.<sup>64</sup> Within this thesis, the term "supportive care" has been chosen as an umbrella term for the services that help support survivors and their caregivers.

The working definition of supportive care suggested by NICE is care that:

*"...helps the patient and their family to cope with cancer and treatment of it, from pre-diagnosis, through the process of diagnosis and treatment, to cure, continuing illness or death and into bereavement. It helps the patient to maximise the benefits of treatment and to live as well as possible with the effects of the disease."* (p.18)<sup>22</sup>

Supportive care needs in cancer patients are diverse and may fall into numerous domains, such as: physical, psychological, practical, information and communication. 'Needs' have been defined as having 'the requirement of some action or resource that is necessary, desirable or useful to attain optimal well-being' (Foot, 1996, as cited in Sanson-Fisher, et al., 2000, p.227).<sup>65</sup>

### **1.5 Thesis objectives**

The overall aim of this PhD was to understand the long-term issues and supportive care needs experienced by teenage and young adult (TYA) childhood brain tumour survivors and their caregivers, at least five years after diagnosis.

The rest of this thesis is motivated by this aim, as well as more specific research questions, throughout the following chapters.

A brief summary of the contents of each chapter is provided below.

## **Chapter 2: Systematic review**

This chapter details the systematic review, which aimed to identify and narratively synthesise evidence on the issues experienced and the supportive care needs of TYA childhood brain tumour survivors and their caregivers. The purpose of the systematic review was to identify what has already been investigated and where the gaps in the literature were. Hence, where the need for new research was most needed. The findings of this systematic review were used to focus the aims of the mixed methods study.

## **Chapter 3: Methodology**

This chapter presents the research methodology and specific design used as the framework for the mixed methods study, based on the research aims and objectives. An introduction of different research methodologies and how they underpin health care research is outlined. The choice for a convergent mixed method approach is then discussed in detail.

## **Chapter 4: Mixed methods study – design, recruitment and analysis**

This chapter details the methods chosen for the quantitative and qualitative phases of the mixed methods study. It starts with a description of the process of obtaining ethical approvals followed by the recruitment of patients, sample size calculation, data collection and analysis for both the quantitative and qualitative phases.

## **Chapter 5: Quantitative results - survey**

This chapter presents the results of the quantitative phase. The specific objectives were to: 1) describe the met and unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers; 2) explore if survivor sociodemographic (i.e. sex, age) and clinical data (i.e. tumour type, location and treatment) are related to unmet needs; 3) determine whether unmet needs are associated with Quality of Life (QoL) outcomes and 4) explore the role and perceived use of support services in TYA survivors and their caregivers. Survivor and caregiver data are presented separately in this Chapter. Tables and figures are used to enable a straightforward reading and understanding of the data.

## **Chapter 6: Qualitative results – interviews**

This chapter outlines the findings of the qualitative phase. The specific objectives were to 1) describe the met and unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers; and 2) explore the role and perceived use of support services in TYA survivors and their caregivers. The chapter starts with the description of the

sociodemographic and clinical characteristics of the interviewed participants. It then presents each theme in detail. Anonymised quotes extracted from participant interviews are included to enable a richer understanding of each theme and category.

### **Chapter 7: Integration of quantitative and qualitative data**

This chapter interprets the findings from both the quantitative and qualitative phases. The integration of these two phases together with the findings of the systematic review enables this study to provide an in-depth understanding of the long-term issues and supportive care needs faced by TYA childhood brain tumour survivors and their caregivers.

### **Chapter 8: Discussion**

The final chapter summarises the key findings, study limitations, recommendations and implications for clinical practice/supportive services/future research, planned future work and a reflective account of the PhD experience. The chapter ends with a conclusion.

## Chapter 2: Systematic review

Chapter 1 provided background about TYA childhood brain tumour survivors and their caregivers and aims of this PhD research. This chapter reports a systematic review of the literature relating to the issues and supportive care needs of TYA brain tumour survivors and their caregivers.

The systematic review has been published in *Psycho-oncology* during the PhD - “*Long-term issues and supportive care needs of adolescent and young adult childhood brain tumour survivors and their caregivers: A systematic review.*”<sup>55</sup>

The value of doing systematic reviews to identify areas requiring further health care research is widely established.<sup>66</sup> It was decided that a systematic review was the most appropriate method of evidence synthesis because of the comprehensive, transparent and rigorous stages. Other review types, such as rapid reviews do not always provide a definitive, unbiased, exhaustive review of the evidence base.

The review helped to identify what had already been investigated and where the gaps in the literature were. Hence, where the need for new research was most needed. The findings of this systematic review were used to focus the aims of the mixed methods study.

### 2.2 Aim

The overall aim of the review was to systematically identify and narratively synthesise evidence on the issues experienced and the needs of TYA childhood brain tumour survivors and their caregivers.

The research questions explored were:

- 1) What issues do TYA childhood brain tumour survivors and their caregivers' experience?
- 2) What are the needs of TYA childhood brain tumour survivors and their caregivers?
- 3) Do survivors and caregivers feel their needs are being met?

#### 2.2.1 Rationale and previous systematic reviews

Previous systematic reviews have focused on the experiences of TYA survivors or caregivers with mixed cancer diagnoses.<sup>67, 68</sup> However, as brain tumour patients have unique

and invasive treatment on the brain, it is important that this patient group is studied separately to other childhood cancers. Other reviews have concentrated on the experiences of young children,<sup>69, 70</sup> adults,<sup>71</sup> or caregivers of adult brain tumour survivors.<sup>72</sup> To date, no systematic review has collated evidence on the issues and supportive care needs of TYA childhood brain tumour survivors and/or their caregivers. Given the complex developmental stages and increased frequency of life events during adolescence and young adulthood, it was expected that the experience and impact of a childhood brain tumour on TYA survivors and their caregivers would be unique to their age group.

## **2.3 Methods**

### **2.3.1 Search strategy**

PRISMA guidelines<sup>73</sup> for systematic reviews were utilized as a template for the methodology. The following electronic databases were searched: Medline (Ovid), Embase (Ovid), PsycInfo (Ovid), Pubmed, CINAHL (Ebsco), and the Cochrane library (Wiley). The grey literature were searched using Web of Science and the NHS Evidence. The end date of the searches was September 2017. The searches were developed for the concepts: brain tumours, children and survivorship. Guided by an Information Specialist (Rocio Rodriguez Lopez, University of Leeds), the searches were developed combining subject headings and free text terms for each concept. See appendix 1 for the full search strategy.

### **2.3.2 Selection criteria**

Original, peer-reviewed articles were included according to the following criteria:

- Human participants with a primary diagnosis of a brain tumour and/or a primary caregiver of a survivor. If the study had a mixed cancer participant group, brain tumour data had to be reported separately.
- Survivors of a paediatric brain tumour, currently aged 14-39 or caregivers of survivors within this age range. The study sample could extend beyond these age parameters provided the results for the target age range were clearly reported.
- Articles reporting data that focused on any issues, needs or unmet needs of brain tumour survivors and/or their caregivers.

As Chapter 1 highlighted there are contradictions across the oncology literature regarding how the TYA age group should be defined.<sup>33, 34</sup> Definitions differ greatly amongst

organizations, countries and TYA literature. For this review, the TYA age range was set at 14-39 to capture as many relevant studies as possible.

Exclusion criteria included: articles not in English, reviews, reports on incidence of brain tumours or treatment trials / intervention programmes not covering TYA patient or caregiver issues and needs. There was no restriction on publication date or study type.

Identified articles titles and abstracts were screened against the inclusion criteria, with a random sample (20%) independently screened by a second reviewer (Florien Boele, PhD supervisor). Reasons for excluding studies were recorded. Any discrepancies between reviewers were discussed until an agreement was reached. If unsure at this stage articles were kept in as a precaution, until the next selection stage. Next, full-text articles of potentially relevant abstracts were further assessed for inclusion, again a random sample (20%) was reviewed independently by the second reviewer.

### **2.3.3 Data extraction and quality assessment**

Data extraction was performed using a standardized template. Extracted data included:

- a) Study details: study objective, design, setting, sample size, outcome measures, primary findings, and conclusions.
- b) Survivors' characteristics: age at diagnosis of brain tumour, age at study enrolment, brain tumour type/location and treatment details (time since treatment completion and types of treatments received).
- c) Caregivers' characteristics (if applicable): age, sex and relationship to the survivor.

All included studies were assessed using the Mixed Methods Appraisal Tool (MMAT) – version 11.<sup>74</sup> The tool was chosen as it is one of the few appraisal tools suitable for critical appraisal of studies with diverse designs, allowing the concomitant appraisal of many study designs using one tool. The tool has also been validated in several studies testing its interrater reliability, usability and content validity.<sup>75, 76</sup>

The tool has separate subsets of items appraising the quality of (1) qualitative methods, (2) quantitative methods (different criteria for different types of quantitative components), and (3) mixed methods - the approach used to combine qualitative and quantitative components. The tool has been designed to appraise the methodological quality of the studies retained for a systematic mixed studies review, not the quality of their reporting. The checklist includes two screening questions which were applied across all studies and further items to assess the quality of specific study types. Items are worded to reflect good quality. For example,

“Are measurements appropriate (clear origin, or validity known, or standard instrument)?” in which each study is rated as “yes,” “no,” or “cannot tell”.

### 2.3.4 Narrative Synthesis

Narrative synthesis methods were used to summarise, integrate and interpret the findings of articles included within the review. It was decided that a narrative synthesis was appropriate given that scoping searches identified that there was little uniformity amongst the methods, findings, or outcomes measures used to assess the issues and needs of survivors and caregivers.

In contrast to a meta-analysis, the defining characteristic of a narrative synthesis is that:

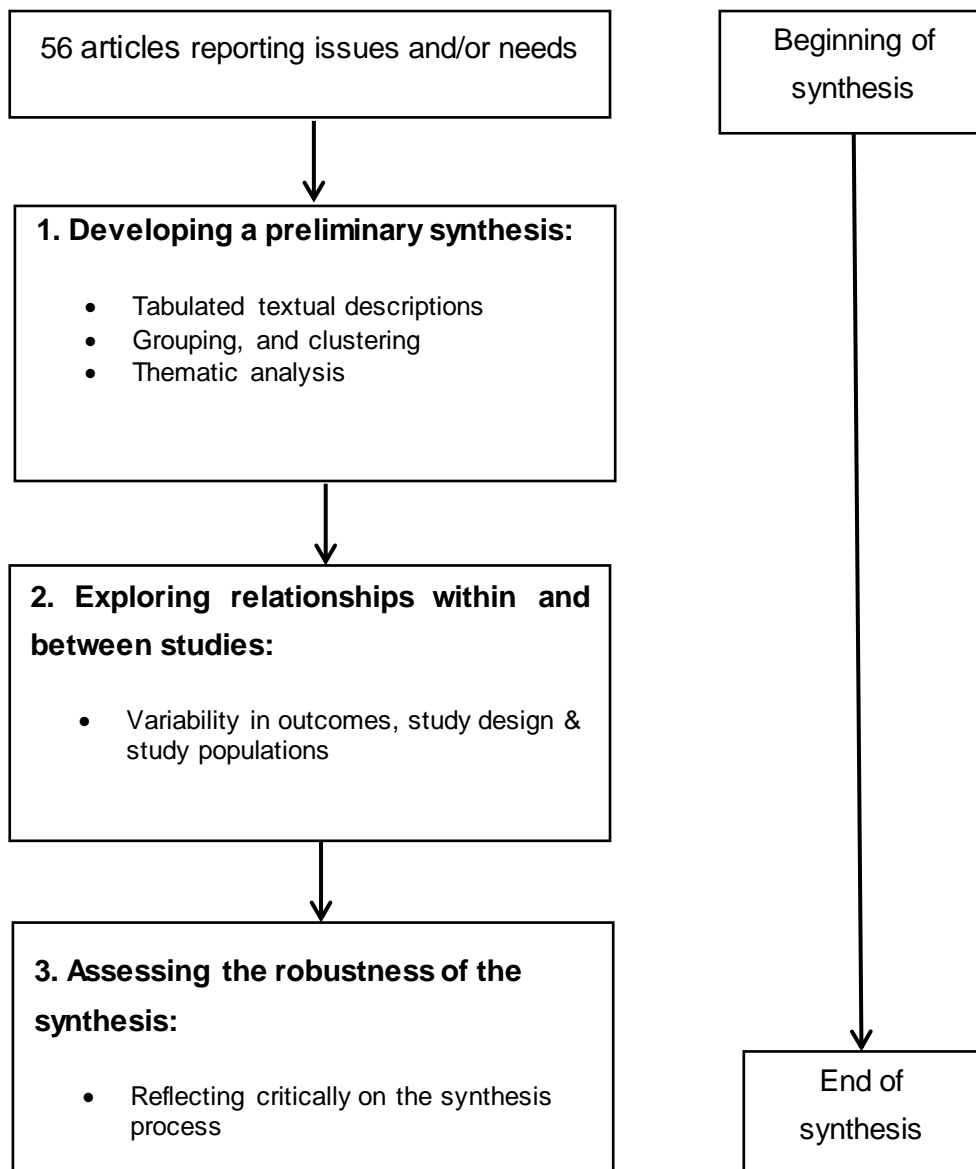
*“it adopts a textual approach to the process of synthesis to ‘tell the story’ of the findings from the included studies.” (p.5)<sup>77</sup>*

A narrative synthesis pursues to generate an understanding of the evidence and provide novel insights that would not otherwise be apparent either from only including certain types of data, e.g., quantitative data.<sup>78</sup>

Figure 1 shows the processes undertaken in the narrative synthesis, guided by Popay et al.<sup>77</sup> and the Cochrane Consumers and Communication review group handbook.<sup>79</sup> The processes undertaken included (1) preliminary analysis, (2) exploration of relationships, and (3) assessment of the robustness of the synthesis. The City of Hope quality of life conceptual model<sup>80</sup> was adapted to map the survivors issues. The original model proposed a quality of life model for long-term cancer survivors: physical wellbeing and symptoms, psychological well-being, social well-being and spiritual well-being. Cognitive symptoms were added to better fit TYA brain tumour survivor issues.



**Figure 1 - Narrative synthesis process**

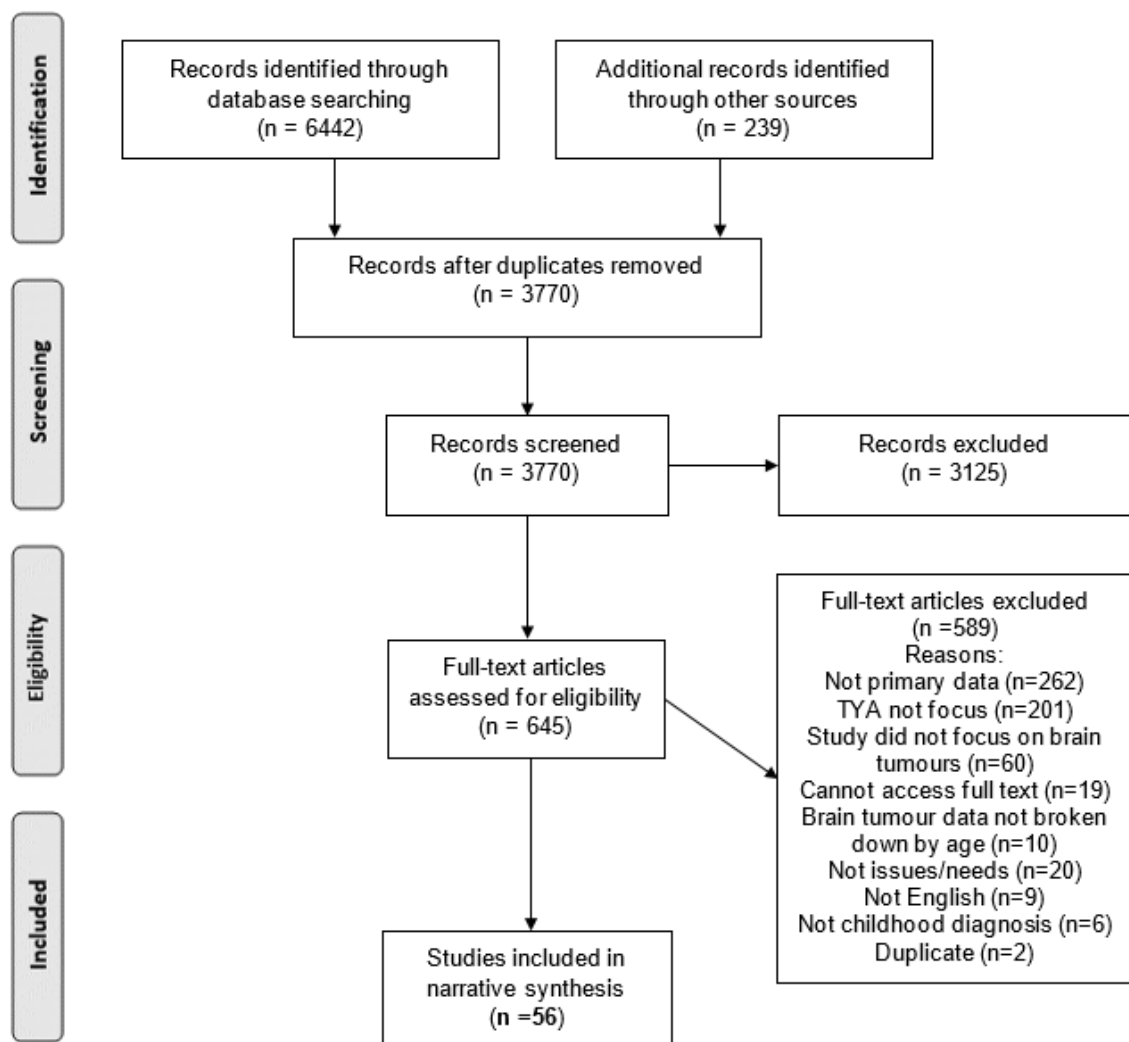


## 2.4 Results

### 2.4.1 Search Results

In total, 6442 article citations were found. 239 additional records were identified through hand searching journals and reference lists. All identified citations were downloaded into Endnote X7 and duplicates removed. Once duplicates were removed a total of 3770 articles remained. After reading the abstracts 3125 were removed because they did not meet the inclusion criteria. Of those remaining, 645 were retrieved in full-text, of these 589 were excluded because they did not meet the inclusion criteria. In total 56 articles (49 studies) remained for inclusion within the narrative synthesis. The process of searching and sifting is shown in Figure 2.

Figure 2- Flow chart showing article selection



### **2.4.2 Study characteristics**

Over half of the studies had been published after 2013 (57%), and 53% of studies originated within the United States. The majority of studies (76%) reported quantitative data; five (10%) were qualitative; and seven (14%) utilised a mixed methods approach. The majority collected data from the survivors only (86%), in three studies (6%) data was solely from caregivers and in four (8%) there was data from both survivors and caregivers. In over a third of studies (35%) there was a comparison or control group.

Over half the studies (55%) reported data from mixed brain tumour samples. A further eight studies (16%) reported one specific brain tumour group. Fourteen studies (29%) had recruited patients with varied cancer types, but reported brain tumour data separately. See appendix 2 for further description of studies.

### **2.4.3 Critical appraisal of included studies**

There was great variety in research designs and in MMAT-scores; scores ranged from 25% (one criteria met) – 100% (all criteria met). For this review no studies were removed because of their appraisal score but lower quality study findings should be interpreted with greater caution and with consideration for the limitations. See Table 2 for individual study scores.

**Table 2 - Critical appraisal scores using the MMAT Tool. See Pluye (2011) for MMAT guidelines and details of screening questions.<sup>81</sup>**

Author/Year	Study type	Score (%)	Screening questions		Qualitative				Quantitative Non-randomised				Quantitative descriptive				Mixed methods		
			A	B	1. 1	1. 2	1. 3	1. 4	3. 1	3. 2	3. 3	3. 4	4. 1	4. 2	4. 3	4. 4	5. 1	5. 2	5. 3
Ahomäki 2017	Cross-sectional	75	Y	Y					Y	Y	Y	N							
Ailion 2016	Cross-sectional	75	Y	Y					C	Y	Y	Y							
Ait Khelifa-Gallois 2015	Cross-sectional	100	Y	Y					Y	Y	Y	Y							
Armuand 2017	Cross-sectional	100	Y	Y					Y	Y	Y	Y							
Barakat 2015; Deatrick 2014; Hobbie 2016; Hocking 2015; Lucas 2014; Lucas 2016; Palma 2015	Mixed methods	75	Y	Y	Y	Y	C	Y					Y	Y	Y	Y	Y	Y	N
Boman 2009	Cohort	100	Y	Y					Y	Y	Y	Y							
Boydell 2008	Qualitative	75	Y	Y	C	Y	Y	Y											
Brinkman 2012	Cross-sectional	75	Y	Y					Y	Y	N	Y							
Chen 2008	Qualitative	75	Y	Y	Y	Y	N	Y											
Chou 2009	Mixed methods	75	Y	Y	Y	Y	N	N	N	Y	Y	Y					Y	Y	N
Cohen 2012	Cohort	75	Y	Y					Y	Y	C	Y							
D'Agostino 2013	Qualitative	100	Y	Y	Y	Y	Y	Y											
de Blank 2016	Cross-sectional	75	Y	Y					Y	Y	Y	N							
Deatrick 2009	Mixed methods	75	Y	Y	Y	Y	C	C					Y	N	Y	Y	Y	Y	N
Demers 2016	Cross-sectional	50	Y	Y					Y	Y	N	C							
Frangé 2009	Cohort	100	Y	Y					Y	Y	Y	Y							
Gray 1992	Mixed methods	25	Y	Y	Y	C	Y	N	N	Y	Y	Y					C	C	N
Gunn 2016	Mixed methods	75	Y	Y	Y	Y	Y	Y	Y	Y	Y	N					Y	Y	N
Heikens 1998	Incidence study	75	Y	Y									Y	Y	Y	C			
Hoffman 2015	Cross-sectional	100	Y	Y					Y	Y	Y	Y							
Jayakar 2015	Cross-sectional	50	Y	Y					N	Y	Y	N							
Jereb 1994	Cohort	50	Y	Y					Y	Y	N	C							
Johannsdottir 2012	Cross-sectional	100	Y	Y					Y	Y	Y	Y							
King 2015 I	Cross-sectional	75	Y	Y					N	Y	Y	Y							
King 2015 II	Cross-sectional	75	Y	Y					N	Y	Y	Y							
King 2015 III	Cross-sectional	75	Y	Y					Y	Y	Y	C							
King 2016	Cross-sectional	50	Y	Y					Y	Y	N	N							
Klosky 2014	Cross-sectional	100	Y	Y					Y	Y	Y	Y							
Koustenis 2013;	Incidence study	75	Y	Y									Y	Y	Y	N			

**Pfizer 2013**

<b>Lehmann I 2017; Lehmann II 2017</b>	Cross-sectional	75	Y	Y					Y	Y	Y	C	T									
<b>Liptak 2016</b>	Mixed methods	75	Y	Y	Y	Y	Y	N						Y	Y	Y	N	Y	Y	N		
<b>Maddrey 2005</b>	Cross-sectional	50	Y	Y					Y	Y	N	N										
<b>Mandrell 2012</b>	Case series	75	Y	Y										Y	N	Y	Y					
<b>Maunsell 2006</b>	Cross-sectional	100	Y	Y					Y	Y	Y	Y										
<b>Maurice Stam 2013</b>	Cross-sectional	75	Y	Y					Y	Y	Y	C	T									
<b>McClennan 2013</b>	Mixed methods	50	Y	Y	C	T	Y	Y	N					C	T	N	C	T	N	Y	Y	N
<b>McCurdy 2016</b>	Cross-sectional	75	Y	Y					Y	Y	Y	N										
<b>Petraroli 2007</b>	Incidence study	50	Y	Y										C	T	Y	Y	C	T			
<b>Pietila 2017</b>	Cross-sectional	100	Y	Y					Y	Y	Y	Y										
<b>Poggi 2005</b>	Incidence study	100	Y	Y										Y	Y	Y	Y					
<b>Strauser 2013</b>	Cross-sectional	75	Y	Y					Y	Y	Y	C	T									
<b>Taiwo 2017</b>	Cross-sectional	75	Y	Y					Y	Y	Y	C	T									
<b>Turkel 2007</b>	Incidence study	75	Y	Y										Y	N	Y	Y					
<b>Uday 2015</b>	Cohort	75	Y	Y					Y	Y	C	T	Y									
<b>Vance 2004</b>	Qualitative	75	Y	Y	Y	Y	N	Y														
<b>Wasilewski-Masker 2016</b>	Incidence study	75	Y	Y										Y	Y	Y	N					
<b>Wilford 2017</b>	Qualitative	75	Y	Y	Y	Y	Y	C	T													
<b>Zebrack 2002</b>	Cross-sectional	75	Y	Y					Y	Y	Y	N										
<b>Zebrack 2010</b>	Cross-sectional	75	Y	Y					Y	Y	Y	N										

- \*Screening question A) are there clear qualitative and quantitative research questions (or objectives\*), or a clear mixed methods question (or objective\*)? B) Do the collected data allow address the research question (objective)?
- 1.1. Are the sources of qualitative data (documents, informants, observations) relevant to address the research question?
  - 1.2. Is the process for analysing qualitative data relevant to address the research question (objective)?
  - 1.3. Is appropriate consideration given to how findings relate to the context, e.g., the setting, in which the data were collected?
  - 1.4. Is appropriate consideration given to how findings relate to researchers' influence?
  - 4.1. Is the sampling strategy relevant to address the quantitative research question?
  - 4.2. Is the sample representative of the population under study?
  - 4.3. Are measurements appropriate (clear origin, or validity known, or standard instrument)?
  - 4.4. Is there an acceptable response rate (60% or above)?
  - 5.1. Is the mixed methods research design relevant to address the qualitative and quantitative research questions?
  - 5.2. Is the integration of qualitative and quantitative data (or results\*) relevant to address the research question (objective)?
  - 5.3. Is appropriate consideration given to the limitations associated with this integration?

Within the MMAT categories the most common study type was 'quantitative non-randomised' (30 articles- including cohort, case-control and cross-sectional studies), the quality as reported ranged from 25 to 100%. Two cohort studies and seven cross-sectional studies obtained a score of 100. The quality of the remaining studies was limited mainly in terms of the recruitment of participants, and acceptable response rates (60% or above). Studies in general scored well in the measurements used being appropriate, clear and valid.

Seven studies were categorised as 'quantitative descriptive studies' (case series and incidence studies) scored 50-100%. 4 out of the 7 studies scored poorly or they did not report if there was an acceptable response rate (60% or above). That said, the majority (6/7) reported a relevant sampling strategy (i.e. a justified sample size -using power calculation for instance) and an appropriate, clear and valid measurement (7/7).

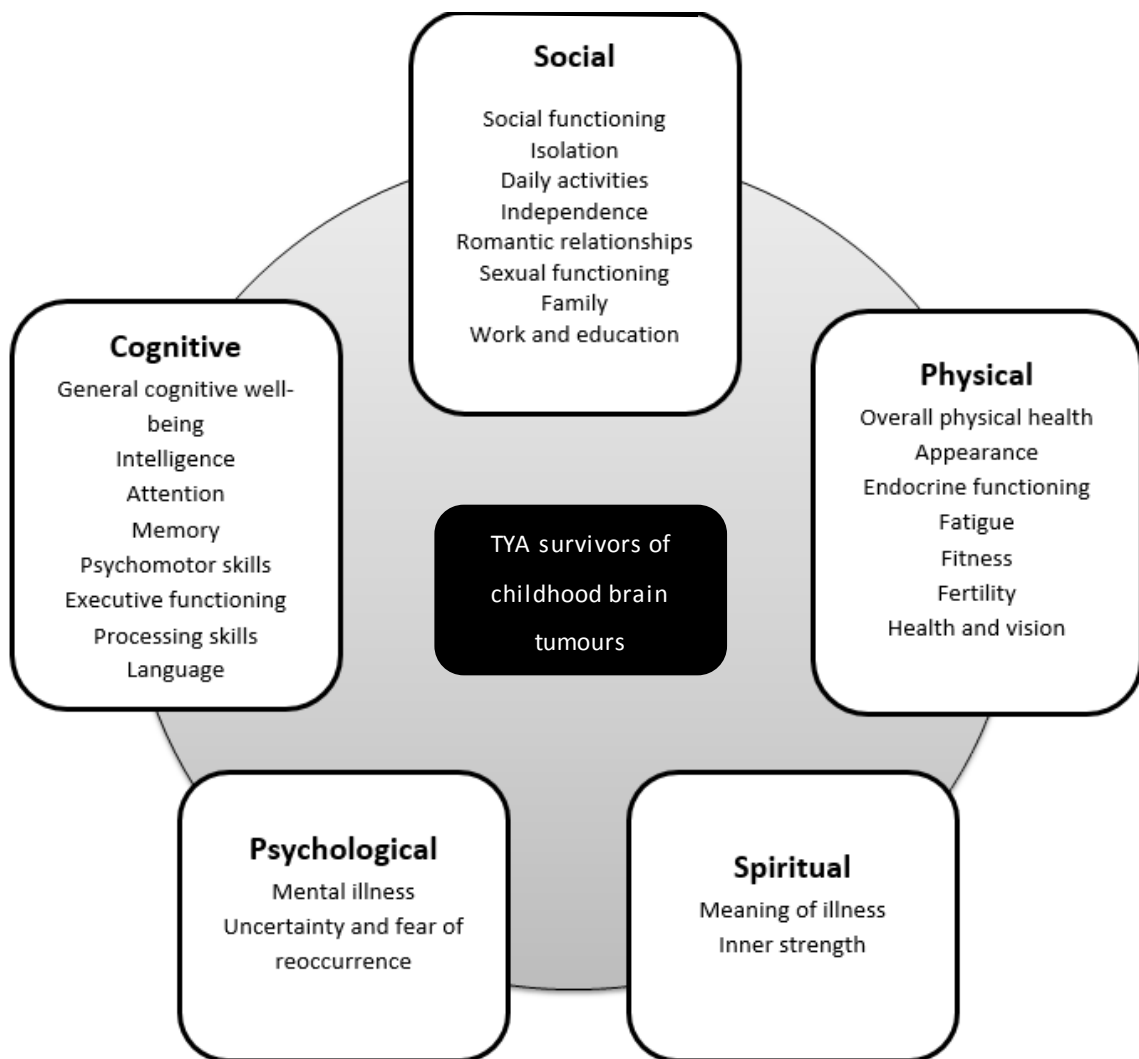
The quality appraisal scores of the five qualitative studies ranged from 75-100%. Qualitative studies scored lowest in regard to reporting the study context i.e. the setting, in which the data were collected.

The seven mixed method studies scored between 25 and 75%. It was recorded that the majority of the mixed methods studies had a relevant design to address the research questions and the qualitative and quantitative components were integrated at some stage to address the research question. Frequently this integration occurred at the interpretation stage but sometimes it occurred during data analysis. No mixed methods studies acknowledged or reflected on the limitations of their mixed methods design or appeared to address the philosophical tensions involved in mixed methods research.

#### 2.4.4 Section 1: Survivor issues

Survivor issues were derived from the main themes found in the articles and categories used in the City of Hope quality of life model.<sup>80</sup> The model has been adapted to illustrate the potential issues TYA brain tumour survivors may encounter which could impact on their quality of life (see Figure 3).

Figure 3 - Adapted Quality of Life Cancer Survivorship model

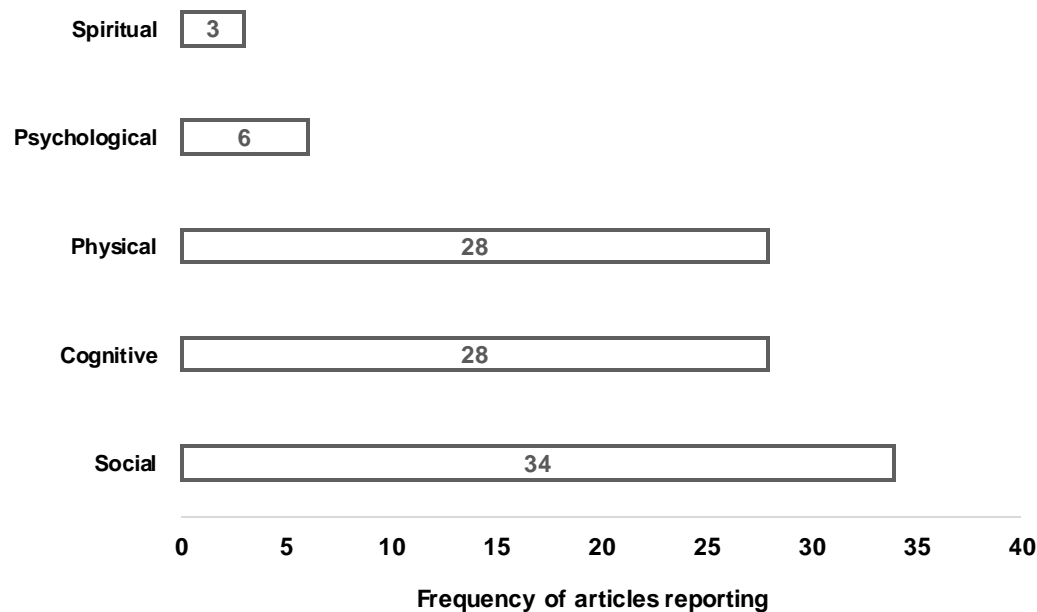


Adapted from: *Quality of Life Conceptual Model Applied to Cancer Survivors*, City of Hope Beckman Research Institute<sup>80</sup>

## Social well-being

Social well-being was the most commonly referred to theme (see Figure 4): Thirty-four articles investigated the impact a childhood brain tumour may have on social domains of a TYA survivor.

Figure 4 - Frequency of articles that reported survivor issues by theoretical framework theme



MMAT Scores by theme: Spiritual issues = All 75, Median = 75; Psychological = 75-100, Median = 75; Physical = 25-100, Median = 75; Cognitive = 25-100, Median = 75; Social = 25-100, Median = 75

Articles reported TYA survivors having impaired social functioning, expressed as e.g., avoiding social situations,<sup>82</sup> or social isolation.<sup>82-87</sup> Poorer social functioning was reported among brain tumour survivors compared to other cancer groups.<sup>88, 89</sup> Specifically, multiple studies reported a link between poorer social functioning and anti-cancer treatment and diagnosis.<sup>83, 85, 90, 91</sup> During treatment, survivors spend a long time in social isolation after which some young people found it difficult to engage with others again.<sup>85</sup> Physical issues, e.g., headaches or impaired fitness, as well as cognitive deficits were linked to social functioning,<sup>82, 92, 93</sup> and impacted on survivors' abilities to join in sports or hobbies.<sup>93</sup> Similarly, parents reported that their child's brain tumour had had a substantial or high impact of their social functioning<sup>84, 91, 94</sup> Parents described their children as "*having no social life*," "*no social outlet*," and "*not socializing outside of school*".<sup>84</sup> Unlike their parents, survivors did not show significant negative emotion to poor social functioning: "*It is not really upsetting to her that she doesn't participate fully*".<sup>84</sup> The lack of upset and distress amongst survivors was



consistently explained by parents as a lack of apathy, and social immaturity regarding social functioning.<sup>84</sup>

Six articles reported impaired daily functioning and/or functional living skills,<sup>83, 92, 95-98</sup> survivor restrictions ranged widely in severity,<sup>95, 96</sup> with only a small minority reporting no limitation in daily activities.<sup>98</sup> Qualitative work linked these restrictions to impaired cognitive functioning through difficulties with reading or handling finances, or managing medications.<sup>83</sup> TYA survivors rarely lived independently,<sup>88, 97, 99, 100</sup> which was linked to feelings of frustration.<sup>86, 92, 93</sup> That said, survivors generally report close relationships with their family.<sup>82, 83, 92</sup> Poorer survivor cognitive functioning<sup>101</sup> and lower income<sup>98, 101</sup> was associated with worse family functioning.

In romantic relationships, TYA survivors see themselves as less valuable mates to a prospective partner.<sup>102</sup> Survivors were likely to be sexually inactive.<sup>88, 92, 97, 103-105</sup> Reduced sexual activity was found when compared to other cancer groups<sup>88, 103, 105</sup> and the general population.<sup>92</sup> Survivors of a childhood brain tumour were also less likely to have children compared to general population controls<sup>106</sup> and other cancer survivors.<sup>107, 108</sup> However, it was also reported that brain tumour survivors' desire to have children was less than the general population.<sup>106</sup>

Many TYA survivors need assistance to perform well in school.<sup>92, 100, 108</sup> Education experiences were often described as problematic: including missing school, cognitive difficulties - feeling misunderstood, facing bullying and social isolation.<sup>85, 86, 93</sup> In qualitative studies, survivors described educators as not understanding their cancer experiences and needs - which was frustrating for some survivors. Whereas some survivors felt in a dilemma of wanting special considerations but not wanting to be different.<sup>85</sup>

Survivors were more likely to be unemployed later in life when compared to age and sex matched controls<sup>108-110</sup> and other cancer survivors,<sup>88, 111</sup> with reported unemployment rates varying from 8-70%.<sup>88, 95, 97-101, 108-113</sup> Issues in attaining or keeping a job included fatigue, poor concentration, physical issues (i.e. epilepsy), cognitive difficulties, and poor social skills.<sup>83, 92, 93</sup> Some survivors had sheltered employment, a setting in which people with disabilities receive services and training to develop work-related skills and behaviours.<sup>99, 100, 109</sup> TYA survivors were found to have significantly lower levels of vocational identity and career readiness (i.e. ability to perform work tasks, social skills) when compared to TYA non-cancer survivors.<sup>114</sup> Vocational identity is an integral part of human development, especially for TYAs. This process includes the formation of career prospects: objectives, goals, aspirations and plans. These results suggest that brain tumour survivors may benefit from comprehensive career/vocational information and services. Financial difficulties were also

common in TYA brain tumour survivors.<sup>101, 115, 116</sup> TYA survivors were more likely than other cancer groups to be receiving disability benefits.<sup>116</sup>

### **Cognitive well-being**

Different aspects of cognitive deficits were reported in 28 articles. Overall, in these articles, cognitive functioning was found to be impaired. TYA brain tumour survivors were often at higher risk of cognitive issues than other cancer survivors.<sup>88, 111, 115, 117</sup> Impaired memory seemed to be the most common cognitive issue.<sup>82-87, 97, 101, 109, 110, 118-121</sup> Qualitative findings<sup>82-85, 92, 109, 118</sup> established that impaired memory was a daily issue. Memory issues were innate in how survivors' and their caregivers assessed their ability to self-care and meet developmental milestones, such as living independently,<sup>83</sup> making friendships,<sup>84</sup> and educational achievements.<sup>92</sup> For some, memory loss was described as an invisible effect.<sup>85</sup> Unlike physical effects, invisible effects may go undetected and mean the appropriate additional support may not be offered. It is then that memory may impair other areas of TYA survivors' lives, such as education<sup>85, 92</sup> : *"I have a hard time retaining knowledge. That's probably the most difficult thing. My memory is just shot."*<sup>85</sup>

Attentional deficits were particularly challenging because of their consequent impact on the development of other cognitive abilities, social functioning and academic achievement.<sup>82, 97, 110, 119, 121</sup> Evidence suggested that TYA survivors scored lower in Intelligence Quotient (IQ) scores and had limited mathematical skills.<sup>90, 95, 96, 109-111, 117, 120, 122</sup> TYAs scored lower in IQ testing than population controls.<sup>96, 122</sup> Survivors treated with radiotherapy were most at risk of limited intellectual capacities.<sup>122</sup>

Many survivors presented with poor processing speeds,<sup>82, 84, 99, 101, 110, 121, 123</sup> leading to poorer physical and/or mental health,<sup>99</sup> social functioning<sup>82, 84</sup> and health-related quality of life.<sup>99, 101</sup> Parents reported that processing speed difficulties were a consistent issue: *"The processing speed is just with everything, it just covers everything. It's the worst thing. Because even socially it affects him. He cannot keep up with the conversation with other kids his age"*.<sup>82</sup> Similarly language and vocabulary issues were challenging.<sup>92, 97, 117, 122</sup> Compared to matched healthy controls, brain tumour survivors scored significantly lower in verbal assessments.<sup>92, 108, 122, 123</sup>

Some survivors suffered from impaired motor skills.<sup>92, 96, 97, 99, 108, 110</sup> Motor deficits contributed to social isolation and vocational limitation by restricting the types of activities open to survivors (i.e. playing sports or writing).<sup>92, 97</sup>

Executive functions are a diverse set of cognitive processes broadly conceptualised according to four primary domains: decision making, planning (e.g. organisation), purposive

action (e.g. set shifting - the ability to move back and forth between tasks), and effective performance (e.g., preservation, goal maintenance). Survivors experienced poor executive functioning including: planning/organising,<sup>96, 110</sup> preservation,<sup>96</sup> set shifting,<sup>96, 97, 110</sup> and flexibility.<sup>110</sup> Survivors self-reported less executive dysfunction in comparison to when their mothers reported symptoms (by proxy). This may be problematic as survivors who perceive fewer executive functioning difficulties may not pursue help or support, such as neuro-rehabilitation programmes, which may have a negative effect on other long-term functional outcomes.<sup>124</sup>

## Physical well-being

Survivors had impaired general physical health.<sup>82, 88, 89, 91, 92, 108, 115</sup> More specifically symptoms included: poor mobility,<sup>82, 92, 100, 108</sup> poor physical functioning,<sup>83, 91</sup> reduced bone mineral density,<sup>125, 126</sup> hearing and/or vision issues,<sup>82, 84, 86, 92, 94, 100, 108</sup> and poor fitness levels.<sup>92, 117</sup>

At a time when physical appearance becomes increasingly more salient, TYA survivors reported having issues with their appearance.<sup>83, 85, 88, 92, 93, 100</sup> Visible effects after the tumour and treatment included –small stature, hair loss, weight issues and scars.<sup>83, 85, 92, 93</sup> Survivors said their physical appearance meant that they were often mocked or felt rejected by others: *“In high school...they thought I was funny cause my head was shaped weird, and called me all weird names. I didn't like it.”*<sup>83</sup> Parents of survivors described that issues with body image contributed to starting/maintaining peer relationships because survivors worried they looked different from their peers.<sup>93, 100</sup>

TYA survivors commonly had endocrine dysfunctions.<sup>82, 84, 92, 99, 100, 111, 120, 127-130</sup> The endocrine system interacts with body organs and tissues, serving as a major contributor to overall health and wellness. Growth hormone deficiency was reported in 22-97% of survivors<sup>95, 100, 111, 127, 128, 130</sup> and more common in brain tumour survivors than other cancer diagnoses.<sup>111</sup> Other frequently reported endocrine disorders included hypothyroidism,<sup>95, 128, 130</sup> hypogonadism<sup>92, 95, 130</sup> and cortisol deficiency.<sup>91</sup> Issues with endocrine functioning during adolescence could impact on growth and development, weight gain, reproductive processes, and mood. Both young age at treatment<sup>127</sup> and chemotherapy and radiotherapy treatment<sup>130</sup> were linked with more prevalent endocrine dysfunctions.

Many survivors experienced increased levels of fatigue as teenagers and young adults.<sup>82, 84, 87, 88, 92, 93, 115, 128, 131, 132</sup> In comparison to other cancer survivors, brain tumour survivors were more likely to report difficulty with tiredness and fatigue.<sup>87, 115</sup> In one study fatigue was linked to sleep apnea.<sup>128</sup> Whilst fatigue is reported as a physical issue following the cancer

survivorship model, it is also a common symptom of many cognitive and psychological problems. Survivors expressed how fatigue had stopped them doing hobbies/sports and socialising.<sup>100</sup>

Regarding fertility, childhood brain tumour survivors fell pregnant significantly less often than healthy peers.<sup>106</sup> That said, brain tumour survivors' desire to have children was significantly lower than healthy peers of the same age.<sup>106</sup> For some fertility was not currently an issue, especially for teenagers, but it was something they were anxious about facing in the future.<sup>93</sup>

### **Psychological well-being**

Survivor mental health did not differ from general population controls.<sup>86, 108</sup> However, one article found that TYA brain tumour survivors reported significantly poorer mental health than other cancer survivors.<sup>89</sup> Another reported that TYA brain tumour survivors had poorer psychological functioning than other cancer survivors.<sup>88</sup> The most frequent psychological problems for survivors were internalising problems and withdrawal.<sup>90</sup> Psychotic symptoms (i.e. delusional thinking and hallucinations) were diagnosed in a small number of survivors, with antipsychotic medication having little effect.<sup>133</sup>

Despite improvements in recurrence-free survival rates for children diagnosed with brain tumours, TYA survivors still worry about recurrence. In two qualitative studies participants expressed that the anxiety of whether the tumour was going to return, was one of the realities of living as a survivor.<sup>86, 118</sup> In some cases the uncertainty of recurrence negatively affected survivors' ability to plan for the future or feel engaged in life.<sup>86, 118</sup> Both studies highlight that the fear of recurrence may become an issue as survivors enter young adulthood; especially if they feel unable to plan a future, they may not reach the same developmental milestones.

### **Spiritual well-being**

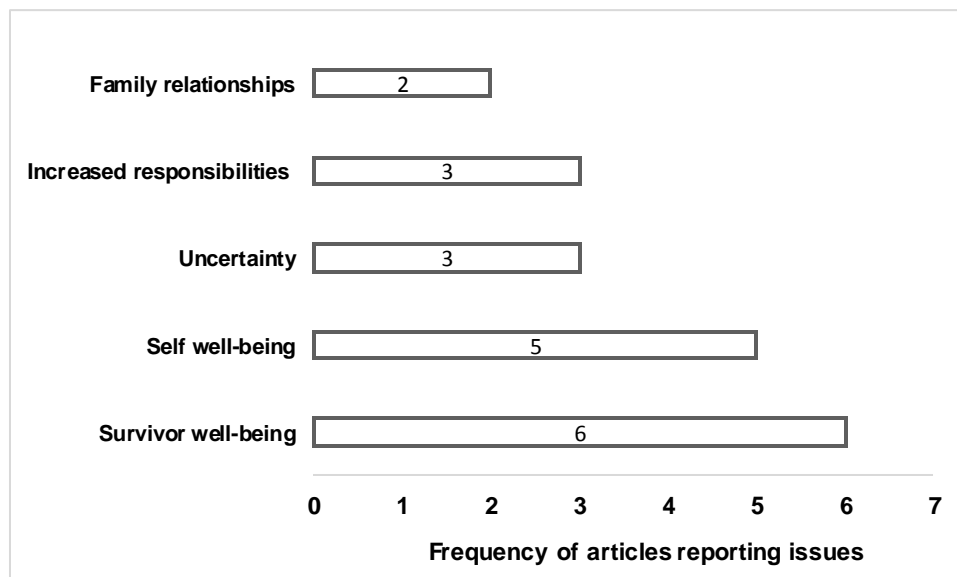
Compared to other TYA cancer survivors, brain tumour survivors had significantly poorer levels of optimism, self-esteem and vitality.<sup>89</sup> However, survivors were not less resilient.<sup>88</sup> The way survivors viewed the meaning of their illness differed vastly.<sup>83, 86, 92</sup> Some focused on the negative effects of illness, seeing themselves as 'losers' for their deficits. Some survivors said they were treated differently because of their cancer history, and felt that others pitied them.<sup>92</sup> Yet in the same study half of survivors described at least one positive consequence of surviving a brain tumour on their world view.<sup>92</sup> Some survivors positively viewed their experience of surviving a brain tumour, feeling it had made them more mature and were trying to move beyond the illness.<sup>86</sup>

## 2.4.5 Section 2: Caregiver Issues

The next section is going to focus on the reported issues that caregivers of TYA brain tumour survivors' experience. There was considerably less reported on caregivers issues, only eight articles reported caregivers' issues,<sup>82, 84, 85, 91, 93, 98, 134, 135</sup> of which four focused specifically on studying the role of the family caregiver.<sup>91, 98, 134, 135</sup> Caregiver participants consisted of parents (mothers and fathers),<sup>82, 91, 136</sup> mothers only<sup>84, 98, 134, 135</sup> and mixed family members (including grandparents).<sup>85</sup>

Parents celebrated their child making it through the cancer experience, although now in survivorship this was coupled with many challenges. The continued demands of caregiving into adolescence and young adulthood weighed heavily on many parents. Caregiver issues had five themes (see Figure 5): survivor well-being (results cover the effects of survivor well-being on caregiver worries/well-being), uncertainty; increased responsibilities; self-well-being and family relationships. Unlike survivor issues, which adapted an existing framework (Cancer survivorship model), this framework was developed to show caregiver issues. The reason it was developed was because existing frameworks were limited and were not applicable to this systematic review findings.

**Figure 5 - Frequency of articles that reported caregiver issue by theoretical framework theme**



*MMAT Scores by theme: Family relationships = All 75, Median = 75; Increased responsibilities = All 75, Median = 75; Uncertainty = All 75, Median = 75; Self well-being = 75, Median = 75; Survivor well-being = 75, Median = 75*

## **Survivor well-being**

Parents were concerned about the survivor's well-being. Caregivers worried that survivors had limited social life and that they weren't accepted into social groups.<sup>85, 93, 98, 134</sup> They acknowledged that survivors' lives were fundamentally different to others their age because of the brain tumour and worried if they would ever recover their previously existing social network.<sup>93</sup> Parents often compared their child's current social life with how it was before the brain tumour: "*Jennifer was very popular, very, always had friends for tea...always got invited to parties and things and none of that happens now... and I find that hurts me.*"<sup>93</sup>

Parents were not only worried about survivors' current issues but about potential issues in the future.<sup>91</sup> In particular their ability to find a romantic partner<sup>82, 91</sup> and have children.<sup>93</sup> Others feared that their dependent children might outlive them and worried who would care for them in the future.<sup>91</sup>

## **Uncertainty**

Uncertainty was a common theme, especially regarding survivors' future health.<sup>91, 93</sup> As seen in survivor reports, the threat of relapse was a pressing issue. In some cases this threat stopped parents from planning future activities, and instead they lived very much in the present.<sup>91, 93</sup> Another source of uncertainty came from whether their child would ever reach independence.<sup>93, 135</sup> In one study the majority of parents believed their child would never be independent.<sup>135</sup> Some worried about leaving them alone in the house or their ability to be financially independent.<sup>93</sup>

## **Increased responsibilities**

Caregivers had increased responsibilities, tasks that usually healthy TYAs can handle independently. Qualitative findings described parents assisting survivors in: everyday tasks (i.e. cooking meals, managing finances and promoting hygiene); managing medications, education; arranging social contacts; and support with hobbies and leisure activities.<sup>85, 93, 134</sup> In some cases parents decided to home school the survivor because they felt they needed extra assistance and support.<sup>85, 93</sup>

## **Self-Well-being**

Five studies reported diminished caregiver well-being.<sup>84, 91, 93, 98, 134</sup> Parents revealed that because of their caregiving demands, they had less time and energy to dedicate to maintaining self. Parents described feeling fatigued,<sup>93, 134</sup> having problems maintaining their

own social relationships,<sup>84, 91, 134</sup>; and not being able to pursue their own careers.<sup>134</sup> Their social lives declined because caring became their main priority, meaning they had less time to engage in social and leisure activities, thereby limiting their social world.<sup>84, 91</sup> Caregiver health plays a major role in family life, one study highlighted the direct association between caregiver well-being and better family functioning.<sup>98</sup>

### **Family relationships**

The pressure of keeping healthy family relationships, when caring for a young brain tumour survivor, could prove to be challenging for caregivers. Families of children who have survived a brain tumour may face issues that make family management more difficult. For some caregivers their marriage had suffered – leading to separation or divorce.<sup>82, 91</sup> Strain was noted because of the added pressure of caring for the survivor and also because of disagreements between partners about how best to care for their child.<sup>91</sup> For others the diagnosis, treatment, and late effects had brought them closer together.<sup>82</sup> Additionally, parents worried about their relationships with survivors' siblings, anxious they felt alone or as if they "*got the short end of the stick*".<sup>82</sup> In some cases siblings showed emotional and/or behavioural problems.<sup>82</sup> In one study, survivors were dependent and frequently in need of their sibling's help during adolescence.<sup>86</sup>

### **2.4.6 Section 3: Supportive care needs**

This next section will address the second and third objective. Firstly, the needs of TYA childhood brain tumour survivors and their caregivers. And secondly, whether survivors and caregivers feel their needs are being met.

#### **Survivor Needs**

Only three studies aimed to specifically identify survivor needs.<sup>84, 118, 137</sup> TYA needs differed from those of paediatric and older adult survivors, including the need for age-specific social resources. TYA brain tumour survivors were more likely to value social activities and support groups, compared to other cancer groups.<sup>137</sup> Social activities and support groups were favoured above informational mailing, weekend retreats, informational workshops or individual counselling.<sup>137</sup> Social resource recommendations included creating several arenas for peer support.<sup>118</sup> Parents highlighted that their child needed more opportunities to merely "hang out" with peers.<sup>82</sup> When a TYA social support programme was evaluated survivors relayed positive experiences.<sup>84</sup> Participation in the programme provided survivors with "*something to look forward to*", a regular social activity, important in addressing some of the social isolation issues. Furthermore, including the use of social media as a part of the

programme was recognised as a manageable and satisfying way to decrease isolation within the survivors.

Some survivors felt the current health care delivery did not meet all their needs. Many were unsure how to discuss issues with physicians, and others felt the information they received was inadequate because the medical staff communicated solely with their parents.<sup>109, 118</sup> It was suggested that providing survivors with age-specific resources about late effects and psychosocial challenges at each follow-up appointment may be helpful.<sup>118</sup>

Although only three studies focused on survivor needs, several studies (as a consequence of their findings) stressed the need for comprehensive follow-up services for childhood survivors.<sup>92, 99, 100, 112, 130, 131</sup> The importance of follow-up in a survivorship clinic to offer coordinated, multidisciplinary care that can address the multiple issues/needs of survivors of brain tumours while addressing caregiver needs and family functioning was highlighted. One study stressed that health professionals should be mindful that many survivors may define themselves as healthy and will not necessarily identify late effects caused by their illness/treatment.<sup>92</sup> Findings specifically identified careful follow-up of endocrine function,<sup>130</sup> liver function,<sup>131</sup> and health related quality of life.<sup>99, 112</sup>

The stress of keeping up with their classmates in an environment that was not equipped to handle their needs proved difficult for many young survivors.<sup>85</sup> Regarding educational support, survivors described needing: extra time to complete assignments/exams, more one-on-one help, and extra encouragement.<sup>85, 93</sup> Some survivors said that by appearing more “normal” meant that they were not always recognised as needing additional support with their academic work.<sup>85</sup> Throughout survivor narratives, the paradox of wanting special considerations within the classroom yet not wanting to be different or singled out among their peers was strongly evident. Another study concluded that academic/vocational goals and expectations must be observed over time, as learning needs may change.<sup>83</sup>

### **Caregiver needs**

Again only three studies investigated and reported caregiver needs. Where reported parents consistently reported that there was not enough support available for themselves (or the survivor during adolescence and young adulthood). Parents felt that when their child was undergoing treatment they had support, but that support declined as they moved further away from the treatment.<sup>91, 93</sup> In one instance, a mother said she felt the safety net was not there and that “if I don’t do it, who will?”<sup>91</sup>



Regarding useful support services, parents expressed that parent support groups and survivorship education classes would be most helpful. Other services mentioned were parental health and self-care classes and practical support such as financial assistance.<sup>82</sup>

Two articles highlighted that different caregivers and families deal with survivorship differently and will have different needs.<sup>91, 135</sup> One study found that clinicians find it challenging to tailor interventions to family and caregiver needs, as tools to assess family functioning and caregiver coping are not available.<sup>91</sup> It was highlighted that nuanced communication is needed, not only during treatment, but also into survivorship with specific approaches to meet caregiver needs and provide coping skills to manage stressful situations.<sup>91, 135</sup>

## **2.5 Discussion**

The systematic review revealed that TYA brain tumour survivors can encounter various social, cognitive, physical, psychological and spiritual issues which have the potential to significantly impact upon their wellbeing. Social, cognitive and physical issues were the most frequently reported - with poor social functioning being the most pressing. A childhood diagnosis of a brain tumour appeared to inhibit or change how TYAs interact with family, friends and employers as well as how they cope with unexpected and unwanted changes in areas ranging from employment stability to romantic relationships. Survivors miss out or struggle with achieving key life-events including: independence, educational achievements and becoming parents.

Cognitive deficits meaningfully contribute to these poor social outcomes. Survivors experience neurocognitive late effects across multiple domains that often hinder their independence - and are associated with poorer social functioning. This is compounded by growing concerns about their physical issues as they move into adolescence and young adulthood.

Identified problems were frequently more prevalent, or more intense, than in other cancer groups, making it more likely that brain tumour survivors will have poorer overall well-being. Similar to the findings of this review, another review (not focusing on TYA survivors specifically) reported poorer quality of life outcomes in paediatric brain tumour patients than those diagnosed with other cancers,<sup>138</sup> emphasising that brain tumour supportive care should be disease-specific and tailored to individual needs.

The majority of identified studies focused on only survivor's experiences rather than their caregivers'. Yet the findings highlight that caregivers may experience momentous stress

related to the pressures associated with their caring responsibilities. These pressures can take their toll on well-being, especially when support is not available. Caregivers indicated that support had declined as they moved further away from the survivor's treatment into long-term survivorship.

There was little data reported that specifically focused on the supportive care needs and/or unmet needs of TYA childhood brain tumour survivors. Equally none of the studies included in our review assessed the needs of the TYA survivor's caregiver. In a sample of mixed cancer survivors (not just those living beyond a diagnosis of a brain tumour),<sup>139</sup> over 50% of TYA survivors indicated that they had unmet needs in relation to information and services. Based on the current review, it would be reasonable to expect that this percentage would be higher for brain tumour survivors because of the higher prevalence and complexity of late morbidities. However, it is still unclear what these specific needs are and whether they are currently being met.

### **2.5.1 Strengths and limitations**

This narrative synthesis provides a first systematic overview of the issues and needs experienced of TYA survivors of childhood brain tumours and their caregivers.

However, there are limitations to the current synthesis. In the guidance published by Popay et al. they recommend a summary that reflects critically on a) included studies and b) the synthesis process.<sup>77</sup>

#### **2.5.1.1 Limitations of included studies**

There are several limitations to the research reported in this review. First, the quality of the evidence varied per study, as reflected in the MMAT scores (see appendix 2), and thus, findings should be interpreted with caution. Second, the majority of the studies involve a cross-sectional design. This is problematic as adolescence is a period of rapid development and change - behavioural patterns are established, cognitive functions mature, physical changes occur, and complex social relationships develop. Future research should focus on collecting longitudinal data that will allow us to examine if and when survivors and caregivers issues/needs change as they progress into adulthood. Third, numerous studies had small sample sizes. Understandably, as the number of childhood brain tumour survivors is relatively low, it can be hard to recruit adequate sample sizes. Therefore, for those studies with small samples, findings should be interpreted with caution, especially when looking at differences between sub-groups. Additionally it was often the case that studies recruited participants from past study samples, meaning that these participants may be

overrepresented in the review. However, when this was the case it was generally clearly reported. Fourth, in the majority of studies with caregiver participants, the views of the mothers dominated the sample, with very little representation from fathers. However, this may not necessarily be a limitation as this could simply reflect what the caregiver population looks like. Fifth, in three studies survivors had been treated up to four decades ago, these results may not be relevant to survivors who have been treated more recently.<sup>87, 111, 127</sup>

Older treatment periods may present varying treatment effects and therefore issues and needs, whilst different levels of supportive care may have been available. More recently, long-term survivorship has prompted many new treatment procedures aimed at reducing late effects while maintaining the survival rates. Treatment is now generally less aggressive: fewer radical surgical procedures, lower doses of radiation therapy, smaller volumes of radiation therapy, and fewer and less intensive courses of chemotherapy.<sup>140</sup> Finally, this is a relatively new area of study, and the majority of the studies included in this review have been conducted in the United States. Therefore, additional studies are needed in other countries to identify specific issues and needs that might be culturally tied or dependent upon differences across health and social care systems.

#### **2.5.1.2 Limitation of synthesis**

There are limitations specific to the systematic review. First, due to the inclusion criteria, some studies with important findings were excluded from this review. For example, the set age criteria (14-39) meant that studies with participants outside this parameter were excluded, even if the majority of the participants were aged 14-39.<sup>141, 142</sup> Second, some potentially relevant articles may have been missed for the following reasons: only 20% of the abstracts were reviewed by a second reviewer, and we were not able to access all full-texts. Also new evidence is constantly emerging, and the review does not include any articles published after the last search date (September 2017). Third, in some studies brain tumour survivors only formed a small percentage of the study population. Although some data were reported separately, allowing us to include the study within the review, this data was not always very detailed. Fourth, only studies published in English were used due to resource limitations and time constraints. This may have meant that studies with different cultural perspectives were missed. Finally, even though the MMAT tool was deemed the best potential instrument to critically appraise the studies, it does have its limitations. At times the tool seemed a little too simplistic, due to only being able to grade each criteria with a 'yes', 'no' or 'cannot tell' answer. For example, one study that meets all aspects of a certain criteria would score a 'yes', whilst another study that only meets some of the criteria would also be scored a 'yes'.

## **2.5.2 Clinical implications**

Currently, there is insufficient knowledge of what TYA brain tumour survivors specifically need from supportive care. Only a small number of studies in the review addressed unmet supportive care needs. Despite the numerous studies reporting survivor issues, it is known that the presence of issues is not always related to the need for, or uptake of supportive interventions.

The systematic review showed that TYA survivors often experience many unique long-term issues as a consequence of the diagnosis and treatment of a childhood brain tumour. These issues are different to older survivors and those still in childhood. Their priorities and unique life events mean that the late effects of treatment impact their lives differently to other age groups. Adolescence is a period of trying to gain independence, but the review highlights that survivors can find this difficult – emotionally and practically. For example, impaired daily functioning and cognitive issues were integral to their ability to self-care. Unlike children, TYAs have to make important decisions about their education and future careers. TYAs also start to plan for the future, with romantic relationships considered a critical developmental marking one's entry into adulthood, alongside starting a family. Therefore, it is important that this group is provided with age-specific information, support and resources that guide them through adolescent life events, such as further education, learning to drive, paid work and relationships. Supportive services should be mindful that adolescence and young adulthood is a period of constant change and that the need for information and support may change rapidly, meaning regular reviews may be necessary. Information, support and resources should be brain-tumour specific, as experiences of brain tumour survivors differ significantly to those with other cancers.

Furthermore, it is recommended that caregivers' potential needs are considered by long-term follow-up teams and support services. Even though there was little depicted in the literature, it is anticipated that they too many require access to information, support and resources for caring for an TYA childhood brain tumour survivor.

## **2.6 Conclusions and implications for research and practice**

Living with the consequences of a childhood brain tumour can be particularly challenging for TYA survivors (aged 14-39) and their caregivers. Survivors and caregivers continue to report long-term issues and unmet needs throughout follow-up. Many of their issues are unique or more prevalent than in other cancer groups, due to many survivors experiencing cognitive impairment as a consequence of their tumour and its treatment. More research is needed on

the specific unmet supportive care needs of both survivors and their caregivers and how support services can best meet these needs. Understanding their unmet needs and recognising what services are required due to the late effects of treatment is critical to improving their quality of survival.

Significant gaps exist in our understanding of the unique needs of TYA brain tumour survivors and their caregivers. The extent to which unmet needs are related to tumour and treatment characteristics requires further research. From the systematic review, three areas of future research are identified. First, studies are needed to describe the needs and more importantly unmet needs of both TYA childhood brain tumour survivors and their caregivers. Increasing our understanding of the unmet needs will help to develop more targeted and effective supportive care models. Second, descriptive studies are needed to fully investigate survivor and caregiver expectations for supportive care and how these expectations comply with the current use of long-term follow-up care and supportive services. Subsequent to the results from descriptive studies, evidence based programmes and services need to be modified and/or developed to address both TYA survivor and caregiver unmet and desired supportive care needs. Third, longitudinal work is needed to help determine how surviving a childhood brain tumour changes overtime. The majority of studies included in this review encompass a cross-sectional design, which are helpful to understand the prevalence of the issues/needs but do not highlight change over time. This may be problematic because adolescence and young adulthood is a period of rapid development. Future research should focus on collecting longitudinal data that will allow us to examine if and when survivors and caregivers issues/needs change as survivors progress into adulthood.

## **2.7 Planning the mixed methods study**

Despite being one of the most common cancers in children, brain tumour research is still underfunded. Spending on brain tumour research is under 1% of the national cancer research spend.<sup>143</sup> Moreover, research has traditionally focused on increasing overall survival and stopping tumour growth, with little focus on the side effects after treatment. Studies to improve quality of life are even rarer and this is especially relevant in this younger patient population which has a favourable prognosis but high level of disease-specific symptoms and burden. In 2017/2018 financial year just 1% of the total spend on brain tumour research was focused on 'survivorship issues'.<sup>144</sup>

The percentage of young people surviving a childhood brain tumour has vastly increased in the last 30 years.<sup>145</sup> Therefore, understanding their needs and recognising what services are required due to the late effects of treatment is critical to improving their quality of life. It is

important that needs are accessed separately for TYA survivors in comparison to children or adults, as the systematic review findings suggest that their needs are influenced by unique developmental issues, life milestones, social relationships, and other age-related issues.

Based on the knowledge gap identified in the systematic review, the research aims and objectives for the mixed methods study were proposed. The overall aim of the research was to gain an in-depth understanding of the supportive care needs of TYA childhood brain tumour survivors and their caregivers.

Specific objectives of the study include:

- 1) To describe unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers.
- 2) To explore if sociodemographic (e.g. sex, age) and clinical data (e.g. time since diagnosis, tumour treatment) are related to unmet supportive care needs.
- 3) To determine whether unmet supportive care needs are associated with QoL outcomes.
- 4) To explore the role and perceived use of support services in TYA survivors and their caregivers.

### ***Reflections, learning and decision making...***

The systematic review highlighted there is a dearth in the brain tumour literature of longitudinal studies, with the majority of studies being cross-sectional. This finding highlighted a need for more longitudinal studies to assess changes in issues and needs over time. Ideally the mixed methods study (discussed in the following chapters) would have collected data over two time points to allow longitudinal analysis. However, due to the time restraints of a PhD, it was decided that it was unfeasible to design a longitudinal mixed methods study with sufficient time to recruit participants, and collect and analyse the data.

This research is important, as gaining information about survivor and caregiver needs is key to redesigning or optimising existing support to better meet survivor and caregiver needs and if necessary developing new supportive services.

## Chapter 3: Methodology

The findings of the previous Chapter were carried forward into the development of the mixed methods study presented in the rest of this thesis. Chapter 3 presents the development of the methodological approach used for the empirical stages of this study. Often the terms methodology and methods are used inter-changeably. However, the terms have different meanings. Research methodology is more than just the methods used to collect data and evidence. Instead, methodology focuses on the underlying concepts and philosophy that support the methods and offers theoretical understanding for the most appropriate methods for exploring a certain phenomenon in depth.<sup>146</sup>

The GRAMMS (Good Reporting of A Mixed Methods Study) guidelines for carrying out and presenting mixed methods research in health research was followed (Table 3).<sup>147</sup>

**Table 3 - Good Reporting of A Mixed Methods Study (GRAMMS) as proposed by O'Cathain (2008)**<sup>147</sup>

	Chapter/section
1. Justification for using a mixed methods approach	3.3
2. Design: purpose, priority and sequence of methods	3.5
3. Each method: sampling, data collection and analysis	4.3 – 4.5
4. Integration: where, how and who	4.5
5. Limitations	7.3.1
6. Insights from mixing or integrating methods	7.3

This Chapter begins with a definition of mixed methods and its history in health research (3.1). Then the rationale for using methods is provided (3.2), followed by the philosophical positioning of the research (3.3). Finally, an overview of mixed method designs is then discussed, outlining the convergent design chosen in this research (3.4). The other aspects of the GRAMMS guidelines are addressed in the subsequent chapters.

### 3.1 Mixed methods research methodology

#### 3.1.1 Definition

There are many comparable definitions to describe mixed methods research. A widely recognised definition of mixed methods research is:

*“An approach to research in the social, behavioural and health sciences in which the investigator gathers both quantitative (closed-ended) and qualitative (open-ended) data, integrates the two, and then draws interpretations based on the combined strengths of both sets of data to understand the research problems.” (p.2) <sup>148</sup>*

Central to the definition is the use of both quantitative and qualitative methods in one study. Hence, isolated quantitative and qualitative studies addressing the same research question independently would not be deemed ‘mixed methods’ as there would be no integration at the design, analysis or presentation stage.<sup>148</sup>

A core assumption of a mixed method approach is that statistical trends (quantitative data) and personal experiences (qualitative data) are combined, providing a better understanding of the research problem than either form of data alone.<sup>148</sup>

### **3.1.2 History of mixed methods in health research**

This section begins with a comparison of quantitative and qualitative approaches, withholding the comparison to mixed methods approaches until the end of the section. The reason for beginning with these two established approaches is that by understanding their separate strengths is important to then understand the benefits of combining their strengths. Table 4 provides a summary of the essential features of both quantitative and qualitative approaches and core underlying assumptions for each.

Historically, quantitative methodologies have dominated health research.<sup>149</sup> Quantitative approaches are based in positivism (or post-positivism), or the belief in a single reality accessible through scientific procedure.<sup>150</sup> Research motivated by the positivist worldview is systematic and places considerable value on ‘rationality, objectivity, prediction and control’.<sup>151</sup> Typically, researchers attempt to study a phenomenon from a neutral, detached standpoint and avoid human bias whenever possible, utilising standardised questionnaires and measuring tools to measure observed variables.<sup>152</sup> In the context of health research, one key part of quantitative research is the ability to measure and evaluate phenomena and relate them mathematically to other observable phenomena. Both reliability and generalisability are particular strengths of quantitative research, these cannot always be judged so easily within a qualitative study.<sup>153</sup>



**Table 4 - Comparing quantitative and qualitative approaches**

	<b>Quantitative Research</b>	<b>Qualitative Research</b>
<b>Theoretical stance</b>	<ul style="list-style-type: none"> <li>- Positivism/Post-positivism</li> <li>- Proposes that there are universal 'truths' that scientific enquiry can uncover.</li> <li>- Objective. Data and its interpretation is, as far as possible, value free</li> </ul>	<ul style="list-style-type: none"> <li>- Constructivism/Interpretivism</li> <li>- No single reality or truth.</li> <li>- Subjective. Researcher interpretation and reflexivity is a feature of the analysis.</li> </ul>
<b>Researchers position</b>	<ul style="list-style-type: none"> <li>- Researcher is detached and distant from the data.</li> </ul>	<ul style="list-style-type: none"> <li>- Researcher is involved, close to the data.</li> </ul>
<b>Study context</b>	<ul style="list-style-type: none"> <li>- Artificial settings (experimental). Researcher attempts to control for confounding factors.</li> <li>- Predetermined design.</li> </ul>	<ul style="list-style-type: none"> <li>- Natural settings. Researcher recognises and examines context and bias.</li> </ul>
<b>Data generated</b>	<ul style="list-style-type: none"> <li>- Usually numerical: generated through questionnaires, surveys etc.</li> <li>- Uses experimental and statistical controls.</li> <li>- Works across a larger number of cases.</li> <li>- Emphasises generalization and replication.</li> </ul>	<ul style="list-style-type: none"> <li>- Usually non numerical: generated through interviews, observations and document analysis etc.</li> <li>- Data are rich, detailed and involve fewer participants.</li> <li>- Tends to seek patterns but accommodates and explores difference and divergence within data.</li> </ul>
<b>Analysis</b>	<ul style="list-style-type: none"> <li>- Deductive, i.e. theory driven.</li> <li>- Test pre-existing hypotheses and theory.</li> </ul>	<ul style="list-style-type: none"> <li>- Inductive, i.e. data driven.</li> <li>- Findings are exploratory and form hypotheses and theory.</li> </ul>
<b>Reporting</b>	<ul style="list-style-type: none"> <li>- Findings supported by evidence of numerical data and statistical analyses.</li> </ul>	<ul style="list-style-type: none"> <li>- Findings supported by evidence of textual data.</li> </ul>

Qualitative methodologies offer alternate ways of exploring human behaviour. Towards the end of the 20th century, interest in qualitative research increased, especially within health research.<sup>154-156</sup> Qualitative research is an approach of naturalistic investigation to understand the meaning individuals or groups assign to a social or human problem.<sup>157, 158</sup> Unlike quantitative research, which is deductive and analyses phenomena in terms of trends and frequencies, qualitative research pursues to understand the meaning of a phenomenon through description, with emphasis on the meaning, feelings, experiences and views of the participants.<sup>159</sup> Instead of using a quantitative measuring device, qualitative researchers often collect data by conducting observations, in-depth interviews and by keeping reflective field notes to help with data interpretation and theory generation.

Qualitative approaches are often associated with a constructivism worldview. This refers to the notion that different experiences construct and form the different perspectives and behaviour of individuals.<sup>159</sup> The researcher's purpose is to make sense of participants' meanings of the world, acknowledging that their own personal backgrounds may shape their interpretation. Rather than starting with a theory (as in positivism/post-positivism), researchers generate or inductively develop themes and/or theories from these meanings.<sup>159</sup>

Qualitative research is rich, diverse and complex. It can aim to do many things, including: giving a 'voice' to a group of people or an issue; provide an in-depth description of experiences; develop theory; and interrogate meanings in texts and discourse.<sup>158</sup> Qualitative approaches have a wide range of uses in health research, often used in research detailing the experiences of chronic illness. Qualitative approaches have been less associated with treatment outcomes, as testing of causal variables requires a more positivist view of science, associated with quantitative methodology. This being said, qualitative approaches were not designed to test causal hypothesis but instead provide complex, in-depth data.<sup>157</sup>

Even though there are numerous methods attributed to both quantitative (i.e. experimental, observational) and qualitative (i.e. ethnography, phenomenology) research, often one strategy of investigation is not adequate to investigate a complex research problem. In the past four decades discussions of the limitations of qualitative and quantitative methodologies led to the development of mixed methods research, named the third methodological movement.<sup>160</sup>

Mixed methods research has been established for more than 50 years as a methodological approach in the social and behavioural sciences and is now well recognised and commonly utilised in health sciences.<sup>161, 162</sup> An article published in 1959 by Campbell and Fiske is often recognised as formally introducing the practice of mixed research methods.<sup>159, 163</sup> Their work described multiple methods to study psychological traits. Early thoughts about the value of mixed methods resided in the idea that all methods had bias and limitations, and the collection of both quantitative and qualitative data counteracted the limitations of each form of data.<sup>159</sup>

Mixed methods has been described as both old and new, with a long history in social science, but with a new set of terminology and methods employed by a new and growing community of researchers, including health researchers.<sup>161</sup> It was argued that there was a need to develop new methodologies to improve the quality and scientific power of data in health research.<sup>164</sup> The proportion of studies classified as mixed methods in English health services research has risen from 17% in the mid-1990s to 30% in the early 2000s.<sup>165</sup> In some areas of health research, namely those that are dominant with quantitative approaches

(i.e. clinical drug trials), qualitative methods had previously been dismissed as “poor science” until it was shown that they could be used in combination with quantitative components of clinical research to further understand the subtleties of the application of health interventions.<sup>166</sup>

### 3.2 Rationale for the choice of a mixed methods approach

There are many rationales for why researchers combine quantitative and qualitative research<sup>167</sup> Bryman (2006) identified 16 rationales to why researchers choose to utilise a mixed method approach (see Table 5).<sup>167</sup> In many mixed methods studies numerous reasons for using mixed methods may be chosen and often new reasons may emerge as the study progresses.

**Table 5- Reasons for using mixed methods, Source: Bryman (2006)<sup>167</sup>**

1.	<b>Triangulation or greater validity</b>	Refers to the traditional view that quantitative and qualitative research might be combined to triangulate.
2.	<b>Offset</b>	Refers to the suggestion that the research methods associated with both quantitative and qualitative research have their own strengths and weaknesses so that combining them allows the researcher to offset their weaknesses to draw on the strengths of both.
3.	<b>Completeness</b>	Refers to the notion that the researcher can bring together a more comprehensive account of the area of enquiry in which he or she is interested if both quantitative and qualitative research are employed.
4.	<b>Process</b>	Quantitative research provides an account of structures in social life but qualitative research provides sense of process.
5.	<b>Different research questions</b>	This is the argument that quantitative and qualitative research can each answer different research questions but this item was coded only if authors explicitly stated that they were doing this.
6.	<b>Explanation</b>	One is used to help explain findings generated by the other.
7.	<b>Unexpected results</b>	Refers to the suggestion that quantitative and qualitative research can be fruitfully combined when one generates surprising results that can be understood by employing the other.

8.	<b>Instrument development</b>	Refers to contexts in which qualitative research is employed to develop questionnaire and scale items – for example, so that better wording or more comprehensive closed answers can be generated.
9.	<b>Sampling</b>	Refers to situations in which one approach is used to facilitate the sampling of respondents or cases.
10.	<b>Credibility</b>	Refers to suggestions that employing both approaches enhances the integrity of findings.
11.	<b>Context</b>	Refers to cases in which the combination is rationalised in terms of qualitative research providing contextual understanding coupled with either generalisable, externally valid findings or broad relationships among variables uncovered through a survey.
12.	<b>Illustration</b>	Refers to the use of qualitative data to illustrate quantitative findings, often referred to as putting ‘meat on the bones’ of ‘dry’ quantitative findings.
13.	<b>Utility or improving the usefulness of findings</b>	Refers to a suggestion, which is more likely to be prominent among articles with an applied focus, that combining the two approaches will be more useful to practitioners and others.
14.	<b>Confirm and discover</b>	This entails using qualitative data to generate hypotheses and using quantitative research to test them within a single project.
15.	<b>Diversity of views</b>	This includes two slightly different rationales – namely, combining researchers’ and participants’ perspectives through quantitative and qualitative research respectively, and uncovering relationships between variables through quantitative research while also revealing meanings among research participants through qualitative research.
16.	<b>Enhancement or building upon quantitative/qualitative findings</b>	This entails a reference to making more of or augmenting either quantitative or qualitative findings by gathering data using a qualitative or quantitative research approach.

In this thesis, the key reasoning behind combining methods was “*completeness*” and to “*answer different research questions*”. Completeness refers to the complementary strengths of quantitative and qualitative approaches addressing complex research questions more comprehensively than using either method alone.<sup>148</sup> Hence, the gaps left by one method

(e.g. a quantitative one) can be filled by another (e.g. a qualitative one).<sup>153</sup> Each method has its strengths and weaknesses, therefore by using both qualitative and quantitative approaches, a fuller picture and a more complete understanding can be gained.<sup>148, 163</sup> This is of particular interest in health research due to the complex nature of health and illness.<sup>168</sup> It was anticipated in regards to this study, that the quantitative results would yield general trends and relationships, while qualitative results would provide in-depth personal perspectives of individual experiences. It was predicted that the qualitative approach would also allow participants to highlight and reference their own issues/needs, rather than having them pre-framed by the researcher (e.g. items in a survey); allowing unanticipated ideas and themes to be expressed.<sup>158</sup> Therefore, by combining both approaches a more complete understanding is provided than either data collection alone.

Another rationale for using a mixed methods approach was “*utility or improving the usefulness of findings*”. This refers to a suggestion that by combining both quantitative and qualitative approaches the findings will be more useful to clinical professionals.<sup>167</sup> In this research, through planning a mixed methods design, both numerical and narrative data would be produced. Quantitative data would present key figures and statistics. It was forecast that the quantitative results would help inform clinical professionals of the key issues and supportive care needs of TYA survivors and their caregivers. Equally, the qualitative data could provide health professions with deep insight into survivor and caregiver views and their experiences of long-term survivorship. In the NHS qualitative data is now widely used to inform quality improvements, design and delivery of services and quality policies.<sup>154, 169</sup>

### **3.2.1 Challenges in choosing mixed methods research**

Despite its considerable strengths as an approach, mixed methods research can present researchers with challenges.<sup>148, 153, 170</sup>

Firstly, combining two methods in one study is time consuming and requires knowledge and skills in both quantitative and qualitative methods. In practice this often means that it is preferable to be carried out by a team, with individuals from different disciplines, to utilise their abilities.<sup>171</sup> As this was a PhD study, I was a lone researcher with a large amount of data collection and analysis to carry out. Therefore, to manage this problem the following decisions were made: 1) the study timeline was designed to allow sufficient allocated time for data analysis and integration of both quantitative and qualitative data 2) specific qualitative, quantitative and mixed methods training was undertaken throughout the study

and 3) support from PhD supervisors and other experienced colleagues was readily available and sought when needed.

### ***Reflections, learning and decision making...***

On reflection I believe that utilising a mixed methods approach enabled me to learn and experience different methods and analysis techniques that will be invaluable in my future career. I was pushed out of my comfort zone as my previous research experience was mainly qualitative in nature, but in this PhD I gained more experiences in quantitative design and analysis.

Not only can it be technically and practically challenging to effectively carry out mixed methods research,<sup>168</sup> but quantitative and qualitative research stem from different theoretical perspectives (as discussed in 3.2). Some researchers argue that it is neither possible nor desirable to combine quantitative and qualitative methods in a study as they signify essentially different and conflicting ways of viewing the world and how we collect information about it.<sup>153, 172</sup> However by adapting a pragmatic view (discussed in more detail in 3.3.), I believe that concerns about the incompatibility of worldviews can be dismissed as the combination of quantitative and qualitative methods was the most suitable way of answering the research questions effectively. This pragmatic view informs much applied mixed methods research in health services or policy.<sup>173</sup>

Finally, reporting mixed methods research has been documented as being a significant challenge.<sup>160, 170</sup> Given that articles, reports and theses are the vehicle for disseminating study findings, it is important that the reporting is fit for purpose.<sup>166</sup> Bryman (2007)<sup>170</sup> states:

*“Mixed methods research is not necessarily just an exercise in testing findings against each other. Instead it is about forging an overall or negotiated account of the findings that brings together both components of the conversation or debate.”* (p.21)

There are many ways in which mixed method data can be presented, but there is no template of how best to report mixed methods studies, like those available with quantitative and qualitative studies. Therefore, special attention needs to be given to the presentation and format used to present a mixed methods study.<sup>166</sup>

### 3.3 Philosophical worldview in mixed methods

Researchers make philosophical assumptions to guide their study at a broad level. These assumptions generally consist of a basic set of beliefs, values and available techniques.<sup>174</sup> As previously mentioned the philosophical challenges around mixed methods research stem from the fact that quantitative approaches are usually associated with positivist or post-positivist epistemologies, whilst qualitative approaches are traditionally connected with constructionist or interpretive epistemologies.<sup>160 175</sup> In the past, supporters of qualitative and quantitative approaches were entangled in a debate about the incompatibility between the two, known as the 'paradigm wars'.<sup>160, 176</sup>

Given the two different philosophical worldviews (also known as paradigms), the challenge for mixed methods researchers is how these two worldviews can coexist. In the early years of mixed methods research there was an argument whether it could actually be called a 'paradigm' with its own philosophical foundations, as opposed to simply a mix of methods from the 'opposing paradigms'.<sup>148</sup> The majority of mixed methods researchers focused on the research questions they were investigating rather than the complex philosophical orientations surrounding the chosen methodology of their study.<sup>177</sup> However, the problem remained as to how two diverse worldviews could coexist, as being suggested in mixed methods research. One answer that some mixed methods researchers provided is the use of several paradigms within one study. Others (whom I align myself with) have argued in favour of a single paradigm stance, such as pragmatism or critical realism. Within a single paradigm stance, the underlying philosophy informs both quantitative and qualitative data collection.<sup>148</sup> Making choices of which paradigm to choose depends on which one is suited best to the particular mixed methods project. In this PhD it was decided that the pragmatic approach was the most appropriate, for reasons discussed in 3.3.1.

#### 3.3.1 A pragmatic approach

Many mixed method researchers adhere to pragmatism.<sup>146</sup> A pragmatic approach, accepts that there are both singular and multiple realities, and looks to bring together quantitative and qualitative methodology to best answer real world questions and address real world problems.<sup>146, 178</sup>

Pragmatism offers an increasingly popular approach to the philosophical challenges of mixed methods research.<sup>146, 160</sup> Pragmatism as an mixed methods approach originated in the United States in the late nineteenth century.<sup>152</sup> Originating from its root word - pragma, a Greek word for "*action*", it points to knowledge coming from taking action and learning from

the actions.<sup>152</sup> A pragmatic perspective employs a “what works” mind-set, giving primacy to the importance of the research problem and question.<sup>146</sup> An advantage of utilising pragmatism is that it is not committed to any one system of philosophy and reality, but instead provides the researcher with a freedom to select the methodology and methods most appropriate to answer the research questions and fulfil the aims of the study.<sup>179</sup>

Table 6 is a framework produced by Morgan (2007) for understanding how a pragmatic approach can work and how it differs from the singular use of quantitative or qualitative methods.<sup>146</sup> The framework looks at three aspects of the different approaches: the connection of theory and data, the relationship to the research process and the applicability of the data gathered. Firstly, the distinction between induction and deduction, Morgan recognises that in practice no research is conducted in a purely inductive or deductive fashion and that research is unlikely to be purely theory or data driven.<sup>146</sup> The abductive approach adopted in pragmatism reasons that in real life the researcher moves back and forth between induction and deduction. Secondly, Morgan describes the traditional differences in quantitative and qualitative research in subjectivity and objectivity as essentially artificial in practical research. The concept of being completely subjective or objective does not seem to apply to conducting real-life research and instead that researchers “*work back and forth between various frames of reference*” (p71),<sup>146</sup> the pragmatic term intersubjectivity adequately represents this duality. The final difference put forward in Table 6 is the applicability of the research to other situations. In this instance, the pragmatic approach once again rejects the need to choose from the extremes where research results are either totally specific to a particular context or universal and generalisable.<sup>146</sup> Transferability is the desire to find ways of appropriately applying what is learned in one situation to benefit and inform another. Once again, this includes a process of working back and forth, in this instance between specific results and their more general implications.<sup>146</sup>

**Table 6 - A pragmatic approach, adapted from Morgan (2007)<sup>146</sup>**

	<b>Qualitative approach</b>	<b>Quantitative approach</b>	<b>Pragmatic approach</b>
<b>Connection of theory and data</b>	Induction	Deduction	Abduction
<b>Relationship to research process</b>	Subjectivity	Objectivity	Intersubjectivity
<b>Applicability of data</b>	Context	Generality	Transferability



As previously stated, it is important when selecting a methodology that the researcher determines their epistemological position in order to be clear about the objectives of their research. In this mixed methods study a pragmatic approach was adopted for the reasons described above, and to arrive at outcomes that can be useful in understanding and advancing the fields of TYA brain tumour supportive care and research.

### 3.4 Mixed methods design

As with either quantitative or qualitative studies, research designs are key in mixed methods research, as they contribute and provide logic to decisions made throughout the study- how data is collected, analysed and interpreted.<sup>162</sup> There are essentially three basic designs at the centre of all mixed method studies:<sup>148</sup>

- 1) *An exploratory sequential design* – this design begins with a qualitative data collection and analysis phase, which shapes the subsequent quantitative phase.
- 2) *An explanatory sequential design* – differing from an exploratory design, this design begins with a quantitative data collection and analysis phase, which informs the follow-up qualitative phase.
- 3) *A convergent design (also known as concurrent)* - involves both quantitative and qualitative data collection and analysis at similar times, followed by an integrated analysis. In this design the researcher normally gives equal priority to both quantitative and qualitative data.<sup>162</sup>

Researchers should carefully select a design that best matches the research questions and purpose.<sup>162</sup> A suitable design makes the study manageable and simple to implement. Each of these three designs have particular strengths and limitations, and each has a different design purpose and mixing strategy. The mixed methods study in this thesis uses a convergent design.

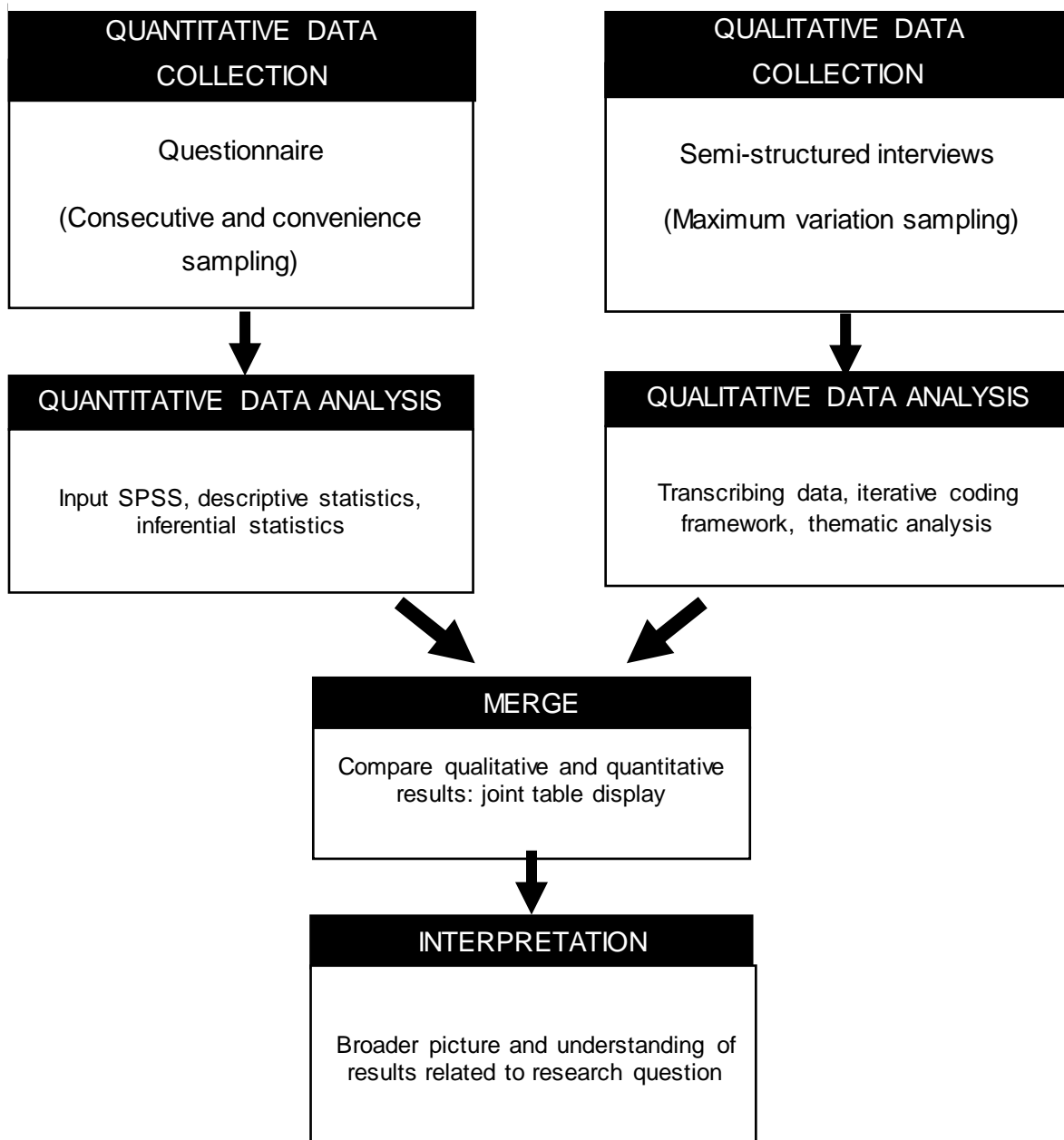
The convergent mixed methods approach is perhaps the most familiar of mixed methods strategies.<sup>148</sup> The main purpose of the convergent design is “*to obtain different but complementary data on the same topic*” (p.122)<sup>180</sup> to best understand the research problem. Qualitative and quantitative methods are utilised to measure overlapping but also different aspects of a phenomenon, enabling an enriched understanding of that phenomenon.<sup>148, 162,</sup>

181

Both the research questions and the data required were the main determinants of the design used. Pragmatically the choice of design was influenced by feasibility, and time constraints

of a PhD study. A strength of a convergent design is that it is an efficient design because the two types of data are collected at the same time. This was important as this was a PhD study with limited time and resources. Furthermore, the assumptions of pragmatism (as discussed in the previous section) are well suited for guiding the work of merging the two approaches into a larger understanding. Figure 6 presents a diagram of the convergent design.

**Figure 6- Convergent mixed methods design (adapted from Creswell and Plano Clark, 2011)**



The basic procedures for using this design are as follows:<sup>148</sup>

- 1) **Data collection.** Quantitative data and qualitative data are collected concurrently but separately — that is, one does not depend on the results of the other. They also typically have equal importance for addressing the study's research questions.
- 2) **Data analysis.** The two data sets are analysed separately and independently from each other using typical quantitative and qualitative analytic procedures.
- 3) **Merge results.** Once both sets of initial results are analysed, the next step is to merge the results of the two data sets. This merging step may include directly comparing the separate results or transforming results to help relate the two data types during additional analysis.
- 4) **Interpretation.** After the results have been merged, both sets of results can be interpreted. Looking for convergence, divergence and relationships of two sources of data. This procedure also includes recommendations for practitioners, limitations and suggestions for further research.

As illustrated in Figure 6, the convergent mixed method parallel design utilised in this study involves a combination of both quantitative and qualitative approaches. Quantitative data addressed objectives 1 - 4 (see 2.7) and qualitative data addressed objectives 1 and 4. Both quantitative and qualitative data are given an equal weighting in this study. The combined results were intended to address the overall aim and provide a more complete and in-depth understanding of the supportive care needs of teenage and young adult childhood brain tumour survivors and their caregivers.

## Chapter 4: Mixed methods study - design, recruitment and analysis

Following the aim and objectives detailed at the end of Chapter 2, a mixed methods approach and convergent mixed methods design was selected and justified (Chapter 3). The mixed methods design combines both a quantitative phase (utilising a survey), and a qualitative phase (semi-structured interviews). This Chapter presents the methods that were used within these two phases, starting with a brief overview of the research design (4.1). Then a description of the process of obtaining ethical and research governance approvals for this study, and details of the procedures that were involved in ensuring the ethical conduct of this study are presented (4.2). The following sections describe the detailed methods used in conducting the quantitative (4.3) and qualitative phases (4.4.) of this study. In each section, the participant sample, recruitment, data collection and data analysis are explained.

### 4.1 Brief overview of research design

*Below is a summary to help place the rest of the Chapter in perspective, more specific details and explanations for the choice of methods follow in the rest of the chapter.*

- The study used a convergent (cross-sectional) mixed methods design including both quantitative data (survey) and qualitative data (semi-structured interviews).
- The overall aim of the study was to gain an in-depth understanding of the supportive care needs of TYA childhood brain tumour survivors and their caregivers.
- The quantitative and qualitative strands addressed specific research objectives under this aim.
- Survivors were eligible if currently aged 13-30, diagnosed before 14 years of age and at least five years from diagnosis. Caregivers were eligible if identified as a primary caregiver of the survivor.
- Participants were recruited both from long-term follow-up clinics (in three NHS Trusts) and online.
- Consenting survivors and caregivers each answered a specific survey. The survey included several validated questionnaires, to assess (unmet) needs; QoL and support service use.
- A sub-group of survivors and caregivers who took part in the survey were asked to partake in semi-structured, face-to-face interviews.

## **4.2 Ethics**

When beginning a research study, any ethical issues that may arise during fieldwork must be considered to ensure the dignity, rights, safety and wellbeing of participants and researchers.<sup>182</sup> These issues were considered as an ongoing and reflexive part of the research process and not just at the study design phase. All aspects of the study was undertaken in accordance with the MRC Good Research Practice guidelines, Good Clinical Practice (GCP) guidelines and the Data Protection Act (2018).

### **4.2.1 Ethical considerations**

The main ethical issues considered in this study were; obtaining informed consent, protection from participant harm and burden, protecting participant anonymity and involving young people in the research. These issues and the minimisation of these issues are covered in the following sections.

#### **Obtaining informed consent**

Informed consent was obtained from all participants prior to any study specific procedures. The right of the participant to refuse participation without giving reasons was always respected. As the study comprised of two phases, two separate consent forms for the quantitative (survey) and qualitative (semi-structured interviews) phases were developed. The interview participants were selected from questionnaire participants who had indicated an interest in participating in interviews.

When obtaining informed consent, the age of the participant is important. Survivors under 16 years of age needed to assent to participating, but also required the consent of a parent or legal guardian. Those 16 years old and above were able to consent themselves. In both cases, the researcher's signature was also required.

Obtaining consent from any young person is a difficult issue. To obtain informed consent, participants must be able to fully understand the information about the aims and methods of the study and what their involvement will entail. It was anticipated prior to recruitment that this may be even more challenging as many survivors would have impaired cognitive functioning as a result of the tumour or its treatment, which may make reading and understanding harder. With this in mind all of the survivor participant information sheets were designed to be simple (reading age of 7) and were purposefully kept clear, simple and short (see appendix 5 for an example).

### ***Reflections, learning and decision making...***

In practice there were other issues that also meant reading the written information was difficult for survivors— including sight problems, poor concentration and physical disabilities (e.g not being able to hold the paper). Therefore, in these cases I always went through the information sheet and assent/consent form with the survivor and extensive verbal information was provided appropriate to their level of understanding.

Every effort was made to ensure participants felt comfortable in the study and no consent issues were experienced throughout the study. As far as it was possible to know, no participants remained in the study against their will or felt that they could not withdraw.

### **Protection from harm and burden**

It was anticipated that the main risk to participants was the potential upset/distress caused by the discussion of their or their loved one' brain tumour experiences. It was foreseen that the survey or interviews may provoke an emotional reaction (e.g., sadness) in the participating survivors or caregivers. Therefore, the following decisions were made. To begin, before any young survivor was approached with information about the study, a clinical member of staff at the centre where they were receiving long-term follow-up care checked the suitability of potential participants. This suitability was based on their knowledge of the individual (fitting the inclusion criteria detailed in 4.3.2). If they were deemed eligible by a clinical staff member, they would then approach the potential participant and ask their permission for the researcher to talk to them about the study. The clinical staff also acted as a gatekeeper ensuring that those eligible were not so cognitively impaired that they were unlikely to be able to complete study procedures. Only one survivor with severe cognitive deficits (which clinical staff believed would prevent successful completion of study procedures) was not approached.

Regarding the survey, it was made clear to all potential participants that they could choose not to answer certain questions if they do not wish to. A statement was also added at the end of the survey that directed participants to an appropriate health professional if they had any problems or concerns after completing the survey.

In the interviews it was almost inevitable that participants would discuss experiences and emotions that might be difficult or upsetting. As such, before each interview participants were reminded that they were not obliged to respond to any interview questions that they did not feel comfortable answering. They were also encouraged to let myself (the interviewer) know if they wanted to stop the interview at any time or take a break.

The study was designed to reduce participation burden. The interviews were all held at the most suitable place for the participants and the majority lasted less than an hour per interview. Similarly, the survey instruments were carefully designed and purposefully chosen to be simple to follow, in an accessible format (available online and on paper) and not too time consuming (under 30 minutes). These decisions were made in consultation with the study Patient and Public Involvement group.

### ***Reflections, learning and decision making...***

Early on in the PhD I decided that a Patient and Public Involvement (PPI) group would be beneficial to help make key decision throughout the PhD and to ensure that the research remained patient-centred.

I advertised for PPI members through the national brain tumour charity - *brainstrust*. They helped share an advert to families in the region and asked people to get in touch if they were interested in taking part. The final PPI group consisted of three parent caregivers of childhood brain tumour survivors and one childhood brain tumour survivor aged 15 years old at the beginning of the study. During the course of the PhD project the PPI representatives provided feedback (in face-to-face meetings and via email) on:

- Study aims
- Study methods (i.e. best way to collect data, participant time commitment, recruitment)
- Study materials (i.e. checking the clarity of language used in information sheets/consent forms)
- Questionnaire/interview design (i.e. content, questionnaires chosen)

The survivor/patient PPI representative helped to make sure the study information/design were appropriate for this age and patient group. Caregiver representatives ensured that any concerns about the research from a caregiver viewpoint were considered. For example, what information they would want to know if their loved one was taking part in the research or what would their concerns be about their loved one taking part (especially if they were under 16 years of age).

## **Protecting participant anonymity**

A core ethical consideration during this process was to keep the data collected from the participants anonymised and confidential.<sup>183</sup> A number of key steps were taken to ensure the confidentiality of the participants and data protection. All the information collected during the study concerning individual participants was treated in the strictest confidence. All data were treated in accordance to the Data Protection Act (2018) and the University of Leeds Information Protection Policy (Version 1.2). The separation between identifiable data and anonymised data occurred at the point of study entry with each participant being allocated a study ID number. The document listing the code link, linking study ID number to participant identity, was stored within a separate password protected file and only available to myself and the PhD supervisors. All completed consent forms, questionnaires and other paper-based data were kept in a locked filing cabinet in a secure office at St James Hospital.

Another ethical concern was the potential for participants to be identified through publication or presentation of their interview data. This was addressed by ensuring to pseudonymise interview extracts and the removal of any personal identifiable data presented in any publications or presentations.

## **Research methods involving children and young people**

As previously discussed in Chapter 1 (1.2.3), there has been a move towards research with rather than on or about young people, and thus an increasing recognition that TYAs need an opportunity to express their opinions on matters which affect them directly. This was highly relevant in this research as this age group needed to be represented. However, to obtain valid TYA-led data, it was important that age-appropriate methods were chosen. Essentially, it was finding a balance between recognising that young people are different from adults, without patronising them. Careful consideration was given to ensure that the research methods and materials used were suitable for young people. The TYA PPI representative helped to ensure study design, methods and materials were age appropriate for this patient group.

### **4.2.2 Ethical approvals**

As this study involved NHS patients and their data, ethical approval to conduct the study was sought from the Yorkshire and the Humber – Bradford Leeds Research Ethics Committee. The University of Leeds acted as the sponsor for this study. The ethics application was submitted on 14th July 2018 and a favourable opinion was given was received on the 26th



September 2018 (REC reference: 18/YH/0312, see appendix 3). A letter of access was granted by Leeds Teaching Hospitals NHS Trust on the 17th October 2018.

During the course of the research I applied for approvals to add two additional NHS recruitment sites and one substantial amendment. A favourable opinion to add additional recruitment sites was received on 21<sup>st</sup> March 2019 (Sheffield Teaching Hospitals NHS Foundation Trust) and 21<sup>st</sup> May 2019 (Sheffield Children's NHS Foundation Trust). A letter of access was granted by Sheffield Teaching Hospitals NHS Foundation Trust on the 24<sup>th</sup> May 2019 and Sheffield Children's NHS Foundation Trust on the 20<sup>th</sup> August 2019.

The substantial amendment involved advertising the survey online with help from brain tumour charities (through their websites, email lists, online bulletins and social media platforms). The purpose of advertising the survey this way was in the hope it would reach a larger group of brain tumour survivors and caregivers, increasing the sample size. A favourable opinion to this recruitment strategy was given on the 10<sup>th</sup> June 2019 (see appendix 4). The survey went live online on the 12<sup>th</sup> June 2019.

### **4.3 Quantitative data: survey**

The following section describes the quantitative data collection strand of the mixed methods study. The quantitative data aimed to address the following objectives:

- 1) To describe unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers.
- 2) To explore if survivor sociodemographic (i.e. sex, age) and clinical data (i.e. treatment) are related to unmet supportive care needs.
- 3) To determine whether unmet supportive care needs are associated with QoL outcomes.
- 4) To explore the role and perceived use of support services in TYA survivors and their caregivers.

The choice of quantitative method was determined by the study aims, methodological considerations (discussed in Chapter 3), the study population, the potential size of available sample, time available and resource constraints. Taking these into consideration it was decided that a survey-based method best suited the aims of the project. The Strengthening the Reporting of Observational studies in Epidemiology (STROBE) guidelines for reporting a cross-sectional study was used for completeness.<sup>184</sup>

Surveys are one of the most common instruments for gathering data.<sup>153</sup> In health research they are vital tools, allowing exploration of a large number of variables and facilitating investigating relationships among them. They can also be an effective means of measuring the behaviour, attitudes, preferences and opinions of a study population. Surveys can provide outcomes that are descriptive, explanatory, and/or explorative.<sup>185</sup>

Until recent years mail or postal surveys had been the most common type administered, but with the rise of the Internet, online survey have become increasingly popular.<sup>153, 186</sup> For this study, the survey was primarily designed as an online version. This decision was guided by the PPI advisory group, who believed that participants, especially TYA participants would prefer to complete the survey online. Alongside the online version, paper surveys were always offered to participants who needed/wanted them. All participants were also given the option to have help (from the researcher) to complete the survey in person or over the phone.

There are many advantages to using surveys in research. Firstly, surveys are convenient for participants, because they can complete them when they want and at the speed that they want to go.<sup>153</sup> The latter point was really important in this research as many TYA survivors experience neurocognitive impairment (as discussed in Chapter 2), which meant that it could take them longer to complete the survey. Therefore, the online version of the survey was designed to accommodate for this by having no 'time-out', this meant that there was no error message or disconnection if the participant took a long time to respond to a question. As recommended by the PPI advisory group, the participants were also able to save the survey and return to it after a break if necessary. Furthermore, it is widely acknowledged that survey data collection has the following advantages: less frequent nonresponse and "don't know" answers, improved data quality (validation checks prompt participants when they enter implausible or incomplete answers), and less researcher data entry errors.<sup>187</sup>

Surveys can also be seen as advantageous when asking personal or sensitive questions. This was an important factor in this research as the survey consisted of personal questions, such as sexual relationships. Additionally, surveys are specifically deemed a useful tool for discovering the views of TYAs, as there is less risk of social-desirability bias than other methods (e.g. face-to-face focus groups).<sup>188</sup>

Finally, surveys are often more cost effective and less resource dependent than qualitative methods, meaning that data can be collected from a larger group of participants than what may be feasible by using a qualitative approached alone.

Surveys, like other data collection tools, also have certain disadvantages. The first disadvantage which was carefully considered, was that a survey format may not be suitable

for populations with poor literacy or visual impairment. In Chapter 2, the systematic review highlighted that TYA survivors experience impaired cognitive functioning and/or poor eyesight that can affect their reading ability. To help resolve this potential issue, every participant was offered several ways to complete the survey, it was not just available online.

### ***Reflections, learning and decision making...***

In practice the way in which participants wanted to fill in the survey varied. Some participants preferred to fill in the survey online, as they could enlarge the writing on the screen or because using an electronic device was easier if they were unable to write very well. Some preferred to use paper, as they could see the text easier. And some TYAs needed help to complete the survey from their caregiver or myself. Equally caregivers differed in how they preferred to complete the survey— some liked the convenience of doing it online and others favoured to complete it on paper.

One disadvantage of using surveys, especially with young people is the concern that other people have filled in the survey for the survivor. Numerous survivors chose to have help from someone else to fill in the survey. I was aware that sometimes the caregiver may try answer for the survivor. To try ensure that it was actually the survivor's voice and thoughts that were captured, I would try to emphasise to the participant that they should try answer the questions by themselves and also explain to the caregiver that help with answers should be kept to a minimum.

#### **4.3.1 Research setting**

The context within which a study takes place is a key influence both on the results obtained, and on the understanding of those results. The next paragraph will describe the research settings in detail.

Participants were recruited via two main strategies: through follow-up clinics and online. Participants were recruited from Leeds Teaching Hospitals NHS Trust, Sheffield Teaching Hospitals NHS Foundation Trust and Sheffield Children's NHS Foundation Trust. Recruitment began in Leeds in October 2018, in Sheffield Teaching Hospitals NHS Foundation Trust in June 2019 and Sheffield Children's NHS Foundation Trust in October 2019 and ended in all sites in February 2020.

Leeds Cancer Centre is one of the largest centres in the UK, treating cancer patients in Leeds and the Yorkshire region. Participants were recruited from the long-term follow-up service, which is offered specifically to people who have finished their cancer treatments and do not need close monitoring for disease recurrence; this is usually from about 5 years after finishing treatment. Patients under age 18 years are treated in the Children's Oncology and Haematology Day Unit at Leeds General Infirmary. Patients aged 18 years and over are cared for by the same team but in the Oncology Outpatients Suite in Bexley Wing at St James's University Hospital.

Sheffield Children's NHS Foundation Trust is the primary treatment centre for children and teenagers with cancer from within South Yorkshire, Lincolnshire and North Derbyshire. The Late Effects Service in Sheffield was one of the first to be established in the UK and provides ongoing care for survivors of childhood cancer. Patients under 18 years attend the Late Effects clinic at Sheffield Children's NHS Foundation Trust, those 18 and over are cared for in the Late Effects clinic at the Royal Hallamshire Hospital (Sheffield Teaching Hospitals NHS Foundation Trust).

In addition to participants being recruited from follow-up hospital clinics, the study survey was also advertised online (from 12<sup>th</sup> June 2019-31<sup>st</sup> January 2020) with help from brain tumour charities. Namely –The Brain Tumour Charity, *brainstrust*, and BTRS (Brain Tumour Research and Support across Yorkshire<sup>1</sup>). These charities provide support for brain tumour survivors and caregivers throughout the UK. Participants were recruited through a range of online channels, including: the charities' websites, email lists, online bulletins and social media platforms. This pragmatic decision was made in the hope that the survey would reach a larger group of brain tumour survivors and caregivers, increasing the sample size.

#### **4.3.2 Participant Sample**

The sampling methods for quantitative and qualitative approaches differ due to their distinctive aims.<sup>189</sup> In quantitative approaches the aim is usually to select a sample representative of the target population so that inferences can be made about this population.<sup>189, 190</sup> The sample strategy and size for both the quantitative method (survey) and qualitative method (interviews) are discussed separately in the following sections.

Sampling is the process of selecting a proportion of the population, when, as in this study, studying the whole population is not practical.<sup>161</sup> In health research, the population is defined as a group of people who share a common character or a condition, usually their diagnosis.

---

<sup>1</sup> The BTRS charity has since been renamed to Yorkshire's Brain Tumour Charity.

In this case the population is TYA brain tumour survivors and their caregivers. There are two common types of sampling designs: random sampling and non-random sampling. Random sampling encompasses random selection, meaning everyone in a population has an equal chance of being chosen for inclusion in the study.<sup>190</sup> Researchers can employ a random sampling design to identify a representative sample in order to achieve generalisability about the entire population.<sup>153</sup> An advantage of random sampling is that it also minimises both sampling and selection bias, arguably making it more rigorous.<sup>153, 190</sup> There are numerous random sampling designs, the main designs include: simple random sampling, systematic sampling, stratified sampling and cluster sampling.<sup>153, 190</sup>

Unlike random sampling, non-random sampling selects individuals in a non-systematic process that does not guarantee equal chances for each subject in the target population.<sup>190</sup> This type of sampling design is less likely to produce a representative sample of the whole population.<sup>153, 189</sup> Despite this, most research samples in health research are non-random samples.<sup>189</sup> This is because many research circumstances mean randomisation is impossible; for example, when the number of individuals in a population is unknown or they cannot be identified before the study is conducted, or researchers have problems in randomly recruiting samples due to time and resource restrictions. The most commonly used non-random designs include: convenience sampling, consecutive sampling, purposive sampling and snowball sampling.

The ideal sampling for minimising bias would have been to use a frame to select a random sample<sup>161, 191</sup> but this would have been unlikely to yield a sufficient sample size given study resources and time restrictions. Therefore, both consecutive and convenience sampling was used in this study. Participants recruited in the long-term follow-up clinics were recruited by consecutive sampling. A consecutive sample involves enrolling every participant who meets the eligibility criteria during a specified time.<sup>192</sup> In this study, all eligible survivors and caregivers attending clinics were approached and informed about the study. Such a sample should be highly representative of the accessible population.<sup>192</sup> And as described earlier in this Chapter, an online convenience sample was also used to maximise recruitment. Convenience samples are common in health research and clinical studies.<sup>193</sup> In short, convenience sampling entails using the most conveniently eligible participants.

When determining a suitable sample size, it is important to consider the practicalities; for example, how feasible it is and how long it may take to recruit such a number of participants. As this was primarily a descriptive and explorative study, no a priori power calculation was necessary or possible. For the survey a sample of approximately 100 participants in both groups (survivors and caregivers) was desired and deemed sufficient to perform the planned

analyses. This was with the feasibility of arranging, conducting and analysing the data within the study time frame.

The end sample size was smaller than initially intended. In part, this was attributed to fewer eligible participants than anticipated attending the long-term follow-up clinics (i.e. patients did not attend), and a lower than initially planned completion rate of the survey following consent. For example, some participants consented to partake in the study in the clinic and took the survey instructions away with them but then did not complete the survey, even after a reminder was sent. Although I had aimed for 100 participants per group so that a larger number of variables could be explored in relation to supportive care needs (in the regression analysis), I adapted the number of variables analysed to still allow a robust analysis with a smaller sample size.

### **Inclusion/exclusion criteria**

Eligible participants in follow-up clinics were identified by either a consultant or clinical nurse specialist. They sought permission from the survivors and their caregiver for the researcher to speak to them about the study. Participants were deemed eligible if they met the inclusion criteria, see Table 7.

**Table 7 - Participant inclusion and exclusion criteria**

<b>Survivor inclusion criteria</b>	<b>Caregiver inclusion criteria</b>
<ul style="list-style-type: none"> <li>• A diagnosis of a primary brain tumour between 0-14 years old</li> <li>• &gt;5 years since diagnosis</li> <li>• Currently aged 13-30</li> <li>• Able and willing to give informed consent</li> <li>• Able to read and understand English</li> </ul>	<ul style="list-style-type: none"> <li>• A primary caregiver (defined by the survivor or themselves) of a primary brain tumour survivor currently aged 13-30, diagnosed between 0-14</li> <li>• Survivor &gt;5 years since diagnosis</li> <li>• Able and willing to give informed consent</li> <li>• Able to read and understand English</li> </ul>
<b>Survivor exclusion criteria</b>	<b>Caregiver exclusion criteria</b>
<ul style="list-style-type: none"> <li>• Cognitive deficits of a severity that would preclude successful completion of study consent or procedures.</li> </ul>	

As discussed in Chapter 1 (1.2.1) the age range for TYAs is variable. The age inclusion boundary for the TYA survivors in this study was set at 13-30. The lower age limit was chosen as in the United Kingdom, the lower limit for TYA supportive care is generally defined as 13 years of age, and services have been developed to cater for these requirements.<sup>34, 37</sup> The upper age limit was more difficult to set, particularly as the transition to adulthood is becoming prolonged.<sup>36</sup> For example, the age of first marriage is higher than in previous decades in post-war Britain. Therefore, the upper age boundary was set at 30 because it was felt that this was high enough to capture the transition events.

### ***Reflections, learning and decision making...***

The Patient and Public Involvement (PPI) group was consulted about the participant age range. The group felt that a starting age of 12-13 years old was acceptable. The group felt this age limit was the start of adolescence and also marked important milestones for young people, such as: starting high school and increasing importance of social interactions with peers.

The chosen upper age limit was also guided by the PPI group. From a discussion in the first meeting, they felt that even though some supportive services may have a limit of 24/25 (i.e. The Brain Tumour Charity), that it would be better to go up to 30 years old. They felt that teenage and young adulthood is more about a stage in life, than about age as a number. Highlighting that by including this age group the data should capture all major transition points, such as education and work (e.g. going to secondary school, College or University, starting work).

When the survey was advertised online, the inclusion and exclusion criteria still applied. However, the cognitive ability of participants could not be measured. Yet, it was presumed (and supported by previous research within the research group that I am based in) that if the survivor had the cognitive ability to read the study information, fill in the survey and submit the answers that they were cognitively able to understand and partake in the study.

### **4.3.3 Recruitment and data collection**

Recruitment and data collection with participants recruited in long-term follow-up clinics was carried out as follows. After an introduction from clinical staff, eligible participants were approached by the researcher who explained the study and relevant study materials (see appendix 5 for an example Participant Information Sheet). All participants were also given an

extra information sheet which describes in further detail data protection in compliance with new General Data Protection Regulation 2018 guidance. Survivors and caregivers had separate information sheets and consent forms. Participants under the age of 16 also had separate study materials with age appropriate information. Potential participants were given as much time as they needed to read the information and ask questions and if they wished to participate were asked to provide written consent. A stamped addressed envelope was supplied if the potential participant preferred to take the information home. The right of the participant to refuse to take part in the study, without giving reasons, was respected at all times.

The majority of survivors attended clinic with their primary caregiver, normally a parent. Hence, contact with both the survivor and caregiver was usually established at the first meeting. When survivors attended the clinic alone, they were asked to identify their primary caregiver and if they would give them the relevant study materials. Their caregiver could then contact the researcher if they would like to take part. In some cases, a survivor consented to partaking in the research, and the caregiver did not, or vice versa. In the latter case the patient's medical record data was not linked to caregivers' self-reported data. All clinical and sociodemographic information about the survivor was obtained through participant self-report, and matched against the medical notes. It is worth noting that this could only be done for the participants recruited in clinic and not those recruited online.

Once the participant had consented, they were able to complete the survey. The survey could be completed in three formats: online, on paper, or with help from the researcher. To complete the survey online, participants were given instructions and a unique username and password which allowed them to login to the QTool system and complete the survey anonymously. QTool is a web-based survey collection system, developed and used as part of the NIHR funded development programme for eRAPID (electronic patient self-Reporting of Adverse events: Patient Information and a Dvice).<sup>194</sup> All data collected with QTool is stored in protected databases. Data stored in QTool is anonymous - only linked through the unique username provided, and no patient identifiable data was held there.

As previously listed participants could also complete the survey on paper. Participants could complete the paper questionnaire in the long-term follow-up clinic or take it home with them to complete later, and post it back to the research group. Participants (mainly survivors) were also offered help to complete the survey, if they wanted it. They could have help completing the paper or online survey, in these cases the researcher would read the questions allowed to the participant.



If participants consented and did not complete the survey within 7 days they were sent a reminder text, email or letter. The choice of how the reminder was sent was chosen by the participant on the consent form.

In June 2019 the survey was advertised online via adverts placed by several brain tumour charities. Both the survivor and caregiver survey was without change except for two minor changes to the sociodemographic information. First, any personally identifying information was removed – we asked participants for their age instead of date of birth (question 2). We also added a question to ask participants to identify which country they lived in (question 3).

Each survey started with written information about the study. Participants were encouraged to contact the researcher with any concerns or queries. Before taking online consent there was a series of tick boxes to check participant eligibility (E.g. “I am currently aged 13-30”). Participants were only able to continue if they meet all the criteria. Before any participant was able to complete the survey they were asked to agree to a statement of consent. Once they consented they were given instructions to complete the survey. Participants were able to opt out of the study, skip questions or withdraw at any time.

#### **4.3.4 Outcome measure selection**

Both the survivor and caregiver survey mainly comprised of several validated, previously published questionnaires/tools that were the best fit to meet the study aims. The use of validated tools helped to save time and resources that were not obtainable during the PhD. Another benefit to using validated questionnaires/tools is that it allows better comparability to other studies which used the same questionnaire. As a part of selecting the most suitable validated tools for the survey, many tools were reviewed and discussed with the supervisory team. During this process a smaller number of tools that were deemed most suitable were left. Next, the PPI advisory group were consulted about which tools/questionnaires they felt addressed the research aims best. The PPI group members were asked to complete the questionnaires to see whether they were understandable and easy to complete. In general, the PPI group were concerned about validated tools that were: too long, had too much text, or were too complex – some tools were described as “confusing” and not visually interesting. Once the PPI group had been consulted a final set of validated questionnaires/tools made up the final survey.

As a reminder the objectives of the quantitative phase were to describe the met and unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers (objective 1), to explore if survivor sociodemographic (e.g. sex, age) and clinical data (e.g. time since diagnosis, treatment) are related to unmet supportive care needs (objective 2), to

investigate whether unmet supportive care needs are associated with QoL outcomes (objective 3) and to explore the role and perceived use of support services in TYA survivors and their caregivers (objective 4). The outcome measures are now described in detail.

#### **4.3.4.1 Survivor survey**

##### ***Sociodemographic and clinical characteristics***

Sociodemographic data collected in this study included patients' sex, age, ethnicity, educational background, employment status, relationship status, and living circumstances.

The clinical characteristics data collected included brain tumour type, grade and location; age at diagnosis and treatment received. The researcher obtained participants' clinical data by reviewing their clinical notes on the NHS system, and then recorded them on a structured data entry form. Participants who were recruited online were asked to self-report this data.

##### ***Quality of life***

Survivor's quality of life was measured using the Paediatric Functional Assessment of Cancer Therapy – Brain (Peds-FACT-Br), using the adolescence specific module.<sup>195</sup> There are a number of questionnaires available for measuring quality of life in brain tumour survivors, such as the Core Quality of Life Questionnaire (QLQ-C30)<sup>196</sup> with the brain tumour disease-specific module BN20,<sup>197</sup> or the MD Anderson Symptom Inventory Brain Tumor Module (MDASI-BT).<sup>198</sup> While each of these questionnaires have their own advantages, limitations and reported validity and reliability, it is beyond the scope of this thesis to examine them all in detail. The decision to choose the Peds-FACT-Br (Adolescence) was based on it being the only tool that was disease specific (for brain tumour survivors) while also being age specific (for TYAs). Also compared with the European Organisation for Research and Treatment of Cancer (EORTC) questionnaires, the Functional Assessment of Cancer Therapy (FACT) modules are more focused on emotional and social concerns, which were identified as a key issue for TYA survivors in the systematic review results (Chapter 2). The FACT tool was also the favoured when compared with others in its category by the PPI group.

The Peds-FACT-Br (Adolescence) tool is comprised of 37 items addressing 5 aspects of QoL: physical wellbeing (7 items), emotional wellbeing & illness experience (13 items), social and family well-being (5 items) and additional concerns (12 items). There are five Likert response options for each item: 'Not at all', 'A little bit', 'Somewhat', 'Quite a bit' and 'Very much'. As with all FACT questionnaires, a high score is good. Therefore, a score of "0" is a severely symptomatic patient and the highest possible score of "148" is an asymptomatic

patient. The instrument has been demonstrated to have robust psychometric properties (high reliability and validity) when validated in patients with primary brain tumours.<sup>199</sup>

### ***Supportive care needs***

Survivor needs were measured using the Supportive Care Needs Survey Short Form (SCNS-SF34).<sup>200, 201</sup> Again there were many cancer need assessment tools to choose from, including, Survivor Unmet Needs Survey (SUNS),<sup>202</sup> Childhood Cancer Survivor Study Needs Assessment Questionnaire (CCSS-NAQ),<sup>203</sup> Cancer Survivors' Unmet Needs (CaSUN)<sup>204</sup> and Cancer Needs Questionnaire Young People (CNQ-YP).<sup>205</sup> The decision to choose the SCNS-SF34 was based on three key factors. The first factor was the number of items in the tool – because some of the survivors might have trouble reading and/or concentrating it was important that the questionnaire was not too long and burdensome. For instance, the CCSS-NAQ had 135 items, and would take someone without cognitive issues around 45 minutes to complete alone. Secondly, the PPI group felt that the SCNS-SF34 had the most relevant content, was concise and was the easiest to understand. Thirdly, the instrument had previously demonstrated high reliability and acceptable convergent and construct validity.<sup>200</sup>

The 34-item instrument assesses needs via five analytically derived domains: 1) psychological (10 items), 2) physical and daily living (5 items), 3) sexuality (3 items), 4) health system and information (11 items), and 5) patient care and support (5 items). The instrument allows survivors to rate their perceived current level of need on a five-point scale. The first response scale consists of two broad categories of need, i.e. 'no need' and 'need'. The 'no need' scale is further divided into 'not applicable' for issues that were no problem to the participant, and 'satisfied' for issues on which a participant needed support but the support was satisfactory. The 'need' category has three subgroups representing the level of need for additional care, namely 'low need', 'moderate need' and 'high need'. Two scoring methods were utilised in the analysis. First, the number of items with low/moderate/high needs were counted for each domain of the SCNS-34. Second, a standardised Likert summated score was calculated for each unmet need domain as recommended in the Supportive Care Needs Survey scoring manual. The score has possible values ranging from 0 to 100, with a higher score indicating more unmet needs.<sup>206</sup>

### ***Support services and information use***

Currently, there is no validated questionnaire that measures TYA brain tumour survivor's service use. Therefore, a modified instrument adapted from a prior research tool on young adult survivors of childhood cancer was used.<sup>207</sup> The tool consists of 17 items which evaluate participants' reported use of and unmet need for: information or information resources, emotional support services and practical support services. The items were

created using information from the systematic review results and known available brain tumour support services in England.

Participants indicate whether they used the service by selecting one of four answers - “YES, and I don’t need it anymore”, “YES, and I would like to use more”, “NO, but I would like to”, and “NO, and I do not need to”. These categories distinguished respondents who indicated a need for service (in the past or currently) from those who did not. They also distinguished survivors who indicated that they have accessed and used a particular service (“met need”) from those who expressed a desire or need but have not, for whatever reason, had that desire or need met (“unmet need”).

### ***Free text questions***

As advised by the PPI group, there were also two free text questions added at the end of the survey, to allow participants the ability to report any thoughts or experiences that were not captured in the survey. The two free text questions asked participants: 1) if they have experienced a need that has not been listed in this survey and 2) to add any additional comments.

#### **4.3.4.2 Caregiver survey**

##### ***Sociodemographic and clinical characteristics***

Sociodemographic data collected included caregivers’ sex, age, ethnicity, educational background, employment status, and relationship status. Caregivers were also asked to state their relationship to the survivor and whether they currently lived with them.

If the survivor had also taken part in the study, then their clinical data was linked to the caregiver’s data. If their loved one (survivor) had not completed the survey or the caregiver had completed the survey after seeing the online advertisement then the caregivers self-reported survivor clinical characteristics data was utilised including survivor: brain tumour type, grade and location; age at diagnosis and treatment received.

##### ***Quality of life***

Caregivers’ quality of life was measured using The Caregiver Quality of Life Index–Cancer (CQOLC).<sup>208</sup> There are other caregiver quality of life validated tools such as Care Related Quality of Life(Carer)<sup>209</sup> and QOLLI-F (Quality of Life in Life Threatening Illness -Family Carer Version).<sup>210</sup> The CQOLC was chosen as it is specifically designed for caregivers of cancer patients, and the content seemed most relevant to this participant group. The CQOLC scale is a 35-item cancer-specific instrument that assesses the carer of a cancer patient’s quality of life, that is, some of the physical, social, emotional, and financial aspects

of well-being, and functioning. There are five Likert response options for each item: 'Not at all', 'A little bit', 'Somewhat', 'Quite a bit' and 'Very much'. The total possible score is 140, with higher scores representing better quality of life.

The CQOLC possesses good validity, test-retest reliability and internal consistency when tested on cancer caregivers.<sup>208</sup>

### ***Caregiver burden***

The Burden Scale for Family Caregivers – short version (BSFC-s) was chosen to measure caregiver burden.<sup>211</sup> There are many other instruments that measure caregiver burden, including the Caregiver Burden Screen (CBS)<sup>212</sup> and Screen for Caregiver Burden (SCB).<sup>213</sup> The BSFC-s was preferred as it was the most efficient instrument for assessing caregiver's burden in a short time frame (10 items).<sup>211</sup> Each item is rated on a scale from 0 (strongly disagree) to 3 (strongly agree). The overall score ranges from 0 to 30 points. Higher scores indicate greater caregiver burden. Studies have supported the convergent and discriminant validity of the BSFC-s when tested for carers of an elderly person<sup>214</sup> and the predictive validity for caring for people with dementia.<sup>211</sup>

### ***Supportive care needs***

Caregiver unmet needs was measured using both the Supportive Care Needs Survey for Partners & Caregivers (SCNS-PC).<sup>201</sup> There are many caregiver supportive needs instruments including: Cancer Survivors' Partners Unmet Needs (CaSPUN),<sup>204</sup> The Health Care Needs Survey (HCNS),<sup>215</sup> The Needs Assessment of Family Caregivers-Cancer (NAFC-C).<sup>216</sup> Reasons for not choosing the prior tools are as follows. The CaSPUN instrument was decided against because it was primarily designed to identify the needs of partners of long-term cancer survivors and majority of caregivers in this study were expected to be primarily parents. The HCNS was thought to be too long as it comprises of 90 items, this was also reiterated by our PPI group. And the NAFC-C had undergone limited psychometric evaluation.

The SCNS-PC is a 45-item instrument comprises health care, psychological, emotional needs, work, social and information needs. The instrument allows caregivers to rate their current level of need on a five-point scale (ranging from no need to high need). Items can be grouped into four domains: health-care service needs, psychological and emotional needs, work and social needs, and information needs, whereby item scores are summed and standardised 0 to 100. This scale has demonstrated high internal validity and reliability in caregivers of cancer patients.<sup>201</sup> The tool was also chosen because it matches the patient version –the SCNS-SF34.

### ***Support services and information use***

We did not find a validated questionnaire that measured caregivers of cancer survivor's service use. Therefore, this section of the questionnaire was specifically designed from the systematic review results and known available support services. The tool consists of 16 items which evaluate participants reported use of and unmet need for: information or information resources, emotional support services and practical support services.

Participants indicate whether they used the service by selecting one of four answers - "YES, and I don't need it anymore", "YES, and I would like to use more", "NO, but I would like to", and "NO, and I do not need to".

### ***Free text questions***

At the end of the survey two free text questions asked participants: 1) If they have experienced a need that has not been listed in this survey and 2) To add any additional comments.

### **4.3.5 Data analysis**

For the quantitative strand of the study, data management and analyses carried out using IBM SPSS Statistics V23.0. The data analysis plan data analyses involved cleaning and preparing the data prior to analyses. Steps in the data analyses plan are outlined below.

#### **Data preparation**

Data was prepared in order to carry out preliminary analyses. To begin all the sociodemographic data collected were first coded numerically, if they were in the form of text, and a codebook of all the created codes was prepared. For example, for highest educational level - High school was coded as 0 and College was coded as 1, and so on. Where necessary questionnaire total scores and subscale scores were calculated.

Time was spent checking the data. First, the responses were checked using frequency plots to ensure that all recorded answers were within the limits of the possible responses. Second, a randomly selected group (10%) of computer records were checked against paper questionnaires to check the quality of data entry. This check identified no incorrect data entry.

#### **Missing data**

Missing data was examined to evaluate if it followed any particular pattern. Missing data were less than 5% (see Table 8). Missing data was not replaced to analyse the first and

fourth objective, which looked at the prevalence of unmet needs and support service use. This has led to some of the sample not being used in some of the descriptive analyses. The reason for this was to ensure that no assumptions were made as to the individual responses of survivor or caregiver needs.

**Table 8 - Percentage of missing survey data by individual questionnaire tools**

Questionnaire tool	Total % of missing data
<b>Survivor individual questionnaire tools</b>	
SCNS-SF34	2.8%
Peds-Fact-Br Adolescence	1.4%
Support services and information use	1.7%
<b>Caregiver individual questionnaire tools</b>	
SCNS-P&CS	4.1%
BSFC-s	3.3%
CQOLC	3.3%
Support services and information use	4.8%

For the analyses (regression and correlation analysis- objective 2 and 3) where questionnaire subscale scores were needed, the subscale scores were prorated where less than half of the items within a domain were missing. Therefore, as long as a participant had completed more than 50% of the items in a single tool, the mean for the individual of the other items in that domain was inputted.<sup>217</sup> The total score was then calculated as the sum of the subscale scores. Where there was more than 50% missing data per each individual questionnaire tool, the participant's data for that scale or total score was excluded from the analysis. By replacing the missing data with the overall mean score for each missing variable on an individual basis was deemed appropriate. As it reflects the survivors or caregivers' other responses in that domain.<sup>217</sup>

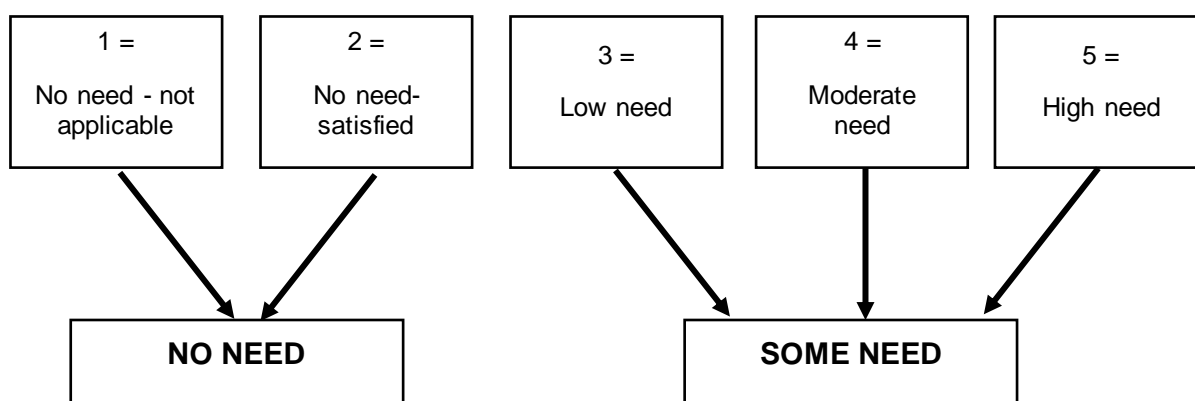
### **Participant characteristics**

Descriptive statistics were calculated to provide information about participant characteristics. Both frequency and percentages were reported to illustrate the sociodemographic and clinical characteristics of the survivors and caregivers, including sex, age group, ethnicity, relationship status, employment status, education level and survivor brain tumour type, grade, time since diagnosis and treatment. The mean value and standard deviation (SD) were used to present the results of outcome measures. The analyses to answer each of the objectives in turn were then undertaken.

### Objective 1: Prevalence of survivor and caregiver unmet supportive care needs

The first objective was to describe the unmet supportive care needs of the TYA survivors and their caregivers. Descriptive statistics were used to gain an understanding of the prevalence of unmet needs and which needs the survivors/caregivers required help with. Survivor unmet needs were investigated using the Supportive Care Needs Surveys – Short Form (SCNS-SF34). Caregiver unmet needs were investigated using the Supportive Care Needs Survey- Partners and Carers (SCNS-P&C). Each questionnaire measures need on a five-point rating scale (1 = no need- not applicable, 2 = no need- satisfied, 3 = low need, 4 = moderate need and 5 = high need). Directed by the questionnaires scoring guidelines,<sup>206</sup> the scores for each need were dichotomised with 0 = 'No need' (scale answer 1 and 2) and 1 = 'Some need' (scale answer 3-5). See Figure 7.

**Figure 7 - How unmet need was dichotomised (SCNS-SF34 and SCNS-P&C)**



It was then possible to calculate the number of unmet needs per participant and the highest ranked/most prevalent items in both the SCNS-SF34 and SCNS-P&C. In Chapter 5, survivor and caregiver needs are presented by the SCNS-SF34 and SCNS-P&C domains.

In addition, the summated scores from each domain in the SCNS-SF34 and SCNS-P&C were calculated in order to understand which domains scored the highest in relation to participants requiring the most help. The summated scores were standardised using the formula provided in the SCNS guidelines.<sup>206</sup> The formula was as followed:  $a \times 100 / (m \times (k - 1))$ , where  $m$  is the number of items in a domain;  $a$  is the adjusted Likert score (crude score –  $m$ ); and  $k$  is the maximum score value for each item.



## **Objective 2: Unmet supportive care needs and social/clinical characteristics**

### ***Regression analysis***

Regression analysis is a statistical process for estimating the relationships among variables. It is widely used for prediction and forecasting. It is also used to understand which among the independent variables are related to the dependent variable, and to explore the forms of these relationships. It is called univariable regression when there is one independent variable (predictor) and a continuous dependent variable. While having at least two variables (predictors) would make the regression multivariable regression.

The second objective was to establish whether social and clinical factors were significant predictors in the reporting of unmet needs of survivors and caregivers. Linear regression analysis was performed to explore the associations between unmet needs and clinical/sociodemographic variables. To begin, univariable linear regression analyses were performed to explore associations between total unmet needs/ individual domain scores (dependent variable) and independent variables: sociodemographic (i.e. age, sex, relationship status and employment status) and survivor clinical characteristics (i.e. time since diagnosis and treatment). The decisions behind the variables were partly exploratory (as there is very little prior research undertaken in this area) and part informed by the systematic review. For instance, from the review highlighted both chemotherapy and radiotherapy treatment were linked with more prevalent endocrine dysfunctions. Variables were also guided by the limitations of the data. For example, the ethnicities of the participants were left out due to a high proportion of participants being white British (survivors = 98.6%). Brain tumour type and location were not included due to the vast number of different tumour types and locations, meaning that many categories have a small number of participants. Similarly, tumour grades were not included as there were a large amount of unknown tumour grades, which would have meant the sample size would have been reduced. Due to multicollinearity concerns between age at diagnosis and time since diagnosis, age at diagnosis was not entered into the univariate analysis.

There are numerous different methods of conducting multivariable linear regression. The variables can be entered into the model using forced entry or through a stepwise method. According to Field (2009) the key factor that will influence this decision is whether you are exploring a concept or testing a hypothesis.<sup>218</sup> This is an exploratory study trying to explore the factors surrounding unmet needs instead of testing a theory and finding a cause. Therefore, a stepwise method was utilised.

The data can be entered in a forwards method, where the model starts with one constant variable and others are added depending on the statistically significant impact that they

make on the unmet needs outcome variable. The backwards method, which was the chosen method for this analysis. This is when the variables are all put into the model and then removed one by one, eliminating variables from the regression model to find a reduced model that best explains the data. This method was preferred as forwards methods of regression run a higher risk of making a type II error due to the suppressor effects which happens when a predictor has a significant effect but needs another variable to be consistent of this.<sup>218</sup>

Potentially many variables could be entered into the model if all the social and clinical characteristics are combined. Therefore, only variables that showed a significant association ( $p < 0.10$ ) with participants having an unmet need in a domain were entered. A significant correlation implies that there is a non-zero relationship between two variables, irrespective of how strong that relationship actually is. A high p value of 0.1 was chosen to ensure that no important association was missed in the multivariable analysis. Therefore, all variables with p value  $< 0.10$  in univariate analyses were entered into the multivariable linear regression model (see appendix 9 for an example). From this point on all tests of significance were two-sided and with p value  $< 0.05$  was considered as statistically significant. The results are presented in Chapter 5 by SCNS-SF and SCNS-P&C total unmet needs and then broken down by the questionnaire domains.

### ***Model assumptions***

Several assumptions must be met when using multivariable linear regression analyses. The main four assumptions include:

1. Linearity
2. Normality
3. No or little multicollinearity
4. Homoscedasticity

The first assumption is to check if there is a linear relationship between the independent and dependent variables. Standard linear and multiple regression can only accurately estimate the relationship between dependent and independent variables if the relationships are linear in nature.<sup>219</sup> The preferred method of linear detection is examination of residual plots. To do this scatterplots of the dependant and independent variables were created. With this visual method I was checking that there was some form of linear relationship, and not an indication of a curvilinear relationship.

The second assumption is normality. In order to make valid inferences from the regression, the residuals of the regression should follow a normal distribution. The residuals are the differences between the observed value of the dependent variable and the predicted value. To check for normality, Predicted Probability (P-P) plots were created to determine if the residuals were normally distributed. If normally distributed the plots conform to the diagonal normality line indicated in the plot. Sometimes, there was some deviation from the line, but normality can be assumed as long as there are no drastic deviations.<sup>220</sup> In all analyses, normality was assumed as there was no drastic deviation from the line. See appendix 10 for an example.

Thirdly, multiple linear regression assumes that there is little or no multicollinearity in the data. Multicollinearity occurs when the independent variables are not independent from each other.<sup>218</sup> Multicollinearity was checked using Variance inflation factors (VIF) values. If the VIF values lies between 1-10, then there is no multicollinearity. The most ideal case being that the VIF values are below 5. In all multivariable analysis VIF values were well within range for all predictor variables, all were below 5, that indicates that the assumption of multicollinearity was met. VIF values are presented in Chapter 5 in the multivariable regression tables.

The last assumption to check is homoscedasticity. Homoscedasticity means that the variance of errors is the same across all levels of the independent variable.<sup>218</sup> Homoscedasticity was checked using a visual examination of generated scatterplots for each model. Ideally, the plotted data should look random and does not have an obvious pattern.<sup>219</sup> The lack of pattern indicatives that the assumption of homoscedasticity has been met. All of the models included in this analyses met this condition. See appendix 10 for an example of the scatter plots created to check for homoscedasticity.

### **Objective 3: Correlation between unmet supportive care needs and quality of life**

The third objective was to investigate the association between the survivors' and caregivers' perceived supportive care needs and QoL. The Pearson's correlation coefficient was used to assess the relationship between unmet needs and QOL. Correlation tests can be used to assess whether two variables have a linear relationship with each other. Correlation tests may be used to evaluate both positive (when one variable increases, the other variable increases) and negative (when one variable increases, the other variable decreases) correlations.

The Pearson correlation test provides a measure of the linear association between two continuous variables. The coefficient,  $r$ , can take a range of values from +1 to -1. A value of 0 indicates that there is no association between the two variables. A value larger than 0

indicates a positive association, meaning as the value of one variable increases, so does the value of the other variable. A value less than 0 indicates a negative association, that is, as the value of one variable increases, the value of the other variable decreases.<sup>218</sup> In this analysis the guidelines depicted in Table 9 were used to interpret the strength of the Pearson's correlation coefficient.<sup>221</sup>

**Table 9 - Guidelines for interpreting the Pearson correlation coefficient**

Strength of association	Coefficient, r	
	Positive	Negative
Weak	.1 to .3	-.1 to -.3
Moderate	.3 to .7	-.3 to -.7
Strong	.7 to 1.0	-.7 to -1.0

Survivor QoL was measured using the Peds-FACT-Br. Before Pearson's correlation could be undertaken, an overall QoL score was calculated, using the questionnaire guidelines. The scoring guidelines also allow the QoL score to be broken down into four QoL subscales, namely physical, emotional, social and additional concerns (as grouped in the Peds-FACT-Br). Once these subscale scores were calculated they could be entered into the Pearson's correlation analysis with the SCNS-SF34 total number of needs per survivor, and also each of the SCNS-SF34 needs domains (Psychological needs, Health system and information needs, Physical and daily living needs, Patient care and support needs, and Sexuality needs).

Caregiver QoL was measured using the CQOLC. As with the survivor analysis, the CQOLC overall score had to be calculated before the Pearson correlation could be performed. The CQOLC overall score was then entered into the Pearson correlation analysis with the SCNS-P&C total number of needs per caregiver, and also each of the SCNS-P&C needs domains (Psychological and emotional needs, Information needs, Health Care Service needs, Work and social needs).

In Chapter 5 all Pearson correlation data are presented in tables, including the correlation coefficients (r), and their p values displayed.

**Objective 4: Prevalence of Service and Information use**

The fourth objective was to explore the role and perceived use of support services in TYA survivors and caregivers. Statistical analyses included a descriptive summary (frequencies and percentages) of participants reporting service use, and also reporting unmet need. Service use was defined if a participant had endorsed having used that service and no

longer had a need of that service or if they had used that service and would like to use it more. Unmet need, was defined if the participant reported not having used the service but would have liked to use it or if they had used the service but would like to use it more. These categories distinguished participants who indicated a need for service (in the past or currently) from those who did not.

### **Free text responses**

Free text responses were analysed alongside the qualitative data using thematic analysis. The free text data are presented alongside the qualitative data in Chapter 6.

## **4.4 Qualitative data: Semi-structured interviews**

The aim of the qualitative strand was to describe the unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers (objective 1), and also to explore the role and perceived use of support services in TYA survivors and their caregivers (objective 4). In response to the aims this qualitative phase, semi-structured, individual face-to-face interviews using open-ended questions were chosen.

Interviews are one of the most common methods of qualitative data collection within social and health research.<sup>158</sup> Compared with other qualitative methods, individual interviews are usually preferred for discussing personal experiences, as they allow for more detailed and personal discussions than group situations will permit.<sup>153, 158</sup> However, some argue that focus groups may be suitable for some marginalised groups because speaking to others 'like you' may be less intimidating than speaking to just the researcher.<sup>158</sup> In this instance interviews were chosen as having had a brain tumour or caring for someone who has had a brain tumour can be a challenging experience, and many survivors have either physical or cognitive issues as a result of the tumour and/or treatment. Therefore, conducting individual face-to-face interviews in their homes gave survivors more privacy and confidentiality in which to talk through their past experiences. Equally by conducting interviews singularly and at home meant that there were less distractions – as it was recognised that people with brain damage often have difficulty participating in group discussions which makes focus groups quite difficult. The PPI group also felt it could be difficult for some participants to express themselves in focus groups, because of issues with hearing, slow processing speeds and shyness.

Interviews can be described as a 'professional conversation',<sup>222</sup> with the aim being to get the participants to talk about their experiences and viewpoints in relation to a topic of your focus.<sup>223</sup> Interviews are often divided into three types: structured, semi-structured and

unstructured. In structured interviews the questions and response categories are all predetermined by the researcher. Whereas in unstructured interviews the interviewer has a list of themes/topics to discuss but the interview is heavily led by the interviewee. Semi-structured interviews include the researcher having a set list of questions (interview guide) but the participant has the opportunity to bring up themes/topics that may have not been anticipated.<sup>153, 158</sup> The most obvious strength of open-ended interviewing is the capacity to follow topics that occur during the course of conversation.<sup>152</sup> Semi-structured interviews have the added benefit of including the topics of interest to the researcher, while allowing the participant to raise their additional insights and thoughts.<sup>152</sup> Some believe that semi-structured interviews are the ideal as they ensure an interview is “*on target but hanging loose*”.<sup>223</sup>

The qualitative interview arose as a method in response to criticism about the depersonalisation of standard social scientific methods of data collection, with face-to-face contact between researchers and participants being viewed as preferential.<sup>158</sup> However, in comparison to telephone or online interviews (i.e. Skype interviews) they can be time consuming, and some participants may be less likely to open up face-to-face, especially those who are ‘hard to engage’.<sup>158</sup> Nevertheless, face-to-face interviews have many strengths, those applicable to this research include: they are ideal for discussing sensitive issues,<sup>158</sup> they are usually preferred when conducting research with vulnerable groups (i.e. young people and those with learning difficulties)<sup>158, 224</sup> and the researcher has more control over the data produced increasing the likelihood of generating useful data.<sup>152, 158</sup>

#### **4.4.1 Participant Sample**

Sampling methods in qualitative research usually focus on permitting a deeper understanding of the phenomena of interest.<sup>225</sup> Qualitative approaches tend to use smaller samples than quantitative approaches, however “*there are no rules for sample size in qualitative inquiry.*” (p. 244)<sup>225</sup> Sample size is determined by what outcomes are necessary, the purpose of the data collection, credibility and what can practically be done (resources and time).<sup>225</sup> Factors including: quality of the data, scope of the study, the amount of useful information obtained from each participant and method used also affect how many participants are needed.<sup>158</sup> Ideally the sample should provide enough data to tell a rich story, but not too much that it impedes data analysis within the time permitted. For qualitative research, an appropriate sample size is one that provides enough information to answer the research question(s).<sup>226</sup>

Qualitative methods place primary emphasis on saturation, through obtaining a comprehensive understanding by continuing to sample until no new substantive information is acquired. Thematic saturation has been defined as meaning that data should continue to be collected until novel themes are no longer generated.<sup>227</sup> It is clearly unknown at the outset of a project at which stage novel themes will cease to be discovered. However, Francis et al. (2012) proposed that it is good practice in qualitative health research to define a-priori criteria for establishing data saturation.<sup>228</sup> One of the main principles for assessing data saturation includes specifying a minimum sample size. For this PhD study the sample size was set at a minimum of ten survivors and caregivers, with the rate of thematic saturation to be continually assessed throughout the process. The rate of saturation was assessed through observation of the content of the interviews with the participants, but also through the analysis process, which was ongoing throughout the interviews and regularly discussed within academic supervision.

Qualitative samples tend to be purposive rather than random.<sup>229</sup> Random sampling is usually inappropriate for qualitative studies because random sampling of a population is likely to produce a representative sample only if the research characteristics are normally distributed within the population. In this study a sub-sample of participants who took part in the survey were asked to partake in an interview. After considering sampling options, purposive sampling was chosen as being most appropriate, due to the exploratory nature of the study questions. Purposive sampling, as its name suggests, is where participants of a sample are purposively selected to represent a particular population in relation to key criteria.<sup>230</sup> The main goal of purposive sampling is to focus on particular characteristics of a population that are of interest, which will best answer the research questions. The sample being studied is not representative of the population, but this is not considered to be a weakness.<sup>225</sup> Rather, it is a choice, the purpose of which varies depending on the type of purposive sampling technique that is used. Even though purposive sampling involves making 'deliberate choices', this does not suggest any bias as the sample is chosen with clear objectivity so that it will stand up to independent scrutiny.<sup>231</sup>

There are many purposive sampling techniques – including homogenous sampling, maximum variation sampling, extreme sampling, snowball sampling and typical case sampling. Central to each purposive sampling strategy is the ability to compare and contrast, to identify similarities and differences in the phenomenon or group of interest. Some strategies (e.g., maximum variation sampling and extreme case sampling) are also used to highlight and expand the range of variation or differences, while other strategies (e.g. homogeneous sampling and snowball sampling) are used to constrict the range of variation and focus on similarities.

In this study a maximum variation sample was utilised. Maximum variation is one of the most frequently used kinds of purposeful sampling.<sup>232</sup> Examples of variations may be race, age, class, sex or other person-related characteristics. Maximum variation sampling allows for variation in perspectives, ranging from those conditions that are viewed as typical through to those that are more extreme in nature.<sup>225</sup> The underlying principle behind maximum variation sampling is to gain greater insights into a phenomenon or group by looking from all angles. This can help the identification of common themes that are evident across the sample.<sup>225</sup>

In this study the purposive variations chosen were survivors that differed in age, sex and tumour type. These three criteria were decided upon for the following reasons. Firstly, both males and females can be diagnosed with childhood brain tumours but their experiences and needs as TYAs may be very different; therefore having a sample of both males and females was important. Age was determined a key criteria as it was anticipated that needs would change with age, according to the developmental milestones that were currently being experienced. For example, a survivor aged 13 would be unlikely to express a need for help with sexual relationships, or similarly a survivor aged 30 would be unlikely to be needing help with education. Finally, the reason for having a varied tumour group was because each tumour diagnosis can be very different, with its own unique treatment. Therefore, late effects, issues and needs may vary between different types of treatment. Despite its wide use and advantages, there are some challenges in identifying and applying maximum variation sampling.<sup>233</sup> Firstly, for small samples, large diversity can be problematic when individual cases are very different. However, maximum variation sampling turns this apparent weakness into a strength in that any common patterns that emerge from variation are of particular interest and value in capturing the core experiences and central shared aspects of a particular topic. Therefore, this approach allows the discovery of both high quality detailed descriptions of each participant while also allowing for important shared patterns to emerge.<sup>225</sup> Another challenge is that the range of variation in a sample is not often known at the outset of a study.<sup>233</sup> Therefore, often an iterative approach of sampling and re-sampling to draw an appropriate sample is usually recommended.<sup>233</sup> In practice, a pragmatic approach was used - those interviewed first were those that agreed to be contacted about an interview and then were willing and available to take part. As the qualitative strand progressed there was a more selective recruitment process. For example, at first the majority of survivor interviewees were aged [18+], which then meant that those aged 13-18 were then invited more frequently to ensure a varied sample.



#### 4.4.2 Data collection

All interviews took place at a place and time that best suited the participant. In most cases this was the participant's home, or a private room in the hospital. Usually survivor and caregiver interviews took place separately, one after another (if both wanted to take part). The aim of the interview and how it would be recorded were described to the participants; they were also assured about the confidentiality of information and their anonymity. Participants were also told the focus of the discussion and the expected length of the interview. They were given the opportunity to ask any questions and then if they still agreed to take part, the participant was asked to sign an assent or consent form. All interviews were audio recorded using an encrypted dictaphone.

To put participants at ease and to also help put their answers into context, all interviews began with the question "*To start, can you please tell me a little about you, in any way that you can/want to?*" Subsequently, several topics were addressed, although the order and the wording of questions sometimes differed from participant to participant, depending on how the interview had progressed. The interview guides and questions can be found in appendix 6. The topic guide was created based on the systematic review findings and the input of the PPI group. The questions asked were designed to explore the research questions as well as allow for the interviewee to have the freedom to express their thoughts and experiences. There were many prompts included, although not always needed they were useful for the interviews with survivors that needed help to recall past experiences. At the end of the interview, participants were thanked for their participation, and they were provided with the opportunity to ask any remaining questions.

After each interview detailed field notes were recorded. The field notes captured the interview setting, observations about the participant, critical reflections on my role as the interviewer and tentative thoughts on analysis.

### ***Reflections, learning and decision making...***

The interviews were designed to be singular interviews with either the survivor or caregiver. However, in practice this was not always the case. With advice from the PPI group, survivors were asked if they would like someone with them during the interview – such as the caregiver. Often survivors did not want anyone in the room, occasionally they did. In other instances, caregivers entered the room during the interview and joined in the interview. There were positives and negatives to this. For example, sometimes the caregiver answered on behalf of the survivor and it felt like the survivor became quieter in the interview. On the other hand, sometimes the caregiver prompted the survivor to think about something and then the survivor articulated experiences and feelings that they may have not if prompted by their caregiver. To deal with the threat of survivors voices getting lost in the interview, I made more eye contact and encouraging words to the survivor, to imply that I was interested in what they had to say, as well as their caregiver. I noted that in some instances caregivers believe that the survivor cannot or will not discuss things with the researcher and feel that they have to do it for them. When in reality many survivors are capable of talking without the aid of their caregiver but may just need extra prompts.

In other instances, interviews with survivors and caregivers were done jointly. For example, in one case both parents wanted to partake in the interview and felt that they both equally had valuable insights to provide. On reflection even though the interview was slightly harder to manage (i.e. sometimes them both trying to talk at the same time, harder to transcribe), it was a very valuable interview as they both had very different experiences of their journey and therefore had different supportive care needs. It was also interesting to see how experiences varied in between two caregivers of the same survivor.

#### **4.4.3 Data management**

During the qualitative phase, all signed consent forms were kept in a locked filing cabinet in a secure research office at St James University Hospital (Leeds). All interviews were recorded on encrypted devices. After each interview, data from the digital recorder was downloaded and stored on a password protected online university server and then deleted

from the recorder. Only the research team (myself and PhD supervisors) had access to these data.

#### **4.4.4 Data Analysis**

A clear and detailed account of the processes of data analysis is necessary to judge the contribution of the qualitative findings to its knowledge base.<sup>234</sup> Qualitative data analysis is a systematic process of sorting and classifying data that has been collected. Thematic analysis was chosen to guide the qualitative analysis of this study. To ensure high quality reporting in this thesis the analytical steps will be reported in detail.

##### **4.3.4.1 Thematic data analysis**

As the main aims of this research are exploratory, a descriptive analysis was deemed most appropriate. Both thematic analysis and interpretative phenomenological analysis (IPA) were considered. Thematic analysis is a method of identifying patterns or themes within qualitative data, providing a detailed account of the overall dataset, through describing participants' experiences and the similarities and differences.<sup>235</sup> IPA similarly identifies patterns across the data, but instead of providing an account of the dataset as a whole, the emphasis of IPA is much more about understanding individualistic experiences, with little attention given to broader social context. To carry out IPA analysis multiple interviews with each participant are usually needed to achieve needed depth.<sup>236</sup> Hence, this approach can produce detailed insights into individual experience, and how these experiences fit within the dataset as a whole, but due to this dual focus it can lack the overall descriptive narrative of thematic analysis. Therefore, even though IPA and thematic analysis could both provide interesting and useful insights into the lives of TYA brain tumour survivors and their caregivers, given that the main aim of this project is to describe and explore supportive care needs rather than a detailed exploration of individual perceptions, thematic analysis was deemed the most appropriate analytical method for this study.

In the past thematic analysis has been seen by some as only a tool for qualitative research rather than a method in its own right. However, Braun and Clarke (2006, 2013)<sup>158, 235</sup> argue it is a foundational method from which all qualitative research skills and methods can be built. The aim of thematic analysis is to identify themes, i.e. patterns in the data that are important or interesting, and use these themes to address the research questions. Thematic analysis is a useful method for examining the perspectives of different research participants, emphasising similarities and differences, and producing unforeseen insights. As a method it is much more than solely summarising the data; thematic analysis should also interpret and explain it.<sup>158, 235</sup>

Unlike many qualitative analytical methods (i.e. IPA, grounded theory and discourse analysis), thematic analysis it is not tied to a particular epistemological or theoretical perspective. Through its theoretical freedom, thematic analysis provides a highly flexible approach that can be adapted for the needs of many studies, facilitating a rich and detailed, yet complex account of data.<sup>235</sup> This was one reason it was chosen for this study, it allowed the maximum amount of flexibility of all the qualitative approaches. Additionally, thematic analysis provided a detailed framework that ensured the data was analysed in a systematic and rigorous way. It enabled the study to be firmly grounded within the data and allowed for previously unforeseen themes to emerge and be analysed.

Thematic analysis has numerous variations and can be driven by existing theory (deductive thematic analysis) or by the data (inductive thematic analysis).<sup>158</sup> It is advised that researchers specify if they are conducting an inductive or deductive thematic analysis as it informs how themes are theorised.<sup>235</sup> As highlighted in Chapter 2, there is very little is known about this topic area, hence the explorative nature of the research objectives. The outcome desired was a coherent picture of the participant experiences and supportive care needs. Therefore, an inductive thematic analysis approach seemed appropriate as the interviews were conducted and analysed with no theory-led position, so that new or unexpected themes could emerge bottom-up from the data. However, it is also unreasonable to suggest that researchers can totally free themselves of their values, assumptions and perspectives. Therefore, by applying a pragmatic stance, I acknowledged that in practice no research can be conducted in a purely inductive or deductive fashion and that it is impossible to be purely theory or data driven.<sup>146</sup> Hence, in this study, a combined approach was deemed appropriate. The pragmatic approach promotes abduction to move back and forth between deduction and induction. Here, a pragmatic approach allows flexibility to adopt the most feasible approach to address research questions. Therefore, the thematic analysis was both deductive – as it aimed to find data to address the research objectives, but also inductive to permit unexpected findings.

For this study, Braun & Clarke's (2013) 6-step framework was followed.<sup>158</sup> This method was chosen because it offered a detailed, clear and practical framework for doing thematic analysis. The rest of this section will detail the 6 steps taken. It is worth noting that all these steps were not done in a linear fashion. Data analysis requires constant movement between transcription, familiarisation, coding and the creation of themes.<sup>234</sup> The stages of analysis are detailed further below. At each stage of analysis, the results were discussed with the research team (myself and PhD supervisors).

### ***Transcription, immersion and familiarisation***

The process of transcription, while it can be time-consuming is an good way to start familiarising yourself with the data.<sup>237</sup> Some researchers even argue it should be seen as a key phase of data analysis, as it enables a far more thorough understanding of the data through having transcribed it.<sup>238, 239</sup> Therefore, I transcribed the interview data (verbatim) upon the completion of each interview. Transcription commenced at the same time as data collection, to enable ongoing analysis.

No notes were taken during the interviews, but detailed field notes were written after each interview. Field notes reflected on the details of the interview, such as the participant's presentation and surroundings, how I felt the interview went and key discussions which related to ideas of supportive care needs.

Once six interviews were transcribed a more structured analysis began, starting with a process of immersion in the data. The aim being to become more familiar with the interviews content, and to recognise things that might be relevant to the research questions.<sup>158</sup>

Immersion in the data early has the added benefit of making analysis more manageable, instead of tackling large amounts of data at one time.<sup>234</sup> During this process each interview transcript was read and re-read, and any initial thoughts were written down. These were not codes but just any brief ideas, written as memory aids and triggers for future analysis. Braun and Clarke (2013) highlight that these initial notes often reflect the researchers position in relation to the data, frequently being the most obvious aspects of the data or the things that are most significant to the researchers interests.<sup>158</sup>

Through reading the interview transcripts, familiarisation started to occur, meaning that the data could then be read actively, analytically and critically – moving beyond the surface of the data.<sup>158</sup>

### ***Coding***

The second step of data analysis was to begin coding the data. In a qualitative study coding generally refers to identifying and labelling aspects of the data that relate to your research question.<sup>158, 234</sup> Data are distilled, sorted and grouped by the process of coding. In thematic analysis coding continues to be developed and defined throughout the entire analysis.<sup>158, 235</sup>

There are many ways to code data. For example, some researchers like to mark paper copies of the transcripts with markers and some prefer to use software packages. In this instance Nvivo 12 was used. NVivo was chosen as there was a large amount of interview data to manage, and the software helped to make the process more efficient and manageable.

In line with thematic analysis guidance, *complete* coding was undertaken, aiming to identify anything and everything of interest or relevant to answering the research question within the entire dataset.<sup>158</sup> This means that rather than focusing on a specific corpus of instances, that all the data relevant to the research question is coded, and only later in the analytical process does the process become more selective. Any data that was not relevant to the research question was not coded. As the research objectives were fairly broad (many types of supportive care needs/supportive services) the coding was done widely, inclusively and comprehensively. For each part of text that was coded, it was decided whether it fitted with a code already used or whether a new code was needed. Open coding was used; this meant that there were no pre-set codes, but instead codes were developed and modified throughout the coding process.<sup>240</sup>

As more interview data was analysed, codes were added and the meanings refined. When this happened, previously coded transcripts were revisited to check that the code still applies or whether the codes needed updating. This process involved moving back and forth through the transcripts, while returning to the study questions and thinking of potential themes. The ultimate aim of this step was to produce a comprehensive set of codes that separated different issues and ideas in the data.<sup>158</sup>

### **Searching for themes**

After coding the data, the focus was looking for larger patterns across the dataset. This phase includes systematically identifying outstanding features of the data, while also integrating and interpreting the patterns identified, and creating themes.<sup>158</sup> A 'theme' can be described as something that "*captures something important about the data in relation to the research question, and represents some level of patterned response or meaning within the data set*" (p.82).<sup>235</sup> A theme is generally wider than a code in that it covers many facets.<sup>158</sup> Braun and Clarke (2013) use the analogy of brick house: a theme is like a wall, made up of many individual bricks (codes).<sup>158</sup>

To identify patterns and themes in the data, all the codes and data relating to the codes were reviewed in order to identify any similarities between codes. After developing a list of possible patterns, these were then refined and sorted into initial themes and subthemes to allow the data to be filtered and classified. During this step, patterns were decided as important for two reasons: their frequency in the data and also those patterns that were most meaningful for answering the research question. Hence, ensuring that analysis is importantly focused on meanings rather than just numbers.<sup>158</sup> Braun and Clarke's guidance promoted that there is no 'magical equation' or limit to what counts as a theme in qualitative analysis. Themes need to be identified across codes in proportion of the data, they do not need to be

present in every data item or even most data items. The questions in Figure 8 were used to help develop codes to themes. From this stage, four potential themes were constructed. Themes developed at this stage were provisional and were revised and refined throughout the analysis process.

**Figure 8 - Questions to aid developing themes - adapted from Braun and Clark's (2013)**

- Is this a theme (or just a code or subtheme)?
- What is the quality of this theme? Does the central organising concept tell me something meaningful about a pattern in the data, in relation to my research question?
- What are the boundaries of this theme – what does it include and exclude?
- Are there enough meaningful data to support this theme?
- Is there too much going on in the theme, so that it lacks coherence? Are the data too diverse and wide-ranging? Would a sub-theme resolve these problems? Or should it be split into two or more themes?
- How does this potential theme relate to other potential themes?
- What's the overall story of my analysis – does this contribute to that overall story?
- Does the central organising concept reflect the title/label I have given the theme?

***Reviewing and revising themes***

This phase was fundamentally about 'quality control' and consistency- checking whether the provisional themes fit well with the coded data and represent the dataset collected.<sup>158</sup> In order to do this several processes were undertaken. Firstly, to improve the credibility of the analysis, one of the academic supervision team (Florien Boele) independently coded 4 interviews. After this process, the coding and themes created were compared and discussed. These discussions lead to the removal and addition of themes. For example, in some cases sub-themes were split into several more specific sub-themes. In other instances the organising concept of the theme was expanded, and codes were added. Throughout this process it was important to remember that 'good' themes are distinctive on their own but also fit together with other themes to form an overall analysis.<sup>158</sup> After each theme had been revised, every transcript was re-read, checking that the themes captured the meaning of the transcripts in relation to the research questions.

### ***Defining themes and writing up the analysis***

A part of finalising analysis is clearly defining themes, stating what is unique and specific about each. For each theme a written rich description of its focus was completed. Data extracts were used to illustrate the different aspects of each theme, a narrative was written around those extracts and the overall theme. Extracts were carefully selected to compellingly illustrate the analytical point being made about the data, while also helping to convince the reader of the credibility of the argument. Alongside the descriptive and illustrative data extracts, a clear analytical narrative was written, which aims to make specific interpretative claims about the data extracts and the theme. The interpretative narrative helps to identify more unspoken or underlying meanings behind the data extracts. In total, 4 themes and 13 sub-themes are presented in Chapter 6.

#### **4.4.5 Quality of qualitative research**

Similar to quantitative research, establishing validity and reliability is a required step in the process of qualitative research. Various measures were employed to ensure methodological quality. Qualitative validity refers to checking the accuracy of the findings by employing certain procedures, while qualitative reliability means ensuring that the approach used is consistent.<sup>241</sup> Creswell (2014)<sup>242</sup> recommends that researchers actively incorporate several validity strategies into their work, to both assess the accuracy of their findings as well as convincing others of that accuracy. These approaches include: triangulation, member checking, rich thick description, clarifying the bias, negative or discrepant information, prolonged engagement, peer debriefing/reviewing and external auditor.

For this study, triangulation, providing a rich thick description, clarifying the bias, presenting negative cases and peer debriefing/reviewing were employed to ensure the accuracy of findings. Triangulation refers to the use of more than one approach and/or data sources in order to establish credibility and better understanding. If themes are established based on joining several sources of data, then this process can be seen as adding to the validity of the study.<sup>242</sup> Triangulation was achieved based on the mixed methods design of this study, both quantitative methods and qualitative methods were utilised. For example, the survey data enabled the identification of unmet needs, and the qualitative interview data facilitated the in-depth understanding of the intricacy of survivors and caregivers needs. Therefore, emergent qualitative themes were checked against surveys responses and were found to be useful for building a coherent explanation for the quantitative results.

Rich, thick description refers to a approach for establishing credibility by ensuring there is a detailed description of the qualitative: setting, participants, methods and themes.<sup>242</sup>



Therefore, the reliability of the research was ensured by providing a rich, comprehensive description of the research setting, sampling strategy, sample size, interviews, topic guide and data analysis earlier in this chapter to ensure transparency and to allow readers to decide the applicability of the findings to other settings.<sup>242</sup>

Clarifying the bias refers to the highlighting bias that the researcher brings to the study. Good qualitative research comprises comments by the researchers about how their interpretation of the findings is shaped by their background (e.g. training and research interests) In Chapter 6 (6.4.2) reflexivity is discussed as an important part of this qualitative research.

As advised by Creswell (2014)<sup>242</sup>, throughout the results negative or discrepant information that runs counter to the themes have been presented. This is because real life is made up of diverse perspectives that do not always consolidate into one version of events. As a maximum variation sampling strategy was used it meant that participants differed in many ways (i.e. sex, age, tumour type), hence experiences and feelings sometimes differed. In the results you can see when any data contradicts the general view of the theme. By discussing and highlighting any conflicting data it adds credibility and validity to the findings.

Peer debriefing/reviewing is a process to enhance the accuracy and validity of the qualitative account. This approach includes asking someone who is in a similar research area to review and ask questions about the study. This strategy enhances the accuracy of the findings by involving an interpretation beyond that of the researcher.<sup>242</sup> In this study, this process was undertaken by involving academic supervisors who reviewed this study from the initial design stage to the final thesis. Throughout the project, monthly supervision meetings were held which allowed the supervision team the opportunity to offer advice, support and guidance on the conduct of the study.

#### **4.5 Integration of quantitative and qualitative data**

After the presentation of the quantitative (Chapter 5) and qualitative findings (Chapter 6), Chapter 7 presents the integrated results and also details the integration methods undertaken.

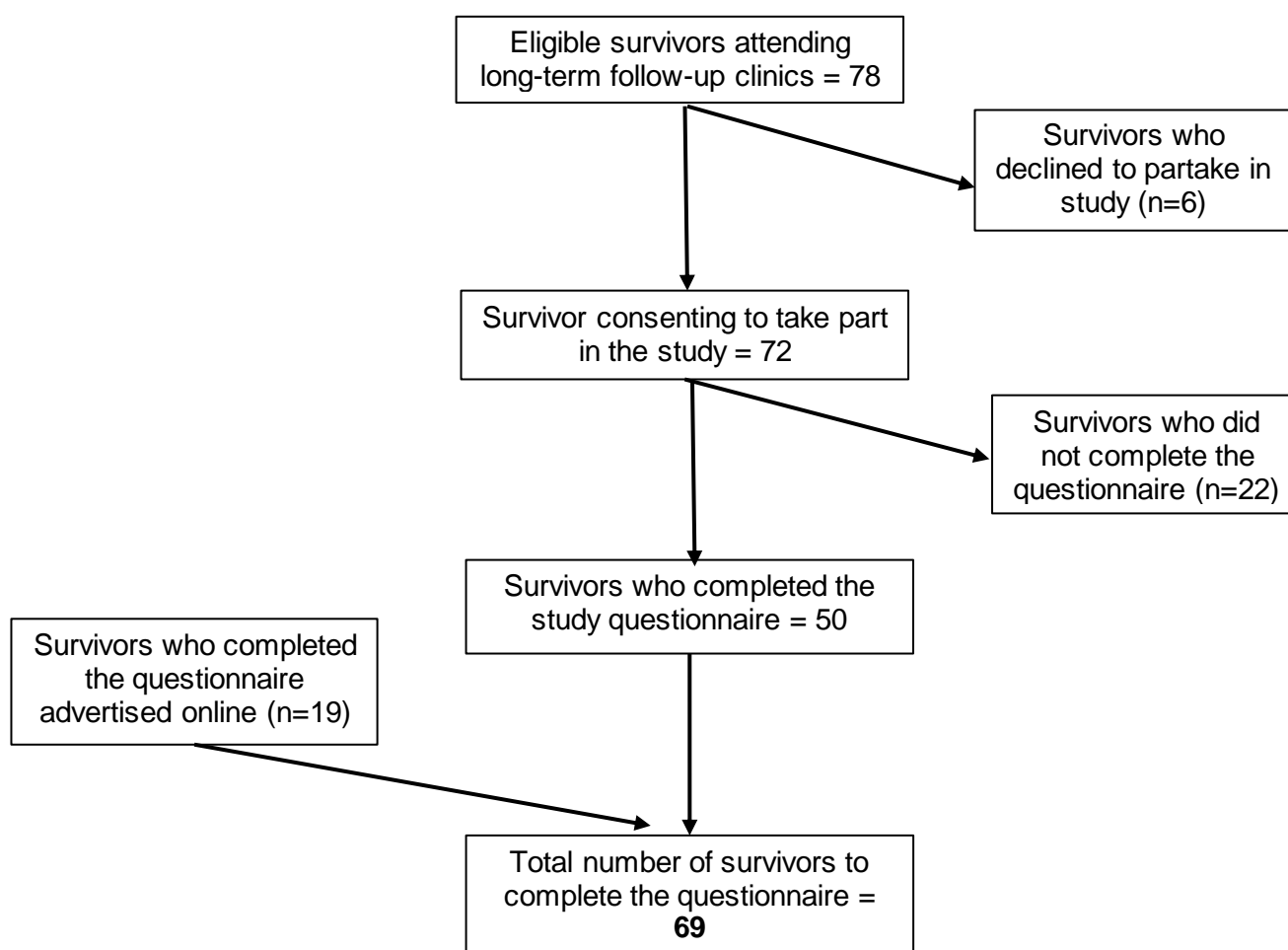
## **Chapter 5: Quantitative Results**

The following Chapter presents the quantitative study results. The methods of recruitment, data collection (4.3.3) and data analysis (4.3.5) were presented in the previous Chapter. The first section of this Chapter describes the response rate and sample size, next the sociodemographics of the sample and then the results in accordance to each of the study objectives are presented. Survivor and caregiver results are presented separately.

### **5.1 Survivor recruitment and response rate**

A total of 69 survivors took part in the quantitative strand of the study. 78 survivors were identified as eligible and approached about partaking in the study by the clinical teams (please see Chapter 4.3.3 for more details of the recruitment process). Of those 78 survivors, 72 consented (92.3%) to take part in the study and 50 went on to complete the survey (response rate = 69.4%). All participants who had consented were sent a reminder (if necessary), data was not collected to why participants did not complete the survey after consenting. Those who did not want to take part in the study did not give particular reasons apart from lack of time. A further 19 survivors completed the survey online after seeing the online advertisement. In total, 69 survivors completed the survey (See Figure 9).

**Figure 9 - A flow diagram to represent survivor response rate**



## 5.2 Survivor participant characteristics

Survivor sociodemographic, social and clinical characteristics are detailed in Table 10.

Within the survivor population, there were slightly more males (53.6%) than females (46.4%). The age of survivors ranged from 13-30, the mean was 22.6 years ( $\pm 4.3$ ). Nearly all survivors identified as White (98.6%). The majority of the respondents were still in education at the time they completed the survey (68.1%), 40.5% were in some form of employment and over a third were unemployed or unable to work (33.3%). Most survivors were living with their caregivers (85.5%), none lived alone. In the main survivors identified as single (79.7%), with only one survivor (1.4%) being currently married.

Clinical characteristics were recorded from both medical records (survivors recruited in long-term follow-up clinic, n=50) and survivor self-report data (survivors who completed the survey advertised online, n=19). The age at brain tumour diagnosis ranged from 0-14, the

mean age was 7.2 ( $\pm$  3.5) years. Time since diagnosis ranged from 7-27 years, on average it was 17.4 ( $\pm$ 4.9). Diagnoses were varied, with medulloblastomas (34.8%) and astrocytomas (26.1%) being the most common. The location of the tumour varied, most commonly located in the posterior fossa (21.7%). Nearly 40% of the respondents had low grade (grade I and II) brain tumours at time of diagnosis. The majority of survivors had received surgical resection (73.9%), radiotherapy (68.1%) and/or chemotherapy treatment (60.9%). A small minority of survivors had been diagnosed with Posterior Fossa Syndrome (5.8%).

There was little difference in the demographics of participants recruited online and long-term follow-up clinic. When compared more online survivors were female and had higher educational attainment. See appendix 8 for further details.

**Table 10 – Survivor sociodemographic, social and clinical characteristics**

Characteristic	Survivors N=69 (%)	Mean ( $\pm$ )
<b>Sex</b>		
Male	37 (53.6)	
Female	32 (46.4)	
<b>Age at completion (years)</b>		
13-18	15 (21.7)	22.6 (4.3)
19-24	31 (44.9)	
25-30	23 (33.3)	
<b>Ethnicity</b>		
White	68 (98.6)	
Other	1 (1.4)	
<b>Currently in education</b>		
Yes	47 (68.1)	
No	21 (30.4)	
Missing	1 (1.4)	
<b>Current highest education level</b>		
High School	14 (20.3)	
College	25 (36.2)	
University	27 (39.1)	
Other	3 (4.3)	
<b>Employment</b>		
Working Full-time	15 (21.7)	
Working Part-time	13 (18.8)	
Unable to work due to illness or disability	12 (17.4)	
Unemployed	11 (15.9)	
Student	15 (21.7)	
Other	1 (1.4)	
Missing	2 (2.9)	
<b>Living status</b>		
With parents	59 (85.5)	
Friends	3 (4.3)	
Partner	4 (5.8)	
Supported living or sheltered accommodation:	1 (1.4)	
<b>Relationship status</b>		

Single	55 (79.7)	
In a relationship	14 (20.3)	
<b>Age at diagnosis (years)</b>		
0-4	22 (31.9)	7.2 (3.5)
5-10	31 (44.9)	
11-14	16 (23.2)	
<b>Time since diagnosis (years)</b>		
7-13	15 (21.7)	17.4 (4.9)
14-19	29 (42.0)	
20-27	25 (36.2)	
<b>Tumour type</b>		
Medulloblastoma	24 (34.8)	
Astrocytoma	18 (26.1)	
Craniopharyngioma	6 (8.7)	
Pineal Tumour	4 (5.8)	
Choroid Plexus Carcinoma	4 (5.8)	
Ependymoma	3 (4.3)	
Other*	10 (14.5)	
<b>Tumour location</b>		
Posterior Fossa	15 (21.7)	
Cerebellum	9 (13.0)	
Pineal	7 (10.1)	
Brain stem	7 (10.1)	
Optic Nerve	4 (5.8)	
Cerebrum	4 (5.8)	
Brain not otherwise specified	4 (5.8)	
Other**	13 (18.8)	
Not sure (Online/self-report group only)	6 (8.7)	
<b>Tumour grade (at diagnosis)</b>		
Grade I	20 (29.0)	
Grade II	6 (8.7)	
Grade III	4 (5.8)	
Grade IV	18 (26.1)	
Unknown	21 (30.4)	
<b>Treatment: Resection</b>		
Yes	51 (73.9)	
No	17 (24.6)	
Missing	1 (1.4)	
<b>Treatment: Re-resection</b>		
Yes	7 (10.1)	
No	61 (88.4)	
Missing	1 (1.4)	
<b>Treatment: Radiotherapy</b>		
Yes	47 (68.1)	
No	26 (37.7)	
Missing	1 (1.4)	
<b>Treatment: Chemotherapy</b>		
Yes	42 (60.9)	
No	26 (37.7)	
Missing	1 (1.4)	
<b>Posterior Fossa Syndrome (PFS)</b>		
Yes	4 (5.8)	
No	62 (89.9)	
Not sure	2 (2.9)	

Missing	1 (1.4)	
---------	---------	--

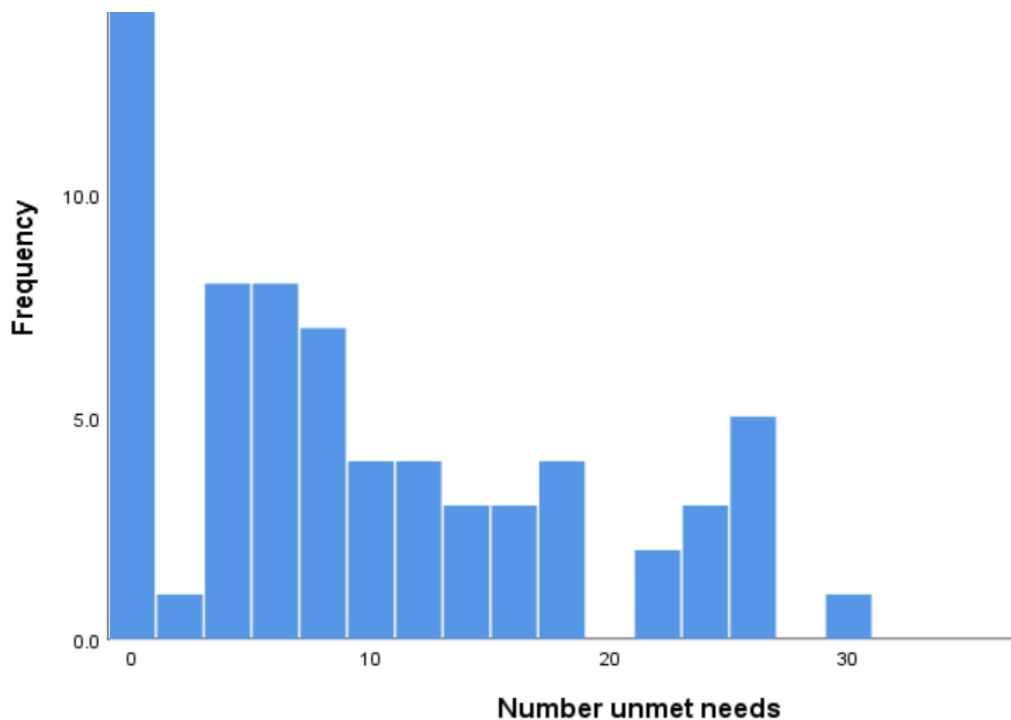
\* Two mixed glioma, Two PNET, one oligodendroglioma, one ganglioma, one germinoma, one neuroblastoma, one pineoblastoma, one 'other-not specified'.

\*\* Three pituitary gland, three parietal lobe, two craniopharyngeal duct, two frontal lobe, one occipital lobe, one temporal lobe and one cranial nerve.

### 5.3 Objective 1: Survivor unmet supportive care needs

The first study objective aims to describe the unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers. In this section survivor needs are outlined, in section 5.9 caregiver needs are detailed, these data combined meet the first objective. Survivor unmet needs were measured using the Supportive Care Needs Survey –Short Form (SCNS-SF34). The scores for each need were dichotomised with 0 = 'No need' - no need for supportive care or the survivor feels their need is being met; and 1= 'Some need' – a low, moderate or high need for supportive care. Survivor unmet needs ranged from 0 (no needs) to 30, the mean number of unmet needs was 9.4 ( $\pm 8.5$ ) per survivor (see Figure 10). Table 11 includes the percentage of survivors experiencing at least one, three, five, ten or fifteen unmet needs. Overall, over three-quarters of survivors (78.3%) reported at least three unmet needs. And over a quarter of survivors (27.5%) have at least fifteen unmet needs. 15 (21.7%) reported no unmet supportive care needs.

**Figure 10 - Histogram to show total number of unmet needs per survivor**



**Table 11 - Frequency of survivor unmet needs**

<b>Survivor unmet needs (answering 3 - 5 on each item)</b>	<b>Survivor N=69 (%)</b>
Mean 9.4 ( $\pm 8.5$ )	
No unmet needs	15 (21.7)
At least one unmet need	54 (78.3)
At least three unmet needs	53 (76.8)
At least five unmet needs	45 (65.2)
At least ten unmet needs	28 (40.6)
At least fifteen unmet needs	19 (27.5)

In accordance with the SCNS-SF34 guidelines unmet needs were divided into 5 domains: the Psychological domain, the Health System and Information (HSI) domain, and the Physical Daily Living (PDL) domain, the Sexuality domain and the Patient Care and Support (PCS) domain. Table 12 lists each SCNS-SF34 items, by domain and the number of people who perceived they required support with this. The results for each domain are then presented. Table 12 also presents the standardised Likert summated scores for each domain. Standardised means were used to compare the level of need (possible range 0-100) across domains. Higher scores indicate a higher perceived unmet need.

**Table 12 - Number of survivors who identified an unmet need, by each item of the SCNS-SF34 by domain. Table also shows the top ten most frequent supportive care needs in survivors.**

SCNS-SF34 domain and items	Standardised Likert summated Mean ( $\pm$ )*	Survivors with a need for help/ number who responded to question (%)	Top ten most frequent needs
<b>PSYCHOLOGICAL</b>	30.2 (23.9)		
Anxiety		41/68 (60.3)	<b>1/10</b>
Feeling down or depressed		33/68 (48.5)	<b>3/10</b>
Feelings of sadness		32/68 (47.1)	<b>=4/10</b>
Fears about the cancer spreading		11/68 (16.2)	
Worry that the results of treatment are beyond your control		15/68 (22.1)	
Uncertainty about the future		34/68 (50.7)	<b>2/10</b>
Learning to feel in control of your situation		26/67 (38.8)	<b>7/10</b>
Keeping a positive outlook		25/67 (37.3)	<b>8/10</b>
Feelings about death and dying		12/67 (17.9)	
Concerns about the worries of those close to you		28/67 (41.8)	<b>6/10</b>
<b>PHYSICAL AND DAILY LIVING</b>	28.0 (20.0)		
Pain		16/68 (23.5)	
Lack of energy/tiredness		32/68 (47.1)	<b>=4/10</b>
Feeling unwell a lot of the time		22/68 (32.4)	
Work around the home		14/68 (20.6)	
Not being able to do the things you used to do		23/68 (33.8)	
<b>PATIENT CARE AND SUPPORT</b>	25.1 (18.2)		
More choice about which cancer specialist you see		9/67 (13.4)	
More choice about which hospital you attend		11/66 (16.7)	
Reassurance by medical staff that the way you feel is normal		18/67 (26.9)	
Hospital staff to attend promptly to your physical needs		13/67 (19.4)	
Hospital staff to acknowledge, and show sensitivity to, your feelings and emotional needs		12/67 (17.9)	
<b>HEALTH SYSTEM AND INFORMATION</b>	18.3 (22.3)		



To be given written information about the important aspects of your care		16/67 (23.9)	
To be given information (written, diagrams, drawings) about aspects of managing your illness and side-effects at home		14/67 (20.9)	
To be given explanations of those tests for which you would like explanations		16/67 (23.9)	
To be adequately informed about the benefits and side-effects of treatments before you choose to have them		11/67 (16.4)	
To be informed about your test results as soon as feasible		16/67 (23.9)	
To be informed about cancer which is under control or diminishing (that is in remission)		8/67 (11.9)	
To be informed about things you can do to help yourself get well		21/67 (31.3)	
Access to professional counselling (e.g., psychologist, social worker, counsellor, nurse specialist) if you/family/friends need it		24/67 (35.8)	<b>9/10</b>
To be treated like a person, not just another case		16/67 (23.9)	
To be treated in a hospital or clinic that is as physically pleasant as possible		18/67 (26.9)	
One member of hospital staff with whom you can talk to about all aspects of your condition, treatment and follow-up		23/67 (34.3)	<b>10/10</b>
<b>SEXUALITY</b>	13.4 (19.5)		
Changes in sexual feelings		9/67 (13.4)	
Changes in sexual relationships		7/67 (10.4)	
To be given information about sexual relationships		15/67 (22.4)	

\* The summated scores were standardised using the formula provided in the SCNS guidelines. The formula was as followed:  $a \times 100 / (m \times (k - 1))$ , where  $m$  is the number of items in a domain;  $a$  is the adjusted Likert score (crude score –  $m$ ); and  $k$  is the maximum score value for each item

### 5.3.1 Psychological needs

The standardised scores (Table 12) indicate that Psychological needs had the highest mean score (30.2) of all the SCNS-SF34 domains. The Psychological domain consists of 10 items of need that people with cancer may require further assistance with in terms of the psychological and emotional well-being. Table 12 lists each item and the number of people who perceived they required help with items listed in all areas of the Supportive Care Needs Survey – short form. The majority of the top 10 ranked unmet needs belonged to the Psychological domain (7/10). The top four unmet needs were all in this domain: ‘Anxiety’ was the need ranked the highest by the participants (60.3%), followed by ‘Uncertainty about

*the future*' (50.7%), *'Feeling down or depressed'*. (48.5%) and *'Feelings of sadness'* (47.1%). The areas in which participants had the least amount of needs in this domain were *'Feelings about death and dying'* (17.9%) and *'Fears about the cancer spreading'* (16.2%).

### **5.3.2 Physical and Daily Living needs**

The Physical and Daily Living domain consists of 5 items that relate to physical problems and symptoms that people with cancer may require further help with. The domain was another highly reported area of need, nearly half of all participants said that they required help with *'Lack of energy/tiredness'* (47.1%). 'Furthermore around a third of participants recorded that they required further support with these issues: *'Not being able to do the things you used to do'* (33.8%) and *'Feeling unwell a lot of the time'* (32.4%).

### **5.3.3 Patient Care and Support needs**

The Patient Care and Support domain consists of 5 questions relating to healthcare provider and the choices and support that are offered to the survivor. This domain was not identified by survivors as a high area of need, none of the items in this domain were in the top 10 ranked unmet needs. The highest unmet need in this domain was *'Reassurance by medical staff that the way you feel is normal'*, reported by around a quarter of participants (26.9%).

### **5.3.4 Health System and Information needs**

The Health System and Information domain consists of 11 items relating to informational needs and treatment centre. The needs in this domain were not reported as frequently as in the Psychological and Physical and Daily Living domains. However, two of the top ten overall ranked needs were in this domain. The highest unmet need, reported by 35.8% of participants, was *'To have access to professional counselling (e.g., psychologist, social worker, counsellor, nurse specialist) if you/family/friends need it'*. The second highest unmet need in this domain was *'To have one member of hospital staff with whom you can talk to about all aspects of your condition, treatment and follow-up'* (34.3%).

### **5.2.5 Sexuality needs**

The standardised scores (Table 12) indicate that Sexuality needs had the lowest mean score (13.4) of all the SCNS-SF34 domains. This domain consists of 3 items that relate to sexual issues that the participants may require more support with. Table 3 also highlights that two of

the items with lowest level of unmet needs across all domains were in this domain: “Changes in sexual relationships” (10.4%) and “Changes in sexual feelings” (13.4%).

## **5.4 Objective 2: Survivor unmet supportive care needs and sociodemographic/clinical data**

The next section presents the data to address the second objective - to explore if survivor sociodemographic or clinical data are significant predictors in the reporting of unmet supportive care needs within a group of TYA survivors. Nine potential predictor variables were chosen to include in the univariate analysis, including: sex, survivor age at study entry, whether they have attended higher education, employment status, relationship status, time since brain tumour diagnosis and whether they have been treated with surgery, chemotherapy and radiotherapy (See 4.3.5 for the reasoning behind the selection of these variables). Univariate regression analysis indicated whether variables were associated at  $p < .10$  level. Variables that were associated at  $p < .10$ , were entered into a stepwise multivariable regression until the best model, with the fewest predictors was left. The results are now presented by the total number of unmet needs in all domains and then individually by the SCNS-SF34 domains.

Since both the survivor and caregiver sample size was smaller than initially planned, it meant that potentially fewer predictor variables could be inputted into the multivariable models. There are many sample size methods available for multiple linear regression. In general, sample size rules are based on the premise that with a large enough ratio of subjects to predictors, the sample regression coefficients will be reliable and will closely estimate the true population values.<sup>243</sup> Several authors suggest that per variable the minimum number of participants should be 10.<sup>244-246</sup> Hence, this was what guided the multivariable regression analysis. It was decided prior to the analysis that if there were more variables that significantly associated at univariate analysis than the guidelines support then the variables with the strongest association would be prioritised. In practice, this did not occur, so no variables had to be prioritised.

### **5.4.1 Total level of unmet needs**

The following analyses were undertaken to explore the association between clinical/social characteristics and the total number of unmet needs as reported by survivors. Univariate regression analysis identified six social or clinical categories that significantly correlated ( $p < .10$ ) with the reporting of unmet needs. The analysis indicates that unmet needs are more prevalent in females ( $r^2 = .89$ ,  $p = .013$ ), survivors further away from diagnosis ( $r^2 = .28$ ,  $p = .090$ ),

those not in a relationship ( $r^2=.42$   $p=.092$ ), those not in employment ( $r^2=.081$   $p=.018$ ), those not treated with surgery ( $r^2=.43$   $p=.093$ ) and chemotherapy ( $r^2=.52$   $p=.065$ ). See Table 13 for more details.

**Table 13 - Univariate regression analysis: associations between total number of unmet needs and clinical/social characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	5.053	.89	.013*	1.083 to 9.023
Current age	-.032	.000	.896	-.527 to .462
Higher education (0=no/1=yes)	2.880	.028	.173	-1.295 to .7055
In a relationship (0=no/1=yes)	-4.296	.42	.092*	-9.310 to .718
In employment (0=no/=1=yes)	-4.889	.081	.018*	-8.924 to -.855
Time since diagnosis	.372	.043	.090*	-0.60 to .804
Surgery (0=no/1=yes)	-3.938	.043	.093*	-8.545 to .670
Chemotherapy (0=no/1=yes)	-3.856	.052	.065*	-7.951 to .240
Radiotherapy	.862	.002	.714	-3.648 to 5.299

\*P<0.10

These six variables were entered into the multivariable regression analysis, using a stepwise selection model. In the final multivariable model sex, time since diagnosis, and employment status remained the best fit with survivor unmet needs ( $r^2=.237$ ,  $p<.01$ ). The analysis indicates that unmet needs were more prevalent in females, survivors further away from treatment and those not in employment. The  $r^2$  for this model was 0.237 that means 23.7 % variability of the outcome is explained by this model. VIF values were calculated to check multicollinearity; each value is below 10, indicating that the assumption is met. See Table 14 for more details.

**Table 14 - Multivariable model: total number of unmet needs**

Model	B	P value	95% CI	VIF
Sex (male =0/female=1)	4.973	.005*	1.299 – 8.647	1.006
In employment (0=no/=1=yes)	-5.704	.002*	-9.452 - -1.955	1.026
Time since diagnosis	.476	.023*	-.086 - .866	1.032

\*p<.05

#### 5.4.2 Psychological needs

After univariate regression analysis, five social and clinical variables were significantly associated with the reporting of psychological unmet needs. The analysis indicates that psychological unmet needs were more prevalent in female survivors ( $r^2=.115$ ,  $p=.005$ ), those

in higher education ( $r^2=.043$ ,  $p=.091$ ), and those not treated with surgery ( $r^2=.071$ ,  $p=.031$ ) and chemotherapy ( $r^2=.051$ ,  $p=.069$ ). See Table 15 for univariate regression analysis results.

**Table 15- Univariate regression analysis: associations between Psychological needs and clinical/social characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	16.163	.115	.005*	1.083 to 9.023
Current age	-.750	.017	.290	14.841 to 79.439
Higher education (0=no/1=yes)	10.079	.043	.091*	-1.649 to 21.806
In a relationship (0=no/1=yes)	-8.588	.22	.235	-22.897 to 5.720
In employment (0=no/1=yes)	-11.671	.081	.048*	-23.240 to -.103
Time since diagnosis	.664	.017	.286	-569 to -1.897
Surgery (0=no/1=yes)	-13.941	.071	.031*	-26.566 to -1.315
Chemotherapy (0=no/1=yes)	-10.582	.051	.069*	-22.001 to .838
Radiotherapy (0=no/1=yes)	-3.076	.004	.623	-15.515 to 9.362

\* $p<.10$

These variables (all associated at  $p<.10$ ) were entered into a stepwise multivariable linear regression analysis. The final model (see Table 7) with the best fit included sex and surgery ( $r^2=.168$ ,  $p<.05$ ). This indicates that psychological unmet needs were more prevalent in females and those who have not been treated with surgery ( $n=24.7\%$ ). The  $r^2$  for this model was 0.168 that means 16.8 % variability of the outcome is explained by this model.

**Table 16- Multivariable model: Psychological needs**

Model	B	P value	95% CI	VIF
Sex (male =0/female=1)	14.398	.008*	3.820 to 24.975	1.000
Surgery (0=no/1=yes)	-13.629	.027*	-25.673 to -1.585	1.000

\* $p<.05$

### 5.4.3 Physical and Daily Living needs

Within the Physical and Daily Living domain, only one variable was significantly associated with the reported level of unmet needs. At the univariate level there was a negative correlation between employment and reporting Physical and Daily Living unmet needs ( $r^2=.134$ ,  $p=.002$ ). Indicating that survivors not in employment report more physical unmet needs. See Table 17 for the results of this analysis.

**Table 17- Univariate regression analysis: associations between Psychological and Daily Living needs and clinical/social characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	7.799	.115	.223	-5.130 to 20.727
Current age	0.42	.000	.957	-1.511 to 1.595
Higher education (0=no/1=yes)	3.907	.005	.559	-9.362 to 17.176
In a relationship (0=no/1=yes)	-7.844	.014	.331	-22.872 to 8.139
In employment (0=no/=1=yes)	-19.696	.134	.002*	-32.005 to -7.387
Time since diagnosis	.667	.014	.330	-.700 to -2.053
Surgery (0=no/1=yes)	-9.906	.027	.181	-24.531 to 4.719
Chemotherapy (0=no/1=yes)	-6.904	.017	.298	-20.035 to 6.227
Radiotherapy (0=no/1=yes)	6.862	.015	.331	-7.137 to 20.861

\*P<0.10

#### 5.4.4 Patient Care and Support needs

After univariate analysis, four variables had an association with the reporting of unmet needs within the Patient Care and Support domain (see Table 18). The analysis indicates that unmet needs were more prevalent in females ( $r^2=.058$ ,  $p=.049$ ), survivors with higher education ( $r^2=.046$ ,  $p=.081$ ), those not in employment ( $r^2=.069$ ,  $p=.032$ ) and those not treated with chemotherapy ( $r^2=.055$ ,  $p=.057$ ).

**Table 18- Univariate regression analysis: associations between Patient Care Support needs and clinical/social characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	10.699	.058	.049*	-.026 to 21.327
Current age	-.294	.003	.657	-1.614 to 1.025
Higher education (0=no/1=yes)	9.699	.046	.081*	-1.219 to 20.617
In a relationship (0=no/1=yes)	-10.020	.034	.136	-23.277 to 3.236
In employment (0=no/=1=yes)	-11.777	.069	.032*	-22.504 to -1.049
Time since diagnosis	.719	.024	.215	-.700 to -2.053
Surgery (0=no/1=yes)	-7.137	.022	.233	-18.965 to 4.692
Chemotherapy (0=no/1=yes)	-10.087	.055	.057*	-20.492 to .318
Radiotherapy (0=no/1=yes)	-2.728	.004	.633	-14.090 to 8.634

p<.10

When entered into the multivariable model higher education and employment remained the best fit with survivor unmet needs ( $r^2=.167$ ,  $p<.05$ ). This indicates that Patient Care and Support unmet needs are more prevalent in those who have higher levels of education (i.e. University degree) and those who are not in employment. The  $r^2$  for this model was 0.167 that means 16.7 % variability of the outcome is explained by this model. See Table 19.

**Table 19- Multivariable model: Patient Care and Support needs**

Model	B	P value	95% CI	VIF
Higher education (0=no/1=yes)	14.369	.006*	4.331 to 24.407	1.055
In employment (0=no/=1=yes)	-13.442	.009*	-.23.428 to -3.456	1.055

$p<.05$

#### 5.4.5 Health system and Information needs

Within the Health System and Information domain, three variables were significantly associated with the reported level of unmet needs. Female survivors ( $r^2=.065$ ,  $p=.037$ ), unemployed survivors ( $r^2=.057$ ,  $p=.051$ ) and those who were not treated with chemotherapy ( $r^2=.074$ ,  $p=.027$ ) were more likely to report more health system and information needs. See Table 20 for further details.

**Table 20- Univariate regression analysis: associations between Health system and Information needs and clinical/social characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	13.518	.065	.037*	.822 to 26.214
Current age	.110	.000	.890	-1.467 to -1.687
Higher education (0=no/1=yes)	9.615	.032	.149	-3.518 to 22.748
In a relationship (0=no/1=yes)	-12.809	.39	.110	-28.596 to 2.978
In employment (0=no/=1=yes)	-12.806	.057	.051*	-25.695 to .083
Time since diagnosis	.992	.031	.151	-.372 to 2.335
Surgery (0=no/1=yes)	-6.103	.011	.411	-20.851 to 8.644
Chemotherapy (0=no/1=yes)	-14.493	.074	.027*	-27.259 to -1.727
Radiotherapy (0=no/1=yes)	.855	.000	.904	-13.252 to 14.961

$P<.10$

The final multivariable model with the best fit included variables employment and chemotherapy as the best predictors of unmet needs ( $r^2=.125$ ,  $p<0.05$ ). Within the Health System and Information domain survivors who were not employed and had not received

chemotherapy had more unmet needs (Table 21). The  $r^2$  for this model was 0.125 that means 12.5 % variability of the outcome is explained by this model.

**Table 21 - Multivariable model: Health system and Information needs**

Model	B	P value	95% CI	VIF
In employment (0=no/1=yes)	-14.879	.019*	-.27.277 to -2.481	1.036
Chemotherapy (0=no/1=yes)	-17.297	.008*	-29.837 to -4.757	1.036

\*P<.05

### 5.4.6 Sexuality needs

Finally, in the sexuality domain, after univariate analysis only one variable was associated with reported unmet needs. Chemotherapy was negatively associated to sexuality unmet needs ( $r^2=.089$ ,  $p=.015$ ). Indicating that survivors who have not been treated with chemotherapy had more unmet needs. See Table 22 for univariate analysis details.

**Table 22- Univariate regression analysis: associations between Sexuality needs and social and clinical characteristics**

Model	B	R <sup>2</sup>	P value	95% CI
Sex (male =0/female=1)	3.017	.0065	.532	-6.576 to 12.610
Current age	.505	.012	.384	-.644 to 1.654
Higher education (0=no/1=yes)	.247	.000	.960	-9.534 to 10.028
In a relationship (0=no/1=yes)	-8.704	.33	.139	-20.306 to 2.898
In employment (0=no/1=yes)	-5.693	.006	.242	-15.318 to 3.932
Time since diagnosis	.695	.029	.170	-.305 to 1.695
Surgery (0=no/1=yes)	1.200	.001	.830	-8.898 to 12.299
Chemotherapy (0=no/1=yes)	-11.875	.089	.015*	-21.359 to -2.391
Radiotherapy (0=no/1=yes)	7.609	.032	.148	-2.784 to 18.001

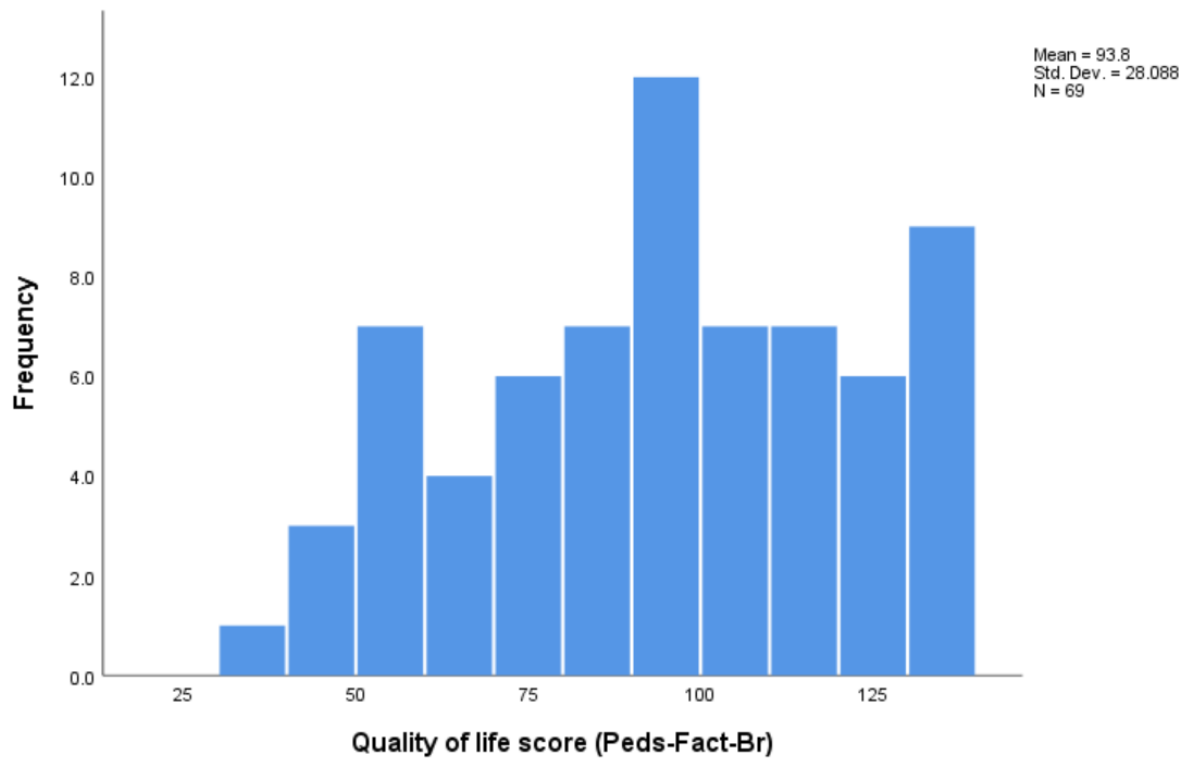
\*P<0.10

## 5.5 Objective 3: Survivor unmet supportive care needs and quality of life

The third objective looked to determine whether unmet supportive care needs are associated with QoL outcomes. Survivor QoL was measured using the Peds-FACT-Br. The total possible score is 148, a higher score represents better QoL. Survivor scores ranged from 33-139, with a mean score of 93.8 ( $\pm 28.088$ ). See Figure 11.



**Figure 11- Histogram to show survivor Quality of Life scores**



To investigate the correlation between the survivors’ perceived needs and quality of life, the Pearson correlation coefficients were calculated. The total unmet needs score was significantly associated with overall QoL score ( $r = -.621, p < .001$ ). With  $r$  being between  $-0.30$  and  $-0.70$  this indicates a moderate negative correlation.<sup>218</sup> This means that as one increases, the other decreases. Indicating that survivors who had more unmet needs reported a lower QoL.

All of the SCNS-SF34 needs domains, including the psychological, health system and information, physical and daily living, patient care and support, and sexuality domains, were also significantly negatively correlated with QoL scores. See Table 23. Indicating that as the number of survivor unmet needs increases, survivor quality of life score decreases. The correlation coefficients ranged from a moderate negative association between QoL overall score and Sexuality needs ( $r = -.358, p = .003$ ) to a strong negative association between QoL overall score and Psychological needs ( $r = -.751, p < .001$ ).

When the QoL scores were broken down into physical, emotional, social and additional concerns (as grouped in the Peds-FACT-Br) there were significant negative correlations between all need domains and individual QoL domains except from survivors’ Social

wellbeing score and Sexuality needs ( $r = -1.90$ ,  $p = .124$ ). The most significant correlation was found between Emotional wellbeing score and the Psychological needs ( $r = -.770$ ,  $p < .001$ ).

**Table 23 - Quality of life scores in correlation to the total number of unmet needs and individual SCNS-SF34 domains**

		Quality of Life overall score (Peds-FACT-Br (Adolescence))	Physical wellbeing score	Emotional wellbeing score	Social wellbeing score	Additional concerns score
Total number unmet needs	Correlation	-.621***	-.536***	-.602***	-.396**	-.551***
	P value	.000	.000	.000	.001	.000
	N	68	68	68	68	68
Sexuality domain	Correlation	-.358**	-.338**	-.331**	-.190	-.354**
	P value	.003	.005	.006	.124	.003
	N	67	67	67	67	67
Psychological domain	Correlation	-.751***	-.506***	-.770***	-.567***	-.652***
	P value	.000	.000	.000	.000	.000
	N	67	68	68	68	68
Physical domain	Correlation	-.729***	-.691***	-.663***	-.513**	-.648***
	P value	.000	.000	.000	.003	.000
	N	68	68	68	68	68
Patient Care and Support domain	Correlation	-.515***	-.467***	-.498***	-.359**	-.427***
	P value	.000	.000	.001	.003	.000
	N	67	67	67	67	67
Health System and Information domain	Correlation	-.449***	-.427***	-.431***	-.294*	-.368**
	P value	.000	.000	.000	.016	.002
	N	67	67	67	67	67

\*  $p < .05$ , \*\*  $p < .01$  \*\*\*  $p < .001$

## 5.6 Objective 4: Survivor service and information use

The next section presents the results to address the fourth objective - to explore the role and perceived use of support services in TYA survivors. Services used by TYA survivors are summarised in Table 24. The most used services were counselling (42.0%), online support groups -such as a Facebook group (36.2%) and cognitive rehabilitation (34.7%). The least used services by survivors included 24/7 online support (7.4%), mentor/buddy system (9.4%) and informational workshops (10.5%). Over half of survivors said there was an unmet need (survivors who stated 'Used and would like to use more' or 'Not used but would like to')

for weekend retreats with other brain tumour survivors (55.2%), monthly social activities with other survivors (52.1%) and counselling (37.6%).

**Table 24 - Survivor support service use**

<b>Support Service</b>	<b>Individual response</b>	<b>N = 69</b>	<b>%</b>
Counselling	Used and have no further need	14	20.3
	Used and would like to use more	15	21.7
	NOT used but I would like to	11	15.9
	NOT used and have no need	29	42.0
Cognitive Rehabilitation	Used and have no further need	19	27.5
	Used and would like to use more	5	7.2
	NOT used but I would like to	8	11.6
	NOT used and have no need	37	53.6
Online information	Used and have no further need	10	14.5
	Used and would like to use more	12	17.4
	NOT used but I would like to	7	10.1
	NOT used and have no need	40	58.0
In- person support group	Used and have no further need	9	13.0
	Used and would like to use more	8	11.6
	NOT used but I would like to	14	20.3
	NOT used and have no need	38	55.1
Online support group	Used and have no further need	12	17.4
	Used and would like to use more	13	18.8
	NOT used but I would like to	8	11.6
	NOT used and have no need	36	52.2
24/7 Telephone support	Used and have no further need	5	7.7
	Used and would like to use more	2	3.1
	NOT used but I would like to	8	12.3
	NOT used and have no need	50	76.9
24/7 Online chat support	Used and have no further need	4	5.9
	Used and would like to use more	1	1.5
	NOT used but I would like to	10	14.7
	NOT used and have no need	53	77.9
Monthly social activities	Used and have no further need	7	10.1
	Used and would like to use more	13	18.8
	NOT used but I would like to	23	33.3
	NOT used and have no need	26	37.7
Weekend retreats	Used and have no further need	4	6.0
	Used and would like to use more	11	16.4
	NOT used but I would like to	26	38.8
	NOT used and have no need	26	38.8
Informational workshops	Used and have no further need	2	3.0
	Used and would like to use more	5	7.5
	NOT used but I would like to	14	20.9
	NOT used and have no need	46	68.7

Mentor/Buddy system	Used and have no further need	2	3.1
	Used and would like to use more	4	6.3
	NOT used but I would like to	17	26.6
	NOT used and have no need	41	64.1

Information resources used by TYA survivors are summarised in Table 25. Most young adult survivors indicate that they have received information related to their brain tumour treatment side effects (86.5%). Over half have also accessed information regarding diet (66.5%) and exercise (55.0%). The information that has least been used were about finances (37.7%) and fertility (40.0%). Not only were these two information resources the least used/received by survivors, they were the most desired. Nearly half of survivors said they would like to receive information (or more information) about their finances (42.0%) and their fertility (43.1%).

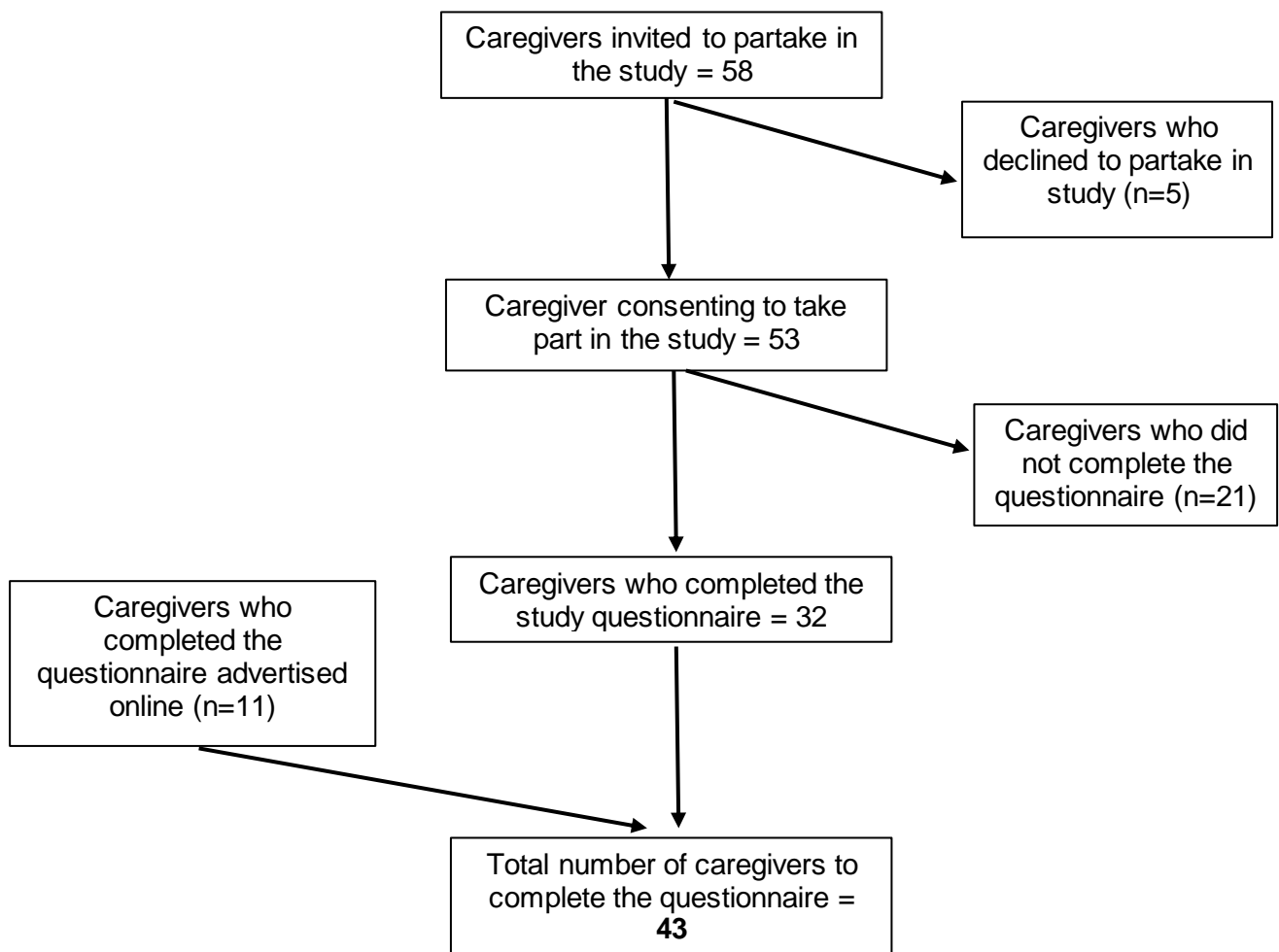
**Table 25 - Survivor information resource use**

Information	Individual response	N = 69	%
<b>Treatment side effects</b>	Information received and no further need	42	61.8
	Information received and would like more	10	14.7
	NOT received but I would like to	9	13.2
	NOT received and have no need	7	10.3
<b>Diet</b>	Information received and no further need	31	44.9
	Information received and would like more	8	11.6
	NOT received but I would like to	12	17.4
	NOT received and have no need	18	26.1
<b>Exercise</b>	Information received and no further need	31	44.9
	Information received and would like more	7	10.1
	NOT received but I would like to	13	18.8
	NOT received and have no need	18	26.1
<b>Employment/ Education</b>	Information received and no further need	21	30.9
	Information received and would like more	10	14.7
	NOT received but I would like to	20	29.4
	NOT received and have no need	17	25.0
<b>Finances</b>	Information received and no further need	16	23.2
	Information received and would like more	10	14.5
	NOT received but I would like to	19	27.5
	NOT received and have no need	24	34.8
<b>Fertility</b>	Information received and no further need	16	24.6
	Used and would like to use more	10	15.4
	NOT received but I would like to	18	27.7
	NOT received and have no need	21	32.3

## 5.7 Caregiver recruitment and response rate

When survivors were approached in the hospital clinics, their caregivers (when applicable) were asked to participate too, in total 58 caregivers were invited. Out of the total number of caregivers identified, 53 consented (93.1%) to take part in the study. For the caregivers who refused to take part in the study the main reason was the lack of benefit for them by taking part. 32 caregivers went on to complete the survey (60.4%), 21 did not complete the survey after consenting and despite a reminder. A further 11 caregivers completed the survey online after seeing the online advertisement. In total, 43 caregivers completed the survey. See Figure 12.

**Figure 12- A flow diagram to represent caregiver response rate**



## 5.8 Caregiver participant characteristics

Caregiver's social characteristics are detailed in Table 26. Caregivers were all parents of the survivors. The majority were female (86.0%), with a mean age of 52.4 ( $\pm$  6.4). Nearly all caregivers identified as White (93.0%). Around a third (34.9%) are educated to University degree level and around half (55.9%) were in full or part-time employment. The majority were married (74.4%), and around a fifth (21.0%) were not in a relationship.

Table 26 also details the caregivers' loved ones (survivor) clinical details. Most caregivers were living with the survivor (90.7%). The survivors they cared for ranged from 13-30 years old, 21.2 ( $\pm$  4.8) on average. It ranged from 5-27 years since their child was diagnosed with a brain tumour, the mean time was 14.1 (5.0). The survivors were mainly diagnosed with medulloblastomas (37.2%) and astrocytomas (25.6%), but there were thirteen different tumour diagnoses in total. Again tumour locations varied widely, around a fifth of survivor's tumours were located in the posterior fossa (20.9%). Nearly half (41.8%) of caregivers' loved ones were diagnosed with a high-grade tumour (grade III and IV).

When compared there was little difference in the demographics of caregivers recruited online and long-term follow-up clinic. Caregivers recruited online were on average looking after younger survivors and closer to diagnosis.

**Table 26 - Caregiver social characteristics and survivor clinical characteristics**

Characteristic	Caregivers N=43 (%)	Mean ( $\pm$ )
<b>Sex</b>		
Male	6 (14.0)	
Female	37 (86.0)	
<b>Age at completion (37-64 years)</b>		
37-46	8 (18.6)	52.4 (6.4)
47-56	26 (60.5)	
57+	9 (20.9)	
<b>Ethnicity</b>		
White	40 (93.0)	
Other	1 (2.3)	
Missing	2 (4.7)	
<b>Highest education level attained</b>		
High School	12 (27.9)	
College	7 (16.3)	
NVQ	6 (14.0)	
University	15 (34.9)	
Masters	1 (2.3)	
Other	2 (4.7)	
<b>Employment</b>		
Working Full-time	10 (23.3)	
Working Part-time	14 (32.6)	
Caring for home/family	13 (30.2)	
Retired	3 (7.0)	

Unable to work due to illness disability	1 (2.3)	
Other	1 (2.3)	
Missing	1 (2.3)	
<b>Relationship status</b>		
Married	32 (74.4)	
Divorced/separated/widowed	5 (11.7)	
Dating/in a relationship	2 (4.7)	
Single	4 (9.3)	
<b>Lives with survivor</b>		
Yes	39 (90.7)	
No	4 (9.3)	
<b>Survivor current age (range 13-30 years)</b>		
13-18	14 (32.6)	21.2 (4.8)
19-24	16 (37.2)	
25-30	12 (27.9)	
Missing	1 (2.3)	
<b>Survivor age at diagnosis (years)</b>		
0-4	14 (32.6)	6.7 (3.7)
5-10	23 (53.5)	
11-14	6 (14.0)	
<b>Time since diagnosis (range 5-27 years)</b>		
5-12	15 (34.9)	14.1 (5.0)
13-19	23 (53.5)	
20-27	4 (9.3)	
Missing	1 (2.3)	
<b>Tumour type</b>		
Medulloblastoma	16 (37.2)	
Astrocytoma	11 (25.6)	
Ependymoma	3 (7.0)	
Choroid Plexus Carcinoma	2 (4.7)	
PNET	2 (4.7)	
Ganglioglioma	2 (4.7)	
Other*	7(16.3)	
<b>Tumour location</b>		
Posterior Fossa	9 (20.9)	
Cerebellum	6 (14.0)	
Optic nerve	6 (14.0)	
Pineal	4 (9.4)	
Cerebrum	3 (7.0)	
Parietal lobe	3 (7.0)	
Brain not otherwise specified	3 (7.0)	
Unknown	3 (7.0)	
Other**	6 (14.0)	
<b>Tumour grade</b>		
Grade I	10 (23.3)	
Grade II	3 (7.0)	
Grade III	5 (11.6)	
Grade IV	13 (30.2)	
Unknown	12 (27.9)	
<b>Treatment : Resection</b>		
Yes	32 (74.4)	
No	11 (25.6)	
<b>Treatment: Re-resection</b>		
Yes	4 (9.3)	

No	39 (90.7)	
<b>Treatment: Radiotherapy</b>		
Yes	31 (72.1)	
No	12 (27.9)	
<b>Treatment: Chemotherapy</b>		
Yes	34 (79.1)	
No	9 (20.9)	
<b>Posterior Fossa Syndrome (PFS)</b>		
Yes	3 (7.0)	
No	40 (93.0)	

*\*One oligodendroglioma, one craniopharyngioma, one mixed glioma, one pineal tumour, one neuroblastoma, one pineoblastoma and one other- brain tumour not specified.*

*\*\* One ventricle, one pituitary gland, one temporal lobe, one choroid plexus, one cranial nerve and one brain stem.*

## 5.9 Objective 1: Caregiver unmet supportive care needs

The first objective of the study aimed to describe the unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers. Caregiver unmet needs were measured using the Supportive Care Needs Survey for Partners & Caregivers (SCNS-P&C). This 45-item instrument comprises four domains (Information needs, Psychological and Emotional needs, Health Care Service needs and Work and Social needs) and assesses the need for help for caregivers across the illness trajectory. The scores for each caregiver need item were dichotomised with 0 = 'No need' - no need for supportive care or the caregiver feels their need is being met; and 1= 'Some need' –a low, moderate or high need for supportive care. Caregiver unmet needs ranged from 0-42, and the mean number of unmet needs was 12.4 ( $\pm 12.3$ ) per caregiver (see Figure 13).



**Figure 13- Histogram to show total number of unmet needs per caregiver**

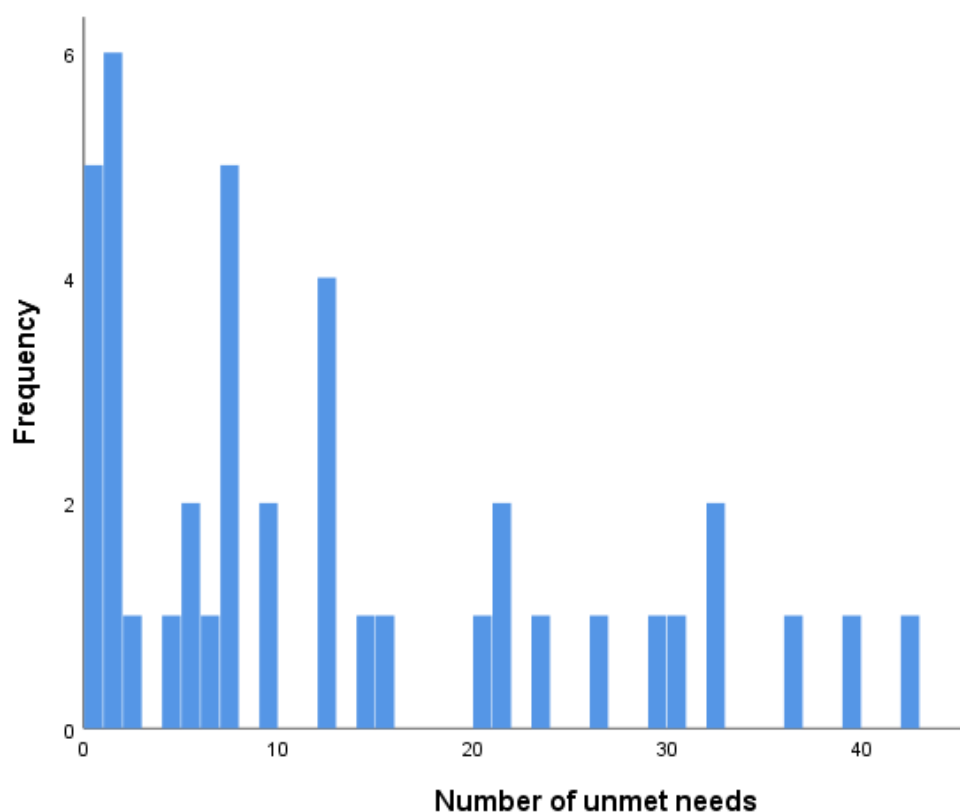


Table 27 details the percentage of carers experiencing at least one, three, five, ten, or fifteen unmet needs. Overall, the majority of caregivers (88.4%) reported at least one unmet needs. Nearly half (46.5%) reported at least ten unmet needs, and over a third (34.9%) identified fifteen unmet needs. Only 5 caregivers (11.6%) reported no unmet supportive care needs.

**Table 27 - Frequency of caregiver unmet needs**

Caregiver unmet needs (answering 3 - 5 on each item)	Caregivers N =41 (%)
Mean 12.4 (±12.3)	
No unmet needs	5 (11.6)
At least one unmet need	38 (88.4)
At least three unmet need	31 (72.1)
At least five unmet need	30 (69.8)
At least ten unmet need	20 (46.5)
At least fifteen unmet need	15 (34.9)

In accordance with the SCNS-P&C guidelines unmet needs are divided into 4 domains: the Psychological and Emotional domain, the Information needs domain, the Health Care Service needs domain and the Work and social needs domain. Table 28 lists each SCNS-P&C item, by domain and the number of people who perceived they required support with this. The results for each domain are then presented. Table 28 also presents the standardised Likert summated scores for each domain. Standardised means were used to compare the level of need (possible range 0- 100) across domains. Higher scores indicate a higher perceived unmet need.

**Table 28 - Number of caregivers who identified an unmet need, by each item of the SCNS-P&C by domain. Table also shows the top ten most prevalent supportive care needs in caregivers**

SCNS-P&C domains and items	Standardised Likert summated Mean ( $\pm$ )*	Caregivers with a need for help/ number who responded to question (%)	Top ten most prevalent
<b>PSYCHOLOGICAL AND EMOTIONAL</b>	28.8 (25.7)		
Managing concerns about recurrence		17/41 (41.5)	<b>=4/10</b>
The influence cancer has had on your relationship with survivor		6/41 (14.6)	
Understanding the experiences of the survivor		9/41 (22.0)	
Balancing own and survivor's needs		16/41 (39.0)	<b>=7/10</b>
Adjustment to changes in survivors body		9/40 (22.5)	
Addressing problems in your sex life		3/40 (7.5)	
Getting emotional support for yourself		16/41 (39.0)	<b>=7/10</b>
Getting emotional support for the people you love		13/41 (31.7)	
Dealing with your emotions about death and dying		13/41 (31.7)	
Dealing with others who don't recognise the effects on your life of looking after the survivor		17/41 (41.5)	<b>=4/10</b>
Dealing with your emotions when the recovery of the person with cancer has not happened as you had expected		12/41 (29.3)	
Making decisions about your life in the midst of uncertainty		17/41 (41.5)	<b>=4/10</b>
Being able to give meaning to the survivor's illness		7/41 (17.1)	
Exploring your spiritual beliefs		3/41 (7.3)	

<b>INFORMATION</b>	27.0 (26.2)		
Information relevant to your carer needs		15/42 (35.7)	
Information about prognosis		6/42 (14.3)	
Information about support services		16/41 (39.0)	<b>=7/10</b>
Information about alternative therapies		9/41 (22.0)	
Information about survivor physical needs		9/41 (22.0)	
Information about side effects of treatment		12/41 (29.3)	
Information about possible fertility problems		18/42 (42.9)	<b>=1/10</b>
Information about financial support and governmental benefits		18/42 (42.9)	<b>=1/10</b>
Information about life and/or travel insurance		15/41 (36.6)	
Information about accessing legal services		8/41 (19.5)	
<b>HEALTH CARE SERVICE</b>	26.7 (26.5)		
Getting the best medical care		10/41 (24.4)	
Accessing local health services		13/40 (32.5)	
Being involved in survivor medical care		7/42 (16.7)	
Opportunity to discuss care with doctor		8/42 (19.0)	
Feeling confident that all the doctors consult with each other to coordinate care		12/41 (29.3)	
A case manager who coordinated services		15/42 (38.7)	<b>=10/10</b>
Complaints regarding care being addressed		6/42 (14.3)	
Reducing stress in the survivor's life		16/42 (38.1)	
Looking after your own health		16/42 (38.1)	
Pain control for survivor		3/42 (7.1)	
Fears about survivor physical and mental deterioration		15/41 (36.6)	
Managing practical caring tasks		10/42 (23.8)	
Accessing hospital parking		16/42 (38.1)	
The opportunity to participate in decision making about survivors treatment		7/37 (18.9)	
<b>WORK AND SOCIAL</b>	25.2 (22.6)		
Changes to survivor working life or usual activities		14/42 (36.6)	

Influence of caring on your working life or usual activities		18/42 (42.9)	<b>=1/10</b>
Communicating with the patient with cancer		9/41 (22.0)	
Communicating with family		6/41 (14.6)	
Getting more support from your family		9/41 (22.0)	
Talking to other cancer carers		6/41 (14.6)	
Discussing the cancer in social situations or at work		9/41 (22.0)	

\* The summated scores were standardised using the formula provided in the SCNS guidelines. The formula was as followed:  $a \times 100 / (m \times (k - 1))$ , where  $m$  is the number of items in a domain;  $a$  is the adjusted Likert score (crude score –  $m$ ); and  $k$  is the maximum score value for each item

### 5.9.1 Psychological and emotional needs

The standardised scores (Table 28) indicate that Psychological and emotional needs had the highest mean score (30.2) of all the SCNS-P&C domains. The Psychological and Emotional domain consists of 14 items of needs pertaining to preserving or managing emotions, thoughts, and/or relationships with the survivor and others. The majority of the top 10 ranked unmet needs belonged to the psychological and emotional domain (5/10). The top unmet needs in this domain were: '*managing concerns about recurrence*' (41.5%), '*dealing with others who don't recognise the effects on your life of looking after the survivor*' (41.5%) and '*making decisions about your life in the midst of uncertainty*' (41.5%). The areas in which caregivers had the least amount of needs in this domain were in reference to '*exploring their spiritual beliefs*' (7.3%) and '*addressing problems in their sex life*' (7.5%).

### 5.9.2 Information needs

This domain has 10 items relating to the caregiver's information needs. Two of the highest ranked items (ranked joint first) were in this domain, with nearly half reporting wanting information about survivor '*fertility problems*' (42.9%) and '*financial support and governmental benefits for themselves and/or the survivor*' (42.9%). The item with the lowest need was for '*Information about prognosis*' (14.3%).

### 5.9.3 Health Care Service needs

The Health Care Service needs domain consists of 14 items relating to receiving optimal health care services and/or appropriate support from health care professionals. The needs in this domain were not reported as frequently as the other domains. Yet, one of the top ten overall ranked needs was in this domain, with over a third (38.7%) of caregivers reporting a

need for support with: *'ensuring there is an ongoing case manager to coordinate services for the survivor'*. The lowest ranked need was also in this domain, with only 7.1% of caregivers identifying *'obtaining adequate pain control for the survivor'* as an unmet need.

#### **5.9.4 Work and Social needs**

The standardised scores (Table 28) indicate that Work and social needs had the lowest mean score (25.2) of all the SCNS-P&C domains. The domain has 7 items and addresses the caregiver or survivor's work, in addition to items relating to interpersonal exchanges and/or talking about the cancer. One of the top ranked unmet needs was in this domain, nearly half (42.9%) of caregivers identified a need for help with *'the impact that caring for the survivor has had on their working life, or usual activities'*. And over a third (36.6%) of caregivers also reported an unmet need in *'adapting to changes to the survivors working life, or usual activities'*.

#### **5.10 Objective 2: Caregiver unmet supportive care needs and sociodemographic/clinical data**

The next section presents the results to address the second objective - to explore if caregiver sociodemographic or their loved ones (survivor) clinical data are significant predictors in the reporting of caregiver unmet needs. Caregiver predictor variables were selected using the systematic review findings (e.g. chemotherapy was a predictor of poorer survivor educational achievement, hence this could be a potential predictor of caregiver needs). Variables were also selected with the knowledge of the significant variables associated with survivor needs (e.g. time since diagnosis) and through utilising a pragmatic approach to considering what variables might influence unmet needs (e.g. caregiver employment status).

Consequently, six potential predictor variables were selected, namely: caregiver age at study entry, whether they have received higher education (i.e. University level and above), relationship status, employment status, survivor current age and time since survivor brain tumour diagnosis. The sample size after inclusion of all variables was n=41. Following the sample size guidelines highlighted earlier in this chapter (5.4), it was decided that a maximum of 3 variables would be entered into the model for multivariable regression. If there were more than 3 variables that significantly associated at univariate analysis then the variables with the strongest association would be prioritised. In practice, there were no more than three significant variables per multivariable regression model, so no variables had to be prioritised.

The results are now presented by the total number of unmet needs in all domains and then individually by the SCNS-P&C domains.

### 5.10.1 Total level of unmet needs

The following analyses were undertaken to explore the association between caregiver characteristics and the total number of unmet needs reported by caregivers. Univariate analysis identified three variables that significantly correlated ( $p < .10$ ) with the reporting of unmet needs. The analysis indicates that unmet needs were more prevalent in single caregivers ( $r^2 = .281$ ,  $p < .001$ ), caregivers caring for younger survivors ( $r^2 = .079$ ,  $p = .079$ ) and those caring for survivors closer to treatment ( $r^2 = .102$ ,  $p = .044$ ). See Table 29 for univariate analysis.

**Table 29 – Univariate regression analysis: associations between total number of unmet needs and caregiver characteristics/survivor clinical factors**

Model	B	R <sup>2</sup>	P value	95% CI
Caregiver current age	-.282	.021	.363	-.902 to .338
Higher education (0=no/1=yes)	2.516	.010	.541	-5.744 to 10.775
In a relationship (0=no/1=yes)	-15.556	.281	.000*	-23.620 to -7.492
In employment (0=no/1=yes)	2.342	.009	.550	-5.514 to 10.198
Survivor current age	-.718	.079	.079*	-1.525 to .088
Time since survivor diagnosis	-.781	.102	.044*	-1.540 to -.021

\* $p < .10$

The three significant variables were entered into the multivariable regression analysis. In the final multivariable model, only relationship status remained the best fit with caregiver unmet needs ( $B = -15.394$ ,  $r^2 = .276$ ,  $p < .001$ ,  $CI = -23.575, -7.213$ ). This indicates that single caregivers were more likely to report unmet needs. The  $r^2$  for this model was 0.276 that means 27.6% variability of the outcome is explained by this predictor alone.

### 5.10.2 Psychological and emotional needs

After univariate analysis, three variables were significantly associated with the reporting of caregiver psychological and emotional unmet needs. The analysis indicates that psychological unmet needs were more prevalent in single caregivers ( $r^2 = .152$ ,  $p = .012$ ), for caregivers caring for younger survivors ( $r^2 = .087$ ,  $p = .064$ ) and those caring for survivors closer to treatment ( $r^2 = .164$ ,  $p = .010$ ). See Table 30 for univariate analysis results.

**Table 30- Univariate regression analysis: associations between Psychological and emotional needs and caregiver characteristics/survivor clinical factors**

Model	B	R <sup>2</sup>	P value	95% CI
Caregiver current age	-.783	.013	.224	-2.067 to .500
Higher education (0=no/1=yes)	-2.008	.001	.816	-19.324 to 15.308
In a relationship (0=no/1=yes)	-23.927	.152	.012*	-42.204 to -5.650
In employment (0=no/=1=yes)	5.054	.010	.537	-11.343 to 21.451
Survivor current age	-1.566	.087	.064*	-3.228 to .096
Time since survivor diagnosis	-2.044	.164	.010*	-1.540 to -.021

\*p<.10

Significant variables (all associated at p<.10) were entered into a stepwise multivariable linear regression analysis. The final best fit model (see Table 30) included caregiver relationship status and time since survivor diagnosis ( $r^2=.255$ ,  $p<.05$ ). This indicates that Psychological and Emotional unmet needs were more likely to be reported by single caregivers and caregivers looking after survivors closer to diagnosis. The  $r^2$  for this model was 0.255 that means 25.5 % variability of the outcome is explained by these two predictors.

**Table 31 - Multivariable model: psychological needs**

Model	B	P value	95% CI	VIF
In a relationship (0=no/1=yes)	-18.798	.040*	-.36.653 to -.943	1.049
Time since survivor diagnosis	-1.704	.026*	-3.192 to -.216	1.049

\*p<.05

### 5.10.3 Informational needs

Within the Informational needs domain, three variables were significantly associated with the reported level of unmet needs. At the univariate level there were negative correlations between informational unmet needs and caregiver relationship status ( $r^2=.285$ ,  $p<.001$ ), survivor current age ( $r^2=.116$ ,  $p=.031$ ) and time since survivor diagnosis ( $r^2=.116$ ,  $p=.031$ ). Indicating that caregivers that are not in relationships that care for younger survivors and those closer to diagnosis have more unmet informational needs. See Table 32 for the results of this analysis.

**Table 32 – Univariate regression analysis: associations between Informational needs and caregiver characteristics/survivor clinical factors**

Model	B	R <sup>2</sup>	P value	95% CI
Caregiver current age	-1.049	.065	.108	-2.340 to .242
Higher education (0=no/1=yes)	-.344	.000	.969	-18.030 to 17.342
In a relationship (0=no/1=yes)	-33.368	.285	.000*	-50.507 to -16.229
In employment (0=no/=1=yes)	4.486	.007	.591	-12.270 to 21.241
Survivor current age	-1.868	.116	.031*	-3.561 to -.176
Time since survivor diagnosis	-1.783	.116	.031*	-3.397 to -.168

\*P<.10

The three significant variables were entered into the multivariable regression analysis. In the final model, only relationship status remained the best fit predictor of caregiver unmet needs (B =-33.368, r<sup>2</sup>=.285, p<.001, CI =-50.507,-16.229). This indicates that single caregivers were more likely to report unmet informational unmet needs. The r<sup>2</sup> for this model was 0.285 that means 28.5 % variability of the outcome is explained by this predictor alone.

### 5.10.3 Health Care Service needs

After univariate analysis, two variables had an association with the reporting of unmet needs within Health Care Service domain (see Table 33). Both caregiver relationship status (r<sup>2</sup> =.329, p<.001) and time since survivor diagnosis (r<sup>2</sup>=.103, p=.043) had a significant negative association with unmet needs. Indicating that caregivers who are not in a relationship and who look after a survivor closer to diagnosis report more Health Care Service needs.

**Table 33 – Univariate regression analysis: association between Patient care and Support needs and caregiver characteristics/ survivor clinical factors**

Model	B	R <sup>2</sup>	P value	95% CI
Caregiver current age	-.443	.011	.508	-1.786 to .899
Higher education (0=no/1=yes)	7.559	.019	.394	-10.161 to 25.278
In a relationship (0=no/1=yes)	-36.266	.329	.000*	-53.059 to -19.473
In employment (0=no/=1=yes)	-.068	.000	.994	-17.079 to 16.942
Survivor current age	-1.389	.064	.116	-3.137 to -.360
Time since survivor diagnosis	-1.684	.103	.043*	-3.316 to -.052

P<.10



In the final best fit model only caregiver relationship status remained as a predictor of caregiver unmet Health Care Service needs (B=-36.266,  $r^2=.329$ ,  $p<.001$ , CI=-53.059,-19.473). This indicates that single caregivers are more likely to report unmet Health Care Service needs. The  $r^2$  for this model was 0.329 that means 32.9 % variability of the outcome is explained by this predictor alone.

#### 5.10.4 Work and social needs

Within the Work and social domain, two variables were significantly associated with the reported level of unmet needs. This indicates that single caregivers ( $r^2=.246$ ,  $p=.001$ ), and caregivers looking after survivors closer to diagnosis ( $r^2=.136$ ,  $p=.019$ ) were more likely to report work and social needs. See Table 34 for further details.

**Table 34- Univariate regression analysis: associations between Work and Social needs and caregiver characteristics/social clinical factors**

Model	B	R <sup>2</sup>	P value	95% CI
Caregiver current age	-.238	.005	.676	-1.384 to .908
Higher education (0=no/1=yes)	.898	.000	.906	-14.319 to 16.114
In a relationship (0=no/1=yes)	-26.724	.246	.001*	-41.859 to -11.588
In employment (0=no/=1=yes)	6.630	.022	.355	-7.682 to 20.943
Survivor current age	-.936	.041	.212	-2.430 to .557
Time since survivor diagnosis	-1.633	.136	.019*	-2.985 to -.280

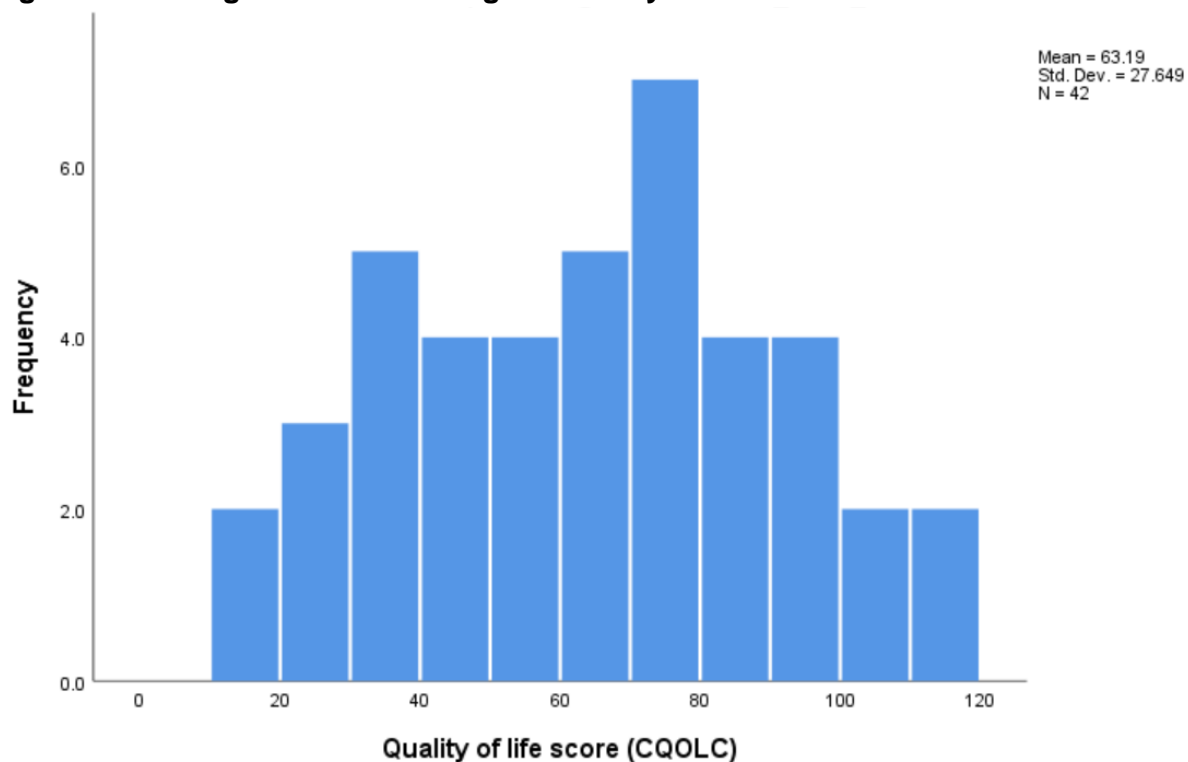
$p<.10$

Once entered into the multivariable model, only caregiver relationship status remained as the best fit predictor of caregiver work and social needs (B=-26.724,  $r^2=.246$ ,  $p<.001$ , CI=-41.859,-11.588). This indicates that single caregivers are more likely to report unmet Work and Social needs. The  $r^2$  for this model was 0.246 that means 24.6 % variability of the outcome is explained by this predictor alone.

#### 5.11 Objective 3: Caregiver unmet supportive care needs and caregiver QoL

As previously highlighted the third objective looked to determine whether unmet needs are associated with Quality of Life (QoL) outcomes. Caregiver QoL was measured using the CQOLC scale. The total possible score is 140, with higher scores representing better QoL. Caregiver scores ranged from 14-117, with a mean score of 63.19 ( $\pm 27.6$ ). See Figure 14.

**Figure 14 - Histogram to show caregiver Quality of life scores**



To investigate the association between caregivers’ perceived needs and QoL, Pearson correlation coefficients and p values were calculated. Correlations between caregiver unmet needs and QoL are presented in Table 35. The total number of unmet needs were significantly associated with overall QoL score measured using the CQOLC ( $r = -.616$ ,  $p < .001$ ). With  $r$  being between  $-0.3$  and  $-0.7$  this indicates a moderate negative relationship between caregiver unmet needs and caregiver QoL. This means that as one increases, the other decreases. Indicating that caregivers who had more unmet needs reported a lower QoL.

All of the SCNS-P&C needs domains were significantly negatively correlated with QoL. See Table 35. Indicating that as the number of caregiver unmet needs increases, caregiver quality of life score decreases (gets worse). The strongest negative association was between QoL overall score and Psychological and Emotional needs ( $r = -.652$ ,  $p < .001$ ).

**Table 35 - Caregiver quality of life score in correlation to total number of unmet needs**

		Quality of Life overall score (CQOLC)
Total number unmet needs	Correlation Coefficient	-.616
	P value	.000**

	N	41
Psychological and Emotional needs	Correlation Coefficient	-.652
	P value	.000**
	N	41
Informational needs	Correlation Coefficient	-.587
	P value	.000**
	N	41
Health Care Service needs	Correlation Coefficient	-.602
	P value	.000**
	N	41
Work and Social needs	Correlation Coefficient	-.530
	P value	.000**
	N	41

\*\*\* P<.001

## 5.12 Objective 4: Caregiver service and information use

Services used by caregivers are summarised in Table 36. The most used service, used by over half of parents was online brain tumour information (58.5%). Followed by counselling (by a social worker, psychologist or psychiatrist - 35.4%) and online support group, such as a Facebook support group (30.0%). The least used services included 24/7 online live-chat support, i.e. communicating through the internet to a trained support worker, in real time (2.5%) and informational workshops (17.1%). Even though these services were the least used services they were reported as desired services by around a third of caregivers (30% and 34.1% respectively). The service with the highest unmet need (caregivers wished to use but have not or have used but wanted to use more) were weekend retreats with other brain tumour survivors and their caregivers (47.5%).

**Table 36- Caregiver support service use**

Support Service	Individual response	N	%
Counselling	Used and have no further need	11	25.6
	Used and would like to use more	4	9.8
	NOT used but I would like to	8	19.5
	NOT used and have no need	18	43.9
Online information	Used and have no further need	16	39.0
	Used and would like to use more	8	19.5
	NOT used but I would like to	4	9.8
	NOT used and have no need	13	31.7
	Used and have no further need	6	15.0

In- person support group	Used and would like to use more	2	5.0
	NOT used but I would like to	7	17.5
	NOT used and have no need	25	62.5
Online support group	Used and have no further need	6	15.0
	Used and would like to use more	6	15.0
	NOT used but I would like to	2	5.0
	NOT used and have no need	26	65.0
24/7 Telephone support	Used and have no further need	5	12.5
	Used and would like to use more	2	5.0
	NOT used but I would like to	8	20.0
	NOT used and have no need	25	62.5
24/7 Online chat support	Used and have no further need	0	0.0
	Used and would like to use more	1	2.5
	NOT used but I would like to	11	27.5
	NOT used and have no need	28	70.0
Monthly social activities	Used and have no further need	1	2.5
	Used and would like to use more	6	15.0
	NOT used but I would like to	7	17.5
	NOT used and have no need	26	65.0
Weekend retreats	Used and have no further need	5	12.5
	Used and would like to use more	4	10.0
	NOT used but I would like to	15	37.5
	NOT used and have no need	16	40.0
Informational workshops	Used and have no further need	4	9.8
	Used and would like to use more	3	7.3
	NOT used but I would like to	11	26.8
	NOT used and have no need	23	56.1

Information resources used by caregivers are summarised in Table 37. Over three-quarters of caregivers indicated that they have received information related to their loved ones brain tumour side effects (78.1%). Over half have also accessed information regarding their family relationships (55.0%), and their loved ones diet and nutrition (53.6%) and future health (55.0%). Caregivers reported the least used/received information about their own health and well-being (35.0%) and their own finances (39.0%). Caregivers said they would most like to receive information (or more information) about their loved ones future health (41.5%) and their loved ones social well-being (39.1%).

**Table 37- Caregiver information resource use**

Information resource	Individual response	N	%
	Information received and no further need	11	27.5
	Information received and would like more	3	7.5

<b>Caregiver health and well-being</b>	NOT received but I would like to	12	30.0
	NOT received and have no need	14	35.0
<b>Caregiver finances</b>	Information received and no further need	13	31.7
	Information received and would like more	3	7.3
	NOT received but I would like to	8	19.5
	NOT received and have no need	17	41.5
<b>Family relationships</b>	Information received and no further need	0	44.9
	Information received and would like more	11	10.1
	NOT received but I would like to	10	18.8
	NOT received and have no need	20	26.1
<b>Loved ones side effects after treatment</b>	Information received and no further need	23	56.1
	Information received and would like more	9	22.0
	NOT received but I would like to	4	9.8
	NOT received and have no need	5	12.2
<b>Loved ones diet and nutrition</b>	Information received and no further need	21	51.2
	Information received and would like more	1	2.4
	NOT received but I would like to	9	22.0
	NOT received and have no need	10	24.9
<b>Loved ones social well-being</b>	Information received and no further need	15	36.6
	Information received and would like more	4	9.8
	NOT received but I would like to	12	29.3
	NOT received and have no need	10	24.9
<b>Loved ones future health</b>	Information received and no further need	18	43.9
	Used and would like to use more	4	9.8
	NOT received but I would like to	13	31.7
	NOT received and have no need	6	14.6

## 5.13 Discussion

### Key results:

- TYA survivors and their caregivers have needs for information and/or support services that remain unmet.
- Unmet psychological needs (e.g. support with anxiety) are the most pressing for survivors. Caregivers most frequently had unmet informational needs (around finance and fertility).
- Female survivors, unemployed survivors and survivors further away from diagnosis were more likely to have more unmet needs. While single caregivers are more likely to report more unmet needs.
- For both survivors and caregivers unmet needs were associated with poorer QoL.
- The strongest association was between survivor psychological needs and poor QoL.
- Counselling was one of the most used services by both survivors and caregivers. Caregivers had accessed online brain tumour information more frequent, while survivors had utilised online support groups more.
- Services most desired by both survivors and caregivers were focused around social interaction with other brain tumour survivors/families (e.g. weekend retreats).

### ***Objective 1: Unmet supportive care needs***

In this cross-sectional survey study, over three-quarters of survivors reported at least one unmet need now they were in long-term survivorship. These findings are similar to previous studies that identify a high number of TYA cancer patients (not brain specific) report having at least one unmet supportive care need.<sup>247, 248</sup> For instance, Zebrack (2008) found that up to 87% of TYAs expressed having at least one unmet information and service need, in most cases in regards to sexuality needs.<sup>247</sup> In this study, the most prevalent unmet needs identified were in the psychological domain, with around half of all survivors wanting support with anxiety, feeling down or depressed, feelings of sadness, and uncertainty about the future. Interestingly, counselling was one of the highest used services by survivors. However, this may have been earlier on in their survivorship, rather than a support service used more recently. The results show that unmet psychological needs were consistently the most pressing for TYA survivors. Therefore, it appears important that psychological support services should be available to patients not only in the short term but also in long-term

survivorship. These findings are reiterated by previous studies, who proposed a greater unmet need for long-term, post-treatment psychological interventions.<sup>249 250</sup> Fern (2013)<sup>249</sup> highlighted this may be due to the reduced availability of and extensive wait times for support services that exist in outpatient settings compared with clinical settings. However, these other studies were looking at needs in TYA mixed cancer cohorts, and did not include brain tumour survivors. There are no other studies that have looked at unmet needs in this survivorship group, so we are unable to compare results directly. This study is among the first to provide data concerning the supportive care needs of TYA, long-term brain tumour survivors.

Like survivors, the majority (88.4%) of caregivers also experienced at least one unmet need. This number is higher than other studies that have assessed caregiver needs. For example, Balfe (2016) found that around half of caregivers caring for an adult brain tumour survivor reported at least one unmet need.<sup>251</sup> The difference in study outcomes may be due to the majority of caregivers in the later study were partners, unlike in this study where all the caregivers were parents of young survivors. On the other hand, another study found that parents of children who are currently in treatment for cancer (mixed diagnoses) reported more unmet needs.<sup>252</sup> Aziza (2019) and colleagues found that 83% of parents reported more than 10 unmet needs, this data also found that under half of caregivers (46.5%) have ten or more needs.<sup>252</sup> These findings suggest that parents of children who are currently in treatment have more unmet needs than parents of long-term survivors. However, very few studies have addressed how caregiver problems and needs change during different stages of the illness trajectory. There is also little research about how variables such as social support, quality of life, employment and relationships change during the course of the caregiving experience. Thus, more longitudinal studies are needed to better understand how issues and needs vary over time.

Similar to survivors, caregiver psychological and emotional needs were pressing, with half (5/10) of the top ten ranked caregiver unmet needs belonging to this domain. Yet, the two most frequent caregiver unmet needs were in the Information domain, with nearly half of all caregivers (42.9%) wanting to know about possible survivor fertility problems and financial support/ governmental benefits. Interestingly, it was these information services that survivors also highlighted as most desired in the service use questionnaire. Therefore, these findings suggest that new information resources should be focused on both these two areas, aimed at both survivors and caregivers. It is likely that these are the most frequent needs of this population of caregivers because their loved ones are younger and these are or will soon be pressing issues. These unmet needs differ from caregivers of adult survivors, whose most pressing need was for support managing fears about recurrence.<sup>251</sup>

## ***Objective 2: Unmet supportive care needs and sociodemographic/clinical data***

The results are also able to highlight associations between survivor unmet needs and sociodemographic and clinical characteristics. The multivariable analyses indicates that unmet needs were more prevalent in females, those not in employment and survivors further away from treatment. Beginning with the first significant variable - female survivors were more likely to report more needs. These findings are similar to Boyes and colleagues (2015) who looked at associations within mixed cancer survivors.<sup>253</sup> They found that sociodemographic variables were more significant predictors of unmet needs than clinical ones and that sex (females) was associated with higher supportive care needs in survivors.<sup>253</sup>

Additionally, survivors who were unemployed were more likely to experience unmet needs. There is little in the literature that highlights the association between unemployment and survivor late effects/needs. Studies of adult survivors indicate that brain tumour survivors have difficulty securing and maintaining jobs, further indicating the need for support in this area.<sup>254</sup> It is reasonable to suggest that those who are unemployed have more needs as they are currently suffering more late effects. Unemployed survivors may also have more needs because of these late effects.

Survivors further away from treatment were also more likely to experience unmet needs. It is often thought that time since diagnosis mitigates the effects of cancer. Yet, this finding highlights the importance of long-term survivorship care, due to the new issues that may arise throughout their survivorship trajectory. These findings are similar to Keir et al. (2007), they found that that long-term adult brain tumour survivors were as likely to be categorised as “stressed” than patients closer to diagnosis.<sup>255</sup>

Some unmet need domains were associated with treatment variables. For example, survivors who had not had chemotherapy were more likely to report Health system and Information needs. These findings are unlike results found in the systematic review that indicated that those treated with chemotherapy and radiotherapy often suffered worse late effects than those who have not. These results may differ for a number of reasons. For instance, it could be that looking at whether someone has had a treatment or not is too simplistic to predict needs, as there are numerous other factors at play, such as chemotherapy dose, length of treatment and other adjuvant therapies. It could also be that those who received chemotherapy, received more/better information after their treatment than those who did not have chemotherapy, meaning that they have less needs.

The findings also highlight that long-term supportive services/care should pay attention to caregivers who are not in relationships. Single caregivers were more likely to experience



more unmet needs, than those in relationships. In fact relationship status was a predictive variable in each need domain of the SCNS-P&C. This may be due to single caregivers having less informal support and needing more formal supportive care services. Findings also highlighted that caregivers looking after survivors closer to treatment may also experience more psychological and emotional needs.

### ***Objective 3: Unmet supportive care needs and QoL***

Correlation analysis indicated that unmet needs were strong predictors for QoL of brain tumour survivors. Survivors with more unmet needs reported poorer quality of life. Additionally, the data found that different domains of supportive care needs made different contributions towards the quality of life scores. The strongest association was between poorer overall quality of life and unmet psychological needs. Again, reiterating psychological care is an essential area to target for significantly improving the general sense of survivors' quality of life. Previous research has examined the association between unmet needs and QoL and suggests unmet needs are a more significant associate of QoL in cancer survivors than sociodemographic or clinical characteristics.<sup>256</sup> In addition, previous studies with other cancer groups have found that addressing unmet needs leads to improved QoL.<sup>257, 258</sup>

As seen in the survivor sample, unmet needs were significantly associated with poorer caregiver QoL. All of the SCNS-P&C needs domains were significantly correlated with quality of life, the strongest being between QoL overall score and Psychological and Emotional needs. Again, this mirrors the survivor results. Therefore, it is important for both survivor and caregiver wellbeing that ways are found to better meet these psychological needs.

### ***Objective 4: The role and use of supportive services***

The findings also provide insight into various supportive care services and information TYA survivors have accessed. It is clear that TYA survivors have in the past or are currently locating and using support services and information. For example, over a third of all survivors have accessed online support groups (i.e. Facebook support group). Some services have rarely been utilised including, telephone support and informational workshops. Interestingly, even though mentor/buddy systems were one of the least used services it was one of the highest desired services that survivors wanted to use. A third of survivors indicated that they have not used but would like use of this service. The most desired support services, identified by over half of survivors was for weekend retreats with other brain tumour survivors and monthly social activities with other survivors.

The majority of survivors seemed content with the information they have received about their treatment side effects, diet and exercise. This may be because survivors receive this information regularly in their long-term follow-up care. However, there appears to be a high demand for information with regard to survivor finances and their fertility. Findings indicate that these information needs are going unmet for approximately half of the survivors, who are needing or desirous of this support.

Caregiver support service use varied. Online brain tumour information was popular, and had been accessed by over half of caregivers. However, other online support services were less popular such as online live-chat support. Caregivers most desired support service (identified by nearly half of caregivers) was weekend retreats with other brain tumour survivors and their caregivers. This was rated the highest survivor unmet need too. Consequently, these findings would support that these services should be prioritised for both survivors and caregivers.

Like survivors, the majority (over three-quarters) of caregivers have received information related to their loved ones brain tumour side effects. Again this may be because this information is provided in long-term follow-up care or because this information is readily available online. The information resource that was most desired was about their loved ones future health, therefore this is an area that information resources should focus.

### **5.13.1 Strengths and limitations**

There is growing agreement across all cancer types that there is a great need to meet the information and supportive care needs of those living with and beyond cancer.<sup>259, 260</sup> The data presented in this Chapter extends the very limited research in this area by gaining an understanding of the supportive care needs of TYA childhood brain tumour survivors and their caregivers. Unmet needs have not been well-researched in this population. The systematic review only found a handful of studies that have attempted to describe or map the supportive care needs of TYA childhood brain tumour survivors and their caregivers. All of these studies have small sample sizes ranging from 19-51 survivors, making this study the largest of its kind. This being said, the final sample size is smaller than anticipated. It was planned that there would be approximately 100 participants in each sample group (survivors and caregivers). However, due to the reasons detailed in Chapter 4, the end sample size was reduced to 69 survivors and 43 caregivers. This being said the literature supports that the overall response rate was acceptable for both survivors (69.4%) and caregivers (60.4%), as a 60% response rate has been set as a goal for researchers<sup>261</sup>

Furthermore, there were sufficient participants to perform the exploratory analysis that was necessary to answer the research questions (as described earlier in this Chapter).

The data highlights the impact that unmet needs can have on survivor and caregiver quality of life. Previously, unmet needs and QoL have not been measured within this patient group, in long-term survivorship. Moreover validated, comprehensive, and multidimensional instruments were used to collect data on QoL. The results showed that unmet needs were associated with poorer survivor and caregiver quality of life. This suggests the importance of identifying the specific needs of survivors and caregivers in order to improve their QOL.

Additionally, I believe the results are useful for both clinical teams and other current providers of supportive care, such as brain tumour charities. Primarily, the data highlights survivors and caregivers who may be more likely to have unmet supportive care needs in the long-term, such as survivors who are unemployed and caregivers not in relationships. Secondly, the data highlights the key areas of support they may be warranted, for instance psychological support (i.e. support with anxiety, or feeling depressed) was ranked highly by both survivors and caregivers as an unmet need. Thirdly, the data highlights the use of support services and what services and information survivors and caregivers would like access to. This data can be used to inform decision making around the design and planning of effective services to achieve tailored support that is effective and efficient.

Despite the numerous strengths of this data there are some limitations. First, sampling bias is a possibility. It may be that some survivors did not complete/return surveys because of high levels of anxiety, depression, or possibly because cognitive limitations prevented them from doing so. For instance, cognitive issues as a result of their treatment may have meant that some survivors encountered difficulty in understanding the content of the survey. In order to minimise the effect of cognitive impairment and/or reading ability, all survivors were offered support to complete the survey (i.e. reading the survey to them). It is also worth noting that the sample was purposively inclusive, only one patient was not invited because of too severe cognitive issues.

Furthermore, the majority of the sample was recruited from three NHS Trusts located in Yorkshire in the UK, which may limit the generalisations of the findings. However, pragmatically some survey participants were recruited via online advertising, this decision was made to increase the number of survey respondents. A concern with online recruitment is the accuracy of medical data recall (i.e. tumour grade/diagnosis/treatment), as you do not have access to their actual medical record data. To investigate the accuracy of self-report I analysed self-reported medical information with medical records data. In short, the analysis supported that survivors and caregivers were accurate in reporting treatment details, but

accuracy was lower for specific tumour information – such as tumour location and grade. Indicating that self-report is an acceptable alternative to medical record abstraction in certain instances, but data from medical record review remains preferred. See appendix 7 for a poster presentation that details this data analysis and methods.

Second, as described previously, the SCNS-SF34 and SCNS-P&C outcome measures are well-validated tools for investigating multiple dimensions of supportive care needs and were developed with diverse samples of individuals diagnosed with cancer (and their caregivers) in terms of cancer type and time since diagnosis.<sup>200</sup> However, it is possible that the SCNS-SF34 and SCNS-P&C may not fully capture the unique needs of cancer survivors and caregivers later in the survivorship phase of care. Therefore, this study may underestimate the prevalence of unmet needs reported by survivors and caregivers. Some of the items in the measures may seem unfitting for long-term survivorship as some items discuss treatment. Additionally, while each measure is relatively comprehensive, (the SCNS-SF34 covers 34 items across five domains and the SCNS-P&C covers 45 items across four domains) there are more brain tumour specific issues that are not included in these measures. For example, as seen in Chapter 2, brain tumour survivors often experience cognitive issues, yet, the SCNS-SF34 does not contain items specifically regarding unmet needs for help with memory or concentration. Furthermore, the SCNS-SF34 was designed as a generic measure applicable to the diversity of cancer survivors. Given that young adulthood is a unique developmental period, the use of the SCNS-SF34 may neglect to identify important concerns specific to young adulthood.

Third, as explained in Chapter 4 multivariable linear regression was chosen to analyse part of the data. Some literature suggests larger numbers to conduct multivariable linear regression models. However, by carefully pre-selecting variables to include in the multivariable models, I ensured that not too many predictors were added into the model. Additionally, alternative methods such as using non-parametric analysis and binary logistic regression were considered. Yet, these methods would have produced less informative results. For example, binary logistic regression cannot predict continuous outcomes. Therefore, logistic regression could not be used to determine the rise in unmet needs/need subscale scores because the scale of measurement is continuous. It would have been possible to attempt to convert the number of needs/need subscale scores into discrete categories (e.g. "<5 unmet need" vs. ">5 unmet needs") but doing so would have sacrificed the precision of the data set. This is an important disadvantage of this model. Therefore, even with smaller sample sizes, linear regression was considered the best method for these analyses.

## Chapter 6: Qualitative results

This chapter presents the results of the qualitative phase of the mixed methods study. The specific objectives of the qualitative phase were to describe the unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers (objective 1), and to explore the role and perceived use of support services in TYA survivors and their caregivers (objective 4).

The findings reported in this chapter are based on data from the in-depth interviews, field notes written during data collection and analysis and the free text responses of the survey. The Chapter begins with a description of the sociodemographic and clinical characteristics of the survivors and caregivers involved, followed by the structure of themes and categories that emerged from the thematic analysis. Each theme and its related categories are then demonstrated in detail. Pseudonymised quotes from the participants, extracted from the interviews are included to enable a rich understanding of each theme and category. Each quote is followed by the participants' sex and age in brackets.

### 6.1 Interview recruitment and procedures

Following completion of the survey, participants were approached to participate in semi-structured qualitative interviews. Of 21 survivors and 18 caregivers who were approached, 22 consented (11 survivors and 11 caregivers) and completed an interview between November 2018 and January 2020. Various reasons for non-participation were given, including not interested, not the right time or they didn't answer their phone. Data saturation was reached with the 22nd interview.

Interviews were conducted in the participant's home, or a private room in the hospital. Interviews averaged 53 minutes and ranged from 15 minutes to 140 minutes. Interviews were based on interview guides (see appendix 6), but enough freedom was left to allow the opportunity to explore potentially insightful avenues. Please see Chapter 4 for more details on the interview design.

### ***Reflections, learning and decision making...***

At around interview 18, I felt that there were little new data/themes being identified. However, as discussed in Chapter 4 the minimum number of interviews set was 20 (10 survivors and 10 caregivers). Another reason to carry on with the interviews was to ensure that the sample was varied enough (maximum variation). For example, there were few younger survivors included in the sample, so the following interviews aimed to include survivors under 18. There were no survivors under 16 included in the interviews, this was for two reasons. First, there were few survivors under 16 in the LTFU clinics – this was believed to be because they are still being treated in the acute setting. Second, one survivor who was 13 years old was asked to partake in an interview but her caregiver believed she was unable to do so because of her cognitive late effects. However, the caregiver did partake in an interview.

## **6.2 Participant characteristics**

The majority of survivors who volunteered to be interviewed were female (n= 6, 55%). Survivors were aged between 16-30 years old (24 on average). All were diagnosed between the ages of 1-10, the average age at diagnosis being 6 years old. Their tumour diagnoses varied but most common were a type of astrocytoma (n=4, 36%). This is representative of childhood brain tumour population as these are one of the most common histologies, as described in Chapter 1. Survivors received different treatment modalities, but all received either resection, re-resection, chemotherapy or radiotherapy. One survivor was diagnosed with Posterior Fossa Syndrome (S7). All but one survivor (S4) lived with their parents, not independently despite being over 20 years old. The majority were single, only two survivors were in a relationship (S4 and S9). Additionally, very few survivors had secured employment (n=2) and the majority were single (n=10). The detailed sociodemographic and clinical characteristics of the survivor participants are shown in Table 38.

Caregivers were all parents, the majority being mothers (n=10, 91%). Caregivers were aged between 40-61, the average age being 52. Caregivers were most commonly caring for a child who had been diagnosed with a Medulloblastoma (n=4, 36%). All caregivers currently lived with the survivor. The majority of caregivers were working either full-time or part-time (n= 9, 82%) and were married (n=8, 73%). More details of the caregiver participants are shown in Table 39.

**Table 38 - Survivor characteristics**

	SEX	AGE	EMPLOYMENT STATUS	AGE AT DIAGNOSIS	DIAGNOSIS	GRADE	LOCATION	TREATMENT
<b>S1</b>	F	16	Student	1	Pilocytic Astrocytoma	2	Cerebrum	R, RR, C and RT
<b>S2</b>	M	30	Unemployed – looking for work	6	Pilocytic Astrocytoma	1	Optic nerve	R, C and RT
<b>S3</b>	M	22	Unable to work due to illness/ disability	10	PNET	Not known	Brain NOS	C and RT
<b>S4*</b>	F	28	Working F/T	10	Pilocytic Astrocytoma	1	Occipital lobe	R, RR and RT
<b>S5</b>	M	18	Student	9	Oligodendroglioma	3	Parietal lobe	R, C and RT
<b>S6*</b>	F	27	Working F/T	5	Pilocytic Astrocytoma	1	Brain NOS	R, RR, C and RT
<b>S7</b>	M	24	Unemployed looking for work	10	Medulloblastoma	4	Cerebellum	R, C and RT
<b>S8</b>	F	25	Unemployed looking for work	7	Medulloblastoma	Not known	Not known	R, C and RT
<b>S9</b>	F	26	Unable to work due to illness or disability	2	Medulloblastoma	4	Brain NOS	C and RT
<b>S10</b>	M	30	Unable to work due to illness or disability	5	PNET	Not known	Cerebellar	R, RT
<b>S11</b>	F	17	Student	4	Anaplastic Ependymoma	3	Parietal lobe	R, C

C= Chemotherapy, F/T = Full-time, NOS= Not otherwise specified, PFS = Fosterior Fossa Syndrome, PNET= Primitive Neuroectodermal tumour, P/T= Part-time, R = Resection, RR= Re-resection, RT=Radiotherapy.

\* indicates survivors who were interviewed and their caregiver not.

**Table 39 - Caregiver characteristics**

	SEX	AGE	RELATIONSHIP STATUS	EMPLOYMENT	SURVIVOR SEX	SURVIVOR DIAGNOSIS	AGE	DIAGNOSIS AGE
<b>C1</b>	F	47	Married	Working FT	F	Pilomyxoid Astrocytoma	16	1
<b>C2</b>	M	58	Married	Working FT	M	Pilocytic Astrocytoma	30	6
<b>C3</b>	F	55	Married	Working PT	M	Pilocytic Astrocytoma	30	6
<b>C4</b>	F	56	Married	Working FT	M	PNET	22	10
<b>C5</b>	F	50	Married	Working FT	M	Oligodendroglioma	18	9
<b>C6</b>	F	53	Married	Working PT	M	Medulloblastoma	24	10
<b>C7</b>	F	61	Divorced	Working FT	F	Medulloblastoma	25	7
<b>C8</b>	F	54	Married	Caring for family/home	F	Medulloblastoma	26	2
<b>C9</b>	F	50	Married	Working PT	M	PNET	30	5
<b>C10</b>	F	49	Separated	Working PT	F	Anaplastic Ependymoma	17	4
<b>C11*</b>	F	40	Single	Caring for family/home	F	Medulloblastoma	13	4

C= Chemotherapy, F/T = Full-time, NOS= Not otherwise specified, PFS = Posterior Fossa Syndrome, PNET= Primitive Neuroectodermal tumour P/T= Part-time, R = Resection, RR= Re-resection, RT= Radiotherapy

\* indicates caregivers who were interviewed but not the survivor.



### **6.3 Themes**

Four over-arching themes were identified from the data analysis: 1) Preferences for support and support services (unmet needs) 2) Decline in support 3) Reasons for not obtaining adequate support 4) The role of long-term follow-up care.

Each theme contained a number of subthemes and codes that were created during the analysis (described in Chapter 4), as shown in Table 40. The detailed description of each theme and subtheme are presented below.

Survivor and caregiver accounts were intertwined in the analysis and narrative. However, it is made clear when there are differences between survivors and caregiver experiences.

Although there were differences within the lived experiences of the survivors and caregivers interviewed, there were themes that emerged across all of the dataset. There were also some themes that were more evident in some accounts than others.

**Table 40 - Summary of themes and subthemes**

<b>THEMES</b>	<b>SUBTHEMES</b>	<b>CODES</b>	
<b>1. Preferences for support and services (unmet needs)</b>	Achieving key life events	Finding employed work	
		Financial support	
		Independent living	
	Developing a social network	Declining friendships	
		Speaking to similar others	
		Organised support groups	
	Personalised, individualised support	Mental health support	
		Mentor/1-1 support	
		Social worker	
	Navigating the future	Preparing for long-term treatment effects	
		Dealing with the ever-changing landscape	
		How the survivor will manage in the future	
<b>2. Decline in support</b>	Life after education	Schools, Colleges, and Universities providing support (met need)	
		Decline in support after education	
<b>3. Barriers to not obtaining adequate support</b>	Practical barriers to accessing support	Not knowing where to go or what is available	
		Location and travel	
		Waiting lists, referrals and funding	
		Decline in older adult services	
		Accessibility of the information/support	
	Emotional barriers to support: "Getting on with it"		
	Reliance on family and friends		
	No confidence in support	Disappointed by support	
		Not suitable for brain tumour survivors	
		Online support reputation	
	<b>4. The role of long-term follow-up care</b>	The transition from children's to adults service	Uncertainty after moving from children's services
			Familiar faces
Change in survivor and caregiver roles			
Need for transition clinic			
The importance of follow-up		Reassurance	
		Around the clock support	
		Providing information for a better life	
		Signposting and finding support	
		Continuity - feels like family	
How follow-up care could be made better		Engaging with the survivor	
		Rounded/holistic care	

### 6.3.1 Theme 1: Preferences for support and services

This theme encompasses the unmet needs for support and services that long-term survivors and their caregivers experience after a childhood brain tumour. The theme covers personalised, individualised support, namely mental health support, mentor support, and having a social worker.

#### 6.3.1.1 Achieving key life events

##### *Employed work*

For both caregivers and survivors achieving 'normal' life goals such as paid employment is very important and the prospect of not achieving these caused concern. Over half of survivors (6/11) shared that they were currently having difficulty or were unable to find employed work. Several survivors had been trying for several years since their education finished:

*"I think what they usually expect once you've been to College is for you to go straight into work but for people like me...it's not as easy as that" [S10, M, 30]*

Due to late effects of treatment including sight problems, fatigue, physical disabilities and cognitive issues (slow processing speeds and memory) survivors have found it difficult to find employment. Not being able to find employed work meant several survivors felt low or depressed:

*"I do get depressed a bit and I did do when I was very much looking for work...so I have stopped looking for jobs" [S8, F, 25]*

Caregivers also expressed that this was also one of the biggest worries for them:

*"I guess the biggest challenge is keeping [survivor] boyant, cos since 2015 it has been a bit soul destroying you looking for work, you know. You apply, you get nowhere, you apply, you get nowhere, you apply, you get an interview, then you get nowhere!" [C7, F, 61]*

Caregivers expressed that they felt if the survivor could gain meaningful employment that it would give them: "confidence", "a reason to get up", "a purpose" and their "own money".

For those survivors who were still in education, their caregivers also worried about them finding employment in the future and the repercussions this may have if they cannot:

*“What if he wants to work and he can’t because they are discriminating against him... does he just have to live at home, not have a normal life, not be able to get his own place?” [C5, F, 50]*

Survivors felt that often employers did not have the knowledge or understanding about their brain tumour diagnosis and late-effect. They wanted employers to be given more information about a childhood brain tumour diagnosis and the potential late effects, which might help them get a job in the future.

*“I actually found it quite difficult to explain to them and to get them to understand that oh sorry I can’t quite pick that child up or whatever or things like that...which I did find quite difficult and I don’t feel that they [employers] quite understood.” [S8, F, 25]*

Survivors (5/11) said that they would like more help with gaining employment. Unmet needs for support included: help with finding local businesses who are ‘disability friendly’, help preparing CVs/application forms, and interview support. One caregiver reiterated that this support was necessary *“especially long-term, after you know your 10 years plus down the line after treatment”* [C4, F, 56]. All survivors currently looking for work said they had received support from the Job Centre. This support had helped some survivors to secure work placements that they had found useful (if only short-term), but for others the service had not been as useful:

*“help with employment, that’s a big thing – employment, cos the Job Centre isn’t really helpful, I went down there before and they just said well basically can’t help – just go on the website” [S7,M, 24]*

Over half of the survivors (8/11) were currently volunteering or had recently finished volunteer work. It seemed that volunteering was incredibly important for survivors while they are unable to do paid work. Survivors and caregivers said volunteering allowed them to: gain skills like *“interacting with people”* [C10, F, 49], made them feel like they were *“doing summat”* [C3, F, 55] and took into account their abilities: *“they just tell you to do as much as you can do”* [S10, F, 49]. Some survivors recognised they would never be able to get paid work because of their late effects, which made volunteering was even more important:

*“because I can’t work...which is really hard for me and I find it really hard an upsetting...cos I’m not that sort of person I can work...and that’s why I’ve got to do volunteering or something like that” [S9, F, 26]*

A long-term volunteering placement had helped one survivor [S6] get paid employment, something she had struggled to achieve for many years because of low educational grades. Through volunteering she had been able to gain the skills and experience necessary to

apply for a job. Caregivers also seem to benefit from survivors volunteering, as they were often responsible for the survivors on a day-to-day basis:

*“it’s lovely [survivor volunteering] because he walks out the door and I say “right I’ll see you later” and he says ok and for me that’s great cos...I get that break as well...cos before the only time [survivor name] got out was if I took him and some days I didn’t want to do it – it’s tough!”* [C9; F, 50]

### **Financial support**

Financial difficulties were a key issue as survivors were getting older and entered young adulthood. For instance, some survivors had their benefits taken away when they turned 18 years old. Survivors’ financial issues were a major concern to caregivers, in particular if and how the survivor could support themselves.

*“The main challenges, well is finance, financial...Its stressful cos I want [survivor name] to have some income, you know, cos I can’t support her, do you know what I mean...and that’s what worries me.”* [C8, F, 54]

Survivors discussed financial support needs less, it appeared this may be because caregivers managed their finances and financial support. However, one survivor did say that she found it upsetting that her mum is “stressed” about completing the forms. She also said it worries her to think she may not receive any financial support in the future:

*“I properly need that money...if I don’t get that money it’s going to be hard for me, because I can’t work...which is really hard for me and I find it upsetting. Every time my Mum has to write a form out, and we don’t know if I’m getting it or not, so it’s just really scary.”* [S9, F, 26]

Interview data and free text survey data highlighted the difficulties of completing financial forms, particularly the Personal Independence Plan (PIP). Over half of the caregivers interviewed (7/11) discussed encountering problems with finance/benefit forms:

*“I mean just filling in a PIP– you need a degree to do it, it’s that difficult! Filling in anything like that is really really hard!”* [C7, F, 61]

Even caregivers who described themselves as “fairly articulate”, educated and having had experience with similar forms, still found this process challenging. Caregivers said that completing these forms could be a lengthy process and particularly difficult alongside their other responsibilities:

*“Those forms [PIP] are monumental aren’t they?! I must have submitted about 50 pages all in all and I was actually trying to get my shoulder treated to at the time, and*

*I was going through the divorce, taking my Dad to different oncology appointments”.*  
[C10, F, 40]

Caregivers also described completing financial support forms as “*emotionally wearing - you have to write down all your child’s faults and short comings*” [C10, F, 49]. For one caregiver it particularly affected her well-being:

*“It makes me feel sick when I fill those forms in. And I have to do it over a week, I have to be on my own, quiet, do it gradually, think about it, write it down – yeah it’s awful.”* [C8, F, 54]

One of the “*big jobs*” in completing the forms is providing evidence to support the claim, however, caregivers said they have found this harder as the survivor moves further away from treatment:

*“I’ve got old things about her treatment that she’s gone through but I have nothing current and that’s what frustrates me cos when you go to the doctors they say they don’t give out reports for ESA (Employment and Support Allowance)...so where do I get all this information about [survivor’s] disability?”* [C8, F, 54]

Only two caregivers had received any support with completing financial forms, this help was sought from cancer charities. Both caregivers had actively sought out the help of the charities to ask for benefits advice and help completing the financial forms. Support was described as a “*god send*” [C11, F, 40], with advisors guiding parents through the form and offering to check the final form. Other caregivers had no formal support but had sometimes called on family or friends to help.

Caregivers identified that more brain tumour specific guidance to finance support and benefits system was needed because “*that information wasn’t just there*” [C10, F, 49]. One caregiver explained that because the survivor does not have a current medical diagnosis that there is in theory “*nothing wrong with him*” which can make it harder:

*“you know if it said that [survivor] was autistic or that he was down syndrome then there are certain guidelines that they can say well actually these things he may encounter, but there’s nothing for [survivor] as there’s nothing wrong with him.”* [C9, F, 50]

Caregivers referred to wanting 1-1 help with financial support:

*“Again I think it comes back to that support, I think it comes back to that one person that can point you in the right direction of knowing what to put on the forms cos the stress level!”* [C9, F, 50]

### ***Independent living***

Difficulties with employment and finances were linked with independent living. Nearly all of the survivors were still living with their parents (10/11). Two survivors, both in their early 20s were actively looking to move out the family home but were waiting for help with supported living:

*“I know he could never live independently, so he’s on the waiting list for sheltered housing round here but unfortunately the average age around here is about 70 so he’s not a priority at the moment”* [C6, F, 53]

Caregivers highlighted that some survivors would never live completely independently, as they required daily support (5/11). Physical late effects of treatment, such as vision impairments meant that they were unable to do daily tasks, such as meal preparation or driving. Other survivors needed assistance with most tasks as a result of their learning disabilities:

*“I have to do everything for her, she can’t really do an awful lot. So the getting up stuff in the morning– breakfast, getting ready, going out, getting on the taxi to go to school.”* [C11, F, 40]

Even survivors who had minimal late effects were still unable to travel independently: “we have to take her everywhere, so she’s not independent in that way” [C1, F, 47]. In most instances this was mainly due to sight or mobility problems: “he can’t go out independently... to get on a bus he has to go across the road and he couldn’t see to get across the road” [C4, F, 56], and their caregivers didn’t see this changing in the future: “she’s always going to be dependent on somebody else to travel” [C10, F, 49].

Many caregivers worried about the survivor’s future independence. For caregivers a major concern was what the survivor would do when they were no longer able to care for them:

*“That’s one of my biggest fears, not saying [name of wife] couldn’t look after him but...erm if owt happened to me then how would they cope?”* [C2, M, 58]

Caregivers expressed a need for more information about the support available to help the survivor transition into independent or assisted living. This would help them plan ahead and decrease their anxiety about the future.

### 6.3.1.2 Developing a social network

#### ***Declining friendships***

Most survivors and/or their caregivers discussed that forming social relationships has been difficult in long-term survivorship. For older survivors this has become harder as they have got older and their peers have moved on:

*“the friends he went to school [with] or the friends he’s sort of like had, because of his age now they’re either married with kids...so he has no friends as such that come to the house or anything like that, it’s more family isn’t it”* [C3, F, 55]

One survivor had struggled to maintain any friendships formed at school and this had been particularly difficult for him as he had got older:

*“Yeah seeing all the people that used to know me – getting jobs and partners and god knows what else and just not seeming to care about me...just yeah growing up and forgetting about me.”* [S7, M, 24]

Some survivors indicated they would like support with making and maintaining friends, finding it difficult to interact in social situations. Three survivors (aged 24-30) said this was currently their main challenges and area of unmet need:

*“I think one of my main issues is probably friendships and relationships more than anything, it’s like – I kind of don’t know...I don’t know much about them to be honest.”* [S10, M, 30]

For other survivors the late effects of treatment had stopped them from being able to go out independently with friends, or they said they worried about being in social situations with their friends, *“like in my head I’m thinking “don’t trip up, don’t trip up”* [S1, F, 16]. This may be one of the reasons that several survivors value virtual friendships, spending a lot of time gaming online with others:

*“Most of the time I just go on the Xbox with my friends and they’re like me – they don’t go out much”* [S5, M, 18]

#### ***Speaking to similar others***

Several survivors, including participants who completed the survey said that they would like to meet others like them. They often felt different to others their age due to the late effects of treatment, and some found it difficult to socialise with others because of this. For example, one free text respondent said that they had low confidence in social situations and socialising in groups as they were conscious of their hearing loss. If survivors had the



opportunity to socialise with others with similar late effects they may feel less uncomfortable and more accepted. Survivors said that they would like the opportunity to talk to other brain tumour survivors of a similar age and to do 'normal things':

*"Like arranging to see something at the cinema or to meet up and have a chat with people, like over a drink or a meal or something."* [S1, F, 16]

Caregivers even more so said that they would like their child to have the opportunity to socialise with similar others:

*"I think it's a brilliant idea though cos they've all got something in common, they don't have to be talking about that all the time but they can all find this common ground."* [C8, F, 54]

For some caregivers they said this would ease one of their biggest concerns, which is worrying about their child's lack of social life *"I think the worst thing for [survivor] at his age is that he's not got a social life"* [C3, F, 55].

### ***Organised support groups***

Survivors were enthusiastic about organised support groups. Two survivors had attended brain tumour specific social groups that were organised by charities. The feedback from their caregivers was very positive and they felt they were a good place for the survivors to feel normal, which is also a relief for them:

*"At school she has had a lot of problems with them bullying her... because people constantly say about her hair and stuff...Where as if you go somewhere with like the braintrust they are not going to say that cos they've been through it, seen it, know it. And she won't be the only one that's like that either, so that's an element you don't have to worry about."* [C11, F, 40]

Others were interested in attending brain-tumour specific social groups but had not attended for reasons including: they did not know of any support groups like this or the groups were too far away/not accessible.

Caregivers also said that they would like the opportunity to speak to other parents that have been through similar experiences. Caregivers discussed that when the survivor was in treatment that they had a lot of contact with other families, but this contact had stopped after treatment. Some caregivers said that they had attended charity support groups with other parents when their child was in treatment or shortly after treatment, but this was not something that had continued in long-term survivorship:

*“They also organised for parents to go for a meal all together...and that was nice to talk to parents who were in a similar situation to us. But that was only the once, but there’s never been anything else we’ve been invited to.” [C3, F, 55]*

Some caregivers said they were still interested in support like this as they think it would still be beneficial as long-term survivorship can be a lonely experience: *“[I’d like] to be put in touch with likeminded mums or I don’t know, anything...someone to talk to really” [C6, F, 53].*

Even with virtual friendships being common, survivors generally said they would prefer to meet others face-to-face in a group instead of online. One survivor said:

*“I feel more confident being in a group and everyone sharing their ideas and what they’ve gone through and whatever, and I know that sometimes I don’t feel confident talking about it and sometimes other times people don’t...but you know what I mean just sitting next to someone...and not even going into detail about it ...just having a general chit chat about life I suppose” [S8, F, 25]*

However, another survivor said that she would be interested in using social media to connect with other survivors, as she said she had never taken part in a “group chat” and said that would be something she would definitely like to do.

### **6.3.1.3 Personalised, individualised support**

#### ***Mental health support***

Mental health services were indicated as a crucial form of support. The majority of families had received counselling or seen a psychologist when the survivor was in treatment, but many had not had any support as the survivor had got older. Survivors explained that this support was still needed during long-term survivorship due to a growing understanding of their diagnosis, and the experience of late effects:

*“I had counselling when I was 7 but I still want help having moved on now I’m 25. I don’t want that help to have just stopped...cos there’s still things that are happening and changing... the side effects never leave you if you know what I mean?” [S8, F, 25]*

In the free text questions on the survey one survivor had said that they had been offered counselling at the time of treatment but they had turned it down because they thought they could deal with it alone. However, they said that as the years had progressed they had become more and more aware of the negative impact the brain tumour had on their

mental health, and realised mental health support now would be helpful. Therefore, this finding suggests that it is important that mental health support is offered to survivors continuously through survivorship and into long-term care.

Two caregivers were actively trying but struggling to access mental health support for their loved one. One caregiver [C9; F, 50] described how they had been desperately trying to access a psychologist's support for her son for nearly three years and had only just accessed the support. Another caregiver was told by local mental health services that they currently had no one in their team that would be able to meet the survivor's mental health needs, so she was still looking for support [C11, F, 40].

Most caregivers said they had not been offered any mental health support as their child had got older. Many caregivers did not seem to prioritise their own mental well-being needs very highly, often coping on their own. When asked many shrugged off their own needs. However, four caregivers identified that they wanted mental health support as the survivor had got older. One caregiver said:

*“There has never been any kind of support for how to deal with the aftermath and how to deal with what might be to come...which is really what you need because at the time your firefighting and you get on with it but then obviously there is a sort of PTSD element for want of a better phrase” [C10, F, 49]*

Survivors and caregivers who had recently received mental health support suggested improvements, most frequently mentioning more support/sessions. For instance, one survivor [S4, F, 28] recognised that the mental health support she received via a brain tumour charity had been helpful but felt more sessions would have been beneficial. However, she was conscious of their limited resources and did not want to prevent others from accessing the support by taking up too much time.

It was important to both survivors and caregivers that mental health support was provided by someone with knowledge of brain tumours, to help guide them through life experiences, complicated by late effects:

*“CAMHS (Child and adolescent mental health services) worked really well for us because she had the knowledge, she really understood it – she'd been a nurse for like 30 years or something so she really really got it and I think that's very rare that you get that...cos I think otherwise it's not going to be as effective” [C11, F, 40]*

Some survivors described the mental health support they had received as not adequate/suitable: one survivor felt the counsellor was trained to a very basic level [S4, F, 28], another said he felt “*under pressure*” and would rather speak to his friends instead [S3,

M, 22], and another survivor did not have the chance to finish the treatment when he badly needed it:

*"[Survivor name] went through a period where he was self-harming...and a psychologist came maybe 2/3 times and then we never saw him again, not even a text to say he wasn't coming anymore." [C6, F, 53]*

### **Mentor/ 1-1 support**

Some survivors stated that now that they were older, they would appreciate being able to contact a mentor or somebody that could provide 1-1 help. Two survivor and caregiver dyads in particular felt this was one of their most important unmet needs. The main need for a mentor seemed to be having someone to contact when they needed information or signposting in direction of support:

*"I would like there to be like a mentor or a person who you could ring...somebody that you could contact to talk to about that certain thing that you want information on. Yeah, yes that would be really useful" [C8, F, 54]*

*"To know that that persons there if you need them, and they will be able to provide you with an answer as to whatever you're going to ask them if you know what I mean" [S8, F, 25]*

For survivors it was important that this mentor specialised in brain tumours, so that they had the knowledge to support them with things like job applications:

*"like when I was filling in my job application form they [mentor] could say, yeah do this, but don't forget you have gone through A,B and C" [S8, F, 25]*

Caregivers said there was more of a need for this mentor role now (as they had got further away from treatment) because they were finding it harder to know where to go for the support:

*"We don't have like a point of access...your just knocking on anybody's door until somebody might answer and for me...that's tough cos you're on your own sort of thing...so I've very much felt that I've been on my own, ever since him being 14/15" [C8, F, 54]*

### **Social worker**

The majority of caregivers described that a social worker had been a key source of support when their child was diagnosed, but this support had stopped as the survivor had got older:

*"I don't have a social worker, we've never had a social worker apart from when [survivor] was first diagnosed, which they do to get you through the cancer treatment and stuff, but now I don't have anybody" [C8, F, 54]*

Caregivers described their social workers during treatments as the person who *"helped us with everything"* [C9, F, 50], *"got all the ball rolling about getting things done for us"* [C3, F, 55] and *"the one that I rung"* [C9, F, 50].

Nearly half of caregivers [5/11] expressed a current need to have a social worker.

Caregivers explained that this would not need to be daily support but instead that it would be useful to have access to a social worker when necessary. One caregiver said that a social worker would be useful because of their knowledge of local available support:

*"I still don't know what I can access in my own town - I still don't have that information but again I think it's because we don't have a social worker...I find things out from other parents sometimes but I don't have anyone to access to ask for the information!" [C11, F, 40]*

#### **6.3.1.4 Navigating the future**

##### ***Preparing for long-term treatment effects***

Caregivers recalled that during the survivor's diagnosis and treatment, they were not always aware of potential late effects. Subsequently, families were then shocked when the survivor had been diagnosed with late effects that they did not realise were a side effect of treatment. For example, one family were surprised when the survivor was diagnosed with epilepsy many years after treatment. Another caregiver felt that she was not fully informed about many of her child's late effects and described the loss and thinning of the survivor's hair as she had got older:

*"With her hair we ultimately ended up in dermatology and Professor dermatology goes "oh yeah you've had radiotherapy that is something that often happens" and it's like well why didn't you just tell us that years ago...I just wish someone would have outlined future problems that could occur" [C7, F, 61]*

Caregivers said that the time during treatment was a "blur" and they could not see beyond treating the cancer. Therefore, they felt this was not the appropriate time to be provided with information about potential long-term effects. One caregiver admitted that she knew the clinical team had talked her about her daughter's chemotherapy treatment and potential late effects, but she had now forgotten that information and is now trying to figure out where to get the information from:

*“I’ve kind of had to go back over it myself to make it make sense again now, cos I think you’re given a lot of information in the early days but it just doesn’t go in... and then it’s now that I need that information but there isn’t anyone there now to give it to me.” [C10, F, 49]*

Another caregiver had been provided comprehensive information by a brain tumour charity, including a large book when her daughter was diagnosed but she did not have the capacity to process the information at that time:

*“It was like I can’t take any of this in? I couldn’t get dressed on some days so I can’t focus on reading something like that” [C11, F, 40]*

Caregivers highlighted that better timing of information on late effects and the need for ongoing contact and support alongside survivors’ changing needs is vital.

### ***Dealing with the ever-changing landscape***

Survivors and caregivers highlighted that their needs were constantly evolving and changing as the survivor grows older.

*“It’s not just one thing with the late-effect that’s the thing, it’s this ongoing thing – you might not think that you have a problem in one area until you get there and then suddenly there is a little problem.” [C9, F, 50]*

As they got older, survivors came across new milestones and hurdles. Some were expected, such as trying to find employment (as discussed earlier). Some were less predictable and harder to cope with, such as two survivors developing epilepsy in their early 20s. For one survivor this meant she felt unable to leave the house or continue looking for employment:

*“When this epilepsy started I was like “oh god not another thing” ...and then that you know unfortunately it has gone into me trying to find a job...you know it’s carried on into different things.” [S8, F, 25]*

While most caregivers had been able to go back to paid work several years after their child’s treatment, some were still adjusting to long-term survivorship. One caregiver had recently stopped paid work to become a carer full-time, as she was struggling to balance her caregiver role and work. She said that other parents in similar situations had also done this:

*“I’m not the only parent out of my group of oncology parents that have given up work...and they’ve only just done it recently so obviously there is a trend there cos they’ve all changed jobs or gone part time or they’ve changed things – but it has been quite a bit down the road and I do think it’s because all of a sudden it’s a bit like – actually this is not going to get better, we can’t sustain what we’re doing now cos*

*actually the young person or child needs more support, there isn't the support there in other formats so it's going to have to be as a family that we change what we do."*  
[C11, F, 40]

### ***How the survivor will manage in the future***

Caregivers shared that often the prospect of what is going to happen to the survivor in the future was a daunting one. Caregivers who provided daily care for their child worried who would take on that responsibility. For survivors this issue played on their mind too:

*"A couple of years ago, it was one New Year...we sat in together and he went "but mum what am I going to do when you're not here?" and I said I'm not going anywhere yet...but I think he's got to the age 20/21 and he's thinking oh mum and dad aren't going to be here forever and I think he just had a bit of a thought what, how is life going to carry on in the future you know, for him" [C4, F, 56]*

Another survivor was upset because their caregiver worried about the future:

*"It's about when I get older and my Mum not being there. She's more scared about the future and what's going to happen, and that upsets me sometimes thinking about it...seeing my Mum get upset and stressed"[S9, F, 26]*

## **6.3.2 Theme 2: Decline in support**

This theme encompasses the decline in support that both survivors and caregivers can feel as the survivor gets older, and the new challenges associated during this time. The theme starts with the support that survivors received during their education. It then goes on to highlight how this support diminishes for many survivors as they finish education and how this can be a struggle for both survivors and their caregivers. This theme also covers the decline in support as the survivor moves further away from treatment and into long-term survivorship.

### **6.3.2.1 Life during and after education**

#### ***Schools, Colleges, and Universities providing support***

The majority of survivors and caregivers described support from education generally as positive. Support varied for each survivor. Only one survivor went to a Special Educational Needs school, the rest received mainstream education. Two survivors needed a support assistant with them at all times. Six survivors received Special Education Needs support in a mainstream school. Other survivors needed extra, 1-1 help due to their cognitive issues such as slower processing speeds:

*“I have needed support in school, like in Geography I really struggled in year 11 in Geography, and like they like arranged for extra support to come in...cos it was just really challenging in the class to just understand what was going on as they were working quite fast and it was really hard.” [S1, F, 16]*

Other support provided included additional time in examinations, access to a reader/writer and adapted educational aids.

It was clear from the interviews that high schools and colleges were often given formal advice and guidance to help them support survivors. For instance, three caregivers told us that vision support officers would come in every term to assess the survivors learning environment and then feedback to teachers and support assistants. Although two survivors said that sometimes they had to remind or “badger” their school for the extra support at times. Both survivors said they felt this was because they often managed without the support that they forgot. One survivor who needed enlarged text due to her vision loss said that quite often this was not provided:

*“I think it’s just because I don’t look like there’s anything wrong and generally there isn’t I just needed those things and I think they just forget because they seem me coping and getting on with it and they just think – oh she’s fine she’s getting on with it! But it shouldn’t be down to me, saying “I do actually need that!” [S11, F, 17]*

Overall caregivers were very complimentary about the support provided by the survivor’s education providers. Most caregivers said that they felt the survivor’s school was adequately informed about the survivor’s tumour history and their needs because of their late effects. For example, survivors were sat at the front of their class if they had hearing or sight issues; staff were given epilepsy training; supply staff were educated on survivor needs; regularly assessed the survivors to see if their needs had changed.

However, some caregivers indicated that support from the schools could have been improved. Three caregivers felt that survivors could have benefited from electronic equipment that would support some of the late effects of treatment, such as memory and slow processing speeds:

*“She could have done with a lot more – so maybe to have things on Kindle’s or electronic devices rather than on a whiteboard which obviously reflects everything, cos it’s the looking up – the processing from looking down to looking up and remembering that” [C10, F, 49]*

Survivors also reported receiving similar support when they went to College and in one case University. Namely, extra time to complete assignments, 1-1 support and modified/extra



materials (e.g. provided laptops). Four survivors completed College courses that were specialised for people who had learning difficulties. These courses were often focused around practical life skills. For many caregivers further education was another way of keeping the survivor occupied after High School:

*“She was on the kind of courses that were for people with learning disabilities she got on very well, she enjoyed going and it was something for her to do more than anything. When she left school that was just another way of keeping her occupied and her mind occupied by sending her to College”* [C8, F, 54]

### ***Decline in support after education***

Both survivors and caregivers shared that it was a difficult period for the survivor after they had stopped education. Firstly, because the support given during education was no longer there:

*“After Uni I just felt left...I felt that I didn’t have any support at all really”* [S8, F, 25]

*“When [survivor] was at school it was a lot easier...yeah because you had the support...cos she was SEN, so she had educational needs support”* [C8, F, 54]

Secondly, and more importantly for survivors they felt at a loss of what to do next:

*“When I finished college it’s like – what am I supposed to do after that?”* [S10, M, 30]

*“After coming out of college from then on everything went downhill. Erm that for me was really difficult”* [S6, F, 27]

For many caregivers this was also a challenging time, as they did not know what to do to support the survivor:

*“He was as happy as Larry going to college. But then it ceased cos he turned 21 didn’t he? There were a gap between there and 30 year olds.”* [C2, M, 58]

For some caregivers it meant that the survivor is more dependent on them for daily support:

*“The boredom aspect – he’s bored a lot of the time, especially now he’s not at Uni – “what are WE doing today?” you know.”* [C6, F, 27]

For parents of younger survivors it is something that was a concern for the future:

*“At the minute it’s very easy cos she goes to school, in theory I could probably keep her in an education setting until she’s 25 but then what? What happens then?”* [C11, F, 40]

### 6.3.2.2 Diminishing support getting further away from treatment

All caregivers were particularly positive about the support they received from clinical teams, charities and support services while their child was in treatment or acute care (usually under 5 years from diagnosis):

*“I think we got the most support when he literally started his chemotherapy and radiotherapy, we seemed to have lots of charities available for us and giving us advice.” [C5, F, 50]*

It is clear from the interviews that this support was instrumental in caregivers being able to cope during the diagnosis and treatment phase. However, many caregivers felt that support fell away as the child moved further away from treatment:

*“At the time it was just hospitals all the time. And I think [survivor] felt quite safe and I think it sounds quite a strange thing to say but we both felt quite safe but when you come out of hospital...you feel really lost” [C4, F, 56]*

Caregivers and survivors described being unable to access the support services they had once they reached adulthood:

*“I think the last time he saw a speech therapist he was 13, the last time he saw an occupational therapist he was 16, the last time he saw a physiotherapist – possibly 18... I mean he used to get hydrotherapy – that got stopped...once he turned 18.” [C6, F, 53]*

Also some support from charities became unavailable or less available as they moved further away from treatment. One caregiver talked about the survivor now being on a charities ‘B list’ as the children who were currently on treatment were prioritised to access the support:

*“He used to go once a month [social event]...but because there’s so many young cancer patients coming up now they had to split into two groups – ‘A’ priority and ‘B’. And because [survivor] has been going since he was in there, he’s on the B list now. So he doesn’t get invited...every month.” [C2, M, 58]*

The support not only ceased for survivors but caregivers lost support too, with relief care stopping for some:

*“I always said once he turned 18 he still needed looking after, he still needed care but a lot of what we had prior to him being 18 was taken away from us and not replaced. Like I used to get 48 hours a month respite” [C6, F, 53]*

### **6.3.3 Theme 3: Barriers to not obtaining adequate support**

This theme covers the many reasons that survivors and caregivers may not receive support even though they have unmet needs. The theme first describes some of the more practical aspects of attaining support, such as long waiting lists or support not being in suitable locations for survivors and caregivers. Next, survivor and caregiver emotional attitudes (“getting on with it”) were explored as a reason to why they have never accessed support. The theme then highlights the importance of informal support from family and friends, and why this support is sometimes preferred to other types of support. Finally, the theme identifies how survivors and caregivers have been let down by support in the past or their worries about support offered that then prevents them from accessing support now.

#### **6.3.3.1 Practical barriers to accessing support**

##### ***Not knowing where to go or what is available***

All caregivers said in at least one instance that they were either not aware of the long-term support available, or how to access support:

*“We didn’t really know what other support groups were in place...so we didn’t really know anyway to where you would go and look.”* [C1, F, 47]

One caregiver talked about her current issue of where to get help with getting her daughter a new wig, she felt the information about available support is not accessible:

*“I think some of the specialist stuff that we don’t know about – I don’t know what’s available in terms of like when she got her wig I didn’t know I could use my NHS prescription to go to any wig maker, I just thought you go to that person cos that’s where they told me to go – but then I got a crap wig... I think there isn’t necessarily the information about what’s available and what you can do, where can you go, who can you use? And I don’t know how you would find that out?”* [C11, F, 40]

Many survivors and caregivers believed that they should be made aware, by clinical teams and charities, what support is available in long-term survivorship and how to access it:

*“People shouldn’t wait for us as the sort of clients you know to have to say...they should be asking us cos we don’t know what’s on offer – they need to tell us”*  
[S8, F, 25]

Two caregivers said that all the support they have received they had to actively “hunt” for it. Caregivers said it would be beneficial if there was someone who could provide information and highlight what support was available:

*“If there was someone that could actually sit you down and go “there’s this and that, and we can help you get that started”, that sort of thing would be good. Just for her life as a young adult.” [C10, F, 49]*

It stands to reason that if survivors and caregivers were not aware of the long-term support available that this support will not be accessed when needed.

### ***Location and travel***

Many caregivers (6/11) said they had not accessed the support they felt they needed because it was not accessible due to its location. Many survivors were reliant on their caregiver for transport to and from support services. Therefore, if they were located a long way from where the survivor and caregiver live it is not feasible for them to attend. Families who lived in more rural areas, further away from cities appeared to suffer most:

*“A lot of the problem we have is because we’re in this dead zone – we’re surrounded by big cities, it means we have to travel to the City to do anything and I get that. I know why but [survivor] can’t travel to them places without me, which then puts the burden back onto me cos it’s like – I’m working, I’m doing this and that...I don’t have time! Which is awful for me cos I feel awful saying, “I don’t have time to take you”.*  
[C9, F, 50]

Many survivors were not able to travel independently, either because they cannot drive or they were not capable of using public transport because of their physical (i.e. mobility or sight deficiencies) and cognitive disabilities. Caregivers were unable to transport survivors to support services due to working/not having the time (especially single parents), money (cost of fuel) or not being able themselves (i.e. the caregiver cannot drive). One caregiver especially thought that where they lived and not being able to access support services had negatively impacted her son in the long-term, saying that he was very “isolated and lonely” but also:

*“I just think that [survivor] would have been a different person had he lived in Wakefield or Leeds or Manchester or Birmingham, I think he would have perhaps been a lot stronger, a lot more mobile than he is now because of where we live. But that isn’t anyone’s fault, it’s not ours, it’s not the NHS, it’s not anybody it’s just as the saying goes and is used a lot – a postcode lottery. But unfortunately it affected my child, my son.” [C6, F, 53]*

Caregivers said that it would be helpful if there was transport provided for survivors who were further away from support services so that they could attend. Two caregivers suggested that if there was a mini-bus or similar to transport survivors to organised social

events that they would be better able to attend regularly. They also said that this would mean that they felt confident knowing that the survivor was safe and in “good hands”. However, they were very aware of the cost aspect of this support.

### ***Waiting lists, referrals and funding***

Caregivers described waiting lists for support services as “*nightmarish!*” [C10]. One caregiver described how they had been trying to access mental health support for the survivor for quite some time, yet so far had not yet received it because of long waiting lists:

*“The waiting lists for everything are just immense...one thing we’ve been trying to get help with is to see a psychologist...we’ve still not seen a psychologist and this is 2 years on”* [C9, F, 50]

The same caregiver described that support had not always been available to her son because there was no agreement on who would pay for the service:

*“Again the funding and the health authorities, the arguing between who’s paying for a service – I find that so frustrating because when you’re here – I don’t care who pays for it as long as one of you does!”* [C9, F, 50]

On the other hand, two survivors who had recently received mental health support via brain tumour charities said they received this support in a timely manner.

Another issue that families face when trying to access support is needing to be referred for the support, and not being aware of where to get a referral from. One caregiver explained that she had been struggling in recent years with anxiety and things “*getting on top*” of her [C11]. She had been referred for psychological support at the hospital where her daughter had been treated and was currently in long-term care. However, the referral was refused because she did not live in the city where the support was being offered. Meaning she could not access the support.

### ***Decline in older adult services***

For three survivors, support they were receiving from community support groups ceased as they had got older. One survivor had attended a community support group for young people with long-term conditions for many years, but when he turned 18 he was no longer eligible to attend. Not only did he lose friends, he lost a place where he could regularly socialise:

*“Well like the short breaks and stuff that was...like I’d go to [City name] and have short breaks, like every last weekend in the month... get takeaways and have a laugh and that...but then I found that when I was 18 they just kick you out and don’t tell you anywhere to go.”* [S7, M, 24]

Another survivor said when he turned 25 several community support groups he attended were no longer available to him:

*College, youth clubs, socials...that all stopped... if you're older than that then I suppose you can't go to them!"* [S10, M, 30]

He said this 'gap' in support made him feel very low "*it was like what's the point of me getting up? I haven't got anything else going, I'll just stay in bed.*" The decline in support not only affects the survivor's happiness, it also has an impact on their caregiver as the support was also respite for them:

*"When [survivor] used to go on the residentials and things like that it were a break for [survivor] of course it were but it was a break for me as well. Cos otherwise it is all year [caring]"* [C9, F, 26]

Survivors as they grow older (age 18-25) may be at risk of needing additional support services or sign posting to new suitable support as other support services become unavailable.

### **Accessibility of the information/support**

Survivors identified that support or information about support is not always in an accessible format. Due to poor vision and cognitive issues many survivors find reading difficult:

*"I wasn't good at reading, if there was a leaflet on the door I wouldn't be able to read that leaflet and understand that leaflet that's actually staring me in the face because I can't read that leaflet I'm not going to know."* [S6, F, 27]

One survivor said that she struggled to interact or receive support over on the telephone due to her processing speeds:

*"I couldn't process what she was saying properly, when I got off the phone I had to get my notepad out and try and remember what she's said"* [S8, F, 25]

Some survivors said that online support/information was not preferable because they did not have a mobile phone or use it much. Another survivor admitted that she "*wasn't good on computers*" [S6]. On the other hand, for some survivor's digital/online information is better than physical information (i.e. book, leaflet) due to their sight difficulties as they can enlarge the texts or zoom in.

### 6.3.3.2 Emotional barriers to support: “Getting on with it”

One of the leading explanations provided by both survivors and caregivers for not having accessed support services was, that they were just “getting on” with life. Caregivers appeared to not prioritise their own needs, often saying they do not need it or choosing to cope on their own:

*“It’s hard but it’s called life and you have to get on with it!”* [C8, F, 54]

*“You try and kind of get on with it yourself and battle it yourself, I don’t know you just feel that you should be able to sort these things”* [C9, F, 50]

One caregiver felt support would remind them of the survivor’s disease and this is not what they wanted: *“we kind of want to move on and live normally really.”* [C1]

Survivors also expressed a positive outlook on life and if they could, they chose not to seek formal support and manage on their own:

*“Like with the emotional stuff I just get on with it. There’s nothing like...like my chemo does, did affect me but I’ve just got over it”* [S5, M, 18]

Both survivors and their caregivers expressed that ‘getting on with it’ is synonymous with ‘seeming to be in control of the situation’.

### 6.3.3.3 Reliance on family and friends

Most caregivers had never received any formal support in the long-term for themselves (e.g., counselling), instead they often discussed the importance of their family and friends for informal support. For example, in cases where the survivor needs daily care, family members occasionally take care of them so that the caregiver can have some time for their social life. Friends provide an outlet for caregivers to talk to and “sound off”:

*“I’ve not asked for any [support] and I’ve not been offered any... I just like to talk to people, certain people at work if you know what I mean, friends I’ve known from the beginning”* [C5, F, 50]

Sadly for some caregivers the support from family members has declined in recent years, as family members had aged or passed away:

*“My Mum has got a lot older now, she’s nearly 80...you know she’s not able to support me like she could. And you know my Dad’s not here anymore so.”* [C6, F, 53]

For many survivors, their family is their central social network:

*“He’s never left out or out. If we do anything, he goes with us. We don’t go out without him do we?” [C2, M, 58]*

Siblings often helped to support and take care of the survivor:

*“[Survivor’s brother name] does encourage him -he says come on, even though he’s feeling a bit tired some days he says “were going to go watch the football” ... I think he could have given up if it wasn’t for [brothers name] being here” [C4, F, 56]*

However, caregivers were aware that the survivor sometimes may benefit from more interaction from others who were like them:

*“She’s got lots of support from my friends and family – you know they all love her, they all know [survivors name] situation and they all treat her fantastically but her going out or doing things isn’t the same with them as it is with people on her level” [C8, F, 54]*

#### **6.3.3.4 No confidence in support**

##### ***Disappointed by support***

Unfortunately, some survivors and caregivers had negative experiences of support services, for some this had made them reluctant to look for other support. Some survivors and caregivers describe bad experiences which include being let down or forgotten about:

*“They [the charity] sent a lady who was going to take him out and meet up with some other young man quite near us but that never transpired.” [C6, F, 53]*

Losing confidence in the small amount of support available is a significant blow.

Two survivors were using a brain tumour charity’s support services until recently, when the charity sadly lost their funding and the support was no longer available. For one 30-year-old survivor this meant he was no longer mixing with similar others of his own age and no longer had any other social interactions outside his family [C3, F, 55]. Another family never fully accessed the support but had been interested in the support it said it could offer:

*“So we did start doing something with [charity name] but then the lady that was running it, she left and it was never taken up...we thought initially it was quite interesting cos it was talking about, you know helping with...they knew solicitors and people like that, that could help with issues like Personal Independence Payment, erm ESA, housing for people with learning disabilities, so it sounded really encouraging so that’s why we went along with it and they also had meet-ups with*



*other young adults that had had brain tumours or still have brain tumours that are in ongoing treatment and so...but it kind of fell through that.” [C8, F, 54]*

### **Not suitable for brain tumour survivors**

Another unmet need identified is specific brain tumour support. Some survivors received support from cancer charities but felt that this was not suitable for their experiences, in particular their late effects. One survivor was receiving counselling from a charity and decided not to carry on receiving the support because she felt they didn't understand her needs as a consequence of having a brain tumour:

*“I didn't feel it helped really. I actually don't think they got me as a person if you get what I mean. I don't think they understood again, the understanding bit of it...even though they were part of the hospital and cancer research sort of area...cos cancers you know a big umbrella really isn't it...so actually digging down into the brain tumour bit” [S8, F, 25]*

Other survivors tried to get help from local community support but said they were not suitable for them:

*I sought out help but I found they always put me in learning disability groups but not brain tumour type” [S7, M, 24]*

Caregivers often felt that these support services were not able to provide adequate support unless they had an understanding about childhood brain tumour (i.e. cognitive issues):

*“There is certain things but they just don't cater for [survivor] if you know what I mean... not to what he wants...see the biggest problem we've got with him is his concentration, it's nil, isn't it?” [C2, M, 58]*

### **Online support reputation**

Even though the survivors interviewed were part of the 'Google Generation' (a popular phrase that refers to a generation of young people growing up in a world dominated by the internet) many were reluctant to seek support or information online. Survivors described online resources as not necessarily relevant to them and in some instances scaremongering: *“I think sometimes I think they try and scare you.” [S5, M, 18]*

Similarly, caregivers also discussed negative experiences when accessing online forum information. Caregivers felt that the information was not relevant to their child's circumstances. For example, one caregiver said that she could only find American forums, but the survivorship care differed to the UKs:

*“They had all the insurance policies so they got daily physiotherapy, they got daily occupational therapy, you know – that we didn’t get so I got fed up of that!” [C6, F, 53]*

Another caregiver uses a Facebook forum for parents whose child has been diagnosed with a brain tumour but felt that the content on the forum is not always useful and sometimes upsetting:

*“I kind of switch off a bit cos it can bring you down a bit sometimes –people compare “oh my child can do this and my child can do that!” [C11, F, 40]*

Both survivors and caregivers were wary of online information as *“anyone can put anything on”* [S8, F, 25]. Many said in order to trust online information they would have to be signposted to it by the hospital late effects teams, to ensure it was an “official” resource they could trust:

*“I wouldn’t go on some random internet site...I wouldn’t do that I would go via the hospital and then from there.” [C5, F, 50]*

*“If there was like a website that had been set up by NHS or something like that and then I can trust it, instead of like actual...like google giving me the information.” [S5, M, 18]*

Survivors and caregivers indicated that a well-informed, reliable resource would be extremely useful. One survivor said it would stop her “stressing” so much about her late effects:

*“I think the fact that it’s been given to you [online resource] by consultants – there not just talking anything, they are talking cos they care, they are talking because it’s a website that has been crafted by themselves or by professors or people like that who are extremely intelligent but the guidance is there and its true, rather than going onto google – where anybody can put any old information in.” [S6, F, 26]*

Caregivers agreed that a brain tumour specific online resource would be useful for when they needed help as their child has got older:

*“[an online website] would be really useful, really useful because you don’t know until it happens do you, you don’t know what is going to crop up and like me if you’ve not been looking for that information but it would be nice to know that...yeah there is somewhere you can go to find out things” [C8, F, 54]*

### 6.3.4 Theme 4: The role of long-term follow-up care

All survivors attended a long-term follow-up clinic at their hospital. This theme looks at the role of this support for both survivors and caregivers. The theme starts with the transition from children's to adults services, which happens when survivors were around 18-22, depending on the circumstances. The theme looks at the challenging aspects of this transition but also how this transition can be made easier for both survivors and caregivers. The theme then highlights the importance of long-term follow-up care - what the main role of this service is for survivors and caregivers and finally how they feel this service could be improved.

#### 6.3.4.1 The transition from children's to adults services

##### *Uncertainty after moving from children's services*

Survivors and caregivers had mixed experiences from their transition from children's to adults long-term term follow-up services. Some said that they felt the care did not change much and the transition was fine. Yet it was apparent that some found the transition confusing, and lacked information about how the transition would work:

*"You're wondering who do I see? Do I see them at [adult hospital] or do I see them at [children's hospital]. It is a bit confusing" [C8, F, 54]*

Some survivors and caregivers said that more needs to be done to support survivors transitioning from children's to adult hospitals as they had found a lack of support at this time. It was apparent that more detailed information about the transition would be beneficial to put survivors and their caregivers at ease. Two families were new to the adult services, one caregiver expressed concern about how the adult service would compare in their support after the support they were used to:

*So I don't know how the [adult hospital] is going to be ...cos children's hospital was brilliant as in quick appointments, always see someone quickly, immediately phonecall back, very very very good" [C5, F, 50]*

In general, caregivers seemed to find the transition more distressing than survivors. One caregiver of a 13 year old survivor was already worrying about the transition from children's to adult services, even though it is several years in the future:

*"I'm dreading it I'm absolutely dreading it! I think that the Children's is very much a safety net and is really protected and I just hear all these constant stories of "No one spoke to me and we just went in and came out and no one acknowledged us!" and*

*obviously they tell the child not the parent anymore and its all those sorts of things...I don't know, I'm not looking forward to it."* [C11, F, 40]

### **Familiar faces**

The transition was easier where there was continuity in clinical staff, and/or hospital. Caregivers said that for the survivor seeing a familiar face made them feel more settled and comfortable when they moved to adult services. Some survivors had known members of the adult clinical team for many years:

*"They put [survivor name] with a consultant at the [adult hospital] who actually did the surgery on [survivor name] because he worked between the adult and children's hospital. So I don't know if that was coincidence but we've known him for a long long time and I think [survivor] feels quite comfortable with him, so that was nice, that was good."* [C5, F, 50]

Caregivers also found it helpful to see a familiar face when they transitioned to adult services, especially as they felt that the medical team knew the survivors background:

*"It was quite an easy transition, it was same people, same place! It is that familiarity, it's that person...I don't have to explain everything, I don't have to explain myself, I don't have to explain everything with [survivor], cos they know – they've been there all the way through and you're not having to go through the same stuff 3 million times cos they know! And that is such a weight off, that the first 10 minutes of any appointment you're not just explaining what happened"* [C9, F, 50]

### **Change in survivor and caregiver roles**

A change in services not only means that the treatment and clinical staff may change, it also means that the clinical staff may expect different things from both survivors and their caregivers. Survivors found that the change from children's to adult services meant they were expected to actively contribute more during appointments:

*"When we first went it were really difficult for him because he's just so used to being asked odd questions and the questions were aimed at us as parents"* [C5, F, 50]

However, one survivor felt they still were not actively involved enough and that even in adult services that clinical teams would still direct their conversation at her parents:

*"I think for me going from the child's side – where the consultant wasn't talking to me, they were talking to my parents...however going from 16 to 17 to 18 again still they're not engaging with you their engaging with your parents. So again the consultants could maybe look at that, to say well actually we talk to the person and the parents*

*are there as an escort...not to talk to them direct. Ask the person themselves cos we are here and we can talk” [S6, F, 27]*

All caregivers interviewed still attended the long-term clinic with the survivor. Many found that attending with the survivor meant they were reassured as the survivor got older:

*“[survivor name] is 24 now but I wouldn’t like to think he was going on his own and I wouldn’t like to think they would ask him to go on his own, because he doesn’t ask the right questions – he just shrugs his shoulders and things like that – so if he went on his own I wouldn’t find it as reassuring as I do because I go with him” [C6, F, 53]*

However, caregivers also found that their role changed. As survivors were being encouraged to contribute more in adult services appointments, parents were encouraged to take a “step back”:

*“It’s hard because at first when we very first went they were obviously looking directly at [survivor] not talking to us...it’s difficult because to me he’s still a child but he’s not is he – he’s an adult.” [C5, F, 50]*

One caregiver who had just attended the first long-term follow-up appointment with her daughter had found it upsetting that they were separated during the appointment. She had found this hard as she said they had been through everything together as a team and she now felt “pushed out”. She also felt that being separated had meant the communication was disjointed because her teenage daughter had not told her about the information that was given to her during that appointment:

*“Bearing mind everything else you deal with and we dealt with it as a team and we still do as a team, you don’t just sort of push the parent out and expect the child to be on their own if that’s not what they want. But we agreed to it eventually because it’s clear it meant so much but as a consequence of that it felt a bit disjointed with the information that we came away with” [C10, F, 49]*

### **Need for transition clinic**

Several survivors (4/11) suggested that there should be an intermediate teenage and young adult (TYA) service between children’s and adult’s services. Survivors said they often felt as a teenager that the children’s service felt too immature for their needs:

*“It’s all little children...like obviously there’s a lot of toys and stuff around so maybe like, so if you were looking for some improvement... maybe somewhere outside that clinic which is more teenagers.” [S1, F, 16]*

Caregivers also suggested that a teenage clinic for survivors would be advantageous, one caregiver [C1, F, 49] said that this may also allow for survivors to interact with others like them. Another caregiver said that they believed having a transition clinic would help the survivor adjust more easily from children's to adult services:

*“when they've gone from the cocoon of the children's hospital and having mum and dad there and all that to then expecting them to go to the next appointment on their own. Yes that's fine and some people might want to do that but I don't know I think there should be something a bit in between.”* [C5, F, 50]

#### **6.3.4.2 The importance of follow-up care**

##### ***Reassurance***

Survivors and caregivers said the main benefit from attending their long-term follow-up appointments was reassurance. Survivors described these appointments as a yearly “MOT” (annual test of vehicle safety in the UK) [S2, M, 30], and gained comfort from knowing that someone was regularly checking their progress as they got older:

*“it is nice to like feel as though people are looking after me knowing, you know checking me over to make sure that things haven't come back and that I'm healthy and stuff.”* [S1, F, 16]

*“it's kind of a good time to just reflect on how things are going and if there is anything that I could do with help with that they could help me with you know. And it's good to have a check up on those type of things.”* [S4, F, 28]

Equally, caregivers found these appointments an important place to ask questions and raise any concerns about any late-effect symptoms that may have occurred:

*“Once a year to just get some reassurance and maybe talk about symptoms that might have arisen during the year that have started up...no it's really good. Especially with her health if there is something that's bothering me that she's not noticed that I might have noticed...and I can talk to them about. Yeah it's really good for that, I do feel better for being able to go once a year to see them.”* [C8, F, 54]

*“I find it useful to touch base occasionally and we kind of, we had a little problem medically this time and it weren't an urgent thing it was just a “oh well when we see [consultant] we'll mention that” and so for us, for me it's that touching base and “oh by the way there's this happening is this normal, is this ok?” So yeah I find it particularly helpful”* [C9, F, 50]

### ***Around the clock support***

The majority of survivors and caregivers said they felt supported by the clinical teams in between appointments. Many interviewees recalled times where they had telephoned the clinical teams with a query or concern to seek help:

*“I’ve got a phone number if I need any help - he once got something wrong with his spine and his chest and I mean they did help us out, more or less straight away. So I know they’re there if I need them” [C6, F, 53]*

Most survivors and caregivers said that they have a direct telephone number or email address for a specialist nurse within the long-term follow-up teams. This direct support is highly valued by survivors and meant they felt comfortable to contact them whenever they needed, not just for medical concerns but also emotional support:

*“I think having somebody’s number and I’ve been given that number to me means that they care about me... if you’re feeling unhappy or unwell – give me a phone call. Yeah so it does make you feel good.” [S6, F, 53]*

For caregivers if something was wrong this available direct contact was often their first point of contact instead of the GP:

*“You know that you can pick the phone up and speak to them, cos they are more in tune with what’s happening....they have the background, they know have the knowledge of what [survivor name] has gone through.” [C8, F, 58]*

However, one survivor said she did not have this direct clinical support:

*“I wouldn’t go to a member in the hospital [for help], I wouldn’t know who to go to, I wouldn’t know who to ring, I don’t think I feel that I would be supported cos I wouldn’t know who they were if you know what I mean? Yeah I’ve only really got friends and family around me.” [S8, F, 25]*

### ***Providing information for a better life***

Many survivors and caregivers felt one of the key roles of the long-term effects clinics is to provide survivors with information on their life after their tumour:

*“I want it [LTFU] to be able to give [survivor] all the facts that if it will affect his life. And what he can do to improve his life...I didn’t even realise that radiotherapy would affect his bone density.” [C5, F, 50]*

Several survivors (4/11) said they were keen to learn more about their tumour history and the potential long-term effects in these appointments: *“I like to know stuff, I like to be aware of things, so everything they said was helpful.”* [S11, F, 17]

In addition, it was important for them that the clinical teams inform them about how they could live a better life:

*“For me to know that I’m living a healthy life and that I’m doing everything I can do and if I’m not doing everything I can do, for somebody to tell me – actually you can do more.”* [S6, F, 27]

One survivor, who was currently 18, wanted to be provided with the information not only about his late effects now but also in the future:

*“I just want them to tell me like, what’s going to happen further down the line. I think they said something about my bones, they might be effected and my growth – like I’m probably not going to grow anymore. They said that’s maybe why, like I’m shorter than all my friends, I’m the shortest one out of all of them. So I just really want them to tell me what’s going to happen further down the line.”* [S5, M, 18]

Caregivers felt that it was these clinics that should provide their children with survivorship information, information that they did not necessarily know about either:

*“It surprised me some of the things that they said, I never even gave them things a thought...it was like they said about insurance and things for everything...through his life. And I never even gave that a thought...So it was fascinating actually cos I didn’t even think about some of those things, so it was good that we saw them.”* [C5, F, 50]

### **Signposting and finding support**

Many caregivers said that when they had shared any concerns with the long-term clinical teams, they were then signposted to other services for support. Many of these concerns were not necessarily medical or physical but about the survivors emotional or psychological wellbeing. For example, one survivor admitted for the first time during an annual appointment that he had been self-harming. His caregiver was unaware at this point and the clinical team referred him to psychological support. By clinical teams signposting survivors to the support they need, they also help the caregivers who were primarily responsible for ensuring the survivor’s wellbeing:

*“We encountered a few problems that involved the police and I didn’t know where to go, I was at the point that I couldn’t protect [survivor] and I didn’t know how to. I couldn’t do it so I had to get some outside help from somewhere – it was no longer*



*something I could deal with. Erm so it was a desperate email to [consultant name] saying, right this has happened – help! You know where can I go from here? How can I get him some help?” [C9, F, 50]*

### **Continuity - feels like family**

Many survivors and caregivers were very complimentary about the long-term team who provided survivorship care for them. For several families, members of the clinical team have been a part of their lives for a long time:

*“It’s like they’re not just the doctors it feels like there a part of his family and friends if you know what I mean cos they are a part of your life in a way, the last 10 years plus” [C4, F, 56]*

Another caregiver said:

*“It’s just been part of like a community in a way...being able to go and they know what she’s been through...yeah them asking how you’re doing. I think [survivor] quite enjoys going to see the consultant and them talking to her about things and making her feel that they’re bothered about her. No I think it’s important really, it keeps that connection and you can fall back on them in a way” [C8, F, 54]*

### **6.3.4.3 How follow-up care could be improved**

#### **Engaging with the survivor**

A small number of survivors (2/11) indicated that clinicians did not engage with them enough, and felt that they could interact more with them instead of their parents at appointments. They felt it was important that they were spoken to directly, in order to understand aspects of their late effects and care, such as medication:

*“They need to engage more with the child than they do with the parents... I didn’t understand why I was on medication, apart from my Mum telling me...but a consultant physically did not tell me why I was on it, so when I would go pick my prescriptions up at the age of 18 and the consultants saying why are you on this...I would say “I don’t know” [S6, F, 27]*

Equally the way in which the clinical staff engage with survivors is important – providing information that is accessible to them. For example, survivors said it is important that the language used is understandable (i.e. not using medical jargon) and mindful of the survivor’s cognitive limitations (i.e. by speaking slower). A point that survivors made was that clinicians should check survivors’ understanding:

*“Sometimes some of the doctors say things...again it’s a bit difficult to understand them or process what their saying, I don’t quite understand the terminology.”*

[S8, F, 25]

### ***Rounded/holistic care***

Overall, both survivors and caregivers were complimentary of the care provided in the long-term follow-up clinics. However, when interviewees were asked how they thought support could be improved, nearly half of the caregivers (5/11) said they thought that the support and information provided could extend beyond medical care to more holistic, rounded care that also includes aspects of social well-being, such as applying for employment:

*“[The clinical team] are brilliant in their own right but as a person you want it all linked up don’t you– you want the social side of it linking– how do you apply for jobs and all of that.”* [C7, F, 61]

*“The medical side of things I haven’t got an issue with at all because they’ve been really really supportive in all the departments...but I think that [help with employment] is maybe something that could be looked at...maybe some links or some local numbers that could help with employment...that could be helpful people especially long-term, after you know your 10 years plus down the line after treatment”*

[C4, F, 56]

Other suggestions included more information about: available grants/benefits, social issues (i.e. accessing social support groups) and psychological support (i.e. counselling):

*“I mean I know [endocrinologist name] is really good and [LTFU consultant name] is really good but obviously there the clinical side of things, its maybe the psychological side of things that could be opened up a little bit more I suppose.”* [C4, F, 56]

## **6.4 Summary of findings**

This chapter provides insight into the experiences of TYA childhood brain tumour survivors and their caregivers. The interview data highlights survivors’ and caregivers’ unmet support needs, barriers to obtaining support, the role of long-term follow-up care and potential improvements that could be made.

Overall survivors and caregivers continue to have unmet needs in long-term survivorship. Both survivors and caregivers report a number of similar goals with subtle differences in their unmet support needs. The findings suggest that survivors were greatly concerned with their ability to live an independent life, find employment, and build and maintain social

connections. Previous survey-based TYA cancer survivor research (not specific to brain tumours) similarly highlighted the need for support with making and maintaining friendships but did not yet cover 'achieving life events'.<sup>137</sup>

Adolescence and young adulthood is a unique and complex developmental phase consisting of sensitive physical and emotional challenges,<sup>262</sup> and even further complicated by survivors' varying degrees of late effects. Which is probably one of the reasons most survivors wanted mental health and one-to-one support during this time. Both survivors and caregivers explained that they felt a drop in availability of services once the survivor completed treatment. This was of particular concern to survivors who, as they grew older, understood more about their health and the implications that late effects had caused. Yet, mental health support is typically not part of usual care, highlighting an area of significant unmet needs. This is in line with previous research that suggests that more multi-disciplinary, comprehensive, follow-up services for childhood survivors are required.<sup>99, 100, 263</sup>

The data highlighted that caregiver issues and needs were essentially focused on helping the survivor to succeed as they grow older, such as helping the survivor gain employment and socialise outside of the family. Caregivers were also anxious about the future and what will happen if they were no longer able to support the survivor both practically through financial and physical support, as well as emotional support. These findings are in line with previous TYA brain tumour research, caregivers reported concerns over inadequate financial support and a decline in support available as they moved further away from treatment.<sup>91, 136</sup> Yet it is worth noting these studies were based in the US.

Participants reported a number of barriers to accessing formal support. With caregivers relying heavily on informal support sources (family and friends), the continuity and sustainability of this support is of great concern. For instance, when family support is no longer available (e.g. when parents/grandparents pass away). The interviews uncovered some of the reasons for dependence on informal support, including: families being unaware of the current formal support available, and issues accessing this support due to location and funding. Survivors stressed the importance of providing lay-friendly information, in a format that is accessible to them, and providing it to them directly instead of via caregivers. This is supported by other studies in childhood cancer survivors.<sup>118</sup> Yet, this may not always be possible/appropriate in cases where the survivor has severe learning difficulties.

Important to both survivors and caregivers was the quality of support and information. Families discussed being let down by support in the past, which has negatively impacted their motivation to look for more support. Therefore, it is integral that support services are clear and transparent about the care they can provide, and dependable to ensure survivors

and caregivers expectations are met. In regards to information resources, there was concern around the reliability of online resources. Families were sceptical about seeking support online unless it had been recommended by someone they trusted (i.e. clinical team). Nevertheless, both survivors and caregivers saw the benefit and saw the appeal in online resources if they were reputable.

Long-term follow-up care was important and valued. Families explained that long-term follow-up was 'easier' when clinical staff remained the same, especially when transitioning from child to adult services. The transition of care from the paediatric to adult health care setting can be complicated for young adult survivors of childhood cancer.<sup>264</sup> A TYA specific transition clinic could help. Allowing families to adjust to a different type of care (e.g. the parent/caregiver becoming less involved in the appointment). In addition, resources or training for clinicians to facilitate the transition from addressing parents to addressing survivors may be beneficial to follow-up care in general. TYA clinics could also be an important place to provide much needed, age-specific information for families (e.g. around employment, finances). As highlighted the timing of information for families is important. A TYA clinic could also be somewhere that survivors could meet others like them, facilitating the potential for survivors and caregivers to socialise. Helping to address a key need highlighted in this chapter.

#### **6.4.1 Strengths and limitations**

The use of in-depth semi-structured interviews allowed a rich understanding of the unmet supportive care needs of survivors and their caregivers. This data could not have been gained through the survey alone. I felt the interviews provided great depth to the findings of the survey, providing explanations to why certain unmet needs were a priority to participants. The qualitative data also filled gaps that were not addressed in the survey, such as the role of long-term follow-up care. These results may help to develop or improve long-term follow-up care.

A strength of semi-structured interviews is the ability to obtain rich descriptions of participants' experiences and attitudes. As highlighted by Barbour (1999), semi-structured interviewing "*allows for the ordering of questions to be employed flexibly to take account of the priority accorded each topic by the interviewee.*" (p.18)<sup>265</sup> A benefit of using this approach is that participants are able to indicate which areas were of the most importance to them, meaning that issues and needs raised by interviewees were those that were at the forefront of their minds and, therefore, of priority. By encouraging participants to 'tell their story' as they had experienced it, allowed participants to initiate the discussion of topics of

importance and express their views in their own way. Interview prompts were generally only used to re-direct the interviews back on track. This being said, some survivors seemed to find the interview process challenging, mainly because of slow processing speeds and their difficulties with their speech. This may have meant that some of survivors were unable to fully articulate their supportive care needs. However, this was anticipated and during the interviews I ensured that survivors knew they could take their time, have a break at any point and ask for further explanation about the questions. Equally, the interview guide was designed (with input from the PPI group) to be concise, simple and clear as to not overwhelm survivors.

This data has contributed much to our knowledge, yet has some limitations. Firstly, sample bias. The qualitative interviews were carried out with a relatively small group of purposefully sampled survivors and caregivers to represent a broad range of sociodemographic and clinical characteristics. Purposeful sampling is commonly used within the discipline of qualitative research. Survivors were varied in relation to their current age, age at diagnosis and tumour: diagnosis, grade and treatment. Therefore, the diversity of the final sample of participants is a strength of this study. Still, there is potential for bias to have been introduced through non-participation. As described at the beginning of the chapter, 10 survivors and 7 caregivers who were approached to take part did not partake in the interviews. Therefore, the research sample could have been more motivated and 'well' than those who did not partake. If the sample was biased in this way, the findings may have underestimated supportive care needs.

As highlighted in Chapter 4 (4.4.4) there were many ways that qualitative methods can be used to investigate narratives.<sup>235</sup> A strength of the analytical method used (thematic analysis) is that it allows themes to be ordered under pre-existing headings. This allowed the data to be analysed from the perspective of the pre-defined interview aims, while remaining grounded in the data and also exploring unanticipated themes emerging within each aim. Other approaches to qualitative research could have been employed, such as a greater focus on the form and style of the stories, or a deductive analysis process using a predetermined coding frame. However, an inductive process was applied to allow participants' stories to be interpreted separately from the theory that was driving the studies.

Finally, the potential influence of the researcher in shaping the analysis is a common criticism of qualitative research. The internal validity of the analysis is subject to rigorous identification of coding data and developing themes, and the reduction of researcher bias or error. Researcher bias/error may lead to the incorrect definition of emerging themes or missing relevant data for coding altogether. The potential for these biases/errors were

reduced using two methods. First, a random selection of the interviews (four) were independently coded by one of my PhD supervisors (Florien Boele) to compare coding decisions and the evolving themes. Secondly, it is hoped that the reflective sections throughout the thesis have made the potential researcher bias/influence as transparent as possible. Ultimately, it is hoped that this influence has been a strength of this piece of research rather than a limitation.

#### **6.4.2 Reflective thinking**

As previously discussed in Chapter 4, a key aspect to ensuring quality and rigour in qualitative research is the practice of self-reflection.<sup>266</sup> This practice can be seen even more important in oncology research:

*“Data collection can be an intense experience, especially if the topic that one has chosen has to do with the illness experience or other stressful human experiences. The stories that the qualitative researcher obtains in interviews will be stories of intense suffering, social in justice, or other things that will shock the researcher.”* (p.78)<sup>267</sup>

It is widely recognised that the researcher is an active agent in the research process. Interviewers are responsible for setting the tone of the interview, following up on participant comments, and asking the questions. It would be naïve to imagine that one can remain completely objective throughout this process, that my own experiences and beliefs have no impact on the interview.

Being able to honestly and openly discuss my role within the research process is a core attribute in ensuring good quality research. In order to do this, I provide an overview of my research background, strengths, weaknesses and standpoints.

First, I discuss my research background. Following the completion of my BA Honours Sociology degree and Social Research MA, I have worked in a number of different research roles, including mental health research and research investigating the quality and safety of hospital care. Over the years developing a keen interest in health and quality of life research. However, oncology research and specifically brain tumours were a new area of interest when I began this PhD. I have found it fascinating learning about this patient group, and I have worked hard to gain as much knowledge as possible around their patient and survivorship journey. However, I am aware of my limitations, I do not have a clinical background. To overcome this shortfall when necessary I have sought advice from knowledgeable professionals. For example, I have built good relationships with the clinical teams where I have been recruiting and I have discussed several aspects of the research and patient group with them. This has been incredibly helpful throughout the PhD. Equally, I

have been fortunate to have knowledgeable PhD supervisors and be placed within a very experienced research team. Both of which have meant that I have been incredibly supported throughout this research.

Prior to completing this PhD I would have described myself as a qualitative researcher, with the majority of my previous roles being qualitative focused. I have had extensive training and experience in conducting interviews with different populations, which I believe helped to conduct these interviews, which could be very emotive at times. I believe my experience meant that I was able to develop rapport with participants to put them at ease, which meant that they felt comfortable discussing sensitive topics. I believe this can be seen in the results of these interviews, as the quotes were insightful and honest.

Finally, I provide my personal standpoint in relation to this research. I, like many others have unfortunately lost family members to cancer. Yet learning more about the devastation a childhood brain tumour can have on a whole family was shocking. Equally, many family and friends have said to me during the PhD that they don't know much about brain tumours, and I think on reflection that is true, it is one of the lesser known cancers. Many people were surprised to learn of the long-term effects that these survivors and their families endure.

I recognise that my feelings and thoughts may have impacted on the content of the interviews. I am aware that I may have been sub-consciously more interested in certain aspects of participant's stories. For example, my brother has learning difficulties and I saw some parallels in the participant stories (especially caregiver stories) that my brother and our family have experienced. One of the similarities being social isolation and the need for social support. I felt I understood some of the pain survivors had experienced and how difficult it could be for parents with a child with little social interaction with peers. This being said I was aware of my feelings and do not think my feelings changed the focus of the interviews for a few reasons. First, the interview guide was there to focus the interviews (collect data to address the research questions), while also allowing plenty of scope for participants to discuss areas of importance to them. And second, I made sure I wrote detailed field notes after each interview, each of which reflected upon my feelings and thoughts about the interview. By doing this it not only made me recognise my thoughts but also encouraged transparency.

Additionally, during the third year of my PhD I gave birth to my first child. Although the majority of my data analysis was complete at this point, this experience has without doubt affected my thoughts and feelings during the write up. Reflections on what participants have said, especially parents experiences were given another dimension. I felt closer to

understanding the pain and disruption they have experienced due to their child being diagnosed with a life changing cancer.



## Chapter 7: Integration

Chapters 5 and 6 presented the quantitative and qualitative analysis separately using appropriate methods. The following Chapter contains the complementary integration of both data sets to define key points for life after a childhood brain tumour for TYA survivors and their caregivers. The Chapter begins by detailing the approach to the integration (7.1), followed by a Joint Display Table and narrative to highlight the integrated findings in relation to the four research objectives (7.2), to finish a summary is provided, including the strengths and limitations of this integration (7.3). This Chapter enables the identification of key components of participants' experiences and clarify what possible interventions could be of benefit to survivors and caregivers.

### 7.1 Approach to mixed methods integration

Integration is a central and challenging aspect of mixed methods research.<sup>148</sup> The integration of quantitative and qualitative data can dramatically enhance the value of mixed methods research.<sup>162, 167</sup> Integration means bringing the quantitative and qualitative elements of a mixed methods study together for analysis and comparison. It has been found that often studies fail to integrate quantitative and qualitative data within the research,<sup>167, 268-270</sup> which can limit the knowledge that these types of studies generate. Often without integration, the knowledge yield is equivalent to that from a qualitative study and a quantitative study completed individually, instead of achieving a "*whole greater than the sum of the parts.*"<sup>265</sup>

There are specific approaches in which to analyse and integrate data, the most appropriate is often dictated by the mixed methods design. The combination of quantitative and qualitative elements must be justified by the overall aim of the study and the research objectives. The three main approaches to integrate qualitative and quantitative data during the reporting stage include: (1) integrating through narrative; (2) integrating through data transformation; and (3) integrating through joint displays. One or all three of these approaches may be used in one mixed methods study.<sup>271</sup>

In this thesis, the results from the quantitative and qualitative data were integrated using a joint display table. A joint display is defined as a way to:

*"Integrate the data by bringing the data together through a visual means to draw out new insights beyond the information gained from the separate quantitative and qualitative results."* (p.2143)<sup>271</sup>

A joint display table was created to make a matrix that set the findings from the results against the research questions and identified key themes.<sup>242, 272</sup> There are multiple steps in developing a joint display table. First, raw data (e.g. percentages and selected quotes) and coded or grouped data (codes/themes, and statistics turned into text) considered important for inclusion in the integration were listed in the joint display, in appropriate columns. The literature suggests that this can either be a comprehensive process (including all codes and data identified in a prior quantitative or qualitative analysis) or selective (including only particular data or emerging themes from an earlier analysis), this decision depends on the focus and purpose of the integration.<sup>273</sup> In this analysis, the relationships between data were explored in terms of the research objectives, to ensure that the integration outcomes were focused on addressing the overall research aim. Once the relevant data was listed in columns, a matching process proceeded - aligning similar data, and refining and organising themes that had been generated by the two sets of data. Second, the most suitable data to display in the table was chosen, this data was selected to illustrate the themes most appropriately. Data was chosen that best matched the other type of data- trying to reflect the information, context and any other content. It is worth noting that not all columns contain data/content, this is where this data has not been collected in relation to a theme (also referred to as silence). Third, the findings under each theme were compared and contrasted. At this stage it is important to identify the “fit” of data integration – the coherence of the quantitative and qualitative findings.<sup>271</sup> The evaluation of fit of integration leads to four possible outcomes: complementary, convergence, divergence and silent relationships. Complementary findings indicate a strong agreement across both data sets; convergent findings indicate partial agreement; divergent findings indicate that the data contradict or are not similar; and a silent relationship indicates no relationship between the datasets. It is worth noting that having a range of findings shows the value of an integrated mixed methods approach.<sup>147</sup> Therefore, convergence, divergence or silence in this analysis does not necessarily represent disagreement across outcomes, rather the potential for one method to produce findings that the other method could not. Hence, by integrating outcomes the full benefit of the mixed methods process could be explored.

The three steps undertaken are displayed in Figure 15. It is important to highlight that this is an emergent process, going backwards and forwards between steps. This iterative process continues until the final joint display tables illustrates the key findings from both sets of data, in a concise clear way.

**Figure 15 - Figure to show the steps taken to build a mixed methods joint display table**



## **7.2 Mixed methods integration: Joint Display Table**

In Table 41, themes, quotes, findings, and statistics from both datasets are presented in a framework. The final column of the Joint Display Table identifies where findings complemented, converged, diverged, or produced silence.<sup>274</sup> Colour matching of the data is used to match visually the quantitative and qualitative responses from the different participant groups, survivors in blue text and caregivers in green text.

**Table 41 - Mixed methods Joint Display Table**

Integrated findings	Quantitative data (Survey)	Qualitative data (Interviews)		Relationship
		Codes	Quotes	
1) To describe unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers.				
<b>Psychological support</b>	<p>Psychological needs had the highest standardised mean score (30.2) of all the SCNS-SF34 domains. 60.3% wanted support with 'anxiety'. 35.8% wanted access to counselling.</p> <p>Half of the top 10 ranked unmet needs belonged to the psychological domain (5/10). Psychological and emotional needs had the highest standardised mean score (30.2) of all the SCNS-P&amp;C domains. 35.4% wanted access to counselling</p>	<p>Mental health support; reliance on family and friends; Getting on with it; decline in older adult services; Waiting lists, referrals and funding.</p>	<p><i>"I had counselling when I was 7 but I still want help now I'm 25, I don't want that help to have just stopped things are still happening and changing the side effects never leave you"</i>[S8]</p>	Complementary
	<p>42.9% wanted 'Information about financial support and governmental benefits' 42.9% wanted support with the 'Influence of caring on your working life or usual activities'</p>		<p><i>"There has never been any kind of support for how to deal with the aftermath and how to deal with what might be to come...which is really what you need because at the time you're firefighting and you get on with it but then obviously there is a sort of PTSD element."</i> [C10]</p>	Complementary
<b>Financial and employment support</b>	<p>33.3% survivors were currently unemployed or unable to work. 42.0% wanted information about their finances. 44.1% wanted information about employment.</p>	<p>Employed work; Financial support; Decline in support after education; Social worker; How the survivor will manage in the future.</p>	<p><i>I properly need that money[benefits]...if I don't get that money it's going to be hard for me, because I can't work...which is really hard for me and I find it upsetting."</i> [S9]</p>	Complementary
	<p>42.9% wanted 'Information about financial support and governmental benefits' 42.9% wanted support with the 'Influence of caring on your working life or usual activities'</p>		<p><i>"The main challenges, well is finance, financial...Its stressful cos I want [survivor] to have some income...cos I can't support her...and that's what worries me."</i> [C8]</p>	Complementary
<b>Socialising with similar others</b>	<p>55.2% wanted to attend weekend retreats with other brain tumour survivors. 52.1% wanted to attend monthly social activities with other survivors. 47.5% wanted to attend weekend retreats with other brain tumour survivors and their caregivers</p>	<p>Declining friendships; Speaking to similar others; Organised support groups; Decline in older adult services; Not knowing where to go or what is available; Location.</p>	<p><i>"One of my main issues is probably friendships and relationships more than anything...I kind of don't know...I don't know much about them to be honest."</i> [S10]</p>	Complementary
			<p><i>"I've not met another medulloblastoma patient yet...I would like to meet more medulloblastoma parents...yeah I'd like to do that."</i> [C11]</p>	Complementary

<b>Supported independent living</b>	Not measured  Not measured	Independent living; Dealing with the ever-changing landscape; How the survivor will manage in the future; Social worker.	<i>"I don't even know how to cook now and I'm 27 and that's because it's very slow...I'm still picking up the pieces now!"</i> <i>"I know he could never live independently, he's on the waiting list for sheltered housing round here but...he's not a priority" [C6]</i>	Silence  Silence
<b>Fertility information</b>	43.1% would like information about fertility. 42.9% would like information about survivor 'fertility problems'.	Fertility was not a prominent theme in survivor or caregiver interviews		Silence  Silence
<b>Romantic relationships</b>	Sexuality needs had the lowest mean score (13.4) of all the SCNS-SF34 domains. Two of the items with lowest level of unmet needs were: "Changes in sexual relationships" (10.4%) and "Changes in sexual feelings" (13.4%). One of the areas in which caregivers had the least amount of needs was in reference to 'addressing problems in their sex life' (7.5%).	Speaking to similar others Organised support groups; Diminishing support getting further away from treatment	<i>"I think one of my main issues is probably friendships and relationships more than anything, it's like – I kind of don't know...I don't know much about them to be honest."</i> [S10]  Caregiver romantic relationships/sexuality needs was not a prominent theme in caregiver stories.	Divergence  Silence
2) To explore if survivor sociodemographic and clinical data are related to unmet needs.				
Unemployed survivors experience more unmet needs	Unemployed survivors were associated with more: overall unmet needs (B=5.704, P<.005); physical and daily living needs (B=19.696; P<.05); patient care needs (B=-13.442, P<.05); health system and information needs (B=-14.879, P<.05).	Employed work; Mental health support.	<i>"I do get depressed a bit and I did do when I was very much looking for work...so I have stopped looking for jobs"</i>	Complementary
Survivors and their caregivers further from diagnosis experience more needs	Survivors further from diagnosis were associated with more prevalent overall unmet needs (B=.476, P<.05).  Caregivers caring for survivors further from diagnosis were associated with more psychological and emotional unmet needs (B=-1.704, P<.05)	Diminishing support getting further away from treatment; Decline in support after education; Decline in older adults services; Dealing with the ever-changing landscape	<i>"Well like the short breaks and stuff...when I was 18 they just kick you out and don't tell you anywhere to go."</i> [S7]  <i>"I always said once he turned 18 he still needed looking after, he still needed care but a lot of what we had prior to him being 18 was taken away from us and not replaced"</i> [C6]	Complementary  Complementary

Single caregivers experience more needs	Single caregivers were associated with more: overall unmet needs (B=-15.556, p<.001); psychological and emotional needs (B=-18.798, P<.05); informational needs (B =-33.368, P<.001); health care service needs (B=-36.266, P<.001); and work and social needs (B=-26.724, P<.001).	Caregiver relationship status was not a prominent theme in caregiver stories.		Silence
3) To determine whether unmet needs are associated with Quality of Life (QoL) outcomes.				
Unmet needs influence quality of life	Survivors who had more unmet needs reported a lower QoL (r= -.621, p<.001). Caregivers who had more unmet needs reported a lower QoL (r = .616, p < .001).	Although many aspects of QoL was discussed in the interviews, the purpose of the interviews was not to make direct associations between needs and QoL.		Silence  Silence
4) To explore the role and perceived use of support services in TYA survivors and their caregivers.				
Co-ordinated/holistic clinical care	34.3% 'One member of hospital staff with whom you can talk to about all aspects of your condition, treatment and follow-up' 38.7% wanted a case manager who coordinated services	Rounded/holistic care; Signposting and finding support; Providing information for a better life	"Actually having some sort of a person who discusses the big picture of every aspect of you that's what's needed"[S7] "[The clinical team] are brilliant in their own right but as a person you want it all linked up don't you- you want the social side of it linking- how do you apply for jobs and all of that." [C7]	Complementary  Complementary
Ensuring families know where/how to access support services	Not measured  39.0% wanted information about support services	Not knowing where to go or what is available; Needing a referral; Decline in older adult services; Accessibility of the information/support	"I've got friends and family around me. But apart from them, I wouldn't know who to go to, and I wouldn't know how to access who to go to." [S8] "We didn't really know what other support groups were in place...so we didn't really know anyway to where you would go and look." [C1]	Silence  Complementary
Online information and supportive services	Survivors who have used online support: online information (31.9%); online support	Online support reputation; Accessibility	"Anyone can put anything on[line]" [S8]. "If there was like a website that had been	Convergent

	<p>group (36.2%); 24/7 online chat support (7.4%).</p> <p>Caregivers who have used support: online information (58.5%) online support group (30.0%); 24/7 online chat support (2.5%).</p>	<p>of the information/support Signposting and finding support; Preparing for long-term treatment effects; Dealing with the ever-changing landscape</p>	<p><i>set up by NHS or something like that and then I can trust it, instead of like google giving me the information.” [S5]</i> <i>“[website] would be really useful, really useful because you don't know until it happens do you, you don't know what is going to crop up and like me if you've not been looking for that information but it would be nice to know that...yeah there is somewhere you can go to find out things.” [C8]</i></p>	<p>Convergent</p>
--	---	--	--	-------------------

### ***Objective 1: Unmet supportive care needs***

In regards to the first research question- to describe unmet supportive care needs of TYA survivors of childhood brain tumours and their caregivers, three themes had complementary findings. Complementary findings suggest a strong alignment across both data sets. Firstly, findings were complementary around the concept of psychological support. Psychological support was identified as a key unmet need in both the surveys and the interviews. The descriptive survey data highlighted that survivors identified '*anxiety*' as the problem they most wanted support with. The interviews further expanded our knowledge by highlighting that survivor anxiety is aggravated by the ever-changing landscape of early adulthood. For example, survivors seemed especially anxious when they left education or when trying to find employment. Equally, psychological support was one of the few areas that caregivers identified as an unmet need for their *own* well-being, unlike the majority of caregiver needs which were in relation to the survivor's well-being.

Secondly, both quantitative and qualitative data emphasised the requirement for support with finance and employment. The descriptive survey data highlighted that nearly half of survivors desired information regarding finances and employment. The interview data complemented and expanded on the survey data, with many survivors discussing problems with getting a paid job due to their brain tumour history. For many, problems around finance and employment had been ongoing since their education ended. Equally caregivers shared their need for support with helping survivors get meaningful employment. The top ranked unmet caregiver needs were wanting '*Information about financial support and governmental benefits*' and support with the '*Influence of caring on your working life or usual activities*'. The interviews expanded that the financial support caregivers needed the most was help with benefit forms, many caregivers discussed the difficulties they encountered completing these governmental forms, even in long-term survivorship. These complementary findings highlight the necessity for supportive services and information in this area. Specifically, brain tumour specific information and support regarding financial forms and finding paid employment.

Thirdly, findings around socialising with similar others were complementary. The survey descriptive data highlighted that over half of survivors and just under half of caregivers wanted to attend weekend retreats with other brain tumour families. The interview data expanded on this data by highlighting the issues that survivors have had making and maintaining friends in young adulthood. Survivors said they want to meet others with similar experiences, so they have the understanding that other peers their age do not have. Equally, parents shared the importance of their child having social relationships with others outside their immediate family. It is fairly unsurprising that participants had unmet social needs as



this was an issue particularly prominent in the systematic review (Chapter 2). However, there were current barriers to accessing this support including location of services, and families not knowing what support is available.

Silence in one study does not mean that there is dissonance in the findings, rather it is an example of how different approaches can reflect different aspects of a phenomenon.<sup>275</sup> It is a strength of mixed methods research that through studying a problem using different methods a more complete understanding is gained. Silence was evident when comparing findings in relation to survivor fertility. Survivor fertility and potential fertility issues were a central area of interest in the quantitative data, nearly half of survivors and caregivers indicated that they wanted more information about fertility. In fact, caregivers identified information about the survivor's fertility as one of the most pressing unmet needs (ranked joint first). Yet the need for information about the survivor's fertility/fertility issues were not discussed in the qualitative interviews. One reason for this could be that the interview topic guide did not include any specific prompts about fertility. Another reason could be because survivors felt uncomfortable/unable to discuss such a sensitive and private matter. Moreover, for many younger survivors fertility may not be currently at the forefront of their priorities. Instead survivors were concerned with trying to establish friendships. However, young adulthood is a forever changing landscape, with new milestones and challenges arising. Therefore, fertility issues may not currently be a main concern for younger survivors, but it may be something they would like information about for the future as they get older (as the quantitative results suggest). Similarly, fertility may not have been discussed by caregivers in the interviews because their focus was on different issues and needs they were currently experiencing and trying to navigate (such as survivors' social life/finance issues). This finding highlights the importance of timely information for TYA survivors and their caregivers, but also highlights the difficulty in getting the timing right for all families.

Findings around independent living were also silent. Questions around independence and potential support around achieving independence for survivors were not included in the survey. None of the validated questionnaires incorporated questions with this focus. For instance, no questionnaire asked about living independently, assistance with personal care, or obtaining a driver's licence. One reason for this might be that some of the questionnaires were not specific for teenagers and young adults, therefore this milestone may have been overlooked. Another reason that independence may not have been measured in the questionnaires could have been because some of the validated questionnaires used were not brain tumour specific but considered suitable for all cancer diagnoses. As highlighted in the systematic review (Chapter 2), childhood brain tumour survivors often suffer worse late effects than other childhood cancer survivors', hence their independence is potentially more

at risk than other groups. Questions around independence were included in the interview guide. Survivors spoke about several areas relating to their independence including their worry about never being financially independent or being able to look after themselves (i.e. not being able to cook). Even more so, caregivers were specifically distressed about their child's ability to live independently especially after they were no longer able to care for them. Independence is evidently a key aspect of growing older and becoming young adults. Long-term supportive care and information should therefore include helping survivors to live independently (where possible).

Interestingly, divergent findings emerge when exploring support with survivor romantic relationships. That is, the findings are not similar in the quantitative and qualitative work streams. The survey data recorded that survivors had low need for support with sexual relationships and feelings. However, the interview data highlighted that many survivors desired romantic relationships. One possible explanation for divergent findings in this area may be due to how the topic was approached differently in the data collection. In the survey, romantic relationships were posed as 'sexual relationships' whilst in the interviews the topic was often discussed from a different angle. For example, survivors would often discuss romantic relationships when discussing friendships/networks or when thinking about the future and marriage/children; not necessarily using the words 'sexual relationships'. As this group were teenagers and young adults, some under 16 years of age, the phrasing 'sexual relationships' may be more suitable for adult populations. Another potential reason for divergent findings, is that many of the survivors were helped to answer their survey (by a parent or researcher) and they perhaps did not feel comfortable/able to indicate their need for support in this personal topic. Similarly, caregiver findings around romantic relationships were silent. The survey data indicated that caregivers had little need for support with their sexual/romantic relationships. Whilst this area was not discussed in the interviews. One reason for this may be due to the interview guide/prompts not specifically including caregiver romantic relationships. Another reason may be that the silence in the interviews actually supports the low need for help in this area as caregivers did not articulate this as a problem.

### ***Objective 2: Unmet supportive care needs and sociodemographic/clinical data***

The second research question looked to explore if survivor sociodemographic and clinical data were associated to unmet needs. Firstly, the regression analysis conducted on the survey data highlighted that unemployed survivors were more likely to report unmet needs overall. This finding complemented the qualitative findings. Whilst it was not a specific aim of the qualitative study to identify sociodemographic/clinical differences in survivor accounts, it was evident that survivors who were unable to work or currently unemployed were

experiencing more issues and therefore had more needs than those in employment. It is likely that survivors who were unable to work/unemployed also have worse late effects of treatment, and were therefore more likely to report more needs.

Quantitative and qualitative results both highlighted that survivors and their caregivers further away from diagnosis were more likely to experience unmet needs, meaning the results were complementary. Regression analysis showed that time since diagnosis was significantly associated with the overall number of unmet needs survivors recorded. The qualitative results expanded on this finding, survivors and caregivers both discussed that supportive services diminished as they got further away from treatment, which meant in some cases they had more unmet needs now than when they were in/closer to treatment.

The survey analysis highlighted that single caregivers were significantly more likely to report needs in comparison to caregivers in a relationship. Yet when telling their stories, relationship status, was not an element that was explicitly highlighted by caregivers. The silence found may have been because questions around relationships were not included in the interview guide. This being said, in one of the interviews field notes I reflected how I believed single parents had extra pressures that other parents did not. I noted that a single mother had little support from anyone else as she lived alone with her daughter, without a wider support network. This meant that all the responsibility for her child's well-being was upon herself. Therefore, it would make sense that parents in this position would report higher needs, than parents with partners and further support networks. It is imperative that supportive services are mindful of the increased needs single parents may experience, especially those who are also without a wider social network.

### ***Objective 3: Unmet supportive care needs and QoL***

The third objective aimed to determine whether unmet needs were associated with Quality of Life (QoL) outcomes. The quantitative results identified that both survivor and caregiver unmet supportive care needs were strongly associated with poorer QoL. The findings highlighted the complexity of being a long-term survivor/caregiver, and the potential impact on QoL when support needs were not adequately met. The concept of QoL was not specifically explored in the qualitative interviews as it was not one of the qualitative aims. Equally it would be difficult to measure if QoL was associated with unmet needs qualitatively, hence why this research objective was designed to be addressed by quantitative data only. However, it could be argued that qualities associated with quality of life were ingrained in survivor and caregiver stories. As described in Chapter 2 - QoL is a subjective, multidimensional construct that encompasses social, physical, psychological, spiritual and in this instance cognitive well-being factors that all relate to the health of an individual. In the

interviews, aspects of QoL especially social well-being and psychological well-being, were central to survivors' stories. For example, survivors described poor social life's, which made them feel socially isolated, impacting their social well-and QoL.

#### ***Objective 4: The role and use of supportive services***

Objective 4 aimed to explore the role and perceived use of support services in TYA survivors and their caregivers. Findings were complementary around the needs for clinical supportive services to offer holistic and coordinated care. Both survivors and caregivers discussed that they wanted long-term follow-up care to offer more rounded care that covered aspects of social well-being, such as information on support services (e.g. charities) that helped survivors to meet other survivors; or the opportunity to access psychological support (being able to speak to a psychologist/counsellor). Quantitative results confirmed that holistic care was important. In the top ten unmet needs, over a third of survivors wanted someone (e.g a key worker) that they could discuss all aspects of themselves with. While nearly 40% caregivers wanted one person who coordinated all aspects of care. This theme indicates the value and complexity of care coordination highlighted by the multifaceted needs of survivors. Advocacy for appropriate and timely educational, vocational, and social support especially is critical as part of comprehensive survivorship care.

Many caregivers described (in the interviews) not having accessed support they/the survivor needed because they were unsure of what was available, or where to go for this help. Equally, the survey identified that nearly 40% of parents also wanted more information about support services. Meaning data around this theme was complementary. Survivors also discussed not being aware of available support, but this data was not measured in the survivor survey, therefore it cannot be classed as complementary but silent instead. Yet as survivors identified unmet needs it can be reasonable to expect that in part, this can be explained by survivors not knowing where or how to access support.

Survivor findings converged around the concept of online support. Convergent findings suggest partial agreement. The qualitative findings highlighted that many survivors had not used online supportive services as they were wary of the reliability of online content and support. The survey results highlighted that around a third of all survivors had accessed online brain tumour information or online support groups. The reason for the converging data may be because interview survivors were only recruited from the long-term follow-up clinics and not online, whereas some of the survey respondents were recruited online. Therefore, a subgroup of survey respondents may have been more open to using online services. Yet, the results partially agree as survivors also discussed in the interviews how they would like to use online resources but only if they deemed it reliable (i.e. sanctioned by the NHS).

Caregiver findings complemented each other and supported that parents had used online services. Over half of all survey respondents said they had used online support services. Like survivors, caregivers said they would be open to accessing support online but only if the content has been provided and signposted by reliable, appropriate professionals. Highlighting that confidence in online support is low but could be valued by families if created by knowledgeable researchers and championed by trusted clinical staff.

### **7.3 Summary of integration**

The process of integration aimed to consolidate the qualitative and quantitative results presented in previous chapters, and to address all four research objectives. This chapter describes the mixed methods analysis integration approach utilised and discusses the insights from the integration. An array of outcomes from across the data were explored and integrated through the creation of a matrix to investigate when findings complemented each other, converged, diverged or produced silence – the Joint Display Table. The production of the matrix highlighted what was learnt from the mixed methods approach that would not have been learnt through a single study, or separate qualitative and quantitative studies. The implications of the integration findings will be discussed in more detail in the following, and final, chapter.

#### **7.3.1 Strengths and limitations**

The development of Joint Displays has emerged as a highly valued approach for integrating qualitative and quantitative findings in mixed methods research. Joint Display Tables enable analysis, interpretation and provide a visual representation of mixed method results to generate new inferences.<sup>271</sup> One of the benefits of utilising a Joint Display Table approach was that a large amount of data was condensed into a relatively, concise table. Yet, it was challenging to make the table detailed and clear at the same time. To aid clarity and readability, colour codes were used and the information in the table kept to a minimum with detailed explanations in the below text.

The Joint Display Table was organised by broad themes (as recommended by Bazeley, 2016),<sup>272</sup> in relation to the research aims. It is possible that different outcomes would have resulted if the matrix was organised in a different way. However, the decision was made in order to ground the data and results to the research aims. On a similar note, as a sole researcher, it is possible that the Joint Display Table may have looked different if completed by a research team. Each individual researcher brings their own analytical outlook to

research (as discussed in 6.4.2), and therefore it is possible that utilising a varied research team may help to minimise researcher bias.

## Chapter 8: Discussion

The overall aim of this thesis was to gain an in-depth understanding of the long-term issues and supportive care needs of TYA childhood brain tumour survivors and their caregivers. A systematic review was conducted to identify any research that had been completed in this area and to design further investigations (Chapter 2). A mixed methods approach using a convergent design was then used to address the overall aim (Chapter 3 and 4). Based on this, a quantitative phase using a cross-sectional survey and a qualitative phase using semi-structured interviews was conducted, and the results of these two phases were presented in previous chapters (Chapter 5 and 6). This chapter presents the discussion and conclusion of this study. It begins with a brief overview of the key findings discovered in the integration of the findings (Chapter 7). Following this the strengths and limitations of this study are considered (8.2) and the implications for clinical care, support services and future research listed (8.3). Next, the plans for future work after this thesis are discussed, followed by a reflective account of the PhD learning experiences (8.4/8.5). The chapter finishes with a conclusion of this thesis (8.6).

### 8.1 Key findings

The integration of the data in Chapter 7 highlighted that key unmet needs for survivors were in relation to psychological support (specifically support with anxiety and depression), occupational support and social support (socialising with similar others). Other unmet needs included brain-tumour specific fertility information, information/support around romantic relationships and support with independent living. The need for social support and fertility information is confirmative with other study findings.<sup>84, 118</sup> The need for psychological support contrasts other study findings, who found survivors to be psychologically well.<sup>276</sup> While unmet needs related to occupation, independent living and romantic relationships are novel and have not been reported before in this survivorship group.

Support services and clinical services should be mindful of unemployed survivors and those further away from diagnosis as they were more likely to experience unmet needs. Moreover, the timing of support is crucial. Extra support may be necessary at specific time-points in TYA survivors' lives. Specifically, once survivors have left education (where they felt supported), when child/teen support services decline and during the transition from children's to adult hospital based long-term follow-up care. These milestones are complicated by the turbulence of becoming a young adult while dealing with the unique late effects from their childhood brain tumour.

Like survivors, caregivers too reported the need for psychological and emotional support. This support was available during diagnosis and treatment but declined in long-term survivorship. Caregivers faced new challenges in caring for the survivor when they become teenagers and young adults. As such, the need for financial support and information was high (e.g. completing financial/benefit forms). Importantly, single parent caregivers reported more unmet needs, and therefore require attention. This research is the first to link caregiver unmet needs to relationship status (single parents), within this population. In this study single parents accounted for over a quarter of survey participants (25.7%), this is higher than the national average of single parents.<sup>277</sup> If single caregivers are more likely to experience needs, it is vital that this group are recognised by support services.

As described earlier in this thesis there are support services (e.g. brain tumour charities) that provide extensive support and resources to families who have experienced a brain tumour. Yet it is clear from the high number of unmet needs that the support available is not yet adequate for this population (e.g. focuses on survivors closer to diagnoses) or is not being fully utilised (e.g. families being unaware what support is available). This thesis identified several practical reasons why families may not access supportive services, including (but not exclusive to) location of services, waiting lists, and accessibility to information/support. Online resources and support have the potential to help those without the ability to access other types of support (such as physical meetups due to their location). However, survivors and caregivers were wary of online support, which means careful consideration should be made promoting existing online resources and in the designing of new platforms.

Significantly, this research found that unmet needs were strong predictors for both survivor and caregiver QoL. This finding highlights the importance of identifying and targeting support to those who are experiencing unmet needs. This is the first study to have investigated the association between unmet needs and QoL among long-term TYA brain tumour survivors and their caregivers. Improving survivor QoL is important as better QoL has the potential to improve long-term survival.<sup>278</sup> Equally, poor caregiver QoL warrants special attention, as the literature has long supported that poor caregiver well-being can not only affect their ability to care but also survivor wellbeing.<sup>279-281</sup>

## **8.2 Strengths and weaknesses of the thesis**

The strengths and weaknesses of the systematic review, quantitative results, qualitative results, and data integration have been presented in previous chapters. This section presents overall strengths and weaknesses of the research and thesis.



Firstly, the research presented in this thesis has provided the first in-depth mixed methods investigation of unmet supportive care needs of TYA survivors and their caregivers. The results previously summarised have added new insights to the existing literature, especially by highlighting the unmet psychological needs of this group, their need for ongoing occupational and financial support and their need for support with independent living. Also, this thesis has been able to identify particular survivors (unemployed and those further from diagnosis) and caregivers (single parents) who are more likely to have unmet needs. These novel findings have the potential to improve clinical care, focus support services and prompt further exploration in future research (8.3). Additionally, the findings of this thesis are timely as the UK government in 2020 have made research a priority to advance “*diagnosis, treatment, support or care of patients with brain tumours, including access to or the delivery of services*” (National Institute of Health Research, 2020).<sup>282</sup>

This thesis has also shown the benefit of mixed methods approaches when investigating the complicated lives of young people who have had a brain tumour in childhood, and that future research should strongly consider using both quantitative and qualitative methods to understand the needs of this population. As Chapter 7 highlighted, there were many findings that complemented each other but also areas that either the qualitative or quantitative findings highlighted that others did not. For example, a clear outcome from survivor and caregiver stories was that survivors were striving for independence, like peers their age, yet the survey questions did not cover this area. Equally, the survey findings highlighted the significant associations between needs and sociodemographic/clinical factors, which the qualitative data could not (systematically). The pragmatic grounding of the study enabled the use of methods that best answer the research objectives. Each method’s strengths allowed the overall results to be more complete and enabled the overall research aim to be addressed more comprehensively.

An important drive of this research was to give TYA survivors the opportunity to discuss their experiences in their own words. Childhood brain tumour survivors are often excluded in studies evaluating late effects of childhood cancer survivors. Partly because of concerns about the impact of their cognitive deficits on validity of assessment or because of the concern that those with a history of a childhood brain tumour may not fairly represent the greater population of children with cancer.<sup>283</sup> Furthermore, the systematic review highlighted that some previous studies within this area had used proxy measurements.<sup>112, 124</sup> Proxy measurements can be problematic, previous research has found poor survivor-proxy agreement in adult brain tumour survivors.<sup>284</sup> Furthermore, proxy agreement is typically worse for non-physical measurements, such as emotional and mental health factors.<sup>285, 286</sup> Consequently, parent and caregiver experiences remain extremely important, but there is

irreplaceable value in hearing survivor's voices too. Their insights were invaluable and future research must facilitate their involvement as much as possible.

Equally, this research has given a unique insight into the experiences and needs of parent caregivers. The systematic review highlighted their role and well-being in long-term survivorship is often overlooked, with little research focusing on caregivers. There were few (three) qualitative studies but no quantitative data that reported caregiver unmet needs, which makes this study the first of its kind, to our knowledge. This thesis highlights that caregivers too need support in long-term survivorship. In fact, the quantitative results showed that they on average experience more unmet needs than survivors. This finding is similar to findings in the wider literature that indicates that cancer caregivers can have considerably more unmet needs than the survivors they care for.<sup>287, 288</sup> With the responsibility of caring for the survivor day to day, it is integral that caregiver needs are highlighted too. However, there was an underrepresentation of male caregivers (fathers) in the sample. This is a consistent limitation in parent caregiver research,<sup>82, 289</sup> and may simply be because mothers identify as the main caregiver. Nonetheless a brain tumour diagnosis impacts an entire family. A recent study has highlighted that paternal caregiver experience has been significantly underexplored in paediatric neuro-oncology research in comparison to maternal experience.<sup>290</sup> A systematic review found that fathers of paediatric cancer patients reported different needs to mothers as healthcare providers addressed mothers as primary caregivers, which led to fathers to feeling less informed and less included.<sup>291</sup> Therefore, it is reasonable to presume that fathers of children in long-term survivorship may have differing needs in comparison to maternal caregivers. Future research should adequately represent both fathers and mothers roles.

Sampling limitations have been discussed in detail throughout the chapters, but it is worth highlighting the limitation generally. In the main participants were recruited from three NHS Trusts located in Yorkshire in the UK (some survey respondents were recruited online). Thus, potentially limiting the generalisability of the results to other regions. Especially as supportive care services will differ greatly across the UK— in relation to how clinical long-term aftercare is delivered and also what local supportive care is offered. With greater funding and time, it would be beneficial to recruit participants from other NHS Trusts throughout the UK. This would improve generalisability and increase the sample size – to allow for further statistical analysis to be carried out.

## 8.3 Implications

This section outlines the key recommendations for the improvement of clinical care, current and future support services and potential future research. It is worth noting that several implications cross all three areas.

### 8.2.1 Clinical care

- Surveillance, assessment, and treatment of the childhood brain tumour late effects should be provided by multidisciplinary follow-up clinics. Due to the multifaceted late effects of childhood brain tumour treatment, a multidisciplinary model of care has the best potential to meet survivors' varied and complex needs. For instance, an ideal clinic should be staffed (have access to): a neuro-oncologist specialising in survivorship, endocrinologist, clinical nurse specialist, (neuro-) psychologist, service coordinator, and other appropriate specialists (i.e. cardiologist, dietician). This team can then holistically address the physical, psychological and social needs of these survivors and their families. The use of a holistic needs assessment, specifically for brain tumour survivors may further enable holistic care.
- Specific TYA clinics are desirable as an interim service to provide the best care for survivors transitioning from paediatric clinics to adult clinics. As this move in services can be a big step for many families, a specific TYA clinic could assist in this transition by accustoming survivors to changes in service. For example, the survivors becoming the focus of the consultation not their parents. Equally this could be a good place for age-specific information to be relayed about upcoming challenges that may arise in young adulthood and the support available.
- Survivor psychological well-being should be closely monitored, and adequate support offered when needed. Implementation of easily administered assessment tools to identify anxiety/depression may allow for better identification of survivors in need of psychological services.
- Academic and occupational expectations must be assessed and examined over time. Occupational and financial struggles were a source of severe distress for both survivors and caregivers. Adequate support and guidance planning for life and goals after education needs to be provided to pre-empt this distress. Additionally, it cannot be assumed that there are no issues because the survivor is successful at one point in time. For example, the results highlighted that many survivors were adequately supported throughout education, but this support diminished when trying to find employment. For survivors currently unable to attain employment, it should be a part

of their clinical monitoring and survivorship plan and referrals for support put into place.

- There must be better linking between clinical support and other voluntary support services (e.g. brain tumour charity support). Specifically, clinical teams should be better educated and informed on the support services available. Many families were not aware of support available provided by charities, such as peer meetups or informational workshops. Meaning that needs continue to be unmet and these resources are not utilised to their full potential.
- Clinical teams can help families normalise the need for on-going support, especially psychological support as survivors become TYAs, which may be important to helping them cope with the numerous aging milestones (i.e. finishing education or attaining employment). Many families need the encouragement to seek support and not suffer in silence or '*get on with it*' when unmet support needs can impact QoL. QoL is especially important in caregivers as poor QoL may affect their ability to care for the survivor, impacting both the caregivers themselves and survivors indirectly.

### **8.2.2 Support services**

- Many families experienced a decline in support as they moved further from diagnosis/treatment. Support services must endeavour to continue supporting survivors and their caregivers in long-term survivorship. Being mindful of certain milestones that may mean survivors need extra support, for instance when survivors have finished education.
- Similarly, support services should target provision to single parent caregivers, as they experienced more unmet needs. It is integral that caregiver needs are met to improve their QoL. Additionally, addressing caregiver unmet needs has wider societal implications. For example, caregiver unmet needs can impact their emotional and physical well-being, which may affect their paid-work ability/productivity, having high societal costs.
- Resources should be developed/made available to help survivors develop independence, self-care, and life skills. Independence is a key issue for TYA brain tumour survivors and their caregivers. Providing resources to improve survivor independency will also reduce dependency on family caregivers.
- Occupational support for this population should be tailored to address the specific needs of individual survivors. TYA unemployed survivors seeking work require support finding appropriate employment, and support in communicating with prospective employers about necessary job-related needs. Support services should

also liaise with employers provide them with strategies to support survivors in the workplace. Some survivors have cognitive impairments that make working impossible. These survivors, and more often their caregivers, need support in obtaining and maintaining financial support (e.g. disability benefits and grants). The development of comprehensive resources in relation to financial forms and benefit guidance is vital.

- Many support services already offer survivor support to develop/maintain social relationships and social skills (e.g. peer meetups). Yet many survivors in need of this support are not currently aware of what is available or are not accessing support due to practical barriers (e.g. locations of services). Support services must attempt to reach these survivors.
- The format of information/support should be carefully considered. Resources must accommodate for those survivors with physical disabilities (i.e. eyesight loss) and cognitive deficits (i.e. slow processing). Online formats (i.e. Apps) may suit some TYA survivors, but it is important to be mindful of those survivors who cannot access this support, with alternative support provided.

### **8.2.3 Future research**

- This thesis highlights some of the barriers families face when accessing supportive care services. However, it was not a specific objective of the research. Future research should specifically aim to identify the barriers and facilitators to accessing support and investigate ways in which these barriers can be reduced.
- Future research may benefit from including the perspective of health and social care providers and support workers. It may be possible to use this approach to comprehensively identify where the barriers are to the successful use of current support services.
- Many survivors and caregivers were not aware of support services available. It would be beneficial to measure and understand more about their knowledge of support services. Such research (i.e. service evaluation) should be conducted locally to inform and guide improvements to local follow-up, due to regional and national differences. As a consequence of this data collection, we could understand how survivors and their families could be better informed of existing support.
- While the causal relationship between unmet needs and QoL is not known, this research supports the view that interventions to reduce survivor and caregiver needs may be a promising strategy for enhancing their QoL. Future studies may investigate whether addressing unmet needs through interventions improves QoL.

- Future survey research in this population should include validated measures of independence, such as the Independent Living Scale (ILS).<sup>292</sup> More data may provide additional information about the risk of no independence among survivors and help to design support to address this need.
- More research is needed to assess the best ways of providing support services and information to TYA survivors. Due to cognitive issues the way in which support is provided is key to its success. For instance, online resources may be acceptable but need careful design. More information is needed about digital formats of support and how best to utilise their functions.

#### **8.4 Planned future work and dissemination**

Following the completion of this thesis there are several plans to disseminate the findings to participants, and the appropriate clinical and support communities (e.g. brain tumour charities). In the survey and interviews participants were asked if they would like to receive a summary of the findings, which all of them indicated they would like. A summary of the findings will therefore be prepared in language appropriate to the population (with help from the PPI group) and distributed to them.

A feedback event was scheduled to take place in April 2020. The event invited a mixture of clinical staff, brain tumour charities and patient/caregiver representatives who had been involved in the study design and/or recruitment. The event was organised to disseminate the PhD findings and also to include an informal discussion about future collaborations and how we might improve support for survivors and their caregivers. Unfortunately, the event had to be cancelled due to the Covid 19 pandemic. I hope to be able to re-schedule this event in 2021. In the meantime, smaller/online feedback events have been arranged with the local long-term follow-up teams.

In addition to the published systematic review, commentary piece and qualitative paper (described on page 1) a minimum of two further peer reviewed publications will be pursued. First, I plan to publish the quantitative results separately. Second, I will endeavour to publish the mixed methods findings. Publishing mixed methods can be difficult partly due to it being a relatively new research method and clinical journals often having little experience with this type of research, and partly due to word limits. Still, I aim to condense the results and prepare an article. It is also possible that an article which practically outlines an approach to long-term survivorship care and support for TYA survivors will also be pursued, which would be based on the list of recommendations in this chapter. Regardless of whether a practical

article is pursued, the practical application of the work in this thesis will be explored with appropriate brain tumour charities and local clinical teams.

Conference presentations will also be used to disseminate this work, including neuro-oncology specific conferences. I plan to submit abstracts for both the: British neuro-oncology society (BNOS) Meeting 2021, and European Association of Neuro-Oncology (EANO) Meeting 2021.

A mindful decision was made when applying for study ethical approval to store personal data from this study for up to 10 years after its completion, in the hope of obtaining further funding for a follow-up study –which may take several years to achieve. A statement was added to all consent forms to allow the research team to contact participants in the future about further research projects. Therefore, I aim to prepare a grant application to complete longitudinal data collection. As highlighted in the systematic review- longitudinal data collection with this population is scarce and warranted.

## **8.5 A reflective account: learning experiences during PhD**

This section has been included to demonstrate my learning experience throughout the PhD. This section builds on the reflective thinking section (in 6.4.2) and recognises how my experiences have been affected as the research project progressed, providing transparency and information about my position in relation to this study.

This PhD journey has provided me with an excellent opportunity to learn new research skills and to develop them further to an advanced level. Prior to starting my PhD I considered myself as a qualitative researcher, as I did not have much research experience of capturing and analysing research data using a quantitative approach. However, I have learnt a lot about quantitative research and the relevant skills and software over the last three years. These skills have given me the confidence to conduct different statistical tests in the quantitative phase of this study. I also developed my research skills in using mixed methods by attending two training courses on mixed methods research and analysis. Therefore, I no longer see myself as simply being capable in just one approach but as being able to conduct mixed methods research and undertake and combine both qualitative and quantitative research.

This PhD experience also provided me with a good understanding of the principles and processes of research ethics and governance in health research. I had previous experience in contributing to research applications and applying for ethical approval, but I had not yet led the whole process from start to finish. In this PhD, whilst I successfully obtained both

ethics and R&I approvals, there were still some issues that I had not been prepared for. The main challenge came with the ethics and governance application processes. I had to wait a substantial amount of time to obtain separate approvals from the National Research Ethics Service Committee and the Local Trusts (three in total). Although I used this period of time well by writing these Chapters (Introduction and Systematic review chapter), on reflection, I under-estimated the time it would take and this delayed the start of the data collection. In future studies this will be accounted for sufficiently in project timelines.

The PhD has advanced my skills and ability to disseminate research to a range of audiences. Throughout my PhD I have been passionate about disseminating the research design and emerging findings. I have presented results to several oncology clinical teams, such as the Paediatric Oncology & Haematology Research Meetings based in Leeds Teaching Hospitals NHS Trust. I have also been invited to present at brain tumour charity conferences including the *brainstrust* Paediatric annual conference and the annual SUCCESS Brain Tumour Charity conference. Both conferences had a mix of delegates including survivors and families, allied professionals, academics and government stakeholders. Presenting to varied audiences can be challenging as it is important to make the presentation understandable to lay people (survivors and their families) but also engaging to those who had a good understanding of this research area. I believe I achieved this as both talks were followed by engaging discussions and I received very positive feedback from both survivors/caregivers and other audience members.

I have also had the opportunity to present twice at the European Association of Neuro-Oncology (EANO) conferences. First, in Stockholm (2018), where I discussed the findings of the systematic review. And then, in 2019 I was invited to speak at the EANO conference in Lyon. I spoke at the Educational day for nurses and allied health professionals, my talk was about my PhD mixed methods research design and initial findings. By attending and presenting at these international conferences I have had the opportunity to meet and network with likeminded researchers and PhD students. These networks I hope will be useful in future research collaborations and projects. See appendix 11 for a full list of completed dissemination.

I have also established good relationships with health professionals and families in the long-term survivorship clinics. I frequently received positive feedback from many families who appreciated the time I gave and the effort I made and thanked me for providing them with an opportunity to talk about their experiences. I believe this is an important skill for a successful researcher.



## **8.6 Conclusion**

Unmet supportive care needs were common in long-term TYA survivors of childhood brain tumours, with some survivors experiencing a very high number of unmet needs. Survivors were faced with wanting to achieve key milestones as they move into young adulthood, but late effects of treatment often made this difficult. Survivors specifically had high unmet needs in relation to their psychological health, social lives (including romantic relationships), employment, and independence. Caregivers experienced even more unmet needs. Caregiver support is most needed in relation to their psychological well-being and the survivors' financial issues. Currently there are barriers preventing survivors and caregivers accessing supportive services. This thesis provides leads to improving supportive care and long-term follow-up services. Understanding unmet needs and recognising what services are required is critical to improving survivor and caregiver quality of long-term survival following a childhood brain tumour diagnosis.

## References

1. McKinney PA. Brain tumours: Incidence, survival, and aetiology. *Neurology in Practice*. 2004;75(2):ii12-ii7.
2. Louis DN, Perry A, Reifenberger G, von Deimling A, et al. The 2016 world health organization classification of tumors of the central nervous system: A summary. *Acta Neuropathol*. 2016;131(6):803-20.
3. Ostrom QT, Gittleman H, Truitt G, Boscia A, et al. Cbtrus statistical report: Primary brain and other central nervous system tumors diagnosed in the united states in 2011–2015. *Neuro-Oncology*. 2018;20(suppl\_4):1-86.
4. Collins VP. Brain tumours: Classification and genes. *Journal of Neurology, Neurosurgery & Psychiatry*. 2004;75:2-11.
5. Office for national statistics. Cancer registration statistics, england: 2016- cancer diagnoses and age-standardised incidence rates for all types of cancer by age, sex and region including breast, prostate, lung and colorectal cancer. [Internet]. Office for National Statistics 2018 [cited 03 February 2019]. Available from: <https://www.ons.gov.uk/peoplepopulationandcommunity/healthandsocialcare/conditionsanddiseases/bulletins/cancerregistrationstatisticsengland/final2016>.
6. Cancer research uk. Brain, other cns and intracranial tumours risk factors [internet]. Cancer research uk. [cited 20 february 2019]. Available from: <Http://www.Cancerresearchuk.Org/health-professional/cancer-statistics/statistics-by-cancer-type/brain-other-cns-and-intracranial-tumours/risk-factors>.
7. Dho Y-S, Jung K-W, Ha J, Seo Y, et al. An updated nationwide epidemiology of primary brain tumors in republic of korea, 2013. *Brain tumor research and treatment*. 2017;5(1):16-23.
8. Arora RS, Alston RD, Eden TOB, Estlin EJ, et al. Age–incidence patterns of primary cns tumors in children, adolescents, and adults in england. *Neuro-Oncology*. 2009;11(4):403-13.
9. Walker EV, Davis FG. Malignant primary brain and other central nervous system tumors diagnosed in canada from 2009 to 2013. *Neuro-Oncology*. 2019;21(3):360-9.
10. Schmidt LS, Schmiegelow K, Lahteenmaki P, Trager C, et al. Incidence of childhood central nervous system tumors in the nordic countries. *Pediatr Blood Cancer*. 2011;56(1):65-9.
11. Desandes E, Guissou S, Chastagner P, Lacour B. Incidence and survival of children with central nervous system primitive tumors in the french national registry of childhood solid tumors. *Neuro-Oncology*. 2014;16(7):975-83.
12. Kaatsch P, Rickert CH, Kuhl J, Schuz J, et al. Population-based epidemiologic data on brain tumors in german children. *Cancer*. 2001;92(12):3155-64.
13. Makino K, Nakamura H, Yano S, Kuratsu J-i, et al. Population-based epidemiological study of primary intracranial tumors in childhood. *Child's Nervous System*. 2010;26(8):1029-34.
14. McKean-Cowdin R, Razavi P, Barrington-Trimis J, Baldwin RT, et al. Trends in childhood brain tumor incidence, 1973-2009. *Journal of neuro-oncology*. 2013;115(2):153-60.
15. Pollack IF, Jakacki RI. Childhood brain tumors: Epidemiology, current management and future directions. *Nature Reviews Neurology*. 2011;7:495.
16. Gibson TM, Mostoufi-Moab S, Stratton KL, Leisenring WM, et al. Temporal patterns in the risk of chronic health conditions in survivors of childhood cancer diagnosed 1970–99: A report from the childhood cancer survivor study cohort. *The Lancet Oncology*. 2018.
17. Packer RJ. Childhood brain tumors: Accomplishments and ongoing challenges. *Journal of child neurology*. 2008;23(10):1122-7.
18. National institute for health and clinical excellence. Improving outcomes for people with brain and other cns tumours the manual. 2006.

19. Heuer GG, Jackson EM, Magge SN, Storm PB. Surgical management of pediatric brain tumors. *Expert Review of Anticancer Therapy*. 2007;7:61-S8.
20. Ross SG, Northman L, Morris M, Green AL, et al. Cerebellar mutism after posterior fossa tumor resection: Case discussion and recommendations for psychoeducational intervention. *Journal of Pediatric Oncology Nursing*. 2014;31(2):78-83.
21. Tobias J, Hochhauser D. *Cancer and its management*. 7th Edition ed: Wiley-Blackwell; 2015.
22. National institute for clinical excellence. Improving supportive and palliative care for adults with cancer. [internet]. National institute for clinical excellence. [cited 11 february 2019]. Available from: <https://www.Nice.Org.Uk/guidance/csg4>.
23. Fischer C, Petriccione M, Donzelli M, Pottenger E. Improving care in pediatric neuro-oncology patients: An overview of the unique needs of children with brain tumors. *Journal of child neurology*. 2016;31(4):488-505.
24. Cancer research uk. Chemotherapy treatment. [internet]. Cancer research uk. [cited 11 november 2018]. Available from: <https://www.Cancerresearchuk.Org/about-cancer/brain-tumours/treatment/chemotherapy/treatment>.
25. Nhs england. Clinical commissioning policy: Proton beam therapy for children, teenagers and young adults in the treatment of malignant and nonmalignant tumours. [internet] nhs england. [cited 02 december 2020] available: <https://www.England.Nhs.Uk/wp-content/uploads/2020/10/proton-beam-therapy-clinical-commissioning-policy.Pdf>.
26. Howlader N, Noone A, Krapcho M, Miller D, et al. *Seer cancer statistics review, 1975-2014, national cancer institute*. . 2017;November 2016 SEER data submission posted to the SEER web site, April 2017.
27. Basta NO, James PW, Gomez-Pozo B, Craft AW, et al. Survival from childhood cancer in northern england, 1968-2005. *British Journal of Cancer*. 2011;105(9):1402-8.
28. Fairley L, Picton SV, McNally RJ, Bailey S, et al. Incidence and survival of children and young people with central nervous system embryonal tumours in the north of england, 1990-2013. *European Journal of Cancer*. 2016;61:36-43.
29. Ostrom QT, Gittleman H, Liao P, Rouse C, et al. *Cbtrus statistical report: Primary brain and central nervous system tumors diagnosed in the united states in 2007-2011*. *Neuro Oncol*. 2014;16 Suppl 4:1-63.
30. Anderson DM, Rennie KM, Ziegler RS, Neglia JP, et al. Medical and neurocognitive late effects among survivors of childhood central nervous system tumors. *Cancer*. 2001;92(10):2709-19.
31. Turner CD, Rey-Casserly C, Liptak CC, Chordas C. Late effects of therapy for pediatric brain tumor survivors. *Journal of Child Neurology*. 2009;24(11):1455-63.
32. Noone AM HN, Krapcho M, Miller D, Brest A, Yu M, Ruhl J, Tatalovich Z, Mariotto A, Lewis DR, Chen HS, Feuer EJ, Cronin KA (eds). *Seer cancer statistics review, 1975-2015, national cancer institute*. [https://seer.cancer.gov/csr/1975\\_2015/2018](https://seer.cancer.gov/csr/1975_2015/2018).
33. Geiger AM, Castellino SM. Delineating the age ranges used to define adolescents and young adults. *J Clin Oncol*. 2011;29(16):492-3.
34. What should the age range be for aya oncology? *Journal of Adolescent & Young Adult Oncology*. 2011;1(1):3-10.
35. World health organization. Adolescence: A period needing special attention. [internet]. World health organization [cited 07 january 2019]. Available from: <http://apps.Who.Int/adolescent/second-decade/section2/page1/recognizing-adolescence.Html>.
36. National cancer institute. [internet]. [cited 09 january 2019]. Available from: <https://www.Cancer.Gov/types/aya>.
37. Teenage cancer trust. [internet]. [cited 09 january 2019]. Available from: <https://www.Teenagecancertrust.Org/about-us>.
38. Hall GS. *Adolescence : Its psychology and its relations to physiology, anthropology, sociology, sex, crime, religion and education*. New York: D. Appleton and Company; 1904.
39. Erikson EH. *Identity: Youth and crisis*. Oxford, England: Norton & Co; 1968.

40. Spear LP. Adolescent neurodevelopment. *Journal of Adolescent Health*. 2013;52:7-13.
41. Claveirole A. Listening to young voices: Challenges of research with adolescent mental health service users. *Journal of Psychiatric and Mental Health Nursing*. 2004;11(3):253-60.
42. Balen R, Blyth E, Calabretto H, Fraser C, et al. Involving children in health and social research: 'Human becomings' or 'active beings'? *Childhood*. 2006;13(1):29-48.
43. Balen RH, C.; Mountain, G. and Wood, B. . Giving children a voice: Methodological & practical implications of research involving children. *Paediatric nursing*. 2000;12(10):24-9.
44. Alderson P. Ethics in: Fraser, s.; lewis, v; ding, s.; kellert, m. And robinson, c. *Doing research with children and young people*, london, uk: Sage. 2004.
45. Mayall B. 'Introduction' in alanen, I and mayall, b (eds) *conceptualising child–adult relations* london: Routledge falmer, 1–10, 81–96. 2002.
46. Gardner MH, Mrug S, Schwebel DC, Phipps S, et al. Benefit finding and quality of life in caregivers of childhood cancer survivors: The moderating roles of demographic and psychosocial factors. *Cancer nursing*. 2017;40(5):28-37.
47. Witt WP, Litzelman K, Wisk LE, Spear HA, et al. Stress-mediated quality of life outcomes in parents of childhood cancer and brain tumor survivors: A case-control study. *Quality of Life Research*. 2010;19(7):995-1005.
48. Bennett E, English MW, Rennoldson M, Starza-Smith A. Predicting parenting stress in caregivers of children with brain tumours. *Psycho-Oncology*. 2013;22(3):629-36.
49. Minaya Flores P, Berbis J, Chinot O, Auquier P. Assessing the quality of life among caregivers of patients with gliomas. *Neuro-oncology practice*. 2014;1(4):191-7.
50. Paula Sherwood CWG, Heidi Donovan, Jason Weimer, Sarah Belcher and Barbara Given. The impact of caregivers' depression on their physical health. IPOS Confernece 2016. 2016.
51. Choi EK, Yoon SJ, Kim J-H, Park HJ, et al. Depression and distress in caregivers of children with brain tumors undergoing treatment: Psychosocial factors as moderators. *Psycho-Oncology*. 2016;25(5):544-50.
52. Boele FW, Given CW, Given BA, Donovan HS, et al. Family caregivers' level of mastery predicts survival of patients with glioblastoma: A preliminary report. *Cancer*. 2017;123(5):832-40.
53. Wadhwa D, Burman D, Swami N, Rodin G, et al. Quality of life and mental health in caregivers of outpatients with advanced cancer. *Psychooncology*. 2013;22(2):403-10.
54. Grunfeld E, Coyle D, Whelan T, Clinch J, et al. Family caregiver burden: Results of a longitudinal study of breast cancer patients and their principal caregivers. *CMAJ : Canadian Medical Association journal = journal de l'Association medicale canadienne*. 2004;170(12):1795-801.
55. Nicklin E, Velikova G, Hulme C, Rodriguez Lopez R, et al. Long-term issues and supportive care needs of adolescent and young adult childhood brain tumour survivors and their caregivers: A systematic review. *Psychooncology*. 2019;28(3):477-87.
56. Miller K, Merry, B. and Miller J. . Seasons of survivorship revisited. *The Cancer Journal* 2008;14(6):369-74.
57. Rowland JH, Bellizzi KM. Cancer survivors and survivorship research: A reflection on today's successes and tomorrow's challenges. *Hematology/oncology clinics of North America*. 2008;22(2):181-200.
58. National cancer insitute. Survivorship. Nci dictionary of cancer terms. [internet]. [cited 12 february 2019] available from: <https://www.Cancer.Gov/publications/dictionaries/cancer-terms/def/survivorship>.
59. Hewitt MG, S. and Stovall, E. *From cancer patient to cancer survivor—lost in transition*. Washington, DC: National Academies Press; 2005.
60. Rowland JH, Kent EE, Forsythe LP, Loge JH, et al. Cancer survivorship research in europe and the united states: Where have we been, where are we going, and what can we learn from each other? *Cancer*. 2013;119 Suppl 11:2094-108.

61. National cancer research institute. The uk top living with and beyond cancer research priorities. [internet]. National cancer research institute [cited: 02 december 2018]. Available from: <https://www.Ncri.Org.Uk/lwbc/2018>
62. D'Angio GJ. Pediatric cancer in perspective: Cure is not enough. *Cancer*. 1975;35(3 suppl):866-70.
63. National collaborating centre for cancer. Improving outcomes for people with brain and other cns tumours. [internet]. National collaborating centre for cancer, 2006 [cited: 21 august 2017]. Available from: <https://www.Nice.Org.Uk/guidance/csg10>.
64. Fialka-Moser V, Crevenna R, Korpan M, Quittan M. Cancer rehabilitation: Particularly with aspects on physical impairments. *Journal of rehabilitation medicine*. 2003;35(4):153-62.
65. Sanson-Fisher R, Girgis A, Boyes A, Bonevski B, et al. The unmet supportive care needs of patients with cancer. *Cancer*. 2000;88(1):226-37.
66. Clarke L, Clarke M, Clarke T. How useful are cochrane reviews in identifying research needs? *Journal of health services research & policy*. 2007;12(2):101-3.
67. Bradford NK, Chan RJ. Health promotion and psychological interventions for adolescent and young adult cancer survivors: A systematic literature review. *Cancer Treat Rev*. 2017;55:57-70.
68. Galan S, de la Vega, R., Miro, J. Needs of adolescents and young adults after cancer treatment: A systematic review. *Eur J Cancer Care (Engl)*. 2016.
69. Wakefield CE, McLoone J, Goodenough B, Lenthén K, et al. The psychosocial impact of completing childhood cancer treatment: A systematic review of the literature. *J Pediatr Psychol*. 2010;35(3):262-74.
70. Bell H, Ownsworth T, Lloyd O, Sheeran N, et al. A systematic review of factors related to children's quality of life and mental health after brain tumor. *Psycho-Oncology*. 2018;27(10):2317-26.
71. Ford E, Catt S, Chalmers A, Fallowfield L. Systematic review of supportive care needs in patients with primary malignant brain tumors. *Neuro Oncol*. 2012;14(4):392-404.
72. Madsen K, Poulsen HS. Needs for everyday life support for brain tumour patients' relatives: Systematic literature review. *Eur J Cancer Care (Engl)*. 2011;20(1):33-43.
73. Moher D, Liberati A, Tetzlaff J, Altman DG, et al. Preferred reporting items for systematic reviews and meta-analyses: The prisma statement. *BMJ*. 2009;339:2535.
74. Pluye p, robert e, cargo m, bartlett g. Et al. Proposal: A mixed methods appraisal tool for systematic mixed studies reviews. [internet]. 2011 [cited 13 september 2017]. Available from: <http://mixedmethodsappraisaltoolpublic.Pbworks.Com>.
75. Pace R, Pluye P, Bartlett G, Macaulay AC, et al. Testing the reliability and efficiency of the pilot mixed methods appraisal tool (mmat) for systematic mixed studies review. *International Journal of Nursing Studies*. 2012;49(1):47-53.
76. Souto RQ, Khanassov V, Hong QN, Bush PL, et al. Systematic mixed studies reviews: Updating results on the reliability and efficiency of the mixed methods appraisal tool. *International Journal of Nursing Studies*. 2015;52(1):500-1.
77. Popay J, Roberts H, Sowden A, Petticrew M, et al. Guidance on the conduct of narrative synthesis in systematic reviews. [internet]. Lancaster university [cited 21 august 2017]. Available from: [www.Lancaster.Ac.Uk/shm/research/nssr/research/.../ns\\_synthesis\\_guidance\\_v1.Pdf](http://www.Lancaster.Ac.Uk/shm/research/nssr/research/.../ns_synthesis_guidance_v1.Pdf)
78. Pope C, Mays, N. and Popay J. Synthesizing qualitative and quantitative health evidence. Berkshire: Open University Press; 2007.
79. Ryan R. Cochrane consumers and communication review group: Data synthesis and analysis. [internet]. [cited 21 august 2017]. Available from: <https://cccrq.Cochrane.Org/sites/cccrq.Cochrane.Org/files/public/uploads/analysis.Pdf>
80. Ferrell BR, Grant M, Funk B, Garcia N, et al. Quality of life in breast cancer. *Cancer Pract*. 1996;4(6):331-40.
81. Pluye P, Robert E, Cargo M, Bartlett G, et al. Proposal: A mixed methods appraisal tool for systematic mixed studies reviews. [internet]. [cited 01 september 2017]. Available at: <http://mixedmethodsappraisaltoolpublic.Pbworks.Com>.

82. Wilford J, Buchbinder D, Fortier MA, Osann K, et al. "She was a little social butterfly": A qualitative analysis of parent perception of social functioning in adolescent and young adult brain tumor survivors. *Journal of Pediatric Oncology Nursing*. 2017;34(4):239-49.
83. Hobbie WL, Ogle S, Reilly M, Barakat L, et al. Adolescent and young adult survivors of childhood brain tumors: Life after treatment in their own words. *Cancer Nursing*. 2016;39(2):134-43.
84. Liptak C, Brinkman T, Bronson A, Delaney B, et al. A social program for adolescent and young adult survivors of pediatric brain tumors: The power of a shared medical experience. *Journal of Psychosocial Oncology*. 2016;34(6):493-511.
85. Boydell KM, Stasiulis E, Greenberg M, Greenberg C, et al. I'll show them: The social construction of (in)competence in survivors of childhood brain tumors. *Journal of Pediatric Oncology Nursing*. 2008;25(3):164-74.
86. Chen CM, Chen YC, Haase JE. Games of lives in surviving childhood brain tumors. *Western Journal of Nursing Research*. 2008;30(4):435-57.
87. Gray RE, Doan BD, Shermer P, Fitzgerald AV, et al. Surviving childhood cancer: A descriptive approach to understanding the impact of life-threatening illness. *Psycho-Oncology*. 1992;1(4):235-45.
88. Chou LN, Hunter A. Factors affecting quality of life in taiwanese survivors of childhood cancer. *Journal of Advanced Nursing*. 2009;65(10):2131-41.
89. Maunsell E, Pogany L, Barrera M, Shaw AK, et al. Quality of life among long-term adolescent and adult survivors of childhood cancer. *Journal of Clinical Oncology*. 2006;24(16):2527-35.
90. Poggi G, Liscio M, Galbiati S, Adduci A, et al. Brain tumors in children and adolescents: Cognitive and psychological disorders at different ages. *Psycho-Oncology*. 2005;14(5):386-95.
91. Deatrck JA, Mullaney EK, Mooney-Doyle K. Exploring family management of childhood brain tumor survivors. *Journal of Pediatric Oncology Nursing*. 2009;26(5):303-11.
92. Gunn ME, Mort S, Arola M, Taskinen M, et al. Quality of life and late-effects among childhood brain tumor survivors: A mixed method analysis. *Psycho-Oncology*. 2016;25(6):677-83.
93. Vance YH, Eiser C, Horne B. Parents' views of the impact of childhood brain tumours and treatment on young people's social and family functioning. *Clinical Child Psychology and Psychiatry*. 2004;9(2):271-88.
94. Pfitzer C, Zynda A, Hohmann C, Keil T, et al. Educational level of childhood brain tumor survivors: Results from a german survey. *Klinische Padiatrie*. 2013;225(3):138-44.
95. King TZ, Na S. Cumulative neurological factors associated with long-term outcomes in adult survivors of childhood brain tumors. *Child Neuropsychology*. 2016;22(6):748-60.
96. King TZ, Smith KM, Ivanisevic M. The mediating role of visuospatial planning skills on adaptive function among young-adult survivors of childhood brain tumor. *Archives of Clinical Neuropsychology*. 2015;30(5):394-403.
97. Maddrey AM, Bergeron JA, Lombardo ER, McDonald NK, et al. Neuropsychological performance and quality of life of 10 year survivors of childhood medulloblastoma. *Journal of Neuro-Oncology*. 2005;72(3):245-53.
98. Deatrck JA, Hobbie W, Ogle S, Fisher MJ, et al. Competence in caregivers of adolescent and young adult childhood brain tumor survivors. *Health Psychology*. 2014;33(10):1103-12.
99. Demers C, Gelinias I, Carret AS. Activities of daily living in survivors of childhood brain tumor. *American Journal of Occupational Therapy*. 2016;70(1):7001220040p1-8.
100. Frange P, Alapetite C, Gaboriaud G, Bours D, et al. From childhood to adulthood: Long-term outcome of medulloblastoma patients. The institut curie experience (1980-2000). *Journal of Neuro-Oncology*. 2009;95(2):271-9.
101. Hocking MC, Hobbie WL, Deatrck JA, Hardie TL, et al. Family functioning mediates the association between neurocognitive functioning and health-related quality of life in young adult survivors of childhood brain tumors. *Journal of Adolescent & Young Adult Oncology*. 2015;4(1):18-25.

102. Lehmann V, Tuinman MA, Keim MC, Hagedoorn M, et al. Am i a 6 or a 10? Mate value among young adult survivors of childhood cancer and healthy peers. *Journal of Adolescent & Young Adult Oncology*. 2017;07:07.
103. Klosky JL, Foster RH, Li ZH, Peasant C, et al. Risky sexual behavior in adolescent survivors of childhood cancer: A report from the childhood cancer survivor study. *Health Psychology*. 2014;33(8):868-77.
104. Zebrack BJ, Foley S, Wittmann D, Leonard M. Sexual functioning in young adult survivors of childhood cancer. *Psycho-Oncology*. 2010;19(8):814-22.
105. Lehmann V, Tuinman MA, Keim MC, Winning AM, et al. Psychosexual development and satisfaction in long-term survivors of childhood cancer: Neurotoxic treatment intensity as a risk indicator. *Cancer*. 2017;123(10):1869-76.
106. Koustenis E, Pfitzer C, Balcerek M, Reinmuth S, et al. Impact of cranial irradiation and brain tumor location on fertility: A survey. *Klinische Padiatrie*. 2013;225(6):320-4.
107. Armuand G, Skoog-Svanberg A, Bladh M, Sydsjo G. Reproductive patterns among childhood and adolescent cancer survivors in sweden: A population-based matched-cohort study. *Journal of Clinical Oncology*. 2017;35(14):1577-83.
108. Boman KK, Hoven E, Anclair M, Lannering B, et al. Health and persistent functional late effects in adult survivors of childhood cns tumours: A population-based cohort study. *European Journal of Cancer*. 2009;45(14):2552-61.
109. Ait Khelifa-Gallois N, Laroussinie F, Puget S, Sainte- Rose C, et al. Long-term functional outcome of patients with cerebellar pilocytic astrocytoma surgically treated in childhood. *Brain Injury*. 2015;29(3):366-73.
110. Brinkman TM, Reddick WE, Luxton J, Glass JO, et al. Cerebral white matter integrity and executive function in adult survivors of childhood medulloblastoma. *Neuro-Oncology*. 2012;14 Suppl 4:iv25-36.
111. Jereb B, Korenjak R, Krzisznik C, Petric-Grabnar G, et al. Late sequelae in children treated for brain tumors and leukemia. *Acta Oncologica*. 1994;33(2):159-64.
112. Barakat LP, Li Y, Hobbie WL, Ogle SK, et al. Health-related quality of life of adolescent and young adult survivors of childhood brain tumors. *Psycho-Oncology*. 2015;24(7):804-11.
113. McCurdy MD, Rane S, Daly BP, Jacobson LA. Associations among treatment-related neurological risk factors and neuropsychological functioning in survivors of childhood brain tumor. *Journal of Neuro-Oncology*. 2016;127(1):137-44.
114. Strauser D, Wagner S, Wong AWK, O'Sullivan D. Career readiness, developmental work personality and age of onset in young adult central nervous system survivors. *Disability & Rehabilitation*. 2013;35(7):543-50.
115. McClellan W, Klemp JR, Krebill H, Ryan R, et al. Understanding the functional late effects and informational needs of adult survivors of childhood cancer. *Oncology Nursing Forum*. 2013;40(3):254-62.
116. Maurice-Stam H, Verhoof EJ, Caron HN, Grootenhuis MA. Are survivors of childhood cancer with an unfavourable psychosocial developmental trajectory more likely to apply for disability benefits? *Psycho-Oncology*. 2013;22(3):708-14.
117. Ahomaki R, Harila-Saari A, Parkkola K, Matomaki J, et al. Compulsory military service as a measure of later physical and cognitive performance in male survivors of childhood cancer. *Acta Oncologica*. 2017:1-8.
118. D'Agostino NM, Edelstein K. Psychosocial challenges and resource needs of young adult cancer survivors: Implications for program development. *Journal of Psychosocial Oncology*. 2013;31(6):585-600.
119. Jayakar R, King TZ, Morris R, Na S. Hippocampal volume and auditory attention on a verbal memory task with adult survivors of pediatric brain tumor. *Neuropsychology*. 2015;29(2):303-19.
120. King TZ, Na S, Mao H. Neural underpinnings of working memory in adult survivors of childhood brain tumors. *Journal of the International Neuropsychological Society*. 2015;21(7):494-505.

121. Taiwo Z, Na S, King TZ. The neurological predictor scale: A predictive tool for long-term core cognitive outcomes in survivors of childhood brain tumors. *Pediatric Blood & Cancer*. 2017;64(1):172-9.
122. King TZ, Wang L, Mao H. Disruption of white matter integrity in adult survivors of childhood brain tumors: Correlates with long-term intellectual outcomes. *PLoS ONE*. 2015;10(7):1-17.
123. Ailion AS, King TZ, Wang L, Fox ME, et al. Cerebellar atrophy in adult survivors of childhood cerebellar tumor. *Journal of the International Neuropsychological Society*. 2016;22(5):501-11.
124. McCurdy MD, Turner EM, Barakat LP, Hobbie WL, et al. Discrepancies among measures of executive functioning in a subsample of young adult survivors of childhood brain tumor: Associations with treatment intensity. *Journal of the International Neuropsychological Society*. 2016;22(9):900-10.
125. Cohen LE, Gordon JH, Popovsky EY, Sainath NN, et al. Bone density in post-pubertal adolescent survivors of childhood brain tumors. *Pediatric Blood & Cancer*. 2012;58(6):959-63.
126. Petraroli M, D'Alessio E, Ausili E, Barini A, et al. Bone mineral density in survivors of childhood brain tumours. *Childs Nervous System*. 2007;23(1):59-65.
127. Heikens J, Michiels EM, Behrendt H, Endert E, et al. Long-term neuro-endocrine sequelae after treatment for childhood medulloblastoma. *European Journal of Cancer*. 1998;34(10):1592-7.
128. Mandrell BN, Wise M, Schoumacher RA, Pritchard M, et al. Excessive daytime sleepiness and sleep-disordered breathing disturbances in survivors of childhood central nervous system tumors. *Pediatric Blood & Cancer*. 2012;58(5):746-51.
129. Pietila S, Makiperna A, Koivisto AM, Lenko HL. Growth impairment and gonadal axis abnormalities are common in survivors of paediatric brain tumours. *Acta Paediatrica*. 2017;06:06.
130. Uday S, Murray RD, Picton S, Chumas P, et al. Endocrine sequelae beyond 10 years in survivors of medulloblastoma. *Clinical Endocrinology*. 2015;83(5):663-70.
131. Hoffmann A, Bootsvelde K, Gebhardt U, Daubenbuchel AM, et al. Nonalcoholic fatty liver disease and fatigue in long-term survivors of childhood-onset craniopharyngioma. *European Journal of Endocrinology*. 2015;173(3):389-97.
132. Johannsdottir IM, Hjermstad MJ, Moum T, Wesenberg F, et al. Increased prevalence of chronic fatigue among survivors of childhood cancers: A population-based study. *Pediatric Blood & Cancer*. 2012;58(3):415-20.
133. Turkel SB, Tishler D, Tavare CJ. Late onset psychosis in survivors of pediatric central nervous system malignancies. *Journal of Neuropsychiatry & Clinical Neurosciences*. 2007;19(3):293-7.
134. Palma E, Deatrack JA, Hobbie WL, Ogle SK, et al. Maternal caregiving demands for adolescent and young adult survivors of pediatric brain tumors. *Oncology Nursing Forum*. 2015;42(3):222-9.
135. Lucas MS, Barakat LP, Ulrich CM, Jones NL, et al. Mother-caregiver expectations for function among survivors of childhood brain tumors. *Supportive Care in Cancer*. 2016;24(5):2147-54.
136. Vance Y, Eiser, C. and Horne, B. . Parents' views of the impact of childhood brain tumours and treatment on young people's social and family functioning. *Clinical Child Psychology and Psychiatry* 2004.
137. Wasilewski-Masker K, Gilleland-Marchak J, Vangile K, Cherven B, et al. Adolescent and young adult perceptions of cancer survivor care and supportive programming. *Journal of Community and Supportive Oncology*. 2016;14(7):291-8.
138. Macartney G, Harrison MB, VanDenKerkhof E, Stacey D, et al. Quality of life and symptoms in pediatric brain tumor survivors: A systematic review. *J Pediatr Oncol Nurs*. 2014;31(2):65-77.



139. Keegan TH, Lichtensztajn DY, Kato I, Kent EE, et al. Unmet adolescent and young adult cancer survivors information and service needs: A population-based cancer registry study. *J Cancer Surviv.* 2012;6(3):239-50.
140. Shaw S. Endocrine late effects in survivors of pediatric brain tumors. *Journal of Pediatric Oncology Nursing.* 2009;26(5):295-302.
141. Hoven E, Lannering B, Gustafsson G, Boman KK. The met and unmet health care needs of adult survivors of childhood central nervous system tumors: A double-informant, population-based study. *Cancer.* 2011;117(18):4294-303.
142. Beek L, Schappin R, Gooskens R, Huisman J, et al. Surviving a brain tumor in childhood: Impact on family functioning in adolescence. *Psycho-Oncology.* 2015;24(1):89-94.
143. Brain tumour research. Addressing the historic underfunding of brain tumour research. [internet]. 2016. [cited 03 January 2018] available from: <https://www.Braintumourresearch.Org/docs/default-source/default-document-library/public-affairs-and-campaigning-documents/brain-tumour-research---report-on-national-research-funding---2016.Pdf>.
144. National cancer research institute. Spend by research category and disease site. [internet]. [cited 13 January 2019] available from: <https://www.Ncri.Org.Uk/ncr-cancer-research-database-old/spend-by-research-category-and-disease-site/>.
145. Office for national statistics. Cancer survival in England: Patients diagnosed between 2010 and 2014 and followed up to 2015. [internet]. Office for national statistics, 2016 [cited 01 May 2019]. Available from: <https://www.Ons.Gov.Uk/peoplepopulationandcommunity/healthandsocialcare/conditionsanddiseases/bulletins/cancersurvivalinenglandadultsdiagnosed/2010and2014andfollowedupto2015>.
146. Morgan DL. Paradigms lost and pragmatism regained: Methodological implications of combining qualitative and quantitative methods. *Journal of Mixed Methods Research.* 2007;1(1):48-76.
147. O'Cathain A, Murphy E, Nicholl J. The quality of mixed methods studies in health services research. *Journal of health services research & policy.* 2008;13(2):92-8.
148. Creswell J. A concise introduction to mixed methods research London: SAGE Publications Ltd.; 2015.
149. Tariq S, Woodman J. Using mixed methods in health research. *JRSM short reports.* 2013;4(6):1-8.
150. Bryman A. Quantity and quality in social research. 2nd ed. London: Routledge; 2004.
151. Speziale HS, Streubert HJ, Carpenter DR. Qualitative research in nursing: Advancing the humanistic imperative: Wolters Kluwer Health/Lippincott Williams & Wilkins; 2011.
152. Morgan D. Integrating qualitative and quantitative methods. London: SAGE Publishers Ltd. ; 2014.
153. Bryman A. Social research methods: Oxford University Press; 2008.
154. Pope C, van Royen P, Baker R. Qualitative methods in research on healthcare quality. *Quality and Safety in Health Care.* 2002;11(2):148-52.
155. Chafe R. The value of qualitative description in health services and policy research. *Healthcare policy = Politiques de sante.* 2017;12(3):12-8.
156. Holloway I. Basic concepts for qualitative research: Wiley; 1997.
157. Bowling A. Research methods in health - investigating health and health services. Buckingham: Open University Press; 1997.
158. Braun V, Clarke V. Successful qualitative research: A practical guide for beginners: SAGE Publications; 2013.
159. Creswell J. Research design: Qualitative, quantitative and mixed methods approaches. 4th ed. London: SAGE Publications; 2014.
160. Johnson RB, Onwuegbuzie AJ. Mixed methods research: A research paradigm whose time has come. *Educational Researcher.* 2004;33(7):14-26.

161. Teddlie CaT, A. Foundations of mixed methods research -integrating quantitative and qualitative approaches in the social and behavioral sciences. California: SAGE Publications; 2009.
162. Creswell J, Plano Clark V. Choosing a mixed methods design [chapter 3 from: Designing and conducting mixed methods research]. In: Creswell JW, Plano Clark VL, editors. Designing and conducting mixed methods research. 2nd ed ed. Los Angeles: SAGE Publications; 2011. p. 53-106.
163. Johnson RB, Onwuegbuzie AJ, Turner LA. Toward a definition of mixed methods research. *Journal of Mixed Methods Research*. 2007;1(2):112-33.
164. Sciences. NOoBaS. Best practices for mixed methods research in the health sciences. Bethesda: National Institutes of Health. ; 2018.
165. O'Cathain A, Murphy E, Nicholl J. Why, and how, mixed methods research is undertaken in health services research in England: A mixed methods study. *BMC Health Services Research*. 2007;7(1):85.
166. O'Cathain A. Reporting mixed methods projects. *Mixed methods research for nursing and the health sciences*; 2009.
167. Bryman A. Integrating quantitative and qualitative research: How is it done? *Qualitative Research*. 2006;6(1):97-113.
168. Morgan D. Practical strategies for combining qualitative and quantitative methods: Applications to health research. *Qual Health Res*. 1998;8:362-76.
169. Nhs England. Building greater insight through qualitative research. [internet]. Nhs England, 2017 [cited 02 April 2019]. Available from: <https://www.England.Nhs.Uk/publication/bite-size-guides-to-patient-insight/>.
170. Bryman A. Barriers to integrating quantitative and qualitative research. *Journal of Mixed Methods Research*. 2007;1(1):8-22.
171. O'Cathain A, Murphy E, Nicholl J. Multidisciplinary, interdisciplinary, or dysfunctional? Team working in mixed-methods research. *Qualitative Health Research*. 2008;18(11):1574-85.
172. Sandelowski M. Combining qualitative and quantitative sampling, data collection, and analysis techniques in mixed-method studies. *Res Nurs Health*. 2000;23(3):246-55.
173. Murphy E, Dingwall R, Greatbatch D, Parker S, et al. Qualitative research methods in health technology assessment: A review of the literature. *Health technology assessment (Winchester, England)*. 1998;2(16):iii-ix, 1-274.
174. Guba EG, Lincoln YS. Paradigmatic controversies, contradictions, and emerging confluences. *The sage handbook of qualitative research*, 3rd ed. Thousand Oaks, CA: Sage Publications Ltd; 2005. p. 191-215.
175. Guba EG, Lincoln YS. Competing paradigms in qualitative research. *Handbook of qualitative research*. Thousand Oaks, CA, US: Sage Publications, Inc; 1994. p. 105-17.
176. Guba EG. The paradigm dialog. Thousand Oaks, CA, US: Sage Publications, Inc; 1990. 424- p.
177. Tashakkori A, Teddlie C, Teddlie CB. *Handbook of mixed methods in social & behavioral research*: SAGE Publications; 2003.
178. Creswell J. *Conducting mixed methods research*. California Sage Publications; 2007.
179. Creswell JW. *Research design: Qualitative, quantitative, and mixed methods approaches*: SAGE Publications; 2009.
180. Morse JM. Approaches to qualitative-quantitative methodological triangulation. *Nursing research*. 1991;40(2):120-3.
181. Greene JC, Caracelli VJ, Graham WF. Toward a conceptual framework for mixed-method evaluation designs. *Educational Evaluation and Policy Analysis*. 1989;11(3):255-74.
182. Guillemin M, Gillam L. Ethics, reflexivity, and "ethically important moments" in research. *Qualitative Inquiry*. 2004;10(2):261-80.
183. Corti L, Day A, Backhouse G. Confidentiality and informed consent: Issues for consideration in the preservation of and provision of access to qualitative data archives. 2000. 2000;1(3).

184. Strobe statement - strengthening the reporting of observational studies in epidemiology. [internet]. [cited 07 november 2018]. Available from: <https://www.Strobe-statement.Org/index.Php?Id=strobe-home> [
185. Alderman AK, Salem B. Survey research. *Plastic and reconstructive surgery*. 2010;126(4):1381-9.
186. Schleyer TK, Forrest JL. Methods for the design and administration of web-based surveys. *J Am Med Inform Assoc*. 2000;7(4):416-25.
187. van Gelder MMHJ, Bretveld RW, Roeleveld N. Web-based questionnaires: The future in epidemiology? *American Journal of Epidemiology*. 2010;172(11):1292-8.
188. Freake H, Barley V, Kent G. Adolescents' views of helping professionals: A review of the literature. *Journal of adolescence*. 2007;30(4):639-53.
189. Polit DF, Beck CT. *Nursing research: Principles and methods*: Lippincott Williams & Wilkins; 2004.
190. Bowling A, Ebrahim S. *Handbook of health research methods : Investigation, measurement and analysis*. Berkshire, UNITED KINGDOM: McGraw-Hill Education; 2005.
191. Parahoo K. *Nursing research: Principles, process and issues*: Palgrave Macmillan; 2014.
192. Roach KE. A clinician's guide to specification and sampling. *The Journal of orthopaedic and sports physical therapy*. 2001;31(12):753-8.
193. Setia MS. Methodology series module 5: Sampling strategies. *Indian J Dermatol*. 2016;61(5):505-9.
194. Hector C, Holch PT, Warrington L, Morris C, et al. Development of online patient-advice for the self-management of low-level chemotherapy related toxicities (erapid). *Psycho-Oncology*. 2013;22:11.
195. Facit. Pediatric functional assessment of cancer therapy – brain tumor survivor (version 2) patient version (age 12 years and older). [internet]. [cited 23 march 2018]. Available from: <http://www.Facit.Org/facitorg/questionnaires>.
196. Eortc. Quality of life of cancer patients (qlq-c30). [internet] [cited 23 march 2018]. Available from: <https://qol.Eortc.Org/questionnaire/eortc-qlq-c30/>.
197. Eortc. Qlq-bn20. [intenet]. [cited 23 march 2018]. Available from: <https://qol.Eortc.Org/questionnaire/bn20-update/23.03.2018>.
198. Md anderson cancer center. Mdasi brain tumor module (mdasi-bt). [Internet]. [cited 23.02.2018]. Available from: <https://www.mdanderson.org/research/departments-labs-institutes/departments-divisions/symptom-research/symptom-assessment-tools/md-anderson-symptom-inventory-brain-tumor.html>.
199. Weitzner MA, Meyers CA, Gelke CK, Byrne KS, et al. The functional assessment of cancer therapy (fact) scale. Development of a brain subscale and revalidation of the general version (fact-g) in patients with primary brain tumors. *Cancer*. 1995;75(5):1151-61.
200. Boyes A, Girgis A, Lecathelinais C. Brief assessment of adult cancer patients' perceived needs: Development and validation of the 34-item supportive care needs survey (scns-sf34). *Journal of evaluation in clinical practice*. 2009;15(4):602-6.
201. Girgis A, Lambert S, Lecathelinais C. The supportive care needs survey for partners and caregivers of cancer survivors: Development and psychometric evaluation. *Psychooncology*. 2011;20(4):387-93.
202. Hall A, D'Este C, Tzelepis F, Sanson-Fisher R, et al. The survivor unmet needs survey (suns) for haematological cancer survivors: A cross-sectional study assessing the relevance and psychometric properties. *BMC health services research*. 2014;14:211-.
203. Cox CL, Sherrill-Mittleman DA, Riley BB, Hudson MM, et al. Development of a comprehensive health-related needs assessment for adult survivors of childhood cancer. *J Cancer Surviv*. 2013;7(1):1-19.
204. Hodgkinson K, Butow P, Hunt GE, Pendlebury S, et al. The development and evaluation of a measure to assess cancer survivors' unmet supportive care needs: The casun (cancer survivors' unmet needs measure). *Psychooncology*. 2007;16(9):796-804.
205. Clinton-McHarg T, Carey M, Sanson-Fisher R, D'Este C, et al. Preliminary development and psychometric evaluation of an unmet needs measure for adolescents and

- young adults with cancer: The cancer needs questionnaire - young people (cnq-yp). Health and quality of life outcomes. 2012;10:13.
206. McElduff P, Boyes A, Zucca A, Girgis A. Supportive care needs survey: A guide to administration, scoring and analysis. 2004.
207. Lim J-w, Zebrack B. Social networks and quality of life for long-term survivors of leukemia and lymphoma. Supportive Care in Cancer. 2006;14(2):185-92.
208. Weitzner MA, Jacobsen PB, Wagner H, Jr., Friedland J, et al. The caregiver quality of life index-cancer (cqolc) scale: Development and validation of an instrument to measure quality of life of the family caregiver of patients with cancer. Qual Life Res. 1999;8(1-2):55-63.
209. Brouwer WB, van Exel NJ, van Gorp B, Redekop WK. The carerqol instrument: A new instrument to measure care-related quality of life of informal caregivers for use in economic evaluations. Qual Life Res. 2006;15(6):1005-21.
210. Cohen R, Leis AM, Kuhl D, Charbonneau C, et al. Qoliti-f: Measuring family carer quality of life. Palliative Medicine. 2006;20(8):755-67.
211. Graessel E, Berth H, Lichte T, Grau H. Subjective caregiver burden: Validity of the 10-item short version of the burden scale for family caregivers bsfc-s. BMC geriatrics. 2014;14:23.
212. Adelman RD, Tmanova LL, Delgado D, Dion S, et al. Caregiver burden: A clinical review. Jama. 2014;311(10):1052-60.
213. Vitaliano PP, Russo J, Young HM, Becker J, et al. The screen for caregiver burden. The Gerontologist. 1991;31(1):76-83.
214. Pendergrass A, Malnis C, Graf U, Engel S, et al. Screening for caregivers at risk: Extended validation of the short version of the burden scale for family caregivers (bsfc-s) with a valid classification system for caregivers caring for an older person at home. BMC Health Serv Res. 2018;18(1):229.
215. Wingate AL, Lackey NR. A description of the needs of noninstitutionalized cancer patients and their primary care givers. Cancer Nurs. 1989;12(4):216-25.
216. Kim Y, Kashy DA, Spillers RL, Evans TV. Needs assessment of family caregivers of cancer survivors: Three cohorts comparison. Psychooncology. 2010;19(6):573-82.
217. Fayers PM, Machin D. Quality of life: Assessment, analysis, and interpretation: Wiley; 2000.
218. Field A. Discovering statistics using spss: SAGE Publications; 2009.
219. Osbourne JW, Waters EM. Four assumptions of multiple regression that researchers should always test. Practical Assessment, Research and Evaluation. 2002;8:2.
220. Statistic solutions. Normality. [internet]. Statistic solutions, 2013 [cited 02 January 2020]. Available from: <https://www.Statisticsolutions.Com/academic-solutions/resources/directory-of-statistical-analyses/normality/>.
221. Ratner B. The correlation coefficient: Its values range between +1/-1, or do they? Journal of Targeting, Measurement and Analysis for Marketing. 2009;17(2):139-42.
222. Kvale S. Doing interviews: SAGE Publications; 2008.
223. Rubin HJ, Rubin I. Qualitative interviewing: The art of hearing data: Sage Publications; 1995.
224. Hill M. Participatory research with children. Child & Family Social Work. 1997;2(3):171-83.
225. Patton MQ, Fund RECM. Qualitative research & evaluation methods: SAGE Publications; 2002.
226. Marshall MN. Sampling for qualitative research. Family practice. 1996;13(6):522-5.
227. Green J, Thorogood N. Qualitative methods for health research: SAGE Publications; 2004.
228. Francis JJ, Johnston M, Robertson C, Glidewell L, et al. What is an adequate sample size? Operationalising data saturation for theory-based interview studies. Psychology & health. 2010;25(10):1229-45.
229. Miles MB, Huberman AM, Saldana J. Qualitative data analysis: A methods sourcebook: SAGE Publications; 2013.

230. Teddlie C, Yu F. Mixed methods sampling: A typology with examples. 2007;1(1):77-100.
231. Ritchie J, Lewis J, Lewis PSPJ, Nicholls CMN, et al. Qualitative research practice: A guide for social science students and researchers: SAGE Publications; 2013.
232. Sandelowski M. Sample size in qualitative research. *Research in Nursing & Health*. 1995;18(2):179-83.
233. Palinkas LA, Horwitz SM, Green CA, Wisdom JP, et al. Purposeful sampling for qualitative data collection and analysis in mixed method implementation research. *Adm Policy Ment Health*. 2015;42(5):533-44.
234. Green J, Willis K, Hughes E, Small R, et al. Generating best evidence from qualitative research: The role of data analysis. *Australian and New Zealand journal of public health*. 2007;31(6):545-50.
235. Braun V, Clarke V. Using thematic analysis in psychology. *Qualitative Research in Psychology*. 2006;3(2):77-101.
236. Padgett D. Qualitative and mixed methods in public health. Thousand Oaks, California: SAGE Publications, Inc.; 2012.
237. Riessman CK. Narrative analysis. Thousand Oaks, CA, US: Sage Publications, Inc; 1993. vii, 79-vii, p.
238. Bird CM. How i stopped dreading and learned to love transcription. *Qualitative Inquiry*. 2005;11(2):226-48.
239. Davidson C. Transcription: Imperatives for qualitative research. *International Journal of Qualitative Methods*. 2009;8(2):35-52.
240. Maguire MaD, B. . Doing a thematic analysis: A practical, step-by-step guide for learning and teaching scholars. *AISHE-J*. 2017;9(3):p3351-33514.
241. Gibbs GR. Analysing qualitative data: SAGE Publications; 2008.
242. Creswell JW. Research design: Qualitative, quantitative, and mixed methods approaches: SAGE Publications; 2014.
243. Brooks G, Barcikowski RS. The pear method for sample sizes in multiple linear regression. Ohio University 2012.
244. Pedhazur EJ, Schmelkin LP. Measurement, design, and analysis: An integrated approach, student ed. Hillsdale, NJ, US: Lawrence Erlbaum Associates, Inc; 1991. xiv, 819-xiv, p.
245. Green SB. How many subjects does it take to do a regression analysis. *Multivariate behavioral research*. 1991;26(3):499-510.
246. Harris RJ, Harris RJ. A primer of multivariate statistics: Academic Press; 1975.
247. Zebrack B. Information and service needs for young adult cancer patients. *Supportive Care in Cancer*. 2008;16(12):1353-60.
248. Sender A, Friedrich M, Leuteritz K, Nowe E, et al. Unmet supportive care needs in young adult cancer patients: Associations and changes over time. Results from the aya-leipzig study. *J Cancer Surviv*. 2019;13(4):611-9.
249. Fern LA, Taylor RM, Whelan J, Pearce S, et al. The art of age-appropriate care: Reflecting on a conceptual model of the cancer experience for teenagers and young adults. *Cancer Nursing*. 2013;36(5).
250. Planning a comprehensive program in adolescent and young adult oncology—a collision with reality. *Journal of Adolescent and Young Adult Oncology*. 2016;5(4):303-9.
251. Balfe M, Maguire R, Hanly P, Butow P, et al. Distress in long-term head and neck cancer carers: A qualitative study of carers' perspectives. *Journal of Clinical Nursing*. 2016;25(15-16):2317-27.
252. Aziza YDA, Wang S-T, Huang M-C. Unmet supportive care needs and psychological distress among parents of children with cancer in indonesia. *Psycho-Oncology*. 2019;28(1):92-8.
253. Boyes AW, Clinton-McHarg T, Waller AE, Steele A, et al. Prevalence and correlates of the unmet supportive care needs of individuals diagnosed with a haematological malignancy. *Acta Oncologica*. 2015;54(4):507-14.

254. de Boer AG, Verbeek JH, van Dijk FJ. Adult survivors of childhood cancer and unemployment: A metaanalysis. *Cancer*. 2006;107(1):1-11.
255. Keir ST, Swartz JJ, Friedman HS. Stress and long-term survivors of brain cancer. *Supportive Care in Cancer*. 2007;15(12):1423-8.
256. Okediji PT, Salako O, Fatiregun OO. Pattern and predictors of unmet supportive care needs in cancer patients. *Cureus*. 2017;9(5):1234.
257. Husson O, Mols F, van de Poll-Franse LV. The relation between information provision and health-related quality of life, anxiety and depression among cancer survivors: A systematic review. *Annals of oncology : official journal of the European Society for Medical Oncology*. 2011;22(4):761-72.
258. Cramarossa G, Chow E, Zhang L, Bedard G, et al. Predictive factors for overall quality of life in patients with advanced cancer. *Supportive care in cancer : official journal of the Multinational Association of Supportive Care in Cancer*. 2013;21(6):1709-16.
259. National cancer survivorship initiative. Living with and beyond cancer: Taking action to improve outcomes. [internet]. National cancer survivorship initiative, 2020 [cited 09 demember 2020]. Available from: <https://www.Gov.Uk/government/publications/living-with-and-beyond-cancer-taking-action-to-improve-outcomes>.
260. National cancer institute. About cancer survivorship research: Survivorship definitions. [internet]. The national cancer institute, 2018 [cited 19 december 2018]. Available from: <https://cancercontrol.Cancer.Gov/ocs/statistics/definitions.Htm>.
261. Fincham JE. Response rates and responsiveness for surveys, standards, and the journal. *American journal of pharmaceutical education*. 2008;72(2):43.
262. Abraham A, Silber TJ, Lyon M. Psychosocial aspects of chronic illness in adolescence. *The Indian Journal of Pediatrics*. 1999;66(3):447-53.
263. Stensvold E, Stadskleiv K, Myklebust TÅ, Wesenberg F, et al. Unmet rehabilitation needs in 86% of norwegian paediatric embryonal brain tumour survivors. *Acta Paediatrica*. 2020;109(9):1875-86.
264. Ganju RG, Nanda RH, Esiashvili N, Switchenko JM, et al. The effect of transition clinics on knowledge of diagnosis and perception of risk in young adult survivors of childhood cancer. *J Pediatr Hematol Oncol*. 2016;38(3):197-201.
265. Barbour RS. The case for combining qualitative and quantitative approaches in health services research. *Journal of health services research & policy*. 1999;4(1):39-43.
266. Richardson L. New writing practices in qualitative research. 2000;17(1):5.
267. Morse JM, Field PA. *Qualitative research methods for health professionals*: SAGE Publications; 1995.
268. Greene J, Caracelli, V. and Graham, W. Toward a conceptual framework for mixed-method evaluation designs. *Educational Evaluation and Policy Analysis*. 1989;11(3):255-74.
269. Lewin S, Glenton C, Oxman AD. Use of qualitative methods alongside randomised controlled trials of complex healthcare interventions: Methodological study. *BMJ*. 2009;339:3496.
270. O'Cathain A, Murphy E, Nicholl J. Integration and publications as indicators of "yield" from mixed methods studies. *Journal of Mixed Methods Research*. 2007;1(2):147-63.
271. Fetters MD, Curry LA, Creswell JW. Achieving integration in mixed methods designs-principles and practices. *Health services research*. 2013;48(6 Pt 2):2134-56.
272. Bazeley P. *Integrating analyses in mixed methods research*. 55 City Road, London 2018.
273. Johnson RE, Grove AL, Clarke A. Pillar integration process: A joint display technique to integrate data in mixed methods research. *Journal of Mixed Methods Research*. 2019;13(3):301-20.
274. O'Cathain A, Murphy E, Nicholl J. Three techniques for integrating data in mixed methods studies. *BMJ*. 2010;341:4587.
275. Moran-Ellis J, Alexander VD, Cronin A, Dickinson M, et al. Triangulation and integration: Processes, claims and implications. *Qualitative Research*. 2006;6(1):45-59.

276. Zebrack BJ, Gurney JG, Oeffinger K, Whitton J, et al. Psychological outcomes in long-term survivors of childhood brain cancer: A report from the childhood cancer survivor study. *Journal of Clinical Oncology*. 2004;22(6):999-1006.
277. Office for national statistics. Families and households in the uk: 2019. [internet]. Office for national statistics, 2019 [cited 19 January 2021]. Available from: <https://www.Ons.Gov.Uk/peoplepopulationandcommunity/birthsdeathsandmarriages/families/bulletins/familiesandhouseholds/2019>.
278. Mehanna HM, Morton RP. Deterioration in quality-of-life of late (10-year) survivors of head and neck cancer. *Clinical otolaryngology : official journal of ENT-UK ; official journal of Netherlands Society for Oto-Rhino-Laryngology & Cervico-Facial Surgery*. 2006;31(3):204-11.
279. Litzelman K, Kent EE, Mollica M, Rowland JH. How does caregiver well-being relate to perceived quality of care in patients with cancer? Exploring associations and pathways. *J Clin Oncol*. 2016;34(29):3554-61.
280. Julkunen J, Gustavsson-Lilius M, Hietanen P. Anger expression, partner support, and quality of life in cancer patients. *J Psychosom Res*. 2009;66(3):235-44.
281. Northouse LL, Katapodi MC, Schafenacker AM, Weiss D. The impact of caregiving on the psychological well-being of family caregivers and cancer patients. *Seminars in Oncology Nursing*. 2012;28(4):236-45.
282. National institute for health research. Highlight notice: Brain tumours. [internet]. National institute for health research, 2020 [cited 02 December 2020]. Available from: [https://www.Nihr.Ac.Uk/documents/a-call-by-nihr-for-research-on-brain-tumours/11656#funded\\_research](https://www.Nihr.Ac.Uk/documents/a-call-by-nihr-for-research-on-brain-tumours/11656#funded_research) [
283. Gamis AS, Nesbit ME. Neuropsychologic (cognitive) disabilities in long-term survivors of childhood cancer. *Pediatrician*. 1991;18(1):11-9.
284. Ediebah DE, Reijneveld JC, Taphoorn MJB, Coens C, et al. Impact of neurocognitive deficits on patient-proxy agreement regarding health-related quality of life in low-grade glioma patients. *Quality of life research : an international journal of quality of life aspects of treatment, care and rehabilitation*. 2017;26(4):869-80.
285. Kim C, Choi H, Ko H, Park CG. Agreement between parent proxy reports and self-reports of adolescent emotional distress. *The Journal of School Nursing*. 2018;36(2):104-11.
286. Hwang H-F, Chen C-Y, Lin M-R. Patient-proxy agreement on the health-related quality of life one year after traumatic brain injury. *Archives of Physical Medicine and Rehabilitation*. 2017;98(12):2540-7.
287. Soothill K, Morris SM, Harman JC, Francis B, et al. Informal carers of cancer patients: What are their unmet psychosocial needs? *Health & Social Care in the Community*. 2001;9(6):464-75.
288. Sklenarova H, Krümpelmann A, Haun MW, Friederich HC, et al. When do we need to care about the caregiver? Supportive care needs, anxiety, and depression among informal caregivers of patients with cancer and cancer survivors. *Cancer*. 2015;121(9):1513-9.
289. Lucas MS, Barakat LP, Jones NL, Ulrich CM, et al. Expectations for function and independence by childhood brain tumors survivors and their mothers. *Narrative Inquiry in Bioethics*. 2014;4(3):233-51.
290. Robinson JE, Huskey D, Schwartz J, Weaver MS. The many roles of the rock: A qualitative inquiry into the roles and responsibilities of fathers of children with brain tumors. *Children (Basel, Switzerland)*. 2019;6(10).
291. Jones BL, Pelletier W, Decker C, Barczyk A, et al. Fathers of children with cancer: A descriptive synthesis of the literature. *Social Work in Health Care*. 2010;49(5):458-93.
292. Fish J. Independent living scales®. In: Kreutzer JS, DeLuca J, Caplan B, editors. *Encyclopedia of clinical neuropsychology*. New York, NY: Springer New York; 2011. p. 1305-

# Appendices

## Appendix 1 – Systematic review search strategy

### Database: MEDLINE (Ovid)

Search covers: 1946- present

- 
- 1 exp brain neoplasm/
  - 2 exp glioma/
  - 3 ((brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or intracranial) adj4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)).ab,ti.
  - 4 (glioma\* or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\* or neurocytoma\* or pinealoma\* or schwannoma\* or craniopharyngioma\* or PNET or DNET).ab,ti.
  - 5 1 or 2 or 3 or 4
  - 6 exp infant/
  - 7 exp child/
  - 8 (newborn\* or "new born\*" or baby or babies or infact\* or infanc\* or child\* or p?ediat\*).ab,ti.
  - 9 6 or 7 or 8
  - 10 5 and 9
  - 11 exp Survivors/
  - 12 exp "Adult Survivors of Child Adverse Events"/
  - 13 "surviv".ti.
  - 14 11 or 12 or 13
  - 15 10 and 14

### Database: Embase (Ovid)

Search covers: 1996- present

1. exp brain tumour/
2. exp glioma/
3. ((brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or intracranial) adj4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)).ab,ti.
4. (glioma\* or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\* or neurocytoma\* or pinealoma\* or schwannoma\* or craniopharyngioma\* or PNET or DNET).ab,ti.
5. 1 or 2 or 3 or 4
6. exp child/
7. (newborn\* or "new born\*" or baby or babies or infact\* or infanc\* or child\* or p?ediat\*).ab,ti.
8. 6 or 7
9. 5 and 8
10. exp survivor/
11. "surviv".ti.
12. 10 or 11
13. 9 and 12

### Database: PsycINFO (Ovid)

Search covers: 1806- present

1. exp brain neoplasms/
2. exp glioma/
3. ((brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or intracranial) adj4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)).ab,ti.



4. (glioma\* or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\* or neurocytoma\* or pinealoma\* or schwannoma\* or craniopharyngioma\* or PNET or DNET).ab,ti.
5. 1 or 2 or 3 or 4
6. (newborn\* or "new born\*" or baby or babies or infact\* or infanc\* or child\* or p?ediat\*).ab,ti.
7. 5 and 6
8. exp survivors/
9. "surviv\*".ti.
10. 8 or 9
11. 7 and 10

**Database: Web of Science**

**Search covers: 1900- present**

**# 1 TOPIC:** (((Brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or intracranial) near/4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)) OR ((glioma\* or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\* or neurocytoma\* or pinealoma\* or schwannoma\* or craniopharyngioma\* or PNET or DNET))

**# 2 TOPIC:** (newborn\* or "new born\*" or baby or babies or infant\* or infanc\* or child or p?ediatr\*)

**# 3 TITLE:** (surviv\*)

**# 4 #1 AND #2 AND #3**

**Database: PUBMED**

**Search covers: 1996- present**

**#12 Search** (#9 AND #10 AND #11)

**#11 Search** (survivors[MeSH Terms]) OR surviv\*[Title]

**#10 Search** (newborn\*[Title/Abstract] OR "new born\*" [Title/Abstract] OR baby[Title/Abstract] OR babies[Title/Abstract] OR infact\*[Title/Abstract] OR infanc\*[Title/Abstract] OR child\*[Title/Abstract] OR paediat\*[Title/Abstract])

**#9 Search** (((brain neoplasm[MeSH Terms]) OR glioma[MeSH Terms]) OR (((brain\*[Title/Abstract] OR cerebr\*[Title/Abstract] OR cerebella\*[Title/Abstract] OR infratentorial[Title/Abstract] OR supratentorial[Title/Abstract] OR "choroid plexus"[Title/Abstract] OR intracranial)[Title/Abstract] AND (tumo?r\*[Title/Abstract] OR neoplas\*[Title/Abstract] OR cancer\*[Title/Abstract] OR oncol\*[Title/Abstract] OR metasta\*[Title/Abstract] OR malignan\*[Title/Abstract]))) OR glioma\*[Title/Abstract] OR astrocytoma\*[Title/Abstract] OR glioblastoma\*[Title/Abstract] OR ependymoma\*[Title/Abstract] OR medulloblastoma\*[Title/Abstract] OR neurocytoma\*[Title/Abstract] OR pinealoma\*[Title/Abstract] OR schwannoma\*[Title/Abstract] OR craniopharyngioma\*[Title/Abstract] OR PNET[Title/Abstract] OR DNET[Title/Abstract])

**Database: CINAHL (Ebsco)**

**Search covers: 1960- present**

#S13 S9 AND S12  
#S12 S10 OR S11  
#S11 TI surviv\*  
#S10 MH survivors OR MH cancer survivors  
#S9 S5 AND S8  
#S8 S6 OR S7  
#S7 TI ( (newborn\* or "new born" or baby or babies or infact\* or child\* or p?ediatr\*) ) OR  
AB ( (newborn\* or "new born" or baby or babies or infact\* or infanc\* or child\* or p?ediat\*) )  
#S6 MH child\*  
#S5 S1 OR S2 OR S3 OR S4  
#S4 TI ( (glioma or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\*  
or neurocytoma\* or pinealoma\* or schwannoma\* or glioblastoma\* or ependymoma\* or  
medulloblastoma\* or neurocytoma\* or pinealoma or schwannoma+ or craniopharyngioma\* or PNET  
or DNET) )  
#S3 TI ( ((brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid  
plexus" or intracranial) N4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)) ) OR  
AB ( ((brain\* or cerbr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or intracranial)  
N4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*)) )  
#S2 MH glioma  
#S1 MH brain neoplasms+

**Database: Cochrane Library (Wiley)**

**Search covers: 2005- present**

#1 MeSH descriptor: [Brain Neoplasms] explode all trees  
#2 MeSH descriptor: [Glioma] explode all trees  
#3 (brain\* or cerebr\* or cerebella\* or infratentorial or supratentorial or "choroid plexus" or  
intracranial) near/4 (tumo?r\* or neoplas\* or cancer\* or oncol\* or metasta\* or malignan\*):ti,ab,kw  
(Word variations have been searched)  
#4 glioma\* or astrocytoma\* or glioblastoma\* or ependymoma\* or medulloblastoma\* or  
neurocytoma\* or pinealoma\* or schwannoma\* or craniopharyngioma\* or PNET or DNET:ti,ab,kw  
(Word variations have been searched)  
#5 #1 or #2 or #3 or #4  
#6 MeSH descriptor: [Infant] explode all trees  
#7 MeSH descriptor: [Child] explode all trees  
#8 newborn\* or "new born\*" or baby or babies or infact\* or infanc\* or child\* or p?ediat\*:ti,ab,kw  
(Word variations have been searched)  
#9 #6 or #7 or #8  
#10 MeSH descriptor: [Survivors] explode all trees  
#11 Surviv\*:ti (Word variations have been searched)  
#12 #10 or #11  
#13 #9 and #12

## Appendix 2 - Study details

ADL=Activities of Daily Living, ALL= Acute Lymphoblastic Leukaemia, TYA =Teenage and Young Adult, BT = Brain tumour, BMD = Bone Mineral Density, BTS = Brain tumour survivor, EF= Executive functioning, HRQOL= Health related quality of life, M= Mean, MD = Medulloblastoma, QOL= Quality of life.

Articles belonging to the same study are marked with either \*, \*\*, \*\*\*.

Author	Year	Country	Brain tumour participants	Care givers	Controls	Methods	Aims/Measures	Summary of findings	MMAT Score (%)
Ahomäki et al.	2017	Finland	1300 cancer survivors (324 BTS) Age: 18 Diagnosed :0-16		7209 population controls	Military service testing	Physical and cognitive performance	BTS were the second highest cancer group to be exempt from military service. TYA/BTS in service scored poorly on fitness and cognitive testing, which was significantly worse than controls.	75
Allion et al.	2016	USA	25 BTS Age: 18-35 (M=24) Diagnosed :1-19 (M=9.3)		25 neuro-typical matched controls	Questionnaires; Brain magnetic resonance images	Measure cerebellar atrophy to determine its neurobehavioral correlates	80% of BTS had some degree of diffuse cerebellar damage; participants experienced on average 15% cerebellar atrophy. Young age at diagnosis and radiation was associated with cerebellar atrophy, which impacts both written and oral processing speeds.	75
Ait Khelifa-Gallois et al.	2015	France	64 astrocytoma survivors (48 aged: 18-30) Diagnosed :0.4-13.4			Telephone semi-structured interviews	Long term outcomes	Around half of survivors reported long-term difficulties associated with cognitive and physical sequelae. Many had received support including - Remedial teaching, Speech therapy, occupational therapy, physiotherapy and psychomotor therapy.	100
Armuan et al.	2017	Sweden	1206 cancer survivors (225 BTS) Age: 18-39 Diagnosed :<21		2412 population controls	Observational- patient databases	Reproductive Patterns	Both males and females in the BT group were less likely to have had a first born child in young adulthood compared to other cancer diagnosis.	100
Barakat et al.*	2015	USA	126 BTS Age: 14-39 (M=20.5) Diagnosed :<9	186 mothers		Questionnaires	Predictors of physical and emotional HRQOL, by evaluating the mediating role of family functioning	TYA CBTS are at risk for poor HRQOL, many do not live fully independently, involvement of their family, and consequently their family's functioning, is likely critical to improving HRQOL.	75
Boman et al.	2009	Sweden	531 BTS Age: 18-36 Diagnosed :<19 (M=10.4)		996 population controls	Postal Questionnaires	Health-related and long term outcomes	TYA CBTS are at high risk for significant persistent functional, and health-related late effects, with female survivors being more vulnerable. Survivors	100

							were significantly more likely to have lower employment/educational status, use governmental subsidies and less likely to become parents.	
<b>Boydell et al.</b>	2008	USA	14 BTS Age: 17-29	22 family members.	Focus groups; Semi-structured Interviews	Experiences and needs	TYA Survivors carry physical and emotional issues resulting from their tumour and treatment well into their years of emerging adulthood. Strategies to support these young people in ways that will cultivate and harness their strengths and determination need to be developed.	75
<b>Brinkman et al.</b>	2012	USA	20 MD Age: 21-36 (M=29) Diagnosed :2-17		Pilot study, Neurocognitive and EF assessments.	Associations between cognitive processes and white matter	Reduced white matter integrity, was associated with poorer performance on tasks of executive function and observed neurocognitive dysfunction. Neurocognitive impairment was common across many specific domains of TYA function; the worst scores being impaired visual memory and motor processing.	75
<b>Chen et al.</b>	2008	Taiwan	7 BTS (6 aged 17-22 Diagnosed :<18		Semi structured interviews	Experiences from a sociocultural perspective.	Neurological, physical and psychosocial issues are a part of TYA survivorship. Numerous physical symptoms included- diabetes, sex hormone deficiency, and fatigue. Prominent psychosocial issues included isolation, relationship rejection.	75
<b>Chou et al.</b>	2009	Taiwan	98 cancer survivors (49 BTS) Age: 18-21 (M=20.0) Diagnosed :<16		Pilot study, Questionnaires; Interviews; Focus groups.	QOL, long term outcomes (i.e. cognitive, social issues)	TYA BTS had poorer QOL amongst other issues (i.e. physical, psychological, social and cognitive) compared to ALL survivors. This is attributed to BTS survivors having less positive protective factors, more illness-related and individual risk factors and less positive resilience scores.	75
<b>Cohen et al.</b>	2012	USA	36 BTS Age: 14-20 (M=16.9) Diagnosed :M=8.4		Observational; Medical assessments.	Bone mineral density (BMD)	TYA BTS had varied low-normal BMD. Lumbar spine BMD was significantly lower in those closer to diagnosis, suggesting that peri-treatment factors (illness, decreased activity) affect BMD and there may be some recovery over time.	75
<b>D'Agostino et al.</b>	2013	Canada	22 cancer survivors (7 BTS) Age: 18-35 Diagnosed :<18		Focus groups	Challenges and resource needs	The impact of cancer seemed more complicated in TYA BTS. Survivors struggled with neurological sequelae which had psychosocial and practical implications (i.e. social relationships, and educational/vocational options). Individual programming to meet the	100

							unique challenges identified by BTS are required.	
<b>de Blank et al.</b>	2016	USA	587 BTS (255 aged 30-39) Diagnosed : <21		Questionnaires	Psychological and social outcomes of astroglial tumours with and without vision loss	The majority of BTS aged 30-39 group had no impairment. Yet made up 41% of total bilaterally blind group. Survivors with bilateral blindness were significantly more likely to be unmarried, live dependently, and be unemployed in comparison with those without vision impairment.	75
<b>Deatrick et al.*</b>	2014	USA	126 BTS Age: 14-39 (M=20.5)	186 mothers	Telephone questionnaires	Caregiver competence in association with caregiver/survivor health, and family functioning	Caregiver health directly predicted caregiving demands which related to family functioning and finally to caregiver competence. Caregiver health becomes important to how they evaluate the demands of caregiving, but may also limit their ability to access resources (their energy/motivation for the effort for caregiving).	75
<b>Deatrick et al.</b>	2009	USA	BTS Age: 14-30 (M=19.3)	22 parents (17 mothers, 5 fathers)	Interviews; Questionnaires	Family management in families of children with chronic conditions and those who survived a BT vs	Most BT parents reported that their child had medical effects, including cognitive, physical, and social issues. Parents voiced concern about reoccurrence and the ability to balance family life with survivor needs. Parents generally had the same interpretations of family management as parents of chronic conditions. Yet they described fears of loss and the future. Tools are not available to assess family functioning and clinicians find it challenging to tailor interventions to family needs.	75
<b>Demers et al.</b>	2016	Canada	36 BTS Age: 16-29 (M=21) Diagnosed : M=8.9	Compared with age norms	Questionnaires; ADL task performance	Activities of Daily Living (ADL) performance and its association with HRQOL.	ADL functioning in TYA/BTS is significantly lower than norms but HRQOL is similar to the general population. The level of poor performance in ADL skills (motor and process skills) was significantly associated with poorer physical health and with mental health.	50
<b>Frange et al.</b>	2009	France	45 MD Age: 15-39 (M=25.2) Diagnosed : 1.4-17 (median=8.8).		Review of medical records; Questionnaires	Long term outcomes	Only a minority of TYA participants were free of sequelae. Most MD survivors suffer persistent deficits in several domains, with a significant impact on their psychosocial functioning. The findings reinforce the importance of early intervention programmes to reduce the psychosocial impacts of their disease.	100

<b>Gray et al.</b>	1992	Canada	62 cancer survivors (16 BTS) Age: 18-37 (M=26.3) Diagnosed :<18	51 healthy peers	Screening questionnaires; Semi-structured telephone interviews	Long term outcomes and experiences	BTS were more likely than other TYA cancer survivors to report difficulty with a variety of physical symptoms - significantly more memory problems, tiredness, coughing and ringing in the ears. However, there was no statistical significance between BTS survivors scoring worse than other cancer survivors in relation to other symptoms, such as pain, difficulties concentrating and shortness of breath.	25
<b>Gunn et al.</b>	2016	Finland	21 BTS Age: 14-35 (median=24) Diagnosed :<16	327 population controls	Semi-structured interviews; Questionnaires	QOL and Long term outcomes	BTS had significantly worse scores in the following dimensions: mobility, vision, hearing, eating, speech, usual activities, mental function, and sexual activity. Scores were lower among MD survivors, but not statistically significant. Still, the majority of the survivors described their QOL positively.	75
<b>Heikens et al.</b>	1998	Holland	20 MD Age: 19-33 (median 25) Diagnosed :4-17 (M=8)		Endocrine evaluation	Long term endocrine sequelae of cranial irradiation	In 75% of TYAs endocrine abnormalities were observed. The main deficiency was GH impairment, with lack of energy as the major complaint. Young age at treatment was a significant determinant of GH deficiency. Post-treatment interval or chemotherapy were not determinants of endocrine impairment.	75
<b>Hobbie et al.*</b>	2016	USA	41 BTS Age: 15-36 (M=23) Diagnosed :3-14		Semi-structured interviews	HRQOL and long term outcomes	Physical health issues and cognitive impairment were embedded in TYA accounts of their daily life. The majority said that their social functioning was most impacted by their BT and treatment. Loneliness was a prevailing theme for all of the survivors. Those with the most neurocognitive impairments had limited insight as to why they did not have friends or why they could not achieve their goals.	75
<b>Hocking et al.*</b>	2015	USA	34 BTS Age: 18 to 30 (M=23.5) Diagnosed :M=7.4	34 mothers	Questionnaires; neurocognitive assessments	Association between survivor neurocognitive functioning, family functioning, and HRQOL.	Poorer survivor neurocognitive functioning was associated with worse family functioning and poorer survivor HRQOL.	75
<b>Hoffman et al.</b>	2015	Germany	19 Craniopharyngioma		Medical assessments	Nonalcoholic fatty liver	NAFLD occurred in over 50% of TYA CP survivors. NAFLD should be planned for and managed as a major adverse	100

			(CP) survivors Aged: 16-30 Diagnosed 2-20			disease (NAFLD)	late effect in follow-up care of CP patients.	
<b>Jayakar et al.</b>	2015	USA	35 BTS Age: 17-36 (M=24) Diagnosed :1-17 (M=8.17)	59 population controls	Questionnaires; Brain imaging data	Verbal memory and hippocampal volumes	TYA BTS exhibited smaller hippocampi compared with controls. Early declines in hippocampal volume are not fully compensated for in later life survivors grow to become young adults, as evidenced by volumes remaining lower than controls. Indices of verbal memory were significantly lower for survivors.	50
<b>Jereb et al.</b>	1994	Slovenia	42 cancer survivors (23 BTS) Age: 15-26 Diagnosed : <21 (M=8)	19 ALL	Physical, endocrine and psychological examinations; Interviews	Physical and psychosocial long term outcomes	Hormonal, emotional and physical deficiencies were more frequent in BTS than ALL. The degree of mental deterioration was lower for ALL survivors compared to BTS. The majority of BTS endocrinologically evaluated had hormonal deficiencies (more frequent than ALL).	50
<b>Johanns dottir et al.</b>	2012	Norway	398 cancer survivors (88 Astrocytoma) Age: 19-34 Diagnosed :1-18	763 population controls	Postal questionnaire	Fatigue	Chronic fatigue was found in 13.6% of astrocytoma survivors, higher than general population controls (5.8%). There was no significant difference in fatigue levels when comparing the three diagnostic groups (Astrocytoma, Wilms Tumour and Acute Myeloid Leukaemia)	100
<b>King et al.</b>	2016	USA	68 BTS Age: 18-35 (M=24) Diagnosed :0-17 (M=7.96)		Intellectual evaluations	Long-term intellectual and adaptive outcomes	TYA BTS on average performed similar to peers in intellectual and adaptive outcomes but a wide range of outcomes were evidenced. 17.6% were impaired on IQ scores and 29% of the sample were identified as impaired in adaptive functioning and day-to-day living skills.	50
<b>King I et al.</b>	2015	USA	17 BTS Age: 17-35 years (M=23.2) Diagnosed :1-17 (Median = 7)	17 demographically matched controls	Working memory, behavioural and neuropsychological measures	Working memory	TYA BTS evidenced lower working memory performance than controls. On all of measures survivors performed significantly lower than controls. High-grade embryonal tumour survivors performed the worst	75
<b>King II et al.</b>	2015	USA	31 BTS Age: 18-30 (M=22.5). Diagnosed :1-19 (M=8.8)	33 demographically matched controls.	Questionnaires and assessments of EF.	Adaptive function and executive performance	Survivors had significantly lower planning and functional community living skills and greater perseveration. The range of scores was varied which suggests a wide range of functioning amongst BTS.	75
<b>King III et al.</b>	2015	USA	27 BTS Age: 18-32 (M=22.7)	27 demographically	Questionnaires;	White matter integrity	Lower long-term intellectual outcomes of BTS are associated with lower white	75

			Diagnosis age: M=9	matched controls.	MRI and DTI (diffusion tensor imaging) data; IQ test		matter integrity. Radiation and adjunct chemotherapy treatment may play a role in greater white matter disruption. Those treated with radiotherapy were significantly different from non-radiotherapy and controls on all indices of intellectual abilities. The white matter disruption of the radiation with or without chemotherapy was positively correlated with IQ and cumulative neurological factors.	
<b>Klosky et al.</b>	2014	USA	307 cancer survivors <b>(40 BTS)</b> Age: 15-20 Diagnosed : <4		Postal questionnaire	Sexual functioning	TYAs with a history of BTS were significantly less likely to report a history of sexual intercourse.	100
<b>Kousteni et al.***</b>	2013	Germany	203 BTS Age: 19-37 (median=22) Diagnosed : <15		Questionnaire; Radiation exposure assessment	Fertility	Irradiation of the pituitary gland >30 gray in TYA BTS was significantly associated with less pregnancies, increased infertility and absence of menstruation.	75
<b>Lehman et al.**</b>	2017	USA	144 cancer survivors <b>(47 BTS)</b> Age: 20-39 (M=28) Diagnosed : 5-18	144 population controls	Online survey	Psychosexual functioning	TYA BTS were less likely to be sexually experienced, partnered, and achieve milestones of psychosexual development compared with those treated with non-neurotoxic modalities. Survivors treated with high-dose neurotoxic modalities were least likely to be sexually experienced, in a relationship, or have children.	75
<b>Lehman et al.**</b>	2017	USA	149 cancer survivors 52 BTS Age: 20-39 Diagnosed : 5-18	149 population controls	Online survey	Romantic relationships - mate value	Comparisons between diagnostic groups indicated that TYA BTS reported significantly lower mate value than lymphoma as well as leukaemia survivors.	75
<b>Liptak et al.</b>	2016	USA	19 BTS Age: 14-26 (M=19.8) Diagnosed : 0.6-19 (M=7.7)	18 mothers	Semi-structured telephone interviews; Questionnaires	Social issues, psychosocial needs and evaluation of a developed social intervention.	Social isolation was prominent among survivors. The combination of lack of social opportunity and medical, neurocognitive, and psychological issues added to the social isolation. Survivors and caregivers described participation in the STEPS programme as a positive experience which reduced isolation and reportedly contributed to the improvement in social confidence.	75
<b>Lucas et al.*</b>	2016	USA	BTS Age: 15-36 (M=23.38)	45 mothers	Semi-structured interviews	Caregivers' expectations about survivors'	Five main themes emerged as integral to mother expectations: realizing a difference in the survivor, noticing limitations to independence in the survivor,	75



						functioning	memories of learning about clinical prognoses as understood from consent meetings and education, managing these realizations, and acknowledging unresolved challenges. Pre-tumour expectations were a basis for post-tumour uncertainty and worry about the potential that the survivor is too different in important ways and that s/he may not become independent of her.	
<b>Maddrey et al.</b>	2005	USA	16 MD Age: 14-28 Diagnosed : <16 years.	General population norms	Neuropsychological tests; Questionnaires	Cognitive performance, psychosocial functioning and QOL	Impairment existed in all cognitive domains (attention, memory, visuospatial ability, motor functioning, language and EF). Survivors 19+ years old were significantly impaired in all indicators of daily functioning (i.e. employment, education, driving) when compared to population norms. Despite impairment no significant differences were noted (compared to norms) in QOL.	50
<b>Mandrell et al.</b>	2012	USA	31 BTS (17 aged 14-25) Diagnosed : <14		Sleep evaluations.	Sleep disorders	The most common reason for sleep treatment in TYAs was excessive daytime sleepiness, snoring sleep apnea. BTS not only interferes with normal sleep patterns and increases daytime sleepiness, but also may further impair cognitive, physical and social functioning.	75
<b>Maunsel et al.</b>	2006	Canada	1334 cancer survivors <b>(238 BTS)</b> Age: 15-37 Diagnosed : <20	1477 age- and sex-matched controls.	Postal questionnaires	QOL	TYA BTS were the only group to have consistently poorer physical and psychosocial functioning. Compared with controls BTS had significantly poorer QOL in several domains: general health, physical function, and role limitations.	100
<b>Maurice-Stam et al.</b>	2013	The Netherlands	363 cancer survivors <b>(38 BTS)</b> Age: 18-31 Diagnosed : <16	508 population controls	Questionnaires	Disability benefits and psychosocial development	BTS made up 40.4% of cancer survivors with disability benefits. Cancer survivors with disability benefits had lower social and psychosexual scores than those without disability benefits.	75
<b>McClennan et al.</b>	2013	USA	271 cancer survivors <b>(51 BTS)</b> Age: 18-38 Diagnosed : M=10.24		Questionnaires	Long term outcomes, experiences, and information needs	BTS in comparison to other TYA cancer survivors reported significantly worse cognitive late effects and financial difficulties. BTS did not experience significantly greater physical side effects, but they did report greater amounts of fatigue than other survivors. There is a need for development of educational materials appropriate for	50

							survivors and providers desiring more information.	
<b>McCurdy et al.</b>	2016	USA	34 BTS Age: 18 to 30 (M=23.5) Diagnosed: M=7	34 Mothers	Medical record review; Survivor performance-based assessments; Questionnaires	Survivor, mother, and performance-based estimates of executive functioning (EF)	Findings suggest that TYA BTS who received high intensity tumour-directed treatments may overestimate executive skills relative to mother reports and performance on objective measures. Both survivors and mothers reported greater executive dysfunction than the normative mean, though were both in the average range. Survivors with more intensive treatment evidenced greater score discrepancies, reporting less executive dysfunction relative to mother-reported.	75
<b>Palma et al.*</b>	2015	USA	BTS: Aged: 15–30 (M=23)	46 mothers	Semi-structured interviews	Caregiver demands/experiences	Four main categories of maternal daily caregiving demands were identified: managing the illness; identifying, accessing, and coordinating resources; assisting with everyday responsibilities; and fostering psychosocial health.	75
<b>Petraroli et al.</b>	2007	Italy	12 BTS (9 aged 14–25) Diagnosed: 5–15		Observational; Medical assessments and tests.	Bone mass bone mineral density (BMD); Endocrine functioning	There was reduced BMD in all the TYA BTS. Most of the survivors had a BMD that was lower than normal in both the lumbar column and in the femoral neck. The main risk factor for bone mass loss in the sample was hypogonadism but also multiple hormonal deficiencies are associated with lower BMD values.	50
<b>Pfizer et al.***</b>	2013	Germany	203 BTS Age: 19–37 (median=22) Diagnosed: 1–15 (M=11)		Questionnaires	Education	A third of CBTS obtained the highest school leaving certificate. Tumour irradiation, and young age at BT diagnosis may reduce chances for BTS better educational level. Patients without chemotherapy had highest education level.	75
<b>Pietila et al.</b>	2017	Finland	52 BTS (16 >18 group) Age: 18–28.7 Diagnosed: 0.1–15.5		Medical record review; Physical examination; Interviews.	Growth, pubertal development and gonadal function	Growth impairment, growth hormone deficiency and hypogonadism were common in childhood BTS. However, young adults aged 18–28 did not have statistically significant lower height standard deviation scores at follow-up.	100
<b>Poggi et al.</b>	2005	Italy	76 BTS (21 aged 14–18) Diagnosed: <18		Cognitive and psychological evaluations	Cognitive and psychological behavioural disorders	All survivors were cognitively impaired, those aged 14–18 scored worse than younger groups. BTS were impaired in communication, daily living skills, social skills and motor skills. The most impaired domain was 'socialisation' and competence decreased as	100

							participants got older. Most frequent psychological and behavioural disorders were: internalising; withdrawn; social problems.	
<b>Strauser et al.</b>	2013	USA	43 BTS Age: 18-30 (M=22) Diagnosed :M=9.5	298 college students	Questionnaires	Career readiness and work personality	TYA BTS have significantly lower levels of work personality and career readiness when compared to controls. Individuals who are diagnosed age 6-12 may be at increased risk of developing lower levels of work personality which may lead to increased problems meeting the contextual demands of the work environment.	75
<b>Taiwo et al.</b>	2017	USA	61 BTS Age: 18-35 (M=24) Diagnosed :0-17 (M=8)		Neuropsychological assessments; Medical record review	Neurocognitive outcomes	Findings suggest that survivors with more treatments and neurological sequelae experience greater working memory, processing speed, and attention deficits.	75
<b>Turkelt et al.</b>	2007	USA	600 BTS (8 aged 14-20) Diagnosed :6-13		Observational; Medical record review	Psychosis late effects	Psychotic symptoms were seen in BTS, but only in a small number. A delay of several years was seen between the time of diagnosis/treatment and onset of psychotic symptoms.	75
<b>Uday et al.</b>	2015	England	35 MD Age: 16-35 Diagnosed :2-14 (M=8)		Longitudinal observational study	Endocrine functioning	Endocrine sequelae in TYA MD survivors was high. 97% of survivors developed either complete or partial GHD following treatment. Faltering height and tiredness were the most common presentation of GHD. Endocrine issues were more prevalent in those treated with concomitant chemotherapy and radiotherapy.	75
<b>Vance et al.</b>	2004	England	BTS Aged: 15-20 Diagnosed :5-9	4 Parents (with survivors in age limit)	Semi-structured interviews	Long term outcomes, experiences	Survivors reported issues with education, cognitive functioning, physical effects, social functioning, relationships, isolation and independence. As TYAs get older parents were concerned about their child's future- especially their inability to self-care. Parents need more support to help their child adjust to their late-effects.	75
<b>Wasilewski-Masker et al.</b>	2016	USA	157 cancer survivors (22 BTS) Age: 15-30 Diagnosed :<20 years		Online questionnaires	Survivors' experiences of supportive services	BTS were more likely to value social activities or support groups compared with other survivors, and in comparison to informational mailing, weekend retreat, informational workshop or individual counselling. BTS considered social activities and support groups most important, this could maybe indicative of	75

							poorer social outcomes observed among BTS.	
<b>Wilford et al.</b>	2017	USA	BTS Age: M=44 BTS- Age: M=15.7 Diagnosed : M=4.5	20 parents	Semi-structured telephone interviews	Long term outcomes	Parents reported TYA survivors having social withdrawal and impaired peer/romantic relationships. Many parents reported impaired cognitive functioning and ongoing physical issues affecting social functioning. Parents worried about the future and reported chronic stress regarding the survivor's current and future health. Most commonly desired support services were parent support groups, parent survivorship education classes, and an age-matched, ability-matched social support group for BTS.	75
<b>Zebrack et al.</b>	2008	USA	176 cancer survivors <b>(19 BTS)</b> Age: 16-28 Diagnosed : <18		Medical record review; Questionnaire	QOL	BTS scored significantly lower on their overall QOL. BTS also scored lower in the social QOL dimension than all other survivors combined. BTS reported diminished wellbeing with regard to social activities, social relationships, and employment.	75
<b>Zebrack et al.</b>	2010	USA	599 cancer survivors <b>(79 BTS)</b> Age: 18-39 Diagnosed : <21		Postal questionnaire	Sexual functioning	Findings suggest that BTS (especially males) are less likely than other paediatric malignancies to be sexually active. However, no statistically significant differences in sexual functioning were observed across cancer types.	75

## Appendix 3 – Ethical approval letter



Dr Florian Boele  
Leeds Institute of Cancer and Pathology  
Patient Centred Outcomes Group  
Level 06, Bexley Wing  
LS9 7TF



Email: [hra\\_approval@nhs.net](mailto:hra_approval@nhs.net)  
[Research-permissions@wales.nhs.uk](mailto:Research-permissions@wales.nhs.uk)

26 September 2018

Dear Dr Boele

**HRA and Health and Care  
Research Wales (HCRW)  
Approval Letter**

<b>Study title:</b>	<b>Supportive care needs of teenage and young adult childhood brain tumour survivors and their caregivers.</b>
<b>IRAS project ID:</b>	<b>245810</b>
<b>Protocol number:</b>	<b>N/A</b>
<b>REC reference:</b>	<b>18/YH/0312</b>
<b>Sponsor</b>	<b>University of Leeds</b>

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.

**How should I continue to work with participating NHS organisations in England and Wales?**

You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of assessment*" section towards the end of this letter.

You should provide, if you have not already done so, detailed instructions to each organisation as to how you will notify them that research activities may commence at site following their confirmation of capacity and capability (e.g. provision by you of a 'green light' email, formal notification following a site initiation visit, activities may commence immediately following confirmation by participating organisation, etc.).

## Appendix 4 – Ethical approval (substantial amendment): online recruitment



# Health Research Authority

## Yorkshire & The Humber - Bradford Leeds Research Ethics Committee

NHSBT Newcastle Blood Donor Centre  
Holland Drive  
Newcastle upon Tyne  
NE2 4NQ

Tel: 0207 104 8018

10 June 2019

Ms Emma Nicklin  
Leeds Institute of Cancer and Pathology  
Patient Centred Outcomes Group  
Level 06, Bexley Wing  
LS9 7TF

Dear Ms Nicklin

**Study title:** Supportive care needs of teenage and young adult childhood brain tumour survivors and their caregivers.  
**REC reference:** 18/YH/0312  
**Protocol number:** N/A  
**Amendment number:** Substantial Amendment 1, 02/04/2019  
**Amendment date:** 13 May 2019  
**IRAS project ID:** 245810

The above amendment was reviewed by the Sub-Committee in correspondence.

### Summary of Amendment

This amendment was to advertise the study questionnaire online and therefore increasing the sample size.

### Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

### Approved documents

The documents reviewed and approved at the meeting were:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Notice of Substantial Amendment (non-CTIMP) [Notice of Substantial Amendment ]	Substantial Amendment 1, 02/04/2019	13 May 2019
Other [Caregiver Online Questionnaire Participant Information]	1	02 April 2019
Other [What is life like for teenagers/young adults and their caregivers after a childhood brain tumour?]	1	02 April 2019

## Appendix 5 - Survivor Participant Information Sheet (over 16's)



### Support Needs of Young Brain Tumour Survivors and their Caregivers

We would like to invite you and your caregiver to take part in a research study. The study is a PhD project funded by a charity called Ellie's Fund and Yorkshire Cancer Research. Before you decide whether you would like to take part, it is important for you to understand why the study is being done and what it will involve.

Please:

- ❖ **Read** this leaflet carefully. You can talk about it with your family, friends, or us.
- ❖ **Ask us** if there is anything you don't understand or if you want more information.
- ❖ **Take time** to decide whether or not you want to join in.

#### Why are we doing the study?

At the moment there is not much information about how having a brain tumour in childhood can affect survivors and their caregivers when they become young adults. With this research, we want to understand more about the issues and support needs experienced by young survivors and their caregivers.

We're inviting you to take part as you had a brain tumour as a child and are now a young adult. We believe that your experiences will be very different from healthy children or older brain tumour survivors. We hope that by collecting information from you and others like you that we can understand more about surviving a brain tumour.

We are also asking your caregiver to take part in the study. When we say caregiver we mean someone who has shared the experience of a brain tumour with you, and may now help you now with your daily care or practical/emotional support. It is OK if you both don't want to take part in the study, one of you can take part without the other.

#### Why this research is important?

More information in this area could help us to see how we can better support survivors (like you) and their caregivers.

#### What happens if I agree to take part?

If you decide to take part in this research study, a member of the research team will answer any questions you have and ask you to sign a consent form. You will then be asked to complete a questionnaire either online or on paper – the choice is yours. We can also help you fill in the questionnaire over the phone if you feel this would be helpful. Please let us know if you would like to do this.

If you chose to do the questionnaire online you will be given full instructions by the research team of how to login and access the questionnaire at home. Or if you would rather complete the questionnaire on paper, you will be given a pre-paid envelope to return the questionnaire by post. If we see after

one week that you have not completed the questionnaire after agreeing to take part, we will send you a reminder by text, email or letter (only if you agree).

The questionnaire should take around 30 minutes to complete. The questions you will be asked in the questionnaire will include things like:

*“After your brain tumour do you have issues with headaches?”*

*“In the last month what was your need for help with work around the home?”*

*“In the last month what was your need for help with anxiety?”*

If you do not know the answer to any questions or you feel they are not relevant to you, that's OK, you can skip these questions.

We also would like to ask your permission for the research team to look at your hospital records to see your brain tumour history – e.g. when you were diagnosed, the location of the tumour and what treatment you had. The information will only be seen by the research team and will be kept anonymised and confidential.

There is a chance that after you have completed the questionnaire that we may ask if you could like to take part in an interview with the researcher. If you are happy to be contacted about an interview at a later date and given more information then please agree to this on the consent form. Not everyone will be asked to partake in an interview.

#### **Will anybody know I am taking part?**

Your personal details (age, name etc.) will be kept confidential. It is very important to us to keep your information safe. The answers you provide in the questionnaire will only be seen by the research team. All your data will be stored securely on a University of Leeds electronic database and/or stored in a locked file cabinet within the Leeds Teaching Hospitals NHS Trust and will only be accessible by the research teams. When we present any findings of our study we will not name or identify you in any way. The only time we might need to share information about you with others would be if the researchers have serious concern about your health, safety or well-being. If this happens the researchers may inform an appropriate professional. We would make every effort to explain to you why we need to share this information before doing so.

#### **Do I have to take part?**

No. It is up to you to decide whether or not to take part. Also, a decision not to take part will not affect the standard of care or treatment you receive in the future.

#### **Is there a downside to taking part?**

If you do decide to take part then we will be asking you for some of your time, but we have planned this study so this would be minimal. There is a possibility in the questionnaire that we may ask you about some things that you find difficult or upsetting, but you can skip any questions you do not want to answer.

#### **What will happen if I don't want to carry on with the study?**



If you agree to take part and then later decide you want to stop being in the study that is OK. You can stop taking part at any time. If you stop being a part of the study, we will keep the information about you that we have already collected.

### Who has reviewed the study?

The study has been reviewed by survivors and caregivers who have also experienced a brain tumour themselves and by independent experts in this area of research. All research in the NHS is also approved by a Research Ethics Committee, an independent group that works to protect your interests.

### What will happen after the study?

We would like to keep your contact details so that we can send you an overview of what we have found in the study for the entire group. We also may want to contact you in the future about other studies. If you agree to this your details would be kept electronically on a secure University of Leeds database, for ten years after the study. The research team will be the only ones able to access your details.

### What if there is a problem?

If you have any concerns about any aspect of the study please speak to the researchers who will do their best to answer your questions. Their contact details are at the end of this information sheet. If you remain unhappy you may wish to contact the Leeds Teaching Hospitals NHS Trust's Patient Advice and Liaison Service (PALS) - T: 0113 2066261 or 0113 2067168 E: [patient.relations@leedsth.nhs.uk](mailto:patient.relations@leedsth.nhs.uk)

### What now?

Please let the researcher know whether or not you would like to help with the study or if you have any further questions they will be happy to help answer them.

#### How to contact us

If you have any questions about this study, please talk to the coordinating researcher:

Emma Nicklin – PhD student

(0113 206 8330 or [ss12ef@leeds.ac.uk](mailto:ss12ef@leeds.ac.uk))

Level 06, Bexley Wing  
St James' Institute of Oncology  
Beckett Street  
Leeds  
LS97TF

### Survivor Interview Guide

A topic guide will guide the interview; this will continue to develop iteratively throughout the research, with adaptations made during each interview and in response to each individual participant.

#### 1. **Opening**

##### **Establish rapport**

Introduce myself and the PhD project.

##### **Purpose and motivation**

Explain the aims of the research, how we hope the results will be used to help improve the long-term follow-up service and current support services.  
Explain how long the interview should take, and explain they can take breaks whenever needed.

### **Consent**

Adhere to ethical standards: read/sign consent, explain the audio-recording, confidentiality, their right to opt-out without affecting care. Ask if any questions.

## **2. To start can you please tell me a little about you, in any way that you can/want to?**

- Your family?
- What you like to do?
- How you like to spend your time?

## **3. After diagnosis/treatment**

*“When you were younger you were diagnosed with a brain tumour in the past. Can you tell me how your life has been since then?”*

- What would you say are the main changes since being diagnosed and treated?
- What kinds of things are hard for you?
- Is there anything you wish you could do but can't?
- How has having a brain tumour influenced you as you have grown older? (*High school/college/university, work, living independently?*)

## **4. Support services and Service use**

- Has there been anything or anyone who has helped you when you needed it?
- *“After your brain tumour what services have been involved in your after care? (for example – have you had any support from charities)”*
  - When did you receive this (during/after treatment)?
  - How did you end up with this form of supportive care or service?
  - What about it was helpful?
  - What about it was not so helpful?
  - Would you recommend this service to others?
- Are there any support services you're aware of that you haven't used? If anything, why?
- Would you want to help in this yourself or would you like your consultant/GP to seek out help for you?
- How do you think these services could be improved?

## **5. Long-term follow-up care**

*“I'd now like to talk about the Leeds follow-up care service that you attend”*

- How long have you been attending the LTFU clinic? How often do you go there? Do you feel this is enough contact? More or less?
- What do you find most useful about the LTFU clinic?
- Do you feel that these clinics/what is discussed is understandable for you as a teenager/young adult?
- Is there anything more you would like from these appointments?

## 6. Support recommendations

- Do you feel like there is anything you would have liked or would now like more support with?  
If yes, what?
- Do you think there are particular resources that should be provided for survivors when they are becoming older? (Teenagers/young adults)?
- What support formats are you most interested in? Would you prefer groups, individual, face-to-face, online, or a combination? And why?
- How do you think information on how to provide support could be improved for your age group?

## 7. Finish interview

*“Just before we finish do you have any additional comments?”*

- Is there anything else that you think we need to know?”
- How have you found this interview?

**Thank them for their time, and turn off audio-recording.**

## Caregiver interview guide

Whilst interviews will be flexible (and modified in response to efficacy and/or problems of prior ones) a general schedule is drafted:

### 1. Opening

#### **Establish rapport**

Introduce myself and the PhD project.

#### **Purpose and motivation**

Explain the aims of the research, how we hope the results will be used to help improve the long-term follow-up service.

Explain how long the interview should take, and explain they can take breaks whenever needed.

#### **Consent**

Adhere to ethical standards: read/sign consent, explain the audio-recording, confidentiality, their right to opt-out without affecting care. Ask if any questions.

### 2. To start can you please tell me a little about you, in any way that you can/want to?

- Your family/children?
- Employment
- What you like to do?

### **3. Survivor - After diagnosis/treatment**

- Can you tell me a little about [survivor name]?
- How has life been since [survivor name] brain tumour diagnosis?
  - What has changed?
  - What kinds of things are hard for [survivor's name]?
- How have these challenges influenced [survivor name] as they have grown older? (*High school/college/university, work, living independently?*)

### **4. Caregiver challenges**

*“Providing care for someone who has had a brain tumour can be challenging. Could you tell me about your experiences?”*

- Main changes/challenges since the diagnosis/treatment?
- Have these challenges changed since [survivor name] entered adolescence? If so how?
- Do you see these challenges changing in the future?
- Do you worry about your role as a caregiver in the future?

### **5. Support services and service use**

*“So you discussed challenges such as [what they have said] have you been able to find the support you’ve needed to deal with these challenges?” [This can be any care or support apart from the medical care from your treatment team, for example, help from supportive charities like brainstrust, conversations with a social worker or psychologist, peer support groups, self-help methods, etc]”*

- When did you receive this (during/after treatment)?
- How did you end up with this form of supportive care or service?
- What about it was helpful?
- What about it was not so helpful?
- Would you recommend this service to others?
- Do you think these services could be improved?
- Would you like to play an active role in searching for the right support/service (and if so, why)?
  - Would you want to help in this yourself or would you like your consultant or GP to seek out help for you?

### **6. Long-term follow-up care**

*“I’d now like to talk about the role of the Leeds follow-up care service that [survivor name] attends”*

- How long has [survivor name] been attending the LTFU clinic?
- For you and [survivor name] what do you get out of going to the clinic?
- How often do you go? Do you feel this is enough contact – once a year? More or less?
- In these appointments do you feel you can ask any questions about anything to do with [survivor name] health, behaviours, issues, needs?
- Do you feel that these clinics/what is discussed is age appropriate for TYAs?
- Is there anything more you would like from these appointments?

## 7. Support recommendations

- Do you feel like there is anything you would have liked or would now like more support with?
  - If yes, what?
  - Are you aware of services that provide this support?
  - Are there any support services you're aware of that you haven't used? If anything, why?

*Distinguish between patient support and caregiver support.*

- What support formats are you most interested in? Would you prefer groups, individual, face-to-face, online, or a combination? And why?
- Do you think there should be particular information that should be provided for parent caregivers at this specific transitioning age (Teenagers/young adults)?
  - When should these resources be provided?

## 8. Finish interview

*"Just before we finish do you have any additional comments?"*

- 1) Is there anything else that you think we need to know?"
- 2) How have you found this interview?

**Thank them for their time, and turn off audio-recording.**

## Appendix 7 – Accuracy of self-report vs. medical data (Poster BPOS 2020)



### How accurate do TYA childhood brain survivors and their caregivers recall medical history: implications for self-report studies

Emma Nicklin<sup>1</sup>, Galina Velikova<sup>1</sup> and Florian Boele<sup>1,2</sup>  
 Leeds Institute of Medical Research at St James's, Leeds, UK <sup>1</sup>  
 Leeds Institute of Health Sciences, University of Leeds, Leeds, UK <sup>2</sup>



#### BACKGROUND

Medical record review and self-or-proxy report are common methods to collect medical history data in oncology research. Especially in multicentre studies there can be financial and practical difficulties to medical record review. Therefore, patient/proxy reports are sometimes used instead of medical record reviews in oncology research. Yet, it is unknown if accuracy of self-/proxy-report medical data is influenced by time (memory bias) and/or presence of cognitive issues. This survivorship group often have cognitive deficits as a consequence of their diagnosis and treatment. Comparing data from different sources can provide insight into which data source is most appropriate to answer a specific research question.

#### AIM

To determine concordance between self-reported medical information with medical record data in teenage and young adult (TYA) childhood brain tumour survivors (BTS) and their caregivers, at least 5 years post-diagnosis.

#### METHODS

Using data collected from a cross-sectional study on the supportive needs of TYA childhood brain tumour survivors (BTS) and their caregivers, we compared participant questionnaire responses with medical data obtained from their medical records. Concordance between self-report and medical record data were analysed descriptively.

#### RESULTS

##### PARTICIPANT CHARACTERISTICS

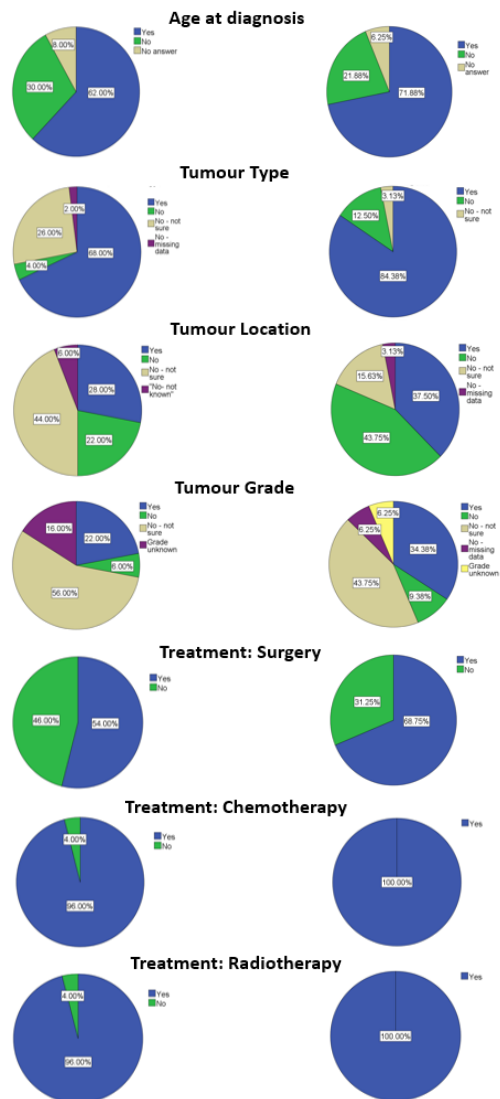
SURVIVORS (n=50)		n (%)	CAREGIVERS (n=32)		n (%)
Sex	Male	31 (62)	Sex	Male	6 (19)
	Female	19 (38)		Female	26 (81)
Age (13-30)	13-18	10 (20)	Age (37-64)	31-40	1 (3)
	19-24	20 (40)		41-50	8 (25)
	25-30	20 (40)		51-60	18 (56)
				60+	5 (16)
Tumour diagnosis	Medulloblastoma	21 (42)	Survivor tumour diagnosis	Medulloblastoma	15 (47)
	Astrocytoma	15 (30)		Astrocytoma	7 (22)
	Ependymoma	3 (6)		Ependymoma	2 (6)
	PNET	2 (4)		PNET	2 (6)
	Other	9 (18)		Other	6 (19)
Age at diagnosis (1-12)	0-4	19 (38)	Survivor age at diagnosis (1-12)	0-4	11 (34)
	5-9	20 (40)		5-9	16 (50)
	10-14	11 (22)		10-14	5 (16)
Time since diagnosis (7-27)	5-10	7 (14)	Time since survivor diagnosis (7-27)	5-10	4 (13)
	11-16	16 (32)		11-16	15 (47)
	17-22	19 (38)		17-22	11 (34)
	23+	8 (16)		23+	2 (6)
				Relationship to survivor	Parent

##### KEY FINDINGS

- Survivor rates of agreement ranged from 22% (tumour grade) – 96% (postsurgical treatment)
- Caregiver rates of agreement ranged from 34% (tumour grade) – 100% (postsurgical treatment)
  - Survivors were most likely to incorrectly recall age at diagnosis (30%).
  - Caregivers were most likely to incorrectly recall tumour location (44%).
- Both survivors and caregivers were less accurate about surgery history (BTS:54%; Caregivers:69%). **\*\*DISCUSSION POINT\*\*** We believe this may be due to the way the question was asked – using the term “resection” not “surgery”, which may have caused confusion. This shows the importance of using appropriate, patient friendly language.

#### SURVIVOR CONCORDANCE

#### CAREGIVER CONCORDANCE



#### CONCLUSION

In a group of long-term BTS and their caregivers, agreement between self-reported medical data and medical record review data varied depending upon the variables. Concordance was highest for postsurgical treatment history, but lower for variables related to diagnosis (i.e. tumour location and grade). Therefore, the use of self-/proxy-report may be an acceptable alternative to medical record abstraction in certain instances, but data from medical record review remains preferred.

**Appendix 8 – Comparison between online responders and participants recruited in LTFU clinics.**

**Survivor data**

	<b>LTFU clinic</b>	<b>Online</b>
<b>Sex</b>		
Male	62.0%	31.6%
Female	38.0%	68.4%
<b>Current age</b>	23.02 (13-30)	21.42 (16-30)
<b>Time since diagnosis</b>	16.54 (7-27)	19.43 (15-27)
<b>Age at diagnosis</b>	6.48 (1-11)	9.16 (2-14)
<b>Tumour type (Top 3)</b>	Medulloblastoma (42.0%) Astrocytoma (30.0) Ependymoma (6.0%)	Craniopharyngioma (26.3%) Medulloblastoma (15.8%) Astrocytoma (15.8%)
<b>Education</b>		
Higher education	32.0%	57.9%
No higher education	68.0%	42.1%
<b>Employment</b>		
Working	42.0%	36.8%
Not working	58.0%	63.2%
<b>Relationship status</b>		
Single	76.0%	89.5%
In a relationship	24.0%	10.5%

**Caregiver data**

	<b>LTFU clinic</b>	<b>Online</b>
<b>Caregiver sex</b>		
Male	18.8%	15.4%
Female	81.2%	84.6%
<b>Age</b>	53.44(37-64)	50.69 (40-63)
<b>Employment</b>		
Working	56.3%	53.9%
Not working	43.7%	36.1%
<b>Relationship status</b>		
Single	87.5%	38.5%
In a relationship	12.5%	61.5%
<b>Survivor age</b>	21.56 14-30)	17.7 (13-25)
<b>Time since diagnosis</b>	15.69 (7-27)	10.17 (5-15)
<b>Survivor age at diagnosis</b>	6.25 (1-12)	7.54 (0-14)
<b>Tumour type (Top 3)</b>	Medulloblastoma (46.9%) Astrocytoma (21.9%) Ependymoma (6.3%)	Medulloblastoma (15.4%) Astrocytoma (38.5%)

## Appendix 9 – Example of multivariable linear regression

### Variables Entered/Removed<sup>a</sup>

Model	Variables Entered	Variables Removed	Method
1	Age_current, Relationship_grouped, Time_since_diagnosis <sup>b</sup>	.	Enter
2	.	Age_current	Backward (criterion: Probability of F-to-remove >= .050).
3	.	Time_since_diagnosis	Backward (criterion: Probability of F-to-remove >= .050).

a. Dependent Variable: SCNSPC\_Total\_needs

b. All requested variables entered

### Model Summary

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate	Change Statistics			Sig. F Change	
					R Square Change	F Change	df1		
1	.568 <sup>a</sup>	.323	.266	10.608	.323	5.720	3	36	.003
2	.566 <sup>b</sup>	.321	.284	10.478	-.002	.100	1	36	.754
3	.526 <sup>c</sup>	.276	.257	10.673	-.045	2.428	1	37	.128

a. Predictors: (Constant), Age\_current, Relationship\_grouped, Time\_since\_diagnosis

b. Predictors: (Constant), Relationship\_grouped, Time\_since\_diagnosis

c. Predictors: (Constant), Relationship\_grouped

**Explanation: The variables removed (age\_current and time\_since\_diagnosis) have been removed as there is not a significant ( $\geq 0.50$ ) decrease in the  $R^2$  value. The R square decrease (predictive value) when current\_age was removed was -.002 and -.045 when time\_since\_diagnosis was removed. Model 3  $R^2$  is .276 meaning that it can account for 27.6% of the outcome.**

### ANOVA<sup>a</sup>

Model		Sum of Squares	df	Mean Square	F	Sig.
1	Regression	1930.681	3	643.560	5.720	.003 <sup>b</sup>
	Residual	4050.694	36	112.519		
	Total	5981.375	39			
2	Regression	1919.476	2	959.738	8.742	.001 <sup>c</sup>
	Residual	4061.899	37	109.781		
	Total	5981.375	39			
3	Regression	1652.959	1	1652.959	14.512	.000 <sup>d</sup>
	Residual	4328.416	38	113.906		



Total	5981.375	39		
-------	----------	----	--	--

- a. Dependent Variable: SCNSPC\_Total\_needs
- b. Predictors: (Constant), Age\_current, Relationship\_grouped, Time\_since\_diagnosis
- c. Predictors: (Constant), Relationship\_grouped, Time\_since\_diagnosis
- d. Predictors: (Constant), Relationship\_grouped

**Explanation: This table shows if the overall model is a significant predictor of the outcome variable, this table tells us the extent to which the individual predictor variables contribute to the model. Model 3 significance = <001.**

### Coefficients<sup>a</sup>

Model		Unstandardized Coefficients		Standardized Coefficients	t	Sig.	95.0% Confidence Interval for B	
		B	Std. Error	Beta			Lower Bound	Upper Bound
1	(Constant)	32.748	7.897		4.147	.000	16.732	48.765
	Relationship_grouped	-13.951	4.120	-.476	-3.386	.002	-22.308	-5.594
	Time_since_diagnosis	-.416	.493	-.170	-.845	.404	-1.415	.583
	Age_current	-.162	.514	-.063	-.316	.754	-1.205	.880
2	(Constant)	30.946	5.387		5.745	.000	20.031	41.861
	Relationship_grouped	-14.023	4.064	-.479	-3.451	.001	-22.257	-5.789
	Time_since_diagnosis	-.528	.339	-.216	-1.558	.128	-1.214	.159
3	(Constant)	24.556	3.558		6.902	.000	17.354	31.757
	Relationship_grouped	-15.394	4.041	-.526	-3.809	.000	-23.575	-7.213

- a. Dependent Variable: SCNSPC\_Total\_needs

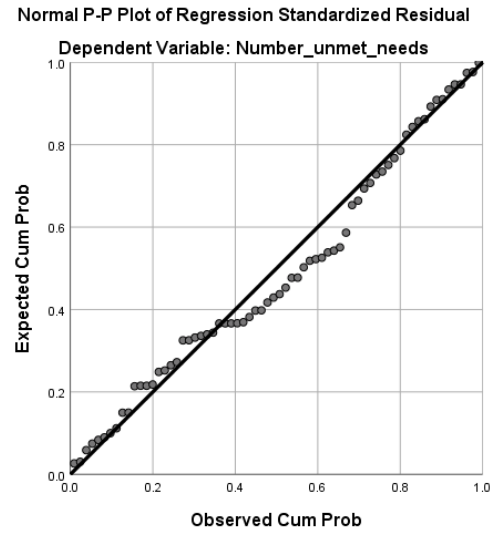
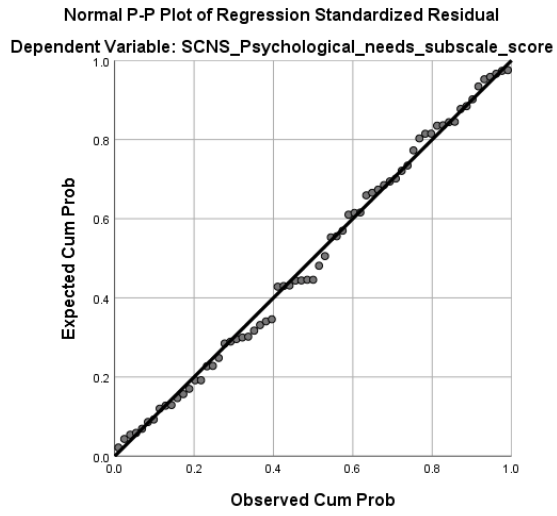
### Excluded Variables<sup>a</sup>

Model		Beta In	t	Sig.	Partial Correlation	Collinearity Statistics Tolerance
2	Age_current	-.063 <sup>b</sup>	-.316	.754	-.053	.466
3	Age_current	-.185 <sup>c</sup>	-1.330	.192	-.214	.962
	Time_since_diagnosis	-.216 <sup>c</sup>	-1.558	.128	-.248	.953

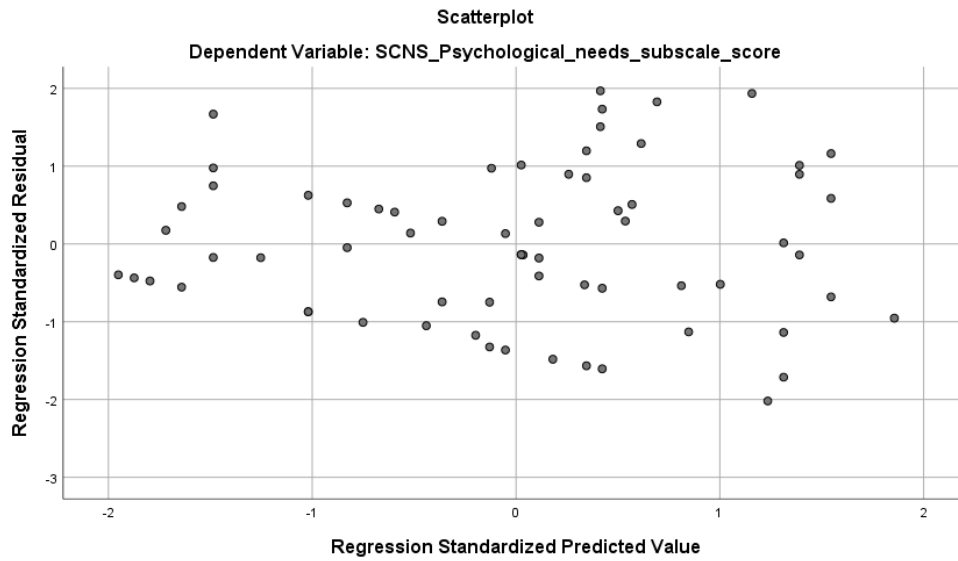
- a. Dependent Variable: SCNSPC\_Total\_needs
- b. Predictors in the Model: (Constant), Relationship\_grouped, Time\_since\_diagnosis
- c. Predictors in the Model: (Constant), Relationship\_grouped

**Appendix 10 – Assumption checks -multivariable linear regression analyses**

## Example P-Plots checking for normality



## Example test to check is homoscedasticity



## **Appendix 11 – Dissemination**

### **Journal articles**

Nicklin E, Pointon L, Glaser A, Sarwar N, Kwok-Williams M, Debono M, Velikova G and Boele F. 2021. "Unmet support needs in teenage and young adult childhood brain tumour survivors and their caregivers: "It's all the aftermath, and then you're forgotten about". [Manuscript accepted for publication] *Journal of Cancer Survivorship*.

Nicklin E, Velikova G, Boele F. 2020. Technology is the future, but who are we leaving behind? *The Lancet Oncology*. 21(1): 29-29

Nicklin E, Velikova G, Hulme C, Rodriguez Lopez R, Glaser A, Kwok-Williams M, et al. 2019. Long-term issues and supportive care needs of adolescent and young adult childhood brain tumour survivors and their caregivers: A systematic review. *Psycho-oncology*. 28 (3):477-87.

### **Oral presentations**

Nicklin, E. Issues and needs experienced by adolescent and young adult brain tumour survivors and their caregivers: A systematic review. Oral presentation at: European Association of Neuro-Oncology; 2018 October 10-14; Stockholm, Sweden.

### **Oral presentations (invited speaker)**

Nicklin, E. The Unmet Supportive Care Needs of Brain Tumour Survivors and Their Families'. Oral presentation at: SUCCESS Annual Charity Conference [Online]; 2020 November 21; United Kingdom.

Nicklin, E. Supportive care needs of young childhood brain tumour survivors and their caregivers. Oral presentation at: Paediatric Oncology & Haematology Research Meeting; 2019 July 11; Leeds, Leeds Teaching Hospitals NHS Trust.

Nicklin, E. Supportive care needs of TYA childhood brain tumour survivors and their caregivers: A mixed method study. Oral presentation at: European Association of Neuro-Oncology; 2019 September 19-22; Lyon, France.

Nicklin, E. Late Effects of Childhood Brain Tumours. Oral presentation at: Paediatric Brain Tumour Symposium; 2018 December 4; Nottingham, United Kingdom.

### **Poster presentations**

Nicklin, E, Velikova, G, and Boele F. Unmet support needs in childhood brain tumour survivors and their caregivers: "It's all the aftermath, you're forgotten about, basically in the

long-term you're left with it". Poster presented at: British Psychosocial Oncology Society; 2020 February 27-28; Edinburgh, United Kingdom

Nicklin, E, Velikova, G, and Boele F. How accurate do TYA childhood brain survivors and their caregivers recall medical history: Implications for self-report studies. Poster presented at: British Pscyhosocial Oncology Society; 2020 February 27-28; Edinburgh, United Kingdom

Nicklin, E, Hulme, C, Velikova G, and Boele F. Childhood brain tumour survivors—issues and needs in adolescence and young adulthood. Poster presented at: University of Leeds - Faculty of Medicine & Health Postgraduate Research Conference; 2018 June 13; Leeds, United Kingdom.

Nicklin, E, Hulme, C, Velikova G, and Boele F. Childhood brain tumour survivors—issues and needs in adolescence and young adulthood. Poster presented at: the British Pscyhosocial Oncology Society; 2018 March 8-9; Southampton, United Kingdom.