Innovative approaches to the self- and shared-management of arthritis by children, their families and professionals: A realist approach

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The candidate confirms that appropriate credit has been given within the thesis where reference has been made to the work of others. The author of this thesis was the first and corresponding author for all publications associated with the research presented in this thesis.

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Mum – this, and everything I will ever do in my life, is for you.

I hope this makes you smile,
I hope you are happy with my life,
At peace with every choice I made,
How I have changed, along the way.
Because I know you believed in all of my dreams
And I owe it all to you…

Dedicated to Sandra Mary Stones

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Abstract

Background: Juvenile idiopathic arthritis (JIA) is a complex long-term condition requiring lifelong management. Children and young people (CYP) should be empowered to self-manage their health and wellbeing (H&W) from diagnosis, while families should be supported in their shared-management role. Self- and shared-management (SSM) interventions can be used to develop SSM capacity; however, few studies have explored SSM in this population.

Aim: To explore how SSM of JIA can be promoted across the lifecourse by CYP, families, and professionals involved in their healthcare, wellbeing, and education.

Methods: Using a realist approach and underpinned by the individual and family self-management theory, evidence syntheses and a qualitative study were undertaken to identify, test, and refine a series of question theories promoting SSM of JIA. Twenty stakeholders were interviewed using a teacher-learner cycle approach. Data were analysed using hybrid deductive-inductive thematic analysis, and were integrated into a framework promoting SSM of JIA.

Results: Six refined question theories outlining the mechanisms by which SSM of JIA is likely to transpire, and the different contexts under which interventions achieve their desired outcomes, were developed and assimilated into a new, JIA-SSM framework. Within the framework, four levels of context related to SSM were identified, at an individual and interpersonal level of CYP, families and professionals; and at institutional and infrastructural levels across health and social care, education, and voluntary sectors. Individual healthcare plans can also act as shared-management tools to facilitate communication between CYP, families, and professionals across healthcare, wellbeing, and education.

Conclusions: The JIA-SSM framework encourages a shift towards a multi-intervention, multi-disciplinary, multi-agency approach which works with CYP and families in equipping them with the knowledge, skills, and behaviours to competently manage their H&W. Further research is recommended to apply and validate this framework in practice, in order to aid future design, delivery, evaluation, and implementation of interventions promoting SSM of JIA.

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List of abbreviations

A full list of abbreviations used in this thesis are included below.

ARMA	Arthritis and Musculoskeletal Alliance
BCW	Behaviour change wheel
bDMARD	Biological/biosimilar disease modifying antirheumatic drug
BSPAR	British Society for Paediatric and Adolescent Rheumatology
CAQDAS	Computer assisted qualitative data analysis software
CBT	Cognitive behavioural therapy
CCAA	Children's Chronic Arthritis Association
CHAQ	Childhood health assessment questionnaire
CI	Confidence interval
CMO	Context, mechanism, outcome
CNS	Clinical nurse specialist
COM-B	Capability, opportunity, motivation, behaviour
COVID-19	Coronavirus disease 2019
csDMARD	Conventional synthetic disease modifying antirheumatic drug
CYP	Children and young people
DMARD	Disease modifying antirheumatic drug
EAST	Easy, attractive, social, timely
EHCP	Education, health, and care plan
ES	Effect size
EULAR	European Alliance of Associations for Rheumatology
GP	General practitioner
H&SC	Health and social care
H&W	Health and wellbeing
HCP	Health care professional
HCSA	Health Conditions in Schools Alliance
HEEADSSS	Home, education, eating, activities, drugs, sexuality, suicide, safety
HRQoL	Health-related quality of life
iCAN	Irish Children's Arthritis Network
IFSMT	Individual and family self-management theory
IHP	Individual healthcare plan
ILAR	International League of Associations for Rheumatology
IQT	Initial question theory

JIA	Juvenile idiopathic Arthritis
LTC	Long-term condition
MDT	Multi-disciplinary team
MMAT	Mixed methods appraisal tool
MRC	Medical Research Council
N&V	Nausea and vomiting
NHS	National Health Service
NRAS	National Rheumatoid Arthritis Society
NSAID	Non-steroidal anti-inflammatory drug
OR	Odds ratio
PAM®	Patient activation measure
PhD	Doctor of Philosophy
PROM	Patient-reported outcome measure
QoL	Quality of life
REC	Research ethics committee
RF	Rheumatoid factor
RMD	Rheumatic and musculoskeletal disease
RQT	Refined question theory
SDM	Shared decision making
SEND	Special educational needs and disabilities
SENDCO	Special educational needs and disabilities co-ordinator
SNAC	Scottish Network for Arthritis in Children
SMI	Self-management intervention
SSM	Self- and shared-management
SSMI	Self- and shared-management intervention
TOAT	Task-orientated activity training
TTC	Teens taking charge
UK	United Kingdom of Great Britain and Northern Ireland
USA	United States of America
VCSE	Voluntary, community, and social enterprise
WWCiH	What? Why? Children in Hospital

Explanation of key terms

- **Arthritis:** A disease causing swelling within a joint, or limitation in the range of joint movement with joint pain, tenderness, stiffness or warmth, which persists for at least six weeks due to inflammation.
- **Children:** Individuals aged up to and including the age of 12 years.
- **Context:** Factors which influence whether an intervention may work or not.
- **Education professionals:** Teachers and other support staff.
- **Families:** Parents, carers, siblings, friends, and other relatives.
- **Grey literature:** Material that can be included in the study which has not been published in peer-reviewed journals.
- **Healthcare professional:** Members of the paediatric rheumatology multi-disciplinary team and wider clinical network.
- **Holistic:** A medical concept upholding people's physical and psychosocial needs, regarding individuals as a whole.
- **Interventions:** A general term covering care, treatment, information, education and support that a practitioner may provide.
- **Juvenile idiopathic arthritis:** A heterogenous group of disorders encompassing all forms of arthritis of unknown aetiology lasting for at least six weeks and with onset before the age of 18 years.
- **Long-term conditions:** Conditions for which there is currently no cure, and which are managed with multiple interventions over time, used interchangeably with the term chronic condition.
- **Mechanisms:** What it is about interventions that generate outcomes – the important changes that an intervention brings about.
- **Middle-range theory:** A theory which people in practice/services can associate with, and which can be tested.
- **Multi-disciplinary team:** A healthcare team including professionals from different disciplines working together to provide comprehensive services.
- **Outcomes:** The consequences of interventions.
- **Parents:** Parents and carers.
- **Programme/question theory:** How an intervention is supposed to work.
- **Proposition:** Statements which theorise about different context-mechanism-outcome components.

- **Realism:** A philosophical approach to conducting research, prioritising how and why things work, supporting the belief that one can work towards understanding causes of change.
- **Rheumatologist:** A specialist doctor caring for people living with rheumatic and musculoskeletal diseases.
- **Rheumatology:** The study of arthritis and other conditions of the joints, muscles and ligaments. When patients are children and young people, this is referred to as paediatric rheumatology.
- **Self-care:** The actions that individuals take for themselves in order to develop, protect, maintain and improve their health and wellbeing.
- **Self-efficacy:** An individual's belief in their innate ability to achieve goals.
- **Self-management:** The manner in which an individual manages the symptoms, treatment, physical and psychosocial impact of a condition.
- **Self- and shared-management:** The knowledge, skills, and behaviours required by individuals with long-term conditions and their support networks to manage collective health and wellbeing.
- **Shared-management:** The manner in which an individual's support network is involved in managing the symptoms, treatment, physical and psychosocial impact of a condition.
- **Transition:** The process of planning, preparing and moving from paediatric to adult health and social care services and settings.
- **Trypanophobia:** A fear of medical procedures involving injections or needles.
- **VCSE professionals:** Any employee or volunteer of a not-for-profit organisation.
- **Young adults:** Individuals aged 17–24 years.
- **Young people:** Individuals aged 13–16 years.

Chapter 1: Background

1.1 Introduction

This thesis presents a research study conducted towards the degree of Doctor of Philosophy (PhD), titled 'Innovative approaches to the Self- and shared-Management of ARThritis by children, their families and professionals' (iSMART). This was completed in the School of Healthcare at the University of Leeds, United Kingdom (UK). The study explores how self- and shared-management (SSM) of juvenile idiopathic arthritis (JIA) can be promoted across the lifecourse, and the stakeholders involved with SSM of JIA, including: children and young people (CYP) up to the age of 24 years, families, healthcare professionals (HCPs), education professionals, and voluntary, community and social enterprise (VCSE) professionals. This thesis provides a detailed overview of SSM interventions (SSMIs) for JIA, a theoretical understanding of SSM needs, and recommendations for promoting SSM of JIA in practice.

This chapter will provide an overview of the wider literature within the field, contextualising the research presented in this thesis, and justifying the need for the study. It concludes with a summary of the structure and overview of this thesis.

1.2 Juvenile idiopathic arthritis (JIA)

JIA comprises a group of heterogenous inflammatory disorders of unknown aetiology, persisting for at least six weeks with onset before the age of 18 years (Martini et al., 2019). JIA is most commonly classified using the International League of Associations for Rheumatology (ILAR) 2001 criteria of six phenotypic subtypes defined by clinical features and serological markers (Table 1) (Petty et al., 2004). These disorders are the most common type of juvenile-onset rheumatic and musculoskeletal diseases (RMDs) (Blazina et al., 2016), a diverse group of over 200 diseases regarded as the most "*prevalent, disabling and burdensome non-communicable diseases in Europe*" (p.4). They are estimated to affect in excess of 120 million Europeans (EULAR, 2019a).

In the UK, the incidence of JIA is estimated at 1 in every 10,000 CYP (Symmons et al., 1996). This equates to at least 1000 new cases each year, and a prevalence of 1 in 1000 CYP aged 16 years and under (ARMA, 2010),

although this likely to be an underestimation (McErlane et al., 2018). While gender distribution and age at disease onset differ by JIA subtype, collectively, JIA is more common in females, with disease onset at a median age of six years (Minden et al., 2002; Costello et al., 2019).

Table 1. ILAR 2001 criteria for JIA (Petty et al., 2004)

Classification	Definition
Systemic	Arthritis in ≥1 joints, with/preceded by fever persisting for ≥2 weeks, documented to be quotidian for ≥3 days, with ≥1 of the following: evanescent erythematous rash; generalised lymphadenopathy; hepatosplenomegaly; serositis.
Oligoarticular (Persistent) (Extended)	Arthritis affecting 1–4 joints (typically knees/ankles) during the first 6 months following disease onset. It is **persistent** when only ever affecting ≤4 joints, and **extended** when affecting ≥5 joints after 6 months of disease duration.
Polyarticular (RF-)	Arthritis affecting ≥5 joints during the first 6 months following disease onset, with a negative RF test.
Polyarticular (RF+)	Arthritis affecting ≥5 joints (typically smaller joints *e.g.*, fingers/wrists) during the first 6 months following disease onset, with ≥2 positive RF tests ≥3 months apart during the first 6 months following disease onset.
Enthesitis-related	Arthritis and/or enthesitis accompanied with ≥2 of the following: human leukocyte antigen-B27; acute anterior uveitis; onset in a male aged ≥6 years; presence/history of sacroiliac joint tenderness and/or inflammatory lumbosacral pain; first-degree relative with ankylosing spondylitis, enthesitis-related arthritis, sacroiliitis with inflammatory bowel disease, or Reiter's syndrome.
Psoriatic	Arthritis and/or psoriasis, with ≥2 of the following: dactylitis; onycholysis and/or nail pitting; first-degree relative with psoriasis.
Undifferentiated	Arthritis that fulfils none of the criteria for other categories or criteria for more than one category.

ILAR: International League for Associations for Rheumatology; JIA: Juvenile idiopathic arthritis; RF: Rheumatoid factor.

1.2.1 Disease course and severity

The disease course and severity of JIA is variable, depending on disease phenotype and individual manifestations (Adunuri and Feldman, 2015). However, general complaints with most ILAR subtypes include morning stiffness, gelling phenomenon (stiffness after extended periods of inactivity), worse arthralgia in the morning (with some improvement through the day), and fatigue (Jacobson and Pham, 2018). CYP can experience waxing and waning of symptoms, which may be experienced as a series of acute exacerbations, or 'flares', which are multi-layered and more complex than a simple increase in pain intensity (Khanom et al., 2020) which should be considered within a developmental biopsychosocial context (Stinson et al., 2012).

Systemic JIA is unique in that it can lead to impaired growth velocity, amyloidosis and a potentially life-threating complication known as macrophage activation syndrome (Martini, 2019). Rheumatoid factor (RF)-positive polyarticular JIA is thought to follow a more aggressive disease course to RF-negative polyarticular JIA, characterised by increased joint deformity (Petty et al., 2004; Espinosa and Gottlieb, 2012). Oligoarticular JIA tends to affect one, often larger, lower extremity joint, and so CYP appear generally well despite ambulating with a limp (Jacobson and Pham, 2018). However, it is associated with the development of iridocyclitis. Psoriatic JIA tends to be mild, though those found to be anti-nuclear antibody and human leukocyte antigen-B27 positive are at a higher risk of developing uveitis (Espinosa and Gottlieb, 2012).

Uveitis is the most common extra-articular complication of JIA, reported to affect 11–30% of CYP (Sen et al., 2015). The most common form of uveitis associated with JIA is iridocyclitis, a chronic anterior uveitis (Jacobson and Pham, 2018). It is typically asymptomatic amongst CYP, although severe inflammation can cause pain, redness and pupil distortion which may be recognised by a parent (Constantin et al., 2018). Although the risk of developing iridocyclitis decreases over time, it can develop many years after the onset of JIA. Therefore, CYP should undergo ophthalmologic screening throughout childhood and adolescence, even when JIA is in remission (PRINTO, 2016).

Although inactive disease and remission can be achieved, JIA remains incurable, characterised by a relapsing and remitting pattern with capacity for progression and impaired functional status without treatment (Gidman et al., 2015). Around one third of those diagnosed with JIA during childhood will

continue to experience active inflammation into adulthood, requiring long-term treatment (Coulson et al., 2014).

1.2.2 Clinical management

Clinical management of JIA requires a multi-faceted, multi-disciplinary, holistic approach, consisting of pharmacological and non-pharmacological interventions (Prakken et al., 2011). Ultimately, disease management goals include achieving remission, maximising function, optimising growth and development, minimising pharmacological toxicity and improving quality of life (QoL) (Gowdie and Tse, 2012). However, despite various evidence-based management guidelines, including the 2010 British Society for Paediatric and Adolescent Rheumatology (BSPAR)/Arthritis and Musculoskeletal Alliance (ARMA) Standards of Care (Davies et al., 2010) and the 2019 American College of Rheumatology recommendations (Ringold et al., 2019), there remains a lack of consensus-driven, evidence-based treatment guidelines for all JIA subtypes, contributing to disparities in access to interventions and practice within and between countries (Webb and Wedderburn, 2015).

There are two key aims of pharmacological interventions. The first is to reduce symptoms, and the second is to control disease activity, with the aim of remission. Aside from analgesia, there are three broad groups of drugs used to treat JIA: non-steroidal anti-inflammatory drugs (NSAIDs); corticosteroids; and disease-modifying anti-rheumatic drugs (DMARDs) – the latter consisting of conventional synthetic (cs), biological/biosimilar (b) and targeted synthetic DMARDs. Pharmacological management has markedly changed in recent decades (Lovell et al., 2013; Ruperto and Martini, 2018), including early and more aggressive use of DMARDs during the ‘window of opportunity’, and the unprecedented safety and efficacy of bDMARDs (Isaacs and Burmester, 2020). This has resulted in an increased number of CYP achieving clinically inactive disease and remission (Wallace et al., 2012; Hinze et al., 2015), in line with a treat-to-target approach which has been advocated for (Ravelli et al., 2018), with the caveat of ensuring CYP and families have an active role in choosing personal targets (Schoemaker et al., 2020).

For many CYP, the csDMARD methotrexate is used as a first line treatment; however, intolerance and non-adherence can be problematic, largely due to side effects including nausea and vomiting (N&V), within 24-36 hours post-

administration (Taketomo et al., 2011; van Dijkhuizen and Wulffraat, 2014). Methotrexate intolerance is estimated to affect around 50% of CYP (Bulatović et al., 2011; Scheuern et al., 2016; Falvey et al., 2017) and is most likely to occur within 6–12 months of commencing methotrexate (van Dijkhuizen et al., 2015). It is the leading causes of discontinuation or dose reduction (Smits et al., 2020), and can lead to a decreased QoL and psychological distress, including the development of anticipatory and associative N&V (Falvey et al., 2017). Various strategies are used to reduce methotrexate-induced N&V, including folate supplementation, switching route of administration from oral to subcutaneous injection, use of anti-emetics, and behavioural therapy, though the results of the latter are inconclusive (Falvey et al., 2017). New approaches being explored include the use of psychobiological principles of pharmacological conditioning (Smits et al., 2020).

While pharmacological interventions play one part of managing disease activity and symptoms, non-pharmacological interventions are imperative for optimising QoL (Fellas et al., 2017), particularly since CYP with JIA display limitations in participating in physical, occupational and social leisure-related activities (Cavallo et al., 2014). Psychosocial support is a critical component of JIA management, given that a number of CYP have anxiety and/or depression secondary to their symptoms, and the emotional distress JIA has on their lives, and their ability to perform childhood activities comparable to their peers (Jacobson and Pham, 2018). Physiotherapy aims to preserve and increase CYP's range of motion, muscle strength, aerobic conditioning, physical fitness and subsequent function while limiting strain on joints (Catania et al., 2017). Research has shown that CYP with JIA are less physically fit and active than their peers (Klepper, 2008), and while muscle atrophy and weakness are common during an active flare of JIA, these may not resolve completely during remission (van der Net et al., 2008; Hulsegge et al., 2015). Therefore, physiotherapists play a role in providing appropriate home exercise regimens (Jones et al., 2009) as well as helping CYP understand the safety and benefit of exercise and physical activity more broadly (Klepper et al., 2019), in order to prevent deconditioning (Catania et al., 2017). Occupational therapy can help to maintain CYP's functioning and ability to participate in occupational and leisure activities, including the use of assistive devices, if commenced early and routinely (PRINTO, 2016). Occupational therapists may also be able to supply

braces and splints to offload mechanical stress, while maintaining joint position to prevent pain, stiffness, contracture and deformity (Fellas et al., 2017). CYP with lower limb pathologies, including biomechanical issues in the foot and ankle, will most likely be referred to a podiatrist and/or orthotist for assessment for orthotic devices and footwear recommendations (Giancane et al., 2016). The most common minimally invasive surgical procedure which CYP experience is arthrocentesis, which can help to decompress the joint, thus relieving pressure and subsequent pain. This normally involves concomitant administration of intraarticular corticosteroids to suppress inflammation, typically under conscious sedation (De Ranieri et al., 2020). Although the need for orthopaedic surgery is less common nowadays, in CYP whose JIA cannot be managed conservatively, or in those with joint damage, surgical intervention may be required to improve their QoL (Giancane et al., 2016).

Adherence to pharmacological and non-pharmacological interventions is critical for improving outcomes and QoL. Reports suggest that as few as 53% of CYP with RMDs have good overall adherence to pharmacological interventions (Coda et al., 2017). Reasons for non-adherence include child refusal, concerns about future consequences of treatments, side effects, pain, personal issues, forgetfulness, embarrassment, lack of medication availability, and financial hardship (Coda et al., 2017; Favier et al., 2018). Favier et al. (2018) describe non-adherence as an "*under-recognised and potentially modifiable obstacle to care*" (p.690).

An additional component of JIA management relates to transitional care, defined as "*the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-oriented healthcare systems*" (p.570) (Blum et al., 1993). Transition extends from early adolescence through to the third decade of life (Palman et al., 2018), although the actual age of transfer from paediatric to adult health and social care (H&SC) typically occurs typically between the ages of 16 and 18. Transition in JIA has received increasing attention in recent years (Foster et al., 2017), with the need for a structured, co-ordinated programme of transitional care well described (Shaw et al., 2007b). The goal of transition is to enable CYP to acquire and develop the necessary knowledge, skills, and behaviours required to be "*independent, empowered and responsible adults*" (p.639) (Foster et al., 2017). Various tools have been developed to assess satisfaction with transitional care

(Shaw et al., 2007a); however, research has shown that although disease activity may not increase after transfer, the number of CYP clinic non-attenders does (van Pelt et al., 2018). Some reports suggest as many as 50% experience an unsuccessful transition, increasing the risk for unfavourable outcomes (Conti et al., 2018). Tattersall (2014) remarks that risk-taking behaviour, appointment non-attendance, and treatment non-adherence are normal in the "*complex neurocognitive development that characterises adolescence*" which "*continues into the mid-20s*" (p.8). Some of these challenges can be proactively identified by HCPs using the Home, Education, Eating, Activities, Drugs, Sexuality, Suicide, Safety (HEEADSSS) 3.0 psychosocial interview (Klein et al., 2014; Tattersall, 2014).

It is also important for CYP and families to be involved in shared decision making (SDM). Research has demonstrated that CYP who are actively engaged in SDM with their parents and HCPs may be more competent at managing JIA into adulthood (Grande et al., 2019). Indeed, SDM should be based upon CYP's values, priorities and experiences, with HCPs recognising contextual influencers of CYP and family behaviour (Tesher and Onel, 2012). Decision aids have been suggested as useful resources to aid SDM, although there is limited comparable evidence among CYP and families (Grande et al., 2019).

1.2.3 Living with JIA

After a diagnosis, CYP and their families are often forced to accept the physical and psychosocial consequences of their disease, which have far-reaching consequences throughout their lives (Foster et al., 2020). They must adapt and integrate JIA into their developing identities. This requires cognitive maturity and familiarity with the disease (Cartwright et al., 2015).

A thematic synthesis of 27 qualitative studies describing the experiences of 542 CYP identified six themes capturing the substantial disruption that JIA has on their sense of normality and capacity for social participation (Tong et al., 2012). Many CYP are averse to being different – an involuntary state resulting from symptoms, impairment, internal disfigurement, differential treatment and forced dependency on others. CYP also strive for normality, by preserving social identity, being resourceful, having a sense of community, focussing on remission, and pushing themselves beyond their physical limitations. Stigma and misunderstanding can also be problematic, due to trivialisation, the invisible

nature of symptoms and treatment side effects, the perception that arthritis is a disease of 'old age', and the future potential of workplace discrimination. CYP also feel suspended in uncertainty, often caught between fluctuating states of control and powerlessness, while attempting to balance hope with disappointment. Managing treatment is another important aspect of life with JIA, with CYP needing to understand the use and benefits of treatment, while feeling respected, involved in SDM, and motivated to be physically active. Finally, CYP have a desire for knowledge, including being kept informed about treatment efficacy and safety, treatment advances which offer hope for the future, and lifestyle management advice so that they can feel empowered to manage their JIA (Tong et al., 2012).

Despite the significant impact of JIA on their lives, many CYP have a remarkable resilience (Cartwright et al., 2015), in part because of their desire to regain a sense of control over their lives. In one qualitative study, four facilitative strategies were identified (Cartwright et al., 2015). The first focuses on taking responsibility and overcoming limitations by adopting an optimistic attitude to coping, as CYP strive to "*assume personal agency*" through "*physical and psychological mastery*" (p.738). Exemplar coping strategies include disclosure, communication skills, maintaining activities with friends, minimising pain and ignoring negative comments (Beneitez et al., 2020). The second includes minimisation, often through downward comparison to other CYP who were comparably worse off; in addition to distraction techniques, although memories of their journey can be "*just below the surface and could be triggered by casual comments… revealing the fragility of this sense of normality*" (p.738). Balancing disclosure and concealment was the third strategy, recognising the potential benefits of support and empathy with disclosure, in contrast to the normalising function of concealment – reflecting the differing levels of acceptance and the complexity of concealment as more than a coping strategy. Finally, familial and peer support were identified as coping enablers, through solidarity and reassurance from peers with a shared understanding of JIA. While some CYP feel content with current friendships, physical and psychosocial isolation often remain, particularly where symptoms such as pain interfere with socialising; hence, CYP have expressed interest in interacting with other CYP with JIA, particularly to learn about how others manage to live with JIA. This desire to build friendships upon shared grounds of empathy and support generally has a

positive impact on CYP's ability to cope with the stigma and misunderstanding associated with JIA, having been shown to positively impress their personal identities (Beneitez et al., 2020). These findings were supported by a UK survey of concerns amongst CYP and families (Stones and Wright, 2015), where the need for ongoing information, advocacy, social support, SDM involvement were identified.

1.2.4 Impact of JIA on outcomes

JIA is known to impact biopsychosocial development, given its multidimensional effects on CYP (Eyckmans et al., 2011; Tong et al., 2012) over time (Weiss et al., 2014). Thus, it is prudent to consider psychosocial, educational and vocational outcomes together with disease and treatment-related outcomes (Palman et al., 2018). The goals of JIA management, recognised from a holistic point of view (Beresford, 2011), are to reduce inflammation, ameliorate symptoms, prevent radiological progression and irreversible damage, improve biopsychosocial functioning, and ultimately improve health-related QoL (HRQoL) (Borchers et al., 2006; Consolaro et al., 2016). Indeed, CYP with JIA have lower HRQoL than their peers (Kip et al., 2019), as do their caregivers (Kuhlmann et al., 2016), in part influenced by JIA symptoms, functional impairment, poor sleep, and treatment side effects, which interfere with daily activities (Haverman et al., 2012). This impact can be particularly severe at disease-onset and during diagnosis (Cartwright et al., 2015), though HRQoL has also been shown to be suboptimal in CYP with no or mild symptoms (Seid et al., 2009). Seid et al. (2014) suggest that non-clinical factors, such as self-efficacy, parental emotional distress and social support, account for 30% of the impact of JIA on HRQoL, with improvements potentially lagging behind measures of disease activity, pain and disability (Oen et al., 2018).

Pain is recognised as one of the most common and distressing symptoms of JIA at all ages (Palman et al., 2018). Pain can disrupt sleep (including delayed sleep-onset, discontinuous sleep and napping behaviour), disrupt school attendance, limit day-to-day activities and impact on psychosocial functioning (Weiss et al., 2014; Giancane et al., 2017). It can also persist when CYP are receiving treatment and inflammation has resolved (Consolaro and Ravelli, 2013; Lomholt et al., 2013; Anink et al., 2015). Pain correlates strongly with the detrimental effect of JIA on physical HRQoL in a longitudinal 30-year follow-up

study (Rashid et al., 2018), and even a minimum decrease in pain intensity has been shown to have a positive effect on health and wellbeing (H&W) (Dhanani et al., 2002). Fatigue is also recognised as a common, frustrating and often debilitating symptom or a side effect of treatment, shown to negatively influence psychosocial health (Armbrust et al., 2016), encompassing the mental (thinking), emotional (feeling), social (relating), and spiritual (being) dimensions of health. Given that three quarters of all adult mental illness are believed to begin during young adulthood (Kessler et al., 2005), early consideration of the psychosocial impact of JIA, including routine screening and intervention should be promoted (Palman et al., 2018), including pre-diagnosis while symptomatic (Foster et al., 2009). Depression during childhood has been shown in some studies to be more common in CYP with JIA (Krause et al., 2017; Hanns et al., 2018), with depressive symptoms accounting for variability in levels of HRQoL (Stevanovic and Susic, 2013); though this has not always been shown (Ding et al., 2008). While depressive symptoms have been associated with worse pain, disability and active disease; depressive symptoms at baseline have been shown to predict future disability and pain, but not active disease (Hanns et al., 2018). CYP with JIA have been found to experience increased adjustment problems and higher rates of internalising behaviours, such as anxiety, depression, social withdrawal, and dysphoria, which have been shown to be linked to rumination and catastrophising, two maladaptive cognitive emotion regulation strategies (Garnefski et al., 2009). To a lesser extent, increases in externalising behaviours, such as aggressiveness, hyperactivity, disruptiveness, oppositional behaviour, and impulsivity, have also been reported (Cartwright et al., 2015; Palman et al., 2018). Factors found to be associated with psychosocial H&W during active disease include pain, physical limitations and poor sleep (Nozoe et al., 2014; Stinson et al., 2014). However, changing body image, social acceptance and future uncertainties contribute to such behaviours, including those in remission (Memari et al., 2016), as well as those with physical changes as a result of JIA and its treatment (Borchers et al., 2006).

As CYP adapt and adjust to living with JIA, fears for the future often come to the fore, including the uncertainty of enduring disease, prospect of worsening health, need for life-long treatment, and the childhood "*irretrievably lost*" (p.737) to JIA (Cartwright et al., 2015). Interestingly, CYP diagnosed with JIA at a

young age, typically in the pre-school years, have been reported to demonstrate better psychosocial adjustment to those diagnosed with JIA later on, as have their parents (Toupin April et al., 2013). This view is supported by Beales' theory of the child's view of chronic illness, which describes younger CYP as seeing their condition as being less severe, and less of a personal disaster, than older CYP (Beales, 1983), which may help them to cope with their JIA more easily. CYP have also described their journey with JIA as character forming and building, which has enabled them to become stronger and more determined individuals (Cartwright et al., 2015). This view is supported by Antonovsky's salutogenic model, which theorises that wellbeing is positively impacted by people's sense of coherence developed during childhood (Antonovsky, 1987). Indeed, CYP with JIA in well-supported families with greater psychosocial resources appear to better adjust to the psychosocial consequences of JIA (Huygen et al., 2000; Rapoff et al., 2003), which may be explained by an altered frame of reference, resulting in a decreased need to compare themselves to peers (Toupin April et al., 2013).

CYP and families alike can find the hospital and medical interventions stressful, with many experiencing increased anxiety, due to the fear of the unknown, uncertainty, the intimidating nature of medical equipment, separation from family, and a sense of losing control (Bray et al., 2020). Quite often, this can be attributed to inadequate information and poor preparation in anticipation of hospital attendance and medical interventions (Duff et al., 2012), potentially leading to disengaged CYP becoming distressed and uncooperative (Bray et al., 2016). Anxiety linked to such interventions can have lasting effects on CYP's H&W, and participation in H&SC (Kahana et al., 2006), while also increasing the likelihood of longer and delayed appointments, and referrals to psychological services (Bray et al., 2020).

In addition, management of trypanophobia (fear of medical procedures involving needles) and pain is problematic (Taddio et al., 2009; McMurtry et al., 2015), a concern given that many CYP regularly require parenteral treatment (Sørensen et al., 2020a; Sørensen et al., 2020b). Research has shown that high levels of fear are associated with perceived pain during needle procedures, adding to the stress CYP and families are already under. CYP often express fears indirectly, as cues and nonverbal signs, more than direct statements. For example, facial expression, body language and crying (Sørensen et al., 2020a).

Intervening to minimise fear is important, since recollection of distress may cause anticipatory fear and increased pain during future procedures (Noel et al., 2012), potentially leading to post-traumatic stress disorder, non-adherence to treatment, and an aversion to H&SC later in life (Sørensen et al., 2020b). Experiencing treatment effects and self-confidence tend to be critical for maintaining motivation for parenteral treatment, alongside distraction strategies, honest and sensitive communication between CYP and adults, allowing CYP to have time to reflect on the situation, and involvement in self-administration when HCPs acknowledge their fears as genuine (Sørensen et al., 2020b).

CYP and their families have identified this lack of appropriate information and preparatory support as an unmet need (Bray et al., 2019b), calling for further resources to enable them to fully prepare for medical interventions (Bray et al., 2019a). Historically, such preparatory material has included leaflets and books (Egert et al., 2017; Tsao et al., 2017), and while serving their purpose, there is an increasing desire for internet- and smartphone application-based interventions (Fortier and Kain, 2015). Bray et al. (2020) identified some of these interventions, but remarked that they predominantly focused on surgical admission and other disciplines, such as radiology. For CYP living with JIA, such a limited focus is unlikely to reflect the wider range of interactions and interventions they will encounter during their lifetime. Recent examples aiming to address this information gap include Xploro®, a digital therapeutic platform using augmented reality, artificial intelligence, gameplay, and conversational agents to inform, educate, and empower CYP (Xploro, 2020); and What? Why? Children in Hospital (WWCiH) procedural videos (What? Why? Children in Hospital, 2020).

JIA management goals are monitored through the regular application of validated outcome measures (Passo and Taylor, 2008; Lovell et al., 2011; Consolaro et al., 2014). Given the complexity of JIA, no single measure can reliably capture all relevant variables (van Mater et al., 2012), and so the use of composite measures has markedly increased (Consolaro et al., 2016). Furthermore, greater emphasis has been placed on collecting patient-reported outcome measures (PROMs), in order to capture the wider impact of JIA and treatment on overall wellbeing and QoL (Palman et al., 2018). This is particularly important in light of discordance between CYP, parents and HCPs (Garcia-Munitis et al., 2006; Sztajnbok et al., 2006; Consolaro et al., 2007).

Evidence has also shown that the use of PROMs in practice may improve the quality of care received (Berard and Laxer, 2011), by ensuring that CYP and families are involved in SDM and all aspects of care (Hersh et al., 2016). One of the most frequently used PROMs for JIA is the Childhood Health Assessment Questionnaire (CHAQ) (Singh et al., 1994). As well as capturing relevant outcomes, it is also pertinent to understand their experience of care, given that experience and outcomes are intrinsically linked. Five patient-reported experience measures were proposed as part of a JIA core dataset, addressing disease and treatment understanding, SDM, adequate support, and favourable experiences within the clinical environment (McErlane et al., 2019).

Many CYP with JIA will experience education and vocational disruption, including a lower attendance to their peers, as a consequence of school absenteeism (Bouaddi et al., 2013). Yet, educational attainment and employment in CYP with JIA has received limited attention (Jetha, 2015), despite recognition that vocational support is uncoordinated, limited and unresponsive (Shaw et al., 2006a). CYP with JIA are at risk of poorer educational and vocational outcomes (Maslow et al., 2011a), although one study has shown a trend for higher education attainment in secondary and tertiary education (Malviya et al., 2012). However, another large, longitudinal cross-sectional study showed that educational attainment was significantly lower compared to the general population (Schlichtiger et al., 2017). Aside from attainment, it is important for CYP to have a positive experience in education, which may become problematic because of JIA symptoms. This can be helped by improving awareness amongst educational professionals, as well as introducing reasonably adjustments (PRINTO, 2016).

CYP also seem to waver between disclosure and concealment, with the latter driven by an eagerness to be ‘normal’ and persevere in the face of adversity (van Gulik et al., 2020). Qualitative studies in this under researched area have called for greater cohesion between H&SC, school and work, in order to improve vocational outcomes for CYP with JIA (Hanson et al., 2018).

1.2.5 The impact of JIA on the family

Parents are often the sole advocates for CYP, particularly when they are younger, and during the diagnostic process, where they must balance vigilance and responsibility to pursue answers about their child’s symptoms with the risk

of being labelled "*neurotic, protective or even exploitative*" (p.402) (Foster et al., 2009). Some parents will consider themselves well-informed and will have the health-seeking knowledge, skills, and behaviours to take prompt action in response to their child's symptoms. However, others may not be able to judge the severity of symptoms and the need to seek advice, through no fault of their own, and may require ongoing support (McGoron and Ondersma, 2015). This latter group of parents may be spurred on to seek HCP input by education professionals *in loco parentis*, who may have observed changes in CYP's functions and/or behaviour (Foster et al., 2009).

Parents have remarked on the difficulty of securing a JIA diagnosis, and the psychosocial toll that JIA inflicts on families (Jones et al., 2009). Life often revolves around JIA, impacting relationships between parents, CYP, and other family members, as well as activities such as work (Jones et al., 2009). Previous research exploring the experiences of parents tend to engage with mothers, whereas fathers are often viewed as peripheral. However, research has been undertaken to explore fathers' experiences, who while also navigating 'loss', appear to conceal their distress by adopting denial and distraction strategies (Waite-Jones and Madill, 2008).

Parents value information from HCPs and those with lived experience of JIA; however, some describe deficits particularly related to psychosocial health, complementary and alternative therapies, and vocational outcomes (Thon and Ullrich, 2009).

Although parent-proxy reports are necessary for very young CYP with JIA, there has been discussion as to whether parent-proxy reports accurately reflect CYP's perceptions (Palman et al., 2018). Variations in agreement between parents and CYP have been suggested, particular in those CYP experiencing invisible manifestations of JIA, such as depression, as well those whose condition is mild or severe (Shaw et al., 2006b; Lal et al., 2011). This acts as a reminder to ensure that the voice of both CYP and parents are heard throughout the SDM process.

1.2.6 H&SC provision

CYP with JIA are managed by a specialist paediatric rheumatology multi-disciplinary team (MDT) in tertiary care (Figure 1), though in many parts of the world, provision is sub-optimal (Foster et al., 2009). The National Health Service

(NHS) England Standard Contract advocates, where appropriate, for specialist teams to work with local teams as part of a network, per shared care protocols, to deliver optimal care as close to CYP's homes as possible (NHS England, 2013). The Contract also recognises that CYP may require the input of other services, such as those from VCSEs, "*in order to optimise outcomes that are focussed on [CYP] and their families*" (p.7) (NHS England, 2013).

Between 2009 and 2010, BSPAR and ARMA published their consensus-derived standards of care for JIA (BSPAR, 2009; ARMA, 2010), focused on empowering CYP and families through an holistic approach to care. However, despite the introduction of these standards and an increasing awareness of the negative impact of delayed access to paediatric rheumatology (Foster and Rapley, 2010), a multi-site UK audit against the standards demonstrated considerable variation in service delivery and referrals (Kavirayani et al., 2013). This led to the development of a national audit tool for JIA, to enable standardised clinical data collection (McErlane et al., 2018).

The diagnosis pathway for JIA is often prolonged, involving complex referrals and delays in access to paediatric rheumatology services, with three in four CYP experiencing a referral delay of 10 or more weeks (Foster et al., 2007). Delays to diagnosis are principally a result of insufficient awareness of JIA, particularly among generalist HCPs in primary care and emergency departments, who are often the first point of contact for CYP and their families (Foster et al., 2009). This can result in erroneous referrals to specialities inadequately equipped with experience of JIA, as well as unnecessary and invasive procedures (Foster et al., 2009). Tools introduced to support identification of JIA include the paediatric Gait, Arms, Legs and Spine tool, (Foster and Jandial, 2013) and the paediatric musculoskeletal matters resource (Smith et al., 2016).

Aside from referral delays, access to care is affected by various other factors, including CYP's residence, family education and insurance status when there is no access to publicly-funded healthcare (Tesher and Onel, 2012). Many CYP can also go undiagnosed or misdiagnosed, with HCPs attributing symptoms to 'growing pains' (Lehman and Carl, 2017). Additionally, JIA is a diagnosis of exclusion, with HCPs excluding other conditions that present with similar clinical manifestations (Giancane et al., 2016).

It is evident that CYP and families must navigate complex care pathways, interacting with several HCPs, in order to receive the care and support they require. Factor input from other education and VCSE professionals, and the landscape becomes increasingly complex. Hence, formal SSMIs for CYP with JIA and their families are pertinent, in order to better support these individuals in coping with the challenges of JIA.

Figure 1. The paediatric rheumatology MDT

Secondary/tertiary care services
Core MDT
Paediatric rheumatology consultants and registrars, paediatric rheumatology CNS, physiotherapists, occupational therapists, clinical psychologists, pharmacists, podiatrists, orthotists, youth and family support workers, clinical secretaries and administrators
Core MDT if part of a shared care network
Adult rheumatologists with an interest in paediatric rheumatology, paediatricians with an interest in paediatric rheumatology
Wider MDT
Ophthalmology consultants and registrars, dentists, orthodontists, dieticians, psychiatrists, radiographers, radiologists, pain specialists, play therapy specialists, and social workers
Primary/community care services
General practitioners, physician associates, community pharmacists, practice nurses, phlebotomists, district nurses, health visitors, and school nurses

CNS: Clinical nurse specialist; MDT: Multi-disciplinary team.

1.2.7 The costs of JIA

Although there have been few economic evaluations in JIA (Gidman et al., 2015), Kuhlmann et al. (2016) remarked that the magnitude of JIA-related direct and indirect H&SC costs are high and pose a considerable financial burden to CYP, families and society. This is illustrated by the estimated cost of care for people with long-term conditions (LTCs) in the UK, estimated to cost around £7 in every £10 of total H&SC expenditure (Department of Health, 2012). Direct costs include the use of pharmacological and non-pharmacological interventions, outpatient and inpatient hospital visits, HCP consultations,

laboratory tests, imaging, splints and devices, supplements, alternative and complementary therapy, treatment administration costs and overhead/fixed resources (Kip et al., 2019). Indirect costs include a range of expenses, including transportation, home adaptations, specialist equipment, childcare, in-patient accommodation, telephone costs, parking, and schooling (Kip et al., 2019). Furthermore, there are productivity costs associated with JIA, particularly amongst parents, including absenteeism, presenteeism, employment cessation, and early retirement (Kip et al., 2019). There are also productivity costs for CYP and their siblings (Shenoi et al., 2018), although these are poorly understand (Gidman et al., 2015). Examples include school absence, additional tutorial time, future employability, future absenteeism/presenteeism, and premature employment cessation (Kip et al., 2019).

1.2.8 VCSE provision

Various VCSEs provide information, education, and support services to CYP with JIA and families, addressing a gap in provision not currently met by statutory H&SC providers. Table 2 outlines the key VCSEs based in the UK and Ireland offering the aforementioned services in JIA, many of which are delivered by people with lived experience of JIA. Some VCSEs are members of ARMA – an umbrella body for rheumatology VCSEs and professional bodies, as well as the Health Conditions in Schools Alliance (HCSA) – a coalition of VCSEs advocating for CYP with medical conditions in school.

Most VCSEs have published a range of readily accessible print and digital media to inform and educate CYP, families and other stakeholders, in addition to various support services offered in specific regions of the country. Indeed, the format and delivery of VCSE services is diverse (Appendix 1, Appendix 2), ranging across the self-care spectrum. Most services utilise a group mode of delivery, with education and support sessions and residential/excursion programmes the most common. Of individual-based services, education, including written and visual information, are predominant. No one VCSE appears to provide services across all formats and elements identified, and there appears to be a limited focus on goal setting, individual level skills training, and enhancing self-management capacity (Stones et al., 2021).

Beyond the UK and Ireland, there are a number of other VCSEs offering similar services. Several of these are members of the European Network for

Children with Arthritis and Autoinflammatory Diseases, a pan-European VCSE network facilitating the exchange of best practices (Stones et al., 2019).

The VCSE landscape in general, and for JIA, is relatively complex. The VCSE sector is often shaped by historical decisions, which can result in an increased number of organisations with similar goals (Krasteva and Yildirim, 2016). The consequences are fragmentation and malalignment of the sector into multiple entities – essentially competing for donor fundraising (Radojev, 2018). While this may be unintentional, the fragmentation may add to the confusion CYP and families face when sourcing relevant information, education and support. Interestingly, the coronavirus disease 2019 (COVID-19) pandemic has enlightened the types of services often disregarded as essential components of statutory H&SC, but which are vital for CYP and families to be able to manage their H&W.

1.2.9 General awareness of JIA

JIA is largely invisible, with ‘arthritis’ often perceived to afflict older people (Tong et al., 2012). Thus, awareness of JIA is generally inadequate amongst the public and many HCPs (Jandial et al., 2009; Egert et al., 2019), prompting for improved understanding while concurrently minimising stigma (Foster et al., 2020). Several annual awareness-raising campaigns have been developed to help address these deficiencies, including World Young Rheumatic Diseases Day (Smith et al., 2020), World Juvenile Arthritis Day (Arthritis Foundation, 2019), Wear Purple for JIA (NRAS, 2021), and World Arthritis Day (EULAR, 2019b).

Table 2. VCSEs in the United Kingdom and Ireland offering JIA information, education, and support services

Name	Constitution	Membership	
		ARMA	HCSA
Focus: JIA			
Children's Chronic Arthritis Association	Charitable incorporated organisation association*	X	X
Irish Children's Arthritis Network	Charitable CLG‡		
JIA-at-National Rheumatoid Arthritis Society	Charitable CLG*†	X	X
Juvenile Arthritis Parents and Families United Kingdom	Unregistered entity		
Juvenile Arthritis Research	Charitable CLG*		
Kids Like Us	Charitable CLG*		
Scottish Network for Arthritis in Children	Unincorporated association†		
Focus: Uveitis			
Olivia's Vision	Charitable CLG*		
Focus: Rheumatic and musculoskeletal diseases			
Arthur's Place	Community interest company registered in Scotland		
Versus Arthritis	Charitable CLG*†, charitable trading company	X	X
Focus: Long-term conditions			
Teapot Trust	Scottish Charitable incorporated organisation†		
What? Why? Children in Hospital	Scottish Charitable incorporated organisation†		

*Registered with the Charity Commission for England and Wales; ‡Registered with An Rialálaí Carthanas, the Irish Charities Regulator; †Registered with the Scottish Charity Regulator. ARMA: Arthritis and Musculoskeletal Alliance; CLG: Company limited by guarantee; HCSA: Health Conditions in Schools Alliance; JIA: Juvenile idiopathic arthritis; VCSE: Voluntary, community and social enterprise.

1.3 Self- and shared-management of JIA

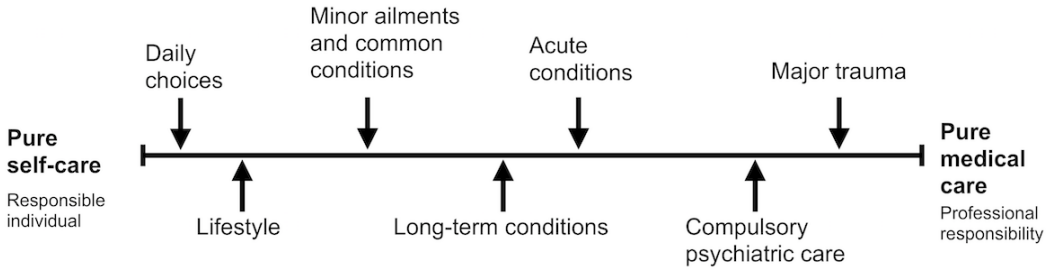
1.3.1 Policy context of LTCs

The prevalence of LTCs is projected to increase (Bee et al., 2018), already affecting in excess of 15 million people in England (Department of Health, 2012). Over a quarter of these are CYP (Wijlaars et al., 2016), whose H&W is likely to be affected across the lifecourse (Mock and Arai, 2011). People with LTCs account for around 50% of general practice appointments, 64% of outpatient appointments and over 70% of inpatient bed days (Coulter et al., 2013); thus, with an increasing demand for H&SC, services must maximise resources without compromising care quality, experience, and outcomes (Bee et al., 2018). The Chronic Care Model advocates for proactive H&SC reinforced by six system areas conducive to high quality LTC management, including supporting people with LTCs to self-manage their H&W (Wagner, 1998), while "*reducing the fiscal burden on healthcare systems*" (p.1) (Bee et al., 2018). However, disparity across the UK has been highlighted (LTCAS and The Scottish Government, 2008), with calls for greater use of legislative levers to promote self-management (Finnis et al., 2016). In England, this was signified in the NHS Long-Term Plan, with a focus on supported self-management of LTCs (NHS England and NHS Improvement, 2020).

1.3.2 Self-management

Self-management can be regarded as a component of self-care (Wilkinson and Whitehead, 2009). Self-care was conceptualised by nurse theorist Dorothea Orem as "*the practice of activities that individuals initiate and perform on their own behalf in maintaining life, health, and wellbeing*" (p.117) (Orem, 1991). Since Orem first described self-care in the 1990s, its definition has evolved into a continuum (Figure 2) from pure self-care through to self-management of acute conditions and LTCs, and pure medical care (Wilkinson and Whitehead, 2009). However, self-management terminology is used interchangeably with self-care, self-help, and patient education (RCN, 2021). Although they do share commonalities, self-management is not synonymous with these other terms (Ryan and Sawin, 2009), since self-management encompasses the totality of actions taken by individuals to manage the medical, role, and emotional aspects of their LTC (Barlow et al., 2002; Ahmad et al., 2014).

Figure 2. The self-care continuum (Self Care Forum, 2020)



The skills and behaviours required to competently self-manage LTCs were first described by Corbin and Strauss (1988), and advanced by nursing scholar, Kate Lorig (Lorig et al., 2001; Lorig and Holman, 2003), who presented six core skills required for self-management. These include: problem solving; decision making; resource utilisation; forming a patient-HCP partnership; taking action; and self-tailoring (Lorig and Holman, 2003). These correspond to the three domains of self-management summarised by Sattoe et al. (2015): medical management (*e.g.*, taking medication); role management (*e.g.*, social participation); and emotional management (*e.g.*, sharing feelings). Ryan and Sawin (2009) went on to describe self-management as three phenomena (Table 3), recognising self-management as a process leading to outcomes, rather than as an outcome or endpoint *per se* (Miller et al., 2015) – thus, viewing interventions as tools to introduce and develop self-management skills amongst people with LTCs (process), across the domains of self-management (outcome) (Ould Brahim, 2019).

In 2011, The Health Foundation published a report emphasising that self-management is not a panacea, or something which can be achieved through the use of solitary interventions (Ould Brahim, 2019). Rather, self-management interventions (SMIs) are most likely to be effective when implemented as part of an holistic approach to H&SC, focused on behaviour change, supporting self-efficacy (de Silva, 2011) and empowering individuals and their families to manage their H&W effectively (Coulter et al., 2008; Davies et al., 2010). This is supported by the notion that information provision alone is unlikely to sufficiently motivate sustainable behavioural change, in comparison to SMIs co-created and co-delivered by CYP and their support network (Funnell, 2010; Tzeng et al., 2010).

Table 3. Self-management phenomena (Ryan and Sawin, 2009)

Phenomena	Description
Process	The use of self-regulation skills to manage LTCs and/or risk factors. Processes may include activities such as: goal setting, decision making, self-monitoring, reflective thinking, planning for and engaging in specific behaviours (Bodenheimer, 2003).
Intervention	Specific interventions designed to prepare individuals to assume responsibility for managing LTCs (Bodenheimer, 2003).
Outcome	Outcomes achieved by individuals' engaging in the self-management process (Chodosh et al., 2005), *e.g.*, treatment adherence can reduce symptoms (Feldman et al., 2007).

LTC: Long-term condition.

Self-efficacy, the main construct of social cognitive theory (Stajkovic and Luthans, 2003), refers to an individual's belief in their ability to complete difficult tasks, achieve goals, and cope in the face of adversity (Bandura, 1977). Marks et al. (2005) remarked that enhanced self-efficacy can improve coping, communication and control, by experiencing small personal achievements which bolsters the confidence required to overcome potential barriers (Duckworth et al., 2007; Fauth and Thompson, 2009; Turner et al., 2015; Burd and Hallsworth, 2016a).

Since the turn of the 21st century, there has also been a shift from reactive to proactive intervention strategies (Coleman et al., 2009) and delivery of remote interventions (Sørensen et al., 2020b), assuming a more active and evolving role for CYP with LTCs and their families (Coulter et al., 2013). This remote management has been intensified during the COVID-19 pandemic (BSR, 2020).

1.3.3 Self-management of juvenile-onset LTCs including JIA

While the historical target group for self-management has been adults with LTCs (Newman et al., 2004), the impact of LTCs on CYP's physical and psychosocial H&W is increasingly recognised in the research literature (NHS Confederation, 2012). In CYP, LTCs like JIA can have a significant negative influence on their QoL and that of their families. Evidence also suggests that CYP growing up with LTCs are less likely to achieve important educational and

vocational milestones, compared to their healthy peers (Maslow et al., 2011b). Therefore, strategic delivery of SMIs at the earliest opportunity, and at times of maximal impact across the lifecourse, could encourage them to adopt self-management behaviours at an early age (Burd and Hallsworth, 2016c) – a rational approach considering that many CYP will face a lifetime of managing their LTC (Modi et al., 2012) with a variety of H&SC needs across the lifecourse (Department of Health, 2008). This is supported by evidence indicating that the age of seven is a developmentally-appropriate age for CYP to begin developing self-management control (Bal et al., 2016). Indeed, there is literature which suggests that early, developmentally-appropriate childhood SMIs have a substantial impact on behaviours and long-term outcomes when effectively negotiated with CYP, their families and HCPs (Lindsay et al., 2014). Indeed, empowered individuals who self-manage their H&W effectively are more likely to have clinical indicators in the normal range (Hibbard and Gilburt, 2014), bearing particular relevance for planning when CYP transition to adult H&SC, where CYP are expected to self-advocate and self-manage their H&W autonomously, often without the complete repertoire of knowledge, skills, and behaviours required (Tattersall, 2014). This is also reflective of changing health literacy and numeracy needs, which have shifted from CYP solely acquiring knowledge from parents and HCPs, to CYP being able to readily access information via the internet, media and peers (Matsuoka et al., 2016).

The process of transition to adulthood is characterised by anticipation for the future, regret for the past which has been lost, anxiety about the future, major psychosocial readjustment and a degree of ambiguity of status during the transition (Colver et al., 2018). During this period, CYP face a number of focal concerns, including: developing/maintaining self-identity and gender identity; gaining independence from parents; accepting/rejecting adult values, acquiring/adjusting to an occupation; and developing friendships and relationships (Coleman and Hendry, 1999). In the pursuit of these focal concerns, CYP must negotiate the physical, psychosocial and cognitive transitions of adolescence, which is even more complex when living with JIA (Kelley et al., 2004). Sattoe et al. (2015) remarked that the focus for self-management in CYP should be experiential, involving ‘mastery experiences’ within their own communities which enable them to observe, learn and model behaviour from peers while feeling accepted (Bandura and Walters, 1971;

Bandura, 1999; Bandura, 2001). However, reference is also made to the need for varying self-management approaches to different LTCs, balancing more technical interventions with cognitive and behavioural interventions (Kirk et al., 2010). In JIA, there is likely to be a balance across technical, cognitive, and behavioural interventions, recognising the complexity and impact of the condition (ARMA, 2010; NHS Confederation, 2012), with the caveat that adult self-management models cannot be effectively translated for use by CYP (Modi et al., 2012).

Some of the more specific SSM activities which CYP with JIA will generally need to perform include: seeking information; seeking advice from peers; developing support networks; consulting HCPs; developing effective partnerships with multiple professionals; problem solving; self-diagnosis, self-monitoring, and self-treatment; adopting health behaviours beneficial to their H&W; adjusting to new social and economic circumstances; and managing the psychosocial consequences of JIA on their lives (Marks et al., 2005; Kirk et al., 2010).

1.3.4 Beyond the 'self' in self-management

Self-management comprises several dimensions which are still under development throughout childhood and adolescence (National Academies of Sciences and Medicine, 2019); therefore self-management in the context of juvenile-onset LTCs like JIA inevitably involves families, as well as other significant adults in CYP's lives, such as education and VCSE professionals.

With a lifecourse perspective, the Shared Management Model implies that as CYP with LTCs like JIA mature, they should increasingly take on responsibility for self-managing their H&W, as agency shifts from parents to CYP (Kieckhefer and Trahms, 2000; Lindsay et al., 2014). This can be facilitated by HCPs and other professionals who listen, understand underlying behaviours, and then empower CYP and their families while leaving space for reciprocal dialogue (Len et al., 2014). Moreover, it is recognised and valued that families play an indisputable role in sharing the management of their child's H&W (Smith et al., 2015). Yet, there is limited formal recognition and support for family members and other involved professionals who take on the shared-management of CYP's H&W, until CYP develop the knowledge, skills, and behaviours to self-manage their LTC effectively. Therefore, a new term was introduced in the study

presented in this thesis – SSM, defined as 'the knowledge, skills, and behaviours required by individuals with LTCs and their support networks to manage collective H&W'. This term recognises the importance of equipping CYP with the capacity to self-manage, while reflecting on the role and needs of families and other professionals who operate in a shared-management role. Shared-management is a complementary yet different role to that of self-management. This is an evolution of other terms, such as supported self-management (NHS England and NHS Improvement, 2019a), by placing direct emphasis on the impact that a shared-management role can have on those individuals providing support, who are in need of assistance themselves to execute their role effectively, while considering their own H&W needs.

Indeed, literature focusing on the families supporting CYP with JIA has been limited, despite broader research evidence of the impact of LTCs like JIA on the entire family, and the recognition of health promoting effects of family relationships for CYP (Stones, 2017b). Thus, a more contemporary approach to self-management as SSM is required, drawing on relational autonomy (Ould Brahim, 2019) to recognise and acknowledge the societal factors and other contextual influencers which shape CYP's behaviour and experiences (Tesher and Onel, 2012), addressing "*the social embeddedness of individuals, the complexities involved in behaviour change, and the environmental constraints and facilitators that impact health outcomes*" (p.8). Such contexts need to be considered not just at the individual and interpersonal level, but also at the institutional and infrastructural level, characterising the interdependency of services and providers in providing holistic, individualised care to CYP and their families (Coulter et al., 2013). This includes educational institutions, which have been recognised as conducive environments for enabling CYP to self-manage with shared-management support from peers and education professionals (Kirk et al., 2012).

1.3.5 Measuring SSM

Aside from the CHAQ, SSM capacity is not routinely measured or assessed in clinical practice, despite availability of validated tools such as the Patient Activation Measure (PAM®) (Hibbard and Gilburt, 2014; Bomba et al., 2018). The concept of patient activation is widely recognised, and describes the knowledge, skills, and confidence individuals have in managing their own H&W

(Foot et al., 2014). The concept, which recognises the relationship between poor health literacy and health status (Nutbeam, 2008; Peerson and Saunders, 2009), can be helpful in understanding groups of CYP and families (Table 4), to enable SSM approaches to be tailored to meet individual and family needs. Evidence shows that people with low levels of activation and health literacy are less likely to play an active role in maintaining healthy behaviours; and therefore, more intensive intervention to increase their level of activation could improve engagement and H&W (Hibbard and Gilburt, 2014). Conversely, those who demonstrate high levels of activation and health literacy may require less intense intervention to promote SSM.

Table 4. The four levels of patient activation (Hibbard and Gilburt, 2014)

Level	Description
1	Individuals tend to be passive, feel overwhelmed by managing their own H&W, and may not understand their role in the care process.
2	Individuals may lack the knowledge and confidence to manage their H&W.
3	Individuals appear to take action but may still lack the confidence and skills to support their behaviours.
4	Individuals have adopted many of the behaviours needed to support their H&W but may not be able to maintain them unwaveringly.

H&W: Health and wellbeing.

1.3.6 Deriving a CYP-friendly SSM framework for JIA

Although SSM is frequently discussed within the H&W arena, the concept generally lacks consensus (Van de Velde et al., 2019), particularly for CYP with JIA and their families. Indeed, adult models of self-management fail to account for key differences with CYP outlined in this chapter – namely childhood development, the shared-management role of families and other professionals (Lozano and Houtrow, 2018), and the shifting agency from parents to CYP. Furthermore, self-management behaviours and overall competency are not attributes which can developed instantaneously through interaction with a solitary, time-constrained SSMI; rather, self-management as a process requires a longer-term approach across the lifecourse (Foot et al., 2014). The disconnect between the use of solitary SSMIs and incremental tactics to develop SSM

competency using a multi-intervention approach may prevent systematic changes to SSM approaches in practice and indeed the impact of interventions on outcomes – alluded to by one evidence synthesis of interventions which implied that improvements in QoL may be of minimal influence (Bee et al., 2018). However, the evidence for SSM of JIA and the use of SSMIs for CYP with JIA first needs to be explored, in order to provide more conclusive judgements to shape future SSM practices in this population.

1.4 Thesis structure and overview

This thesis consists of eight chapters. Following this background Chapter 1, Chapter 2 discusses the evidence base pertaining to SSMIs in the form of an integrative review. Chapters 3 and 4 outline the overall study methodology and the methods used in the qualitative study, respectively. Chapter 5 presents the initial question theories (IQTs) based upon the literature presented in Chapters 1 and 2, providing a tentative theoretical understanding of SSM needs in JIA. Chapter 6 presents the qualitative testing of IQTs with key stakeholders, discussed thematically under refined question theories (RQTs) emerging from the evaluation. Chapter 7 summarises the RQTs, providing a more definitive theoretical understanding of SSM needs in JIA. This is then followed by a proposed framework promoting SSM of JIA. Finally, Chapter 8 summarises the overall results of the study, considering the findings within the wider literature. This chapter also reflects on the overall strengths and limitations of the thesis, its theoretical and practical implications, and recommendations for supporting SSM of JIA in practice.

1.5 Situating the researcher's perspective

The motivation for the research presented in this thesis derives from the researcher's personal experience of living with JIA since early childhood. He witnessed first-hand the impact of JIA on his physical and psychosocial H&W, as well as the impact it had on his family and peers. In addition, he has a decade of experience as an independent patient advocate in the UK and internationally. In this role, he has worked in partnership with countless CYP, families, HCPs and VCSE professionals in understanding their experience, amplifying their voice, and facilitating their involvement in research and service development. Combined with a background in Biomedical Sciences and a

career in science and medical communications, his clarity of vision combined with a deep rooted understanding of the nuances of SSM of JIA, ideally position him to explore this topic further. This approach is substantiated by Pawson and Tilley's (1997) sentiment in their seminal text:

> *"We believe, in short, that the strength of evaluation research depends on the perspicacity of its view of explanation"* (p.219).

1.6 Summary

This chapter has outlined the background to the study through a description of JIA, illuminated by a summary of the literature on living with JIA and the provision of services to support CYP and families. The phenomena of interest in the study, SSM, was summarised, with reference to its utility for CYP with JIA and families, and the researcher's motivation for researching SSM of JIA. However, there is limited empirical evidence regarding SSM of JIA and the use of SSMIs amongst this population. Therefore, a review of the literature was undertaken to identify and describe SSMIs for CYP with JIA and families, which is presented in Chapter 2.

Chapter 2: Literature review

2.1 Introduction

An initial scope of the literature revealed an extensive array of SSMI studies in certain LTCs, such as asthma and type one diabetes mellitus, though fewer in juvenile-onset RMDs, including JIA (Stones, 2018; Saxby et al., 2019). In addition, there appeared to be an emphasis on promoting self-management around the age of transfer to adult H&SC services, compared to across the lifecourse. Therefore, it was necessary to summarise the empirical evidence of SSMIs for CYP with JIA RMDs, and their families across the lifecourse. At this early stage of the study, an integrative review was deemed to be the most appropriate review design to establish a more comprehensive understanding of all potentially-relevant SSMIs for CYP with JIA and other juvenile-onset RMDs. Indeed, an integrative review allows for data from a methodologically diverse range of studies to be critiqued, categorised, and thematically summarised (Noble and Smith, 2018), thus providing a current overview of the landscape which could be further explored, including evidence gaps requiring further investigation. It is these evidence gaps which prompted the focus of the study to shift towards a realist approach to evaluation, in order to identify mechanisms operating within SSMIs, and their influence on outcomes under different contexts.

2.2 Aim of the review

The aim of the integrative review was to identify and describe the empirical evidence base on the use of SSMIs for CYP with RMDs and their families. The following questions guided the review:

- What are the characteristics, content and components of SSMIs?
- How effective are SSMIs at achieving their anticipated outcomes?
- Are SSMIs feasible, usable, and acceptable?

2.3 Methods

2.3.1 Design

The integrative review followed a six-stage process (de Souza et al., 2010), beginning with the problem identification stage, and the establishment of a clear

review purpose. This was followed by a systematic and well defined literature search, and then data extraction. A critical analysis of studies was then performed using validated tools to assess for methodological quality. Subsequently, extracted data were ordered, and compared, so as to allow themes and patterns to arise. Finally, a synthesis was undertaken to comprehensively portray the process of integration of data from the included studies. Unlike other review methods, integrative reviews allow for the inclusion of experimental and non-experimental research (Whittemore and Knafl, 2005). Consequently, an integrative review was the most appropriate method to use here to identify and describe SSMIs for juvenile-onset RMDs across a diverse range of methodologies.

2.3.2 Inclusion criteria

Inclusion criteria were outlined *a priori* of the review being conducted, based upon the types of studies, participants, interventions, outcomes and reporting language. Any studies not meeting these inclusion criteria were excluded from the review.

2.3.2.1 Types of studies

All qualitative, quantitative, and multi/mixed methods studies were included.

2.3.2.2 Types of participants

The sample included CYP and young adults aged up to 24 years, so as to capture self-management across the lifecourse in accordance with the United Nations definition of youth as up to the age of 24 years (Nandigiri, 2012). The aforementioned CYP were diagnosed with one or more RMDs and/or multimorbidities. Studies targeted at a broader group of LTCs were eligible for inclusion if they included RMDs. The sample also included parents and families.

2.3.2.3 Types of interventions

Any form of programme, project or initiative applied at the individual or group level which aimed to improve CYP's self-manage capacity (or families shared-management role) through the provision of information, education, and/or support, were included. This included any reference to concepts aiding self-management, such as problem solving, decision making, healthcare utilisation,

patient-HCP relationships, taking action or control, goal setting, and confidence-building mechanisms. Interventions were reviewed against the nine evidence-based intervention functions of the second layer of the Behaviour Change Wheel (BCW) (Michie et al., 2011), to provide a systematic approach to grouping interventions by their intended behaviour change technique (Table 5).

2.3.2.4 Types of outcomes

Any relevant medical, role, and emotional outcomes were included. These included condition-specific and general outcomes, and could either be HCP-reported, patient-reported, parent-reported, proxy-reported or physiological. Economical outcomes were also be included.

2.3.2.5 Language

Only studies published in the English language were included.

2.3.3 Search methods

Studies published between 01 January 2010 and 08 January 2021 were identified through a comprehensive search of eight bibliographic databases: Ovid MEDLINE, Embase, and HMIC; EBSCO PsycINFO, CINAHL, and ERIC; BEI; and Wiley Cochrane Library. One grey literature database, Social Care Online, was searched. The systematic search was restricted to 2010 to reflect the contemporary nature of paediatric SSM with newer treatments. However, various studies (Lavigne et al., 1992; Rapoff et al., 2002; McDonagh et al., 2007; Lomholt et al., 2015) pre-dating 2010 were included through citation tracking and additional searches since they were theoretically relevant to answer the first review question regarding the characteristics, content and components of SSMIs. The search strategy was informed by a university library information specialist, and included MeSH subject headings, free-text search terms and variations relating to self-management, CYP and families, and RMDs (Appendix 3). All literature was screened against inclusion criteria at the title and abstract and full-text stage (Appendix 4).

Table 5. Intervention functions (Michie et al., 2011)

Intervention	Definition	Example
Education	Increasing knowledge or understanding.	Providing information about JIA.
Persuasion	Inducing positive or negative feelings or stimulating action.	Showing videos of CYP undergoing procedures.
Incentivisation	Creating expectation of reward.	Using a lucky dip bag of prizes after an injection.
Coercion	Creating expectation of punishment or cost.	Limiting access to social opportunities if non-adherent to treatment.
Training	Imparting skills.	One-to-one sessions with a CNS on self-injecting.
Restriction	Reducing the opportunity to engage in target behaviours (or to increase target behaviours by reducing the opportunity to engage in competing behaviours).	Identifying boundaries for participation in youth group activities and contact with professional facilitators out-of-hours.
Environmental restructuring	Changing the physical or social context.	Using smartphone applications to prompt CYP to take their medication.
Modelling	Providing an example for people to aspire to or imitate.	Involving young adults living well with JIA as speakers at family days.
Enablement	Increasing means/reducing barriers to increase capability beyond education and training, or opportunity beyond environmental restructuring.	Providing splints, aids and exercises to promote participation in educational and vocational activities, including physical activity.

CNS: Clinical nurse specialist; CYP: Children and young people; JIA: Juvenile idiopathic arthritis.

2.3.4 Search outcome and article selection process

Details of the article identification and selection process are presented in Figure 3. The initial search yielded 1048 references. These were exported from the databases and imported into EndNote X9 (The EndNote Team, 2013). After de-duplication, 858 references proceed for title and abstract screening. One hundred and twelve articles were subsequently retrieved for full-text screening, of which 26 articles met the inclusion criteria and were included in the review. Of the 26 articles, there were 24 research articles (describing 17 unique interventions) and two review articles. From the review articles, four relevant and otherwise uncaptured research articles were examined, describing four unique interventions. Therefore in total, 28 studies were extracted describing 21 unique interventions. It is worth noting that of the articles excluded during full text screening, 49 were conference proceedings without full text articles. While these conference proceedings were relevant to the aims of the integrative review, because their quality could not be reasonably assessed, the amount of presented data diminutive, and with the restricted timelines of the study, a decision was made to exclude them from the integrative review.

2.3.5 Quality appraisal

Given the review included qualitative, quantitative, and mixed methods studies, the methodological quality of included articles was assessed using the Mixed Methods Appraisal Tool (MMAT; Appendix 5) (Hong et al., 2018a; Hong et al., 2018b). The MMAT was designed for the quality appraisal of systematic mixed studies reviews, providing criteria to appraise qualitative studies, randomised controlled trials, non-randomised controlled studies, quantitative descriptive studies, and mixed methods studies. Therefore, the MMAT was chosen since it is a validated tool with accompanying guidance, and enabled concomitant appraisal of included studies with different designs (Hong et al., 2018a). Since review articles were also included, the methodological quality of included reviews was assessed using the Critical Appraisal Skills Programme Systematic Review Checklist (Appendix 6) (Critical Appraisal Skills Programme, 2018).

2.3.6 Data extraction and synthesis

Given the heterogenous design of included studies, data could not be combined statistically, and so a thematic analysis and synthesis was completed. Firstly,

data from quantitative studies were converted into textual descriptions to aid integration with data from qualitative studies. Data from the included articles were individually coded, and then summarised and recorded within two data extraction forms, addressing study characteristics (Appendix 7); and intervention characteristics (Appendix 8). This assisted the researcher in identifying patterns across the included studies, which were grouped into broad themes through an iterative process of moving within and across the studies to provide a intelligible narrative summary of the literature. This is presented as a narrative summary of SSMI characteristics, content, components, effectiveness, and acceptability. Where reported in the included studies, the results of statistical tests are included in the text discussing SSMI effectiveness.

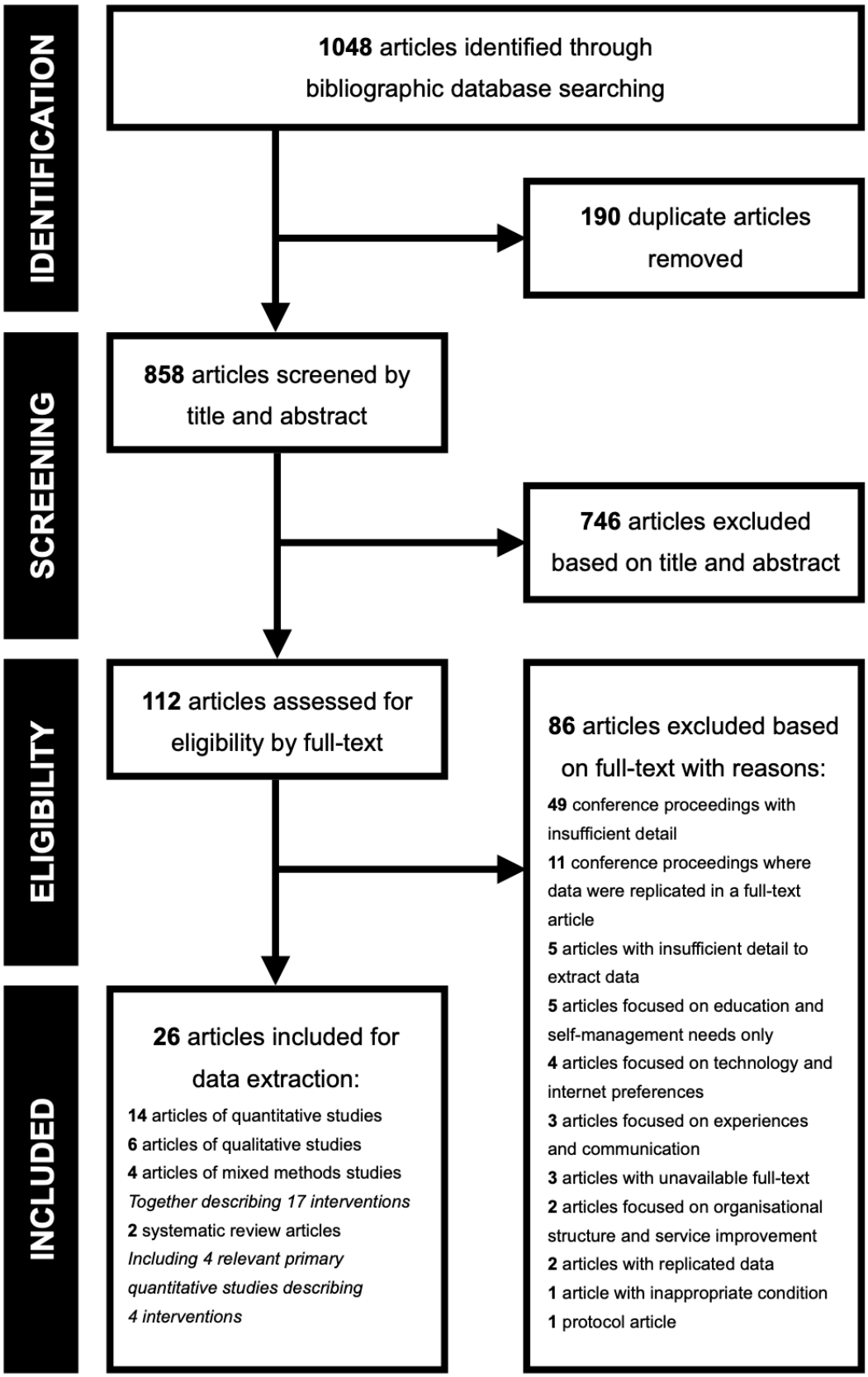
2.4 Results

The key study characteristics from each article are outlined in Appendix 7. First, a description of the studies is provided, followed by the characteristics, content, components, and effectiveness of SSMIs. Results are described for each article, despite some articles reporting on multiple steps of a phased development of a single intervention.

2.4.1 Description of the studies

Although the 26 articles proceeded to data extraction, a total of 28 articles were identified: 24 articles were identified from the systematic search, and a further four articles (Lavigne et al., 1992; Rapoff et al., 2002; McDonagh et al., 2007; Lomholt et al., 2015) extracted from the two included systematic reviews (Lindsay et al., 2014; Sheng et al., 2019). The inclusion of these articles enabled relevant studies published prior to 2010 to be captured. The nature of the integrative review meant the 28 included articles were heterogenous.

Figure 3. PRISMA flow diagram outlining the article selection process



PRISMA: Preferred reporting items for systematic reviews and meta-analyses.

2.4.1.1 Design

Around two-thirds of included studies (64%) were quantitative. Ten were randomised controlled or crossover trials (including waitlist controlled); six were quasi-experimental (including before and after, cohort, and multiple baseline single-subject designs); and two were descriptive quantitative (including surveys). Six studies had a qualitative design, utilising semi-structured individual interviews, focus groups, observation, and surveys. Four studies also had a sequential explanatory mixed methods design; two were RCTs followed by qualitative studies, while two were quasi-experimental studies followed by qualitative studies.

2.4.1.2 Country

By continent, there was an even number of study sites between North America (*n*=14) and Europe and the Middle East (*n*=14). One study was undertaken in Africa. By country, eight studies were conducted in Canada, six in the United States of America (USA), four in the Netherlands, two in Belgium, two in Denmark, one in Egypt, one in Hungary, one in Iceland, one in Israel, one in Switzerland, one in Turkey, and one in the UK. There was only one multi-national study, which recruited participants from Canada and the USA.

2.4.1.3 Diseases

All 28 studies included CYP with JIA. The majority (*n*=24) were JIA-specific, two of which were captured within the reviews using obsolete JIA nomenclature. Four studies included other types of RMDs and LTCs. Review articles also included other diseases, but only those studies including JIA were extracted.

2.4.1.4 Participants

Most studies (*n*=25) targeted CYP, with some inclusive of parents. Two targeted parents only, and only one collectively targeted CYP and their families (including parents and siblings). Studies varied in sample size, ranging from 10-333 participants, with an age range of 2–25 years. Of the 22 studies reporting a mean or median participant age, the majority included those in early adolescence (10–14 years) (*n*=12) and middle adolescence (15–17 years) (*n*=6). This was followed by late adolescence/young adulthood (18–24 years) (*n*=3) and middle/late childhood (6–9 years) (*n*=1). No studies with a mean or

median participant age of 0–5 years (infancy/early childhood) were identified. Of the 25 studies reporting CYP's gender, around three quarters (74%) identified as female. Of three studies reporting the gender of parents, around 80% identified as female.

2.4.1.5 Outcome variables assessed

The studies assessed a range of outcome variables using multiple measures to determine the effectiveness of interventions. Grouped by the overall domain of SSM (Table 6), the most commonly assessed outcomes were medical management-related (*n*=93), followed by role management-related (*n*=48), and emotional management-related (*n*=25). Across all three domains, HRQoL was the most commonly assessed outcome variable, the predominant component being the physical domain. Nineteen outcomes were also identified related to intervention acceptability/feasibility. No economic outcomes were identified.

Table 6. Outcome variables assessed within included studies

Outcome variable		***n***
Medical management (*n*=93)		
HRQoL (physical domain)		20
Health outcomes	Pain (including intensity/interference)	14
	Disease activity	11
	Functional disability/ability/status	10
	Strength/dexterity	4
	Fatigue	3
	Stiffness/tenderness	2
	Physical activity/exercise capacity	2
	Extraarticular symptoms	1
Treatment adherence		8
Disease knowledge		6
Disease-related self-efficacy		6
Appointment attendance		2
Self-management/self-care		2
Family disease beliefs		1
Referral to other HCP (psychologist)		1

Outcome variable		*n*
Role management (*n*=48)		
HRQoL (social domain)		13
Self-efficacy		6
SDM/therapeutic alliances/decisional conflict		5
Satisfaction with care		4
Self-management (including self-medication/independent consultation attendance)		3
Educational/ vocational participation	School absenteeism	2
	Wellbeing at school	1
	Participation in physical education class	1
	Perceived occupational performance	1
	Pre-vocational experience	1
Parent influence	Promoting independence	2
	Supporting autonomy	2
	Behavioural control	2
	Psychological control	2
Perceived social support		1
Perception of family support from healthcare professionals		1
Motivation		1
Emotional management (*n*=25)		
HRQoL (emotion domain)		13
Pain behaviour/beliefs/catastrophising		3
Coping		2
Stress		1
Anxiety		1
Depression		1
Behaviour		1
Disease perception		1
Emotional adjustment		1
Impact on family health		1
Acceptability and/or feasibility of intervention (*n*=19)		
Level of interaction/engagement (including with technical aspects)		4
Level of satisfaction/credibility		4

Outcome variable	*n*
Recruitment/participation/withdrawal rates	3
Adherence/commitment/completion of questionnaires	3
Acceptability	3
Usefulness/comprehensibility	1
Intervention deployment	1

HCP: Healthcare professional; HRQoL: Health-related quality of life; SDM: Shared decision making.

2.4.1.6 Data analysis methods

Of the qualitative or mixed methods studies with a qualitative element, the majority (*n*=7) used content analysis. Two used thematic analysis, and one referred to an open coding approach, indicative of grounded theory, though this was not explicitly described. One study did not report the analysis employed. Aside from descriptive statistical methods, a variety of inferential statistical methods were reported in quantitative studies (Appendix 7).

2.4.1.7 Methodological quality

The methodological quality of studies and reviews is presented in Appendix 5 and 6. The median (interquartile range) score was 80% (80–100%). Some of the key issues identified from quantitative studies included: unblinded outcome assessments, confounding variables unaccounted for in study designs and/or analyses, and assessments of the risk of non-response bias. For qualitative studies, although out of scope of the MMAT, the role of reflexivity was not reported. Reflexivity is an important concept, since it mitigates the social interaction component of the researcher-participant relationship, which is an underlying threat to the accuracy of qualitative research outcomes (Jootun et al., 2009). However, some of these inadequacies may not necessarily suggest a lack of methodological rigour, and may be due to author oversight or publisher-imposed character limitations.

2.4.2 SSMI characteristics

2.4.2.1 Format and setting

The majority of SSMIs were delivered with a face-to-face component (*n*=17), followed by online (*n*=11), telephone (*n*=6), smartphone application (*n*=1), and comic book (*n*=1). Some SSMIs utilised a combination of formats, such as face-

to-face plus online, face-to-face plus telephone, or online plus telephone. With regards to setting, a slight majority of SSMIs were delivered within a remote setting (*e.g.*, at home) (*n*=15). This was closely followed by the hospital (*n*=14). Two SSMIs were delivered in a camp/activity centre venue, and one was delivered in an undisclosed local venue.

2.4.2.2 Number, length, and frequency

The number, length, and frequency of sessions offered in SSMIs varied. Where reported, 1–10 sessions took place, lasting from 20 minutes up to several hours over activity days. The frequency of sessions varied from one-off instances, to multiple times per week, weekly, and monthly over the duration of the evaluation. The most frequent SSMI duration periods were 12 months (*n*=4), 12 weeks (*n*=4), eight weeks (*n*=3), and six weeks (*n*=2), with a range of other periods from 30 minutes to three years.

2.4.2.3 Interventionists

Aside from self-guided SSMIs (*n*=8), the most common interventionists were paediatric rheumatologists (*n*=5) and nurses (*n*=4). Other interventionists included transition/local programme co-ordinators (*n*=3), trained peer mentors (*n*=3), non-HCP health coaches (*n*=3), psychologists (*n*=3), unidentifiable role HCPs (*n*=2), VCSE professionals (*n*=2), physiotherapists (*n*=2), psychology research assistants/students (*n*=2), and pedagogical agents (*n*=1).

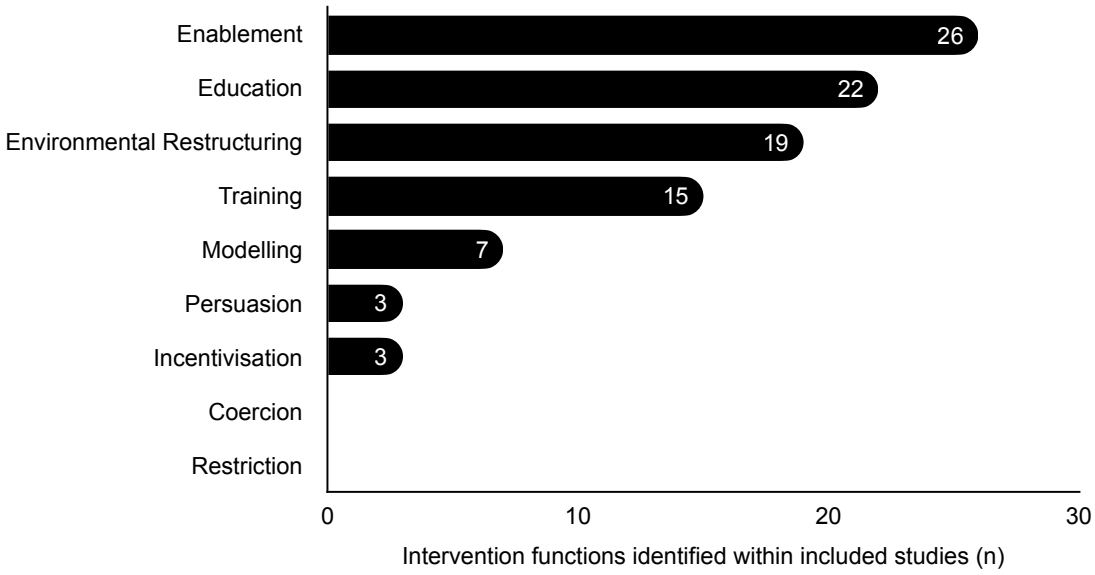
2.4.2.4 Recipients

Most studies (*n*=27) included CYP as recipients of the SSMI, with parents included in over half (*n*=16). Only one study was targeted solely at parents (Svavarsdottir et al., 2020), and only one included siblings as a direct recipient (Burbage et al., 2015). Paediatric rheumatologists were also included as recipients in the electronic PRO intervention by Haverman et al. (2013). Most SSMIs were aimed for individual use (*n*=12) by CYP or parents. Other delivery mechanisms included combined individual/family-based (*n*=8), combined individual/group-based (*n*=5), group-based (*n*=2), and family-based (*n*=1).

2.4.2.5 Functions

SSMIs served a range of BCW intervention functions (Michie et al., 2011). The majority (*n*=26) served two or more functions (Figure 4). The most common was enablement (*n*=26), followed by education (*n*=22), environmental restructuring (*n*=19), training (*n*=15), modelling (*n*=7), persuasion (*n*=3), and incentivisation (*n*=3). No SSMIs used coercion or restriction.

Figure 4. Intervention functions identified within included studies



2.4.2.6 Guiding theories, frameworks, and models

Eighteen studies explicitly identified an underlying theory, framework and/or model which had guided intervention development. Only three theories were identified: Bandura's self-efficacy theory (*n*=1), Bordin's theory of therapeutic alliance (*n*=1), and applied behaviour analytic theory (*n*=1). Four frameworks were identified: the Medical Research Council (MRC) framework for developing, evaluating, and implementing complex interventions (*n*=2), international patient decisions aids framework (*n*=1), Ottawa decision support framework (*n*=1), and peer support within a healthcare context conceptual framework (*n*=1). Seven models were identified: Pender's health promotion model (*n*= 2), breakthrough series model (*n*=1), Canadian occupational performance measure role model (*n*=1), Cox's interaction model of client health behaviour (*n*=1), illness beliefs model (*n*=1), Teens Taking Charge (TTC) conceptual model (*n*=1), and Wright and Leahey's Calgary family assessment and intervention models (*n*=1). Other principles highlighted by studies include: cognitive behavioural (*n*=2),

therapeutic recreation (*n*=1), and thermal biofeedback (*n*=1). One study also utilised the international patient decision aid standards.

2.4.3 SSMI content and components

SSMIs identified were heterogenous in terms of their content and components. The most common type were internet self-guided self-management programmes (with and without social support) (*n*=6). This was followed by cognitive behavioural therapy (CBT) interventions/psychological treatment packages (*n*=4), transition programmes/clinics (*n*=4), decision aids (*n*=3), therapeutic recreation camps/family retreats (*n*=2), internet peer mentoring programmes (*n*=2), telenursing (*n*=2), therapeutic family nursing conversations (*n*=1), comic books (*n*=1), internet-based HRQoL assessment tools (*n*=1), smartphone applications (*n*=1), and video games-based task-orientated activity training (*n*=1).

2.4.3.1 Internet self-guided self-management programmes

The Canadian TTC 12-week internet-based self-management programme consisted of 12 multimedia-based modules for CYP, each taking 20–30 minutes to complete. The modules addressed topics such as the JIA subtypes, diagnosis, treatment, managing symptoms, managing stress, self-monitoring, transition, self-management strategies, and social support. A non-HCP health coach provided weekly telephone support. There were also two modules for parents, focused on the impact of JIA, and strategies for parents to support CYP's effective self-management (Stinson et al., 2010a; Stinson et al., 2010b; Stinson et al., 2020). The weekly telephone support element of TTC was further explored in a pilot study of therapeutic alliance (White et al., 2012).

Connelly et al. (2019) adapted the TTC programme for use in the USA for both English and Spanish-speaking participants. This American adaptation of TTC was similar to the Canadian TTC, in that it was also a 12-week internet-based self-management programme consisting of 12 multimedia-based modules for CYP, each taking 20–30 minutes to complete. However, a restriction was imposed so that no more than two modules could be completed per week. The modules including psychoeducation, cognitive-behavioural coping skills, stress management, and other self-management topics. A non-HCP health coach also provided telephone support, but this was monthly for

three months, with each call lasting no more than 30 minutes. There were also two modules for parents about facilitating their child's self-management skills.

Ammerlaan et al. (2017) developed a 6-week Dutch internet self-guided self-management programme, called 'Change your arthritis', with weekly social support from peer trainers aged 20–30 years with JIA. The programme consisted of three components: a chat section, home exercises, and a discussion board. Once a week, six CYP and two trainers would convene virtually for a planned 90-minute group chat, during which they worked through the following weekly themes: individual capacities, needs, and goals; friends, family and communication; psychosocial impact of JIA and treatment; sport and exercises; relations and intimacy; and having control over life and JIA. In the chats, peer trainers would support CYP in setting goals, practicing how to achieve goals, encouraging questioning and seeking/providing feedback. They also watched real life stories, and played games. After the chats ended, CYP would work through the SSMI and completed assigned exercises, taking no more than one hour per week.

2.4.3.2 Cognitive behavioural and psychological therapy

The Rheumates@Work 14-week internet- and group-based CBT programme aimed to increase physical activity amongst CYP with JIA. Each week, a new topic was introduced through film, animation, puzzles, a spoken text, brain twisters, and/or homework assignments. Topics included JIA health education and physical activity, information on barriers preventing physical activity, explanation of the benefits of physical activity, and self-efficacy towards becoming more physically active. CYP were able to decide whether or not they completed the programme with their parents. The online component included an individualised dashboard and a pedagogical agent called Buddy that guided CYP through the programme. There were also four group sessions with a maximum number of 15 CYP and parents. These were facilitated by various members of the paediatric rheumatology MDT. One chat session was also organised, where staff acted in the place of the pedagogical agent. To facilitate commitment to the plan of action, CYP were invited to sign a declaration of commitment at the end of the first group session, which was rewarded at the last session with a certificate (Armbrust et al., 2015; Armbrust et al., 2017).

Lomholt et al. (2015) delivered a CBT programme, consisting of six, two hour sessions over a 6-week period. The SSMI was manualised, with CYP and parents receiving a workbook, worksheets, and a guide for home practice. The SSMI focused on psychoeducation, relating to pain mechanisms, cognitive restructuring of pain-related negative automatic thinking, and confronting pain-related avoidance situations. Distraction exercises, the exposure ladder, social skills, assertiveness training with roleplay, and re-valuation of family goals were also discussed. Each session followed the same structure, commencing with an evaluation of the homework previously set, followed by a discussion of the session theme, parents leaving the room, a therapist working with CYP, parents reflecting on the homework topic, a therapist working with parents while CYP had a break, before re-joining together to be briefed for the upcoming homework activity.

Rapoff et al. (2002) delivered an SSMI aimed at preventing medication non-adherence in newly diagnosed CYP with JIA. The SSMI, which ran over 12 months, involved one 30 minute session in clinic with a nurse, and 14 telephone calls, initially every two weeks for two months, then monthly for 10 months. The SSMI addressed adherence-enhancing strategies, including monitoring, positive reinforcement, and discipline. These were provided in the form a 10-minute audiovisual programme and a booklet. During the follow-up telephone calls, the nurse reviewed and problem-solved with CYP and parents about adherence improvement strategies.

Lavigne et al. (1992) explored the usefulness of a psychological treatment package for CYP with JIA experiencing pain. The SSMI consisted of six sessions, each 60-90 minutes in length, scheduled fortnightly over a 3-month period. The SSMI covered education and training addressing progressive muscle relaxation, electromyogram biofeedback, thermal biofeedback with autogenic exercises, and operant pain management techniques (parents only). Homework assignments were set after each session for review at the subsequent session.

2.4.3.3 Transition programmes and clinics

The Danish transition clinic, ‘transition for unge i børnereumatologisk ambulatorium’, consisted of two annual appointments over a 3-year period, supplementary and in parallel to routine consultations (Hanghoj et al., 2018).

Each appointment involved same doctor and nurse dyad trained in adolescent medicine and motivational interviewing. Each 60-minute split appointment was held in the afternoon between 14:00 and 18:00, with parents participating in the second half of the appointment only. Topics such as school, friendships, relationships, sex, alcohol and drugs were addressed, as well as any issues CYP wanted to raise. CYP also had an opportunity to meet other CYP with JIA if they desired.

Hilderson and colleagues developed a 5-step brief transition programme (synonymous to a transfer programme) over a 2-year period, starting with an outpatient appointment where a transition co-ordinator was introduced. This was followed six months later by a face-to-face appointment with the transition co-ordinator; an information day for CYP and their families; development on an individualised transfer plan; and the actual transfer from paediatric to adult rheumatology, coinciding with the third face-to-face appointment with the transition co-ordinator. The five steps consisted of eight components: provision of a transition co-ordinator; information and education about JIA, symptom management, medication management, health behaviours, school and friends; telephone availability of the transition co-ordinator for support; information about the adult rheumatology programme, including contact with them; guidance for parents; meetings with other CYP; the transfer plan; and then the actual transfer (Hilderson et al., 2013; Hilderson et al., 2016).

The final programme of transitional care was UK-based and focused on templates for individualised transition plans, designed to reflect adolescent development. These were supplemented by age- and developmentally-appropriate information resources for CYP and families, a local programme co-ordinator, and department transition policies. The intervention was conducted over a 12-month period, including a minimum of three sessions. Once templates were completed at each session, CYP would move onto the next plan in the programme (McDonagh et al., 2007).

2.4.3.4 Decision aids

Brinkman et al. (2017) developed a decision aid consisting of six JIA medication choice cards using plain language and pictorial representations. Each card covered a key issue on how medications differ, including dose frequency, effect time, side effects, cost, length of time to be taken, and other evidence to be

considered before starting, or continuing a medication. The cards were designed to promote SDM, and were accompanied by an instruction sheet, short training video, and summary pamphlet.

El Miedany et al. (2019) developed an SDM aid to facilitate treatment decisions for csDMARDs and bDMARDs. CYP and parents were given 30 minutes in clinic to use the tool. Five visual aids were utilised: emojis to enhance health numeracy; illustrated visual aids to explain treatment targets; virtual risk tools for potential side effects; a visual progressometer to provide some flexibility to customise treatment options; and visual aids for assessing risks and benefits of treatment.

The pain management decision aid, JIA Option Map, was developed as a paper-based prototype to inform an internet-based aid, containing: an assessment of pain and current treatment; a values exercise; a list of 33 treatment options with evidence-based information; and a goal-setting exercise (Toupin-April et al., 2020a; Toupin-April et al., 2020b).

2.4.3.5 Therapeutic recreation camps and family retreats

The Hungarian Bátor Tábor therapeutic recreation summer camp programme took place over an 8-day period and was managed by a VCSE, which took a non-categorical approach to its services for CYP with multiple LTCs (Békési et al., 2011). The programme aimed to provide a free of charge, fun-filled, age-appropriate experience where CYP could acquire activity-related skills. Supervised by trained volunteers including counsellors, it also encouraged the development of a self-sufficient attitude, positive self-esteem, opportunities for a sense of mastery and efficacy in peer relationships, and disease education, through formal learning and informal interaction with peers. The camp focused around challenging CYP to work through their perceived limitations, in a supportive and safe environment.

Another family retreat weekend, held in an American camp-based setting, consisted of lectures and discussions targeted at CYP, their parents, and siblings (Burbage et al., 2015). It aimed to boost knowledge of the whole family, including understanding treatments to assist CYP in coping with JIA symptoms. Small group activities were held to develop family plans to enhance CYP and family coping. These activities were facilitated by VCSE professionals.

Additional activities including camp-based activities to build self-esteem about family members while fostering a sense of group cohesiveness.

2.4.3.6 Internet peer mentoring

Stinson et al. (2016) evaluated iPeer2Peer, an 8-week internet peer mentoring programme previously developed for paediatric chronic pain. The programme consisted of 10 sessions of Skype™ video calls of 20–30 minutes in length, twice per week for two weeks, then once weekly for the remaining six weeks. CYP were sex-matched with a trained peer mentor aged 16–25 and successfully managing JIA. The topics of the conversations were undetermined and unscripted, enabling some individualisation for CYP to discuss topics of importance. However, peer mentors were trained to focus the calls on providing social support and encouraging self-management skills.

2.4.3.7 Telenursing

Ramelet et al. (2017) evaluated a Swiss clinic-home telenursing intervention over a 12-month period. The intervention began with a face-to-face appointment with a doctor and nurse, allowing the nurse to introduce themselves, outline the intervention, and begin to familiarise themselves with CYP and their families. Over the 12-month period, CYP and parents received a monthly telephone call, following a two-part standardised interviewing technique. The first part involved a call description, actions to be taken, and a brief summary of previous conversations and planned actions. The second part focused on the intervention, including eight questions on: school, socialising and everyday life; medication; physiotherapy; occupational therapy; pain symptoms; schedules; administrative issues; and additional topics prioritised by CYP and parents. They were also provided with a telephone number should they need to seek input from the nurse.

2.4.3.8 Therapeutic family nursing conversations

The ‘Family Strengths Oriented Therapeutic Conversation’ intervention was delivered over two sessions, 4–10 weeks apart, by advance practice nurses to parents of CYP with JIA and other LTCs in Iceland (Svavarsdottir et al., 2020). Access to the intervention began within one week of a confirmed diagnosis. The main components of the intervention centred around: establishing a therapeutic

relationship with the parent; drawing a family genogram; exploring the quality of family relationships; encouraging family disease story narratives; asking therapeutic questions; identifying strengths, resilience, and resources; facilitating helpful illness beliefs; challenging constraining illness beliefs; and offering evidence-based information.

2.4.3.9 Comic books

The influence of a comic book titled ‘Neta and the Medikidz explain JIA’ on CYP’s disease-related knowledge and treatment adherence in Israel was explored in another study (Mendelson et al., 2017). The book’s protagonist, Neta, lived with JIA, but struggled to participate in physical activity at school, and found it challenging that her friends did not understand her situation. Using text and illustrations, the Medikidz helped Neta to understand JIA in a way that is not delivered in the clinic, so that she could better communicate with others about her needs and abilities.

2.4.3.10 Internet-based HRQoL assessment tools

Haverman et al. (2013) developed a Dutch internet-based application to monitor HRQoL using electronic PROMs. CYP and parents completed online questionnaires at home via a website before two clinic appointments within a 12-month period. The answers were then automatically converted to a colour coded ePROM profile, to be used by the paediatric rheumatologist on screen during the appointment to focus on identifying, monitoring, and discussing HRQoL issues.

2.4.3.11 Smartphone applications

Lalloo et al. (2021) evaluated iCanCope, a Canadian pain self-management smartphone application, in CYP over 55 days. The application had various features, including a: daily symptom tracker; interactive historical reporting calendar; selectable body maps for pain and inflammation areas; personalised goal setting; disease education and pain coping strategies; and peer-based social support in the form of a ‘question of the week’ forum moderated by the research team.

2.4.3.12 Video games-based task-orientated activity training

Arman et al. (2019) evaluated an 8-week individualised rehabilitation approach known as task-orientated activity training (TOAT), which included intensive training, variable practices, and intermittent feedback. There were two TOAT groups: TOAT in daily living conditions. using items such as clothes and exercise bands); and video games-based TOAT, using Kinect™ for Xbox 360® with a pedagogical agent. CYP completed three, one hour sessions per week for the duration of the study, supervised by a physiotherapist.

2.4.4 SSMI effectiveness

The outcome variables measured varied across the studies (Appendix 7), addressing medical, role, and emotional management domains of self-management. HRQoL was the most commonly assessed outcome the domains.

2.4.4.1 HRQoL

TTC demonstrated improvements in HRQoL domains of problems with pain (F=5.40, ddf=281, P=0.02) and daily activities (F=6.39, ddf=281, P=0.01), as well as treatment problems over time (F=4.94, ddf=281, P=0.008), compared with controls, indicative of the changing difference between groups over time (Stinson et al., 2020). Analysis by Connelly et al. (2019) also found modest improvements in HRQoL from baseline through follow-up (b ± SE=0.37 ± 0.05, β=0.13, t=7.27, P<0.05), though this was comparable amongst those accessing TTC and controls with access to online information only, as indicated by the group by time effects which were not significant. One CBT programme indicated an increase in overall HRQoL (F=1.96, partial η^2=0.12, P=0.18) and JIA-specific HRQoL on the worry (F=4.22, partial η^2=0.22, P=0.06) and communication (F=3.99, partial η^2=0.21, P=0.06) scales in the intervention condition compared to the waitlist condition, albeit not significant (Lomholt et al., 2015). The SSMI based on Pender's health promotion model (Armbrust et al., 2017) and the SSMI based on self-efficacy theory (Ammerlaan et al., 2017) had no effect on HRQoL. A programme of transitional care utilising individualised transition plans had a significant impact on HRQoL, evidenced by an improved adolescent-rated score on the Juvenile Arthritis Quality of Life Questionnaire at 6 and 12 months (P<0.001 and P<0.01, respectively), irrespective of age (McDonagh et al., 2007), as did the brief transition programme (effect size [ES]=0.51) (Hilderson

et al., 2013; Hilderson et al., 2016). The use of a decision aid also appeared to improve HRQoL compared to controls (P<0.01) (El Miedany et al., 2019), as did the internet-based HRQoL application (P<0.01), which flagged potential issues in advance of appointments so that paediatric rheumatologists could increase their focus on discussing psychosocial H&W (Haverman et al., 2013). Meanwhile, the therapeutic recreation camping programme had a significant positive impact on HRQoL, with analysis showing a significant main effect for time (F=3.28, P<0.01) independent of condition, age, gender, and previous camp experience (Békési et al., 2011). Finally, parents in the study by Svavarsdottir et al. (2020) reported a significantly higher QoL (total score) for their family after two sessions of therapeutic family nursing conversations (t=–3.17, df=30, P=0.004), including physical (t=–2.53, df=30, P=0.017) and emotional (t=–2.31, df=30, P=0.028) functioning. Parents also said they were significantly less worried (t=–4.07, df=30, P<0.001) and communicated better as a family (t=–2.60, df=30, P=0.014).

2.4.4.2 Health outcomes

Findings from TTC indicated how self-management amongst CYP with JIA can be foundational to improving health-related outcomes, showing significant improvement in pain intensity (F=5.44, ddf=281, P=0.02) and interference (F=7.40, ddf=281, P=0.007), after adjusting for baseline differences between groups (Stinson et al., 2020), in particular when used over time (Stinson et al., 2010b). Modest improvements in pain intensity (b ± SE=–0.04 ± 0.01, β=–0.08, t=–3.47, P<0.05) and pain interference (b ± SE=–0.04 ± 0.01, β=–0.09, t=–3.99, P<0.05) were also seen in a separate trial of CYP accessing TTC and online information alone, though the group by time effects was not significant (Connelly et al., 2019). Such improvements can be aided by individual contact with trained health coaches, as with TTC (White et al., 2012). In their psychological treatment package, Lavigne et al. (1992) identified a statistically significant trend in improvements in self-reported pain (F=5.72, df=2,14, P=0.02) and more severe pain periods (F=11.81, df=2,14, P=0.02), as well as proxy-reported pain (F=5.32, df=2,10, P=0.03) and more severe pain periods (F=7.00, df=2,10, P=0.01) by mothers. No significant reductions in pain intensity and functional disability (both F=0.06, partial η^2=0.00, P=0.81) were observed in the CBT intervention (Lomholt et al., 2015). Rheumates@Work had

a limited but significant positive effect on physical activity levels (*P*=0.04) and exercise capacity (*P*=0.02) without exacerbating disease status; improvements continued for three months and lasted for 12 months (Armbrust et al., 2015; Armbrust et al., 2017). Interestingly, CYP who started the intervention in winter showed the most improvement. The brief transition programme was shown to improve rheumatology-specific health outcomes including psychosocial health (ES=0.46), treatment (ES=0.33), communication (ES=0.22), pain (ES=0.22), daily activities (ES=0.21), and physical health (ES=0.11) (Hilderson et al., 2013; Hilderson et al., 2016). Meanwhile, El Miedany et al. (2019) identified trends towards better disease activity control in CYP who utilised their SDM aid, though this was not significant. Improvements in morning stiffness (odds ratio [OR] 3.20, 95% confidence interval [CI]: 0.97–7.15) and pain (OR 2.64, 95% CI: 0.97–7.15) were indicated in the telenursing intervention over a 12-month period (Ramelet et al., 2017); while a clinically-meaningful reduction in pain was observed among CYP who utilised the iCanCope smartphone application. However, this did not statistically differ between the intervention and control groups (*P*=0.24) (Lalloo et al., 2021). Finally, both forms of TOAT were effective for CYP's muscle and grip strength (*P*<0.05), with video games-based TOAT being statistically superior to TOAT in daily living conditions for upper limb muscle strength, palmar pinch strength and activity performance (*P*<0.05) (Arman et al., 2019).

2.4.4.3 Treatment adherence and appointment attendance

Rapoff et al. (2002) observed improved treatment adherence amongst newly diagnosed CYP with JIA; however, improved adherence did not have any significant effect on disease activity or functional status (*P*>0.05). Stinson et al. (2010b) indicated how contact with a trained health coach was important for improving motivation and adherence, though no statistical significant differences were found when compared to the control arm for adherence to medication (*F*=0.42, *d*=0.26, *P*=0.52) or exercise (*F*=3.31, *d*=1.11, *P*=0.09). El Miedany et al. (2019) found that CYP who utilised their SDM aid had significantly higher adherence to treatment compared to those without access to the aid (*P*<0.01).

2.4.4.4 Disease knowledge

The comic book significantly improved CYP's disease-specific knowledge (P<0.0001) – the information of which was retained after one year (Mendelson et al., 2017). The authors remarked that improvement in knowledge was greatest among CYP fluent in Arabic. The comic book was able to provide these CYP with a level of information that were unlikely to have received before from their Hebrew-speaking HCPs. Interestingly, Hilderson et al. (2016) did not identify any significant improvements in disease-related knowledge among CYP who participated in their transition programme, only a small positive effect (ES=0.21). They attributed this to its focus, which was on broader experiences and developmental issues, rather than specifically educating CYP about JIA. CYP later confirmed that they did not remember the information received, and had not read the written information provided.

Stinson et al. (2010b) specified that TTC had the greatest observable impact on disease-specific knowledge (F=19.64, d=132, P<0.001), though understanding the longer-term benefits of such interventions on knowledge and subsequent translation to SSM competency would require longitudinal evaluation. The UK programme of transitional care also improved disease knowledge amongst CYP and parents, including continuous improvement over a 12-month period, particularly among younger adolescents (P=0.002) (McDonagh et al., 2007).

2.4.4.5 Self-efficacy and self-management

Although some CYP valued interventions aimed at improving their self-management capacity, this did not always translate into demonstrable improvements in outcomes. This was observed with the Dutch internet-based intervention, 'Challenge your arthritis' (Ammerlaan et al., 2017), where no significant differences between the intervention and control groups were found on self-efficacy at 3 and 6 months (F=2.07, P=0.14). This may have in part been influenced by study design and participant characteristics, such as high baseline scores pre-exposure. It was, however, regarded to influence an active coping style among CYP by sharing experiences, enhancing social support, increasing autonomy, and increasing goal-setting behaviour. Furthermore, CYP in the iPeer2Peer study demonstrated significant improvements in self-management skills (Z=2.11, d=0.72, P<0.05) (Stinson et al., 2016), which also

appeared to positively influenced peer mentors' self-management capacity (Ahola Kohut et al., 2017). Although increases in disease activity were seen in the CBT programme (Lomholt et al., 2015), improvements in adaptive pain cognitions and self-efficacy were reported, but were not significant. These included catastrophising (F=1.67, partial η^2=0.10, P=0.21), control beliefs (F=4.27, partial η^2=0.22, P=0.06), harm beliefs (F=0.29, partial η^2=0.02, P=0.60), disability beliefs (F=1.06, partial η^2=0.07, P=0.32), symptom self-efficacy (F=3.21, partial η^2=0.18, P=0.09), activity self-efficacy (F=3.04, partial η^2=0.17, P=0.10), and emotion self-efficacy (F=3.75, partial η^2=0.20, P=0.07). The summer camp programme also had a positive effect on CYP's self-perception (F=3.31, df=2,112, P<0.05), recognising their generally low self-concept, which when raised, was more likely to be associated with increased coping abilities (Békési et al., 2011). Interestingly, a decrease in autonomy scores in CYP under 14 years was reported, which the authors indicated was a result of CYP maturing beyond the years as a result of their condition, whereas at the camp, they were able to be CYP once more.

2.4.4.6 SDM and therapeutic alliances

Toupin-April et al. (2020b) highlighted how decision aids can facilitate SDM between CYP, families, and HCPs. With the JIA Option Map, it was preferred when potential pain management options were tailored to CYP's preferences, and aided closer discussions with HCPs. Although the included study was a paper-based prototype, the final decision aid would be web-based, enabling options to be individually tailored. This study also highlighted the need to communicate that pain management treatment options are complementary to pharmacological treatment. Importantly, all types of pain management options needed to be presented, including those recommended by clinical practice guidelines, and those not (*e.g.*, complementary therapies), in order to provide evidence for or against their use, to meet expressed needs from CYP and families for such information. Another decision aid using JIA medication choice cards did not appear to enhance SDM (P>0.05) compared to standard care (Brinkman et al., 2017). However, CYP and parents who utilised another decision aid felt that their doctor wanted to know more about their involvement in SDM (P<0.01), and that they were more involved in the selection of treatment options and reached agreement with their doctor on how best to proceed (El

Miedany et al., 2019). White et al. (2012) demonstrated how a therapeutic alliance as part of TTC could be developed in the absence of face-to-face contact. In particular, this study highlighted how such an alliance can be formed between CYP and a trained non-clinical health coach. They also highlighted the perceived sense of anonymity and subsequent level of comfort with a health coach when that support was provided remotely by telephone. The involvement of a health coach was deemed integral to TTC; in particular for motivating CYP and encouraging adherence to the intervention. However, no significant difference was found between this study and a paediatric face-to-face treatment comparison group (*t*=1.37, *P*=0.18, 95% CI: –2.83–14.07) and a paediatric distance/remote treatment comparison group (*t*=1.51, *P*=0.15, 95% CI: –2.01–12.97). Despite the promise of internet-based interventions, the value of human support, even amongst younger generations, was evident (Stinson et al., 2010b). HCPs, in particular nurses, can also play an important and critical role in supporting CYP and their families initially after diagnosis, and during follow-up. Using validated therapeutic family nursing conversations between families and nurses in particular, demonstrated how, over a brief period, family functioning, H&W, and support can be positively influenced (Svavarsdottir et al., 2020).

2.4.4.7 Family illness beliefs and parent influence

Svavarsdottir et al. (2020) demonstrated how therapeutic family nursing conversations enabled parents to have greater conviction about their illness beliefs (*t*=–3.01, *df*=27, *P*=0.006), indicative of their increased confidence in understanding their child's condition and its impact on their family. Parents also reported significantly higher family support after just two sessions (*t*=–5.65, *df*=25, *P*<0.001), including cognitive support (*t*=–4.63, *df*=27, *P*<0.001) and emotional support (*t*=–5.83, *df*=25, *P*<0.001); they also felt that communication in their family improved after they had participated (*t*=–2.60, *df*=30, *P*=0.014). The intervention appeared to benefit parents of CYP of all ages, with greater conviction about illness beliefs seemingly among those with CYP aged eight years and under (*t*=–2.57, *df*=13, *P*=0.023) compared to those with CYP aged 9–17 years (*P*=0.108). Hilderson et al. (2016) found that their brief transition programme had a small, positive effect on shaping certain parent behaviours, such increased autonomy support (ES=0.28), promotion of independence

(ES=0.23), and a reduction in behavioural control (ES=0.21). The authors recognised that changes in parenting behaviours could facilitate CYP's independence, especially since parents of CYP with LTCs can be overprotective. The impact of this overprotection was seen when parents felt the need to monitor their child participating in a peer support programme, which mentors felt negatively influenced subsequent social connections between mentees and mentors (Ahola Kohut et al., 2017).

2.4.4.8 Satisfaction and experience of care

Increasing motivation is an important underlying factor of self-management, exhibited by the use of a video games-based TOAT (Arman et al., 2019). Focussing on the fun of the game, rather than the perception of receiving treatment, highlighted the value of video-based games and serious games more broadly, including their role in incentivising achievement and increasing motivation and satisfaction through interactive elements (P<0.05), as well as visual and verbal feedback. Stinson et al. (2016) also found that CYP were satisfied with iPeer2Peer (mean engagement level of 8.53 out of 10 [standard deviation (range): 1.08 (7–10)]), because they were able to meet and speak to others with JIA who they could relate to, and because peer mentors had already experienced what they were going through. Satisfaction was also markedly improved in the study by McDonagh et al. (2007) at 6- and 12-months for CYP (P<0.01) and parents (P<0.001), respectively. Telenursing support was shown to significantly increase satisfaction among CYP and parents (OR=7.7, 95% CI: 1.8–33.6), through a combination of affective support, health information, and SDM assistance, particularly during the newly-diagnosed period, highlighting the important role played by clinical nurse specialists (CNS) (Ramelet et al., 2017). Finally, from a paediatric rheumatologist perspective, the use of ePROMs increased satisfaction with the level of care they could provide in appointments (P<0.01), providing them with more knowledge, particularly around emotional support, to meet the individual needs of CYP and their families (Haverman et al., 2013).

2.4.4.9 Psychosocial wellbeing and peer support

Peer support can be invaluable for CYP, particularly when provided by slightly older young adults, who are competently self-managing JIA. In their pilot RCT,

Stinson et al. (2016) found that greater flexibility and individualisation is required to tailor peer support to each mentee – each have differing mentoring needs. Conversations often focussed around JIA-related issues, such as lifestyle management, treatment, and future concerns, as well as general life issues (Ahola Kohut et al., 2017). Though in its current form, iPeer2Peer may only be appropriate for certain populations of CYP with JIA who are in need of individualised psychosocial peer support, for example, due to active/refractory disease, or a poor social support network (Stinson et al., 2016). While the benefits of peer support for mentees is often the key focus, mentoring can also have a beneficial impact on mentors, by allowing personal growth, social connection, and positively influencing their own self-management capacity (Ahola Kohut et al., 2017). Within iPeer2Peer, although mentors received intensive training, it became clear that they required further training and support to focus on goal setting with mentees, while being more assertive in scheduling meetings (Stinson et al., 2016). In their brief transition programme, Hilderson and colleagues identified improvements in psychosocial H&W (ES=0.46); however, small negative effects were noted in the reduction in worry (ES=–0.12), and psychological control (ES=–0.20) (Hilderson et al., 2016). One weekend retreat for families living with JIA also appeared to have a beneficial effect CYP, parents, and siblings, with 97% rating the retreat as ‘excellent’ or ‘good’. They felt that the provision of social and educational support to the entire family was important, since it helped to strengthened the family unit, and enabled all of them to learn about JIA. It also helped CYP to realise that they were not alone, and the group setting away from the clinic enabled families to better support each other (Burbage et al., 2015).

2.4.4.10 Educational and vocational participation

Significant improvements, albeit small, were reported for work experience and career advice by McDonagh et al. (2007), which markedly increased with older adolescents at each assessment (P<0.001). Rheumates@Work had a limited positive effect on participation in school (P=0.02) and physical education (P<0.01), without exacerbating disease status (Armbrust et al., 2015; Armbrust et al., 2017). El Miedany et al. (2019) also identified a significant reduction in school absenteeism utilising their SDM aid (P<0.01).

2.5 SSMI acceptability, feasibility, and usability

Usability testing of SSMIs is an important part of their development, to ensure CYP and their families can access, understand, and use the contents of SSMIs, as was demonstrated in TTC (Stinson et al., 2010a). iPeer2Peer went on to highlight the need for flexibility and individualisation of such interventions for CYP, who have different mentoring needs and desires. Examples included differences in the number of calls, and intervention length (Stinson et al., 2016). Armbrust et al. (2015) found that at the end of their intervention, 93.8% of CYP had completely fulfilled all of their assignments for the 14 weeks, demonstrating an understanding of the contents. They attributed this to various factors, namely a motivated group, familiarity with the intervention team which may have encouraged them to feel more committed, and the observation that many CYP had recently participated in a rehabilitation programme. This further suggests that approaches to SSM may need to be multi-pronged in order to provide additive resources across the lifecourse for CYP and their families. Other reasons for this high level of usability included the selection of burdensome topics prioritised by CYP, a signed declaration of commitment, and an interactive pedagogical agent. In their transition programme, Hanghoj et al. (2018) found that many CYP had difficulties understanding the intervention aims, and had poor expectations; highlighting the importance of clear and efficient communication, as well as explicitly setting out expectations at every stage of the process. Those who dropped out of the intervention decided they no longer wanted to participate, without reason; meanwhile those who did not attend said it was because they forgot or were too busy. Other aspects which CYP disliked were the thought of meeting others with JIA in a peer support setting, too infrequent conversations with HCPs, and the practicalities of getting to the appointment. However, they also identified several advantages, including participation without parents, being able to set the agenda, and the ability to build trust with a doctor-nurse dyad who were responsive of their individual needs and preferences (Hanghoj et al., 2018). Hilderson et al. (2013) also felt that a brief transition programme may be more feasible to implement, given estimations that one full-time equivalent employee in the role of transition co-ordinator could take on a case load of 250–300 CYP transitioning from paediatric to adult rheumatology. Although the CBT programme presented by Lomholt et al. (2015) appeared to be acceptable to CYP and their families,

feasibility in general was unclear because of a low participation rate. This could have been due to a number of reasons, namely some families perceiving the intervention as irrelevant due to the pain threshold imposed by the study inclusion criteria. The authors acknowledged that this may not be advisable given the fluctuating nature of JIA symptoms. Brinkman et al. (2017) found their decision aid to be highly acceptable among parents of CYP with JIA, despite not influencing SDM. However, the challenge was the reliable implementation of such a decision aid into routine clinical practice. Meanwhile with the JIA Option Map, CYP and parents opted for a web-based decision aid presenting fewer treatment options consistent with their preference, followed by a discussion with HCPs. They did, however, find the aid useful and wanted to comprehensively understand the evidence underpinning different treatment options, including complementary medicines and nutrition, which are seldom discussed within consultations given some HCPs may feel this is beyond their remit or area of knowledge (Toupin-April et al., 2020b). Lalloo et al. (2021) successfully deployed their smartphone application and found it to be highly acceptable among CYP. Those in the control group who only had access to the symptom tracking features of the application may have engaged more with it since it was the only feature offered within their version of the application. The authors highlighted that of all the resources offered to those in the active arm of the intervention, less than half chose to engage with all of the application features, prompting future work to focus on boosting engagement with daily messages, CYP-friendly aesthetics and algorithm-based suggested content.

2.6 SSMI cost-effectiveness

None of the studies explored the cost-effectiveness of SSMIs or their impact on healthcare utilisation, or indeed utilisation of VCSE services, for example. This is potentially concerning, given that cost-effectiveness analyses are necessary for prioritising H&W expenditure, and the increasing emphasis on self-management by healthcare systems to improve H&W while lowering utilisation in order to minimise waste and reduce costs (Barker et al., 2018). Only one study explicitly mentioned the costs involved with their intervention, stating that the total direct costs of Rheumates@Work amounted to less than € 1,500, with staff management equating to 30 minutes per week (Armbrust et al., 2015). Therefore, further research exploring the SSMI cost-effectiveness is warranted,

as well as a consolidated understanding of overall SSM cost-effectiveness across interventions delivered by public and VCSE services.

2.7 Discussion

Three questions guided the integrative review: the first asked about the characteristics, content and components of SSMIs, the second asked how effective SSMIs are at achieving their anticipated outcomes, and the third asked whether SSMIs are feasible, usable, and acceptable. Findings from the 28 reviewed articles helped to address all three of these questions. The complexity and diversity of SSMIs for CYP with JIA and their families was evident, with study authors using variable terms in reference to SSM, with some failing to explicitly describe their intervention as one aiding SSM, despite measuring relevant SSM outcomes. This highlighted the heterogenous nature of the field, but also a lack of conceptuality clarity, influenced by non-standardised nomenclature, and inconsideration of the lifecourse approach to SSM, likely causing confusion and adding another layer of unnecessary complexity to SSM, in particular for CYP and their families.

SSMIs demonstrated varied levels of significant effectiveness across role, emotional, and medical domains of self-management, consistent with previous findings of their moderate effectiveness (Lindsay et al., 2014). In part, this could have been influenced by study designs and sample characteristics, such as a biased population at baseline, control comparisons, and inclusion criteria. However, the purpose of the integrative review was not to perform a comparative analysis of across different SSMIs; although generating this efficacy data would certainly add to the evidence base, as suggested by the COMPAR-EU study aimed at developing a network meta-analysis of SMI effectiveness high priority LTCs in Europe (Ballester et al., 2020). Lomholt et al. (2015) also highlighted how changes in disease activity, given the variable nature of JIA, could influence CYP's symptom experiences, thus affecting the perceived efficacy of SSMIs under evaluation. Measuring disease activity as well as adjusting for changes is just one example of how more realistic assessment of outcomes in light of specific contexts could be performed in the future.

The review highlighted how SSM can be foundational to improving HRQoL and other role, emotional and medical domains of self-management; though it is

harder to observe distal outcomes and the longer-term impact of SSMIs in time-constrained evaluations. Self-management is a longer-term process – capacity of which does not always translate into improved demonstrable outcomes such as self-efficacy, at least not in the relative short-term. Therefore, it is reasonable to warrant longitudinal evaluations of SSMIs, accounting for a lifecourse approach to SSM.

The review revealed that most published SSMIs focus on medical management, synonymous with adult-focused SMIs, since medical-related tasks are the cornerstone of healthcare – every routine JIA consultation is guaranteed to address symptoms and treatment management. One can view this as the common ground between HCPs, CYP, and families, and the familiarity of the medical domain amongst those professionals, typically HCPs, who have largely been responsible for designing and evaluating SSMIs. This potentially explains why fewer interventions address role or emotional management alone. However, 44% of outcomes assessed identified in the review did fall into the role and emotional domains, and several interventions were aimed at multiple domains (mostly through assessment of HRQoL using comprehensive tools). This indicates a shift in focus towards holistic SSM support. However, HCPs must continue to increase their focus on role and emotional management to ensure that CYP and their families are holistically supported.

SSMIs were heterogenous and although there was no single SSMI characteristic, content, or component that appeared to improve outcomes, there were some common features among included studies. These included face-to-face components, as well as self-guided electronic programmes delivered by professionals and/or peers, and spaces for CYP to participate in interventions, particularly those involving peer and social support, with and without their parents. This is similar to conclusions from a previous review (Kirk et al., 2013), where peer and social support were identified as fundamental SSMI mechanisms for enhancing coping strategies and helping CYP to feel accepted. This review also highlighted the beneficial effect that peer support and mentoring can have on peer mentors, highlighting the evolving process of self-management competency into young adulthood. Although young people are more familiar with, and have greater access to digital forms of social interaction, such as through social media (Orben et al., 2020), the value of human support

for CYP was evident, as it was for parents (Sheng et al., 2019). This is supported by research from Young Minds during the COVID-19 pandemic which confirmed that young people value face-to-face support and do not wish for it to be replaced entirely by digital forms of social interaction (Young Minds, 2020). That is not to say that therapeutic alliances cannot be built remotely, as was seen in some of studies, such as with certain HCPs (*e.g.*, CNS) and health coaches; rather, it appears to be quality of those interactions to support and motivate CYP and their families.

Involvement of parents in the same SSMI as their child also raised a number of important questions, recognising the positive impact of parents on the shared-management of JIA. However, the converse is that parent involvement can itself be a barrier for CYP being able to achieve optimal self-management capacity (Lindsay et al., 2011). Thus, interventions educating and influencing the behaviours of parents are important. Furthermore, a family-focused approach was evidently lacking from included studies, with only one delivering an intervention to CYP, their parents, and siblings. Recognising the impact that JIA has on the whole family, and the importance of supporting and strengthening the family unit is certainly an area for improvement, contextualising CYP's H&W decisions in terms of their broader life experiences and family unit (Clay and Parsh, 2016).

With regards to intervention functions, enablement and education were the most commonly observed, followed by environmental restructuring and training. This indicates that existing SSMIs captured in this integrative review tend to focus on educating CYP and their families about JIA and how to self-manage their H&W, while attempting to provide tools to facilitate SSM behaviours. Changing the environment to make it more conducive to optimum SSM, and the provision of skills-based training were seen, but were less common, indicative of SSMIs operationalised with short-term objectives and goals, despite the lifelong nature of these conditions. Interestingly, very few SSMIs focused on modelling good self-management behaviours, or persuasive and incentivised approaches to engage CYP, despite an understanding that increased motivation can enable CYP to adopt positive self-management behaviours (Service et al., 2014). While education is an important part of SSM, alone it is insufficient in providing CYP with the skills and experiences to become competent at managing their H&W, in an enjoyable and engaging way. This

was indicated in some qualitative studies, where CYP neither read or absorbed the information that they received during SSMIs. In their systematic review in other paediatric disease areas, Saxby and colleagues identified eight educational components useful in enabling CYP to become competent self-managers, encompassing most of the BCW intervention functions. These included: structured and sequenced curricula, reinforcement, active participation, collaboration, autonomy, feedback, multiple exposures, and problem-solving (Saxby et al., 2019).

Intriguingly, some CYP expressed caution and uncertainty about being exposed to face-to-face interventions, including events with peers, which they felt unfamiliar with. Although studies did not explore the rationale against face-to-face interventions in greater detail with these CYP, it may seem logical for remote, and internet-based SSMIs to be useful as a component of a stepped approach to SSM, moving from online to more intensive face-to-face interventions on an individual basis as trust and confidence is built, mitigating against the effect of social anxiety when attending such interventions for the first time, commonly because of the fear of the unknown.

The review identified two relatively large age groups of CYP who are either excluded from interventions or were not accessing them in an evaluative setting. These were pre-adolescents (≤9 years), and late adolescents/young adults (18–24 years). Similar to observations by Lindsay et al. (2014), there were little variation of demographics, with samples consisting mostly of White females. Modest consideration appear to have been paid by authors as to the influence of demographics and some broader contexts on observed results. As Stinson et al. (2016) remarked, young males may prefer less intensive or differently styled interventions to females, to better reflect their attitudes. However, in-depth exploration of the influence of other participant characteristics, such as ethnicity, socioeconomic status, on how CYP and their families respond to SSMIs was not explored, highlighting a fundamental limitation in the design and evaluation of these SSMIs, indicative of their failure to account for the influence of different contexts on outcomes, which has been shown to moderate the efficacy of SSMI in people with LTCs (Hardman et al., 2020). There are many contexts which could influence CYP's awareness of SSMIs in the first place, their subsequent willingness and/or ability to participate, and the outcomes (observed and unobserved) from their

participation. For example, newly diagnosed CYP may not have navigated the acceptance phase of their diagnosis, and so could be unwilling to think beyond their immediate next steps. Whereas, those with active and/or refractory disease may be more motivated to play an active part in managing their H&W in order to achieve some level of normality in their lives.

Although some SSMIs identified in the review appeared to be acceptable to CYP and their families, and were deemed feasible, it is prudent to remember that their effectiveness, feasibility, acceptability, or usability were evaluated in a time-constrained experimental context. This may be considerably different to the real-life settings where such SSMIs would be delivered and used. Implementation challenges were seen in some of the time-constrained interventions, such as the treatment decision aid (Brinkman et al., 2017), stimulating the question of how such interventions would be implemented as part of routine clinical practice. The same study also indicated that acceptability does not necessarily translate into a demonstrable effect on outcomes, such as improved SDM (Brinkman et al., 2017). Overall, participant impressions from included studies regarding acceptability focused on the needed for flexibility, good communication, and individualisation of SSMIs to accommodate specific needs and abilities.

In addition, as has already been mentioned, most studies did not appear to consider the complex confounders at play, such as disease state and severity, and time since diagnosis, as well as the wider context for CYP and their families living with JIA, who shift between interacting with SSMIs and living their everyday lives over time (Craig et al., 2008). Given the complexity of SSMIs and the specific resources available to CYP with JIA, the responses that CYP with JIA make to such resources required further investigation, so as to better understand how SSMIs can be designed and subsequently tailored to meet the individual needs of CYP and their families in light of varying contexts.

2.7.1 Limitations of the review

The key limitations of the review related to the search methods used. Although a systematic search of several bibliographic databases was undertaken to meet the relatively broad inclusion criteria, it was the researcher's intention to identify SSMIs promoting SSM. Consequently, other studies presenting interventions focused on improving specific outcomes (*e.g.*, school attendance), may have

been overlooked. This was not facilitated by inconsistent use of terminology and indexing of articles focused on SSM, as has previously been highlighted in the literature (Grady and Gough, 2014). Steps were taken during the development of the search strategy to mitigate against this issue through the inclusion of multiple medical subject headings and search terms. However, the aim of the review was to identify specific interventions promoting SSM of JIA, and so there can be some confidence that this has been achieved by the range of interventions presented. In addition, while the value of grey literature is recognised, a limited search of the grey literature was performed to keep the retrieved number of records within a manageable number. Although some potentially relevant documents may have been excluded, a further search of grey literature was performed during the first phase of the study presented in Chapter 5. Therefore, it is anticipated that as much relevant data will have been captured as possible within the scope of the aims and objectives of the study presented in this thesis.

2.8 Summary and gaps in the literature

An integrative review of the literature was performed to identify and describe interventions promoting self-management of JIA by CYP, and shared-management of JIA by families; while better understanding the evidence base regarding the SSM of JIA by CYP and their families. The 28 included articles were heterogenous in study design, outcomes assessed, and intervention described, revealing the complexity of SSMIs operating within a complex social system of SSM. Intervention recipients were mostly early-to-mid adolescents and parents, with few SSMIs designed to span the lifecourse from birth to young adulthood, while supporting all members of the family unit. Fewer studies included those in mid-to-late childhood, late adolescence, and young adulthood; with no age- and developmentally-appropriate SSMIs identified for those in infancy and early childhood. Interventions had varied levels of effectiveness across medical, role, and emotional domains, though medical domains were the main focus of evaluation, and role and emotional domains to a lesser extent, mostly through assessing HRQoL. Among the included studies, there was some evidence which suggests that SSMIs impact pain intensity and interference, especially among those SSMIs involving coaches, as well as SDM when interventions included incentivisation functions, which appear closely linked to

motivating CYP and families to be more involved in the SSM of JIA. From the included studies, the most evaluated SSMI was TTC (Stinson et al., 2010a; Stinson et al., 2010b; Connelly et al., 2019; Stinson et al., 2020). Remote enjoyable learning, face-to-face interactions, and professional/peer support were favourably identified among CYP and their families, with remote interventions potentially forming part of a stepped approach to more intensive, face-to-face SSMIs. The vital role of coaches and mentors was evident from included studies, in order to motivate CYP and their families. However, fewer studies used a combination of formats to appeal to a broader group of CYP and families, and similarly, fewer interventions consisted of incentivisation, persuasion, and modelling functions to promote SSM.

A number of gaps were identified in the literature. Firstly, there is a general lack of SSMIs delivered across the lifecourse for CYP with JIA, indicating the need for a shift towards longitudinal evaluation of SSMIs, recognising the longer-term investment in SSM over time. Studies identified also had a varied theoretical underpinning, indicative of the complexity of SSM of JIA. However, there was no common use of a particular theory or model, highlighting the need for further work to explore and illuminate the collective theory behind promoting the SSM of JIA. Secondly, the content and components of SSMIs was heterogenous, with no uniform approach to promoting SSM in this population, indicating the need for a multi-intervention approach to promoting SSM, while also improving equity in access to a standardised suite of interventions for all CYP with JIA and their families to access. Furthermore, few SSMIs appeared to have multi-stakeholder involvement, in particular with peers and VCSEs, despite an awareness of the role of VCSEs in promoting SSM of JIA among CYP and their families. It also remains unclear as to how effective SSMIs are when delivered by different professionals, and whether the role of the intervention provider influences how CYP and families interact with SSMIs, and the subsequent outcomes (observed and unobserved). Similarly, there is insufficient evidence to confirm whether SSMIs have a greater impact on CYP or families. Interestingly, no economic assessments were reported in studies identified in this review, despite recognition of the need for SSMIs to be cost-effective in order to be implemented in routine practice. Finally, studies identified in this integrative literature review generally failed to consider contextual influences on the effect of SSMIs, which are known to influence the

way in which interventions operate to yield outcomes. To address these gaps in knowledge, the following study aims and objectives were developed.

2.9 Aim and objectives of the study

The overall aim of the study presented in this thesis was to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. The specific objectives were as follows:

- To identify possible mechanisms and associated contexts through which SSMIs may promote SSM of JIA by CYP and their families;
- To test and refine IQTs promoting the SSM of JIA by CYP and their families with a range of stakeholders involved in JIA management;
- To develop a preliminary framework promoting the SSM of JIA by CYP and their families, at individual, interpersonal, institutional, and infrastructural levels of context using RQTs tested with different groups of stakeholders.

Chapter 3: Methodology

3.1 Introduction

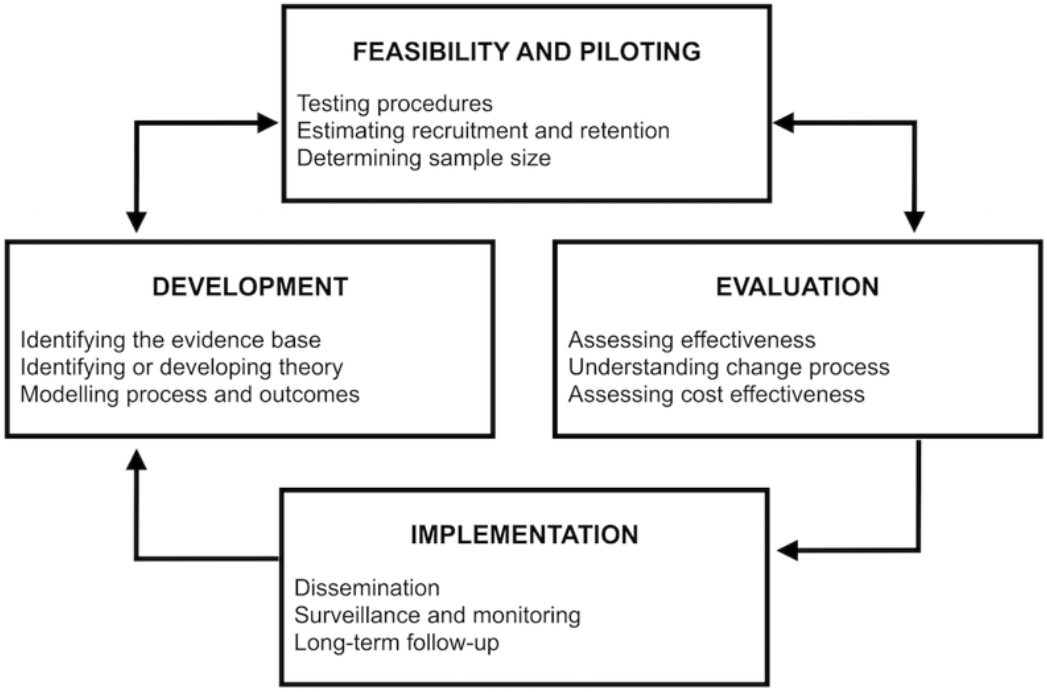
This chapter introduces the theoretical, philosophical, and methodological underpinnings of the study to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. The chapter will argue why qualitative research using a realist approach to evaluation was utilised, and how this was directed by the study aims and objectives.

3.2 SSMIs as complex interventions

Complex interventions are commonly described as interventions containing several interacting components, with a number of additional characteristics (Craig et al., 2008; Petticrew, 2011), including: the number of interacting components within the experimental and control interventions; the number and difficulty of behaviours required by those delivering or receiving the intervention; number of groups or organisational levels targeted by the intervention; number and variability of outcomes; and degree of flexibility or tailoring of the intervention permitted. There are no clear boundaries distinguishing a complex from a simple intervention, though in reality, few interventions can truly be described as simple (Craig et al., 2008). The way in which the characteristics of a complex intervention are dealt with depends upon the aims of the evaluation (Haynes, 1999), though key questions which need to be considered relate to: intervention effectiveness in everyday practice; the active ingredients of interventions; and how the active ingredients of interventions exert their effect. By accepting unpredictability and responding flexibly to emerging patterns and opportunities (Plsek and Greenhalgh, 2001), Michie and Abraham argue that it is the answers to these types of questions which are needed to design more effective and appropriate interventions (Michie and Abraham, 2004). The MRC framework for developing, evaluating, and implementing complex interventions characterised the process of development through to implementation of a complex intervention in terms of the phases of drug development (Tonkens, 2005). Although it can be useful to think in terms of phases in the context of complex interventions, in practice, the process is unlikely to follow a linear or

even a cyclical sequence (Figure 5), indicative of the need to embrace creativity and disorder operating in complex situations (Plsek and Greenhalgh, 2001). By virtue, SSMIs are complex interventions; therefore, the study presented in this thesis attends to the first two steps of the development phase of the framework, in order to identify the evidence base and theory underpinning SSM of JIA by CYP and their families.

Figure 5. MRC framework for complex interventions (Craig et al., 2008)



MRC: Medical Research Council.

3.3 Underpinning middle range theory: The Individual and Family Self-Management Theory

The study adopted a realist approach to the development of a preliminary framework promoting the SSM of JIA, informed by the Individual and Family Self-management Theory (IFSMT) – a middle range descriptive theory proposed by Rodgers (2005) and further discussed by Ryan and Sawin (2009). The IFSMT provides a foundation for expanding the concept of self-management from CYP and their family towards an individual as a member of a social unit (Figure 6) (Ryan and Sawin, 2009). Although there are other candidate middle range theories, such as the Middle Range Theory of Self-Care of Chronic Illness (Riegel et al., 2012); the IFSMT details the diversity of

responsibilities faced by patients and their families by analysing the impact of different individual, family, physical, and environmental factors (*i.e.*, context) on patients' lives. This builds upon the evidence gaps identified in the integrative review presented in Chapter 2 with regards to a limited focus on CYP and family-focused interventions. Moreover, the IFSMT describes self-management as a complex dynamic phenomenon consisting of three dimensions: context, process and outcomes (Fawcett et al., 2001; Meleis, 2011), mirroring a realist approach to evaluation, with process analogous to mechanisms, albeit visible (Figure 7).

Indeed, the IFSMT attends to some of the contextual factors known to affect self-management, and the subsequent linear impact on self-management processes, and outcomes (both proximal and distal). Having been tested and applied in multiple therapeutic areas and populations (including CYP and parents of CYP with LTCs such as type one diabetes mellitus) (Marek et al., 2013; Verchota and Sawin, 2016; Utami et al., 2020), the IFSMT highlights how individual and family-focused interventions can influence SSM by targeting the context and process dimensions of self-management. For example, interventions aimed at influencing the context can help to foster conditions that support SM; and interventions aimed at modulating the process dimension of self-management can enhance people's knowledge and beliefs. It is for this reason why the IFSMT was selected as an appropriate middle range theory underpinning this qualitative evaluation using a realist approach, to further explore how interventions can influence context and process (referred to as mechanisms in the study to capture resources and responses which may evade empirical capture) and subsequent proximal and distal outcomes. Selection of a middle range theory, as opposed to other frameworks or models developed in the area of SSM, is the conventional approach to realist evaluation, in order to bridge the gap between grand theory and clinical practice (Risjord, 2019) by starting and ending with theory at a level of abstraction that can provide explanation of commonalities operating through an intervention in order to improve the generalisability of the study findings (Merton and Merton, 1968).

However, the IFSMT is not without its limitations. Firstly, the IFSMT presents self-management as a linear concept without feedback loops, with context influencing process, leading to proximal and then distal outcomes, with interventions targeted at modulating the context and/or process. The reality of

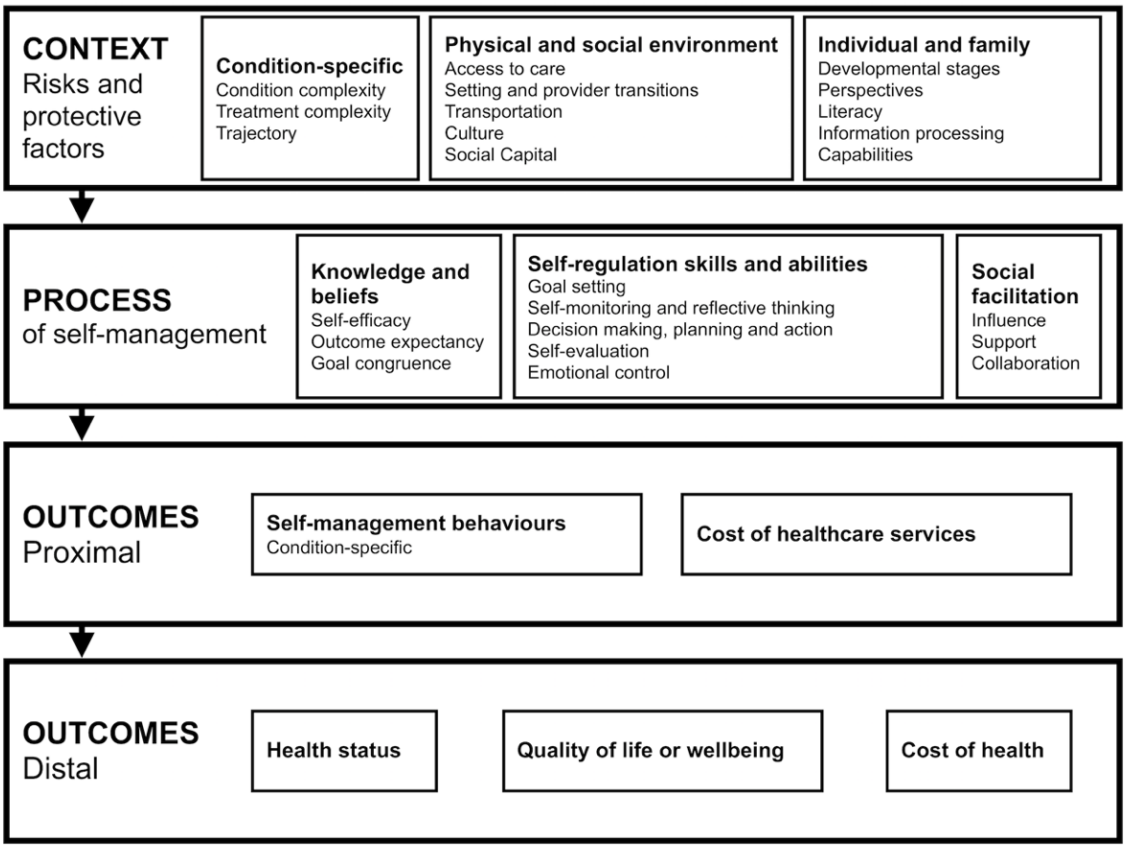
SSM is that many feedback loops are likely to exist, with outcomes in turn shaping context and process variables, given the complexity of SSM. Moreover, it has been suggested that the IFSMT does not fully address all of the complexities associated with SSM, such as a failure to differentiate between received social support and perceived availability of social support, and misaligned outcome variables (Kamp, 2018). Hence in Chapter 5, while the IFMST is used to guide the study, other theories, frameworks, and models inspire the theories which are subsequently tested and refined in the study to inform the new framework promoting the SSM of JIA presented in Chapter 7.

Figure 6. Assumptions of the IFSMT (Ryan and Sawin, 2009)

- Individuals engage in behaviours for personally meaningful reasons that may or may not be directly related to optimising their health status.
- Many factors influence behaviour, including: personal preferences, culture, social norms, and family rules and boundaries.
- Numerous contextual factors affect the ability and desire to engage in SSM.
- Perceptions of resources affect engagement in SSM behaviours.
- SSM involves iterative processes requiring time, repetition and reflection.
- Social facilitation can direct, encourage and support engagement in SSM behaviours and achievement of outcomes.
- Individual/family-centred SSMIs are most effective in fostering engagement in SSM behaviours and achievement of proximal and distal outcomes.
- Concepts of adherence, alliance and compliance contradict SSM, dismissing the notion of individual/family primary responsibility and control.
- Individuals actively engage in SSM by collaborating with professionals in the healthcare system in order to achieve personal health goals.
- Engaging in health promotion behaviours may or may not collaborate with persons in the healthcare system.

IFSMT: Individual and Family Self-management Theory; SSM: Self- and shared-management; SSMI: Self- and shared-management intervention.

Figure 7. IFSMT model (Ryan and Sawin, 2009)



IFSMT: Individual and Family Self-management Theory.

3.4 Underpinning philosophy of scientific realism

Testing of hypotheses, framed in terms of '*what might work, for whom, in what circumstances, and why*' requires a realist approach. Realist evaluation stems from scientific realism, a philosophical approach to social science research. Pawson and Tilley (1997) discuss how scientific realism can help to determine not only which outcomes were observed as a result of interventions, but also how such outcomes are produced, and the influence of the conditions in which interventions take place (Tilley, 2000). This aligns with one of the gaps identified within the integrative review presented in Chapter 2, with regards to the failure of studies evaluating interventions to consider the relationship between contextual influences, SSM processes, and the subsequent influence of SSMIs in yielding outcomes. When developing SSMIs, scientific realism reinforces the stance that real world problems are not always proven by causal relationships inferred from quantitative data (Cornish and Gillespie, 2009; Dalkin et al., 2015). Moreover, it recognises that the goal of evaluation is not to seek to establish universal truths – acknowledging through evaluation that new understandings

come into focus. A realist approach is consistent with the development of complex interventions, offering flexibility to avoid polar paradigms while appropriately blending methods to understand and incorporate stakeholder insights to answer the hypotheses under investigation (Pawson and Tilley, 1997). Furthermore, there is an increased recognition and desire to use theory within H&SC research to facilitate implementation of interventions into practice (Bauer et al., 2015).

3.4.1 Paradigms

According to Weaver and Olson (2006), "*paradigms are patterns of beliefs and practices that regulate inquiry within a discipline by providing lenses, frames and processes through which investigation is accomplished*" (p.460). Paradigms enable evaluators to identify philosophical assumptions informing methodological choices, aiding inquiry to be structured appropriately to address research questions (Weaver and Olson, 2006). In a simplified continuum of positivist-interpretivist paradigms (Figure 8), realism is equidistant (Morgan, 2007) – recognising that human inquiry involves interpretation grounded in empirical analysis (Willig and Rogers, 2017). Table 7 indicates some key characteristics across the paradigms, adapted from Mackenzie and Knipe (2006) and Lawson (2017), which according to Guba (1990), can be characterised by their ontology, epistemology and methodology.

Figure 8. Simplified continuum of research paradigms

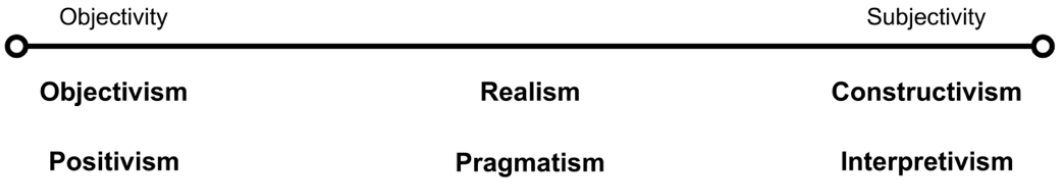


Table 7. Research paradigms, adapted from Mackenzie and Knipe (2006) and Lawson (2017)

	Objectivist/Positivist	**Realist**	**Constructivist/Interpretivist**
Ontology	Reality is objective and exists independently of us	There is a material and social reality that we interact with	Reality is subjective and created by us
Epistemology	Truth and final knowledge exist	Truth and final knowledge do not exist but improved knowledge does	Truth and knowledge exist as what we believe it to be
Methodology	*e.g.*, randomised controlled trial	*e.g.*, realist evaluation	*e.g.*, phenomenological research
Methods	Predominantly quantitative	Qualitative and/or quantitative	Predominantly qualitative
Data collection tools	*e.g.*, experiments, surveys, and scales	Combination of objectivist/constructivist tools	*e.g.*, interviews, observations, documentary analysis
Purpose of data collection and reporting	To describe and analyse the results of controlled inputs-outputs; theory testing by deduction; reporting facts, generalisable knowledge	To accumulate knowledge from theories of causality to inform theories of what works, for whom, in what circumstances and why; generatively theory-driven, by retroduction; reporting transferable theories, accumulating knowledge	To interpret inputs-action-outcomes; theory building by induction; describe participant interpretations, meaning
Causation in interventions	Interventions lead to outcomes	Causality arises from people and their contextualised responses to the resources offered by interventions operating differently in different contexts	Interpretations lead to actions and outcomes

3.4.2 Ontology

Ontology deals with the nature of existence (Rawnsley, 1998), which Bryman (2016) describes as being concerned with the "*nature of social entities*" (p.28). There are two ontological positions: objectivism and constructionism – the former questions whether social entities are "*objective entities that have a reality external to social actors*" (p.28); while the latter questions whether social entities are "*social constructions built up from the perceptions and actions of social actors*" (p.28). Scientific realism is ontologically objectivist (Lawson, 2017), in that social reality exists and has effects, which are largely independent of human cognition, language, perspectives and practices; but rather, interpreted through it. Reality is organised into existing systems which are subject to change.

3.4.3 Epistemology

Epistemology encompasses "*philosophical problems concerned with the origin and structure of knowledge*" (p.3) (Rawnsley, 1998), and is often used interchangeably with the phrase "*theory of knowledge*" (p.3). Epistemological positions should be considered once one has considered ontological position (Bryman, 2016). Within the social sciences, the question is whether the social world can and should be studied using the same concepts as the natural sciences – the epistemological position of which is positivism. The antagonistic epistemological position to positivism is interpretivism, which is founded upon the belief that there are differences between people and the objects of the natural sciences (Bryman, 2016). Scientific realism is epistemologically interpretivist, in that social reality is accessed and understood individually and collaboratively (Lawson, 2017).

3.4.4 Methodology

Methodology is concerned with the steps undertaken for retrieving plausible information (Rawnsley, 1998) – likened to the practice of science. Simplistically, methodology can be viewed as the translation of ontology and epistemology into a system of methods. In the study presented within this thesis, realist evaluation was identified as the most appropriate methodology to guide a theory-driven approach to evaluation (Wong et al., 2013) to aid a better understanding of reality, resulting in a realist approach using qualitative

research methods to identify, consolidate and refine theory promoting the SSM of JIA, to answer '*what works for whom in what circumstances and why*' (Nielsen and Miraglia, 2017). Several scholars have used realist approaches related to SSM of LTCs and SSMIs for people with LTCs (Kousoulis et al., 2014; van Hooft et al., 2017; Desveaux et al., 2018), including intervention guideline development (Powell et al., 2019). In their realist evaluation, Powell et al. (2019) demonstrate how realist approaches are ideally suited for developing complex interventions for complex populations such as CYP with attention deficit hyperactivity disorder, to aid the successful implementation of interventions into practice. However, unlike conventional realist evaluations of a specific intervention, the study presented in this thesis, similar to the guideline development study previously mentioned (Powell et al., 2019), explores the broader phenomena of SSM of JIA, evaluated as a complex social system encompassing multiple interventions operating in other complex systems (Pawson, 2006b). Consequently, the theories developed and tested in the study are question theories, akin to programme or intervention theories, written at a middle level of abstraction to map theory for future research (Westhorp, 2017).

By recognising SSMIs as complex interventions, a realist approach enables an in-depth understanding to be gained of the way in which complex interventions involving human actions may or may not work in real-world situations and social systems that are equally as complex (Miller and Page, 2009). Realist evaluation encourage precision and a depth of analysis which may elude other theory-based approaches to evaluation (Punton et al., 2020), hence its capacity to generate new knowledge, and provide a different perspective on existing knowledge. In particular, its aim is to identify tendencies of outcomes that are the result of different combinations of causal mechanisms, and thus the sorts of contexts that will be most supportive of the introduction of SSMIs. According to Porter and O'Halloran (2012), "*confidence that the most pertinent mechanisms have been identified can be increased through the comparison of different contexts*" (p.19). In the study presented in this thesis, attempts were made to identify as many contextual factors as possible, achieved by including multiple stakeholders groups.

Human societies are complex adaptive systems, whereby characteristics emerge from the collective behaviour of the entire system, as opposed to the behaviour of the system's individual components. In other words, the whole is

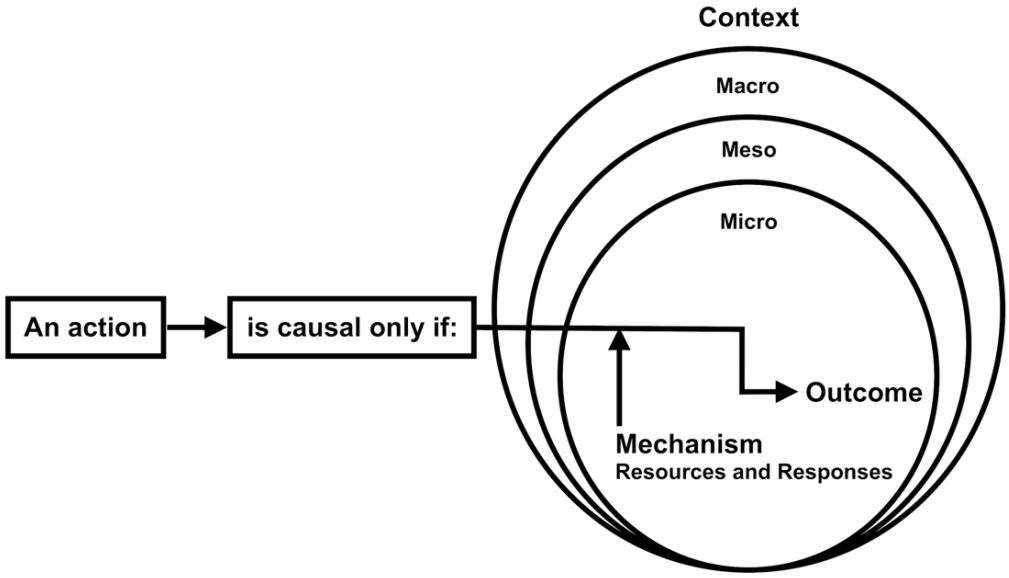
more than the sum of its parts (Eckersley, 2015). The general characteristics of such systems include a large number of elements and interactions, non-linearity of characteristics depicting behaviour, hierarchical structure, unpredictability and self-organisation (Mesjasz, 2010). Change in one part of the system can cause changes in other parts, which may well be non-linear and unpredictable (Eckersley, 2015). This complex reality is interpreted by the individuals within it by identifying underlying mechanisms and identifying how they work under specific conditions (Rycroft-Malone et al., 2012). According to Pawson (2013), policies and interventions are theories of what may happen within the system they create, because "*every programme is a complex system inserted into a complex system*" (p.82). It is only through the workings of entire systems that changes in behaviours and social conditions are affected (Pawson, 2006b).

A realist approach considers the interactions between context, mechanism and outcome (CMO) using a wide range of evidence sources (Pawson and Tilley, 1997). Combining CMO as a question theory is also known as a CMO configuration (Figure 9). CMO configurations seeks to demonstrate the causal relationship between actions and outcomes, describing the influence of context and mechanisms on outcomes. Although Pawson and Manzano-Santaella (2012) describe CMO nomenclature as an "*ugly circumlocution*" (p.183), they defend its utility as a propositional tool to be used in evaluation in order to unearth the underlying workings of interventions:

> *"A [CMO] is a hypothesis that the programme works (O) because of the action of some underlying mechanisms (M), which only comes into operation in particular contexts (C). If the right processes operate in the right conditions, then the programme with prevail. They emphasise the causal and conditional nature of this conjecture… presented formulaically as: C + M = O (sometimes better rendered C + M → O). The action of a particular mechanism in a particular context will generate a particular outcome pattern"* (p.184).

While the evaluation of interventions is not a novel concept, existing research often assumes a positivist approach to evaluation (Bonell et al., 2018), seeking linear causal relationships irrespective of context which may influence such relationships.

Figure 9. CMO configuration, adapted from Pawson and Tilley (1997)



CMO: Context, mechanism, outcome.

Positivist evaluators tend to ask, '*Does intervention X lead to Outcome Y?*'; whereas using a realist approach, evaluators ask, '*What are the mechanisms by which intervention X operate, in which contexts, to lead to Outcome Y?*' By framing questions as CMO configurations, evaluators are more likely to understand the specific conditions under which context-sensitive mechanisms operate, thereby shedding light on when and where interventions may be most appropriate to use (Bonell et al., 2012). Accounting for, and embracing context, as opposed to ignorance or attempting to control context under experimental settings (Bate et al., 2014), is a significant advantage of a realist approach to determining causality – intervention outcomes (O) occur, not because of an intervention *per se*, but because of the manner in which individuals respond (response mechanism [M_2]) when triggered by resources (resource mechanism [M_1]) offered by an intervention, according to the characteristics and circumstances (C) within which an individual operates Pawson (2006b):

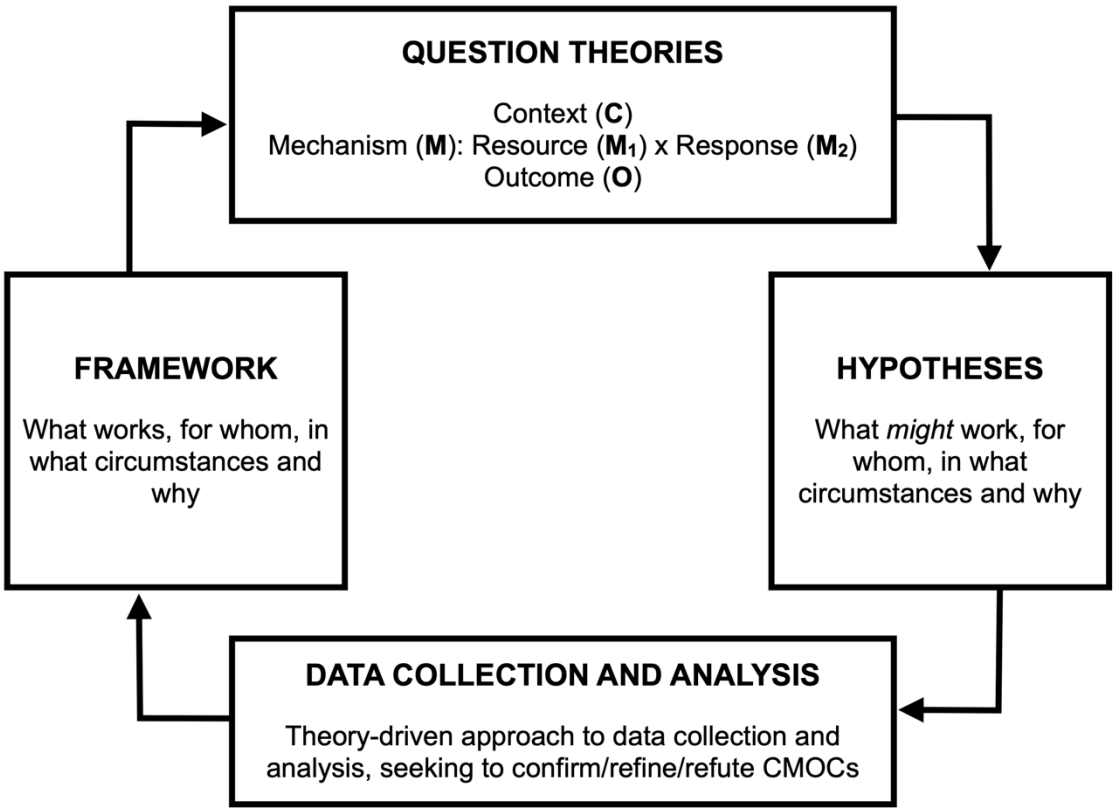
> *"These three locations are the key sources of evidence. In realist jargon the causal connections are established [CMOs]. Although this is a clumsy term it does present a stark contrast with the successionist view, which prioritises the search for outcome regularities"* (p.25).

However, one of the consequences of realist approaches to evaluation is that their "*predictive claims are more modest than those of experimental science*"

(p.19) (Porter and O'Halloran, 2012). This naturally contrasts the applicability and generalisability of findings from experimental evaluation, indicative of the complementary and added value that a realist approach can bring, particularly to the phased approach to complex interventions. Furthermore, the development of CMO configurations can be a difficult process to undertake, in particular when distinguishing between contexts, mechanisms and outcomes (Wong et al., 2017; Greenhalgh and Manzano, 2021). Some scholars also argue that fragmentation of complex phenomena into Pawson and Tilley's CMO configuration heuristic may be seen as oversimplifying the very essence of the topic under study (Marchal et al., 2012; Porter, 2015; De Weger et al., 2020; Haudenhuyse and Debognies, 2021).

Realist approaches typically use research designs underpinned by the same logic of inquiry as that used in other areas of social and natural science research (Pawson and Tilley, 1997). The realist cycle starts with IQTs about how mechanisms are fired in certain contexts to yield specific outcomes. Hypotheses are then derived from these IQTs, in terms of '*what may work, for whom, in what circumstances, and why'*, which are then tested through primary data collection. Data obtained then inform a set of RQTs about '*what works, for whom, in what circumstances, and why*', thus refining theory (Figure 10). A question (or programme/intervention) theory sets out how and why outcomes occur within a specific intervention or intervention area (casually referred to as 'small theory'), compared to mid-range theory ('big theory') and grand theory, which are formulated at higher levels of abstraction (Davidoff et al., 2015).

Figure 10. The cycle of realist evaluation (Pawson and Tilley, 1997, p.185)



CMOC: Context, mechanism, outcome configuration.

3.4.4.1 Contexts

Contexts broadly encompass the circumstances, conditions and/or factors that operate in the background of any intervention and its participants, who will respond upon the resources offered by an intervention if they are conducive (Pawson and Tilley, 1997). Across multiple interventions, there are likely to be a number of contextual constraints, though it is possible to provide a general picture of how contexts work (Pawson, 2006b):

> *"It operates by constraining the choices of stakeholders in a programme. [Participants] are always faced with a choice, but it is both a limited and a loaded one. They have different pre-given characteristics that leave some well-disposed, and some badly disposed, to the [question] theory. They enjoy different pre-existing relationships that leave some well-placed and some ill placed to take up the opportunities provided by the intervention. They come to [interventions] with power, or a lack of it, which enables some to resist and some to embrace the ideas of the [intervention]. There is always choice but it is never a matter of free will. [Interventions] are met with constrained choices, located in pre-existing conditions, and these, as well as the processes internal to the intervention, determine the balance of success and failure"* (p.25).

Westhorp (2011) provide examples of contexts which are "*required to fire the mechanism (or which prevent intended mechanisms from firing)*" (p.8). They are the modifiable and non-modifiable factors which can either facilitate or constrain responses to resources offered as part of SSMIs. Aside from non-modifiable factors such as age, gender, developmental stage and family structure, on an individual level, these may include: CYP's mindset and behaviours, and condition-specific factors related to JIA and treatment. For families, this may include parental anxiety and pre-existing adaptations; while for individual HCPs, this could cover their level of knowledge and interest in SSM.

Pawson (2006b) states that "*interventions are conditioned by the action of layer upon layer of contextual influences*" (p.31) (Figure 11), summarised by at least four contextual layers (Table 8). Reflecting on contexts in this way emphasises the argument that interventions do not work *per se* (Pawson, 2006b); rather, the "*causal powers within the objects or agents or structures*" (p.21) influence change because interventions "*work only if people choose to make them work*" (p.24).

Figure 11. The intervention as the product of its context (Pawson, 2006b)

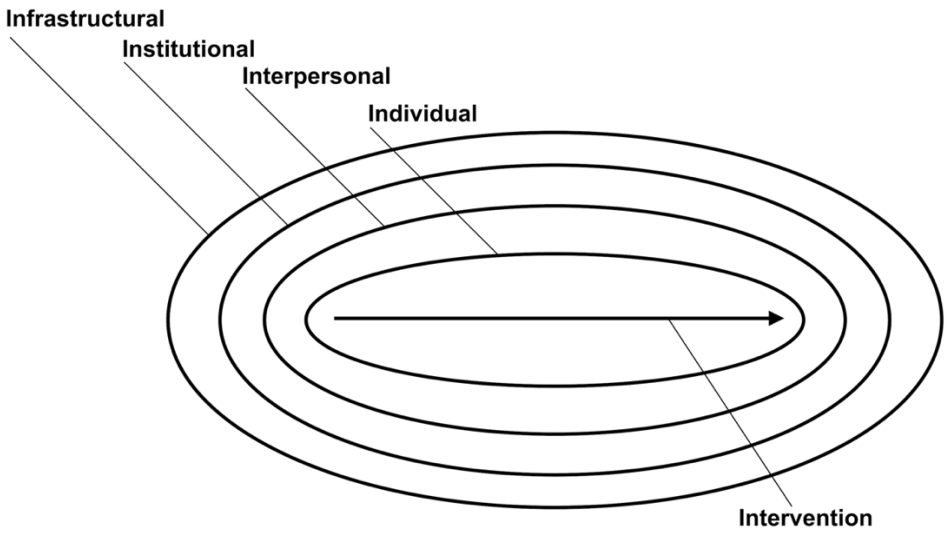


Table 8. The four contextual layers influencing interventions (Pawson, 2006b)

Context	Description
Individual	The capacity of the individuals utilising an intervention, which may include their experience, behaviour and motivations (*e.g.*, CYP with JIA, their families, and individual HCPs)
Interpersonal	The relationships between individuals utilising the intervention and those supporting the intervention (*e.g.*, relationships between CYP with JIA, their families, HCPs, VCSE professionals, and education professionals)
Institutional	The operational setting in which either individuals are based, or the intervention has been developed and implemented, which may include culture, character and ethos (*e.g.*, hospital, school, VCSE facilities)
Infrastructural	The strategic influences underpinning the intervention at a policy and evidence level (*e.g.*, SSM guidance, H&SC provision, VCSE provision, educational policies)

CYP: Children and young people; H&SC: Health and social care; HCP: Healthcare professional; JIA: Juvenile idiopathic arthritis; SSM: Self- and shared-management; VCSE: Voluntary, community, and social enterprise.

While Pawson's four contextual layers influencing interventions aid in stratifying contextual complexity (Pawson, 2006b), it is opportune to reflect on the hierarchy of healthcare to frame how organisational and policy-level discourse command and control the actions of professionals supporting CYP with JIA and their families. While acknowledging the command-and-control thinking (Ham, 2014) guiding decision-making and H&SC provision to a degree, ultimately, it is the CYP with JIA and other patients who represent the majority in the system whose needs the system serves and whose preferences the system should reflect. Systems should exist to support CYP with JIA and their families; rather than nesting them at the base of a top-down hierarchy.

Furthermore, one of the challenges in a realist approach to evaluation is distinguishing between contexts and mechanisms, which can sometimes overlap or interchange, depending on the circumstances. For example, one person's context may well be one person's mechanism, adding to the complexity of reality. Interestingly, while many have focused on context as being static and fixed, Bate et al. (2014) identify context as a dynamic and fluid process, synonymous to the constantly changing seas and seasons.

3.4.4.2 Mechanisms

Mechanisms embody the essence of SSM – it is where SSMIs exert their effect, captured by responses to available resources (Lee et al., 2019). Unlike processes, mechanisms are often invisible and ontologically deep, hidden from empirical capture (Westhorp, 2018; Jagosh, 2020). While resources can be identified at individual, interpersonal, institutional and infrastructural levels to a degree, responses are associated directly with people individually, and through their interpersonal relationships (Pietromonaco and Collins, 2017). At the individual level, SSM resources may include: different provision and styles of education best suited to CYP and their families. Meanwhile, social facilitation and support through negotiation and an open environment; familiar, trusted and relatable intervention facilitators; and individualised, CYP- and family-focused care and support encompass the engagement between CYP, their families and other professionals involved in their care may be at an interpersonal level. At institutional and infrastructural level of organisations, evidence and policy, individualised, CYP- and family-focused care and support may be a core resource. With regards to responses from CYP and families, knowledge and belief about managing JIA, self-regulation skills and abilities, and trust with and amongst others may be the anticipated responses at individual and interpersonal levels.

3.4.4.3 Outcomes

Outcomes from SSMIs must be met on an individual level for CYP with JIA and their families, so that their efforts in engaging with SSMIs are not wasted (Lindsay et al., 2014). Outcomes may either be: intended and realised; intended and unrealised; unintended; or unanticipatable (Jagosh, 2020). Given the complexity of SSM, and the impact of interventions in the short- and longer-term, in the study presented in this thesis, outcomes are grouped as proximal and distal (Arah et al., 2005), so as to capture the more immediate, ‘quick wins’, potentially of most importance for CYP and families, as well as the long-term, aspirant outcomes of living well with JIA into the future. For CYP and families, proximal outcomes are also grouped by role, emotional, and medical management domains (Bal et al., 2021), so as to capture the physical and psychosocial components of living with JIA which SSMIs largely target. Distal outcomes of interest may include health status, QoL, and educational

attainment. At an institutional and infrastructural level for H&SC providers, proximal outcomes of interest may include H&SC utilisation, while distal outcomes may include cost of health. Meanwhile, for VCSEs, service utilisation may be a proximal outcome, while for education providers, attainment may be a distal outcome of interest.

3.4.5 Types of realist methodology

Unlike methodologies aligned with the positivist and interpretivist paradigms, realist inquiry uses retroductive theorising to overcome the deficiencies of inductive and deductive reasoning to offer causal explanations (Meyer and Lunnay, 2013), which may take the form of middle-range theories. Fundamentally, there are two types of realist methodology, connected by their theory-driven approach, but distinguished by the source of data under interrogation: realist evaluation focuses on primary data, while realist synthesis (also referred to as realist review) explores secondary data sources (Pawson, 2013). Regardless of the approach, both types of methodology are robust and involve the testing and refinement of theories (Bonell et al., 2012). In the study presented within this thesis, an approach to realist evaluation was used involving primary data collection with a range of stakeholders.

3.5 Summary

This chapter has explored the rational and choice for taking a realist approach to exploring how SSM of JIA can be promoted across the lifecourse, aimed at identifying the key contextual influencers conducive for SSM of JIA by CYP, their families, and professionals involved in their healthcare, wellbeing, and education; and their potential influence on generative mechanisms – thus answering the question: '*What works for whom, in what circumstances, how and why'.* The theoretical, philosophical, and methodological principles underpinning the study were discussed, utilising realist evaluation and the IFSMT to partially address the development step of the MRC Framework for developing, evaluating, and implementing complex interventions. Chapter 4 of this thesis goes on to outline the methods used in the study.

Chapter 4: Methods

4.1 Introduction

This chapters outlines the methods used to address the study aims and objectives, which are re-visited below. The chapter begins with an overview of the study, followed by the methods of sampling, recruitment, data collection, and data analysis. Steps to ensure quality and rigour are discussed, including reflexivity, before concluding with a summary of ethical and research governance considerations, and how these were addressed.

4.2 Aims and objectives of the study

The overall aim of the study presented in this thesis was to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. The specific objectives were as follows:

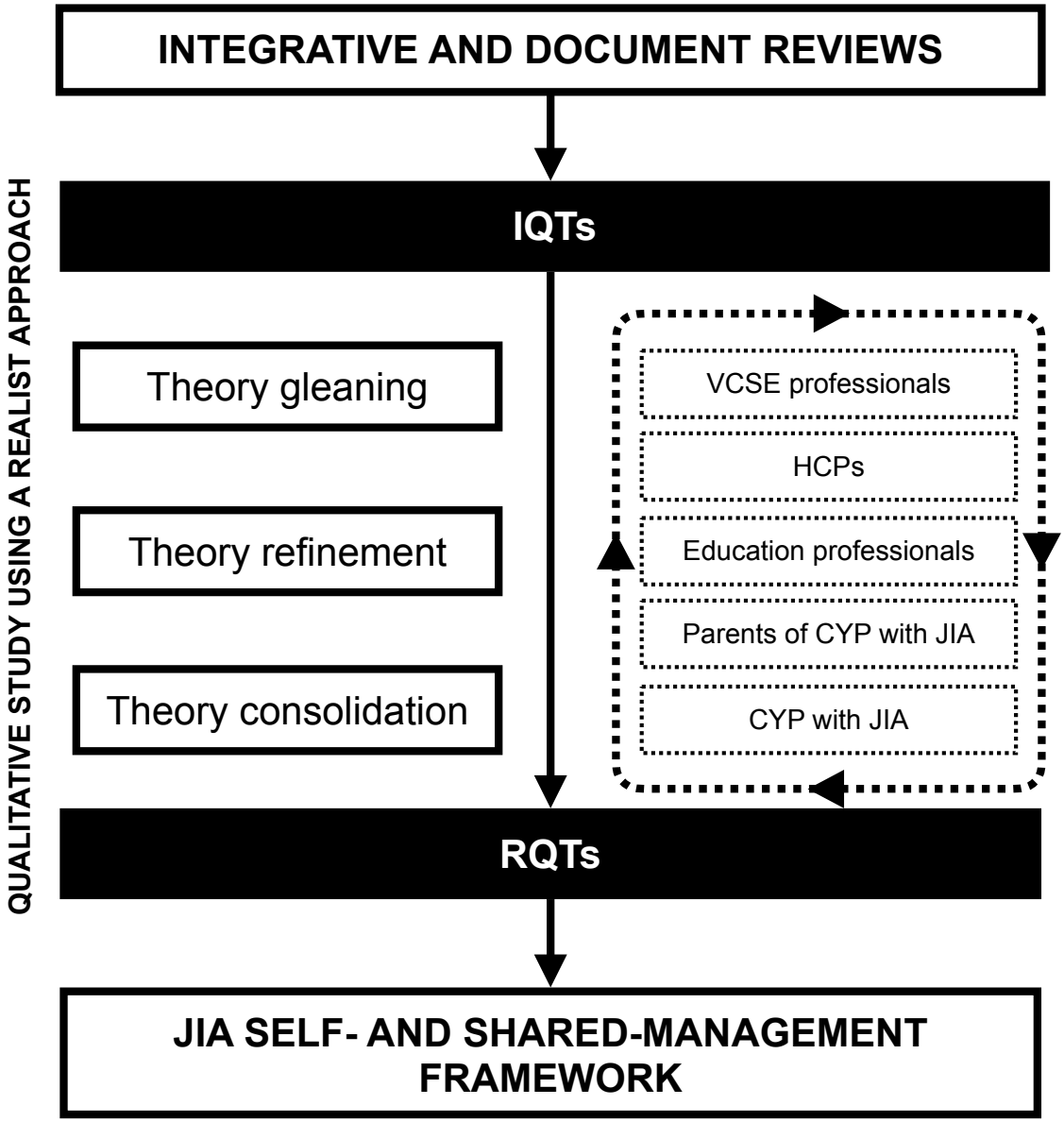
- To identify possible mechanisms and associated contexts through which SSMIs may promote SSM of JIA by CYP and their families;
- To test and refine IQTs promoting the SSM of JIA by CYP and their families with a range of stakeholders involved in JIA management;
- To develop a preliminary framework promoting the SSM of JIA by CYP and their families, at individual, interpersonal, institutional, and infrastructural levels of context using RQTs tested with different groups of stakeholders.

4.3 Study overview

A study overview is shown in Figure 12. The study used a realist approach to evaluation, beginning with a series of IQTs, presented in Chapter 5, which were developed from the integrative literature review presented in Chapter 2, and a document review summarised in Chapter 5. These were tested through primary qualitative data collection involving a three-phased process of gleaning, refining and consolidating theory (Manzano, 2016), resulting in a set of RQTs. Although interviews are presented linearly in Figure 12, this was not necessarily the order in which different stakeholder groups were interviewed, given the iterative nature of data collection. The RQTs were then assimilated into a preliminary

framework defining the SSM of JIA by CYP and their families, which is presented in Chapter 7. The approach to realist evaluation in the study was guided by the RAMESES II reporting standards for realist evaluations (Wong et al., 2016), orientated by the 20-item checklist to be included when reporting realist evaluations.

Figure 12. Study overview



CYP: Children and young people; HCP: Healthcare professional; IQTs: Initial question theories; JIA: Juvenile Idiopathic Arthritis, RQTs: Refined question theories; VCSE: Voluntary, community, and social enterprise.

4.4 Sampling

A range of different stakeholders were involved as participants in the study, to develop a comprehensive perspective to developing RQTs, in an attempt to accurately reflect the current situation regarding SSM of JIA by CYP and their families.

4.4.1 Determining the sample size

There is widespread agreement among qualitative methodologists that there are no set number of interviews that can be assumed to achieve data saturation (Patton, 2005; Given, 2008). However, common professional practice suggests that the acceptable number of interviews for conventional qualitative studies is 20–30 participants (Mason, 2010), accounting for attrition throughout the study (Teddlie and Tashakkori, 2009). However, this rule of thumb is not strictly reliable for studies using a realist approach, since realist hypotheses are not confirmed or refuted through data saturation (Strauss and Corbin, 1990); rather, through relevance and rigour (Manzano, 2016). Substantial amounts of data were needed, even though the sample was relatively small, to be able to move from constructions to an explanation of causal mechanisms. Since the unit of analysis in realist evaluation is not the person, but the events and processes around them, every unique participant uncovers a collection of micro events and processes, each of which were explored (Manzano, 2016).

4.4.2 Sampling technique

Purposive sampling with a realist lens was used as the primary sampling strategy. Purposive sampling is the most commonly used sampling technique in qualitative research (Marshall, 1996), as it facilitates the identification of a sample that reflects the research aims, while striving for maximum variation. In realist terms, the sampling technique was theory-driven, since fragments of evidence and information from different stakeholders were explored, interpreted and pieced together (Emmel, 2013) in order to uncover patterns and regularities about the SSM of JIA. Snowball sampling was also used to identify other relevant stakeholders with a relationship to participants who were eligible to participate (Marshall, 1996); such as other family members and colleagues. Convenience sampling was also utilised for self-referring participants through the internet and social media who met the inclusion criteria (Taherdoost, 2016).

4.5 Recruitment

Participants were recruited through several mechanisms, including: 1) a children's hospital in the North of England; 2) the study's website and social networking platforms; and 3) communication with VCSEs. To minimise inconvenience and to be as flexible as possible to meet participants' needs and preferences, data collection was arranged at mutually convenient times and locations, depending on their availability. Participants were provided with at least 24 hours to review the information about the study that they had received (Appendix 9), prior to providing informed assent or consent (Appendix 10) and arranging a time to proceed with data collection.

4.5.1 Sample identification

Two key strategies were used to identify participants. Firstly, using purposive sampling, CYP aged 5–16 years with JIA were identified by means of a patient database held by a paediatric rheumatology service at a children's hospital in the North of England. Suitable participants were discussed at the MDT's weekly meeting, with two consultant rheumatologists and a CNS acting as gatekeepers to identifying and screening appropriate participants per the study's inclusion criteria. Parents were contacted initially through having a child on the database by the clinical team. They then received an approved study pack containing a letter of invitation, a poster, an information sheet (Appendix 9), an assent form and a consent form (Appendix 10). Those expressing an interest by returning the enclosed consent and/or assent forms by email or in stamp-addressed envelopes provided in the invitation pack were then contacted by the researcher, and interviews were arranged. Recruited parents were free to share information about the study with other families interested in taking part if they wished to do so. HCPs were recruited using purposive sampling that aimed to achieve maximum sampling variation regarding professional disciplines and roles. Contact was made with staff members suggested by participating HCPs, so as to gain a broader picture by including as many MDT perspectives as possible. All potential participants were provided with either a print or digital study pack including a letter of invitation, poster, information sheet and a consent form. Interviews were arranged with those who signed and return the consent form.

Secondly, snowball and convenience sampling were used with self-selecting participants (including families, HCPs, education professionals, and VCSE professionals) who encountered the study through publicity of the study poster via: the internet; social networking platforms; VCSE websites; VCSE-administered mailing lists; and word of mouth. Upon contacting the researcher, they received a relevant electronic study pack, including a letter of invitation, poster, information sheet and a consent form. Interviews were then arranged with those who signed and return the consent forms. When study information packs were provided to prospective participants, they were recommended to take at least 24 hours after receiving the invitation to consider the study, providing them with the opportunity to ask questions to the researcher before providing informed consent.

4.5.2 Eligibility criteria

The inclusion criteria for the study was as follows:

- CYP aged 5–16 years at the time of recruitment with a diagnosis of JIA;
- Parents of a child with JIA;
- Other family members related to a child with JIA;
- HCPs from the paediatric rheumatology MDT with experience of caring for CYP with JIA;
- Education professionals, including teachers and school support staff, with experience of supporting CYP with JIA and/or other LTCs;
- VCSE professionals working in the UK with a remit of informing, educating, supporting, and/or advocating for CYP with JIA;
- An ability to speak, and understand English given resource limitations within a PhD study;
- For face-to-face interviews, to be located within the North of England;
- For virtual interviews, to have access to a landline or mobile telephone and/or access to the internet to use video conferencing software.

4.6 Unearthing IQTs for evaluation

Question theories encompass the assumptions and perspectives of those responsible for designing and implementing interventions – the intervention actors (Pawson and Tilley, 1997). They can be defined as the set of

assumptions, often implicit, that describe how intervention actors explain how they expect an intervention to achieve its objective(s) (Mukumbang et al., 2016).

Explaining what intervention actors think about SSMIs for CYP people with JIA and their families means seeking an understanding of how stakeholders involved in such interventions cognise the influence of various contextual factors, what mechanisms are in action, and how SSMIs would trigger the activation of these mechanisms under specific contextual conditions, thus leading to a set of outcomes, intended and unintended (Jagosh, 2020). Since interventions work differently in different contexts and through different mechanisms, interventions cannot simply be replicated from one context to another, and be expected to achieve the same outcomes. However, theory-based understandings about '*what works, for whom, in what circumstances, and why*' are transferable (Wong et al., 2016).

However, identifying question theories to subsequently test with participants was not straightforward. Within evidence sources, question theories were rarely labelled as such, and statements of the characteristics underpinning a question theory were seldom provided. Therefore, in addition to the integrative review in Chapter 2, a more applied definition of question theory (Pawson et al., 2010) was used while 'digging for nuggets' (Pawson, 2006a) in a document review to inform the IQTs outlined in Chapter 5. Pearson et al. (2015) described these applied definitions as: ideas about what is going wrong; ideas about how to remedy the deficiency; ideas about how the remedy itself may be undermined, and; ideas about how to counter these counter-threats.

In order to track IQTs, after an assessment of the conceptual-richness of sources (Appendix 11) (Ritzer, 1991; Roen et al., 2006), a table was constructed to record sources (Appendix 12), in light of the convoluted process of eliciting question theories, Generally, quantitative evaluations tended to discuss why an intervention worked (or why it did not); while qualitative studies usually extracted question theories during their discussion. However, this was not always the case, given that some evaluations had a strong theoretical basis, and some qualitative studies considered their findings only in the context of comparative work. Consequently, the way to accommodate the irregularity in reporting of underlying question theories was to scrutinise all potential sources, using retroduction to unearth potential causal mechanisms (Jagosh, 2020).

4.7 Linking theory and causality as a process

As IQTs were unearthed, Table 9 summarises the way in which theory and causality are linked throughout the study, using retroductive theorising to unearth latent, activated and actualised mechanisms (Jagosh, 2020).

Table 9. The process of linking theory and causality

<table>
<tr><td colspan="2">What works, for whom?</td><td rowspan="5">Emerging from and evaluated through data, retroductively</td></tr>
<tr><td colspan="2">IQTs</td></tr>
<tr><td colspan="2">In what circumstances and why?</td></tr>
<tr><td>Themes and CMO configurations</td><td rowspan="2">Informed by substantive and middle-range theory</td></tr>
<tr><td>RQTs</td></tr>
</table>

CMO: Context, mechanism, outcome; CYP: Children and young people; IQTs: Initial question theories; JIA: Juvenile idiopathic arthritis; RQTs: Refined question theories; SSM: Self- and shared-management.

4.8 Data collection

Two key methods of data collection were utilised in order to meet the needs and preferences of participants, as far as possible. These included semi-structured individual interviews (Ritchie and Lewis, 2003) and dyadic interviews (Sohier, 1995), including family interviews (Eggenberger and Nelms, 2007). In total, 17 semi-structured interviews were conducted. There were 14 individual interviews, and 3 dyadic interviews with participants who shared a pre-existing relationship (one parent-child dyad, one parent-parent dyad, and one VCSE professional dyad of colleagues employed within the same team). The overall number of participants was 20. Although face-to-face and virtual interviews opportunities were offered to participants, all interviews were conducted virtually either by telephone or video conference, whichever was most convenient and familiar for participants. Given the location of most participants in relation to the researcher, virtual interviews were the most viable possibility. For remaining participants eligible for face-to-face interviews, prevailing government guidance on social distancing during the COVID-19 pandemic resulted in interviews being conducted virtually.

Although dyadic interviews are a relatively new addition to qualitative methods of data collection (Morgan et al., 2016), they offer a number of advantages over individual interviews. Firstly, the multidimensional perspective which can be obtained from dyadic interviews aids the researcher in developing

a more realistic, and complete picture of a given topic or scenario, increasing the credibility of the data collected (Sohier, 1995). Furthermore, interaction between participants in dyadic interviews enabled in-depth concepts to be built upon or contrasted through dialogue (Morgan et al., 2016), sometimes without little prompt from the researcher. However, this could also be problematic, especially in a realist evaluation, when the researcher wants to provide a more directive style of questioning around tentative theories. At times during dyadic interviews, the researcher allowed participants the time and space to discuss the topic at hand between themselves, while prepared to bring the conversation back to the tentative theories under investigation. There is also the concern of participants disclosing too much information which may cause conflict within the dyad, as well as participants disagreeing, which the researcher anticipated for by always bringing the conversation back to the theories being presented in interviews, rather than lived experience. Similarly, the researcher was consciously aware of the potential for participants, particularly CYP and parents, to become emotionally overwhelmed when recounting their lived experience (Melville and Hincks, 2016). While such instances did not occur in the study, the researcher was poised to either wait for participants to regain composure, or suggest rescheduling the interview if participants preferred to do so.

Similarly, virtual interviews have their benefits and challenges. While they overcome geographic limitations and can be more convenient for participants and the researcher to conduct, they do restrict face-to-face interaction. Moreover, not all participants have access to virtual devices and internet or cellular connectivity; therefore, virtual interviews could unintentionally exclude potential participants. The researcher attempted to mitigate this concern by enabling participants to select their preferred interview method, prior to the COVID-19 pandemic which forced data collection to become exclusively virtual. However, it is worth recognising that virtual methods of data collection have gained traction in recent years, in part due to increasingly familiar with video conferencing software as a result of the COVID-19 pandemic (Roberts et al., 2021). Drabble et al. (2016) indicated that telephone interviews as a virtual method of data collection are effective and often preferable for participants when talking about emotional experiences. Indeed, participants may respond differently over the telephone than they would in a face-to-face setting. This could present potential advantages with regards to the amount and quality of

data collected if participants felt more comfortable sharing information; however, the opposite effect could also be seen if interpersonal communication is a challenge, resulting in a lack of trust between the researcher and participant (Block and Erskine, 2012). Participants also seem to be willing to engage in lengthy interviews by telephone, as was also seen in the study, when participants are made to feel engaged and heard. In the absence of non-verbal cues over the telephone, filler sounds and words such as “uh-huh” and “yeah”, respectively, served as cues that the researcher was listening, or the participant was ready to move on to the next point. Other obvious benefits of telephone interviews include cost effectiveness and time efficiency (Block and Erskine, 2012). However, telephone interviews (and other forms of virtual interviews to an extent) can limit the tools available to participants in an interview – for example, access to visual aids which may be used in a face-to-face setting.

4.8.1 Building rapport with participants

The researcher established a dialogue with participants prior to the interviews taking place, either by email or telephone. In the participant information sheet, all participants were made aware of the reasons for the research. During the start of each interview, the researcher attempted to develop rapport between himself and participants, to strengthen the sense of trust and understanding (McGrath et al., 2019). The researcher introduced the study, and himself, which for transparency, included his role as a postgraduate, ‘expert by experience’ researcher. The researcher openly acknowledged his experience as a patient advocate, and his diagnosis as a child with JIA, which had motivated him to complete the study. The researcher had previously engaged with eight participants in other roles; thus a rapport was already established, enabling dialogue to flow more naturally. Participants were also asked a series of general questions to ease them into the interview, before focusing on the IQTs using age- and developmentally-appropriate discussion guide prompts (Appendix 13), which evolved and were adapted in light of ongoing analysis as the study progressed. Throughout the study, the researcher continued to acknowledge the potential role of bias and interviewer assumptions. These were mitigated through continuous reflection and the use of observational notes (Phillippi and Lauderdale, 2018), the use of a literature-informed discussion guide, a realist approach to interviewing with participants using the teacher-learner cycle,

including bringing the conversation back to the tentative theories, and regular communication with supervisors to address any areas of concern.

4.8.2 Teacher-learner cycle approach

Qualitative interviews used an approach from realist evaluation referred to as the teacher-learner cycle (Pawson and Tilley, 1997). Unlike conventional qualitative interviews, in realist interviews, the researcher presents theories under investigation to participants, with the interview then directed by the researcher to 'test' the hypothesis contained within the presented theories (Manzano, 2016). Realist interviews purposely gather additional data to confirm, refute or refine aspects of IQTs (Manzano, 2016). During interviews using the teacher-learner cycle approach, potentially relevant data about aspects of theories that were not previously identified, may be unearthed. Although IQTs were presented to participants supported by a semi-structured discussion guide (Appendix 13), which was developed after the IQTs were drafted, participants were questioned about tentative theories in an open and inclusive manner, in order for them to take a leading and participatory role in guiding the direction of the discussion. An innovative use of the teacher-learner cycle was used in the study when interviewing participants in dyads. To the researcher's best knowledge, this has not previously been conducted. This dynamic enabled the researcher to present tentative theories to both participants, who would test theories between themselves, as well as with the researcher. The teacher-learner cycle was planned to be adapted for use with CYP, using creative approaches to visualise tentative theories in an age- and developmentally-appropriate manner. In face-to-face settings, this would involve presenting a series of visual prompts to CYP (shared in advance of the interview), before allowing CYP to select a preferred method of communication to share their thoughts (*e.g.*, drawing, or role play).

Furthermore, although stakeholders were not members of the research team, they played an important role in the development of theories. Unlike some research studies where participants are simply viewed as the researcher's 'subjects', realist approaches to evaluation enable stakeholders to be key informants, recognising the power and worth of their knowledge (Patton, 1999). Taking a realist approach meant it was important that participants had experiential knowledge of managing JIA; however, it was also important that the

researcher had a firm understanding of the topic area, and entered interviews armed with hypotheses to guide the data collection process. Indeed, Manzano (2016) recommends that the researcher arrives at interviews with an in-depth knowledge of what happens in the natural setting for participants. Unlike traditional qualitative interviews where the researcher is often described as someone friendly and intelligent but lacking the knowledge of the specific situation, realist interviews embrace and use the knowledge of the researcher in helping to guide the discussions that take place, and thus the refinement and consolidation of theory. Therefore, the researcher was well placed to do this, given his lived experience of JIA since childhood, combined with his international advocacy and research experience in the field.

4.8.3 Theory gleaning, refinement, and consolidation

Manzano (2016) suggests that data collection is a seamless, three-stage process of gleaning, refining, and consolidating theory, as the researcher adjusts and shapes the interview, keeping theory as the common denominator. Theory gleaning began at the start of data collection, where participants supported the researcher in articulating the first order theories, derived from the IQTs. These interviews began with the 'architects' – the participants most familiar with the institutional and infrastructural tenets of SSM, including VCSE professionals, HCPs, and some parent advocates in the JIA community. As theories were further refined and consolidated, parents, CYP, and teachers were interviewed to further 'test' and 'refine' the theories, as well as with other VCSE professionals and HCPs.

Data collection was iterative in light of ongoing analysis, similar to other studies using a realist approach (Randell et al., 2020), with additional interviews conducted to refine and consolidate evolving theories. However, no repeat interviews were conducted with the same participants. Unlike conventional qualitative interviews, realist interviews are used as a means to explore theories, and not just as a means to an end (Hammersley, 2008). One of the purposes of interviews was to build knowledge of variations in what happens in natural settings, with this knowledge contributing to the construction, testing, and refining of theories throughout the study.

4.9 Data analysis

4.9.1 Management of data

Interview data were audio recorded, downloaded and transcribed using verbatim, either by the researcher or through an external, University of Leeds approved transcription service. Interview transcripts were then imported into the computer assisted qualitative data analysis software (CAQDAS), NVivo12, for analysis (Bazeley and Jackson, 2013). The transcripts were checked by the researcher against the original audio recorded interviews to ensure they accurately reflected the data collected. Observational notes made during data collection were also entered into NVivo for analysis. CAQDAS such as NVivo12 aid researchers in unearthing accurate and transparent interpretations of qualitative data while providing an audit of the analysis process (Welsh, 2002), and has been used previously for studies using a realist approach (Dalkin et al., 2021). Data saturation was not considered relevant for the aim of the study.

4.9.2 Hybrid deductive-inductive thematic analysis

The researcher began data analysis by reading the transcripts and immersing himself in the data, subsequently analysed using a hybrid deductive-inductive thematic analysis (Fereday and Muir-Cochrane, 2006). This approach was selected because of its integration of theory-driven deductive analysis through an *a priori* lens of IQTs, together with data-driven inductive analysis. This is consistent with realist approaches to evaluation, which use deductive approaches to validate theory; while also providing an opportunity for new or refined CMOs to emerge from the data inductively.

Initially, the researcher intended to analyse the data using a CMO lens; however, unlike some studies involving a realist evaluation of a specific intervention, the study presented in this thesis used a realist approach to promote SSM in JIA. As a result, a large amount of data was collected, and an approach was needed to balance breadth with depth. After an attempt to analyse a sample of transcripts using a CMO lens, the researcher realised that analysis using a CMO lens (*i.e.*, coding for individual C, M, and O) would result in an unmanageable amount of fragmented themes. Therefore, a decision was made to code transcripts using a question theory lens, which others have also applied to their work (Dalkin et al., 2021), recognising that it is near impossible to uncover every possible CMO configuration (Pawson and Tilley, 1997).

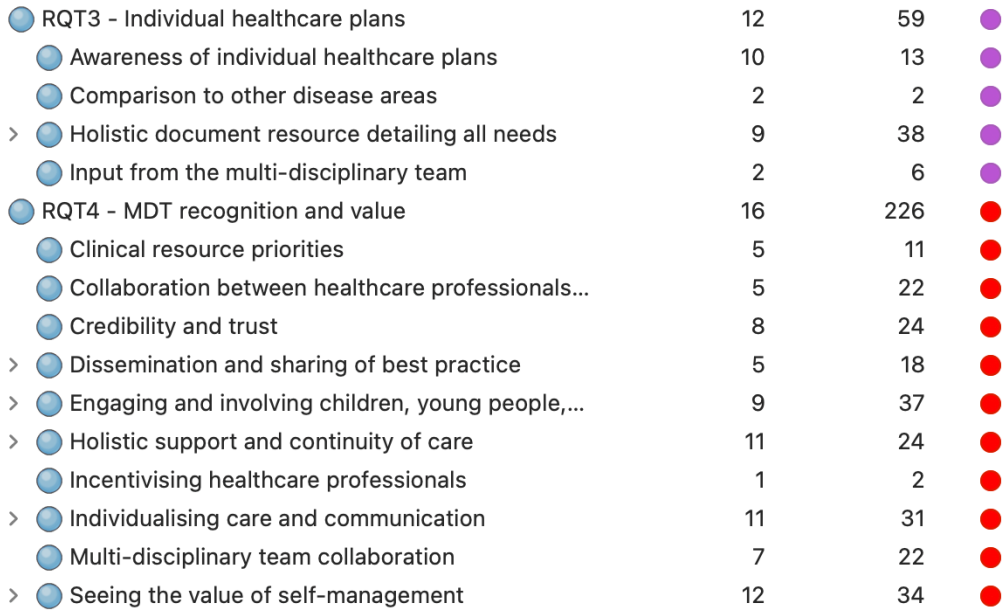
Subsequently, the IQTs outlined in Chapter 5 were used as a set of *a priori* themes and applied initially to a sample of transcripts to test the approach. The initial theories were summarised, and applied across the entire data set, with IQTs coded as 'parent nodes'. This broad, initial approach aided the researcher to become familiar with the entire data set, while looking for connections across transcripts, keeping in mind the goal of developing RQTs linking context with mechanisms and outcomes.

Data were then analysed using an inductive analysis approach (Braun and Clarke, 2006), to identify concepts proposed by participants within IQT 'parent nodes', to confirm, consolidate, or refute question theories. Nodes were colour coded to aid easy identification of which question theory data corresponded with (Figure 13), and annotations within NVivo12 were used to capture the thought processes behind themes coded from the data set. Data analysed inductively were thematically coded as 'child nodes', resulting in a coding tree of question theories and inductive themes containing CMO configurations. As themes were inductively analysed, annotations were made within NVivo12 to theorise the connection between context, mechanisms, and outcomes within each theme under each question theory. Paper annotations were also used to aid the researcher in identifying connections between context, mechanisms, and outcomes within emergent themes. Themes were then reviewed and similar conceptual nodes were combined into a final set of themes. The researcher was the only data coder; however, the coding tree was shared and discussed as it evolved with his supervisors to corroborate the process of moving between IQTs and RQTs. Findings were constantly compared with the IQTs to determine whether they support, refute, or suggest a revision or addition to the question theories by exploring the different CMO components associated with the SSM of JIA – with retroductive reasoning used to move between initial and emergent themes (Jagosh, 2020); thus preserving connections within the data to assist in understanding causality (Maxwell, 2012).

As data analysis unfolded, it became increasingly clear that one question theory was redundant, as themes in the remaining question theories were corroborated and consolidated. During the process of theory refinement and consolidation, these themes were allocated to other question theories, while searching for the connected threads linking context to mechanisms and outcomes. The final stage of abstraction took place as the themes identified

under each question theory were collectively condensed into a set of CMO configurations within each of the RQTs. These findings are discussed thematically in Chapter 6 under their respective RQT.

Figure 13. Exemplar parent and child nodes during analysis in NVivo12



MDT: Multi-disciplinary team; RQT: Refined question theory.

However, thematic analysis is not without limitations or criticisms. The researcher's role can be highly subjective, risking the introduction of bias, especially in the process of generating codes and identifying them thematically within the data. However, the use of a hybrid approach to thematic analysis helped to address such concerns by applying deductive analysis of themes identified within the literature, together with emergent data from participants. Furthermore, credibility was established through broad discussions throughout the study with patient and parent research partners, colleagues, and supervisors, and through including quotations from all participants as part of the analytical process to support or refute the refining question theories.

4.9.3 Framework development

Once theory was gleaned, refined and consolidated, RQTs were integrated into a preliminary framework promoting SSM of JIA, presented in Chapter 7. This included: the most important mechanisms that need to be 'triggered' by SSMIs; and the contexts related to these 'key' mechanisms. Ultimately, the key SSMI

strategies capable of recognising contextual factors in such a way that 'key' mechanisms are triggered were presented.

4.10 Patient and public involvement and engagement

Patient and public involvement and engagement in the design and conduct of research can positively impact the quality, appropriateness and understanding of research (Brett et al., 2014). As previously highlighted, the researcher has lived experience of JIA, and conducting the research as an 'expert by experience' researcher. Throughout the study, he seeked involvement from patient and parent research partners, in addition to a meeting with a CYP's advisory group (Preston, 2018), who collectively assisted in the following tasks:

- Identifying and developing the aims of the research, to address gaps in information, education, and support for CYP and their families;
- Selecting the study's acronym, title, and branding;
- Identifying suitable methods of data collection;
- Developing and reviewing recruitment materials such as the participant information sheet;
- Reviewing the interview discussion guide;
- Identifying appropriate dissemination strategies of research findings, including as plain language summaries through social media.

Unfortunately, the limited funding associated with a PhD study meant that the researcher was unable to achieve some of his goals regarding patient and public involvement, such as training CYP and other patient and parent research partners to be co-researchers.

From an engagement perspective, study updates were shared in addition to information about JIA and research more broadly, via a dedicated website and three social networking platforms – Facebook, Instagram, and Twitter. Furthermore, the researcher together with patient and parent research partners hosted a public activity and awareness stall at the Be Curious event in March 2018, hosted by the University of Leeds.

4.11 Ethical considerations and research governance

Consideration of ethical dilemmas and concerns is an integral component of all forms of research, irrespective of the methodology and specific methods used

to acquire data (Guillemin and Gillam, 2004). In their ethics policy, the University of Leeds provides guidance to aid the identification and management of ethical issues, relating directly to the rights of participants and the obligations of researchers (University of Leeds, 2019b). In the context of the study presented in this thesis, the key ethical considerations from the outset included: obtaining informed consent and assent to participation; anonymity, confidentiality and data management; protection of participants and the researcher from harm; the balance of risk and benefit; and transparency and conflicts of interest. When applying these principles to research related to CYP, there are particular issues which need to be carefully considered with greater attention (Phelan and Kinsella, 2013), requiring a sensitive and individualised approach, recognising CYP's right to participant in, and benefit from research (Unicef, 1989).

4.11.1 Ethical approval

The UK policy framework for H&SC research across the four devolved nations states that all research conducted with people through H&SC services must meet ethical standards, and should only start if a research ethics committee (REC) have favourably reviewed the research proposal or protocol and related information (NHS Health Research Authority, 2020). NHS Health Research Authority and Health and Care Research Wales ethical approval for the study presented in this thesis was provided by the London – Surrey REC (18/LO/1087) on 10 August 2018 (Appendix 14). The University of Leeds acted as the sponsor for the study. One non-substantial amendment requesting an extension to the study end date was approved on 25 February 2020.

4.11.2 Research governance approval

Research governance approval was required for the researcher to be able to commence the study at the participating NHS Trust operating as a participant identification centre (NHS Health Research Authority, 2021). The researcher was issued with occupational health clearance, and an enhanced Disclosure and Barring Service certificate, after completion of good clinical practice training (NHS Health Research Authority, 2020). Research governance was granted with a letter of access on 06 September 2018 (PA18/110968).

4.11.3 Informed assent and consent

The University of Leeds' informed consent protocol was adhered to throughout the study (University of Leeds, 2019a). All participants received an invitation to participate which they were able to keep for their information. Documents for participation were also available to participants in several ways, including online, from the researcher, or from the participating NHS Trust. Participants were reminded that they could ask questions of the researcher at any point, that their participation was voluntary, and that they had the right to withdraw from the study at any time without giving a reason, which would not affect their clinical care. Permission for the use of direct quotes was obtained, and agreement was sought for anonymised information collected during the study to be used in future research. In addition, in the event of a disclosure, participants were informed of the safeguarding process the researcher would need to follow. Participants had at least 24 hours to review the information about the study that they have received, prior to providing written, informed assent and/or consent. This was obtained from participants and was stored securely in accordance with the study's data management plan. CYP aged ≤16 years were required to have consent from their parents; however, their written assent was also obtained, respecting the rights of CYP to information and the choice of participation (Council for Disabled Children, 2011). Importantly, informed assent and consent was viewed as an ongoing process throughout the study, ensuring participants were provided with sufficient information about the research to make an informed choice over time.

4.11.4 Protection of participants and the researcher from harm

Safeguarding the physical and psychosocial H&W and safety of participants and the researcher is one of the most important ethical considerations of any study. The researcher received training related to safeguarding and child protection. Details were also provided to parents about breaking confidentiality in the event of a safeguarding concern with CYP, following an agreed process, beginning with an assessment as to whether there was evidence that CYP were experiencing or likely to suffer significant harm, with a 'reporting a concern form' used to document the safeguarding concern. However, this was not required to be implemented.

4.11.5 Maintaining anonymity and confidentiality

When considering any study to be ethical, maintaining anonymity and confidentiality is an important consideration from the outset (Corti et al., 2000). The approach to anonymising data was informed by the anonymisation code of practice (ICO, 2012). Data was classified using the University of Leeds data protection code of practice, ensuring data access and restrictions were appropriate to the level of confidentiality. The nature of interviews meant that responses were not anonymous to the researcher; however, procedures were in place to ensure the confidentiality of such interviews, unless there was a safeguarding concern. In addition, participants' responses were anonymised, so that participants were not identifiable from their data. Real names were replaced by alphanumeric codes and pseudonyms during enrolment.

The researcher complied with the requirement of the General Data Protection Regulation and Data Protection Act 2018 with regards to the collection, storage, processing, and disclosure of personal information. Ethical and legal guidance was followed, with all personal identifiable data handled in strict confidence. The purpose of collecting this information was to communicate with participants, once they had agreed to take part. Any personal identifiable data, such as consent forms, were kept in a locked filing cabinet in the researcher's secure office at the University of Leeds, in duplicate to electronically-scanned copies stored on an encrypted University of Leeds server. Similarly, audio data were downloaded and saved to the encrypted server after each interview, before permanent deletion on the audio recording device. Participants were reminded that personal identifiable data would be permanently deleted at the end of the study, while assent/consent forms would be treated as essential documents, retained electronically on the encrypted University of Leeds server for six years post-study completion (HM Revenue & Customs, 2019). Physical copies of assent/consent forms were permanently deleted at the end of the study, simultaneously with other personal identifiable data outlined above. With specific reference to the recruitment of CYP and parents from the participating NHS Trust, only members of CYP's existing clinical team had access to their clinical records, who followed the NHS Confidentiality Code of Practice (Department of Health, 2003).

4.11.6 Data management

From the outset, a data management plan was created, to assist in the process of collecting, managing and storing of physical and digital data collected during the study. As previously detailed, security arrangements for the storage of personal data during the study included physical storage that was secure and accessible only to the researcher, and digital storage on University of Leeds encrypted servers. Only the researcher and his supervisors had access to any data stored digitally, and the researcher had received appropriate data management training. For the purposes of access to digitally stored data after the end of the study, the researcher named his lead academic supervisor as the contactable data custodian. Permission for data sharing was obtained during the assent and consent processes, including whether participants were willing for their anonymised data to be published on the Research Data Leeds Repository for future use (University of Leeds, 2021b). This was considered an ethical approach to data collection, given that a lot of valuable data was collected during the study with a variety of different stakeholders. During the ethical approval process, it was agreed that this data would be stored on the Research Data Leeds Repository for 10 years after the study ended, per the repository deposit agreement (University of Leeds, 2021a). For the purposes of transcription of audio data recorded during interviews and focus groups, bespoke data sharing agreements were produced when University of Leeds suppliers of transcription services were used.

4.11.7 Transparency and conflicts of interest

Transparency and the declaration of potential conflicts of interest were made explicit in the study from the outset, including during the process of seeking ethical approval. The researcher identified as an ‘expert by experience’ researcher conducting a piece of user-led research. User-led research implies that individuals with lived experience of health conditions are responsible for designing, conducting and evaluating research (NIHR INVOLVE, 2018), thus ensuring that the voice of those individuals is embedded into the research agenda (Rose, 2015). Furthermore, user-led research undoubtedly aids the establishment of anti-authoritarian relationship between the researcher and participants (Råheim et al., 2016), enabling participants to feel valued and at ease with the researcher. This approach is also entirely appropriate for realist

approaches to evaluation, given that an insider perspective is valuable for exploring and developing theory that can be applied and translated into practice (Manzano, 2016). In the process of demonstrating transparency, the researcher described his background, while demonstrating his capacity and capability as a competent and professional researcher. In addition, procedures were in place from the outset to ensure the study was credible (Creswell and Miller, 2000). These included extensive training modelled on the Vitae Researcher Development Framework (Bray and Boon, 2011), the maintenance of a risks register, regular supervision contact with senior, clinically-trained researchers, collaboration with practising HCPs, and consultancy with patients and parents research partners.

4.12 Reflexivity

Råheim et al. (2016) advocates for the practice of continuous reflexive awareness during the conduct of research using qualitative methods to aid confirmability, stating that "*a lack of critical awareness about the impact of the research context, perspectives chosen, methodological choices made, and… the presence of the researcher, might seriously hamper the knowledge claims made*" (p.10). The researcher continuously reflected on his role throughout the study, which informed the approach to conducting the study as an impartial but informed researcher capable of understanding JIA. He also reflected on interpersonal and subjective processes within interviews, including regular discussions with supervisors, clinical collaborators and patient/parent research partners. As an 'expert by experience', the researcher was acutely aware of his privileged position. He was transparent with participants regarding his experience, but emphasised both in the participant information sheet and during the start of each interview that he was acting in the role as a researcher for their interaction, rather than a patient advocate. His in-depth understanding of JIA and SSM enabled him to foresee when narratives may become too focussed on a topic beyond the remit of the study, so that he could intervene and bring the conversation back to discussing the question theories at hand. Furthermore, some argue that the researcher could be too involved in the topic to provide an objective, and valid analysis of data. Having anticipated this as a potential challenge, the researcher implemented a series of steps to ensure this was not a concern. To ensure credibility of the data, the study had a clear focus and was

designed with the intended focus on the theory underpinning the SSM of JIA. Using a realist approach, the focus of data collection and analysis was always 'the theory', informed by the literature and refined by expert stakeholders. Observational notes were also recorded during each interview to capture key pieces of information, and the relationship between participants and the researcher. In addition, a reflexive diary was used throughout the entire study. This diary contained records of all decisions made and the rationale for those decisions, as well as processes the researcher followed, reflections on the study progress, assumptions that were uncovered during data collection and analysis, along with other general thoughts and remarks. In addition, to further ensure credibility, several forms of triangulation (Flick, 2004), including data, theory, and multidisciplinary triangulation, were used, and review of the process and findings of the study provided by academic supervisors assist in addressing the issue of dependability. The researcher also reflected on the diversity of the participants in his study. Although data on ethnicity, sexuality, and socioeconomic status were not collected during recruitment, the majority of participants were female. The need to increase diversity in research has been widely discussed (Willis et al., 2021), with health inequality and disproportionate disease burden having been spotlighted recently during the COVID-19 pandemic. Indeed, in the study, the diversity of participants is unclear, especially those non-HCPs. Steps to improve the diversity of participants could have included a wider recruitment strategy to target participants who do not typically engage with research, embedding a quota into the inclusion criteria to strive for maximum diversity, and patient/parent research partners trained as co-researchers to facilitate the recruitment process within their own communities and networks.

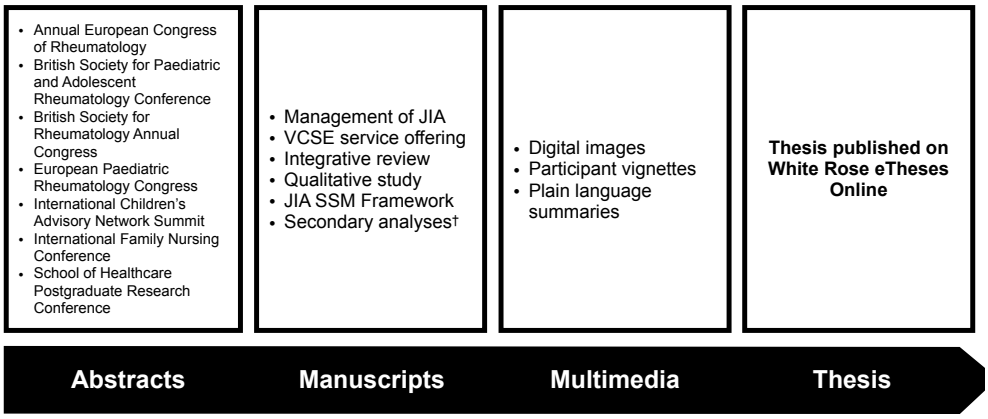
Overall, reflexivity as part of the study echoes a general desire in academia to shift the focus away from a view of research methods as objectified procedures to be learnt by researchers, towards the development of researchers who are capable of crafting procedures integral to the environments in which they operate (Attia and Edge, 2017).

4.13 Publication planning

The main goal of the study was to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their

healthcare, wellbeing, and education. The researcher intended to contribute to the growing evidence-base regarding SSM needs of juvenile-onset LTCs, especially in rheumatology. In pursuing this goal, interpretative research approaches were used to make sense, in depth, of the issues and challenges in this area. Outputs were disseminated locally, nationally and internationally, with the belief that an increased understanding, even prior to intervention development, could yield value to a variety of stakeholders. A publication plan was developed to aid this effective and timely publication of research findings (Figure 14).

Figure 14. Publication plan



†Tentative at the time of publication of this thesis. JIA: juvenile idiopathic arthritis; SSM: Self- and shared-management; VCSE: voluntary, community, and social enterprise.

4.14 Summary

This chapter has outlined the methods used to address the aims and objectives of the study. Using a realist approach to evaluation, the teacher-learner cycle was applied to interviews, using hybrid deductive-inductive thematic analysis to move from initial to refined question theories promoting SSM of JIA across the lifecourse. Considerable attention was paid to the ethics of conducting the study, ensuring the safety of participants and the researcher. Consideration was also given to dissemination of findings, including a publication plan spanning the duration of the study. Chapter 5 of this thesis goes on to present the IQTs developed for subsequent testing with participants.

Chapter 5: Initial question theories

5.1 Introduction

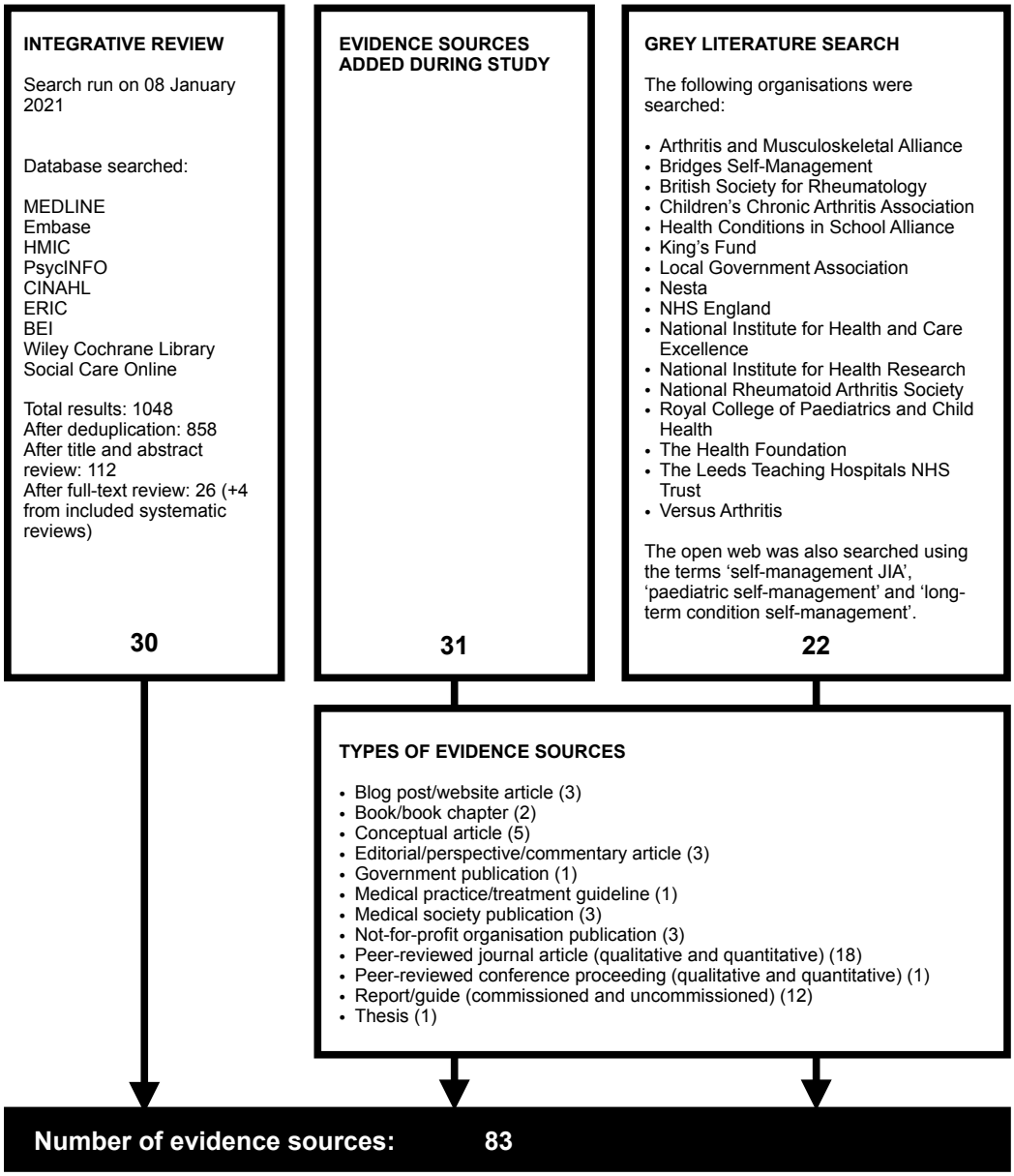
This chapter sets out the IQTs promoting SSM of JIA by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. The IQTs are informed by the experiences of CYP with JIA detailed in Chapter 1, an integrative review of SSMIs described in Chapter 2, and a document review of evidence sources summarised in this Chapter. The thought process by which the researcher navigated evidence sources to develop initial CMO components and IQTs for subsequent testing with participants is also presented.

5.2 Developing IQTs

Pawson and Manzano-Santaella (2012) remark that the purpose of a realist approach to evaluation is to test question theories, and in order to do so, theories "*must be cast as an if-then proposition*", rendered "*into its constituent and interconnected elements*" (p.184). The '*if-then propositions*' are presented towards the end of this Chapter, informed by CMO components. In order to develop the IQTs, the researcher returned to some of the literature discussed in previous Chapters, plus additional evidence sources (Appendix 12) to identify the components underpinning the IQTs. This section sets out the logic the researcher navigated through to establish the IQTs promoting the SSM of JIA by CYP and their families (Figure 15), including a narrative summary of a document review detailing grey literature and additional evidence sources identified through backward and forward citation searching, and internet searches. This is in addition to the literature from the integrative review presented in Chapter 2, and was used to begin identifying relevant contexts, mechanisms, and outcomes underpinning SSMIs for CYP with JIA.

Six key areas of focus were identified to inform the IQTs. These included: individual and family self-management practices; supporting and encouraging SSM; SSM behaviours; timing of SSMIs; approaches and support to access SSMIs; and champions facilitating SSM. These are now briefly discussed, in order to set the scene for the subsequent IQTs to be tested with participants.

Figure 15. Document review evidence sources informing IQTs



JIA: Juvenile idiopathic arthritis; NHS: National Health Service.

5.2.1 Individual and family self-management practices

The process of developing IQTs began with the IFSMT, a mid-range theory proposed by Rodgers (2005) and further worked on by Ryan and Sawin (2009). The IFSMT splits self-management into three dimensions: context, process and outcomes (Fawcett et al., 2001; Meleis, 2011), making this a particularly useful place to start on the journey towards identifying CMO components and IQTs pertaining to the SSM of JIA. Concepts from the Self and Family Management Framework (Grey et al., 2006; Grey et al., 2015), Family Style Management Framework (Knafl et al., 2008) and Paediatric Self-management Model (Modi et

al., 2012) were also used to identify relevant concepts for inclusion in the IQTs, having been identified as candidate theoretical guides underpinning SSM during initial scoping exercises undertaken during study conception.

Ryan and Sawin (2009) suggest that several contextual factors affect the ability and desire for individuals and their families to engage in self-management practices. Some may facilitate SSM, others may present barriers (Grey et al., 2006; Grey et al., 2015). Such contextual factors broadly include condition-specific factors, environmental factors and individual and family characteristics, and modify the dynamic process of self- and family-management of JIA (Knafl and Deatrick, 1990; Grey et al., 2006; Knafl et al., 2008). Condition-specific factors encompass the physical, structural and functional characteristics of JIA, including treatment and the prevention of complications, joint damage and disease progression which impact on the amount, type and nature of behaviours required to self-manage. Environmental factors encompass physical and social contexts, such as access to H&SC, transition from one HCP, provider or service to another, transportation, neighbourhood, school, family working circumstances, culture and social capital. Individual and family characteristics are those of the CYP and their family directly, such as development stages, perspectives, health literacy/numeracy, information processing and capabilities.

The underlying assumptions of the IFSMT emphasise the importance of individuals engaging in behaviours for personally meaningful reasons – which may or may not be conducive to health outcomes. This reflects the notion that SSMIs may be more effective at encouraging self-management behaviours and achieving proximal outcomes, rather than distal outcomes. These underlying postulations also indicate that self-management in CYP with JIA is really shared-management with parents, other family members and professionals involved in CYP's healthcare, wellbeing, and education (Lozano and Houtrow, 2018). To support SSM, evidence identified in this document review suggests that individual healthcare plans (IHPs) could be useful (ARMA, 2010), to enable CYP and their families to document their individual needs, goals and abilities. This could in turn enable more efficient sharing of information with other professionals, including HCPs outside of the paediatric rheumatology MDT and teachers (Health Conditions in School Alliance, 2015; Versus Arthritis, 2019).

There is also strong evidence for SSMIs targeted at CYP, as well as those utilising digital and group-based methods delivered within the community. However, focusing solely on parents, or hospital-only interventions are unlikely to be effective (Kirk et al., 2013). Furthermore, terms commonly used in self-management research such as adherence and compliance contradict the concept of self-management, whereby responsibility lies with the individual, and/or their family to help them overcome barriers to adherence in order to achieve their personal health goals (Rapoff, 2018). This is in the context of their everyday lives (Grey et al., 2006), as active and knowledgeable individuals, instead of passive recipients of H&SC (Kirk et al., 2010). This may be where previous evaluations have failed to understand the broader perception of self-management, the shifting of capacity and capability to competently self-manage amidst daily life, and the impact of interventions initially on shorter-term (proximal) outcomes, coupled with the understanding that non-adherence can be a result of inadequate self-management behaviours (Modi et al., 2012).

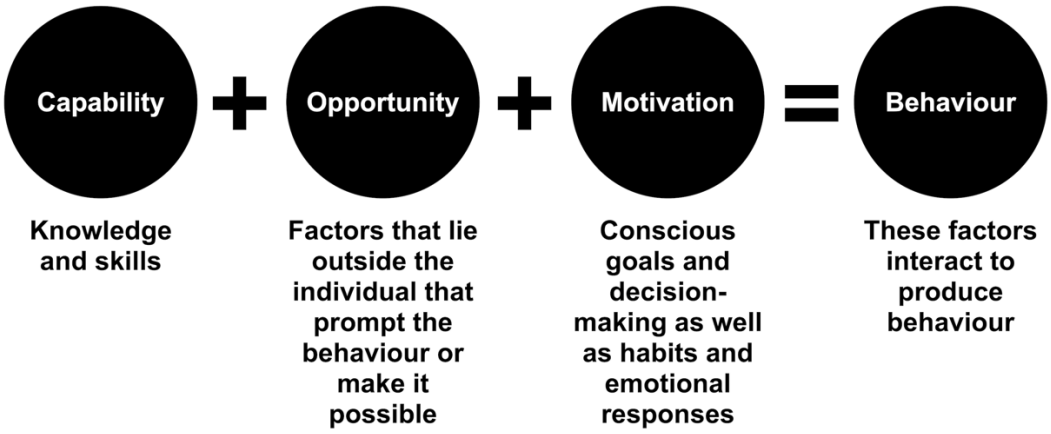
5.2.2 Supporting and encouraging SSM

Kirk et al. (2012) incorporated SSM activities into a 'menu of choices' for SSMIs, defined in relation to four dimensions: providing a sense of community; promoting independence and confidence; developing knowledge and skills; and engaging CYP. They emphasised that the values, attitudes, knowledge and skills required to support SSM should be embedded within interventions and professional practice. Sattoe et al. (2015) went on to propose a content-based measurement framework for the selection of outcome measures in the evaluation of SSMIs based on domains of self-management, which is used in the study and the CMO components detailed later in this chapter (Table 10). From previous studies, the only outcome related to all three domains is HRQoL. Unsurprisingly, almost half of the 78 interventions included their systematic review focused on medical management, with fewer focusing on role and emotional management, which have been recognised as important components of self-management, as also identified in the integrative review presented in Chapter 2.

5.2.3 SSM behaviours

In order to best support SSM, one must reflect upon the underlying behaviours required by CYP and families to want to engage in practices conducive to their H&W (*i.e.*, the underlying mechanisms of SSMIs). This was exemplified in the NHS England-funded Realising the Value programme (Finnis et al., 2016), which was informed by the work of Michie et al. (2011) and the capability, opportunity, motivation, behaviour (COM-B) model (Figure 16). Different factors may be present within each condition, making a behaviour more or less likely to happen (Burd and Hallsworth, 2016a). Five enabling factors were identified in the programme, including: growth mindset, self-efficacy and 'grit' (capability); removing friction costs (*e.g.*, removing increases in effort required to perform a behaviour) (opportunity); social connections (opportunity); intrinsic motivation (motivation); and goal setting and feedback (motivation). These enabling factors are most likely to be conducive when individuals have a sense of purpose, the confidence to act and supportive life circumstances (Wilson et al., 2018).

Figure 16. COM-B system for understanding behaviour (Michie et al., 2011)



COM-B: Capability, opportunity, motivation-behaviour.

People's behaviour has a strong influence on their H&W (Fisher et al., 2011); however, even when people are aware of the influence of their behaviour, and may intend to perform the behaviours conducive to H&W, they often encounter barriers. This can apply to CYP with LTCs like JIA, and professionals supporting those individuals (Greenhalgh et al., 2004), for a number of reasons, including processing excess information, seeking to minimise effort, avoiding change, and using mental shortcuts to interpret facts (Burd and Hallsworth, 2016c).

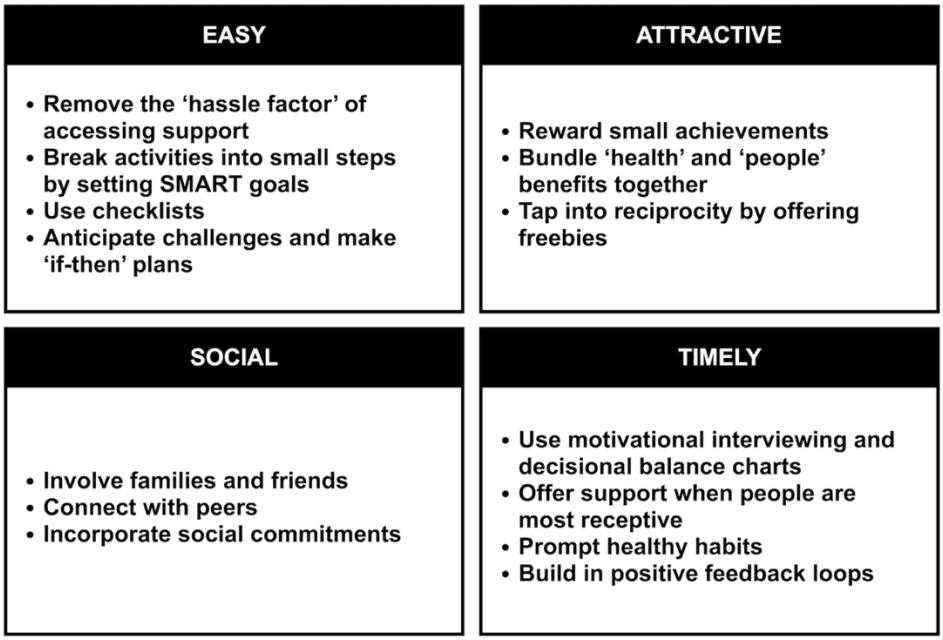
Table 10. Content of self- and shared-management interventions, grouped by self-management domains (Sattoe et al., 2015)

Domain	**Content of self- and shared-management intervention(s)**	
Medical management	Condition-specific	Understanding the condition, understanding (the necessity of) medication and treatment; understanding side effects; adherence; understanding the use of specific treatment devices or techniques; dealing with symptoms; drafting an individual healthcare plan; self-monitoring of clinical outcomes
	General	Accessing healthcare; communication with healthcare professionals; managing consultations; coping with hospitalisations; goals and dreams for the future related to health and wellbeing; Children and young people-parent sharing/teamwork related to condition-specific medical management; knowing where to find specific disease information; knowing when to ask for medical help; risk behaviour (*e.g.*, unsafe sex, drug abuse)
Role management	Social initiation and friendship making; social networks; family and romantic relationships; sexuality; relationships and sex; managing teasing and bullying; conflict resolution; participating in normal social activities; keeping up with peers; internet and social media; goals and dreams for the future related to school, work, community, living, housing, recreation and leisure; school issues; explaining the condition to others (disclosure); educating peers; setting (life) goals and becoming assertive; growing up; communication and social problem solving; organisational skills; independent living; travelling/staying abroad; social rights and benefits	

Domain	Content of self- and shared-management intervention(s)
Emotional management	Self-confidence and self-esteem building; developing a positive body image; body esteem; self-appreciation; enhancing hope; enhancing self-efficacy; empathy; fear-related thinking; feelings related to condition; sharing of feelings and experiences; accepting condition; self-reflection; healthy expressions of anger and transforming or managing anger; helpful/positive thoughts; stress management; decreasing negative thoughts; decreasing stress and boredom; decreasing social isolation; spirituality; emotions

Frey and Rogers extended this notion by proposing four ‘persistence pathways’, elaborating on the ways that interventions may lead to long-term sustained behavioural changes (Frey and Rogers, 2014). The most convincing pathway is habit, since this is known to encourage automatic behaviour. Other pathways include: changing how or what people think; changing future costs; and external reinforcement (Perry et al., 2015). These pathways can be materialised with the easy, attractive, social, and timely (EAST) framework (Service et al., 2014) consisting of four principles (Figure 17) which should apply when encouraging a behaviour (Dolan et al., 2010). However, these principles can only be applied when clearly defining the outcome(s), understanding the context, and how the intervention operates (*i.e.*, the intervention mechanisms) (Finnis et al., 2016).

Figure 17. The EAST Framework (Burd and Hallsworth, 2016c)



EAST: Easy, attractive, social, timely; SMART: Specific, measurable, attainable, realistic, time-bound.

Nonetheless, the behavioural science field has been critiqued for focusing too much on individuals and interpersonal factors, rather than wider institutional and infrastructural factors (Marteau et al., 2011). Indeed, while focus must be paid to individuals, it’s important to consider the systems, structures and cultures which enable or restrict individuals from performing certain behaviours. This is where the simple intervention of ‘nudging’ bears relevance, whereby the environment (*i.e.*, the context), is altered to change (or indeed instil) behaviour (Hauser et al.,

2018), and it is why institutional and infrastructural factors are considered in the study, in addition in individual and interpersonal factors.

5.2.4 Timing of SSMIs

Having briefly discussed some of the enabling factors for optimum SSM behaviour, it is necessary to reflect on the willingness and capacity of CYP and families to adopt such behaviours changes over time; recognising that that they tend to pass through different stages of motivation or activation for adopting behaviours (Prochaska and DiClemente, 1982; Hibbard and Gilburt, 2014). For CYP with JIA and their families, appreciating this variability of JIA symptoms could be critical for the timing of certain SSMIs to aid optimum SSM, in light of the waxing and waning of symptoms, and transition between disease flare and remission. However, physiological changes in the course of JIA are just one component of the puzzle, with psychosocial factors playing an important part in the 'shifting perspective' as to whether illness or wellness is in the psychological foreground (Paterson, 2001). One tool which Burd and Hallsworth (2016c) highlight to aid people in moving towards a readiness for change is a decisional balance to deliberate the pros and cons of certain behaviours, which when used with motivational interviewing (Erickson et al., 2005), can support conversations with CYP and families hesitant or resistant to change (Miller and Rollnick, 2002; Miller and Rollnick, 2009). This could be useful during periods of uncertainty and at times when CYP are most receptive, such as during transition from paediatric to adult H&SC (ARMA, 2010), or at a time point shortly after diagnosis, as CYP come to terms with a diagnosis and begin seeking information and support. In addition, the importance of frequent 'touchpoints' for information, education, and support has emerged from both the literature and researcher's experience as valuable points over time when SSMIs may help CYP and families to build confidence and take the actions needed for them to achieve their personal goals (Wilson et al., 2018).

5.2.5 Approaches and support to access SSMIs

Finnis et al. (2016) describe person- and community-centred approaches as practices encompassing 'more than medicine' holistic approaches to complement clinical care (such as access to peer support, self-management education and health coaching), to community activities that enable people to

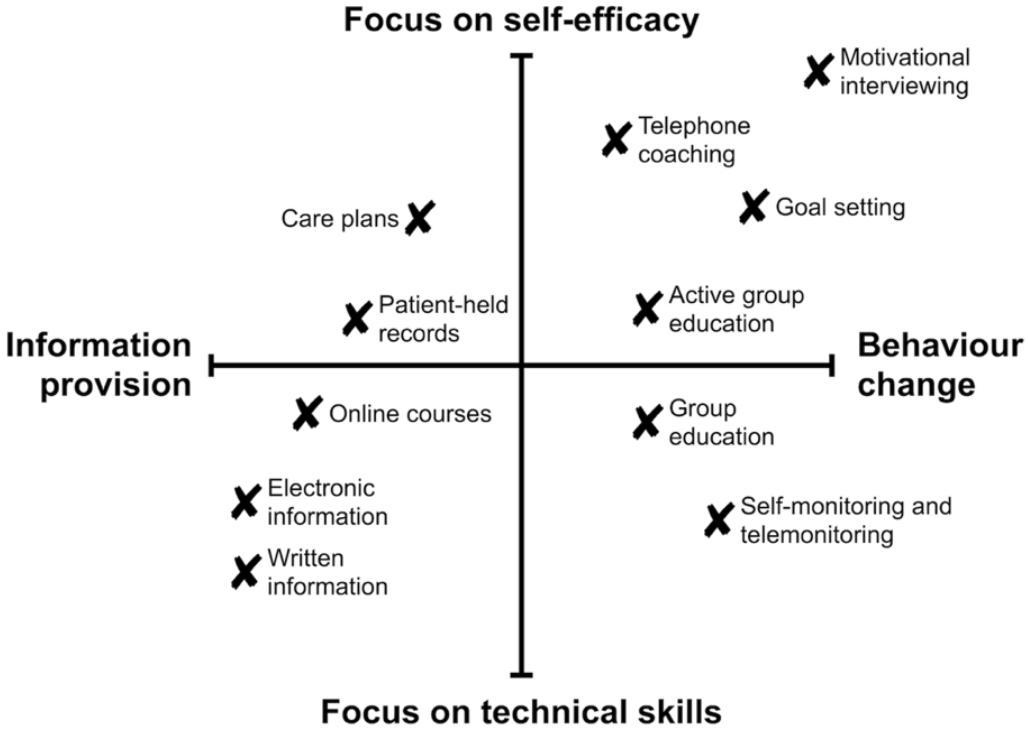
improve their H&W, regardless of their condition (such as youth group activities and asset-based activities). While the nuisances vary, person-and community-centred approaches must always:

> *"Put people and communities at the heart of health and wellbeing, focusing on what is important to people, what skills and attributes they have and on the role of their family, friends, and communities."* (p.13).

A number of enabling mechanisms of these person- and community-centred approaches are described in the Realise the Value report (Finnis et al., 2016), which are essentially resource mechanisms which would connect people with SSMIs. These include: personalised care and support planning, personal health budgets, social prescribing and bridging/facilitator roles, such as health trainers, community navigators, youth and family support workers and transition co-ordinators (Wood et al., 2016). These approaches, coined 'activation', enable people to look after themselves (or their loved ones) better, to have meaningful relationships and to collaboratively work with professionals. In doing so, the focus on developing knowledge, skills and confidence should lead to behavioural change. This change has been measured using the PAM® in adults; however, using the PAM® to tailor support to different CYP, and importantly their families, could be useful in maximising SSM support as well as health inequalities (Hibbard and Gilburt, 2014); thus ensuring timely access to the right services, for the right groups of CYP and families, at the right time.

There are a multitude of approaches which have been described to support SSM, and it is clear that multi-setting approaches are beneficial. These range from more passive interventions pertaining to information provision, to more active interventions targeting behaviour; and from focussing on technical skills to focussing on building self-efficacy (de Silva, 2011). These typologies and positions are illustrated in Figure 18, to aid the development of IQTs, and to illuminate the importance of SSM as a longer-term investment in enabling CYP with JIA and families to live well with their condition (distal outcomes), which does not necessarily happen after participating in a solitary intervention.

Figure 18. Typologies and positions of SSM support (de Silva, 2011)



There has been a notable focus on ‘nudges’ as a type of behavioural change intervention in recent years to encourage people to adopt certain behaviours, appreciating that many behaviours are automatic and not made consciously (Perry et al., 2015). In nudge-type interventions, encouraging certain behaviours is achieved while maintaining people’s freedom of choice (Sunstein, 2016), with the goal of reducing choice overload, simplifying processes and removing potential friction costs. While nudge-type interventions may be useful to aid certain in aspects of the SSM of JIA, nudges tend to only work when the context is stable, since they are designed to effect immediate (proximal) behaviour (Van Lieren et al., 2018) – prompting for a more nuanced approach to support CYP and families into the most appropriate mindset at the right time. Schools are also a key area where understanding of LTCs like JIA are conducive to enabling CYP to self-manage their condition, with shared-management support from their peers and professionals (Kirk et al., 2012).

5.2.6 Champions facilitating SSM

The role of SSM champions, whether they be CYP, families, HCPs, VCSE professionals or education professionals, can help to engage others in

practicing and/or promoting SSM (Burd and Hallsworth, 2016b). By acting as role models, champions can help to nudge individuals within their network to behave differently (Bodenheimer, 2007). For CYP and families, such champions may be young adults and other parents, since this shared connection can help CYP and families to identify, believe and aspire to follow the actions of influential peers (Perry et al., 2015). The influence and nature of the relationship between CYP, families and their HCPs, in particular the CNS and doctors (Oliver, 2018) can influence whether or not SSM becomes an integral function of managing JIA (Kirk et al., 2010). Meanwhile for HCPs, fellow colleagues are mostly likely to inspire change and uptake via peer networks (Greenhalgh et al., 2004), alongside patient and parent champions amplifying the value of SSM (Harkins and Petty, 1981). This is important, particularly when driving positive change among HCPs who may not see the full value of SSM (Burd and Hallsworth, 2016b), in terms of potential for themselves and the H&SC system, or indeed CYP and their families – now, and for their future selves. Finnis et al. (2016) call for new and more constructive engagements between H&SC and VCSE providers, and those with LTCs like JIA to be involved in shaping and supporting such care for others. This is reflected in the IQTs to encompass all of the relevant stakeholders potentially involved in the SSM of JIA.

There have also been calls to upskill HCPs and non-clinical staff across different teams and services to be able to more confidently integrate SSM support into everyday practice (Bridges Self-Management, 2019). This aligns to priorities for H&SC services to be commissioned in ways that incentivise and support person- and community-centred approaches (Kirk et al., 2010; Finnis et al., 2016), and sub-speciality learning outcomes in paediatric rheumatology on organising patient and parent education and empowering CYP to negotiate their own care (RCPCH, 2018).

Furthermore, a challenge reported in the literature and observed in reality is a lack of awareness among HCPs around the range of VCSE services and available for CYP with JIA and families. Calls have been made to facilitate signposting to VCSEs, making the process as easy as possible for HCPs, as well as CYP and their families (Burd and Hallsworth, 2016b). This is likely to become of increasing relevance with an increasing focus on social prescribing within and beyond primary care (Local Government Association, 2016), as outlined in the NHS Long-term Plan (NHS England and NHS Improvement,

2019a); the aim of which is to connect people with LTCs with 'more than medicine' approaches to H&W through linkage with VCSEs.

VCSE organisations provide essential information, education, and support and services for many CYP with JIA and families, often filling gaps where H&SC services simply do not have the resources to address. As detailed in Chapter 1, VCSEs work to support CYP with JIA, as well as parents and siblings in various manners. Beyond providing trusted information, education, and support on living with JIA, some also provide recreational activities and workshops for CYP and their siblings, to build their confidence and self-esteem while also tackling sensitive subjects such as body image (ARMA, 2010). However, widespread knowledge of VCSEs among CYP, families and HCPs is inconsistent, as is signposting to such information, education, and support, despite the ARMA Standards of Care in 2010 stating that "*children and their families should be made aware at the time of diagnosis*" (p.8) of such organisations (BSPAR, 2009; ARMA, 2010).

One approach of connecting people with 'more than medicine' approaches, albeit from adult rheumatology, is the 'New2RA Right Start Service' (NRAS, 2020), which aims to improve the outcomes of newly diagnosed people with rheumatoid arthritis through a collaborative framework of self-management tailored to individuals (Bosworth and Galloway, 2020). Implementation of such services exemplifies how H&SC and VCSE providers can be complementary 'system partners' (Finnis et al., 2016). With VCSEs investing resources into SSM support, it seems discernible for HCPs to leverage available support services while advocating for greater integration (Lozano and Houtrow, 2018).

Burd and Hallsworth (2016b) go on to highlight the importance of enabling different individuals and organisations to seek ownership of SSMIs, recognising that "*lifting and shifting programmes of person- or community-centred approaches from one area to another rarely encourages interventions to spread meaningfully*" (p.15) (Greenhalgh et al., 2004). Therefore, they suggest that individuals and organisations design and adapt their own interventions to suit their specific needs, without losing the "*hard core of effective practice at its heart*" (p.15). This is evidenced by people's tendency to value their own creations (Burd and Hallsworth, 2016b). In practical terms, this is where the findings from the study and the subsequent framework set out in Chapter 7 may be useful, to provide a basis of *what works, for whom, in what circumstances,*

and why, for VCSEs to build upon, and then operationalise with different groups of CYP and families they may come into contact with.

5.3 Condensing concepts into initial CMOCs and IQTs

The researcher needed to condense and clarify the aforementioned concepts from the document review presented in this chapter, along with the integrative review findings from Chapter 2 and background literature in Chapter 1, into a set of IQTs promoting SSM of JIA to be tested with key stakeholders. To aid this narrowing process, the researcher used Pawson's four levels of contextual influence and the Paediatric Self-management Model (Modi et al., 2012), to place the primary focus on individual and interpersonal levels, and the secondary focus on institutional and infrastructural levels, recognising the wider role of organisations and policy in promoting and enabling SSM of JIA. The CMO components identified from the literature were itemised as a list of concepts under the four levels of contextual influence, and are visualised in a matrix in an attempt to portray the thought process by which concepts were grouped as contexts, mechanisms, and outcomes underpinning the SSM of JIA. These were kept at a higher level of abstraction and not typified as CMO configurations at this point in the study, since the contexts cut across the mechanism-outcome patterns, and so it was anticipated that the link between context and the mechanism-outcome coupling would become clearer during the evaluation, in particular after gleaning theory during initial interviews. The CMO component matrix informed the subsequent IQTs derived from these tentative ideas of how and why SSMIs exert their effect on CYP with JIA and their families, set as 'if… then' statements combining contexts, mechanisms, and outcomes from the CMO component matrix, for testing through primary data collection. These were proposed by the stakeholder group potentially involved in the SSM of JIA (*i.e.*, at the level of CYP, families, and HCPs in IQT1, 2, and 4), by the organisations and policies involved in promoting the SSM of JIA (*i.e.*, VCSE organisations, H&SC services, and education in IQT5, 6, and 7), and by specific tools which could promote SSM of JIA with the involvement of multiple stakeholders and organisations (*i.e.*, IHPs, IQT3). Presentation of the CMO components in this way also illuminates the close interplay and state of balance between people operating individually, and people operating collaboratively; realising that the dynamic between lived experience and professional

experience, corresponding with the notion that interventions work only if people invest in their goals (Pawson, 2006b).

5.4 Initial CMOs and IQTs

Initial CMO components were tabulated as a matrix, reiterating that causality is located at the individual and interpersonal level – amongst the actions people take, individually and collaboratively, in response to the resources made available to them through SSMIs (Table 11), with clear reference to the role of organisations, evidence, and policy shaping the actions people take regarding the SSM of JIA. The initial CMO matrix corresponds with IQTs shown subsequently in order to map causality, with IQTs drawing on the tentative relationship between outcomes and mechanisms, and the role of context. The matrix can be depicted as a series of possible CMO configurations which are captured in the IQTs at a higher level of abstraction, for example, 'Within an optimistic, proactive and supportive family environment (C), CYP with JIA can engage in a family support weekend (SSMI) in the presence of other CYP with JIA and their families (M_1), to increase their confidence in managing JIA (M_2), so that they can feel more in control ($O_{proximal}$)'. Four IQTs at the individual and interpersonal level evolved during the aforementioned development process (Table 12), while three IQTs were developed at the institutional and infrastructural level (

Table 13). These IQTs informed the semi-structured discussion guide which was used in interviews with participants.

5.5 Summary

Seven IQTs pertaining to the SSM of JIA by CYP, their families, and professionals involved in their healthcare, wellbeing, and education were set out in this chapter. The IQTs suggest that holistic SSM support needs to be consistently provided across the lifecourse CYP and families in meaningful ways, which best reflects the specific needs and preference of all CYP. These were developed comprehensively, using background literature about SSM and JIA presented in Chapter 1, literature on SSMIs presented in Chapter 2, and a document review summarised in this chapter, akin to ‘digging for nuggets’ of valuable concepts to unearth the underlying processes of SSM applicable to JIA, to aid subsequent gleaning, refinement and consolidation of the question theories with key stakeholders in Chapter 6. Consequently, this process enables a deeper understanding of the nuances of SSM of JIA by CYP and their families to be shared, by accumulating knowledge about SSM in this population.

Table 11. Initial CMO component matrix

	Contexts	Mechanisms	Outcomes
Individual	• Non-modifiable demographics by intervention (*e.g.*, ethnicity, age) • Behaviours • Condition-specific factors • Family structure and function • Interest in SSM	**Resource** • Different provision and styles of education **Responses** • Knowledge and beliefs • Self-regulation skills and abilities	**Proximal** • Medical management (*e.g.*, managing symptoms) • Role management (*e.g.*, social and vocational participation) • Emotional management (*e.g.*, self-confidence and self-esteem) **Distal** • Health status (*e.g.*, remission) • QoL and perceived wellbeing • Attainment (*e.g.*, qualifications)
Interpersonal	• CYP and family relationship, support and time • Receptiveness for change • Shifting of responsibilities • CYP/family and relationship with service providers • Cultural norms and stigmatisation	**Resources** • Social facilitation and support through negotiation and an open environment • Familiar, trusted and relatable intervention facilitators/co-ordinators • Individualised, person- and family-focused care and support **Response** • Trust	
Institutional and infrastructural	• Physical and social environment factors (*e.g.*, social incentives, safe environment and transportation) • Health and social care service provision and access • Education provision and access • VCSE organisation provision and access	**Resource** • Individualised, person- and family-focused care and support	**Proximal** • Healthcare service utilisation (*e.g.*, hospitalisation) • VCSE service utilisation (*e.g.*, event attendance) **Distal** • Health status (*e.g.,* remission) • QoL • Cost of health • Attainment (*e.g.*, qualifications) • Intervention design

CMO: Context, mechanism, outcome; CYP: Children and young people; QoL: Quality of life; SSM: Self- and shared-management; VCSE: Voluntary, community and social enterprise.

Table 12. IQTs at the individual and interpersonal levels

IQT as an 'if… then' statement
1. Meaningful self-management support across the lifecourse for CYP with JIA
If self-management support opportunities for CYP with JIA are introduced, easily accessed, attractive, socially-orientated and consistently endorsed from the point of diagnosis in an age, developmentally-appropriate and personally meaningful way for each CYP, within a trusted and familiar network across H&SC, education and VCSE organisations, **then** CYP will have the opportunity over time to become increasingly competent at the medical, role and emotional management of their H&W, by improving their knowledge, beliefs, self-regulation skills and abilities during childhood, in anticipation for adulthood.
2. Meaningful shared-management support for families supporting CYP with JIA
If shared-management support opportunities for parents, siblings and other family members supporting CYP with JIA are available and accessible over time from the point of diagnosis, with an emphasis on the gradual transfer of responsibility at appropriate timepoints to CYP, **then** families will generate their own strategies to aid CYP and the entire family to more competently manage the medical, role and emotional aspects of JIA now and in the future.
3. IHPs as shared-management tools to aid other professionals in supporting the specific needs and preferences of CYP with JIA and their families
If IHPs are made available to all CYP with JIA and their families from the point of diagnosis, are unfailingly used and regularly updated over time to reflect on the specific needs and preferences of CYP with JIA, **then** this will facilitate how key stakeholders (such as school staff, other HCPs and VCSE professionals) support the shared-management of JIA for CYP under their professional care, in a manner which reflects the medical, psychological and social needs of every CYP with JIA.
4. Consistent recognition and approaches within the paediatric rheumatology MDT towards the value of SSM support for CYP with JIA and their families

IQT as an 'if… then' statement
If all members of the paediatric rheumatology MDT (internally and externally) recognise the value of SSM support, are aware of all available support opportunities for CYP with JIA and their families, and consistently refer and/or signpost them to such support from the point of diagnosis and throughout long-term follow-up, **then** it is more likely that CYP and families will secure access to information and support from the outset, in order to aid the medical, role and emotional management of JIA in a more consistent manner.

H&SC: Health and social care; H&W: Health and wellbeing; IHP: Individual healthcare plan; IQT: Initial question theory; JIA: Juvenile idiopathic arthritis; MDT: Multi-disciplinary team; SSM: Self- and shared-management; VCSE: Voluntary, community and social enterprise.

Table 13. IQTs at the institutional and infrastructural levels

IQT as an 'if… then' statement
5. SSM support services commissioned with statutory services as a component of routine care for CYP with JIA and their families
If SSM support services are commissioned in a way that are integrated with statutory services (for example, VCSE-delivered services within H&SC services), so as to be available and accessible in a timely manner for all CYP with JIA and their families as part of routine care, **then** those CYP and families will have quick and easy access to the resources required for them to be able to more competently manage all aspects of JIA on a more equitable and informed level, while reducing their reliance on constrained H&SC services that could otherwise be avoidable.
6. CYP- and family-centred holistic care across the lifecourse for those living with JIA
If paediatric rheumatology H&SC services are designed to facilitate access to, and navigation of services in a CYP- and family-centred way, including an holistic focus on integrated SSM support within clinical practice, shared care across specialities and settings, continuity of care, seamless transition and involvement of CYP and families in all decision-making processes, **then** the H&W of CYP with JIA, as well as their families, is likely to be improved.
7. Inclusive and proactive educational settings to enable CYP with JIA to secure equivalent educational attainment and social development to their peers
If schools are inclusive for all CYP with special educational needs and disabilities (including LTCs like JIA) with appropriate policies and provisions in place, and if relevant education professionals within schools (and the local authority) are involved in the shared-management of JIA by working with CYP, families, HCPs, and VCSEs in identifying and communicating specific support needs for each CYP, **then** CYP with JIA will be more likely to enjoy school and have a positive experience, akin to their peers, so as to not be at a disadvantage in terms of educational attainment and social development.

H&SC: Health and social care; H&W: Health and wellbeing; IQT: Initial question theory; JIA: Juvenile idiopathic arthritis; LTCs: Long-term conditions; SSM: Self- and shared-management; VCSE: Voluntary, community and social enterprise.

Chapter 6: Qualitative findings

6.1 Introduction

The following chapter presents the findings of the qualitative study using a realist approach. The study aimed to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education, in order to confirm or refute contextual influencers and generative mechanisms for developing SSM capacity amongst CYP with JIA and their families. This was achieved by testing, refining and consolidating the IQTs presented in Chapter 5, which tentatively described the SSM of JIA. In this chapter, the themes identified are discussed thematically, informing the RQTs in which they are presented.

6.2 Participant characteristics

Out of 22 interested participants, 21 consented and one was lost to follow-up. Therefore, 20 participants were recruited; 17 of whom identified as female. Eight participants were recruited via the partner NHS Trust, and 12 responded to study advertisements by email and on social media. There were participants from three of the four devolved nations of the UK, although the majority were from England (*n*=16). The demographics of the participants are presented in Table 14. Participants were recruited between 15 October 2018 and 03 April 2020, with interviews taking place between 05 November 2018 and 06 April 2020. The researcher conducted interviews independently. Six participants were interviewed in dyads (see Table 14), with the remaining 14 interviewed individually. Sixteen participants were interviewed by telephone; while four were interviewed by video conferencing software – either Skype™ (*n*=3) or Zoom™ (*n*=1). In total, there were 1030 minutes of audio recordings, equating to 170,486 words across 376 pages of transcripts. The average interview lasted 61 minutes, and the average transcript consisted of 10,029 words across 22 pages. Some participants had multiple experiences (*e.g.*, parent and VCSE professional). To protect participants' identity, pseudonyms are used, and those with multiple experiences are identified only by their primary focus at the time of interview. However, for context, additional experiences not shown in Table 14

include one parent, two VCSE professionals, and four young adults/adults living with JIA.

Table 14. Participant characteristics

Participant	Experience	UK nation
Audrey	VCSE professional	England
Ava	HCP (Paediatrician)	England
Betsy	Teacher	England
Caoimhe	HCP (CNS)	England
Chanda*	VCSE professional	Scotland
Clara†	Parent	England
Darcie‡	Child aged 5-7 with JIA	England
Esther	Parent	England
Helen‡	Parent	England
Hope	VCSE professional	Northern Ireland
Judi	VCSE professional	Scotland
Kendra	HCP (Youth and Family Support Worker)	England
Krishna	HCP (Paediatrician)	England
Martha	Teacher	England
Megan*	VCSE professional	Scotland
Miriam	VCSE professional	England
Toby†	Parent	England
Tom	HCP (Paediatric Rheumatologist)	England
Victoria	HCP (CNS)	England
Xena	VCSE professional	England

*Dyad 1 (VCSE professional–VCSE professional); †Dyad 2 (parent-parent); ‡Dyad 3 (child-parent). CNS: Clinical nurse specialist; JIA: Juvenile idiopathic arthritis; UK: United Kingdom; VCSE: Voluntary, community and social enterprise.

6.3 RQT refinement and consolidation

As data analysis progressed, it became clear that there was overlap between IQT6, 'CYP- and family-centred holistic care across the lifecourse for those living with JIA', and other IQTs. As a consequence, IQT6 was disbanded and its themes mapped to IQT5. Therefore, six RQTs emerged: RQT1-4 operating at the individual and interpersonal level, and RQT5-6 operating largely at the

institutional and infrastructural level, with some level of interpersonal context. The six RQTs and their themes are discussed below, with direct quotations used to illustrate the findings while enabling judgments to be made about their credibility. The themes presented below explore the relationship between contexts, mechanisms, and outcomes, to inform the CMO configurations and RQTs presented and discussed in Chapter 7.

6.4 RQT1: Self-management of JIA by CYP

6.4.1 Age, development, and cognitive functioning

Self-management was deemed by participants to be more appropriate for older CYP; however, the relevance of the age of disease-onset and the time of diagnosis as contextual influencers became clear, as well as assumptions and perceptions of age-related capacity and capability. One participant described there being no HCP assumption that parents would perform all tasks for older CYP, while HCPs such as Krishna, a paediatrician, were generally more confident in gauging CYP's developmental capacity. Participants remarked that for those diagnosed at a younger age, SDM will be the parents' responsibility, and the likelihood of re-visiting disease understanding and self-management with CYP over time is unlikely, thus CYP in early adolescence are most likely in need of more intensive SSMIs:

> *"The younger group… who seem to be coping okay… we don't go back and do the discussion again, as they get to an age where they can start to understand it… we just carry on and carry on with treatment, but we might not check how much they know about it."* (Krishna, HCP – Paediatrician)

However, age is not the only potentially influencing context, with developmental stage/capacity playing a role in CYP's self-management readiness and willingness. Participants reflected on CYP's uniqueness, and the need for individualised education and empowerment. Childhood and early adolescence were seen as periods where CYP are constantly learning, and receptive to new information and strategies if pitched appropriately. Miriam, a VCSE professional, used the simile of 'soaking information like a sponge' to describe this optimum period of learning:

> *"Like a sponge… soaking up information… that's a really important stage where they ought to be learning about taking on their own care before they're a teenager and they get stroppy and the hormones all kick in."* (Miriam, VCSE professional)

Enabling CYP to foresee causation was seen as an important step for when they are developmentally capable to assimilate knowledge and observations in anticipation for self-management. Similarly, participants such as teacher, Betsy, felt that explaining key concepts, using age- and developmentally-appropriate language, could help CYP as they grow up with JIA:

> *"When you're teaching phonics [method for teaching how to read and write an alphabetic language]… you use the correct words for the graphemes [smallest functional unit of a writing system] and the digraphs [two letters combined to make a single sound]… the kids all understand what they mean… they know the different sounds… because then the older they get, you don't have to continually keep introducing something new."* (Betsy, Teacher)

The impact of JIA on cognitive development was raised by educational professionals, such as Martha, who remarked that symptoms and consequences of JIA, such as fatigue, over-exertion, and anxiety, can exacerbate symptoms, while impacting motor control and cognitive development, including high functioning CYP who are academically and socially capable. This was felt to be something which existing interventions may overlook, given that JIA is typically seen as a physical condition:

> *"Clearly [there is] a physical need… that might impact… cognition and learning… when you are experiencing pain, that… interferes with all your other core cognitive processes because your brain is trying to deal with the pain… how can you then be expected to learn [and] socially interact?"* (Martha, Teacher)

6.4.2 Stigmatisation and normalising life

CYP with JIA often carry the emotional burden of their diagnosis, feeling different to their healthy peers. CYP can feel disabled by their diagnosis, requiring perspective and support to help them adapt their lifestyle so that they can perform their preferred activities:

> *"I always think of it now like a rucksack of baggage… children who feel different in some way carry that with them and that affects how they feel when they mix with their peers, which… is what school is about."* (Miriam, VCSE professional)

Finding and enjoying hobbies is another important part of childhood, but something which can be more complicated and challenging for CYP with JIA. Participants highlighted how for some, hobbies undertaken with others living with additional needs, can help them to feel accepted and valued.

Unfortunately, a poor public understanding of JIA, as well as a lack of empathy, can impact on CYP's behaviours, such as when compared to osteoarthritis affecting older people, and to those with noticeable, physical impairments. Participants remarked that this can, in turn, make CYP internalise and introject negative thoughts from the external world, impacting emotional and role management outcomes.

6.4.3 Condition-specific factors

Parents highlighted the need for flexibility, reflecting the variable nature of JIA. During periods of minimal disease activity and remission, there may be less need and preference for intervention. Participants remarked on CYP's frustrations during a relapse, and the occurrence of interconsultation crises, where CYP must recognise, react, and adapt to changing symptoms.

HCPs also remarked that fewer CYP nowadays require hospitalisation for JIA. Therefore, a greater level of care and responsibility falls to CYP, as well as parents, with more treatment provided at home. This may alter the dynamic of how consultations are performed, with greater independence ensuing when CYP have the opportunity to see HCPs in the absence of their parents. Participants felt that parents' presence can hinder CYP from discussing sensitive topics with HCPs. Therefore, HCPs believed that split appointments could be a reasonable compromise, so that CYP have the opportunity to self-advocate and disclose information to HCPs, while also allowing parents some level of input.

Dealing with, and integrating JIA-related medical interventions into daily life can be a challenge for many CYP, especially when they have not received the necessary support to manage anxiety. Trypanophobia appears to be a common issue. For younger CYP, getting to the point of feeling comfortable having an

injection was viewed as the first goal, before considering self-administration. As one parent highlighted, hearing the words 'injection' and 'methotrexate', can cause her daughter anxiety:

> *"We have to be careful in how we describe these things because [she] is not very keen on sharp, pointy things or naming the drugs… it's something of a battle every time… [her] phobia of it extends to watching videos."* (Helen, Parent)

Beyond trypanophobia, treatment for JIA can cause pharmacophobia (fear of pharmacological interventions) and anticipatory responses, such as anticipatory N&V and associative interference (mistaken association between different stimuli) – which can profoundly impact CYP's psychosocial H&W:

> *"We went through a phase where we'd only do injections after having iced her leg… then, she got so phobic about cold that she wouldn't go into the chiller section in [the supermarket] because she felt cold, and the cold made her feel sick because it reminded her of methotrexate."* (Miriam, VCSE professional)

These negative experiences, particularly with methotrexate administration, are seen to influence treatment adherence, though this may not be immediately clear within a consultation with a HCP. Hope, a VCSE professional, found a remarkable difference in CYP's honest response regarding treatment adherence between the clinical and VCSE settings, highlighting the need for more open and honest conversations, where CYP feel more comfortable to disclose their true feelings and experiences:

> *"Twenty kids were asked at the clinic, are you taking your methotrexate regularly… all fine… then we asked the same 20 kids on a residential… and the answers were completely different… they just say… 'I don't want to upset anybody'… they're not trying to be difficult, they're just being kids."* (Hope, VCSE professional)

The process for training CYP to self-administer subcutaneous injections also appeared to vary, with regards to the HCPs involved, the resources available (*e.g.*, practice devices), and how CYP and/or parents were involved. This was perceived to be additionally challenging when CYP were taking control from their parents without receipt of formal training. Greater consistency and CYP-friendly approaches were felt to aid CYP in feeling more confident with self-

administration, such as knowing how much pressure is required to successfully self-inject.

Furthermore, enabling CYP to contextualise their treatment and how they self-manage their H&W as part of everyday life can be important, and is influenced by those supporting them, especially parents. Participants such as Toby, whose daughter has JIA, felt that self-management should be regarded within the remit of self-care practices, so as to not seem overwhelming. The prospect of taking a tablet every day for the rest of their lives may seem overwhelming, yet the need to brush teeth twice daily does not. Thus, helping CYP to normalise self-management can be beneficial, to the point where CYP can be resourceful and proactive in managing their treatment, and broader H&W:

> *"She reminds us if she hasn't had her folic acid or her vitamin D, even though I've got a reminder on my phone… she'll be remembering before that and goes and gets it out the cupboard."* (Toby, Parent)

6.4.4 Family factors and relationships

Another influencing context is CYP's living environment, including the changing dynamic with parents. While the expected involvement of older CYP was considered by participants, attention was focused on those diagnosed at younger ages, when parental proxy management is necessary. Participants felt that younger CYP's JIA information and education needs related to JIA are seldom re-visited over the lifecourse, especially if they seem to be coping. Participants felt that these CYP could benefit from an experience akin to adolescents receiving information and support at diagnosis.

Cultural comparisons were also drawn, with one participant observing a greater dependency on parents in the UK compared to the rest of Europe, with UK CYP appearing more reliant on parents to manage their H&W, up to, and beyond, the point of transfer to adult H&SC services. HCPs felt that CYP do not self-manage their H&W themselves entirely, with a smaller proportion seemingly motivated and determined to be in control, often when encouraged to do so in a safe and controlled manner within a strong and supportive home environment. The opposite could be argued, particularly amongst adolescents, rebelling about instruction from parents, as Judi had observed in her VCSE role:

"It's much easier, isn't it, just to say you're not allowed to do this… and then expect your children to listen. But of course that's not going to happen… they'll do what they want to do anyway, so you might as well be honest about it." (Judi, VCSE professional)

6.4.5 Growth mindset in the face of uncertainty

JIA can often feel like a punishment for CYP – whether that be the diagnosis itself, the treatment, or the restrictions imposed on them by others for their perceived benefit. Accepting a diagnosis can be challenging, and CYP diagnosed during early childhood whose disease later relapses, can experience delayed realisation, akin to managing a new diagnosis:

"It was only with her turning eight or nine… having a bad flare [of JIA], back on medication when she'd been off for a while… that was when she started being really cross and angry, and saying… why am I the only person in the world [with JIA]." (Miriam, VCSE professional)

The bridge between accepting and confidently living with JIA also appears varied. Participants commented that not all CYP will want to be vocal about their condition, but felt it was important that they learned to acknowledge and accept it, since this is often a key hurdle towards becoming competent self-managers. Confidence and self-confidence were seen as two sides of the same coin, as Esther remarked, contrasting her daughter's public speaking ability with reservations to converse with HCPs:

"They say how are you… and how has it been? And she'll be like oh yeah, okay fine… It's like, just tell them actually what the problem is. You need to learn to stop saying that you're fine." (Esther, Parent)

As a result, CYP may persevere with their symptoms, out of embarrassment or avoidance to draw attention to themselves. This can be problematic for those in school, who may need to pace themselves, but do not wish to stand out from their peers. Some VCSEs, including the organisation Audrey works with, aim to develop the skills and confidence to empower CYP to be able to talk more openly about their condition and experiences, while promoting self-esteem, through their services and activities:

> *"[After] coming along to a [VCSE] event, learning more about her condition, knowing that she wasn't the only one [with JIA]… she suddenly had the confidence to… say [to the HCP]… this is what's happening, I'm not happy about it… can we talk about what other options are available?"* (Audrey, VCSE professional)

Adolescence is a period of great change and uncertainty for all CYP, who may seem to cope well; however, when compounded by JIA, their ability to calmly and confidently incorporate change and uncertainty can be volatile and overwhelming, especially in light of tension between independence and dependence during such periods:

> *"When your medication has to change, and medication fails, or uveitis gets involved… something new comes to the party, that's when I think people struggle… I'm trying to encourage her to see if she'd like to do the injection herself… physically… of course she can do it... but it's psychological … she'd quite like to control everything, but actually, she's so used to me doing it, she doesn't want to change anything."* (Miriam, VCSE professional)

The inseparable link between physical and psychosocial H&W was highlighted, especially in the context of behavioural response and in understanding the reasons underpinning observed behaviour. One participant drew comparison between JIA and the positive and negative symptoms of psychosis – positive (physical) symptoms reflecting the addition of new phenomena (*e.g.*, joint inflammation), and negative (psychosocial) symptoms contributing to social and functional decline (*e.g.*, depression, anxiety, social isolation, and aggression). Helping CYP to see the difference between physical and psychosocial symptoms, while realising that they are intrinsically linked, was viewed as a positive step forwards.

One participant also highlighted the importance of teaching CYP with JIA about emotional intelligence, citing inadequate access to psychosocial support as unhelpful. This participant eloquently described the hand model of the brain, a psychological model used in counselling to describe the different parts of the brain and what happens when CYP temporarily lose control of their behaviour (Siegel, 2010). The participant felt that reframing behaviours in this way, and equipping CYP to be more emotionally intelligent, would aid development of their self-regulation skills.

6.4.6 Honesty, respect, and encouragement

CYP with JIA are often bombarded with new terms, treatments, and procedures to familiarise themselves with. Although communication should be individualised, participants such as parents, Toby and Clara, felt information should nonetheless be accurate, honest, trustworthy, and evidence-informed, with CYP given credit for their developing knowledge:

> *"When all her friends were two and three and learning mummy, daddy, whatever… she was learning methotrexate, X-ray, MRI. So, she knows all the right words and she knows what they all mean."* (Toby and Clara, Parents)

A repeated point was made for information to be explained in a way that respects the context surrounding CYP's H&W, *i.e.*, the 'what, when and why', to promote positive behaviours early on:

> *"Instead of just going, 'you're going to take this tablet'… Why am I taking this tablet? Why is it the size of a horse tranquiliser? Actually if I knew that if I took one of those, then I wouldn't have to take another one for a week, oh, fine, I'll take it, but nobody ever said that to me."* (Betsy, Teacher)

Appreciation of the fact that CYP will have JIA for life, meant that it should be accepted that they will be responsible for self-managing JIA in the future. At the very least, it was felt that CYP should know the name of their disease, their medicines, and how often they take them, without seeking confirmation from parents. It was also felt that there may be benefit in providing education days, which some hospitals already provide, with specific resources provided to help CYP understand the different roles and responsibilities related to self-managing their H&W, to facilitate the transfer of responsibility from parents.

Providing CYP with the skills and specific tools to manage different aspects of their life with JIA was deemed important by participants, caveated with the understanding that there is no expectation for them to perform an activity immediately – rather, that they have the skill as part of their self-management armamentarium.

Another important skill for CYP to develop includes self-assessment of symptoms and side effects. In the waiting room before the consultation in paediatric rheumatology, CYP are asked to complete the CHAQ, though

participants like CNS, Caoimhe, felt that few CYP actually completed the questionnaire independently:

> *"You give it to the young person but still very regularly find that actually it's the parent that's filled it out."* (Caoimhe, HCP – CNS)

One HCP suggested that during this time in the waiting area, greater focus could be made to plan and prepare for the consultation, rather than simply filling out a form without thorough consideration. An extension of self-assessment includes supporting CYP to self-assess risk and benefits, in order to make informed decisions – particularly for topics that may not be addressed directly by HCPs and the literature. VCSE professionals remarked that such topics are those which they tend to discuss with CYP, highlighting the benefit of hearing from older peers living fulfilling lives, who are also aware of, and managing, the risks associated with their decisions.

Furthermore, helping CYP to pace was felt to be particularly important, as was supporting CYP with decisions related to disclosure and support entitlements. The need for H&W record summaries for CYP was also highlighted:

> *"They can't actually remember their own history about what drugs they've had and when they had them and what reaction they had to it, which I think that kind of thing is going to be quite difficult when they're on their own."* (Esther, Parent)

Perceived benefits of a non-categorical approach to building knowledge and skills was also identified as an opportunity to widen CYP's perspective, helping them to realise that there are others going through similar experiences:

> *"Perhaps it's quite good that some of the stuff they're given to watch is about other conditions… realising 'it's not just me, this child has a different condition – they have a daily injection, I've only got a weekly one. Well actually, mine's not so bad now…' [otherwise it is] just perpetuating the JIA, JIA, JIA thing..."* (Miriam, VCSE professional)

6.4.7 Timely access to support

Early intervention to prepare CYP in advance of medical interventions and self-management activities, were seen as important steps in ensuring they felt

informed, empowered, and in control – even at the most basic levels of handing over small amounts of control (*e.g.*, enabling CYP to choose which limb they will have their injection in). Participants felt that those who have been more involved with their care from the outset are seen to have more favourable outcomes, including a better understanding of their disease, and are more competent at self-managing their H&W.

Preparation for transition to adult H&SC services was brought into focus, highlighting the need for early discussions, typically during early adolescence. Although transition was sometimes seen as the final destination of paediatric care, some felt that transfer at the of 16 was too young, and that an age cut off is unhelpful. Participants indicated that more credit should be given to CYP who are already familiar with other transitions in life, such as moving from primary to secondary school, and how they cope with such changes. Viewing this retrospectively, and engaging CYP in conversation about how they feel about their increasing self-management role, was seen by CNS, Victoria, to be an important step in encouraging them to become more independent:

> *"It's having that discussion about, if your parents are still giving the medication, how do you feel about self-administering? Because if these children are going to end up going into adulthood with these devices and this treatment… mum's not going to be there to give it."* (Victoria, HCP – CNS)

Preparation for hospital and medical interventions were seen to ease CYP's anxieties and concerns, when approached sensitively. Some imaging procedures can be daunting, and while some hospitals provide written information, earlier use of such materials, and exposure to more interactive learning experiences, were seen to be potentially helpful, helping to make experiences more palatable:

> *"There's still quite a number of parents who take their child to hospital and avoid telling them they're going to hospital… of course for children that's so scary, if they have no idea what's going to happen, they are in a panic and then the procedure often ends in real upset."* (Judi, VCSE professional)

The psychosocial challenges of early intervention for younger CYP were discussed. In particular, with regards to enabling relative comparison (*i.e.*, CYP observing differences before and after an intervention and associating them

with particular outcomes). Recollection of this association was seen to be important:

> *"She remembered that before that she couldn't walk and she was in a lot of pain… she was happy having a needle stuck into her… [now], she doesn't remember not being able to walk… all she remembers of the pain… is the injection. So that's why she doesn't like it…"* (Toby, Parent)

VCSE professionals Megan and Chanda believed that early intervention to prevent crises was seen to be an important attribute for CYP to be aware of, so that they had instant access to a repertoire of information and strategies to execute in order to prevent worsening of symptoms, unfavourable outcomes, and potentially the need for unnecessary intervention:

> *"It's much more preferable for people to have the self-management skills before they need them, so that they've got that toolbox there to dip into…."* (Megan and Chanda, VCSE professionals)

6.4.8 Multichannel approach

The varied usefulness of printed, written information for CYP was touched upon; however, participants felt the future pointed towards digital and interactive channels, including for hosting written information like information leaflets. The increasing influence of online and smartphone applications was noticeable among participants, with prompts for further exploration on utilising and ensuring the longevity of different digital interventions in the context of self-management for JIA. Examples of serious games such as Toca Life: Hospital, were mentioned by some participants, as tools to help familiarise CYP with the hospital environment. Gamification was seen as an opportunity to optimise education and support, particularly for those in late childhood and early adolescence, who are often too old for resources aimed at younger CYP, but too young for resources aimed at those in late adolescence, approaching young adulthood.

Participants highlighted how videos about JIA, as well as different medical interventions, have the capacity to increase engagement and ease anxiety; often complementary to written information. Examples mentioned included NHS Trust-specific health-related videos, VCSE-based video sites, such as WWCiH;

and television-based videos and programmes, such as My Life, Operation Ouch! and Get Well Soon:

> *"She absolutely loves this programme… every episode takes the life of a child who has… a disability… she's incredibly knowledgeable about all these different conditions… she likes to hear about other people who feel slightly different, because she feels slightly different."* (Miriam, VCSE professional)

Interestingly, participants felt there was preference for live action content as opposed to animated content – with the former helping to contextualise real life for CYP. Indeed, the other health-related videos would often involve CYP undergoing different procedures, while featuring their families and HCPs. Exploration of this area with CYP would be an interesting future piece of work. Several participants discussed intentions within their organisation to produce videos, but highlighted resource constraints as a barrier, supporting the need for collaboration and sharing of information.

Play therapy and the role of the play therapist was discussed, particularly when preparing CYP for medical interventions. As well as aiding with distraction, play therapy was seen as an educative tool to ease anxiety. However, despite planning different strategies with play therapists (and psychologists in certain circumstances) around medical interventions, participants felt that strategies were disbanded during fraught periods. Roleplay was also seen as a useful way to examine and discuss how CYP can navigate challenges and positively change their behaviour; as long as it was used as a demonstrative tool which did not diminish CYP's capacity:

> *"They put… a tourniquet on a teddy, and… used the needle… I was sat there looking at this nurse going, is she stupid? I think sometimes we forget that yes, children are children… [but] they've got that cognitive response… they know that it's a teddy."* (Betsy, Teacher)

Play therapy and roleplay were quickly associated with younger CYP, but remarks were also made for the absence of such interventions for older CYP, in a more age- and developmentally-appropriate format, such as serious games, virtual reality, and augmented reality.

Meanwhile for older CYP, platforms like Arthur's Place (Stones, 2017a) seemed to be more appealing, where there is a mixture of content developed for and by young adults addressing relevant topics pertinent to self-

management. However, there was a feeling from Kendra, one of the Youth and Family Support Workers, that for the time being, there is no comprehensive replacement for face-to-face interaction:

> *"I don't think there'll ever be a replacement for the face-to-face stuff… there's a lot of apps… it's kind of… the reason probably why that person is having a difficulty… because… current relationships aren't giving them what they need."* (Kendra, HCP – Youth and Family Support Worker)

6.4.9 Social influence and peer support

VCSEs provided several age-appropriate support opportunities for CYP, though provision can be challenging when ensuring all age groups are catered for. While informal parent peer support gatherings with younger CYP may be suitable, older CYP may wish to grow beyond the confines of being with their parents in a support setting, as Xena alluded to:

> *"When you become an adolescent, you're wanting to do things separate from your families. You're building your own friendship groups… so that's why our events… are just for the young person."* (Xena, VCSE professional)

Access to activities which encourage peer support may also be those considered unconventional SSMIs, but those which provide a safe space for CYP to socialise and experience 'normal' activities that their healthy peers may take for granted. Some of these activities, including outdoor activities, are offered by VCSEs, as Darcie had experienced herself:

> *"We did some rock climbing thing and went in a cave [at the CCAA (Children's Chronic Arthritis Association) weekend]… [nodded when asked if it was nice to be with other people who understand JIA]."* (Darcie, Child aged 5–7 with JIA)

It was also highlighted that those in early and middle adolescence can often be overlooked, yet still require enjoyable, inexpensive, age-appropriate support. One example given was a CYP meet-up, led by two members of the paediatric rheumatology MDT, involving an escape room experience and dinner one weekend. Enabling support to naturally happen in an informal, and unstructured setting was felt to be beneficial to CYP, supported by the familiarity of known

HCPs *in loco parentis*, which was a key reason for families allowing CYP to attend.

Peer support can also be implicit – in that, CYP may not speak about JIA, but there is a general appreciation that everyone understands their perspective, which is why social features of SSMIs appear to be useful:

> *"[She] had a lot of sickness… she'd never met anybody else with arthritis. She came along… was chatting away and somebody on methotrexate said, 'oh, yeah'... and she said, 'what, you also get sick?' She couldn't believe [it]… she [thought she] was the only one that… got sick."* (Audrey, VCSE professional)

This implicit peer support may also extend into observation and modelling behaviours, even amongst those during infancy and early childhood, such as seeing other CYP experience venepuncture. However, it may not be in the manner in which is often presented in evaluations of existing SSMIs:

> *"I don't think that can always work through forced groups, like forced support groups. I think you're lucky if you find someone locally going through the same thing and your kids bond."* (Toby and Clara, Parents)

This raises an important point about the reality of peer support, and obliging different CYP with diverse backgrounds and preferences, to befriend each other in an unnatural environment, which may be counterintuitive to the underlying goal of providing peer support:

> *"It's the same as putting me with somebody… and being like, I expect you two to be friends. And for some people, people with long-term conditions, they think, oh, yeah, we'll put them [together]… if they've got nothing in common, don't do that."* (Kendra, HCP – Youth and Family Support Worker)

However, socialising with healthy peers was considered as important as formal peer support, though this can be challenging and frustrating if friends do not understand or empathise with CYP's difficulties. There are also examples where older CYP have messaged each other via social networking platforms, but the longevity and strength of these connections appeared varied according to participants. One example included CYP doing their physiotherapy exercises via live video chat, which motivated them to adhere to their treatment by modelling their peers. A more implicit value of peer presence for some CYP

could be the exposure to a wider number of peers with varying LTCs – including some in worse health, but with different outlooks. VCSE professionals remarked that such exposure could help those with low self-esteem and narcissistic traits:

> *"Someone said that while being at the workshop, they realise that they're not the most affected person with arthritis… it maybe something silly that you know you are comparing yourself, but I think in a way… if you feel pity for yourself and then you see that someone is actually in a worse situation, that… could be empowering."* (Megan and Chanda, VCSE professionals)

Another example of peer support in a clinical setting included a noncategorical youth group, which was regularly attended by the same group of CYP who formed a bond and took initiative to engage in conversation beyond the group. However, one of the downsides was the spiralling effect of negativity, which worsened their mood in some instances. The youth and family support worker highlighted how this instance prompted them to provide more structure for the group, while prompting CYP to set boundaries to protect one another.

However, a difficulty for some families was having the contacts to link CYP up with each other, especially for those unaware of existing opportunities via VCSEs, which is where conversations around peer support were felt to start. Beyond peer support, the value of peer encouragement in education- and skills-based self-management training was viewed as essential for motivating CYP to take control:

> *"Your friends are key, and therefore having JIA friends is going to be a big factor in whether you're prepared to take stuff on yourself."* (Miriam, VCSE professional)

An extension of peer support includes the presence and influence of role models, such as older adolescents and young adults with JIA, who are successfully self-managing their H&W, and see the benefit of looking after themselves. Participants felt that CYP are reticent about self-management because they tend not to see positive examples of how it would practically work:

> *"Young people are more likely to listen to other young people than… doctors… I think we need to use that for information for teenagers. They need to hear it from someone else their own age"* (Judi, VCSE professional)

Providing the space and opportunity for CYP to be able to talk with such role models could facilitate how they envisage their future, helping to put them at ease, particularly given the anxiety that future uncertainty can cause:

> *"I just want there to be a talk… about somebody who got to university… all of school and got a job. I just want to see somebody's normal life and how that progressed."* (Kendra, HCP – Youth and Family Support Worker)

However, one of the more unique aspects of VCSEs offering JIA SSM services is that many of the volunteers, and in some cases staff, have a personal experience of JIA. For certain VCSEs, their services are supported and delivered by young adult or parent volunteers:

> *"All the volunteers are… young people with arthritis… [they] are leading the discussions. These people are really positive role models… a lot of [other VCSEs] run… events with parents. I think having parents with teenagers totally changes the dynamic… I think there is absolutely scope for some of that, but I just think… there should be a balance."* (Hope, VCSE professional)

Furthermore, the presence and visibility of youth workers with lived experience of JIA linked to their rheumatology MDT can be comforting for CYP, encouraging them to relate and be able to feel more at ease:

> *"When they come along to their clinic appointments, I don't think they expect to meet someone else who has the condition… some people are quite surprised when… [I] mention that I [have] arthritis too… that does help them to relate."* (Xena, VCSE professional)

6.4.10 Positive reinforcement and experiential learning

Various methods of positive reinforcement to motivate, incentivise, and distract CYP were suggested, particularly for promoting adherence to treatment and agreement to undergo medical interventions. These included stickers, stamps, and confectionery, generally seen to be most useful for younger CYP. Agreeing on appropriate forms of positive reinforcement for adolescents seemed more challenging. Shifting focus from bribery to positive reinforcement was felt to be an important perspective for everyone involved, in an attempt to remove the negative connotations associated with bribery and coaxing:

"Positive reinforcement doesn't need to be a thing, it can be an experience. So anything else that might be a bit scary, you can then do something good, and it has the same effect." (Betsy, Teacher)

"I get ten stamps and I get a lucky dip." (Darcie, Child aged 5–7 with JIA)

VCSE professionals remarked how they had witnessed a number of CYP experimenting with non-adherence to treatment. This 'trial and error phase' appears to be a common experience, and therefore, enabling CYP to learning from their actions, was felt to be part of the process of CYP becoming competent self-managers:

"There probably are young people that haven't taken their medication [on a weekend away], but have realised on Monday morning they are sorer for it… I think it's really important that they learn… they know the difference then if they take [their treatment]." (Megan and Chanda, VCSE professionals)

Experiential learning was also viewed in terms of modelling the behaviour of others, either by CYP with JIA being in the presence of their peers with JIA, or simply knowing that there are other CYP like themselves having to go through similar experiences:

"When she met this other little girl who's similar in age… that was such an encouragement… she thought oh, that same day that she has her injections this other girl [is] having her injection." (Toby and Clara, Parents)

Participants remarked on other ways to incentivise CYP, including developing strategies to cope with JIA while achieving their goals, and performing self-management roles in order to stay over with friends, for example. Moreover, HCPs were seen to play a role in incentivising CYP by setting goals. Participants felt this was strongest when it came from the consultant. By helping CYP to progressively achieve different self-management-related tasks, participants said that this could be one way in which goal setting towards competent self-management could be achieved:

"If the consultant… said… I'm seeing you now you're 10… by the time you're 11, let's pick two of the things off this list that you can take on yourself'… I can see my daughter going for that, because actually, somebody else has told her." (Miriam, VCSE professional)

VCSEs appear to be increasingly interested in using goal setting as part of their SSMIs at a group level, such as through accountability partners, whereby CYP could build reciprocal relationships over time, while coaching one another to achieve their goals:

> *"A lot of that comes from that peer support environment, where it's encouraged… a safe place… it becomes a much easier place to set a goal and work towards it and know that you're going to see those people again and continue to be encouraged."* (Megan and Chanda, VCSE professionals)

6.4.11 Autonomy

Participants felt that parents can help to encourage a sense of autonomy amongst CYP by setting clear and consistent expectations, communicating openly, staying calm, allowing CYP control over decisions, and by discouraging rebellion, thus complementing their growing desire to start to take responsibility for different tasks:

> *"I missed out on a lot of things, but when they [parents] had the opportunity for me to be able to choose something, then that's what I did."* (Betsy, Teacher)

Listening to, and understanding CYP's individual outlook may help them on their journey to be in control. For some, being in control may provide them with an increased level of freedom, such as self-medicating. HCPs were seen to have a particularly important part to play in encouraging CYP's developing sense of autonomy, by allowing them the space and opportunity to speak in clinic, involving them in SDM, and in encouraging them to take ownership. This was viewed to be as simple as listening to CYP when they are ready and showing interest in doing things for themselves:

> *"If a child's directly involved in their care… encouraged to talk, to bring their own questions to clinic and to describe their own symptoms… that's a help towards them thinking about what's needed to manage [JIA]."* (Krishna, HCP – Paediatrician)

The influential and powerful role of HCPs to promote autonomy was referenced several times, when HCPs remarked on the importance of directing questions

and responses towards CYP, instead of parents, while also counselling CYP to be informed as to what their treatment entails:

> *"For your older children, we make sure that when we are going to do counselling, we are involving them… the consent comes from the parents, but it's them that have to adhere to the treatment, and the understanding of why they're having it… best to get them on board, from day one."* (Victoria, HCP – CNS)

This extends to supporting CYP in performing medical interventions necessary for managing their H&W, such as self-administering injections. Resources are available to safely help CYP feel comfortable with self-administration, such as demonstration pens, and HCPs remarked that most are willing to get more involved, if given the opportunity to do so.

However, CYP have got to want to take on some responsibility for their H&W, thus finding themselves at the right place, psychologically, to feel able to take control. Participants felt that this may be where psychological intervention to support CYP in understanding that JIA is their condition and they need to be responsible for it, may be useful.

Providing CYP with the opportunity to be self-reliant, by making choices and taking ownership, as early as possible, was seen to be key to equipping them with the confidence to self-manage their H&W over time. Examples included seemingly small decisions such as sitting on their parent's knee or in the chair on their own for venepuncture. Subtle choices were seen to give CYP the tools to enable them to have a say in their treatment, and thus some level of control.

6.4.12 Willingness to accept responsibility

Some participants, including paediatric rheumatologist, Tom, felt that CYP are quite capable of undertaking certain self-management tasks, such as self-administering treatment, but when their parents do it for them, they will quite happily allow them to proceed. This can be problematic for CYP who are generally more reserved, dependent on their parents, and hesitant to change:

> *"When the young person comes to my clinic with a flare up of arthritis and I ask them, 'what are the joints that are painful or swollen in your body…', they are… not able to even answer this simple question… they're confused, they look at their parents…"* (Tom, HCP – Paediatric Rheumatologist)

Participants recognised that cognitive dissonance may contribute to CYP's perceived lack of willingness to self-manage their H&W – adhering to protective features which feels safe and familiar. Strategies to help overcome cognitive dissonance often revolve around the family approach to the transfer of responsibility, where parents provide prompts for CYP to take control:

> *"We've offered for her to do her own methotrexate injections and she just freaks out at the idea. But it is an open offer."* (Toby, Parent)

Similar to other aspects of increasing self-management responsibility, a balanced approach was seen to be required, so that CYP can become more involved in their care, in an age- and developmentally-appropriate manner, requiring the necessary input and preparation beforehand to ensure they feel comfortable and confident:

> *"You look at the positives and think, well now, he can sit in a consultation without welling up and crying… he can ask questions if he wants to… he does reach out if he needs help… it's the small wins…"* (Kendra, HCP – Youth and Family Support Worker)

An aspect of readiness for young adulthood involves building character and resilience, which can be unnerving when entering unchartered territory. HCPs remarked that on paper, transition from paediatric to adult H&SC services appears to be working, but the reality tells a different story, with many CYP entering adults non-adherent to treatment, consequently experiencing disease activity requiring more intensive treatment. During this period of transition, participants felt that current support does not necessarily set expectations of how adult services are different, and how CYP may struggle with certain routines:

> *"The access to the service is so much different… sometimes it can be a bit of a shock… they do have helplines, but it might not necessarily be a question that's answered for a few days."* (Victoria, HCP – CNS)

The need for some form of keyworker support was highlighted, whether that be a CNS or youth and family support worker, to help bridge the gap between the support offered by the paediatric rheumatology MDT, and the void in adult rheumatology, especially in the absence of specific adolescent and young adult

services. Having somebody to advocate for them, other than their parents, may be the support CYP need to help identify what they would like to discuss with HCPs, in order to navigate the new adult environment until they feel comfortable, competent, and independent:

> *"They need a cheerleader that's not their parents… you do really have to break things down for some people, in order to make them more comfortable with what they've got to do."* (Kendra, HCP – Youth and Family Support Worker)

6.4.13 RQT1 Summary

CYP must navigate a stigmatising environment in the pursuit of normalising life with a variable disease like JIA, with different support structures in place depending on individual CYP and families. Timely access to individualised information, education, and support, using a multi-channel approach embedded within a peer support structure can impact on CYP's knowledge and beliefs, self-regulation skills and abilities, and sense of autonomy, thus improving their sense of self-management capacity throughout childhood into adulthood.

6.5 RQT2: Shared-management of JIA by families

6.5.1 Family dynamic

The home environment and the functioning of the family unit can impact the way that families cope with the shared-management of JIA. It can also influence how families interact with HCPs, with participants commenting on those who feel competent to be self-sufficient in-between consultations, compared to those who require constant medical advice. With advances in care and treatment, more management can take place in the home, which also shifts the responsibility of care from HCPs to families. Recognising the uniqueness of each family, and tailoring information and support to their needs was seen to be important, but also quite challenging practically. To help meet this need, participants felt it was important for HCPs to build relationships with families, so that they have an opportunity to develop a better understanding of the family dynamic:

> *"I would want to know [at diagnosis]… what is the family dynamic, who is around, what other siblings are there, how do you think this is going to impact them, whose going to be responsible for giving medication, what if you have to be away, will there be a backup, do you have family nearby? I don't know how families would feel about being asked those kind of questions… but it's to do with how it's explained to them."* (Miriam, VCSE professional)

Indeed, families come in a variety of shapes and sizes, with varying levels of support networks in place. Participants related this back to the types of families accessing VCSE services, in particular those who self-refer. The likelihood is that the families accessing VCSE services are those who are able to proactively seek support, leaving a large proportion of families without access. The responsibility may also fall to one parent in two parent families, which can influence the way in which CYP relate to, and trust their parents, and how this impacts on their willingness to accept responsibility over time.

JIA can have a profound impact on the entire family, with some families seldom meeting others living with JIA, particularly if they are unaware of support opportunities. As well as the physical and practical demands of JIA, such as taking time off work, JIA was felt to be emotionally demanding too:

> *"If you think about the time parents have to take off work to look after children, to take them to these appointments, to deal with them when they're off ill, that's huge, and it's not necessarily the health service's problem, but it is a knock on impact…"* (Miriam, VCSE professional)

For siblings, the SSM of JIA was seen to intrude on family life, providing unpleasant and traumatic experiences for siblings. Participants remarked that attention may be diverted unintentionally towards CYP with JIA, since they may require more help or support. This can come at the expense of other family members, namely siblings, who may feel resentment or frustration that they are being restricted or overlooked:

> *"I saw… a talk… it was… entitled 'I wish I had JIA', and the focus was on siblings… I don't know if he actually would think that he wishes he had it but… the amount of attention can be a bit unequal."* (Helen, Parent)

However, there was some conflict regarding this stance, since some HCPs said that they had not experienced these issues in practice, prompting thought on whether there are variations in the level of impact on siblings, or whether the

impact is simply not discussed. Siblings can, however, be a source of support, though this may often go unrecognised. It was identified that such siblings may benefit from being registered as young carers with their local authority, providing them with opportunities to access support themselves. Similarly, siblings also found solace from attending VCSE events, since this helped to reaffirm that they were not the only CYP who has a sibling with JIA. Unfortunately, siblings may also become more anxious for their loved one with JIA:

> *"The youngest one has found it really difficult… it's just little things like… we have to think about what we're doing all the time… that impacts on [the younger sister] and the stress and everything as well… she worries about [her sister with JIA]."* (Esther, Parent)

Some HCPs remarked how they had utilised services offered by non-JIA VCSEs, such as Rays of Sunshine, to offer families an opportunity to have an enjoyable event where they could momentarily forget about JIA:

> *"We've just helped a family… they've had a whole family trip down to London… the child's been so unwell recently, it's just been taken over by what's been going on with the JIA, and the treatments… we utilise the charities… to try and bring that family back together."* (Victoria, HCP – CNS)

6.5.2 Parental perspectives

Proactive parents tend to be those who are more confident at the shared-management of JIA, while also recognising the need to engage and empower CYP to become increasingly competent at self-managing their JIA over time:

> *"We try to keep her involved in what's going on and why. So, she knows the names of most of her doctors. She knows all the medication she's on. We try to explain why she has them and why she has MRIs and things like that. So, we can obviously keep going forward with doing that and explaining what's happening."* (Toby and Clara, Parents)

Higher levels of education amongst parents were referenced as being potentially supportive of their ability to research and interpret JIA literature, highlighting the disparity against parents who do not have the education or skills to be able to search for information independently:

> *"I've got a very academic background and so for me… researching all of these things, I can do it. I'm pretty confident at talking to people and finding out information… I'm probably making [my daughter's life] better because I'm able to do those things but a lot of people wouldn't be able to."* (Esther, Parent)

Different activities which parents had proactively undertaken included diary symptom monitoring; however, in retrospect, some wish they had documented more items and events, such as symptoms, timelines, and each treatment trialled – at the time believing everything would be documented clearly in the medical record.

Parents also recognised when proactive shared-management behaviours had become lapsed, such as adhering to physiotherapy exercises; though they also recognised different patterns and adapted physiotherapy according. For example, if CYP were more physical activity during the day, physiotherapy was overlooked. Similarly, those who were informed and supported to be able to self-assess their child's H&W felt in a better position when identifying when to seek input from HCPs (*e.g.*, General Practitioner [GP], CNS, or emergency department), while others may have struggled unnecessarily waiting for the next consultation with the paediatric rheumatologist.

For many CYP, their parents will manage their JIA by proxy; often out of necessity for younger CYP lacking capacity to self-manage, but also for older CYP and adolescents, since participants felt that parents generally feel a natural and subconscious responsibility to care for their child, regardless of age, and potentially because it can be more convenient, especially if their child's JIA is well managed:

> *"I think I was probably guilty of this, of thinking I didn't really need to do anything about it because she was fine and I was fine, and her condition was managed. So therefore, why bother bringing it up again and making it a big thing."* (Miriam, VCSE professional)

However, this can begin to be detrimental over time, especially when CYP are not involved in SDM with families, conditioning CYP to passively rely on others to make decisions on their behalf:

> *"Just ask them [parents] how they would feel if somebody just came up to them and did it [injection] without communicating or without asking them anything, just because it was going to be quicker."* (Betsy, Teacher)

This may prompt the need to revisit different aspects of the diagnosis over time as CYP develop, while supporting parents to take a step back from their caregiver role, as paediatrician, Ava, alluded to:

> *"We try and talk to [CYP] and say, how are you, what are you doing, was this happening? They then turn to mum or dad and say you tell them… parents then will generally step in and that carries on with lots of them… there are certain ones where you… feel I really do need this [parent] not to be in the room because they are probably hampering how this is going."* (Ava, HCP – Paediatrician)

The parental perspective may be different from CYP's perspective, particularly when considering how CYP feel 'different' to their peers – parents will likely know this, but do not necessarily view it in the same light, or not to the gravity that CYP may experience. Helping parents to have more informed and honest conversations with CYP was seen as important for building trust between CYP, their parents, and HCPs:

> *"If you tell your child you're going to McDonalds and you end up in hospital, your child's not going to trust you next time you're going somewhere. The same about… 'needles don't hurt, it's fine, you won't feel it', and of course you will. So it's important that parents are honest with their child."* (Judi, VCSE professional)

How parents are coping, and their mindset, will inevitably reflect on CYP's long-term coping ability. Parental anxiety is a phenomenon which can affect CYP's symptomatology and outlook, though parents are often overlooked as being in need for support, despite the link between their anxiety and CYP's anxiety. Enabling parents to be optimistic and more confident in managing JIA was regarded to have a positive effect on CYP's H&W:

> *"If the parents are… visibly upset and they've got loads of anxiety and they express [that], then the child will detect that… and the child would then quite often not cope as well… it's really much better… if they project… confidence… 'we will be able to manage this'… even if that's not what the parents are thinking, it really gives the child a strong sense of resilience and a positive approach to managing the condition."* (Krishna, HCP – Paediatrician)

Participants remarked that parents living with LTCs themselves could sometimes project their own experiences onto CYP, which if parents were

unsupported to self-manage effectively, may be reflected by CYP's self-management behaviours. An example of where information has enabled parents to feel more informed include the WWCiH videos, which has helped to reassure some parents, who consequently feel calmer – breaking the negative spiral of panic between parents and CYP:

> *"They struggle seeing their child in pain… it really increases anxiety for parents, so the parent is often very stressed… [but with WWCiH videos, the] parent can watch it and understand better what's going to happen… and when the parents are calmer, the child is often calmer as well."* (Judi, VCSE professional)

Families often find themselves juxtaposed between dealing with the present, and envisioning the future. Participants highlighted how for some parents, the uncertainty of their child's future is too overwhelming. Parents may want to ask questions about prognosis, but may feel reluctant to do so in the presence of CYP out of fear of causing anxiety:

> *"There's a sort of aspect of not wanting to consider that we might need [VCSE support] down the line, but we have to have that at the back of our minds as well… our focus… is that we're just dealing with the now…"* (Helen, Parent)

Interestingly, for those families whose CYP may be coping reasonably well, there can be anxiety around reading stories from others who experienced deterioration or additional challenges:

> *"At the time when [my daughter] was reasonably well… reading stories about how people were okay and then got a lot worse afterwards… I just though, actually that could be more detrimental to be reading all of that."* (Esther, Parent)

The overwhelming news of a diagnosis, and the grieving process that families may go through, can often be exacerbated when the diagnostic process is poorly managed, in terms of information and access to support:

> *"After that appointment there was no follow-up… it was here's your appointment, here's your information, off you go, goodbye… there wasn't anyone who then stepped in and said actually, you've just had a really… major diagnosis… it'd be… very helpful to know there's someone available, perhaps phoning out, rather than a parent having to go in."* (Toby and Clara, Parents)

Access to such information and support is not only relevant at diagnosis, but strategically over time too. Too much input too soon may be counterintuitive, requiring a balance between awareness of what is available, but without the pressure of processing excess information at an already difficult period. Information regarding transition, for example, would be inappropriate for the families of young CYP at diagnosis, but will be of greater importance in future years. Therefore, having a way of getting the right information, at the right time, in the most accessible ways possible, was seen to be an important consideration. This also recognises the need to look beyond the medical focus of 'diagnosis, treatment, and remission', to pre-empt support, which may not be needed, but would be more appropriately addressed from the outset:

> *"For a lot of people, probably they get the treatment started… [it's] effective… they don't necessarily think that they need anything beyond that. But… as we all know, it's a bit of a rollercoaster, and it's only when the wheels all come off, then actually it would have been helpful if you had that support network in place."* (Miriam, VCSE professional)

Participants acknowledged that there are likely to be large groups of families who may be perceived as 'hard-to-reach', but are in fact just not being reached, and it is these families who are most likely in need of information, education, and support. Indeed, consideration has not always been paid to the H&W of CYP's families, which influences how they and their child interact:

> *"You… need to be careful because the parents that you have who are engaging with you are parents… who want to see a way forward, who are quite articulate… onboard… but there is probably a cohort of parents that you are not touching and they are the ones who are not engaged."* (Martha, Teacher)

6.5.3 Relationships with professionals

Cementing the relationship between families and HCPs was seen to be an important part of building trust. Examples were provided of how certain centres proactively engage families by hosting new patient/family coffee mornings. Indeed, by building better relationships between families and HCPs, the likelihood for improved communication and SDM may be higher. It may, however, involve some 'difficult' conversations, such as HCPs understanding the specific needs and abilities of families, communicated appropriately and sensitively:

> *"There are times in the journey where families need a lot more reassurance than others… it's to do with how it's explained to them, that they are going to have to be part of their child's care, which is a good thing, because it keeps their child out of hospital, but it's the hospital's responsibility to ensure that they're well set up and with the right support in place."* (Miriam, VCSE professional)

Continuity of care is important for many families, particularly amongst certain HCPs, such as the CNS. Families feel reassured when they know there is somebody they can reach out to for support, even though they may not need regular contact. Participants felt it was more the principle of knowing they had a direct contact at the hospital, who recognised that CYP and parents cannot be separated from a practical and emotional support point of view:

> *"[The daughter] went in for a scan… I went and had a coffee with her mum, because I could see her mum was really struggling… To disconnect the two is silly, because they're supporting [the child] and your work will go further… it doesn't take anything to just ask how they are, because they know that you care about all of them. Treating them as an isolated person, it doesn't really work, because then you don't see how the family works together."* (Kendra, HCP – Youth and Family Support Worker)

For certain families, it was recognised that the same, named HCP was pivotal for maintaining balance, and so teams would do what they could to help ensure such continuity of care could be provided. As is discussed in RQT5, service delivery varies across the UK. For those with access to community nurses or CNS performing outreach services, it became increasingly clear as to how those HCPs develop close relationships with CYP and families. By seeing them in clinic and in their home environment, these HCPs said that they

surreptitiously generated a picture of the family unit so that they could tailor information and support on a case-by-case basis. They also helped coordinate practical aspects of managing JIA, such as blood monitoring, counselling CYP on new treatments, and providing telephone helplines, text messaging and email support:

> *"It definitely works by going through everything with them… that's definitely something that's good about our team… we have the time to be able to do that, and again, it's building those relationships up…"* (Victoria, HCP – CNS)

While families appreciated the workload and pressures that HCPs are under, they also strongly remember many aspects of their interactions with HCPs, including body language, expression, tone, and specific words used, which can subsequently impact the way in which families interact with HCPs:

> *"We then got taken to a separate room with the rheumatology nurse… she didn't even sit down. She kind of stood, almost like she was in a hurry to get away… and it just felt a little bit of a rushed five minutes... and then when we did try and make contact… she never got back."* (Toby and Clara, Parents)

6.5.4 Parental education and understanding

Parental education cuts across most of RQT2, demonstrating the power of knowledge in influencing behaviours conducive to the shared-management of JIA. At the point of diagnosis, unless they have prior experience, many families felt unaware of the different care available, including knowledge that paediatric rheumatology is a specialism. Some other paediatric specialities hold an education day for families post-diagnosis, though this does not happen consistently. A challenge thus remains as to who is responsible for educating parents, who are not seen as patients, but equally, when informed will influence their child's outcomes:

> *"Unfortunately, too often, units don't see educating the parents as an important part of their job… they do it if it's around health and safety… but I think that's very little and needs more education for parents generally… the more training and education… the better the outcomes for that child."* (Miriam, VCSE professional)

The level of understanding of JIA varies enormously, as anecdotally evidenced within closed groups on social networking sites where parents appeared to have a poor understanding of JIA:

> *"We're on a couple of different Facebook groups… and the number of people on there… from their questions have very little understanding of [JIA] or what the treatments are… yet we assume that they're given the same information. They've got the same access to information… but they just have no real concept of what that means… and that's not the fault of the person receiving the information… so there has to be a way of interpreting that into something that makes sense for them."* (Toby and Clara, Parents)

Equipping families with the knowledge and skills to be able to identify credible and accurate information about JIA is one of the core aspects of shared-management. Participants felt this has become increasingly difficult in the internet-era where there is an abundance of readily-available information via search engines and social networking sites, though families who are at the start of their journey may not know which sources they can trust, or indeed what to search for. These challenges highlighted the need for an objective summary of different information, education, and support services that are available. Evidently, there needs to be a balance of information, in terms of quantity, and quality, ensuring information covers every aspect of JIA, so that families feel they have a rounded and well-informed overview:

> *"Some people are given too much information, because we had an awful lot of detail about methotrexate and lots of different phone numbers were given… but actually, overall, we don't get enough information about the condition and general support… there was a leaflet, a little bit on the emotional side of things… but not in any level of detail."* (Toby and Clara, Parents)

Participants felt that information should be provided in a manner that does not overwhelm families, especially at the point of diagnosis, but equally, not in a way where the information can be quickly disregarded. This can be aided by HCPs empowering families to ask question and develop their understanding:

> *"If you send it out in the post, it's just easily… thrown to the side… and then they've not got the right information, before they've started… there might be something they don't understand… and there's no one to ask… and they've got to then make a point of ringing us… it's definitely easier to just go through it with them."* (Victoria, HCP – CNS)

Furthermore, participants felt that a balance was required between excess information on certain topics, balanced with a more strategic impression of the road ahead, such as all treatment options available, and their mechanisms of action, which could help to demystify some of the concerns families may have. While information is available about different treatments, participants felt that there are fewer examples of the process and order in which they are used. It seemed that there is not necessarily a need for more information; rather, a process for assembling information so that families can access the right information, at the right time:

> *"I wonder if there is a way of helping parents to become aware of that bigger picture… yes, it is a big deal for you but in the overall scheme of things this is what's needed… just to reassure them."* (Toby and Clara, Parents)

While comments were made regarding parents having access to a central contact point at their hospital, it was also raised that parents need to be able to self-assess their child's H&W and level of urgency, seeking appropriate medical intervention as and when appropriate, such as through the GP or emergency department.

A common, and somewhat unhelpful goal conjectured by families is that of a 'cure', or 'growing out of JIA'. While that may be the goal in an ideal world free from JIA, it is not the reality, and so aiming towards 'cure' may do more harm than good. Helping families to understand that remission is a more realistic goal, could help them to conceive that while JIA is a life-long diagnosis, it does not necessarily mean that symptoms will always be the same, even though treatment may be maintained when asymptomatic:

> *"Sometimes we do find that when their children are well, it's like, 'why's he on treatment, if he's well? Why do we need to give him this injection every week?' But it's the injection keeping him well. So you don't want to be taking that away."* (Victoria, HCP – CNS)

Similar to the multichannel approached required for CYP, there is also need to ensure that information is accessible in various formats and through various channels for families, recognising their preferences for receiving information, with written formats still favoured for getting across key points, particularly during the initial post-diagnosis period.

6.5.5 Recognising parental expertise

Through their shared-management role, parents often become expertised in JIA – they know their own child best. Consequently, parents need to be recognised and valued for their expertise, as they are essential members of the MDT involved in caring and supporting CYP with JIA, perhaps more so than in the past, with an increasing level of care shifting to the home environment:

> *"Like it or not, the parent is part of the MDT. They might not choose to see it like that or accept that that is the case, but 24/7 that parent or parents are giving that care, and being asked to give that care, and have that worry and that responsibility on their shoulder."* (Miriam, VCSE professionals)

As a result of their experiences, some parents also go on to advocate for other CYP and families:

> *"I write about the emotions we go through, because I don't want other people to feel alone and ashamed… I think that's something that needs to come to the fore a lot more."* (Toby and Clara, Parents)

6.5.6 Receptiveness to accept and access support

Parents seem themselves as the protectors and supporters of CYP, appearing strong in the face of adversity. This can, however, cause them to overlook or refuse support:

> *"They don't want to be seen as weak and needing extra help… they don't want to be seen as not having the capacity to support their own children."* (Betsy, Teacher)

Reminding parents that accepting support is not a weakness, was felt to be important from the point of diagnosis. However, their receptiveness to accepting such support, or information about support, may vary between individuals, but also temporally. Participants remarked that for some parents, the immediate post-diagnosis period is a time when they find as much information and support as possible to manage post-diagnosis anxiety, whereas for others, they may need time to accept and adjust, potentially going through a period of denial:

> *"We saw they came back… a year later, and the difference was amazing… they weren't as anxious, and they were great in terms of what they were sharing with other families…"* (Audrey, VCSE professional)

When parents have come to accept their child's condition, participants remarked that there can then be further hurdles of accepting that they and their child require support, which may feel unnatural. This may require intervention from HCPs, to support parents in seeing the value of support, and the potential beneficial impact it could have:

> *"Accepting and turning up for that support… isn't always natural to all families… there's sometimes stigma around it… [but] to almost force the parents to accept that they might not naturally want to go to meet a bunch of strangers, but gosh what a difference it would make."* (Miriam, VCSE professional)

There are, though, many who feel isolated from other families living with JIA. Although there are VCSEs across the UK providing different types of peer support, access is variable, and awareness sometimes materialises incidentally:

> *"So on Facebook, I'm a member of a parents of kids with JIA group and I think there was a video shared on there… when I saw the CCAA weekend last year… I leapt at the chance to go… it was brilliant, [my daughter] absolutely loved it. We went as a family and so everybody was joining in together and [my son] really enjoyed it and made some friends."* (Helen, Parent)

Indeed, social networking sites can be readily accessed, enabling parents to feel more confident in asking questions and discussing different points that they would not otherwise feel comfortable doing in a face-to-face setting. It has been shown to be particularly useful during crises, when families need a quick answer from a group of people whom they can trust:

> *"If you're having a meltdown in the middle of the night… and you've just lost it because you can't accept things anymore, somebody will be online. A lot of the hospitals are scared of the online stuff, but it really is a lifeline."* (Miriam, VCSE professional)

This aligns to what has previously been alluded to, in that online support may be a bridge to face-to-face support, with the latter still valued for developing lasting bonds and friendships:

> *"You don't want a leaflet, you want a hug and a cup of tea, and somebody who can say, 'don't worry, we've been there, things will get better, and there's things you can do to help'…"* (Miriam, VCSE professional)

However, online support can sometimes become competitive, where some families begin to feel inferior to others who may feel that they have it worse than others. Families like to see positive stories from the community, including young adults coping well with JIA, which gives them hope. HCPs highlighted how the JIA community anecdotally appears to be quite active in terms of sharing information and support, compared to other disease areas.

Frustrations can appear, though, as a result of inconsistent, or non-existent, signposting to available support, namely from HCPs, some of whom may be averse to signposting to social networking sites in particular.

There are also specific VCSE groups for JIA across the country which encourage local gatherings, when permitted. However, families said they are still looking for the chance to connect with others in their area. One logical, potential solution suggested by participants was to encourage peer support and networking within the clinic waiting room, where there are likely to be one or more families living with JIA waiting to see the paediatric rheumatologist. Suggestions were provided for indicators to help families identify each other, such as wristbands or designated networking areas of the outpatient department, to highlight that they are a JIA family and they are happy to talk to others. Wristbands appeared to be quite popular amongst participants, featuring VCSE logos, and given at the point of diagnosis, acting dually as a prompt for HCPs to highlight the support available for families – thus potentially aiding families in reaching out to support services, while providing them with a tool to utilise. Interestingly, some centres have trialled formal support group-styled conversations within the clinic after consultations with the CNS and paediatric rheumatologist, according to one participant. Though participants recognised that people may not be in the right frame of mind to have conversations in the clinic:

> *"If you knew you were in a clinic where all the people that were at that clinic were other children with JIA, you kind of might just sit and talk to people… I guess some people might not want to talk and that's fine… but… a kind of, JIA badge with a green light, yes, come and talk to me might be quite nice."* (Esther, Parent)

Similarly to CYP, it can be problematic to begin 'matching' different families with each other. HCPs and VCSEs are often asked to connect families, though this can be challenging since not every family is going to get along, whereas if

they were able to decipher such relationships for themselves, then more natural and valued peer support connections may follow:

> *"I do think peer support would be at the top of the list, because that's something that we struggle with… it's actually really hard because linking one family with another family…. Would that be the right family for them? Whereas if they do it themselves… talking to each other, they make their own decisions."* (Caoimhe, HCP – CNS)

Professional support for parents was also highlighted, in terms of people advocating for parents, and more practically in terms of access to psychosocial support for parents who may be struggling with their child's diagnosis. Indeed, some VCSE professionals identified that families could benefit from some of the SSMIs delivered specifically to CYP, such as managing pain.

6.5.7 Encouraging attendance at support events

Despite the need for support events, suboptimal attendance became noticeable during the study, leaving HCPs and VCSEs feeling at a loss as to how they can increase attendance. This was frustrating for those advocating for more support from HCPs, to then struggle to attract attendees. Various reasons were conferred as to the potential barriers for attendance, including inaccessible locations, inconvenient meeting times, competing priorities, cultural differences, and family dynamics. However, several appeared to be linked to social anxiety, with support events in a face-to-face setting seen as daunting and outside of people's comfort zones:

> *"I think reducing the travel time for people [will help]… having them in places that people know is sometimes useful… they don't know what to expect when they get there… it's quite a lot of unknowns, and I think people like to have things that they know… I think it's helpful that if I've met families before [and say]… I'm going to be at the event… [they] know one person who's going to be there… they felt that people being connected prior to day events might encourage people to come along more."* (Xena, VCSE professional)

Some parents also had concerns that such settings may not be appropriate for CYP, who may not want to go with their parents, suggesting how hosts of support events may need to communicate goals more clearly, including how events would be tailored to CYP of different ages, and more clearly defining the benefits of attending. Participants felt this could be demonstrated to families in

advance through video and social media. Steps to help overcome such barriers could include a digital bridge towards face-to-face support, which could help families to familiarise and embed themselves within the support setting. Participants agreed that the key principle is to open up more opportunities for more families, giving them a choice in identifying what is right for them at any given moment in time. Consistent encouragement and repetition from HCPs from whom families trust was also suggested as a way to help families to see the value of attending support events:

> *"A lot of people who I know who have been online for a while, and I've then been able to say, well look, if you went to physical groups, you might actually find people in your areas to have a coffee with, and I think by then, they've seen the value of chatting online, and begin to see that there might be some value in meeting up."* (Miriam, VCSE professional)

6.5.8 Protectiveness and catastrophisation

Participants felt that some parents may be subconsciously overly protective of CYP. This behaviour could ultimately influence their willingness to transfer responsibility to CYP, and thus stall CYP's increasing self-management capacity. This clearly differs between families, and also by the different parent figures in families. One example was provided in the context of a VCSE activity weekend, where the parents had concerns over self-medication. However, during the weekend, trained staff had no issues with CYP not taking their medication or being unwell as a result, highlighting how CYP are often more capable than their parents may realise. This period of experimentation with self-medicating, where parents take a step back, was seen to be a good opportunity for CYP to realise the repercussions of non-adherence, and that by allowing them to make those mistakes within a safe environment, that they are learning the benefit of treatment, and the value of adhering to treatment, and the subsequent impact on their H&W.

Indeed, supporting families to make more informed decisions that are in the best interest of CYP's H&W are important, including instances where parents feel uneasy about their child attending an event, such as a residential activity, because of their JIA. Emphasis was placed on the fact that CYP should be actively encouraged to attend events, particularly for their psychosocial H&W,

understanding that reasonable measures may need to be in place to account for every eventuality, while allowing CYP some freedom and normality:

> *"I've worked with a lad… he is 18 now… and we are saying, you know, let's get you a bus pass. Even if I got on the bus with him, his mum doesn't want him doing it… he's 18… and he has the life of a sheltered… 11 year old… it's very hard because we're saying… you could do this, you could do that, and he's like, 'but I'm not allowed'… it's the fundamental thing you can't fly at the end of the day, unless you end up getting through to the parents. It's the one thing you can't move."* (Kendra, HCP – Youth and Family Support Worker)

Catastrophisation can also be problematic for some parents, particularly amongst those who are less familiar with the nuances of JIA and how to adapt to new challenges. While more experienced parents were seen to be more confident in assessing situations, others may not, highlighting the need for instant access to information and support:

> *"I think families do jump on, when they've got a flare, and they're wanting to be seen, like, tomorrow. Sometimes they might not need to be seen, if they've had a viral infection or something, it might just settle down by itself. We do see obviously, heightened anxieties when they're flaring."* (Victoria, HCP – CNS)

6.5.9 Willingness to transfer responsibility

The first step for parents transferring self-management responsibility to CYP begins with recognition of the need to transfer responsibility in the first instance:

> *"I recognise that one day she's going to have to do this herself, and those baby steps are trickier to take… I don't know if you ever let go of that completely… that was what made me think, that actually, going to some support things isn't about me, it's about her this time – she needs it. I am beginning to think about trying to hand over little bits to her, but I'm getting a big resistance, and so I suppose I'm exactly in that headspace about trying to think about how do I empower her…"* (Miriam, VCSE professional)

Participants remarked that teaching CYP to manage their condition independently should be seen as a component of parenting, akin to other self-care activities. However, some families may not regard self-management in the same light, particularly if they have not received guidance from HCPs, and may feel as though their child's H&W is going to worsen as a result, because they are not going to adequately self-manage. Participants remarked that this can be

challenging in practice since there is no formal process for supporting CYP throughout childhood and adolescence to begin taking responsibility over from their parents. Supporting parents to develop skills for encouraging self-management could help them with the process of transferring responsibility and shifting agency to CYP:

> *"The parents find it really hard to let go sometimes… there's a number of reasons, especially if their son or daughter's had a diagnosis from a really young age… taking a step back can be really hard and really difficult… that's why in the appointments, I… say… how about coming in for ten minutes on your own… sometimes that's actually harder for the parents to cope with… you don't want them to feel that they're no longer part of the care…"* (Audrey, VCSE professional)

Remarks were made as to CYP having the capacity to perform various self-management-related tasks, but when their parents are present and willing to execute such tasks, then CYP will allow them to do so. Such examples may include self-administration of medication, as well as arranging appointments, scheduling blood tests, and ordering repeat prescriptions. Participants felt that it would take the parent saying 'no', and prompting their child to perform said task, for them to realise that agency is shifting.

6.5.10 RQT2 Summary

Families play an important role in the shared-management of JIA, though this is shaped by the dynamic of the family unit, parental perspectives, shifting responsibilities, and relationships with professionals. Parents require timely access to information, education, and support, in the same way as CYP, using a multi-channel approach which recognises their expertise. However, they need to be receptive to accepting and accessing such services, which requires encouragement from supporting professionals. A focus on the gradual transfer of responsibility from parents to CYP is needed, influenced by changes in their knowledge, beliefs, and self-regulation skills and abilities, in order to improve their ability to competently management JIA on their child's behalf, while positively influencing CYP's self-management behaviours and independence.

6.6 RQT3: IHPs as shared-management tools

6.6.1 Awareness of IHPs

There appeared to be a poor awareness of IHPs and the existence of a JIA-IHP template on the HCSA website. This lack of awareness applied across all stakeholder groups, including HCPs – some of whom were aware of IHPs, but had never used them or directed families to them. Similarly, some parents were aware of IHPs, but stated that their child had not been given one. IHPs were sometimes confused with Education, Health and Care Plans (EHCPs) (GOV.UK, 2021), which replaced the statement of Special Educational Needs. Unlike EHCPs, there are no requirements or thresholds for IHPs, though confusion with EHCPs may cause people to consider them based on judgments of perceived severity and misconceptions of what constitutes a disability:

> *"People don't always realise that you can have an individual healthcare plan without actually having to have a leg missing… you can actually have that for all sorts of medical conditions, including JIA."* (Miriam, VCSE professional)

When IHPs became part of the conversation, it appeared to be in response to when issues surfaced, particularly at school. Moreover, when IHPs were in place, additional challenges materialised in ensuring relevant members of staff were aware of IHPs, and then able to interpret the contents and apply the recommendations consistently and appropriately.

Some participants referenced the longstanding use of IHPs in other disease areas. In particular, comparison to CYP with neurodevelopmental conditions was used as a prompt for how provision should be CYP with JIA. As previously mentioned, the HCSA have provided a generic IHP template which can, and has been used by different schools. There are other IHPs hosted on the website which have been co-produced by other VCSE organisations.

Adapted versions of IHPs, in the form of disease passports and 'all about me' summaries, were also mentioned, where all of the relevant information about CYP were appropriately detailed in a summarised fashion, and would be utilised to educate new staff, for example, when CYP moved year groups or schools. The first part of these resources centre on CYP's characteristics and preferences, and other general descriptions. The focus then goes onto specific challenges. For example, if CYP have mobility difficulties, detailing what they

find difficult, such as fastening shoe laces after participating in physical education. In doing so, suitable adjustments can be agreed upon, and detailed.

6.6.2 Holistic communication tool of CYP's needs

IHPs were considered useful resources in helping CYP and their families document and articulate their H&W needs. Particular relevance of IHPs was suggested for newly diagnosed CYP, as well as those with refractory disease. Ensuring IHPs were bespoke to individual CYP was highlighted, reflecting their unique and evolving needs and the way in which their condition impacts their life over time; ensuring they are maintained and regularly updated, at least annually.

Some parents explained how they felt responsible for completing IHPs, and in those cases, they would populate IHPs based on the 'worst case scenario'. This extended into documenting for every eventuality – including when CYP could be physically present in school, and when they could not, as well as clarifying the indicators of a flare-up or worsening of symptoms, and the proactive (or reactive) interventions required. Participants felt this could be challenging in the immediate post-diagnosis timeframe, when CYP and families are still learning how JIA affects them, as well as what support is available, and what interventions would be helpful. However, on the other hand, when used accordingly, they could guide the process by which CYP and their families identify challenges and how to overcome them. Participants felt that at the heart of IHPs though, must be CYP, and all other stakeholders should use this as an opportunity to engage CYP and hear their voice:

> *"Get the child involved… it's okay for the school to say okay, we'll do everything, we'll do this… the child actually then feels more isolated because of all… that's been put in… just having open conversations with the young person about things that they could try and things that are possible."* (Xena, VCSE professional)

Extending beyond their utility in documenting CYP's needs, IHPs were regarded as useful facilitators of communication and SDM; better equipping families in proactively engaging with their child's school to help them learn, particularly if they were hesitant to approach and speak to staff members:

"Giving parents some materials that they can use to go in and have that conversation with school, because not everybody's happy to speak to teachers and [don't] necessarily feel it's their place to go into school."
(Miriam, VCSE professional)

Even if specific provision was not required, families felt it was important that appropriate staff members were armed with explicit instructions of how to support CYP when they needed it, as well as when and how to raise concerns. However, when specific actions were needed –proactive and reactive, IHPs were seen to be useful in outlining processes, such as school needing to know the severity of seemingly minor ailments. One example provided was the increased risk of serious complications in CYP people receiving DMARD therapy after infection with varicella zoster. Parents highlighted to school that they needed to be made swiftly aware when other CYP in school were infectious, so that they could take swift, preventative measures to treatment.

Parents also highlighted how IHPs had helped them to challenge existing school policies which could have impacted on their child's H&W; for example, safeguarding issues regarding the application of sunscreen. Parents emphasised that documenting their explicit permission and instruction for staff to apply sunscreen to their child in the IHP helped school to understand that there was a genuine medical need for this action.

6.6.3 HCP influence over IHPs

The need for MDT input towards IHPs was highlighted, though it was less clear as to who was responsible for initiating the conversation about IHPs amongst the MDT, and subsequent completion and implementation. Some parents described instances where the onus was on them to complete the IHP and co-ordinate the relevant input from HCPs before sharing with their child's school, and this was something they would proactively undertake. However, this was not the case for all families, and further, completion of IHPs could be difficult, especially in the immediate post-diagnosis timeframe, as CYP and families learn how JIA affects them, while identifying helpful interventions. This is where exemplar prompts were suggested, as well as having informed professionals, including VCSE professionals, help CYP and families work through completing their IHP:

> *"I've sat with the young person and gone, okay, do you think this might be helpful… and worked through sort of the list… with that they would then go to school."* (Audrey, VCSE professional)

It became clear from participants that there was a need for greater focus on IHPs from HCPs, with more regular discussions in clinic. One participant hypothesised that the IHP could guide the conversation between HCPs and CYP, by working through the IHP, addressing different topics such as participation at school, physical activity, psychosocial H&W, and socialising, which may or may not be asked during conventional consultations. This approach at looking at the wider impact of JIA and the need for suitable interventions was seen to fall under the remit of allied health professionals, namely occupational therapists; though these professionals are not uniformly available to all CYP with JIA across the country.

6.6.4 RQT3 Summary

IHPs play a potentially important role as a holistic, shared-management communication tool between CYP, families, HCPs, VCSEs, and education professionals. However, awareness of IHPs is variable, with no current, standardised approach for their application into routine practice. With templated IHPs for JIA already available, assignment of responsibility for their completion, and regular maintenance, could help CYP and families articulate their needs and preferences more consistently, thus enhancing their involvement in SDM in order to improve the way in which they manage their H&W.

6.7 RQT4: Paediatric rheumatology MDT approach to SSM

6.7.1 Clinical resource priorities

Conversations between HCPs, CYP and families are often quite clinical in nature, focusing on treatment and other practical aspects of JIA management. As a result, participants felt that there tends to be less focus on psychosocial H&W and other holistic topics, such as SSM. HCPs attributed this focus on limited resource, attitudes, and inadequate training:

> *"You don't often sit back and think, well, what happened in that conversation? I don't think it's something that we actively do very well."* (Krishna, HCP – Paediatrician)

HCPs acknowledged that appointment time constraints often meant that they are unable to practically see CYP without their parents in the consultation room, despite an appreciation of the benefits of investing more resource into allowing CYP to be able to converse independently. Remarks were made to the increasing number of referrals to paediatric rheumatology which are not matched with increased staffing capacity, thus limiting what HCPs can practically do within the time that they have. Since there does not appear to be a readily-available remedy to combat these strains, participants agreed that attention must be paid to increased collaboration beyond H&SC, through integrated care services with local authorities and VCSEs, to help ensure CYP and families have access to the support that they require.

6.7.2 Sharing of best practices

Sharing of best practices amongst paediatric rheumatology colleagues, including VCSEs, tends to be at national and international conferences and organisational meetings. Interestingly, the youth and family support workers embedded within paediatric rheumatology MDTs from VCSEs were seen to have a broad perspective and exposure to different resources, helping to bring such resources and support to the attention of HCPs through their role, particularly when involved in MDT meetings, where new knowledge could be shared and explored.

Participants also highlighted the benefit of interdisciplinary networks, looking beyond rheumatology to other specialities for best practice, as was highlighted by community nurses sharing office facilities with different specialities. This can also be useful for CYP with multimorbidities. Participants went on to underscore the utility and need to update standards for paediatric rheumatology, including speciality-wide audit and evaluation to demonstrate improvement:

> *"You put together standards for… what are people doing really well and then you do a peer review… one team from one area go to another centre and they go through… they have a discussion about the set up in that team and they look at their operational policies… their outcomes… their plans… it's a tool for trying to get change, trying to improve a service. If you then have audit and peer review showing there is a need to be improved, then that's a prompt to the managers of an organisation to make changes."* (Krishna, HCP – Paediatrician)

6.7.3 Social capital

Collaboration amongst the paediatric rheumatology MDT was seen to be welcomed by CYP and families, as part of a person- and family-centred approach to H&SC. Seeing all HCPs in one room, or at least closely together, and with consistent messaging, was seen to be helpful in identifying and integrating different H&W-related needs:

> *"We [have] all got the same aim, to help young people. But sometimes we are looking at the same situation from different angles and because of that we see it from different perspectives… it doesn't mean… neither of those persons are right… I think losing the pettiness… we can't expect young people [to] self-manage well if the adults… around them… can't agree on things."* (Megan and Chanda, VCSE professionals)

However, it was acknowledged that the MDT composition varies between centres, with few having designated pathway co-ordinators to help deal with the co-ordination of requests and responses from CYP and families. The increasing need for a member of the MDT responsible for effectively supporting the psychosocial H&W of CYP and families was also identified:

> *"That person is there anyway, as a standard… you meet your rheumatology nurse, you meet your family mental health manager, you meet your rheumatologist, you meet your pharmacist… it's just… another person, because what we do is we say, physical physical, physical… mental health, give us a shout when everything goes to sh*t."* (Betsy, Teacher)

More recently, youth and family support workers have been integrated as part of some paediatric rheumatology MDTs, to help address disparities and variations in partnerships and knowledge of holistic services. This differs between centres, depending on the locality, set up (*i.e.*, working within or across paediatric and/or adult rheumatology) and their funding (*i.e.*, NHS or VCSE funded). They attend clinics where they meet CYP and families attending follow-up reviews, and receive referrals for newly diagnosed CYP to whom they can reach out to and outline available support. Furthermore, by being part of the MDT, they can more easily identifying CYP and families requiring additional support. The real benefit of an MDT approach was regarded when each professional pools their collective expertise and perspective on different CYP

and families, to paint a more complete and accurate picture of their circumstances, and how best to support them:

> *"[The youth and family support worker is] an embedded member of the team… she was at our meeting at the end of clinic… discussing… the patients they'd seen that afternoon... the consultant had met with this girl and he had said, 'oh my goodness, she's… on top of everything. She seems… more than capable and I have no concerns.' And then, [the youth and family support worker] and the psychologist were able to say actually, there's some real issues… what was happening was she was over-compensating, so she seemed incredibly competent for her age but actually she was almost too competent. So it was nothing wrong with the consultant; he'd based that information on the interactions that he had… it's about working together… there is no point doing [a] jigsaw puzzle with everyone holding onto their pieces."* (Megan and Chanda, VCSE professionals)

6.7.4 Understanding and valuing SSM

Participants felt that HCPs needed to understand and recognise the value of SSM, the impact of SSM on outcomes, and how SSM can be embedded into clinical care from the outset. Some participants attributed the limited focus on SSM to constrained resources, attitudes and skillset:

> *"Because the professionals are always stretched for time, you don't necessarily get the explanation that goes hand in hand with that…quite a lot of the time… it's a bit of lip service. Here's the leaflet, and then they can tick the box saying, 'Have you offered the family support?'… If you're in an okay place after that appointment, that leaflet is going to go in the hospital folder… and will be forgotten about."* (Miriam, VCSE professional)

Indeed, for many HCPs, there may be a limited understanding of what SSM means, requiring clearer definitions in the context of paediatric rheumatology to reflect on the physical and psychosocial H&W of CYP and their families:

> *"Self-management needs to be supported by every professional that is in contact with a young person… one thing that I think doesn't help with that is that there is a difference in understanding on what self-management means… [in] their context, [it will be seen as] taking your medication… physical aspects of your condition, because that's where their focus in, whereas for us self-management is obviously much bigger… more holistic… I think that health professionals will often think that they are encouraging self-management and thinking about it maybe by recommending an app… and while that is self-management, it's not the whole story."* (Megan and Chanda, VCSE professionals)

HCPs were not aware of the scale of VCSE provision in the UK, including their benefits. Generally, it seemed to be the CNS and youth and family support workers who were most familiar and proactive in identifying and promoting VCSEs; highlighting how the wider MDT could potentially benefit from attending some of the events and services themselves:

> *"We were asked to do it this year… I really enjoyed it… I really got a lot out of it. And you could see that the families get a lot out of it."* (Caoimhe, HCP – CNS)

However, HCPs generally recognised the value of VCSEs in filling the service gaps that they are unable to address, but were less clear on the practicalities of establishing cross-sector partnerships:

> *"The gaps that we feel, in our service, is that extra time with patients and families to talk about the other stuff. So, somebody who's got that time for the education side of it and just the knowledge and sort of support to families… I think, [would be] really useful. I don't know whether charities have the money and time to send somebody in the clinic [though]."* (Ava, HCP – Paediatrician)

6.7.5 Credibility and trust

Personalities were seen to influence the level of trust between HCPs, CYP, and families, particularly by the way HCPs communicate. Participants remarked that the credibility of different HCPs within the MDT depended on their roles and responsibilities, and how those intersect with the needs of CYP and families. However, the medical hierarchy was referenced, with the consultant considered an omnipotent individual to whom CYP and families listen to the most, including adhering to their instructions:

> *"Regarding the disease or… diagnosis… plan of the treatment, it is absolutely true that it is the consultant. It's even not a registrar, it has to be the consultant, who they trust, who the family and the patient trust. They usually do a circle… [asking] friends… family members… the GP… the local paediatricians… the registrars and nurses, then they go to the consultant, and if we say so, they trust us."* (Tom, HCP – Paediatric Rheumatologist)

Therefore, participants felt it was pertinent for consultants to remember the power and influence that they hold, and to be acutely aware that CYP and families take in what they say and credit it with great value, even if misplaced:

"There's still a bit of a stigma around the consultant is the top dog… therefore if he or she says something, that word is Gospel… it's an old thing… doctor is somebody you should always listen to and respect, and that… is why people struggle when consultants are very flippant… I think consultants can often do some damage in what they say." (Miriam, VCSE professional)

However, nurses were generally seen as the HCPs who build one of the strongest relationship with families, and whom families sincerely trust, since they tend to be the first port of call whilst requiring support:

"They see the same people when they come to clinic, the same people coming out to see them when they need to be seen at home. So, it's getting that relationship with them… I think definitely helps." (Victoria, HCP – CNS)

Collectively, if CYP and families trust, and have confidence in their HCPs, then participants felt this influenced the manner in which they take on the SSM of JIA. This was related back to principles of informed consent, SDM, and family-focused care, placing CYP and families at the heart of paediatric rheumatology H&SC:

"If you do ask a child are you ready and the child replied no, I'm not ready, if you then ignore them and still go ahead and do whatever you've planned to do, then what did it mean that you asked them for permission to have a procedure and then you overrule them anyway… I think there's a lot of questions around if you do ask children's consent are you listening to them?" (Judi, VCSE professional)

6.7.6 Engaging and involving CYP and their families

HCPs engaging and involving CYP and their families equally in conversations and SDM related to JIA was seen as a step towards supporting them in the SSM of JIA, even if the process takes longer than it would with engaging parents. However, participants felt this was often suboptimal, citing reasons such as limited social skills, lack of resource, or lack of consideration. Participants remarked how conversations, particularly with the paediatric rheumatologist, were generally clinical and gravitated towards the parents:

> *"When we've been to see them with our daughter, they tend to only focus on us, they don't really ask her anything at all… most of the time, she could not be in the room and it wouldn't make any difference."* (Toby and Clara, Parents)

Each consultation is conducted differently, though participants thought it may be helpful for HCPs to gauge CYP's self-management capacity, and thus begin to introduce tangible actions to build towards the goal of self-management:

> *"What I typically do… is… say hello… introduce myself… and then… I just summarise their case: what is the disease, what is the treatment, what is the plan, and what we know from the last visit… then, I try to ask the patient… how are the joints, how is the response to treatment… if they have any troubles with taking the medicine… how is school, how are they at home… what is the plan for the next weekend… it gives me quite a good picture about the family, the independence of the child, and whether they are able to do some self-management or not, and if no, then I try to help. Some of the parents are very grateful… because they say, 'wow, nobody did this with us before'…"* (Tom, HCP – Paediatric Rheumatologist)

The relationships with different members of the MDT appeared to influence how much CYP and families felt engaged. Generally, this appeared to be strongest with allied health professionals:

> *"With her physios, they're much more interested in her than in us. So, we're there and they'll talk to us, but actually the conversation is with our daughter, because she's the one going through the exercise and she can explain to an extent how she's feeling when she does them… so that's much more focused on her than us."* (Toby and Clara, Parents)

Families also valued when HCPs came to them for advice on how to support their child, recognising their expertise, which acts as a powerful driver for SDM:

> *"Once when we've ended up in the hospital… the… junior doctor… she came to me and she said, you tell me what I need to do, because you're the expert in this. So, I said by the point we get to this stage it usually is [an] infection… they usually put her on antibiotics and that does the trick… and she took my guidance and advice as to what to do."* (Toby and Clara, Parents)

6.7.7 Holistic and rapid access to support

Holistic support for CYP, and families was seen to begin with HCPs recognising the physical and psychosocial aspects of JIA, and the impact it can have on families – not just from a health perspective, but from multiple angles, before

transparently setting out the available support. However, there was a sense that pre-empting psychosocial support remains a more reactive rather than proactive response, with less upfront discussion around the likelihood of JIA impacting on the psychosocial H&W of CYP and their families:

> *"We don't tell parents, this is how you need to react… these are some of the emotions… because they have this condition, and… these are some strategies for how to manage that… we don't say that to families, we just try and answer questions as they come up."* (Krishna, HCP – Paediatrician)

Nurses were seen to play an important role in using pharmacy counselling to support CYP and their families in getting the most from treatment, often using checklists to ensure they have addressed key points. Indeed, remarks were made that it is not always dealing with JIA which is particularly problematic; rather, it is the interventions required to manage JIA which can make life more difficult. For example, when considering trypanophobia and the associated anxiety with that, participants felt that relatively few CYP and families were reliably educated on techniques to help them cope:

> *"The parents have a role to play, they can help their child through the procedure with distraction and with techniques that give them the feeling that they have a role and the child will know they can trust the parent… Parents often feel desperate and guilty for having to take their child to hospital to have these treatments… so to give them the tools, it must be much wider than just about the information, what's going to happen, it needs to be about what can you do, how can you help."* (Judi, VCSE professional)

Furthermore, participants felt it could be challenging for opinions to be aligned as to the interventions required to holistically support CYP while doing so in a way that enhances their QoL:

> *"It is difficult because you will get different views about what is the best way for that child to manage… and the consultants aren't always necessarily… sympathetic about the fact that [the] child is in school with 29 of their peers and… what might be an obvious solution to the consultant, 'oh sit on a chair then, it's fine' isn't always that easy to implement."* (Miriam, VCSE professional)

Participants felt that families should have instant access to the contact details of their MDT, ensuring they know all members of the MDT. In return, HCPs were seen to be well placed to proactively identify and communicate with

families who may be struggling, to ensure any potential issues can be flagged. Participants agreed that closer monitoring can be facilitated when CYP and families experience continuity of care, particularly with the same paediatric rheumatologist and CNS. This was seen to contribute to the level of mutual trust experienced between all involved parties, especially for CYP and their families, who feel reassured that somebody cares for them:

> *"I think there's no cost-benefit to giving patients reassurance. You're not giving them a medication or a treatment. It's… holistic care, it's very difficult to prove a cost on, but it's the cost of somebody's mental health, as a parent, and it's the cost of that child being able to carry on with normal family life... it's a different measure to how many joints and flares does this child have…"* (Miriam, VCSE professional)

6.7.8 Individualised, co-ordinated, and continuous care

Experience of H&SC and communication were seen to be interlinked, with clear and honest dialogue, delivered compassionately and empathetically, proving to be critical. Individualised approaches to H&SC were alluded to, with examples of how HCPs proactively engage CYP and families in-between consultations, particularly if they are non-adherent to treatment and require closer monitoring:

> *"It's like on Friday, I sent somebody two links via email, one was for a HIIT [high-intensity interval training] workout, because they were struggling with sleep, and another one was something related to anxiety…"* (Kendra, HCP – Youth and Family Support Worker)

Participants believed that centres where additional provision is in place to support CYP, helped to bridge the gap between home and hospital, helping CYP to feel more familiar with the clinical environment while getting to know certain members of the wider MDT through informal activities in a 'safe' space. Unfortunately, administrative challenges were seen to result in inadequate communication, impacting on how families engage with HCPs, and how competent and trusted they may be perceived:

> *"We've found that the best way is to drop them an email and we know that someone looks at it, but they don't always get back to you… we were warned of that when we were… referred. We were told the administration is not fantastic."* (Toby and Clara, Parents)

This was seen to depend on local circumstances for how enquiries are managed, including separation and integration of administrative staff duties with those of CNS and other HCPs responsible for managing helplines and advice email accounts.

The clinical management of JIA was viewed to be the core focus of conversations between CYP, families, and HCPs. Although attention was seen to be paid to comorbidities, like uveitis, participants felt it was less clear as to who initiates these discussions, and how CYP and families are educated about related conditions. Families also said they felt a great deal of frustration when relatively little is discussed in the consultation, and then the clinic letter for the GP arrives at the family's home, detailing information that was not discussed or explained in the consultation:

> *"I see a lot of scenarios where consultants will say not very much, then the clinic letter will appear, and there'll be all sorts in there that wasn't discussed… and that to me is absolutely outrageous because any words or particularly naming conditions that are on a letter that haven't been discussed with the family cause the worse amount of worry… they've got to be providing enough information that you don't want to go to Google first off because you don't understand."* (Miriam, VCSE professional)

Similarly, during heightened situations, where CYP and families may be experiencing increased levels of anxiety, participants felt it was quite easy for information to be misinterpreted, particularly if incorrectly communicated:

> *"What the doctor said was not to get pregnant on methotrexate but it wasn't explained properly, so the person thought that they never will be able to have children."* (Megan and Chanda, VCSE professionals)

An interesting and evolving dynamic discussed with participants was that of the relationship between HCPs and CYP and/or families who have become advocates within the community – a role evolving the conventional patient-HCP relationship into collaborative working relationships and friendships, which was noted by some participants as potentially off-putting when taken out of context:"

> *"It put me off a little bit… when [at the conference]… it was really lovely, what they said, but I think it kind of freaked a few people out and they were like, 'oh, the patients are like our friends, or our family… and I think some people were a bit, like… boundary."* (Kendra, HCP – Youth and Family Support Worker)

This remark reflects the changing role of paediatric rheumatology, in that HCPs and researchers are now joined by patient and parent advocates, as well as VCSEs, in driving change, which should be in a collaborative but professional manner.

6.7.9 Incentivising HCPs

However, sharing of best practices appeared to be *ad hoc* and inconsistent. The reasons for these irregularities were unclear, but in part, were believed to be caused by limited concise summaries of best practices and how to implement these locally. MDTs were seen to be already under excess pressure, and so one of the easiest ways to influence activities may be to provide more directive advice for centres, incentivising them to utilise support services available for CYP and families without using additional resource requirements:

> *"How do we get all these hospitals [to use these resources]… what's been really effective is if I'm invited as a speaker at a conference and I share… one of the videos and then tell the staff that's there we can provide you with these free of cost hospital leaflets… we'll print and we'll send them to you, you don't have to pay anything, but this is how you can help your patients... people are very impressed … also other healthcare professionals who say it saves us a lot of time not having to go through the details of the procedure because the family will come in and say yes… I know what to expect… so because… it saves them time and it helps to calm the children down… it's in their benefit to share the videos."* (Judi, VCSE professional)

6.7.10 RQT4 Summary

HCPs play a fundamental role in promoting SSM of JIA amongst CYP and families. Through multi- and inter-disciplinary collaboration, the sharing of best practices, recognising the value of SSM, and reflecting on their influence, HCPs can proactively contribute to enhancing SSM capacity amongst CYP and families. This can be supported by HCPs engaging and involving CYP and families in their H&SC, which should be holistic, individualised, co-ordinated, and continuous where possible, in order to build trust with CYP and families over time. This may be aided by incentivising HCPs to recognise the many benefits of SSM.

6.8 RQT5: CYP- and family-focused care across the lifecourse

6.8.1 General awareness and understanding of JIA

Despite an increasing number of awareness-raising campaigns, participants remarked on inadequate awareness and understanding of JIA and its impact – amongst the public, and generalist HCPs. Families described arduous journeys to diagnosis, which can have a devastating impact on the psychosocial H&W of CYP and their families, as well as increasing the risk of physical damage as a result of uncontrolled inflammation:

> *"We had multiple trips to the GP and onward referrals… to the fracture clinic… went back a week later… still couldn't see anything… I think it was on the third occasion… by then [we had] been digging around online and finding out… what the possible alternatives might be, and came across juvenile arthritis."* (Toby and Clara, Parents)

For awareness-raising campaigns, it also seemed important for childhood-onset arthritis to be referred to as JIA, rather than some of the messaging such as '*children get arthritis too*', since participants felt that this may cause greater confusion given the many different types of arthritis. Participants also felt that people generally fail to understand the relapsing-remitting and volatile nature of JIA, including the variability of symptoms, which while largely invisible, can be difficult to appreciate:

> *"How you can be… 100% fine to do whatever it is, a one mile run round the park one day, and then the next day, absolutely not appropriate… I think [that] is what schools do struggle with, and yet to all intents and purposes, the child to the untrained eye would look the same to them both days… the invisibility is a big, big part of why that's so hard, and a big part around what needs to be done around education and awareness."* (Miriam, VCSE professional)

6.8.2 CYP- and family-centred approaches to integrated care

Participants believed that care was at its best when services were integrated, and focused on the holistic needs of CYP and families, with them more involved in processes related to their care. VCSE professionals were keen to highlight how their services can be useful within and beyond the clinical environment:

> *"They are always going to end up being [at the] hospital. So for us, it was the logical place to go and I think the more that we have worked in that setting*

> *the more we realise that there is a role supporting families there… I think it's got to be seen as a key priority from those that have got the power to make change in the shape and the funding… I think it is actually part of what will support children and young people and their families to live well, to manage their condition… and actually see a reduction in the amount of hospital visits."* (Megan and Chanda, VCSE professionals)

Despite multi-disciplinary working in paediatric rheumatology, particularly between the paediatric rheumatologist and CNS, other members of the MDT, such as allied health professionals, may still be situated outside of the clinic when CYP are in attendance. Questions were raised as to whether it would be more suitable for CYP to be seen in clinic with the entire MDT present, so that decisions could be jointly made with everyone 'reading from the same page'. When asked about entering a room with the MDT present, Darcie as a child with JIA did not seem phased by their presence, and appeared to be amused by their approach to performing joint assessments:

> *"No [it's not scary, it's]… tickly, because they move your arms and legs around. And sometimes I laugh."* (Darcie, Child aged 5–7 with JIA)

Limited resources were also seen to impact the level of CYP- and family-centred approaches in clinic:

> *"We're very much doing the business of, are you well, have you got any new… inflammation, what treatments are you on, have you got your bloods done? And that's kind of our 15 minutes and that's all we can do."* (Ava, HCP – Paediatrician)

In reality, to account for all CYP and families, participants felt that multiple opportunities are required to ensure everyone has rapid and continuous access to CYP- and family-focused care and support that is bespoke, personally relevant, and integrated with existing services:

> *"Humans are individuals, they're all going to access different things at different times in different ways. I think you need all sorts of different support in all different forms for all different stages of the journey… there's no one size fits all… people may cope fine with a certain amount of it, but then suddenly… something new comes to the party, that's when I think people struggle… I think there's a certain tendency with some centres to keep people at arm's length, and I think it's a false economy."* (Miriam, VCSE professional)

6.8.3 Variations in service delivery

The way in which services are delivered naturally influences CYP's ability and need to self-manage their H&W, depending on which services they access. Participants remarked that regardless of the service offering, CYP and families should have access to an equitable level of care and support, regardless of their location:

> *"There are differences in need and geography dictates a certain way that a service is set up… it's not to say that it is necessarily right for it to be the same everywhere, but I do think that families should have the same support and the same standard of service even if it's set up differently."* (Miriam, VCSE professional)

Unfortunately, there are a finite amount of paediatric rheumatology tertiary care services across the UK, and so for a number of CYP, their care may be delivered as part of a hub and spoke model under a shared care agreement with local secondary care services. Participants highlighted how this may be independent or as part of an outreach clinic, reflective of the need for care to be delivered as close to CYP's homes as possible. This can be helpful for families who are further away from tertiary care centres, though may mean that they have less frequent access to the wider MDT, and potentially no access to certain HCPs, such as occupational therapists and psychologists. This was seen to impact overall access to information and support to promote SSM:

> *"It can be every six months and it can be annually that they see the young people because they're then having shared care with other hospitals… so, while some patients [the tertiary centre] is their only hospital and they see [the MDT] quite regularly… other patients… may only see them once a year."* (Audrey, VCSE professional)

This also appeared to have implications for when families were in attendance at VCSE events, for example, and HCPs would describe their local services; which for some, was irrelevant given the contextual influence of geographical locations dictating what they can access:

> *"If you are in an areas that has a paediatrician instead of a rheumatologist, then that shouldn't restrict you from accessing things… it's acknowledging those differences… [that] those differences will always be there, there will always be discrepancies in NHS funding and ability to access different*

services in different areas, but then finding a way round it instead of just going, 'ah well'…" (Betsy, Teacher)

Some of the tertiary care services described by participants also offered outreach clinics, which may or may not involve the local paediatrician or adult rheumatologist, adding to the dynamic of how some CYP receive care across the lifecourse, which may also facilitate transition into adult H&SC by becoming more familiar with the adult rheumatologist early on:

"I do a clinic a month… with an adult rheumatologist who has an interest in young people… all the children are shared care... so, we might get new referrals where we think they've got JIA, but we would ask the [tertiary centre] to see them as well and usually any sort of decisions about treatment are made by [tertiary centre] and then we will prescribe them methotrexate and see the children in between the [tertiary centre] appointments so they haven't got to travel… we're sort of like a little triaging… system." (Ava, HCP – Paediatrician)

For CYP under a shared care agreement, knowledge of a contact at their local hospital was seen to be important, particularly when urgent care was required, or input regarding treatment was needed. However, the agreement may add complexity to the way in which CYP, and their families access H&SC services. Given the complexity in models of care, how services operate, and who to contact, participants felt it may be timely to provide consistent guidance to CYP and families, so that they can visualise how their care is delivered, and what this means for them:

"They think that you go to your GP for one thing and then you get sent to a hospital. They don't… realise that you might have all these different people. You know, why have I got to [go] here and there… like how paediatric rheumatology is done… you'll be seeing people in [tertiary centre]… and [then] your local paediatrician. You can go to either place for information… but that's why you need to go to [tertiary centre] because things like joint injections are more experienced there… maybe sort of system information about that kind of idea [would be useful]." (Ava, HCP – Paediatrician)

The tendency for good practice to be undertaken on a small scale by enthusiastic teams was mentioned, calling for best practice to be scaled up across the country, including the implementation and audit of relevant standards and guidelines. One participant described how two paediatric rheumatologists formulated a business case to employ a youth and family support worker,

recognising the critical need to holistically support CYP and families across paediatric and adult rheumatology, thus providing a continuity service across the lifecourse by the same individual.

Unfortunately, participants alluded to a field which can, at times, be politicised, including reluctancy to identify and share best practice, refusal to evolve and adapt, rivalry, and personality clashes, which can be potentially damaging to wider collaboration and learning, as well as the H&SC and support available across the country:

> *"I've been quite shocked since I've been involved in the JIA world… the politics involved in it all…. There's a real reluctance to look at best practice and say, 'oh, that's working fantastically at this hospital, maybe we could try the same'… there's a lot of egos involved, and people just want to do things their way… I've realised that there are certain consultants whose names aren't popular… I've learned to keep my mouth shut a little bit."* (Miriam, VCSE professional)

6.8.4 Systems, data sharing, and networking

System communications, including data management and sharing, were seen to be a challenge for HCPs and families alike. From the family perspective, insufficient access rights to CYP's medical records by different HCPs was seen to make the process of person-centred, integrated care, a difficult goal. Frustrations with inconsistent reporting, including the continued use of written notes in place of electronic patient records, were seen as illogical in a technological era, with a seeming lack of intelligible documentation of CYP's medical history, treatment history, and treatment outcomes. These challenges were also expressed by HCPs:

> *"Certainly the ability to see notes in different places. Even within our own hospital it's very difficult for me to see the physiotherapist's notes. You'd think it would be completely simple but it's not."* (Ava, HCP – Paediatrician)

Poor system communications were also seen to impact on clinical care, through inconsistent and contradictory information for families and other affiliated HCPs, such as those in primary care:

"A whole palaver about the flu vaccination this year, because the guidelines did change, and some hospitals issued new guidelines but without really explaining them, and other hospitals stuck with the old ones and said no, they hadn't even heard about the new ones… it causes families no end of… worry." (Miriam, VCSE professional)

Poor system communications can also result in a number of inefficiencies, both for families, and the H&SC system, such as appointment scheduling when CYP are under shared care agreements. While some may feel that families should be proactive in cancelling appointments which are close together, participants remarked that this is not necessarily their role, and could be better facilitated by administrative schedulers ensuring that appointments are more evenly spread:

"If there was some way that somebody was able to look at the clinic appointments out of all the places and sort of see what was happening because it is a waste for them just to be seen by two different paediatricians a week apart when they actually need to be seen three months apart. So, definitely sort of the system bits could be far more efficient." (Ava, HCP – Paediatrician)

Finally, it was suggested that in order to embed SSM practices successfully within paediatric rheumatology, a network or alliance may be required, to pool expertise, agree on standardised processes, and then support the community to embed SSM practice across the country:

"There have been several attempts to bring everyone together in some way and I know there's challenges but actually there's power in pulling that together as you've done and it would be great to see some continued work in that because that's how we can all learn… and support one another." (Megan and Chanda, VCSE professionals)

6.8.5 Primary and community care and support

Although paediatric rheumatology is largely based within secondary and tertiary care, there is an existing, and increasing level of input from primary and community care. Participants reflected on how community nurses may be involved with going out and meeting families in their home environment, allowing for trusting relationships to be built over time, as previously discussed.

Families discussed varied relationships with their GPs, often compounded by an inadequate understanding of JIA – with instances of GPs failing to identify

the hallmarks of JIA, knowing where to refer patients to, and when diagnosed, not following recommendations from paediatric rheumatology MDTs. This added to frustration and burden of responsibility experienced by families:

> *"They didn't know that they should have… sent us to [tertiary centre]. They were kind of like, oh which hospital do you want to go to? He should have said to me there's an expert paediatric team… I think you should go there."* (Esther, Parent)

HCPs with paediatric rheumatology experience felt that GPs were so busy that when requesting their input, specialist instructions must be concise and straightforward:

> *"If I need GPs to do something, it must be simple and straightforward, such things like continuous prescriptions… sometimes, they can do the blood tests, but sometimes they even cannot do that. And if they cannot, then we ask the general paediatrician from the local town to do so. Or if it is not necessary to be done too often, we can do it regularly when they come to our clinic appointments."* (Tom, HCP – Paediatric Rheumatologist)

6.8.6 VCSE services and their visibility

VCSEs provide a range of information, education, and support services for CYP and families, as previously outlined. The number of services differ amongst VCSEs, dependent on several factors, such as capacity, capability, and funding – with many dependent on charitable donations:

> *"It's a shame not everyone can have that opportunity. I think that's the only thing with projects like this, it's always time-limited and there's never enough people to help everyone…"* (Judi, VCSE professional)

Furthermore, few VCSEs appear to have the capability to formally and rigorously evaluate their services, thus, change tends to be driven by anecdotal evidence and experience. This may also limit the opportunities for VCSEs, in terms of funding, since they have less robust evidence to demonstrate and support the services that they are providing:

> *"There's not enough evidence out there as to the impact of groups and support groups."* (Hope, VCSE professional)

Participants believed that more consistent cross-sector collaboration built upon clear, consistent communication and transparency, could help to ease these concerns, and some of the challenges already identified. VCSEs can help to provide the holistic support which H&SC services are unable to currently deliver, acting as both a responsive and preventative service. However, a challenge remains in that many HCPs are unaware of VCSE services:

> *"Sometimes professionals… or even the parents look at us as a crisis service… but also… we are mostly a preventative service… I think linking up with [the] NHS we can provide holistic support for young people and their families because we can jump into the gaps, specially this kind of emotional, social… gap… [that] the NHS sometimes don't have time to fill… we've got more flexibility so we can be more responsive to NHS needs as well… so I think working together is the way forward… [but] there is not always the willingness to do that."* (Megan and Chanda, VCSE professionals)

Participants commented on how many VCSEs within paediatric rheumatology historically focussed on JIA as the most common juvenile-onset RMD; and while there is benefit and a need in providing specific information and education around JIA, when looking towards the SSM of JIA, there are many similarities with related conditions, hence why an increasing number of VCSE opportunities are aiming to cover SSM topics for all CYP with juvenile-onset RMDs.

Indeed, as well as cross-sector collaboration (within and beyond rheumatology), participants indicated that greater collaboration amongst VCSEs in the UK and internationally could benefit how CYP and families are supported. VCSEs have differing aims, target audiences, and coverage, and so signposting from one VCSE to another could also be beneficial. In addition, when the sector is appropriately networked, participants sensed that VCSEs could collectively reach and benefit a greater number of CYP and families across the country, while avoiding unnecessary duplication:

> *"There's politics around all of that… why can't we just, 'right, you do that and we'll do this?' I think, oh for goodness sake… I wish it… wasn't that way… I don't understand why people need to do… the same work in the same areas…"* (Hope, VCSE professional)

Consequently, visibility and access to VCSE services varies across the country, with many CYP and families identifying these organisations and their services fortuitously while seeking information, education, and support, typically from

other families and advocates. Participants agreed that the visibility of VCSE services could be enhanced when there are VCSE-affiliated youth and family support workers embedded within paediatric rheumatology MDTs, since they can then inform CYP and families about services and the potential impact they could have, as well as when HCPs are aware and proactively share this information with CYP and their families. Many VCSEs also have their own mailing lists, and so for those families who have consented to receiving information, they can be kept updated with SSM events. A challenge for VCSEs is ensuring that there are events available in every region of the country, to people from all walks of life, which can be difficult for smaller VCSEs with limited budgets. If families have to travel beyond their locality, as well as to unfamiliar locations, participants felt that was unlikely that they would participate:

> *"They have these events… but a lot of it seems to be a very long away from us… so it's kind of difficult to access those sort of things… [also] the audiences or the members of those panels… they tend to be of a particular social class and actually you are missing out on sort of engaging with a lot of other people for a variety of reasons… social class kind of puts them off or they don't have… often money is not provided to kind of do the travel and things."* (Esther, Parent)

Some participants described how widening the reach of VCSE services can be achieved in some circumstances online, though emphasised that CYP and families need to feel engaged and involved, requiring greater thinking rather than simply recording an event or directly publishing material online. Additional suggestions also included short, planned promotional videos of VCSE services, featuring CYP and family testimonials, to help others witness first-hand the benefit of VCSE services, as well as raising awareness of their existence.

6.8.7 Standardised clinical signposting

Participants discussed the various layers of signposting to information, education, and support in relation to JIA. Signposting by HCPs appeared to be inconsistent, both within and across different settings. In particular, there seemed to be varied signposting to VCSE services, with biased recommendations appearing when HCPs had direct experience of particular VCSEs:

> *"It's so bitty, and people get shoved this leaflet and that leaflet, depending who their consultant is or their nurse or whoever it is, but it's not ever the whole picture."* (Miriam, VCSE professional)

Consequently, CYP and their families may feel unable to envisage the wide range of services available. In cases where information was passed on, participants believed this was often passive in the form of literature, when what may be most useful would be a conversation about the SSM opportunities available. There were also instances where HCPs, in particular, the paediatric rheumatologist or paediatrician, did not signpost to any information or support at all. One paediatrician highlighted that they do not signpost, and were also unsure if nurses in their team did either, highlighting how the lack of integrated working on information and support may be lacking for many CYP and families across the country. These HCPs recognised this as a limitation in the wider support that they offer:

> *"There might be specific things occasionally but no, we don't really go back to any sort of education things. We probably should, but we don't."* (Ava, HCP – Paediatrician)

Indeed, families and HCPs alike felt that a more transparent and detailed overview of accurate, relevant, and trusted information, education, and support services would be useful – like a roadmap of SSM support, so that families would not necessarily have to struggle, or rely on searching the internet for information, education, and support services, which currently feels disjointed:

> *"I think it's very bitty at the moment. I just had this vision of… a roadmap, a one pager, that would have… what support was available, particularly from the big five charities, depending on where you were in the country, what the age of your child was, the different types of support (online… face-to-face… residential)… and just somehow so that people can see… 'well we live in Scotland, so okay, SNAC [Scottish Network for Arthritis in Children]'s going to be what we need'… or, our daughter is only a toddler, so that's not appropriate, but CCAA would be."* (Miriam, VCSE professional)

Similarly, with regards to signposting to information, participants indicated that that there is greater focus towards clinically-related information, such as about the disease and treatments, focusing less on the wider psychosocial impact of JIA, as well as how to practically manage JIA. A number of NHS Trusts have developed their own information to this effect, despite many VCSEs

have produced a range of literature addressing all aspects of JIA. While some VCSEs appeared to contact different hospitals, participants were unclear whether this is done regularly, and whether all VCSEs are doing the same or not. More systematic and consistent approaches to delivering resources into clinic was called for, to help avoid the patched approach currently seen. HCPs who did use different materials remarked that they should not just be given to people to read; rather, they should be used as a tool to address different aspects of JIA, before allowing CYP and families to digest the information and peruse at their leisure:

> *"It's something that we would take out to the visits with us… we tend not to try and post it out… [but] go out and physically go through everything with them, that's in the booklet, and then leave the booklet for them."* (Victoria, HCP – CNS)

6.8.8 Timely access to the most appropriate HCPs

Timely access to the most appropriate HCPs was seen to be increasingly important so that the needs of CYP and families are met promptly and effectively by those with the correct skillset to help make a difference, particularly when it comes to psychosocial H&W. Similarly, knowledge that there are certain HCPs to whom families can contact when required, can be a notable point of reassurance for families, in that they feel more confident that they can reach a known and trusted individual. Invariably, this can happen at different time points, and there are likely to be different points in the journey with JIA where families require more reassurance:

> *"If you know that… you could speak to somebody who knows your child or who can access the correct information about your child, I think then you don't panic and you don't ever worry nearly as much… if you give them the right reassurance at that time… then actually they may well tick along quite happily for another long time… I think the families who I'm in touch with who have the worst time are the ones who can't get hold of anybody at the hospital… when families don't know what to do and can't access that help, that's when they really struggle."* (Miriam, VCSE professional)

Where staffing may be more challenging, triage lines offering information and advice were suggested. The key principle of access to such information and support maps to the positive effect of early intervention on various outcomes,

both related to the physical and psychosocial H&W of CYP and families, but also those related to H&SC utilisation:

> *"The hospitals that do it well do it really well, and I think they would probably see, overall, need for appointments would go down, because a lot of things get nipped in the bud. Families just need a quick answer, and things don't escalate… they key thing is being able to get hold of somebody when something happens, and sometimes all you need is a quick 'yes that's fine'..."* (Miriam, VCSE professional)

Access to psychology services appeared to be a challenge for many as a consequence of delayed referrals and waiting lists, despite the increasing awareness and understanding of the psychosocial impact of JIA, including the impact of treatment and development of phobias, and how these have the potential to influence short- and long-term adherence if unaddressed:

> *"One of the things we've tried to do and it's taken a very long time, we still haven't got the appointment, is trying to get her referred to the psychologist to help with the phobia of the methotrexate injections."* (Toby and Clara, Parents)

Participants felt that the model for psychological support required investigation. Although some paediatric rheumatology MDTs work closely with psychologists via direct referrals, several participants felt that CYP and families tend to be referred reactively when problems arise, rather than proactively which would make more sense, with adequate resource, to pre-empt issues and ensure that CYP and families have the support and skills in the SSM of JIA:

> *"We tend to refer to psychology when there's a problem, and I suppose sometimes it's a bit too late. It would be a very ideal world, but if you had the opportunity where psychology could see families right from the beginning before there was an issue…"* (Caoimhe, HCP – CNS)

While existing HCPs said that they try to provide psychological support beyond their clinical remit, they recognised that they are not experts and lack formal training. Moreover, psychological intervention was not seen to be a quick fix that can be resolved within one session. To help address the shortage of trained psychologists, participants felt that there may be an increasing role for mental health practitioners, either based within statutory H&SC services or VCSEs, who are able to support CYP and families as they try to live with JIA.

Participants did not feel that it mattered where the provision was, as long as it is there and easily accessible.

CNS were also seen to play an important part in supporting CYP and families in the SSM of JIA, often being the first port-of-call for families when engaging with the paediatric rheumatology MDT in-between consultations, as well as facilitating blood monitoring and other aspects of JIA management. Indeed, the demand for CNS in light of existing capacity has influenced staffing provision, as some participants confirmed, given the increasing number of CYP requiring care and support from CNS.

As previously alluded, youth and family support workers are a relatively new additions to the paediatric rheumatology MDT, but are playing a critical role, where available, in providing much needed SSM support that is individualised to CYP and families, typically with adolescents upwards. Participants remarked on the various models of youth and family support worker embedment into MDTs – the most common being direct employment by NHS Trusts, or employment by VCSEs with an honorary NHS contract. Some workers are based solely within paediatrics, while some work across paediatric and adult rheumatology to bridge the gap in service provision to provide continuity of care and support. Participants said that youth and family support workers may come into contact with CYP through various ways, such as referral from HCPs, information discussions in the clinic, self-referral, or through inpatient ward rounds. Embedding youth and family support workers as part of the MDT was seen to make the worker's role easier, while enabling them to be exposed to CYP and families within and beyond the clinic, observing different behaviours which HCPs may not have identified. Thus, they can facilitate the flagging of issues within the MDT, and with CYP or family:

> *"I've seen them at a weekend, they're really hobbling, they can't move… I then report back to the clinic and say, look, this kid's really struggling… can we… up their appointment? So it's a very much two-way-street… similarly… they'll say, we've had a letter… that this kid's really struggling at school and not able to do things. And I will say, well, you know, I had them away at a weekend… and they were completely fine and in a really good place… can we investigate that?"* (Hope, VCSE professional)

Participants highlighted how youth and family support workers need to be familiar with JIA and its management, which can benefit from lived experience.

However, participants were keen to emphasise that psychosocial and practical support, not clinical management, is not their core focus:

> *"I will like to signpost to Citizens Advice. I thought it'd help initially. But certainly with a lot of people I've set quite a few appointments up for them. They're not quite sure where to go, who their local one is, and I might have made a phone call for them and set it up."* (Audrey, VCSE professional)

Moreover, youth and family support workers were also considered to bridge the gap between other professionals, such psychologists, social workers, and play therapists, identifying and helping to address issues early-on, which may not require the input of said professionals. This was seen to be an important consideration given the access challenges described:

> *"I suppose it also fits a gap of social services that isn't, you know, social worker aligned through the paediatric team. Probably a lot of the kids' needs wouldn't meet the threshold of a social work service anyway but it bridges that gap."* (Hope, VCSE professional)

> *"The feedback that I've had, is that it's good because… it can deal with issues that aren't straight to psychology. Because half the time, you wouldn't have an in between, would you? I think it works well for the issues that wouldn't quite reach the threshold for psychology, and those people that are [coping] alright."* (Kendra, HCP – Youth and Family Support Worker)

Youth and family support workers were seen to add a unique lens to the management of JIA, by advocating for CYP from the earliest opportunity, while enabling them to become increasingly confident at self-managing their JIA:

> *"She is sitting down with young people… asking them how they are… helping them navigate those first appointments, which we know can be very anxious times for young people… and [saying]… have you got a list of things you want to discuss.. let's take a moment and help you write down a list so you know you get everything answered. That's self-management but that couldn't necessarily be done by a consultant in the time and things that they have [to do]."* (Megan and Chanda, VCSE professionals)

Their flexibility means that they may engage CYP and families before, during, and after consultations with HCPs, dependent on the needs of individuals and families. Seemingly, there is also no referral or discharge process that needs to be followed, meaning that CYP and families can receive support occasionally or regularly, depending on their needs. They also have the flexibility to see them

outside of the hospital, including at home, a public meeting place, or at a VCSE activity or event. This was said to be aided by communicating with CYP and families using preferred mediums, such as instant messaging:

> *"A guy moved over to adults two years ago… he texted me a few months ago, being like, I'm having trouble with appointments, what do I do? He then knows that he can get an answer and it doesn't mean that he has to see me. He just knows it's a point of reference, where he can get information or help if he wants it, and then I might not speak to him for another year... I often explain to people… you can contact me any time on a week day… half eight [to] half five, and I will answer you or get back to you… after the weekend… I've never got something back like, 'why didn't you reply?'… I think as long as you're clear with the boundaries."* (Kendra, HCP – Youth and Family Support Worker)

6.8.9 Transition planning

An integral part of CYP- and family-focused care across the lifecourse involves readiness and planning for transition from paediatric to adult H&SC services, though it seemed as though not all centres systematically utilise interventions such as Ready Steady Go, as indicated in some transition guidelines:

> *"They are providing a clinic for young people but actually for doing the paperwork that goes… Ready Steady Go, they don't have time to do it… they don't have time to give that priority with the young people… we think that [Ready Steady Go] could be a self-management tool in terms of helping young people to take that control and helping young people to understand their condition."* (Megan and Chanda, VCSE professionals)

Participants believed that preparation for transition (and the actual process of transfer) should begin early on, typically during early adolescence, as a phased approach. They felt this linked to the process of increasing CYP's self-management capacity, and the importance of doing so, readying CYP for independently managing their H&W as young adults. Participants pointed out that while different centres may be meeting transitional requirements on paper, many problems are often experienced amongst young adults under the care of adult rheumatologists, such as non-adherence to treatment, and non-attendance in clinic. This highlighted how a deficiency in self-management capacity may be the underlying cause to such behaviours in adult H&SC.

Participants remarked that during the period of transition between late adolescence and young adulthood, CYP may value the support of a keyworker

or co-ordinator, such as a youth and family support worker, or CNS, to help provide individualised support bridging the void between paediatric and adult H&SC services:

> *"What I think they lack, the thing that's missing, is keyworker support from a specialist nurse at that age, 17, 18, 19, 20, mid 20s, when they have had a lot of support… they then move to a service where, like all adult clinics, they get given an appointment and they may or may not attend, that's up to them. They don't get phoned up, and if they choose not to have treatment, then… they will present with more difficult disease."* (Krishna, HCP – Paediatrician)

6.8.10 Annual review appointment

The potential of an annual review appointment in paediatric rheumatology was raised by some participants, similar to what is performed in other paediatric specialities, such as diabetes, as part of a focus on the wider physical and psychosocial H&W of CYP and their families. The purpose of such appointments would be to focus less on the clinical situation, and more on the broader issues, taking a more holistic, CYP- and family-centred approach to enable JIA to be optimally managed:

> *"What tends to happen in clinic, is you have… almost all the discussion is about… what's currently happening that week, with someone's joints. But actually, you might go then, for several years without discussing exercise. But if you had an annual review which included everything which was important, as well as the immediate management… the benefit of that is that it's just a slightly different discussion to the normal clinic discussion… we do a checklist of things to go through… it's part of a fairly widespread approach in pediatric diabetes, to have once a year checks, including psychology screening and dietetic review and health checks… [and] maybe the annual review process would be a way of just prompting the family and… the team to think about… those wider things."* (Krishna, HCP – Paediatrician)

Culture amongst HCPs appears to be a part of this approach, particularly within diabetes, alongside the fact that a significant part of diabetes is self-management, patient education, and adherence to treatment, which may come across as more important than in JIA. However, providing these opportunities, in particular psychology screening, was viewed as a positive step to identifying CYP who may slip under the radar, but whom require support. One specific example of a review-type clinic held in a paediatric rheumatology centre was then provided, where CYP had consultations with their paediatric

rheumatologist and CNS, before having the opportunity to meet other CYP attending the same clinic for a peer support session facilitated by the youth and family support worker.

6.8.11 Utilising technology

Finally, participants commented on the utilisation of technology within paediatric rheumatology, which has come to the forefront during the COVID-19 pandemic. The tensions and barriers in place prior to COVID-19, such as restrictions on the use of video conferencing software, seemed to have been relaxed once their requirement became urgent. While participants acknowledged the limitations in the use of technology to perform physical examinations, they felt that there are benefits for its use in other areas, such as psychosocial H&W, peer support, and self-management training, such as self-administration of treatment. Mention of electronic PROMs were also made, enabling CYP and families to complete PROMs before their consultation via a smartphone- or internet-based application, so that the results could be used in clinic to guide the conversation around CYP's needs and priorities.

During the COVID-19 pandemic, HCPs remarked that had remotely assessed CYP in order to triage whether they required face-to-face appointments, and it appears that post-COVID-19, H&SC systems may explore how care can be delivered. However, participants hinted to some concerns, namely overlooking potential issues in the absence of face-to-face contact, hence why a hybrid approach may be most useful. However, if increased utilisation of technology is to become a reality, participants felt strongly that the infrastructure needs to be fit for purpose to enable HCPs to perform their roles successfully:

> *"If I had a smart phone from the trust, yeah, I think I'd use it a lot more… we've all got an iPad now as CNSs, which is great in theory, and it was brilliant when we first got them, but actually the reality is I've lost all 4G on mine now… it's just like if you've got these devices we want to be able to use them properly, but it's the constraints of the IT as well… [when they do work], we do get things up on them, but it's not as easy as it sounds because it's not very quick, and you're in the busy clinic and you've only got a set amount of time you know you can give to this family."* (Caoimhe, HCP – CNS)

6.8.12 RQT5 Summary

CYP- and family-focused care across the lifecourse is foundational to promoting SSM amongst CYP with JIA and their families. The provision and quality of services can be influenced by a variety of physical and social environmental factors, including general awareness of JIA, CYP- and family-centred practices, variations in service delivery and system communication, integrated interdisciplinary services, and valuing SSM across the lifecourse. By taking a multi-disciplinary, biopsychosocial approach to care across the lifecourse, including consistent signposting to readily available information, education, and support, alongside timely, co-ordinated access to appropriate HCPs, CYP and families are more likely to feel informed, empowered, and better supported to competently manage their H&W.

6.9 RQT6: Inclusive and holistic education

6.9.1 Implementing legislation

Many families appeared to be unaware of legislation relevant to CYP with JIA, such as the Equalities Act 2010 and the medical conditions in school statutory guidance, which some VCSEs have summarised to aid their understanding. It was sensed from participants that schools seem to have different approaches to their medical conditions policies, and although statutory guidance is in place, implementation appears varied. This subsequently affects the manner in which schools support CYP with JIA. For example, some schools had refused to store and administer medication for CYP during the school day despite written permission, which in one example, resulted in the parent having to go to school every lunchtime. Clearly, an agreed protocol is required to protect everyone involved, while ensuring CYP's H&W is not neglected:

> *"That's all very highly policed and monitored. So… they go down and they get their tablets and there's a sign-in process… they have to sign for the tablet… phone the parent and tell them they've taken it… I would expect… all that needs to be agreed… in the care plan."* (Martha, Teacher)

There also appeared to be some confusion and inconsistencies with how CYP with JIA were included under the remit of the Special Educational Needs and Disabilities Co-ordinator (SENDCO):

"We weren't even down on the [SENDCO] register… the [SENDCO] had never even heard of [her]… they just don't seem to understand… the kind of illnesses like JIA. They don't know where to put them." (Esther, Parent)

One participant highlighted that the lack of ring-fenced funding for CYP with conditions like JIA and additional needs is an issue, in particular if they are not in receipt of EHCPs. They provided a vision whereby grants would be awarded to CYP with conditions like JIA – ineligible for EHCPs, to help support them in education, while providing them with the tools and holistic treatment required to self-manage their JIA and improve their QoL:

"I would say if you have a diagnosed medical condition… I think [Department for H&SC] should give that child a bursary… like an education healthcare plan, which comes from the government, so it comes a little bit [out of] Health a little bit out of Education, and we try to meet the child's needs within the school setting… we need some sort of agreement between Health and Education…to enable schools to have that flexibility... you think, God, there's all these children with health needs who are not getting anything, who are suffering in silence or else just not coming to school, because there's no funding." (Martha, Teacher)

6.9.2 Understanding, empathy, and appreciation of JIA

The level of understanding about JIA amongst schools appeared varied, with tension often arising between school and families when there was a breakdown in communication. Unfortunately, this can happen when schools do not identify with supporting CYP beyond their formal education:

"I know at the school I was at, they were like, we are a school, our job is to educate, that is it. We're only interested in her being in school or doing her learning… they'll say… that's a health need. Like, yes, but there's a part of that child and, even though it's a health need, it is impacting on their education." (Martha, Teacher)

There was an appreciation that school's may encounter lots of different conditions, and so they may not necessarily be able to understand every specific detail about every condition. However, what was seen to be more achievable was for school's to be more understanding, empathetic and appreciative of the individual CYP's needs, and what works best for them. This was seen to require close co-operation with CYP and their families so that everyone is aligned and appreciates the context of CYP's life beyond the

classroom, and how conditions like JIA are long-term, often invisible, and cannot be compared to acute conditions:

> *"I think the invisibility of JIA is what makes it quite a tricky one… they know the kids who've got the asthma pumps because… they've got to carry them around… they're a visible tool. I sometimes think it would be more helpful if JIA was more visible."* (Miriam, VCSE professional)

One participant touched upon the potential for lack of trust between school, CYP, and families, especially when CYP may have persevered unnecessary without seeking help – sometimes making them feel like fraudsters:

> *"I mean she looks [like] death by the end of the day and she can't walk… and they are like, oh well, you haven't put anything in place before… you're just making it up now… because you want extra time in your GCSEs... and I'm just thinking she's going in every month for infusions and stuff… insinuating that she was making up the disease and thinking they're not spending thousands of pounds on her medication just because she says it's bad… for her she would just keep coming back in tears thinking that she was having to prove that she wasn't a fake."* (Esther, Parent)

Participants believed that schools need to be aware of the complex impact of JIA on CYP's life at school, including their physical and psychosocial H&W, as well as cognition and learning. At the point of diagnosis, some felt it may be useful for the paediatric rheumatology MDT to consistently provide a letter to the school outlining the child's diagnosis, reflecting all aspects of JIA, not just the physical component; while also providing a list of suggestions for what the school need to evaluate with CYP and their families in order to ensure they are fully supported and included in education, both academically and socially:

> *"It's not just about their learning… it's about their whole school experience and feeling included and feeling involved… I make that very clear to the parent and say, we need to work together, because actually… it's just harder to return… friendship groups move on so quickly."* (Martha, Teacher)

It was also acknowledged that there is a complex interplay between absence, symptoms, and CYP's ability to keep up-to-date with the curriculum. Absence may mean that CYP may fall behind, which may contribute to, and exacerbate symptoms, such as anxiety and fatigue. Consequently, participants suggested that absences have to be accounted for as part of planning, with CYP and

families potentially benefitting from advanced notice of the curriculum, including readily-accessible materials to help ensure that CYP are not left behind when absent from school:

> *"The mum was saying that the… lesson was too quick, and I was thinking, yeah, it's too quick because she's missed so much… she has missed so much schooling that now she's finding the pace too quick because she's missing so much of the key concepts… and obviously it's fatiguing her much more because she's having to work so much harder in a lesson just to be able to keep afloat."* (Martha, Teacher)

6.9.3 Minimising attendance pressures

Attendance and absence from school are particular areas of concern for CYP and families. Some schools appear to lack understanding as to why it may be necessarily for CYP to be absent, while in some cases, CYP are being penalised as a result of their absence record, even when absences are authorised. Participants acknowledged that school attendance is a positive part of childhood, contributing to CYP's learning and psychosocial development, though in some instances, it seems to be approached in an oppressive manner, causing tension between school and families. Participants believed that schools should appreciate that for CYP with LTCs like JIA, they are likely to require multiple absences throughout their education:

> *"We do have issues with attendance, and we have a lot of families coming and saying to us that we need our letters for our clinic appointments, 'cause school won't let the child come out, without their clinic letter to prove we're going."* (Victoria, HCP – CNS)

Families described receiving generic letters regarding unsatisfactory attendance, which often felt insulting and caused immense anxiety:

> *"I rang up and I was like, you know, I was a bit surprised to get this letter… she was like, oh no, no, we do understand but they are just automatically generated… but it can be quite upsetting to people. I've seen loads on the Facebook page of people getting very angry about attendance awards and things that their children can never access."* (Esther, Parent)

Furthermore, many schools incentivise attendance, through various means, such as certification, awards, and other reward experiences. However, for many CYP with JIA, participants highlighted how they had been excluded from receipt

of such incentives, as a result of their absence, even when authorised and as a result of attending medical appointments, or a consequence of their JIA and the treatment required. When Darcie (aged 5–7) was asked about her experiences of school rewarding those with full attendance, she replied:

> *"But I don't get anything."* (Darcie, Child aged 5–7 with JIA)

This can lead to CYP being singled out as the individual who has lowered the class attendance percentage, placing unnecessary pressure and potential for bullying on CYP for events beyond their control:

> *"We hear, unfortunately, all the time how their children have been excluded from things like award ceremonies… special discos, and things, because their attendance hasn't been great… it seems that lots of children are almost… penalised… for being ill."* (Audrey, VCSE professional)

Although there is rationale for attendance policies, and it was recognised that schools are under certain amounts of pressure from authorities to monitor and address attendance for reasons such as truancy, greater sensitivity across the education sector in light of these experiences could help to eliminate unnecessary anxiety amongst CYP and families, who through no fault of their own, may require absence from school related to their H&W. Participants highlighted that absences may be inevitable on multiple occasions, including for medical appointments, but also when they are recovering from procedures and experiencing worse symptoms:

> *"Do they know about the Tuesdays? [when recovering from a medical appointment]… last time, I couldn't actually walk because of the thing."* (Darcie, Child aged 5–7 with JIA)

Teachers recognised that CYP need to be at home when they are feeling unwell, but on the other hand, when they are feeling more able, that school is the best place for them to be, with the correct support in place; caveated with the knowledge that should they deteriorate during the school day, that they should be allowed to go home. It was also recognised that parents may decide to keep CYP off school while they are in a flare, and when they do not necessarily know what to do for the best. This point related back to ensuring

families feel knowledgeable in self-assessing situations, and in seeking timely input from HCPs.

In occasional instances, a breakdown of communication between families and school may ensue, prompting the involvement of a school attendance officer. Though from some experiences shared, this has also happened inappropriately for CYP with JIA. Participants suggested that in order to change systemic attitudes towards attendance, influence has to emanate from government, and the inspectors and regulators responsible for assessing education providers:

> *"It's even above the school really, because… they're judged on attendance from Ofsted… it's like they're having to try and stick within a certain limit…"* (Xena, VCSE professional)

6.9.4 Communication between families and school

Communication between families and school appeared to be varied. However, a common finding was one of families having to proactively and in some cases, repeatedly, fight to get CYP the support that they required to succeed in education, which can be difficult for those who are disempowered and may struggle to talk confidently with schools:

> *"It was me that had to battle for ages with the school about getting her, you know, the sort of extra time, rest breaks… making sure that they'd got it set up. And I've had to keep going back to them because they keep mucking it up all the time."* (Esther, Parent)

Families described how documenting in writing their child's condition, its impact on their H&W, and how best to support them in school could be useful for school to gain a clearer understanding of JIA, supplemented by educational literature and a face-to-face conversation to inform staff on how to support CYP. This does put the onus onto families though. One family suggested using the end of one academic year as a prompt for preparing for the upcoming year:

> *"We put everything in writing and sent it through, and they read it and came back to us the next day and said we'd never realised… because when she's at school she's so happy and bubbly. She gets on with life. We didn't realise she has all this stuff going on. I think it helped that they were shocked by the great long list… they got a greater insight into some of the things that she has to go through… NRAS [National Rheumatoid Arthritis Society] do the*

[booklet] about young people with schools… so, what I did, when I had a physical copy of that booklet, I highlighted certain bits that I really wanted the school to take board… I gave them that copy so that they could see at a glance… the issues I'd be most concerned about." (Toby and Clara, Parents)

Parental anxiety, often rooted in mistrust, was said to be lessened by reassurance that CYP will be fully supported and monitored while in school – so that all CYP feel happy, safe, and cared for. Trust between families and school was seen to be important, and if broken, could cause distress to CYP and families, antagonising the relationship. Indeed, misaligned perceptions between families, school, and HCPs may also cause problems, particularly when stakeholders are not communicating openly and acknowledging the true impact of JIA, which may be more difficult in secondary schools when there are a greater number of staff involved:

"I have to say… a lot of the times when I speak to consultants, they'll say, no, I would expect over 90 per cent attendance or… you know, they will say what's reasonable for the school to expect." (Martha)
"I mean secondary schools should have mechanisms for passing those messages on, but in a primary school, I think it's easier to tackle and should be quite straight forward to get them all on side… but I've heard it both ways." (Miriam, VCSE professional)

Advanced planning and cooperation between school, families, and HCPs was viewed as potentially helping to avoid instances where there are differing opinions, for something seemingly as simple as avoiding clashes between medical appointments and mock examinations:

"Quite rightly a health appointment comes before everything… but… there have been a couple of times where I've had to… phone a parent and say… we've got our mocks, we'd much rather the child was here… and parents have said that [the hospital] hasn't been great in sort of supporting that, particularly if it's come from the school… there just seems to be this real kind of… like we're locking heads somewhat… and in actual fact every conversation I have with the health professionals we're each blaming each other… and I get onto them sometimes saying, will you stop telling parents we are going to X, Y and Z; we can't, we have not got the capacity. And they're like, but you should be… I could point the finger and say, you should be doing X, Y and Z [too]… it's just this public sector thing which does make it hard." (Martha, Teacher)

6.9.5 Communication between HCPs, VCSEs, and school

More consistent approaches to informing school about JIA, and their subsequent tailoring of support for CYP was highlighted, reflecting on the need for assessing and supporting CYP's holistic H&W needs, as well as their educational and cognitive development needs. This included transparently documenting what schools should implement, such as administering analgesics during the school day, and examination access arrangements, so that every school receives the same level of information from HCPs to better enable them to make actions, which could be through the IHP:

> *"A letter could go from the health services to the school… that if you have a child with [JIA]… the first thing you might need to look at is… their social and emotional and mental health… there may be absences and attendance issues… there may be issues to do with them eating because of the medication and lack of appetite, and that can bring on fatigue as well… Secondly… you might want to look at cognition and learning…"* (Martha, Teacher)

An ideal world was painted whereby the SENDCO, HCPs, CYP, and families would sit down and talk about all aspects related to CYP's H&W, and what would help them in school. Some believed the dialogue should be automatic for all CYP with JIA, beginning proactively at diagnosis, instead of reactively when issues arise. Participants felt that this could help to overcome some of the conflicts emerging from agreements on reasonable adjustments. VCSE professionals also highlighted how they had occasionally gone into school to help support the process, particularly for families who were struggling:

> *"If they feel… apprehension about going in on their own I've always offered and said, we're very happy to come in with you."* (Audrey, VCSE professional)

It was highlighted how from the pediatric rheumatology MDT, the occupational therapist, physiotherapist, and CNS may be best suited to engage with schools in helping them to identify how to practically support CYP, with written information from the consultant where appropriate and necessary. When this was raised with some HCPs, they were unclear within their MDT as to who has conversations with schools, indicating that the process of communication is inconsistent:

"I think we only have contact with schools, if there's an issue… there have been some occasions where we've had to write supporting letters for, if the child hasn't been too well… we'll get involved that way, but not as a general rule, we wouldn't contact every school for every patient, 'cause it's just too much." (Victoria, HCP – CNS)

6.9.6 Flexible and bespoke support

For some CYP, school may be an escape, where they can feel relatively normal, surrounded by friends, without necessarily having to think about JIA. Indeed it is a big part of their life, but one which may be lower down the priority list when considering the SSM of JIA:

"It's not hospital, and mum isn't around poking and prodding them with stuff, and they can just get on with it… these kids are properly fed up with the whole thing, and don't then want to have a big drama about it at school." (Miriam, VCSE professional)

While reasonable adjustments in school were seen to be a step in the right direction, participants felt strongly that this should not compromise CYP's psychosocial H&W – namely, feeling different to their peers. This prompts schools to think of discrete ways in which support can be provided inconspicuously, potentially involving all CYP in a certain class, so as to not isolate specific individuals:

"I think it's a real balancing act… you can have young people that are just really struggling… but if they did have the support, [like] being able to move around in a classroom, it will really benefit them. But they're like 'no, I'd rather struggle and not do that, because I don't want to seem like the odd one out…' if there were times within the classroom where there is a lot of moving around and stuff, that's a way bigger area of education… that will make the difference." (Xena, VCSE professional)

Unfortunately, not all CYP with JIA and their families are aware of the support that can be available to them in school, such as examination access arrangements. This was viewed to be overlooked by school if the child was not linked to the SEND register. Yet when they are, the process of securing such arrangements can be traumatic. Participants suggested that the assessment process used to gather evidence strips the situation of all context, and does not necessarily reflect the impact of JIA symptoms, including fatigue, on their cognitive abilities:

"They gave her a test to see how quickly she could type and then they kind of said, oh well, I don't think you're going to get it because you can clearly type… then they said, okay, we'll give you these cognitive tests… but of course she can write a small bit like that reasonably quickly. But the point is she's got an hour and a half exam… by the time you've finished writing there's a big difference between that and writing your first sentence… I was like these tests are not measuring what… her issue is at all… to me this is just ridiculous." (Esther, Parent)

Indeed, it is imperative to remember that JIA affects CYP differently – there is no one standardised support package; rather, support should be flexible and bespoke to individual CYP, and may require giving CYP the opportunity to trial different ways of working to find the one that best suits their needs and preferences. Teachers said that identification of these routines early on can then be used as evidence to the Joint Council for Qualifications to state that this is CYP's normal way of working, and with the appropriate medical evidence, could help to secure the necessary examination access arrangements required:

"I give them the full range of access arrangements to try out in the classroom, so that when it does come then to… year 10 and 11, that they can say, actually Miss, I don't want a reader, or I don't want a scribe, I'd rather use a laptop, you know, or whatever it is… subtle little things but they matter hugely." (Martha, Teacher)

Teachers highlighted that schools can implement a number of discrete and reasonable adjustments to help make life easier for CYP with JIA, without necessarily being made to appear visibly different to their peers. Aside from examination access arrangements, examples included access to lockers throughout their time at school, reduced timetables, additional processing time in lessons, differentiated homework requests, more time to move between classrooms, lift passes, and adapted scheduling to mock examinations:

"We kind of need to help them live with it, manage it and self-strategise as best they can… obviously I'm a teacher and I'll say, the main thing is you've got to get them into school and, once in, we can be, and we should be as flexible as that child needs for us to be… there's all sorts of wraparound little provision that… need to be put in place." (Martha, Teacher)

In addition, the provision of remote learning was also seen to be an important, proactive measure to have in place, pre-empting future scenarios where CYP may be absent from school. Two examples of remote learning

referenced were Bednet and Klassecontact, which allow CYP with conditions like JIA to connect to their classroom online. However, since the COVID-19 pandemic, a greater number of CYP and schools have used more readily-available software, such as Zoom, and so the implementation of such provision may be more easily available. Providing this opportunity for remote learning could help CYP who may be at home or in the hospital, to connect with their class and learn:

> *"It wouldn't have benefitted me when I was really poorly to do stuff, but actually, if I'm at home with a swollen knee and I've got have peas on it and sit on the sofa, I can [take part]… what I didn't feel there was, was it didn't feel like there was a set process… it's not about. Making any more work, it's about utilising what they do already."* (Betsy, Teacher)

6.9.7 Access to a safe space

It was suggested that CYP with JIA may benefit from access to a physical safe space, away from their classroom, where they could go, feel calm, and reduce cognitive overload. There, they could bring their work, or simply rest, and it would be preferable to them sitting in an office on a plastic school chair, or going home, while providing families with confidence that school is the best place for them to be:

> *"When you are experiencing pain, that really interferes with all your other core cognitive processes… how can you then be expected to learn, to socially interact? I would rather have a child in a… meditation room… mindfulness room on a nice yoga mat with some nice music… nice lighting… they could go and they could do sort of breathing techniques… [rather than being] at home. The pain won't go away just because you're at home…I certainly think for the majority of patients, if there was more confidence that the school had a little area that the child could go to… the parent [would have] some reassurance."* (Martha, Teacher)

This extended into the concept of school becoming more of an holistic centre for CYP with JIA, with access to CBT, for example, to help them manage their symptoms and be able to enjoy school life to the full:

> *"In an ideal world, for them to have… cognitive behavioural therapy around how to manage pain… I'm sure they're not doing that at home but school is the right setting. I would say… send your child in, they can come to the… meditation room… then with cognitive behavioural processes with other child[ren] that I've worked with, they say… they become much quicker at*

being able to get into the zone and override that feeling of anxiety, in the majority of cases… so that's what I would like to see and have the child in school… because this is a tragedy that the child needs for life." (Martha, Teacher)

6.9.8 Key trusted contacts

The need for key, trusted contacts in school was evident. Various suggestions were provided, including teaching assistants, academic mentors, class teachers/form tutors, and SENDCOs. Participants remarked that these contacts should be someone or a small group of people with whom CYP feel comfortable with, and confident to go to with their worries or concerns, so that the onus is not on CYP to seek help. Participants felt that it was important for at least a few professionals in school to have a strong understanding of individual CYP with JIA, including their characteristics, and what works best for them, as well as the capability within school to initiate change, or at least to initiate a conversation on CYP's behalf:

"Now she's got an academic mentor… [she] can go to her and say they still haven't sorted out what's happening about my exams, or whatever… that's been really helpful, because it just means that she's got somebody that she trusts that she can talk to… that person actually understands all of her." (Esther, Parent)

The role of the school nurse or a designated member of staff with more advanced training who can support CYP with their H&W in school was raised, while providing reassurance to families that school are looking out for their child's H&W:

"That's one thing I would want to ensure… is that there is a full-time medical officer in the school. They don't necessarily have to be a qualified nurse… but they can just be the medical person who has all the first aid… and they can be tracking… someone to feedback to the parents and say, they seem to be in… a lot of pain today… it just makes the parent feel that somebody is really looking after their child." (Martha, Teacher)

6.9.9 Proactive, responsive, and informed staff

Families felt that school staff could have been more responsiveness to their requests to support CYP, as well as speeding up the implementation of reasonable adjustments. Also, as part of making reasonable adjustments, families valued when schools were able to put contingencies in place for every

eventuality. This was seen to be aided by teachers getting to know CYP, both by engaging in discussion with them but also through observation:

> *"I remember going in and just observing… I just saw all the visual stress of him having to read this PowerPoint and I thought, you know, no wonder he's then getting headaches and he's still fatigued… he really needs not to be doing so much reading because it's bringing on the headaches, which is then causing the fatigue, and we know that inherently there can be difficulties with their eyes… the [SENDCO]… needs to get to know the child really well, and needs to have an open door."* (Martha, Teacher)

Participants felt that teachers must recognise that CYP may often persevere with their symptoms, without reaching out for help. This was seen as an experiential learning as part of CYP developing self-management competency, in being able to detect changes and seek appropriate help, though for many CYP, they do not necessarily have the level of self-efficacy required to do this. Hence, as adults *in loco parentis*, teachers should to be proactive in understanding CYP's behaviour and how it links to their H&W; prompting them to seek help when appropriate:

> *"I didn't take pain medication as much as I should have done, because I didn't want to… go out of class, but if on my book it said, sometimes I get really quiet when I'm in pain, or sometimes I lash out when I'm in pain, if a teacher had that, and they saw something that was out of the ordinary, they might say, do you need to go and get your meds, are you in pain? It takes a… very mature young person to recognise that they're struggling, to be prepared to put themselves out…"* (Betsy, Teacher)

However, participants felt this must be balanced with overprotecting CYP too much, so as to force them to behave differently out of fear or concern:

> *"They were just going to treat me like I was made of… ceramic, and I couldn't do anything, and I couldn't go anywhere, just in case something happened… and that was really hard… it was almost like they were saying, oh, we don't have a risk assessment."* (Betsy, Teacher)

Finally, it was recognised that other members of staff may need to be made aware of CYP's needs beyond the immediate teachers *in loco parentis*, so that they could look out for them and ensure they were supported. This may include lunchtime welfare staff and sports coaches, who may come into contact with CYP and may require some additional context, so as to not impact on their

H&W, while also observing and paying attention to their functioning in-between lessons.

6.9.10 CYP's adaptive behaviours

As previously mentioned, JIA can impact on CYP's behaviours in school, with many seen to persevere through their symptoms and treatment side effects, in order to achieve some normality akin to their peers. Some may also try to avoid their families having input in school, so as to not be seen as seeking attention or inflicting additional work for their teachers, for which some CYP feel guilty for doing. In addition, some of the steps which CYP must go through in order to secure additional support, such as examination access arrangements, can cause them great amounts of stress, sometimes making them feel that they must prove how ill they really are:

> *"She was feeling like she had to prove that she was poorly and I think anybody just looking at her every day just trying to struggle round the school, you know, knows that she is… it's really stressful, feeling like she was being tested the whole time."* (Esther, Parent)

Furthermore, there can be unintended consequences of reasonable adjustments and additional provision within school for CYP with JIA. Families, teachers and HCPs may view adjustments as positive, whereas for CYP, they may be seen as counterintuitive, having unintended consequences of adding further stigma and isolation from peers:

> *"Giving her all the things she needed, would make her feel even more different… even… a chair in assembly, instead of sitting on the floor, again just puts a big arrow over their head, saying I'm different… so my daughter quite soon began to refuse the things that were put in place to help her…"* (Miriam, VCSE professional)

Participants jokingly remarked on some occasions, CYP may be seen to take advantage of their diagnosis or manipulate others to be more lenient towards them. CYP have different characters and personalities, and this is something which participants felt should be acknowledged, but not used as an excuse for teachers not to believe CYP – but also for CYP to learn that they must take responsibility for their actions too, including appropriate use of adjustments in school. One of the most likely reasons to do this may to be excused from

participating in physical education, with teachers sometimes spotting the patterns:

> *"Some of our older children have got pass lifts… but we encourage them to… use the stairs, 'cause it's good exercise, and good to strengthen up your legs… but I think because they've got it, they just use it, and they're missing out on that, you know, activity. So, there's the other end… of the spectrum for that, which we've seen with some of ours."* (Victoria, HCP – CNS)

6.9.11 Peer education and support

Peer support in school was seen to be important for CYP with JIA, to help reduce feelings of isolation and segregation, as well as enabling CYP's psychosocial development to progress:

> *"Two of my friends also have something… it isn't JIA but it's something a little bit similar and it hurts for them to sit on the floor so they sit with me."* (Darcie, Child aged 5–7 with JIA)

However, not all CYP necessarily understand the implications of LTC like JIA, hence why increased levels of peer education could be useful in promoting a more empathetic and non-judgmental culture among CYP in general, removing the stigma associated with LTCs like JIA:

> *"Every child goes to a new class, [they] don't have to tell [their] new teacher and all [their] classmates, there is just a little video… about juvenile arthritis, I think that would be really helpful for children, because it must be so difficult and stressful to have to keep telling your class."* (Judi, VCSE professional)

A 'circle of friends group' was highlighted as an example by one teacher of how they have identified a group of trusted CYP to form a support network:

> *"I get… five or six kids together and I educate them about your condition. I tell them about your up days and your bad days… I'm really honest and really open with them, and I've spoken to all their parents and they've agreed to have their child participate in this… and then I just say to them… if [CYP with JIA] has a day of absence… go up and say, hi… we missed you yesterday, guess what? And then tell [them] a bit of news or a bit of gossip… we've got this project due in whenever… just sort of bring you back into the fold… I think it's made the child feel that actually, they want to be in, they don't want to be missed."* (Martha, Teacher)

Similarly, when teachers are planning their seating plans, participants felt it was a good reminder for teachers to consider where they seat CYP with JIA, so that they can maybe leave the room more discretely if required, or move around more easily. Additionally, consideration for who they are seated near was also flagged, so that they are near to a group of other CYP who may help to 'carry' them along.

6.9.12 RQT6 Summary

An inclusive and holistic education is foundational to the SSM of JIA by CYP and families. Consistent and correct implementation of relevant legislation, together with an inclusive and supportive culture, proactive and responsive communication, and minimal attendance pressures, can allow CYP to feel safe and happy in school, while reassuring families that CYP's H&W is school's key priority. This can be facilitated by the co-ordination of communication with key stakeholders, including HCPs and VCSEs, transparent and regularly documented needs (including contingency plans), proactive awareness of support opportunities, and constant involvement of CYP in all decision making.

6.10 Chapter summary

Six RQTs promoting SSM of JIA by CYP, their families, and professionals involved in their healthcare, wellbeing, and education were described in this chapter. Under each RQT, a range of themes describing key contexts, and mechanisms linked to outcomes were discussed, supported by excerpts from participants. CYP- and family-focused care across the lifecourse, as well as inclusive and holistic education, are key to influencing how CYP and families are involved in SSM of JIA, and the way in which they interact with other professionals involved in their healthcare, wellbeing, and education. The data presented in this chapter will now summarised as a set of RQTs in Chapter 7, informing the JIA-SSM framework introduced towards the end of Chapter 7.

Chapter 7: Refined question theories and framework

7.1 Introduction

This chapter assimilates the findings from the qualitative study presented in Chapter 6 into a set of RQTs promoting SSM of JIA by CYP, their families, and professionals involved in their healthcare, wellbeing, and education, presented at a higher level of abstraction. These RQTs have been tested and refined from the IQTs set out in Chapter 5. The JIA-SSM framework is then introduced, addressing the third objective of the study, to develop a preliminary framework promoting the SSM of JIA by CYP and their families, at individual, interpersonal, institutional, and infrastructural levels of context using RQTs tested with different groups of stakeholders.

7.2 Moving from IQTs to RQTs

The qualitative study presented in Chapter 6 used a realist approach to test, refine, and consolidate question theories promoting SSM of JIA by CYP, their families, and professionals involved their healthcare, wellbeing, and education. A set of seven IQTs were formulated in Chapter 5 based on an integrative literature review, presented in Chapter 2, and a document review of evidence related to SSM of LTCs, summarised in Chapter 5. The process of developing the RQTs is akin to 'digging for nuggets' of valuable concepts to unearth the underlying processes promoting the SSM of JIA (Pawson, 2006a). The process by which RQTs emerged from the evaluation are subsequently described.

7.2.1 Starting and ending with theory

This process of developing RQTs began and ended with the IFSMT (Ryan and Sawin, 2009). The middle-range theory aided the formulation of IQTs due to its three dimensions of context, process and outcomes (Fawcett et al., 2001; Meleis, 2011), providing a basis upon which to draft IQTs, essentially splitting 'process' into mechanism resources and responses. A number of other relevant frameworks and models identified during preliminary scoping exercises also guided the early development of the IQTs as previously outlined, including The Self and Family Management Framework (Grey et al., 2006; Grey et al., 2015), Family Style Management Framework (Knafl et al., 2008) and Paediatric Self-

management Model (Modi et al., 2012). Each framework or model added a different lens and 'nuggets' of information related to the context, mechanisms, and outcomes related to SSM management. The result was a set of seven IQTs which summarised key contexts, resource mechanisms, response mechanisms, and outcomes believed to promote the SSM of JIA. These were used as stimuli in interviews with key stakeholders to contribute to the refinement and consolidation of the IQTs into RQTs.

As data analysis proceeded, it became clear that there was naturally overlap between some of the question theories, which was anticipated given the complexity of the SSM of JIA. However, there was noteworthy overlap between IQT5 and IQT6, both of which related to the commissioning and prioritisation of services that were CYP- and family-focused. Hence, these two IQTs were integrated to become RQT5, with remnant concepts unsuitable for inclusion in this RQT incorporated into appropriate themes under remaining RQTs.

7.2.2 Question theories akin to programme theories

As previously remarked, the study, which used a realist approach to evaluation, did not examine a specific intervention *per se*, rather the more generic principles of SSM of JIA. This is one of the key differences compared to many other realist evaluations previously published, and is why the theories are labelled question theories, akin in type to programme theories, written at a middle level of abstraction (Westhorp, 2017). Although the RQTs have been refined, these are embryonic given the broad approach taken in this evaluation, and will most likely benefit from further testing and refinement to further explore causative patterns between outcomes, mechanisms, and contexts at the level of each RQT.

7.2.3 Building a picture of question theory connections

As the hybrid deductive-inductive thematic analysis of all qualitative data was completed (Fereday and Muir-Cochrane, 2006), the researcher used the annotation function of NVivo12 to capture his thought process making the connections between context, mechanism, and outcome within the themes identified. Written notes were also used to review the themes which emerged from the analysis, in order to identify and strengthen the connections between context, mechanisms, and outcomes under each RQT. The researcher used

retroduction to move from the tentative theories presented in Chapter 5 to the refined theories presented in this chapter, building upon, and refining, the CMO components underpinning each IQT first presented in Chapter 5 into sets of CMO configurations. This was a convoluted process of constant comparison across the themes identified and presented in Chapter 6, and the literature informing those theories as previously described. The IFSMT was continuously used throughout the process as a reference point, particularly when summarising the CMO configurations at higher level of abstraction as 'if… then' statements under their respective RQTs, to close the loop of starting and ending with theory.

At the individual level of CYP, and families, RQT1 and RQT2 identified a number of similar contexts and mechanisms, indicating the duality of SSM (*i.e.*, you cannot have self-management without some degree of shared-management), and the need to deliver SSMIs to CYP and families at a minimum. As highlighted in the integrative review presented in Chapter 2, family-focused interventions are uncommon for CYP with JIA and their families, and may represent an area for improvement to begin to address the disparity in support offered to CYP and families. The RQTs highlighted that contexts can be enabling or inhibiting in terms of their influence on the ability and desire for CYP and their families to engage in SSM practices; however the notion that context deserves attention when exploring SSM has only gained more attention in recent years (Sattoe et al., 2015). This was indicated at the individual level in RQT1 and RQT2, but also in the remaining RQTs at interpersonal, institutional, and infrastructural levels. Representing CMO configurations over the six RQTs assists in building a picture of how question theories are connected. CMO components operating at the institutional and infrastructural levels (*i.e.*, RQT5 H&SC and VCSE services and RQT6 inclusive and holistic education), inclusive of the social determinants of health, prime the environment for CMO configurations operating at interpersonal levels (*i.e.*, RQT4 amongst paediatric rheumatology MDTs and RQT3 with IHPs), as well as the CMO configurations operating at individual and interpersonal levels with CYP and their families (RQT1/2). With regards to outcomes, a content-based framework proved to be useful in grouping outcomes identified by participants in the study (Sattoe et al., 2015), with reference to the role, emotional, and medical management of JIA.

7.3 Refined RQTs

The six RQTs and their constituent CMO configurations are shown below (Figures 19-24), with direct comparisons drawn between IQTs and RQTs, showing where additions have been made from the findings of the qualitative study, referenced in Appendix 15. The plausible mechanisms (consisting of resources and responses) address the 'what', while the contexts address 'for whom' and 'in what circumstances', leading to possible outcomes. As each RQT is discussed, CMO configurations are presented in the text; however, the respective figures also display CMO configurations as a matrix of components, with multiple mechanisms likely to operate in light of the numerous contexts detailed.

7.3.1 RQT1: Self-management of JIA by CYP

The first set of CMO configurations underpinning RQT1 (Figure 19) relate to self-management support across the lifecourse for CYP with JIA. Positioned at the individual and interpersonal level, RQT1 is also influenced by question theories at institutional and infrastructural level, the outcomes of which play a role in priming the contextual environment for self-management of JIA by CYP. With regards to outcomes, promoting self-management among CYP with JIA in the short-term (proximal outcome) tended to focus more on improving role management (*e.g.*, socialising and communication skills), with emotional management (*e.g.*, self-efficacy) and medical management (*e.g.*, treatment adherence) regarded as longer-term (distal) outcomes from promoting self-management.

Providing individualised information, education, and support in a timely manner using a multichannel approach may improve CYP's knowledge and beliefs, as well as their self-regulation skills and abilities, accounting for CYP's specific circumstances and preferences (*e.g.*, growth mindset, personality, and health literacy and numeracy), in order to improve their role management skills in the short-term so that they do not feel overwhelmed, and emotional and medical management skills in the longer-term.

CYP also require respect, encouragement, and support from their social networks and professionals involved in their healthcare, wellbeing, and education, whom they have faith and trust in, in order to feel accepted and

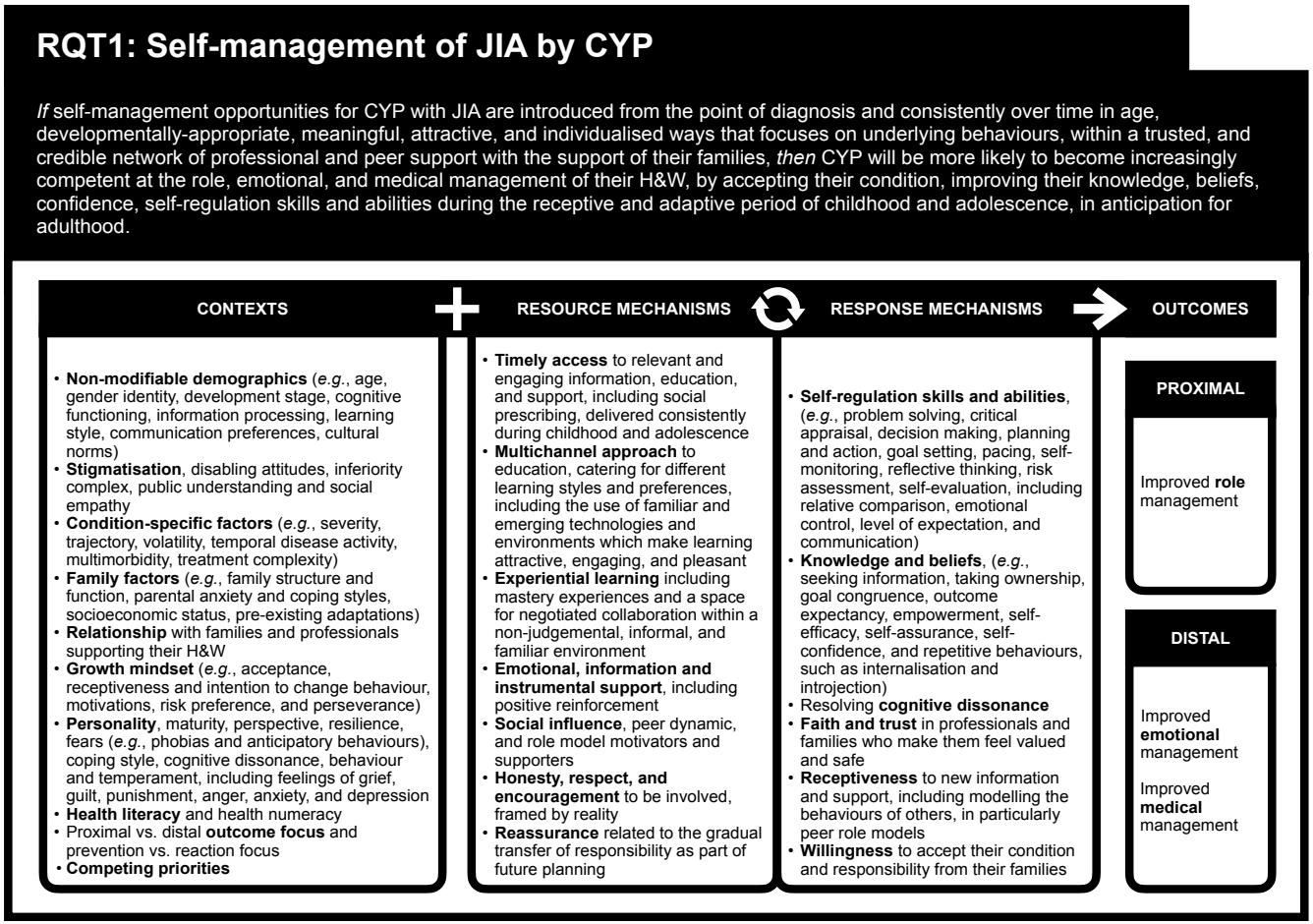
valued as an individual with JIA, in light of the stigma associated with LTCs like JIA.

Furthermore, CYP require reassurance and flexibility from professionals involved in their healthcare, wellbeing, and education, in order to assist them in feeling willing to accept their condition and share increasing levels of responsibility in cooperation with their families, so that they can improve their role management skills, in light of their individual condition-specific factors.

Experiential learning, including mastery experiences and a space for negotiated collaboration within a non-judgemental, informal, and familiar environment throughout childhood and adolescence can help CYP to have faith and trust in their family and professionals who make them feel valued and safe, which in turn can aid CYP in becoming autonomous in the role management of JIA, and subsequent emotional and medical management of JIA. This can be influenced by family factors, such as parental anxiety and coping styles, as well as CYP's relationship with families, and their approach to outcome focus (*i.e.*, focusing on short-term goals and the aggregation to long-term goals).

Finally, social influence, in particular peer support and role models living with JIA can motivate CYP to be receptive to new information and support, including modelling the behaviours of others, enabling them to become increasingly competent at the role, emotional, and medical management of JIA. This does, however, depend on CYP's personality and growth mindset, as well as competing priorities, which may deflect their attention away from socialising with those who have JIA or indeed other LTCs.

Figure 19. RQT1 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; H&W: Health and wellbeing; JIA: Juvenile idiopathic arthritis; RQT: Refined question theory; SSM: Self- and shared-management.

7.3.2 RQT2: Shared-management of JIA by families

The second set of CMO configurations underpinning RQT2 (Figure 20) concern the shared-management support for families supporting CYP with JIA. Similarly to RQT1, RQT2 is positioned at the individual and interpersonal level, and so is also influenced by question theories at institutional and infrastructural level, the outcomes of which play a role in priming the contextual environment for the shared-management of JIA by families. With regards to outcomes, promoting shared-management of JIA by families in the short-term (proximal outcome) tended to focus more on improving role management (*e.g.*, communication skills) and influencing CYP's self-management behaviours and autonomy, while emotional management (*e.g.*, self-efficacy) and medical management (*e.g.*, treatment adherence) were often regarded as longer-term (distal) outcomes, though perhaps with a more immediate focus for families, given their desire to improve their child's H&W as quickly as possible.

Firstly, HCPs and other professionals involved in the healthcare, wellbeing, and education of their CYP should build relationships with each family, recognising and embracing their unique family dynamic and needs (*e.g.*, condition-specific factors, level of previous disruption, and pre-existing adaptations), in order to build faith and trust with families through continuity of interactions and allowing a space for discussion, in order to improve how they interact with professionals, and subsequently support their CYP's H&W.

Families, like CYP with JIA, also require the provision of individualised and family-focused information, education, and support in a timely manner using a multichannel approach to improve the knowledge and beliefs of the family unit, as well as their self-regulation skills and abilities. In doing so, the family's expertise should be recognised, and their specific circumstances and preferences accounted for (*e.g.*, growth mindset, perspective, socioeconomic status, cultural norms, and health literacy and numeracy), in order to improve their role management skills in the short-term so that they do not feel overwhelmed, and emotional and medical management skills in the medium- to long-term.

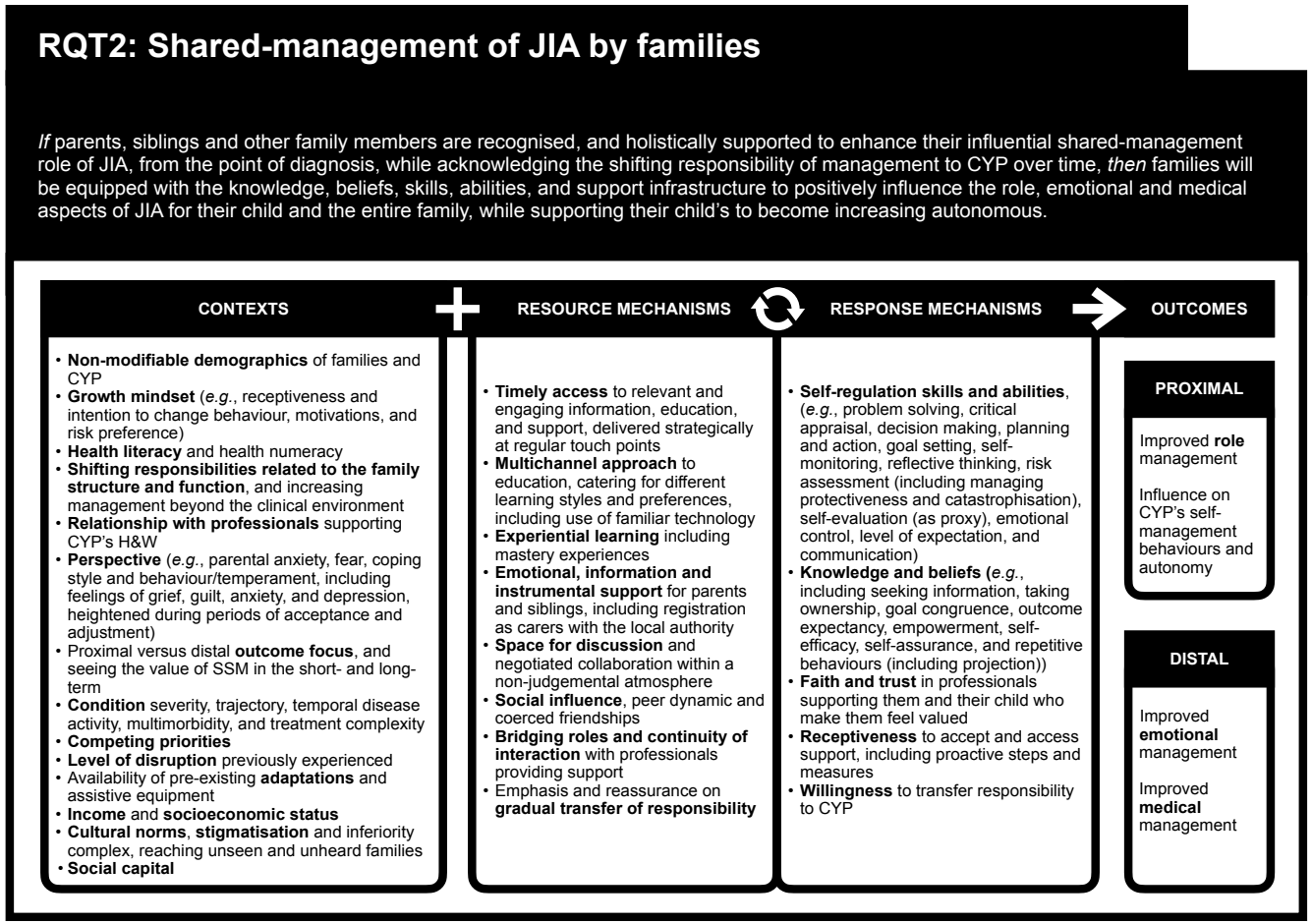
Emphasis and reassurance on the gradual transfer of responsibility from parents to CYP with JIA can aid parent's willingness to transfer responsibility when age- and developmentally-appropriate to influence CYP's self-

management behaviours and autonomy, when the shifting responsibilities related to the family structure and function are discussed, and other factors such as competing priorities, level of disruption previously experienced, and availability of pre-existing adaptations and assistive equipment are discussed.

Providing a space for discussion and negotiated collaboration within a non-judgemental atmosphere, including experiential learning, can help families to be receptive to accepting and accessing support, including proactive steps and measures as a result of their developing knowledge, beliefs, self-regulation skills and abilities, in order to aid the role, emotional, and medical management of their child's JIA.

Finally, social influence, in particular parental support and fellow parent and patient role models, can motivate families to be receptive to new information and support, including modelling the behaviours of others, enabling them to become increasingly competent at the role, emotional, and medical management of JIA, as well as influencing their CYP's self-management behaviours and autonomy. This does, however, depend on the perspective and growth mindset of family members, as well as competing priorities and social capital.

Figure 20. RQT2 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; H&W: Health and wellbeing; JIA: Juvenile idiopathic arthritis; RQT: Refined question theory; SSM: Self- and shared-management.

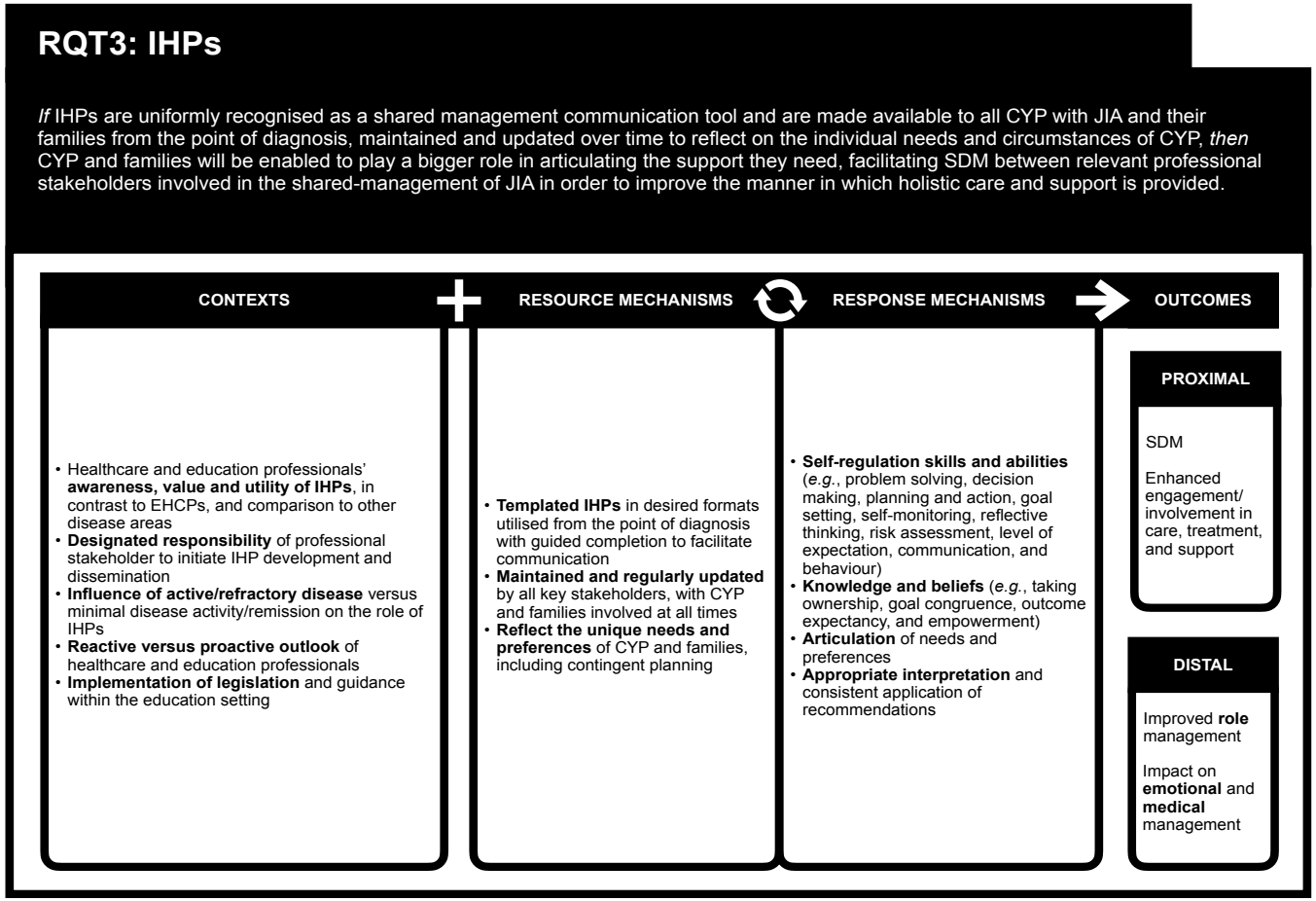
7.3.3 RQT3: IHPs

The third set of CMO configurations underpinning RQT3 (Figure 21) relate to IHPs as shared-management tools to aid other professionals in supporting the specific needs and preferences of CYP with JIA and their families. RQT3 is positioned more at the interpersonal level of context, as a tool to facilitate communication between CYP, families, and professionals involved in CYP's healthcare, wellbeing, and education.

Templated IHPs in desired formats utilised from the point of diagnosis can facilitate appropriate interpretation and consistent application of recommendations for individual CYP, when maintained and regularly updated, in order to enhance SDM and CYP's engagement and involvement in their care, treatment, and support. However, this is dependent upon an awareness of IHPs among HCPs and education professionals, including their value and utility, alongside implementation of appropriate legislation and guidance within the education setting to support CYP with JIA, and designation of responsibility among professionals stakeholders to initiate IHP development and dissemination.

Furthermore, IHPs should reflect and articulate the unique needs and preferences of CYP and their families, including contingent planning, in order to enhance CYP's self-regulation skills, abilities, knowledge, and beliefs, and subsequently to enhance their engagement and involvement in SDM in the short-term; improving their overall management of JIA in the long-term.

Figure 21. RQT3 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; EHCP: Education, health and care plan; IHP: Individual healthcare plan; JIA: Juvenile idiopathic arthritis; RQT: Refined question theory; SDM: Shared decision making.

7.3.4 RQT4: Paediatric rheumatology MDT approach

The fourth set of CMO configurations underpinning RQT4 (Figure 22) relate to consistent recognition and approaches within the paediatric rheumatology MDT towards the SSM of JIA by CYP and their families. RQT4 is positioned at the individual level of HCPs within the paediatric rheumatology MDT, but focuses on the interpersonal relationship with CYP and families, and the HCP's role in promoting SSM among CYP and their families, touching on some of the institutional levels of context which influence their approach and outlook. Regarding outcomes, distal outcomes typically focus on the role, emotional, and medical management of JIA, with proximal outcomes representing tangible short- to medium-term outcomes in the form of enhanced experiences of care and support.

Trusted, credible, and familiar professionals in multi- and inter-disciplinary teams should provide co-ordinated and continuous holistic care, by working in partnership with CYP and families to make them feel valued, respected, and part of the conversation, in order to develop a mutual understanding, level of familiarity, and trust with HCPs. Care should be guided by the biopsychosocial model of health, extended beyond the conventional clinical focus, in order to allow for compassionate and empathetic interactions, when appropriate training, social skills, and administrative support are in place. This is likely to enhance the experience of care to more accurately reflect the lives of CYP and their families, as well as improving HCP's relationships with CYP and families, benefiting the role, emotional, and medical management of JIA in the long-term.

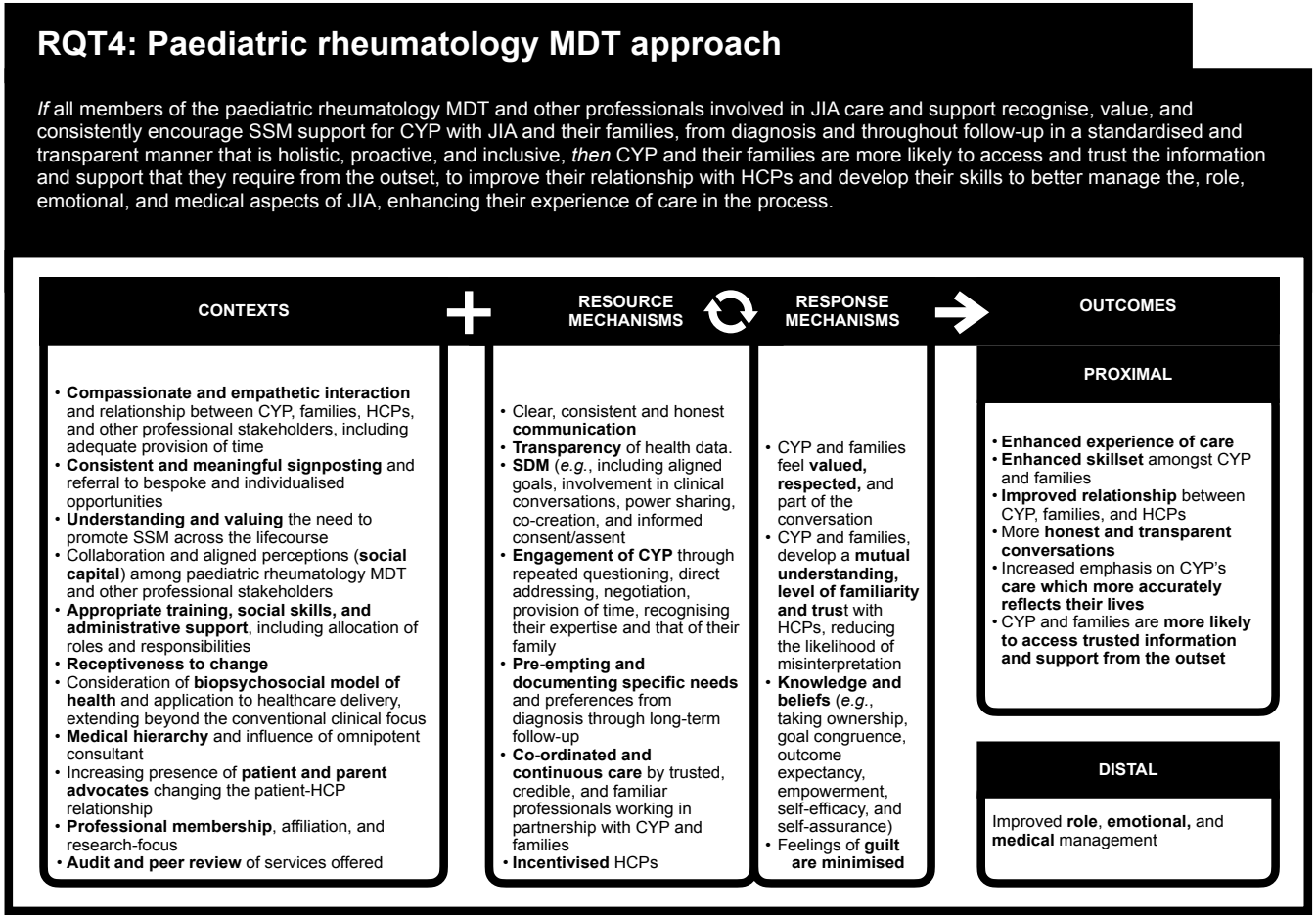
If HCPs actively engage CYP and their families in SDM, and pre-empt and document their specific needs and preferences from the point of diagnosis through long-term follow-up, CYP and families should not feel guilty for requiring support, and rather feel valued and respected, and have improved knowledge and beliefs related to managing JIA. Consequently, this will enhance experience of care to reflect their lives, improve relationships, and the skillset among CYP and families including their likelihood to access trusted information and support from the outset.

Clear, consistent, and honest communication including transparency of health data with CYP and families, can enhance how valued and respected CYP and families feel, in particular developing a mutual understanding between HCPs, CYP and families, to allow for more honest and transparent

conversations. This can be influenced by compassionate and empathic interactions, including adequate provision of contact time, social capital among the MDT and other professional stakeholders (*i.e.*, collaboration and aligned perceptions), as well as acknowledging medical hierarchies.

Finally, incentivising HCPs to promote SSM among CYP and families requires their own professional knowledge and beliefs to be influenced by an appreciation of the value and utility of SSM in practice. This could be facilitated by the sharing of best practices, through interactions with patient/parent advocates, participating in professional membership activities, and through auditing services offered by their team, and other teams. In turn, This might enhance the experience of care for CYP and families, allowing them to become more competent and the SSM of JIA.

Figure 22. RQT4 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; HCPs: Healthcare professionals; JIA: Juvenile idiopathic arthritis; MDT: Multi-disciplinary team; RQT: Refined question theory; SDM: Shared decision making; SSM: Self- and shared-management.

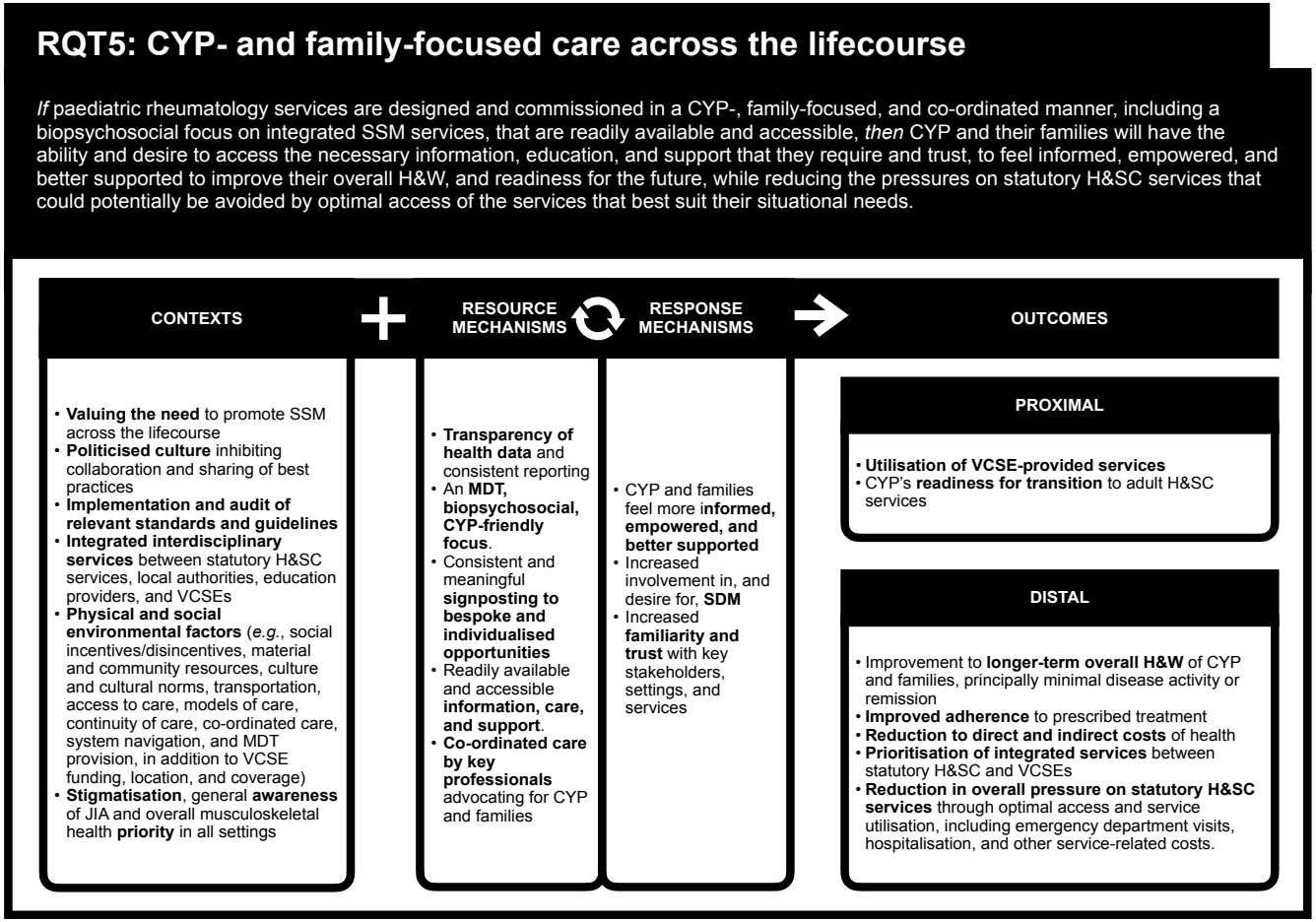
7.3.5 RQT5: CYP- and family-focused care across the lifecourse

The fifth set of CMO configurations sustaining RQT5 (Figure 23) relate to CYP- and family-focused care across the lifecourse for those living with JIA. This question theory is positioned at the institutional and infrastructural level, considering commissioned services aimed at treating and supporting CYP with JIA and their families; providing the framework for SSM at individual and interpersonal levels as described in earlier RQTs. Outcome patterns for RQT5 shift heavily to distal outcomes, recognising the longer-term impact of services designed to promote SSM across the lifecourse.

By taking a multi- and inter-disciplinary, biopsychosocial approach to care across the lifecourse, including consistent and meaningful signposting to bespoke and individualised opportunities and readily available information, education, and support, then CYP and families are more likely to feel informed, empowered, and better supported, resulting in utilisation of VCSE-provided services, as well as improvements to treatment adherence and longer-term overall H&W, which could reduce overall pressures on statutory H&SC services, when interdisciplinary services are implemented between statutory H&SC services, local authorities, education providers, and VCSEs who value the need to promote SSM across the lifecourse.

Co-ordinated care, including transition planning, by key professionals advocating for CYP and families, relies upon transparency of health data and consistent reporting, to allow for CYP and their families to be encouraged and empowered to be involved in SDM, through increasing familiarity and trust with key stakeholders, settings, and services supporting CYP with JIA. In turn, this could improve treatment adherence and longer-term H&W, while reducing costs of health and overall pressures on H&SC services. However, this requires consideration of various physical and social environmental factors, such as models of care and politicised cultures inhibiting collaboration, as well as prioritisation of paediatric musculoskeletal health across the lifecourse through implementation of relevant standards and guidelines.

Figure 23. RQT5 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; H&SC: Health and social care; H&W: Health and wellbeing; HCPs: Healthcare professionals; JIA: Juvenile idiopathic arthritis; MDT: Multi-disciplinary team; RQT: Refined question theory; SSM: Self- and shared-management; VCSE: Voluntary, community, and social enterprise.

7.3.6 RQT6: Inclusive and holistic education

The final set of CMO configurations sustaining RQT6 (Figure 24) relate to inclusive and holistic educational settings to enable CYP with JIA to secure equivalent educational attainment and social development to their peers. Like RQT5, this question theory is positioned at the institutional and infrastructural level, considering the role of education providers in supporting CYP with JIA and their families to flourish academically and socially, regardless of their condition. Outcome patterns for RQT6 are both proximal and distal; in the short-term, school (or other educational institutes) should be safe, supportive, and inclusive environments enabling open and regular dialogue with home; while distal outcomes focus more on long-term academic and social development milestones, as well as overall attendance.

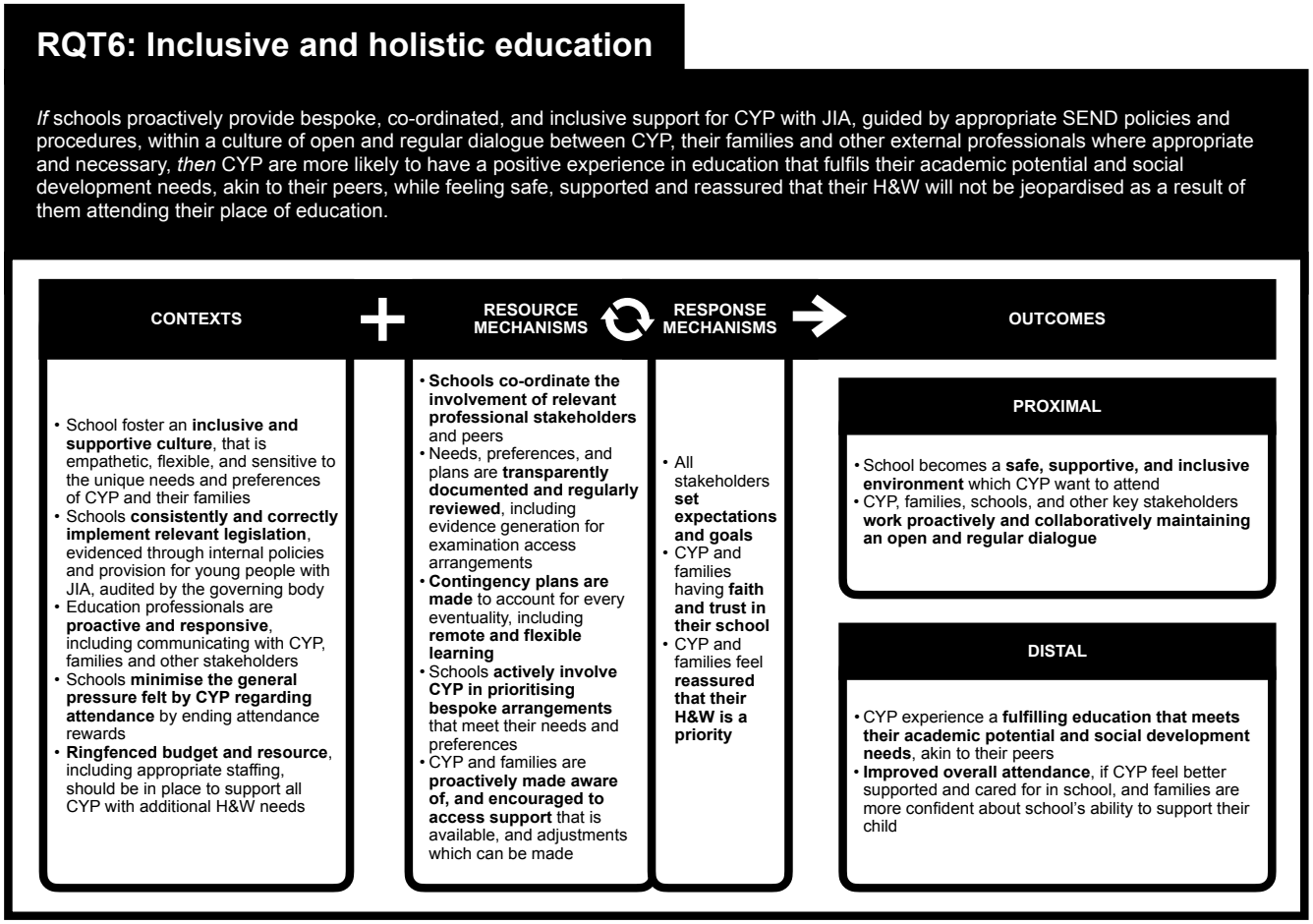
When CYP and their families are proactively made aware of, and encouraged to access support that is available, and adjustments which can be made, CYP and their families are more likely to have faith and trust in their school, as well as feeling reassured that their H&W is a priority. When schools consistently and correctly implement relevant legislation, evidenced through internal policies and ringfenced budgetary provision for CYP with JIA, school should become a safe, supportive and inclusive environment which CYP want to attend, aided by an open and regular dialogue between CYP, families, school, and other professionals. In turn, this should enable CYP to experience a fulfilling education that meets their academic potential and social development needs, akin to their peers.

Furthermore, when schools actively involve CYP in prioritising bespoke arrangements that meet their needs and preferences which are transparently documented and regularly reviewed (*e.g.*, safe spaces, trusted contacts, examination access arrangements), and co-ordinate the involvement of relevant professional stakeholders and peers to action such arrangements, all stakeholders involved are able to set realistic expectations and goals, to enable CYP and families to have faith and trust in their school, while feeling reassured that their H&W is a priority. Consequently, CYP, families, school, and other key stakeholders work proactively and collaboratively to maintain an open and regular dialogue, so that CYP feel school is a safe, supportive and inclusive environment. In turn, this could improve overall attendance if CYP feeling better

supported for and cared for in school, and families should feel more confident about school's ability to support their child.

Finally, if schools make contingency plans to account for every eventuality, including remote and flexible learning, all stakeholders will have clear expectations and goals for when CYP are unable to be physically present in school, or require a modified timetable, meaning that CYP and their families feel reassured that their H&W is a priority. However, for this to be a reality, and for CYP to feel supported and included in school so as to fulfil their academic potential and social development needs, schools and regulatory bodies must minimise the general pressure felt by CYP with JIA regarding attendance, by ending attendance rewards at a minimum.

Figure 24. RQT6 and its constituent CMO components



CMO: Context, mechanism, outcome; CYP: Children and young people; H&W: Health and wellbeing; HCPs: Healthcare professionals; JIA: Juvenile idiopathic arthritis; RQT: Refined question theory; SEND: Special educational needs and disabilities; VCSE: Voluntary, community, and social enterprise.

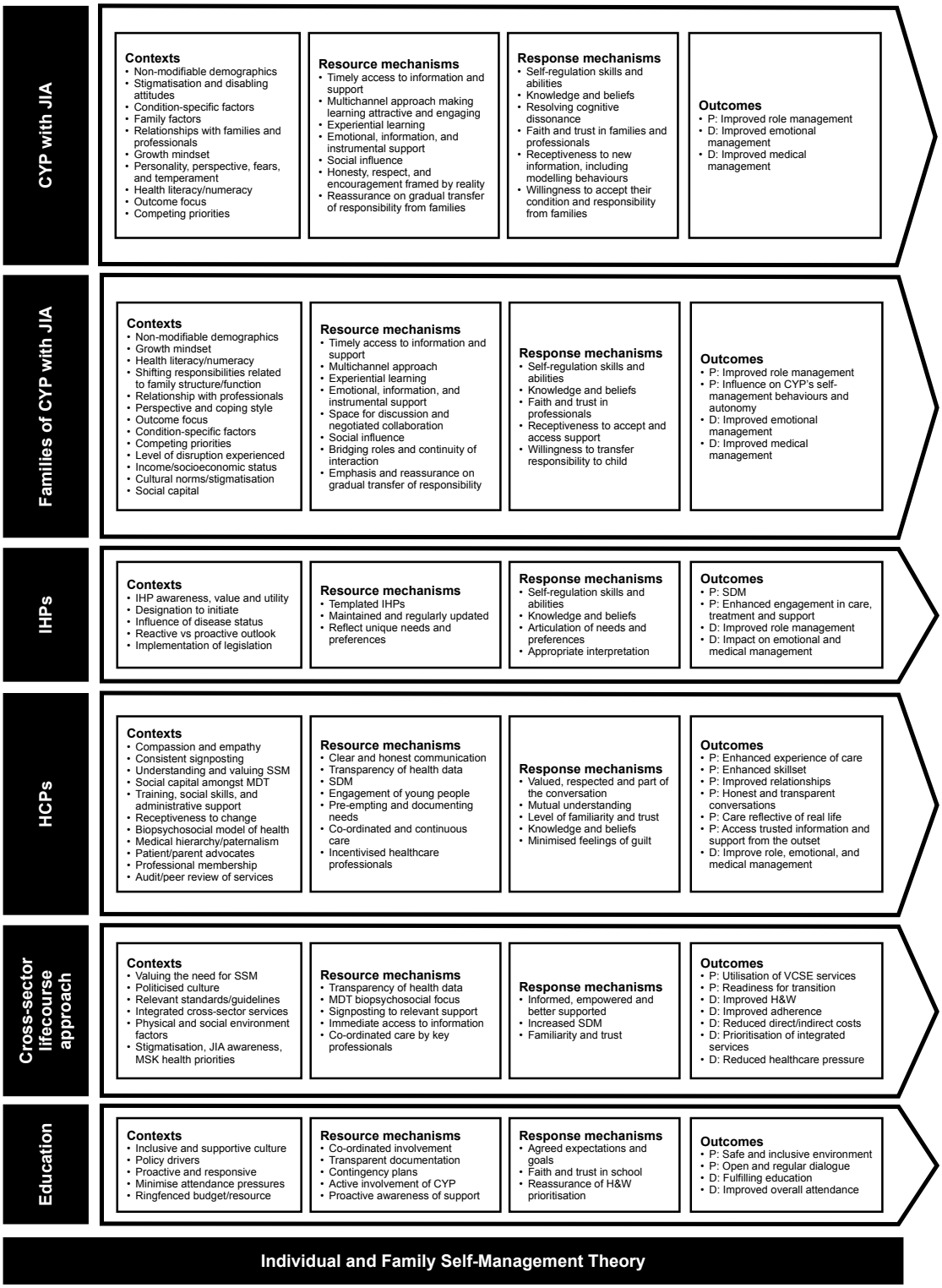
7.4 The JIA-SSM framework

The six RQTs and their constituent CMO configurations were combined at a higher level of abstraction into a preliminary framework promoting the SSM of JIA, underpinned by the IFSMT. The JIA-SSM framework (Figure 25), sets out the key principles believed to promote the SSM of JIA by stakeholders and organisations playing a role in influencing how CYP self-manage their H&W, as well as how families are involved in the shared-management of their child's H&W. Visualising the RQTs on a split level in the framework first highlights the complexity of SSM practices, while enabling respective stakeholders groups to interpret the question theory directly related to their discipline. The framework thus provides stakeholders with an appreciation that their actions are part of a larger phenomenon dependent on outcomes influenced by the interplay of contexts and mechanisms at varying levels, as well as how one set of outcomes can prime the context for subsequent actions, in particular those at an infrastructural and institutional level paving the way for an environment conducive for SSM by CYP and their families.

7.5 Summary

A preliminary framework, the JIA-SSM framework, was formulated in light of the RQTs promoting the SSM of JIA which evolved from the study after testing with participants in Chapter 6. This is guided by the underpinning mid-range theory, the IFSMT. The JIA-SSM framework provides an overview of six key foci pertinent to promoting SSM behaviours under the influence of different contexts. This includes two at the individual and interpersonal level for CYP people and families; one at the interpersonal level through the use of IHPs to facilitate communication; one at the individual and interpersonal level amongst HCPs; and two at the interpersonal, institutional and infrastructural level amongst cross-sector H&SC, VCSE, and educational providers. Visualising the RQTs on a split level in the framework enables respective stakeholders groups to interpret the question theory of direct relevance, while emphasising that their actions are part of a much larger process dependent on outcomes influenced by the interaction of contexts and mechanisms at multiple levels.

Figure 25. The JIA-SSM framework



CYP: Children and young people; D: Distal; H&W: Health and wellbeing; HCPs: Healthcare professionals; IHP: Individual healthcare plan; JIA: Juvenile idiopathic arthritis; MDT: Multi-disciplinary team; MSK: Musculoskeletal; P: Proximal; SDM: Shared decision making; SSM: Self- and shared-management; VCSE: Voluntary, community, and social enterprise.

Chapter 8: Discussion and conclusions

8.1 Introduction

A realist approach to evaluation was conducted with a range of stakeholders to promote SSM of JIA by CYP, families, and professionals involved in their healthcare, wellbeing, and education. This resulted in a new, preliminary framework promoting SSM of JIA, titled the JIA-SSM framework. This was the first study to use a realist approach to understand the underlying mechanisms and the influence of context on SSM-related outcomes, in addition to applying the IFSMT within a paediatric rheumatology context.

The overall aim of the study was to explore how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. An integrative review of the literature was designed to identify and describe the empirical evidence base on the use of SSMIs for CYP with JIA and their families (Chapter 2). The review identified research groups, refined the aim and objectives of the study, the methodology and methods of which were described in Chapters 3 and 4, to inform the question theories (Chapter 5) which were subsequently tested in a qualitative study (Chapter 6). Deductive, inductive, and retroductive analysis of the data revealed a collective set of previously undefined contexts at differing levels (individual, interpersonal, institutional, infrastructural) and their influence on SSM-related mechanisms and outcomes for CYP with JIA (Chapters 6 and 7).

This final chapter discusses the unique contribution to knowledge that is provided through the findings of the study, including how the JIA-SSM framework extends existing theory pertaining to the SSM of juvenile-onset LTCs, as well implications for research, policy, and practice; recommendations for future research; a critique of the strengths and limitations of the research; and personal reflections from the researcher.

8.2 Unique contributions to knowledge in the context of the existing literature

There were several aspects of the research presented in this thesis which make a unique and important contribution to the literature, and wider understanding of SSM of JIA which could potentially apply to other juvenile-onset LTCs. A new,

preliminary framework promoting the SSM of JIA, titled the JIA-SSM framework, emerged from the study. This is the first framework in paediatric rheumatology focusing on SSM, which applies and extends the work of Ryan and Sawin (2009) with their middle-range theory, the IFSMT, by adding granularity to their model, particularly around contexts, and by breaking the process of self-management down into mechanistic resources and responses. Recognising the two-way interaction between resources and responses can aid scholars in theorising how an intervention may exert its effect (*i.e.*, how people respond to resources offered to them through interventions). There are various published frameworks and models pertaining to the SSM of juvenile-onset LTCs from different perspectives which the study presented in this thesis aligns with (Grey et al., 2006; Knafl et al., 2008; Modi et al., 2012; Grey et al., 2015). However, each of these respective frameworks and models focus on different aspects of self-management and shared-management, without comprehensively addressing all aspects related to SSM, in particular the contextual influencers at interpersonal, institutional, and infrastructural levels. Thus, the JIA-SSM framework produced in the study extends these previously published frameworks and models relating to self-management of LTCs in CYP by explicitly identifying and describing the contexts, mechanisms, and outcomes at multiple contextual levels, adding clarity behind the underlying forces operating within SSMIs and the environments within which they are implemented.

Interestingly, while the research presented in this thesis was ongoing, a new overarching model of self-management in CYP with LTCs was published, incorporating key elements of 13 self-management conceptual models, with an emphasis on the shift of agency from families to CYP (Dall'Oglio et al., 2021). This overarching model resonates with the JIA-SSM framework, insofar as addressing the different contexts, processes, and outcomes related to SSM, and the unequivocal role of CYP and their families in managing LTCs. The JIA-SSM framework does, however, further extend this model, to more clearly describe the types of contexts, the level at which they operate, a more granular details of the mechanisms at force, and a content-based approach to proximal and distal outcome assessment. Theory inevitably plays an important role in guiding the design, evaluation, and implementation of SSMIs; however, many scholars often feel ‘alienated’ and confused by theory which discourages them from applying theory to their work (Davidoff et al., 2015). The JIA-SSM

framework attempts to demystify some of the concerns scholars may have regarding the use of theory in designing, evaluating, and implementing SSMIs by providing a theory-informed framework, guided by the literature and a small group of participants, which details '*what works, for whom, in what circumstances, and why*'.

Furthermore, there are a number of other novel contributions from the study to the wider research literature. There is no awareness of studies using a realist approach to understand outcomes, underlying mechanisms and the influence of context at multiple levels on SSM of JIA, and there is no awareness of studies having considered the complexity of SSM of JIA, not only by CYP and parents, but other professionals involved in their healthcare, wellbeing, and education. In addition, the study was based on a middle-range theory, the IFSMT (Ryan and Sawin, 2009), which is believed to be a unique application of this theory in JIA and the wider speciality of paediatric rheumatology. Finally, to best knowledge, few qualitative studies in JIA have included multiple stakeholders from different backgrounds together in a single study, in particular professionals from VCSEs and education.

The research presented in this thesis is also intended to stimulate reflection and discussion regarding SSM of JIA with multiple stakeholders, including consideration on how SSMIs can be better designed over the lifecourse to improve outcomes for CYP and families, while recognising the need for longer-term investment into building SSM capacity (Coulter et al., 2013). A recent integrative review exploring theoretical and contextual considerations for SMIs for CYP with LTCs also suggested that involving CYP in the design and delivery of SMIs is most effective when said interventions do not outweigh cognitive ability or maturity level (Camp-Spivey et al., 2021). The reviews of the literature presented in this thesis have also provided a picture of current provision of SSM across the UK and Ireland, including the services offered by VCSEs, which has not been explored to this level previously across the sector (Stones et al., 2021). The focus on SSM also reaffirms the reality of juvenile-onset conditions such as JIA which often requires CYP and their families to develop a range of complex skills to manage their H&W effectively (Nightingale et al., 2015).

The research presented in this thesis has also contributed some evidence towards the unmet needs identified by the European Alliance of Associations for Rheumatology (EULAR) RheumaMap (EULAR, 2019a), including: 'optimising

the international network of expertise and information in JIA' by consolidating information about services across sectors; 'identifying and elucidating psychosocial support systems for CYP with JIA and their families', and 'identifying optimal mechanisms to enable CYP with JIA to have educational attainment'.

This thesis and the focus on SSM of JIA is timely given the rapidly changing policy landscape in the UK, such as with the NHS Long Term Plan (NHS England and NHS Improvement, 2019a) and its focus on supported self-management (NHS England and NHS Improvement, 2020). This focus is likely to be followed by an increasing number of initiatives developing new or modified SSMIs; therefore, it is hoped that the findings from the research presented in this thesis will bear relevance not just to paediatric rheumatology, but to others working within paediatrics, and indeed adults, in supporting thinking around the underlying mechanisms operating within SSMIs, and more broadly the environment within which SSMIs may be implemented.

8.3 The JIA-SSM framework prompts a shift towards a multi-intervention, multi-disciplinary, and multi-agency approach

The JIA-SSM framework provides a panorama of forces determining how CYP self-manage their H&W, as well as how families share the responsibility for said management. The framework proposed firstly confirms the contribution of CYP, as well as their families, in managing their H&W (Ryan and Sawin, 2009), and secondly, emphasises the role of other professionals with responsibilities for healthcare, wellbeing, and education, and the influence of organisations, evidence, and policy more broadly on the way in which SSM becomes a reality (Grady and Gough, 2014). The JIA-SSM framework demonstrates the complexity of SSM, prompting a deviation away from solitary endeavours aimed at mastering specific outcomes – towards a lifecourse, multi-intervention, multi-disciplinary, multi-agency approach which works with CYP, and families as partners from the point of diagnosis, in equipping them with the information, skills, and behaviours to be able to competently take control of their H&W, with support of relevant professionals and potential use of innovative technologies which CYP are familiar with (Edwards et al., 2021).

Ould Brahim (2019) calls for a social-ecological approach to SSM, emphasising the importance of SSMIs influencing multiple levels to address

systemic effects. As evidenced by the findings from the research presented in this thesis, SSM policies and practices must shift focus from individuals towards a 'patient nexus' encompassing the other individuals and structures which influence people's H&W (Tausig, 2013). The JIA-SSM framework provides a basis for this shift towards a 'patient nexus', supported by the 2021 EULAR recommendations for the implementation of self-management strategies for people with inflammatory arthritis (Nikiphorou et al., 2021). SSMI architects should also consider other concepts pertinent to SSM of JIA, such as: assessment of patient activation (Hibbard and Gilburt, 2014) and self-management behaviour phenotypes (van Dongen, 2014); consideration of other relevant frameworks regarding behaviour change, including the BCW (Michie et al., 2011) and EAST (Service et al., 2014; Burd and Hallsworth, 2016c); tools to support goal setting (*e.g.*, WOOP) (Adriaanse et al., 2010); and intervention functions and typologies (de Silva, 2011; Michie et al., 2011).

Considering infrastructural levels of context, reform appears to be on the horizon in the UK, in part as a result of the COVID-19 pandemic and the collaborations and co-ordination of services which ensued during this unprecedented period (Department of Health and Social Care, 2021). These proposed changes appear to recognise the pivotal role of local government, VCSEs, and education providers in supporting people's H&W alongside the NHS. In their white paper setting out legislative proposals for a health and care bill (Department of Health and Social Care, 2021), the Department for H&SC expresses its intention to build upon the NHS Long Term Plan, by elevating the role of integrated care systems to provide better joined up H&SC across the lifecourse, while reducing bureaucracy impeding decision making. Moreover, NHS England and NHS Improvement have pledged to move to a 0–25 years' service by 2028, enabling "*person-centred and age-appropriate care for individual needs, rather than an arbitrary transition to adult services based on age*" (p.50) (NHS England and NHS Improvement, 2019a). However, it is disappointing to see little emphasis on self-management in the 'Getting It Right First Time' review of rheumatology, where self-management, regarded by the report authors as a component of patient education, is only mentioned in the context of COVID-19 to ensure patients are not disadvantaged by not receiving a face-to-face consultation (Kay, 2021). Although paediatric rheumatology fell outside the scope of this report, it is concerning that there is not a greater

emphasis on the importance of self-management in rheumatology more broadly. A further avenue of exploration in relation to SSM at infrastructural levels is the Health Anchors Learning Network, a joint initiative between NHS England and NHS Improvement, and The Health Foundation, aimed at developing the role of the NHS as an anchor institution for the community, using principles of co-design to tackle health inequalities (The Health Foundation, 2021). Other potentially relevant areas of infrastructural reform include NHS Personalised Care (NHS England and NHS Improvement, 2019b), intended to give people more choice and control based around what matters to them, improved access to primary and community care, SSM, social prescribing, H&W coaches, and care co-ordinators. These prospective changes provide an opportunity for SSM to come into focus, in a way in which is embedded across the NHS, local government, public agencies, VCSEs, and education providers, together with policy makers, senior managers, and international experts, so that CYP and their families have equitable access to information, education, and support. The influential role of infrastructure was evident in the research presented in this thesis, and so changes which move towards more integrated ways of service delivery are welcomed.

From an education perspective, the HCSA have called for the Department for Education and Ofsted to take joint action in understanding their responsibilities and creating quality medical conditions policies which are subsequently published on schools' websites. Secondly, for Ofsted to instruct inspectors to routinely scrutinise schools' medical conditions policies as a routine component of inspections, in order to foster a culture of prioritising H&W needs of CYP with LTCs like JIA (Health Conditions in School Alliance, 2021). The findings from the research presented in this thesis add to the narrative underpinning this action towards making education more inclusive and accessible to CYP with LTCs like JIA, recognising school as a place of education and socialisation, where H&W is promoted (Colao et al., 2020).

The findings from the study presented in this thesis also emphasise the importance of recognising and valuing the expertise of CYP and their families, which can be difficult when a power imbalance remains between HCPs and CYP (Boland et al., 2019), which was evidenced by participants. Indeed, the dynamic of the patient-HCP relationship has evolved from medical paternalism to one that increasingly respects patient and caregiver autonomy (Kilbride and

Joffe, 2018). However, true respect of patient autonomy and SSM expertise is inconsistent across the board. This was highlighted in a qualitative patient-led study by the late Rosamund Snow, who demonstrated some of the obstacles that patients can experience when HCPs feel uncomfortable with their high level of expertise. This can be compounded when HCPs deny access to SSM resources (Snow et al., 2013). Although this study was in adults, the general tone resonates with the experiences of families of CYP with JIA, who desire to be valued and recognised as part of the MDT in supporting their child's H&W, with self-assurance and reassurance forming a firm basis to their subsequent behaviours. One of the key principles in this regard is to afford autonomy to CYP and their families, supporting their confidence at every stage of their journey with JIA, while balancing autonomy with developmental capacity and maturity (Grootens-Wiegers et al., 2017). This was supported by an integrative review exploring theoretical and contextual considerations for self-management of LTCs by CYP, which demonstrated that CYP with adequate support from their families, HCPs, and peers, can use age-appropriate self-management strategies effectively (Camp-Spivey et al., 2021).

The contrast between subconscious and conscious thinking also became clear during the study presented in this thesis, at the point of interaction between individual CYP and family members and the external world. Small changes to subconscious beliefs and values can result in longer lasting changes to behaviour with less struggle, whereas changes to conscious thoughts and emotions requires greater force and struggle (Hauser et al., 2018). Influencing the subconscious through the right contexts is one step towards influencing positive changes to thoughts and emotions – focusing particularly on empowerment and growth, rather than problems or issues (van Staa et al., 2021). Experiential approaches to learning and 'mastery experiences' seemed to be more appealing for CYP, incorporating social learning theory (Bandura and Walters, 1971), which argues that people learn best from others. This learning by 'mastery experiences' may be more powerful for CYP in contrast to being told what to do, particularly when having to weigh medical advantages against social advantages, as they strive to be 'normal' (Ghio et al., 2021).

As the study proceeded, it also became increasingly clear that there was a lack of systemic SSM support for CYP and families, which should be a continuum of SSMIs and strategies aimed at different tasks requiring different

skills (Sattoe et al., 2015). To achieve this, short sightedness must be overcome to envision the long-term impact and outcomes of SSMIs to enable CYP and their families, to competently manage their H&W across the lifecourse. Greater investment upfront may pay dividends later (Kemperman et al., 2017). While current SSMIs may serve their purpose, there is a broader regard for developing SSM capacity across the lifecourse, requiring a multi-lens approach.

8.4 Strengths and limitations

Methodologically, there were strengths and limitations for various aspects of the research presented in this thesis. Overall, a realist approach provided non-prescriptive framework for promoting SSM of JIA, offering flexibility in terms of the scope of the research, though equally challenging at the same time, due to limited standardised methods (Pawson and Tilley, 1997). Such theory-driven approaches provide an understanding of the outcomes which interventions are designed to address, by elucidating the underlying mechanisms which mediate people's responses to resources offered by interventions. This resonated with the overall aim of the study presented in this thesis, going beneath the surface to identify underlying processes operating within SSMIs aimed at CYP with JIA and their families. However, one may argue whether the realist approach of uncovering contexts, mechanisms and outcomes may be overly ambitious due to the potential infinite amount of underlying generative mechanisms. Similarly, it can be impractical to explore every piece of literature regarding patient education and SSM. However, by implementing various steps throughout the study, including an integrative review, IQT development and subsequent testing, across broad areas pertaining to SSM in JIA, it is anticipated that the key principles and 'nuggets of evidence' have been captured, balancing rigour, relevance, and richness. Realist evaluation has also been argued against, on the basis that the approach can be deterministic when categorising behaviour change as a series of static CMOs (Porter, 2015), and risks the researcher imposing leading questions and encouraging participant to agree with their proposition. However, appropriate application of the teacher-learner cycle (Manzano, 2016) demonstrates how suitable questioning can stimulate thinking which accepts, refines, or refutes IQTs, making the participant an active partner in the research process, instead of a mere contributor.

One limitation of the realist approach used in the study was its lack of mixed methods, with only qualitative data used to test and refine IQTs. Observations of SSMIs may have been useful, and a survey to further test and refine theories with a wider group of participants, especially CYP, parents, and other family members, would have added value, and increased the credibility of the findings. However, these could form future components of research to further test and refine theories identified within the JIA-SSM framework. Others may question why a realist approach was selected over alternative methodologies and frameworks, such as process evaluation (Moore et al., 2015) and the Intervention Mapping protocol (Bartholomew et al., 1998). Indeed, all of these approaches are theory-driven and recognise the challenge of inadequate implementation of interventions which can compromise their intended effectiveness (Fernandez et al., 2019). However, the researcher felt that realist evaluation went beyond description of the context and expected outcomes, by focusing on the generative mechanisms firing under certain contexts. Indeed, all mechanisms are processes, but not all processes are mechanisms in the realist sense, since they are invisible and can evade empirical capture (Westhorp, 2018). Process evaluations, for example, are not usually focused on this invisible level; rather, targeted at what is visible. When the study was being planned, a realist approach seemed the most appropriate and logical methodology since it explores both the visible and invisible mechanisms at work, and can be applied to more than one intervention *per se* – rather a broader approach potentially across many interventions.

There were also challenges along the way in terms of dealing with uncertainty, disagreement and apparent conflicting data during analysis and the most appropriate approach to take. This involved trial and error, shifting from coding for CMOs to taking a question theory lens, given the infinite number of possible constituent CMO components. However, these conflicts in hindsight are sources of rich data and where higher level interpretation can evolve from examining the data repeatedly and from different perspectives. This was compounded by the fact that there were few published, practical examples of analysing data with a realist lens using CAQDAS (Dalkin et al., 2021). The paper by Dalkin and colleagues, published towards the end of the study, was particularly useful for the researcher in proceeding with analysis using a question theory lens, with some adaptations to suit the study's aims and

objectives. This was also the researcher's first exposure to realist approaches to evaluation, as it was for his academic supervisors, which provided a unique and at times, difficult, learning experience. Upon reflection, if the researcher was to conduct a realist evaluation again, he would make a number of changes. Firstly, he would set boundaries early on, recognising that a realist evaluation, in particular as a PhD study, cannot possibly explore all dimensions of a complex intervention or indeed a set of complex interventions. He would also ensure that adequate resource is allowed for the realist evaluation, given that the time allowance was severely underestimated given the iterative nature of realist evaluation. Finally, he would look to budget for greater stakeholder involvement throughout the realist evaluation cycle, including patient/parent research partners to be co-researchers, since this could add greater depth of clarity to how theory is gleaned, refined, and consolidated. As Punton et al. (2020) remark, "*realist evaluation can provide deeper understanding which can lead to better policy – it can be hard work, but it is worth it*." (p.11).

The study benefited from various forms of triangulation to strengthen the quality of the data collected from IQT development through to RQTs informing the preliminary JIA-SSM framework. These included data, theory, and multidisciplinary triangulation. Added benefit may have been sought from investigator and methodological triangulation, though this was not feasible within the scope of the research presented in this thesis.

Recruitment of participants was also challenging for a number of reasons. Firstly, it was problematic to identify CYP, especially once COVID-19 pandemic social distancing guidance was imposed (Public Health England, 2020). The nature of how recruitment occurred also meant that no interviews could be conducted in a face-to-face setting. While this has limitations in terms of being able to build rapport and observe non-verbal communication, it did not necessarily restrict the quality and quantity of data collected, and also meant that participants from across the UK were able to participate. Remote interviewing also meant that the researcher was able to follow the interview schedule more easier, record fieldnotes, and redirect questions in response to answers. Unfortunately, the way in which NHS REC approval was sought meant that recruitment of CYP had to be undertaken via one participant identification centre, and was not permitted through other avenues, such as through VCSEs and self-referral on social media. This resulted in several self-referring CYP

keen to participate being unable to do so. Combined with the COVID-19 pandemic, this meant that only one child participated. While the researcher was disappointed that he could not recruit more CYP, or other intended stakeholders, such as siblings and grandparents, the aim of the study was not to seek personal narratives; rather, to refine question theories pertaining to SSM. This also helped to account for a sample of participants who may have had more interest in SSM than the general population, providing some objectivity to the discussion. While the researcher believes that the limited participation of CYP (and to a lesser extent, parents) in the study will not have negatively impacted the study findings, given the theory-driven approach to evaluation with relevant literature of their experiences, there remains some level of uncertainty as to whether the framework truly addresses their needs and preferences regarding SSM. This does, however, present an opportunity in the future to explore the JIA-SSM framework with CYP using age- and developmentally-appropriate methods. Under different circumstances, the researcher would have explored other avenues for widening recruitment, one of which may have been to apply for ethical approval from a University-based REC for more flexibility regarding recruitment of CYP from VCSEs, or request a different NHS REC with more experience of qualitative research, research with CYP, and user-led research to review the study application. The researcher's experience with the NHS REC also prompts for inquiry into inadequate training and awareness among REC members about 'expert by experience' researchers, as well as qualitative research and research with CYP more broadly, since deficiencies in these areas are suspected to have caused greater restriction on what the researcher was able to do within the study, in particular, regarding recruitment.

In addition, the discussion guide was not piloted ahead of conducting interviews with participants. This could be viewed as a weakness in ensuring the questions asked were valid and reliable; however, the realist approach to interviewing facilitated this process using the teacher-learning cycle, which aided the refinement of interview questions over time, moving from theory gleaning to refinement and consolidation. A realist approach lends itself to a more informal conversation between the researcher and participant, guiding the discussion around key areas related to SSM, though for some interviews, data obtained may have lacked depth or spread across all question theories, hence

why it was important to conduct the number of interviews that were previously outlined, to have a reasonably secure appreciation that the majority of points had been captured.

Given the researcher's perspective, some participants were already familiar with the researcher which helped to build rapport and a more honest conversation on the topic. Measures taken to reduce researcher bias were already outlined. Too close or over-immersion in a research setting can obscure the objectivity and critical analysis of data, particularly of qualitative data; however, the realist approach counteracted that notion. In reality, tension between the patient and researcher role were uncommon. However, familiarity of the study context facilitated collection of rich data, since the researcher was able to understand and empathise with participants, and explore different aspects given his familiarity with presented topics that may have otherwise been overlooked. He acknowledged his beliefs and perspective, and was transparent throughout in terms of his duality as an 'expert by experience' researcher, aided by frequent contact with experienced supervisors. Other opportunities to engage with different people with different perspectives were also beneficial to evolving the study design and subsequent interpretation of the data, through various opportunities while undertaking the research presented in this thesis. These included research seminars, conference presentations, and discussion with colleagues from multiple stakeholder backgrounds.

8.5 Implications for research, policy, and practice

The findings from the study have implications for research, policy, and practice related to SSM of JIA. Firstly, the study has highlighted the importance of theory in guiding the development and implementation of more suitable SSMIs that have the capacity to be more effective by recognising mechanism-outcome patterns, and the influence of different contexts; prompting for more widespread use of theory by SSMI architects, and explicit reporting of how theory has guided research (Davidoff et al., 2015). The JIA-SSM framework presented carefully uses language reflecting the Social Model of Disability (Goering, 2015), reframing medical or 'negative' language with a more 'positive' language that recognises CYP with JIA as human beings, which others professionals should consider in their own practice. The study has also shown that there is a wealth of information, education, and support available for CYP with JIA and

their families, to an extent where excessive amounts of such information can overwhelm CYP and families (RCPCH, 2021). Therefore, the answer is not necessarily for more information, but for a more targeted approach which condenses and consolidates information, and delivers it in an individualised manner using preferred channels and mediums which CYP and families feel comfortable with. This became clear during the study, where participants found difficulties in being able to see the 'bigger picture' of what was available, and how to access such information and support. Finally, the findings of the study have also spotlighted the role of VCSEs and education providers in collaborating with HCPs and H&SC services to support CYP with JIA and their families in the SSM of JIA. Attention should be paid by all stakeholders involved to identify opportunities to enable greater co-operation between all stakeholders, keeping CYP and their families at the heart of all decisions, and most importantly, involved at every step of the process. One clear shared-management tool to facilitate this collaboration is through IHPs, as demonstrated in the study.

8.6 Recommendations for research, policy, and practice

Finally, the study has illuminated several points-to-consider in order to promote SSM of JIA. These include:

- Theory-driven approaches to intervention development and evaluation, which reveal underlying mechanisms, and the influence of context on outcomes should be considered for designing and evaluating SSMIs;
- Routine clinic assessments to support SSM should include psychosocial screening and assessment of patient activation;
- Health literacy and numeracy should be considered when developing SSMIs;
- Cultural changes and partnerships, which are people-focussed, involve shared priority, responsibility, and power, should be prioritised;
- The process of finding the right information, education, and support, at the right time, in the most accessible format and route possible should be improved;
- A paediatric, adolescent, and young adult rheumatology SSM network across the four devolved nations of the UK should be established to

promote cohesion and integration of services across sectors, by involving all relevant stakeholders, with the possibility of pooling resources, reducing/sharing costs, and improving ways of working;

- Existing infrastructure in the UK should be leveraged to promote greater collaboration within and beyond rheumatology across sectors, in addition to collaboration with international networks;
- A cross-sector roadmap of SSM information, education, and support services across the UK should be produced;
- A clinical summary document detailing H&SC and support provision for CYP and families, relevant to their local context, should be produced by local H&SC providers.

8.7 Recommendations for future research

The study also identified the need for research in the following areas:

- Potential further testing of the RQTs developed and presented in this thesis, in particular with CYP with JIA and parents, and other stakeholders from a wider range of backgrounds, to confirm, refine, or refute whether the RQTs developed are fit-for-purpose;
- Application of the JIA-SSM framework in relating to ongoing and future SSMI development, to assess whether the framework is applicable and capable of influencing intervention development per the MRC framework for the development, evaluation, and implementation of complex interventions;
- Employment of mixed methods for evaluating SSMIs, so that qualitative methods can relevantly capture relevant contexts and mechanisms influencing the outcomes assessed using quantitative methods;
- Assessment of the impact of IHPs on SSM of JIA, and communication between professionals from healthcare, VCSEs, and education.

Finally, given the amount of data collected from the qualitative study presented in Chapter 6 of this thesis, it would be immoral to disregard anonymised data of value. Therefore, anonymised research data of long-term value will be deposited in the Research Data Leeds Repository (University of Leeds, 2021b), in accordance with the University of Leeds Research Data Management Policy,

so that it can be repurposed by other researchers in the future. All participants consented for this during recruitment.

8.8 Personal reflections of the PhD journey

My PhD has been a rollercoaster in every sense. After graduating with a Bachelor's degree in Biomedical Sciences, I made the 'leap' to a PhD in healthcare. I embraced every opportunity to develop my knowledge and skillset, within the scope of achieving goals related to the research presented in this thesis. I created a training and development plan, mapped across the four domains of the Vitae Researcher Development Framework (Bray and Boon, 2011). I viewed my PhD as a development opportunity, while also conducting publishable research to impact the community I serve. I have thoroughly enjoyed the experiences and challenges since I commenced my candidature in 2016, having pushed myself beyond my comfort zone in every sense. There have been, though, some more sombre occasions, which I have a duty to raise. I have witnessed first-hand, the injustices that 'expert by experience' researchers face as a result of paternalistic attitudes within a system plagued by inherent, unconscious biases regarding patients to be inferior to those in positions of authority and power – who have claimed our contributions to be *"unrepresentative, invalid, and unobjective"* (p.1507) (Riggare, 2020). Thankfully, I have worked with many individuals who value and respect my skills, experiences, and integrity, though sadly, it only take one experience to lower the tone. Unfortunately, my PhD has also associated with a significant amount of sadness – I started this journey with my mum by my side, and sadly she is no longer here to see me complete it. When mum was diagnosed with metastatic pancreatic adenocarcinoma in July 2018, my world was shattered. It is in these times when you realise what really matters in life – and although I adore my work, family took precedent. Throughout 2018 into 2019, I continued my PhD while caring for my mum. However, in 2019, the pressure was unbearable, and so I temporarily suspended my studies, which evidently impacted participant recruitment. During this time, mum died, and I returned to work in 2020 with a piece of me missing. At this point, I re-energised my study, which after several weeks of re-approvals, was immobilised by the COVID-19 pandemic. Inevitably, my plans have changed from those set out at the start of

my PhD, but nonetheless, I am proud of the research that I have conducted, and the independent researcher I have become during this time.

8.9 Final conclusion

This research has fulfilled its aims of exploring how SSM of JIA can be promoted across the lifecourse by CYP, their families, and professionals involved in their healthcare, wellbeing, and education. Using a realist approach, a qualitative study involving multiple stakeholders has enabled a new, in-depth appreciation of the contexts, underlying mechanisms, and outcomes associated with SSM of JIA, thus adding knowledge to the field. The JIA-SSM framework, underpinned by the IFSMT, was presented as a new, preliminary framework to guide the design, evaluation, and implementation of SSMIs for CYP with JIA and their families. The JIA-SSM framework encourages a shift towards a multi-intervention, multi-disciplinary, multi-agency approach to working with CYP, and families as partners from the point of diagnosis, in equipping them with the information, skills, and behaviours to competently take control of their H&W across the lifecourse, when permitted to do so in a conducive and supportive environment. Further work is recommended to contextualise, apply, and validate the JIA-SSM framework with SSMIs being applied in practice, recognising the fluidity of contexts and how their dynamic has the potential to change over time, and subsequent influence the underlying mechanisms under which SSMIs operate.

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Appendices

Appendix 1. VCSE services for JIA in the UK and Ireland

VCSE	Coverage	Project/programme name	Project/programme description*
Arthur's Place	UK	Online magazine and YouTube channel	Digital articles and videos for and by young adults (~18-35 years) on topics related to living with JIA and other RMDs.
		Arthur's Place Social	A closed Facebook group of >1,200 young adults (~18-35 years) with JIA and other RMDs.
CCAA	England and Wales	Family support weekend	Annual weekends enabling CYP and families to meet others with JIA. The weekend provides insight into JIA management and aims to provide CYP with the confidence to achieve their goals and ambitions using a combination of fun activities and informative support sessions.
		CCAA local groups (formerly JIA Matters)	A network of local groups managed by volunteer parents, to share experiences and mutual support.
		Award badges	A set of six award badges for CYP with JIA (and siblings) to recognise their individual efforts. Nominations for award badges are made via the CCAA website.
iCAN	Ireland	iCAN Just for Teens	A support network just for CYP with JIA aged 13-19 years, including: i) a private Facebook group moderated by youth representatives; ii) organised events (*e.g.,* pizza making, cinema trips, and make-up tutorials); iii) annual weekend away; and iv) workshops (*e.g.,* transition).
		iCAN family days	Family days hosted 2-3 times per year in various locations around Ireland. CYP with JIA participate in several activities (*e.g.,* indoor rock climbing and kayaking) supervised by qualified instructors. Parents participate in a series of presentations and discussions from guest speakers. These days provide opportunities for families to meet each other, share their experiences, and learn more about JIA.
		iCAN coffee and chats	Hosted several times a year, these informal coffee and chat sessions enable parents to get together in a local café, share their stories and experiences, learn new information, and gain new friends.
		Social media peer support	A private Facebook Group for parents to join for peer support.
		Local support	A team of local parent representatives based in several counties across Ireland, offering telephone conversations with other parents. Medical advice is not provided.
		Kipo	A book for CYP about JIA, aimed to open communication channels between parents and CYP. The book gives parents tools to understand how their child is feeling, and to explain what JIA is, through identifying with Kipo the monkey, who has JIA.
JIA@NRAS	England and Wales	Family fun day	Supervised outdoor adventures for CYP with JIA (*e.g.*, pond dipping, and team games). Paediatric rheumatology experts are also present for discussions with parents. Refreshments are provided.
		Bushcraft day	Targeted at CYP with JIA aged 10-16 years, involving a series of fun and informative activities helping to equip CYP with the skills and knowledge to thrive in the natural environment.
	England, Scotland and Wales	JIA and me art competition	A free art competition open to CYP with JIA aged 4-16 years, who are invited to prepare artwork in advance, and then attend a location for judging, prize giving and the opportunity to meet other families.
		RheumaBuddy	A free smartphone application designed for CYP with JIA to record, manage and discuss their symptoms, as well as a resource for parents to understand their child's arthritis, record levels of pain, and photograph flares.

VCSE	Coverage	Project/programme name	Project/programme description*
Juvenile Arthritis Parents and Families UK	UK	Juvenile Arthritis Parents and Families UK	A closed Facebook group of >2,700 parents and families of CYP with JIA.
Juvenile Arthritis Research	UK	A Little Box of Hope	A package of information, practical items and a plush Monkey toy named Kipo, for newly diagnosed CYP with JIA and their families.
		Kipo's Hero	A book for CYP about JIA, aimed to open communication channels between parents and CYP. The book gives parents tools to understand how their child is feeling, and to explain what JIA is, through identifying with Kipo the monkey, who has JIA. A personalised 'Kipo's Hero Award' certificate is also provided.
Kids Like Us	West Midlands	Family day	A one day activity event for CYP with JIA and families (*e.g.*, building dens and having a picnic).
		Family weekend	For all of the family to be with other people living with JIA, supported by HCPs to answer questions.
		Break away	For CYP with JIA to enjoy a holiday along with other CYP, and a full team of HCPs. The aim is to enable CYP to have some fun, while learning to self-manage and providing families with respite.
		Outreach events	Social outreach events for CYP with JIA and families (*e.g.*, ten pin bowling and pottery painting).
Olivia's Vision	UK	Forum	A website-based forum to enable people living with uveitis and families to ask questions and share experiences.
SNAC	Scotland	Annual parent days	Parent information and support days with HCPs presenting updated information.
		Family weekends at Crieff Hydro	An annual fun information and networking weekend offering the whole family a break together while learning more about JIA, supported by HCPs.
		Fun activity and networking days	Various activities for CYP with JIA (*e.g.*, soft play, climbing, picnics, swimming, pantomimes, and visits to leisure venues), with a chance for families to meet and network.
		Family contacts and Facebook group	Local area family contacts (*e.g.*, telephone conversations) offer information and support, in addition to a closed Facebook group for parents.
		My Journal	A journal co-designed by CYP with JIA to: i) record test results, hospital appointments, and other important dates; ii) write/draw how CYP feel; iii) map joint pain; iv) aid communication with HCPs; v) Read CYP testimonies; vi) Learn to say terms (including glossary); and vii) Learn things to help.
		Buzzy® devices	Supply Buzzy® devices which help CYP to cope with the pain of injections using a patented combination of cold and vibration to replace pain with temperature and movement.
Teapot Trust	UK	One-to-one art psychotherapy	One-to-one art psychotherapy services for CYP and their families, primarily in hospital settings, upon referral from HCPs.
		Art Therapy at Home	One-to-one virtual art therapy services delivered online via secure video conferencing, in addition to a range of freely downloaded resources to use at home.
Versus Arthritis	UK	Arthritis Tracker	A smartphone application created for and with CYP aged 13-25 years, allowing CYP to rate their symptoms and visualise summaries for use in SDM (*e.g.* pain, side effects, activity, sleep, and emotions). Versus Arthritis support is also signposted.
		Arthritis Virtual Assistant	A conversational agent known as Arthritis Virtual Assistant, powered by artificial intelligence to help people with RMDs to answer their questions 24 hours a day.

VCSE	Coverage	Project/programme name	Project/programme description*
		Helpline	A free national helpline, available 09:00 – 20:00 GMT/GMT+1 Monday to Friday, for people to access information and support. Enquiries are made and answered by phone call, social media, email and post.
		Online community	An online community, hosted via the Versus Arthritis website, providing peer-to-peer support for people with RMDs and their families. There is a specific 'young people's community thread' for those aged <25 years.
		Young People and Families Service	A service across the UK between Versus Arthritis and paediatric rheumatology MDTs, where CYP with RMDs and families can self-refer, and HCPs can refer CYP into the service, which includes contact in the clinic with Youth and Family Support Workers, as well as phone and email support, and access to wider Versus Arthritis offerings, detailed below.
	England	Family fun days	Sporadic family fun days of sports and wellbeing, enabling CYP with RMDs to participate in a range of activities (*e.g.*, tennis, golf, and yoga). These are open to all abilities, and designed to enable CYP to enjoy themselves while taking part alongside their families. Other VCSEs are sometimes present, and refreshments are provided.
		Local social meet ups	Regular small, social meet ups for CYP with RMDs aged 11-18 years in Birmingham, London, Manchester, and online. These are fully funded, insured and managed by qualified and experienced staff, and provide a chance for CYP to meet others, listen to talks, and experience different activities.
		Virtual events	Virtual events organised in light of the COVID-19 pandemic, for CYP with RMDs, typically organised on the last Saturday of every month from 16:00 to 17:30 GMT/GMT+1.
		Residential events	Residential events held in the North and South of England to bring CYP with RMDs and families together for a series of informational talks and activities.
		Social media peer support	A private Instagram account for CYP with RMDs in England to join for peer support.
	Northern Ireland	Jointz	A series of fun and discussion-based activities are delivered to support CYP with RMDs and families. These include: i) residential activity weekends for 11-18-year olds (*e.g.*, rock climbing and paddle boarding); ii) one-day activity events for 12-17-year old; iii) one-day workshops for 12-18-year olds and their friends/family (*e.g.*, adulting with arthritis and pain management); iv) CYP and family events; v) parents information seminar; and vi) online quiz and catch up for CYP aged 13-25 years.
		Online community	A private Facebook Group and Instagram account for CYP with RMDs aged 13-18 years to join for peer support, as well as a private Facebook Group for parents.
		Pantomime and family party	An annual group trip for CYP with RMDs to the pantomime, followed by a party and dinner.
		Volunteering opportunities	For CYP with RMDs aged 16-25 years, to become event youth leaders, and social media moderators.
	Scotland	Joint Potential	A self-management and personal development programme, integrated within rheumatology services, designed to establish a sustainable and integrated pathway for CYP with RMDs aged 16-25 years. Self-management is on the agenda at each consultation. There are five key outputs: i) sessions for CYP and families in clinic; ii) one day/virtual one hour self-management workshops (*e.g.*, pacing, sleep, and pain); iii) residential personal development and self-management weekends (*e.g.*, activity, self-image, and good relationships); iv) peer support network; and v) information sessions for HCPs.
		Take Control	A self-management and personal development programme for CYP with RMDs aged 10-18 years, including: i) joint creativity, an arts-based project; ii) one-day personal development workshops on sleep and pain management; iii) activity weekend; and iv) 'Kids Have a Say' forum which meets three times a year.

VCSE	Coverage	Project/programme name	Project/programme description*
	Wales	Beyond Limits	A programme providing emotional and peer support for CYP with RMDs. Specific activities include information drop-in sessions, workshops (*e.g.*, pain management, transition, sleep, and sex and relationships), family days, activity weekends, and friendship groups.
		Taking the Next Step	A twelve month pilot programme (commenced February 2019) to support CYP with RMDs aged 16-25 years to build employability skills and support them into further and higher education.
What? Why? Children in Hospital	UK	Video library and colouring pages	A range of over 50 videos featuring CYP and families undergoing a range of medical procedures, as well as videos to support anxiety management, and colouring pages.

*All organisations also produce informational literature which are not detailed unless they are the primary objective of the project/programme. CCAA: Children's Chronic Arthritis Association; COVID-19: Coronavirus disease 2019; CYP: Children and young people; GMT: Greenwich Mean Time; HCP: Healthcare professional; iCAN: Irish Children's Arthritis Network; JIA; Juvenile idiopathic arthritis; NRAS: National Rheumatoid Arthritis Society; RMD: Rheumatic and musculoskeletal disease; SDM: Shared decision making; SNAC: Scottish Network for Arthritis in Children; UK: United Kingdom; VCSE: Voluntary, community, and social enterprise.

Appendix 2. Format and elements of VCSE services

Formats	Elements	CCAA	iCAN	JAR	KLU	NRAS	OV	SNAC	VA	JAPF	AP	TT	WWCIH
INDIVIDUAL													
Educational sessions (with or without parents), including written and visual information	Informational books, comics, leaflets, and videos	X	X	X	X	X	X	X	X		X	X	X
	Daily diaries or notebooks (with or without incentivisation)							X					
	Certificates/kits for positive reinforcement	X		X									
	Check-in or follow-up telephone calls by an interventionist								X				
Family sessions	Written materials								X				
Telemedicine system (*e.g.*, through smartphone devices, text messaging, website, or web-based systems, including conversational agents)	Monitoring through self-report diaries					X			X				
	Trends in condition-specific outcomes					X			X				
	Individualised feedback					X			X				
	Reminders or cueing					X			X				
	Social media communication or online discussion board	X	X	X	X	X	X	X	X	X	X	X	X
	Gamification (feedback/rewards), role-playing, or knowledge quizzes												
	Goal setting or action plans												
	Information messages, animated lessons, or tips								X				
	Skills training												
	Modules with homework												
	Helpline								X				
	Possibility to contact healthcare professionals												
Peer support	Mentorship								X				
	Volunteering opportunities	X	X	X	X	X	X	X	X		X		
IHP	Age- and developmentally appropriate template/plan												
	Goal setting												
	Signposting to IHP	X				X			X				

Formats	Elements	CCAA	iCAN	JAR	KLU	NRAS	OV	SNAC	VA	JAPF	AP	TT	WWCIH
GROUP													
Art therapy sessions	Discussion of scheduled topics											X	
	Art making					X			X			X	
	Discussing art and related feelings								X			X	
Residential/excursion programmes	Outdoor activities	X	X		X	X		X	X				
	Condition-specific activities	X	X		X	X		X	X				
Skills training/workshops	Symptom management, quality of life, and coping strategies								X				
	Transition		X						X				
	Education and employment								X				
	Goal setting, practice, and assessment								X				
Educational and/or support sessions	Didactic presentations	X	X		X	X		X	X				
	Question and answer sessions	X	X		X	X		X	X				
	Discussions and problem solving	X	X		X	X		X	X				
	Quizzes and games	X	X		X	X		X	X				
	Self-management plans				X				X				
	Peer education	X	X			X		X	X		X		
	Sharing experiences	X	X		X	X		X	X	X	X		
	Social activities		X		X			X	X				
School programme	Written information for school			X		X		X	X				
	Didactic presentation about JIA			X				X					
	Peer education												

AP: Arthur's Place; CCAA: Children's Chronic Arthritis Association; iCAN: Irish Children's Arthritis Network; IHP: Individual healthcare plan; JAPF: Juvenile Arthritis Parents and Families UK; JAR: Juvenile Arthritis Research; JIA: Juvenile idiopathic arthritis; KLU: Kids Like Us; NRAS: National Rheumatoid Arthritis Society; OV: Olivia's Vision; SNAC: Scottish Network for Arthritis in Children; TT: Teapot Trust; VA: Versus Arthritis; WWCiH: What? Why? Children in Hospital.

Appendix 3. Integrative review search strategy

1 arthritis/
2 Musculoskeletal Diseases/
3 (juvenile idiopathic arthritis or juvenile arthritis or JIA or JA).tw.
4 exp Self Care/
5 (self adj (care or help or manag* or led or directed or monitor*)).tw.
6 Self Efficacy/
7 Patient Participation/
8 Self-Help Groups/
9 health communication/
10 reminder systems/
11 social networking/
12 Patient-Centered Care/
13 patient care planning/
14 Patient Education as Topic/
15 models, educational/
16 models, psychological/
17 exp consumer health information/
18 access to information/
19 information dissemination/
20 health education/
21 health promotion/
22 Health Knowledge, Attitudes, Practice/
23 patient medication knowledge/
24 ((social or psychosocial or practical or group*) adj3 (information or advice or help or support or network)).tw.
25 ((education* or information* or communicat* or material* or resource*) adj5 (book* or leaflet* or pack* or video* or tape* or phone* or telephone* or manual* or advice* or audiovisual or audio visual or internet or social media or online or app*)).tw.
26 ((education* or information*) adj3 (program* or intervention* or material* or resource* or provision or provid* or session* or consultation* or class or classes or discussion* or meeting*)).tw.
27 (shared manag* or shared-manag*).tw.
28 ((personal or individual) adj development).tw.
29 Schools/
30 child/
31 infant/
32 (young people or young person or teenage* or adolescen*).tw.
33 parents/
34 siblings/
35 or/1-3
36 or/4-28
37 or/29-34
38 35 and 36 and 37
39 limit 38 to (english language and humans and yr="2010 -Current")
40 remove duplicates from 39

Appendix 4. Integrative review screening tool

Criteria	Stage one: Title/abstract screening	Stage two: Full text screening
Population – Participants	CYP (0-24 years), parent(s), family members.	
Population – Condition	Rheumatic and musculoskeletal diseases* and/or multimorbidities of these conditions or targeted generally at all long-term conditions.	
Intervention – Self-management	Any form of programme, project or initiative applied at the individual or group level that aims to improve the way individuals self-manage their condition through the provision of information, education, and/or support.	Stage one, plus reference or inference to one or more of the following concepts aiding self-management: problem solving, decision-making, healthcare utilization, patient-HCP relationships, taking action or control, goal setting, and confidence building mechanisms.
Outcome	Any relevant clinical, humanistic and economical outcomes (medical, role and emotional management), including disease-specific and general outcomes such as quality of life, health service use, psychological outcomes, behavioural outcomes, social functioning, adherence to treatment and costs. Outcomes can either by clinician-reported, patient-reported, caregiver-reported, proxy-reported or physiological.	
Study design	Any type of qualitative, quantitative, or mixed methods study.	

CYP: Children and young people; HCP: Healthcare professional.

Appendix 5. Quality appraisal of articles

First author (year)	Qualitative study designs criteria*					
	Is the qualitative approach appropriate to answer the research question?	**Are the qualitative data collection methods adequate to address the research question?**	**Are the findings adequately derived from the data?**	**Is the interpretation of results sufficiently substantiated by data?**	**Is there coherence between qualitative data sources, collection, analysis, and interpretation?**	**Overall score**
Ahola Kohut (2017)	1	1	1	1	1	100%
Armbrust (2015)	0	0	1	1	0	40%
Burbage (2015)	1	0	1	1	1	80%
Hanghøj (2018)	1	1	1	1	1	100%
Stinson (2010a)	1	1	1	1	1	100%
Toupin-April (2020)	1	1	1	1	1	100%
	Quantitative randomised controlled study designs criteria*					
	Is randomisation appropriate performed?	**Are the groups comparable at baseline?**	**Are there complete outcome data?**	**Are outcome assessors blinded to the intervention provided?**	**Did the participants adhere to the assigned intervention?**	**Overall score**
Arman (2019)	1	1	1	0	1	80%
Armbrust (2017)	1	1	1	1	1	100%
Connelly (2019)	1	1	1	0	1	80%
El Miedany (2019)	1	1	1	1	1	100%
Lalloo (2021)	1	1	1	1	1	100%
Ramelet (2017)	1	1	1	0	1	80%
Stinson (2010b)	1	1	1	0	1	80%
Stinson (2020)	1	1	1	0	0	60%
	Quantitative non-randomised study designs criteria*					
	Are the participants representative of the target population?	**Are measurements appropriate regarding both the outcome and intervention (or exposure)?**	**Are there complete outcome data?**	**Are the confounders accounted for in the design and analysis?**	**During the study period, is the intervention administered (or exposure occurred) as intended?**	**Overall score**
Békési (2011)	1	1	0	0	1	60%
Haverman (2013)	1	1	1	0	1	80%
Lavigne (1992)	0	1	1	0	1	60%
Lomholt (2015)	1	1	1	0	1	80%
McDonagh (2006)	1	1	1	0	1	80%
Mendelson (2017)	0	0	1	0	1	40%

First author (year)	Qualitative study designs criteria*					
	Is the qualitative approach appropriate to answer the research question?	**Are the qualitative data collection methods adequate to address the research question?**	**Are the findings adequately derived from the data?**	**Is the interpretation of results sufficiently substantiated by data?**	**Is there coherence between qualitative data sources, collection, analysis, and interpretation?**	**Overall score**
	Quantitative descriptive study designs criteria*					
	Is the sampling strategy relevant to address the research question?	**Is the sample representative of the target population?**	**Are the measurements appropriate?**	**Is the risk of non-response bias low?**	**Is the statistical analysis appropriate to answer the research question?**	**Overall score**
Brinkman (2017)	1	1	1	0	0	60%
Svavarsadottir (2020)	1	1	1	0	1	80%
White (2012)	1	1	1	0	1	80%
	Mixed methods study designs criteria*					
	Is there an adequate rationale for using a mixed methods design to address the research question?	**Are the different components of the study effectively integrated to answer the research question?**	**Are the outputs of the integration of qualitative and quantitative components adequately interpreted?**	**Are divergencies and inconsistencies between quantitative and qualitative results adequately addressed?**	**Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?**	**Overall score**
Ammerlaan (2017)	1	1	1	1	1	100%
Hilderson (2013)	1	1	1	1	1	100%
Hilderson (2016)	1	1	1	1	1	100%
Stinson (2016)	1	1	1	1	0	80%

*Quality appraisal undertaken using Mixed Methods Appraisal Tool. 1=Yes, 0=No/unable to tell.

Appendix 6. Quality appraisal of review articles

Criteria*	First author (year)	
	Lindsay (2014)	Sheng (2019)
Did the review address a clearly focused question?	1	1
Did the authors look for the right type of papers?	1	1
Are all the important, relevant studies included?	1	0
Did the review's authors do enough to assess quality of the included studies?	0	1
If the results of the review have been combined, was it reasonable to do so?	0	1
What are the overall results of the review? *Adapted to ask if they are clear and expressed appropriately*	1	1
How precise are the results? *Adapted to ask if confidence intervals were given*	0	1
Can the results be applied to the local population?	1	1
Were all important outcomes considered?	1	1
Are the benefits worth the harms and costs?	1	1
Overall score	70%	90%

*Quality appraisal undertaken using the CASP Systematic Review Checklist. 1=Yes, 0=No/unable to tell.

Appendix 7. Results of included studies

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
Ahola Kohut (2017)^ Canada JIA Chronic pain	To explore the perceived benefits and challenges of acting as a young adult peer mentor to CYP. Qualitative: Descriptive study using semi-structured individual interviews and a focus group	• Ten young adult peer mentors (90% female), six had JIA (83.3% female) • Mean (SD) age 19.8 (1.1), range 17-21 years	NA	NA	Inductive qualitative content analysis
Ammerlaan (2017) Netherlands JIA	To investigate the effectiveness of an internet-based SMI guided by peer trainers. Mixed methods: RCT with 6-month follow-up period, and qualitative analysis of chat scripts	• 72 CYP randomised, 67 at analysis (88% female) • Mean (SD) age 19.1 (2.7), range 16-25 years	• Self-efficacy • Self-management related outcomes • QoL; physical functioning • QoL; pain, fatigue, general wellbeing, and disease activity	• Dutch Arthritis Self-efficacy Scale • Dutch Health Education Impact Questionnaire • Dutch Consensus Health Assessment Questionnaire Disability Index • NRS	• Linear mixed models • Thematic analysis
Arman (2019) Turkey JIA	To compare the effects of two different task-oriented activity training programmes on activity performance and participation. Quantitative: RCT	• 62 CYP randomised, 50 at analysis (84% female) • Mean (SD) age 13.6 (3.4) [intervention] and *12.4 (3.0) [control]* range 6-18 years	• Functional ability • Hand ability in performing household tasks • Perceived occupational performance • Pain • Muscular strength • Grip strength • Pinch strengths	• CHAQ • Duruoz Hand Index • Canadian Occupational Performance Measure • NRS • MicroFET 2 hand-held dynamometer • Standard adjustable handle Jamar Plus+Hand dynamometer • Hydraulic pinch gauge	• Independent *t* test • Paired *t* test
Armbrust (2015)† Netherlands JIA	To assess the acceptance of Rheumates@Work in terms of commitment, level of interaction, technical aspects, costs, effort, satisfaction, educational content and perceived benefit; and to describe the theoretical background and design.	• 64 CYP (64% female) • Mean (SD) age 10.0 (1.4) years	• Commitment • Technical aspects • Level of interaction • Level of satisfaction	• Number of participants who had completed weekly assignments on Monday • Emails detailing technical problems • Count of emails sent by patients to initiate contact	NR

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
	Qualitative: Descriptive acceptability study			with supervisor, and by registering participation in chat sessions • Three/four point scales, plus suggested improvements	
Armbrust (2017)† Netherlands JIA	To determine the effects of Rheumates@Work, an internet-based programme supplemented with group sessions, on physical activity, exercise capacity, HRQoL, and participation. Quantitative: RCT	• 49 CYP randomised (67.4% female) • Median (IQR) age 9.7 (8.7-11.3) [intervention], *10.2 (9.0-10.8) [control]* years	• Physical activity • Exercise capacity • HRQoL • Disease activity • Functional ability • Pain • Wellbeing • School absenteeism • Physical education class participation	• 7-day activity diary and accelerometer • Bruce Treadmill protocol • PedsQL • Physician-reported VAS • Dutch CHAQ • VAS • Attendance record	• Wilcoxon's test (baseline comparison) • Mann-Whitney *U* test (between-arm comparison from baseline) • Friedman's test (longitudinal intervention effect) • Linear mixed models
Békési (2011) Hungary JIA Cancer T1DM	To evaluate the effectiveness of a therapeutic recreation summer camp programme on HRQoL changes in CYP with LTCs (including JIA). Quantitative: Quasi-experimental	• 28 CYP with JIA (75% female) • Mean (SD) age 13.4 (2.2) years	• HRQoL • Revised Illness Perception Questionnaire	Hungarian version of KIDSCREEN-52	• Repeated measures of multivariate analysis of variance • Morris and DeShon's equation 8 • Reliable Change Index
Brinkman (2017) USA JIA	To develop and reliably implement a decision aid for treatment of CYP with JIA. Quantitative: Descriptive study using questionnaires	171 parents	• SDM • Decisional conflict	• CollaboRATE • 4-item SURE	Chi-square tests

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
Burbage (2015) USA JIA	To evaluate a weekend family retreat designed to educate and provide support for families of CYP with JIA, as well as to assess parent perceptions. Qualitative: Survey	• 31 parents (58% female) • 22 CYP attended the retreat, age range 7-18 years • 15 siblings attended the retreat	NA	NA	Open coding approach using memos
Connelly (2019) USA JIA	To determine the efficacy in improving pain and HRQoL of an internet SMI for CYP with JIA. Quantitative: RCT	• 289 CYP randomised • 144 CYP in intervention group (68% female) • Mean (SD) age 14.6 (1.8) years • *145 CYP in control group (77% female)* • *Mean (SD) age 14.5 (1.7) years*	• Pain intensity and interference • HRQoL • Self-efficacy • Pain coping • Emotional adjustment • Disease knowledge • Adherence	• NRS • PedsQL 3.0 Rheumatology Module, self-report version • Children's Arthritis Self-Efficacy scale • Pain Coping Questionnaire • PROMIS Paediatric Anxiety and Depression Short Forms • Medical Issues, Exercise, Pain, and Social Support Questionnaire • Analytical tracking of logins and pages accessed, as well as health coach logs	Conditional multilevel models
El Miedany (2019) Egypt JIA	To develop and evaluate an illustrated, stand-alone, interactive evidence-based SDM aid for CYP with JIA. Quantitative: RCT	• 220 CYP randomised • 94 at analysis [intervention] (56% female) • Mean (SD) age 12.7 (1.3), 6.1 – 15.25 years • *95 at analysis [control] (57% female)* • *Mean (SD) age 12.8 (1.5), 6.25 – 15.5 years*	• Disease activity • Functional disability • QoL • School absenteeism • Patient self-reported joint tenderness • Patient global assessment • Patient motivation • Patient's perceived involvement in SDM • Adherence to therapy (compliance/ persistence) • Comprehensibility and usefulness	• Juvenile Arthritis Disease Activity Score-27 • CHAQ • Undisclosed c-PROM • Self-reported days absent, in relation to total school days • 9-item shared-decision making Questionnaire • Medication possession ratio / time from treatment initiation to discontinuation with no medication refill gap	• Mann-Whitney *U* test and Fisher's exact test

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
				for a period of 30+ days during period of interest • VAS	
Hanghøj (2018) Denmark JIA	To investigate the feasibility of a transition intervention aimed at CYP with JIA, focused on declines, drop-outs, no-shows and participation outcomes. Qualitative: Descriptive study using focus groups to investigate participation in a feasibility RCT	• 14 CYP in the focus group • 116 CYP randomised in the original RCT • Range 12-20 years	NA *For feasibility RCT:* • *Adherence to medication* • *Patient experience* • *QoL*	NA *For feasibility RCT:* • *Morisky Medication Adherence Scale-8* • *Mind the Gap* • *PedsQL 4.0* • *JAMAR*	Thematic analysis
Haverman (2013) Netherlands JIA	To investigate the effectiveness of electronic PROMs in clinical paediatric rheumatology care. Quantitative: Quasi-experimental	• 176 CYP (66.8% female) • Mean (SD) age 11.6 (4.5), 10.8 (4.5) [intervention], 13.0 (4.2) [control] years	• Patient-reported HRQoL • Disease-specific outcomes • Functional ability • Pain and overall wellbeing • Communication about HRQoL • Referral to psychologist • Satisfaction • Evaluation of the ePROfile	• Preschool Children Quality of Life • PedsQL Generic Core Scale • Self-composed questionnaire based on DISABKIDS arthritis module • CHAQ • VAS • Yes/no • Dichotomy scale • Adapted Patient Satisfaction Questionnaire • Ad-hoc designed 5-point/ 3-point scale questionnaire	• Mann-Whitney *U* test • Independent *t* test • χ^2 test
Hilderson (2013)‡ Belgium JIA	To describe: the content of a brief transition programme for CYP with JIA; the rationale and design of a mixed-methods study evaluating the clinical impact of the programme; and baseline data of the intervention group. Mixed methods:	• 33 CYP (75.8% female) • Median (IQR) age 16 (15.2-17.1)	• Perceived health status (HRQoL) • Medication adherence • Illness-related knowledge • Global quality of life • Fatigue • Functional status • *Parenting dimensions*	• PedsQL 4.0 Generic Core Scale • PedsQL 3.0 Rheumatology Module • VAS • SWISS HIV Cohort Study Adherence Questionnaire • Modified Patient Knowledge Questionnaire • Linear Analogue Scale	• Wilcoxon test • Qualitative content analysis

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
	Sequential explanatory (quasi-experimental study followed by qualitative study)		• Promotion of independence • Support of autonomy • Behavioural control • Psychological control • Absence of disease activity	• Multidimensional Fatigue Inventory-20 • CHAQ-DI • Promotional Independence Scale • Autonomy Support Scale • Parental Regulation Scale revised to parent self-report • Psychological Control Scale revised to parent self-report • Clinical remission on medication or clinical remission off medication	
Hilderson (2016)‡ Belgium JIA	To investigate the clinical impact of a brief transition programme for CYP with JIA. Mixed methods: Sequential explanatory (quasi-experimental study followed by qualitative study)	• 23 CYP [intervention group comparative analysis] (78.3% female) • Median (IQR) age 17.7 (16.6-18.7) *years* • *23 CYP [control group comparative analysis] (73.9% female)* • *Median (IQR) age 18.9 (17.5; 20.2) years* • 27 CYP in longitudinal analysis (74.1% female) • Median (IQR) age at T0 16.1 (15.2-17.3), at T2 17.5 (16.4- 18.7) years	• Perceived health status • Medication adherence • Illness-related knowledge • Global quality of life • Fatigue • Functional status • *Parenting dimensions* • Promotion of independence • Support of autonomy • Behavioural control • Psychological control • Absence of disease activity	• PedsQL 4.0 Generic Core Scale • PedsQL 3.0 Rheumatology Module • SWISS HIV Cohort Study Adherence Questionnaire • Modified Patient Knowledge Questionnaire • Linear Analogue Scale • Multidimensional Fatigue Inventory-20 • CHAQ • Promotional Independence Scale • Autonomy Support Scale • Parental Regulation Scale revised to parent self-report • Psychological Control Scale revised to parent self-report • Clinical remission on medication or clinical remission off medication	• Wilcoxon test • Qualitative content analysis

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
Lalloo (2021) Canada JIA	To evaluate the feasibility and preliminary effectiveness of iCanCope with Pain, a smartphone-based pain self-management programme, in CYP with JIA. Quantitative: RCT	• 60 CYP randomised (78.3% female) • Mean (SD) age 15.0 (1.7), 14.9 (1.7) [intervention], 15.1 (1.6) [control] years	• Pain intensity • Pain-related activity limitations • HRQoL • Participant accrual and attrition rates • Successful app deployment • Acceptability • Adherence • App engagement • Disease activity	• 11-point NRS • Child Activity Limitations Interview-21 • PedsQL 3.0 Arthritis Module • Acceptability e-Scale • Analytics Platform for Evaluating Effective Engagement • Physician Global Assessment	Linear mixed models
Mendelson (2017) Israel JIA	To examine whether a comic book would improve disease-related knowledge and treatment adherence among CYP with JIA. Quantitative: Prospective cohort study	• 61 CYP (67% female) • Mean (SD) age 14 (3.3), range 8-18 years	• Disease knowledge • HRQoL • Treatment adherence (medication use, physiotherapy appointment attendance, rheumatology appointment attendance)	• Bespoke 20-item multiple choice questionnaire • Childhood Health Questionnaire Parent Form 50 • NRS	• χ^2 test • Paired *t* test • Wilcoxon test • *t* test • Mann-Whitney *U* test • Pearson's r or Spearman's ρ
Ramelet (2017) Switzerland JIA Behçets Chronic osteomyelitis JDM JSLE Autoinflammatory disease NOMID Crohn's disease	To evaluate the impact of a nurse-led telephone intervention on satisfaction and health outcomes of CYP with inflammatory RMDs and their parents. Quantitative: Randomised crossover trial	• 55 CYP and parents randomised • 24 CYP (58.3% female) • Mean age 13.1 years • 31 parents (96.8% female)	• Satisfaction • Clinical health status (morning stiffness, pain, functional capacity, disease status, HRQoL, extraarticular symptoms)	• Client Satisfaction Questionnaire-8 • JAMAR French version	• Random intercept mixed effect linear models (for continuous outcomes) • Random intercept logistic mixed models (for binary outcomes)

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
Undetermined diagnosis					
Stinson (2010a)[#] Canada JIA	To explore the usability of the TTC internet-based SMI with CYP and their parents to refine the prototype. Qualitative: Usability study using semi-structured interviews and observation	• 19 CYP (73.7% female) • 11 English-speaking, 8 French-speaking • Mean (SD) age 15.7 (1.5) years	NA	NA	Simple content analysis
Stinson (2010b)[#] Canada JIA	To determine the feasibility of the 12-week TTC internet-based SMI with telephone support in reducing symptoms and improving HRQoL. Quantitative: Nonblind pilot RCT	• 46 CYP (67.4% female) • Mean (SD) age 14.6 (1.5), 14.4 (1.3) [intervention], 14.9 (1.7) [control], range 12-18 years	• HRQoL • Pain • Stress • JIA-specific knowledge • Self-efficacy • Adherence to treatment	• Juvenile Arthritis Quality of Life Questionnaire • Recalled Pain Inventory • Perceived severity of stress questionnaire • 24-item Medical Issues, Exercise, Pain, and Social Support Questionnaire • Children's Arthritis Self-Efficacy scale • JIA-specific Child Adherence Report Questionnaire / Parent Adherence Report Questionnaire	• *t* test • χ^2 test • Linear mixed models using an analysis of covariance approach
Stinson (2016)^ Canada JIA	To examine the feasibility and acceptability of an online peer mentoring program. Mixed methods: Waitlist pilot RCT and explanatory qualitative study using semi-structured interviews	• 39 CYP randomised • 30 CYP at analysis (96.7% female) • Mean (SD) age 14.3 (1.7), 14.1 (1.5) [intervention], 14.4 (2.0) [control], range 12-17 years	• Engagement and satisfaction • Recruitment • Withdrawal • Adherence with programme • Proportion of completed questionnaires • Self-management • Pain	• Measured through semi-structured interview • Rate • Defined as 100% when participate completed ten calls over eight weeks • Defined as 10)% when all measures completed • Medical Issues, Exercise, Pain and Social Support	• Qualitative content analysis • Marginal linear models

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
			• Perceived social support • Self-efficacy • HRQoL	Questionnaire, including social support subscale • Recalled Pain Inventory • Children's Arthritis Self-Efficacy • PedsQL Arthritis Module	
Stinson (2020)[#] Canada JIA	To evaluate the effectiveness of the TTC internet-based SMI with telephone support in reducing symptoms and improving HRQoL compared with education alone. Quantitative: RCT	• 333 CYP randomised • 219 CYP at analysis (70.3% female) • Mean (SD) 14.4 (1.6), 14 (1.5) [intervention], 14.5 (1.7) [control], range 12-18 years • 197 parents included in analysis	• Pain • HRQoL • Anxiety • Depression • Adherence to treatment • Pain coping • JIA knowledge • Self-efficacy	• Recalled Pain Inventory—Short Form • PedsQL Rheumatology Modules • PROMIS Pediatric Anxiety Short-Form • PROMIS Depressive Symptoms Short Form • Child Adherence Report Questionnaire and Parent Adherence Report Questionnaire • Pain Coping Questionnaire • Medical Issues, Exercise, Pain and Social Support Questionnaire • Children's Arthritis Self-Efficacy; Parent's Arthritis Self-Efficacy	Linear mixed models
Svavarsadottir (2020) Iceland JIA Epilepsy T1DM Sleeping disorder with ADHD	To evaluate the benefits of two sessions of a Family Strengths Oriented Therapeutic Conversation to mothers of newly-diagnosed CYP with LTCs. Quantitative: Quasi-experimental study with one group pre- and post-comparison	31 parents (9 of CYP with JIA; 100% female)	• Perceptions of family support offered by HCPs • Illness beliefs of family members • Impact of paediatric LTCs on family's health • Satisfaction with healthcare service	• Iceland Family Perceived Support Questionnaire • Iceland Family Illness Beliefs Questionnaire • PedsQL – Family Impact Module • PedsQL – Health Care Satisfaction Generic Module	• Paired *t* test • Independent *t* test to compare families with different LTCs

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
Toupin-April (2020) Canada, USA JIA	To develop and assess the acceptability of a paper-based prototype of a web-based patient decision aid. Qualitative: Acceptability study using in-depth interviews	• Twelve CYP (83.3% female) • Median 12.5, range 8-17 years • 12 parents • 11 HCPs	NA	NA	Simple descriptive content analysis
White (2012)[#] Canada JIA	To examine if a therapeutic alliance is achievable with an adolescent internet-based SMI. Quantitative: Descriptive study using validated questionnaires	• Fourteen CYP (71.4% female) • Mean (SD) 14.57 (1.23), range 12-18 years	• Therapeutic alliance • Meaning of distance treatment experience	• Working Alliance Inventory Client Scale • Distance Experience Questionnaire	• Welch's *t* tests • Content analysis
Lindsay (2014)	*Systematic review aim:* To understand how SMIs for CYP with physical disabilities influence health behaviours, and to identify the common components of effective SMIs for CYP with physical disabilities.				
Lavigne (1992) USA JIA[$]	To explore the utility of a psychological treatment procedure for CYP with high levels of pain associated with JIA. Quantitative: Quasi-experimental	• 8 CYP (87.5% female) • Age range 9-17 years	• Pain (via 28-day pain diary) • Pain-related behaviours • Clinical state of disease (physician-reported) • Overall pain experienced during physical therapy (physiotherapist-reported) • Behaviour problems	• VAS • Paediatric Pain Behaviour Questionnaire • Laboratory markers and clinical examination • Child Behaviour Checklist	Mann-Whitney *U* test
McDonagh (2007) UK JIA	To determine whether the QoL of CYP with JIA can be improved by a co-ordinated, evidenced-based programme of transitional care. Quantitative: Quasi-experimental	• 308 CYP (60% female) • Median age 14.2, range 11-17 years • 303 parents (83.2% female)	• HRQoL • Arthritis-related knowledge • Satisfaction with rheumatology care (CYP/parent) • Independent health behaviours (self-medication and independent consultations) • Pre-vocational experience (household chores, work	• Juvenile Arthritis Quality of Life Questionnaire • 16-item disease-specific multidimensional measure with multiple-choice response format • 22-/27-item measures using 7-point Likert scales • Closed questions • Closed and open questions • CHAQ	• χ^2 test • Mann-Whitney *U* / Kruskal-Wallis / Jonckheere-Terpstra tests • Spearman's ρ • Wilcoxon signed-rank test • McNemar's test

First author (year), country, condition	Study aim and design	Participants	Outcome variables	Outcome measures*	Analysis
			experience, career advice, career aspirations) • Health/functional status		• Stepwise linear regression analyses
Rapoff (2002) USA JIA[$]	To evaluate a clinic-based, nurse-administered educational and behavioural intervention to prevent anticipated drop in adherence of NSAIDs among newly diagnosed CYP with JIA. Quantitative: RCT	• 34 CYP (68% female) • Mean (SD) age 8.44 (3.96), range 2-16 years	• Adherence • Disease activity • Functional status	• Medical Event Monitoring Event System • Number of active joints • Number of minutes of morning stiffness • Global Disease Activity rating • CHAQ	• Mann-Whitney *U* test • Wilcoxon tests
Sheng (2019)	*Systematic review aim:* To determine the effectiveness of caregiver-involved interventions on the QoL of CYP and/or caregivers, and to investigate the factors associated with improved QoL.				
Lomholt (2015) Denmark JIA	To evaluate the feasibility of and preliminary efficacy of a cognitive behavioural therapy group intervention for CYP with JIA and their parents. Quantitative: RCT	• 19 CYP • [Intervention] 9 CYP (89% female) • Mean (SD) age 11.4 (2.0) years • *[Control] 10 CYP (70% female)* • *Mean (SD) age 12.0 (1.4) years*	• Pain intensity • Functional disability • HRQoL • Pain catastrophising • Pain-specific beliefs • Self-efficacy • Disease activity • Credibility of the intervention • Satisfaction with the intervention	• Revised Faces Pain Scale • Functional Disability Inventory • PedsQL 4.0 Generic Core Scale • Internalising/ catastrophising subscale of the Pain Coping Questionnaire • Revised version of the Survey of Pain Attitudes. children's version • Children's Arthritis Self-Efficacy Scale • Composite Arthritis Activity Score • Bespoke 5-item questionnaire using 5-point Likert scale • Modified version of the Experience of Service Questionnaire	• χ^2 test • Mann-Whitney *U* test • Analysis of covariance

*Measures used to collect participant characteristics not included. [$]The studies described the condition as juvenile rheumatoid arthritis (JRA), a legacy term for JIA. [^]Studies describing iPeer2Peer. [†]Studies describing Rheumates@Work. [‡]Studies describing the brief transition programme. [#]Studies describing the Canadian TTC. ADHD: Attention deficit hyperactivity disorder; CHAQ: Childhood Health Assessment Questionnaire; CYP: Children and young people; DI: Disability index; HRQoL: Health-related quality of life; IQR: Interquartile range; JAMAR: Juvenile Arthritis Multidimensional Assessment Report; JDM: Juvenile dermatomyositis; JIA: Juvenile idiopathic arthritis; JSLE: Juvenile-onset systemic lupus erythematosus; LTC: Long-term condition; NA: Not available; NOMID: Neonatal onset multisystem inflammatory disease; NR: Not reported; NRS: Numeric rating scale; NSAID: Non-steroidal anti-inflammatory drug; PedsQL: Pediatric Quality of Life Inventory; PROM: Patient-reported outcome measure; PROMIS: Patient-Reported Outcomes Measurement Information System; QoL: Quality of life; RCT: Randomised controlled trial; RMD: Rheumatic and musculoskeletal disease; SD: Standard deviation; SDM: Shared decision making; SMI: Self-management intervention; T1DM: Type 1 diabetes mellitus; TTC: Teens Taking Charge; UK: United Kingdom; USA: United States of America; VAS: Visual analogue scale.

Appendix 8. Intervention characteristics and key findings

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
Ahola Kohut (2017)^	Peer support within a health care context conceptual framework	*Name:* iPeer2Peer *Type:* Internet peer mentoring programme *Deliverer:* Trained peer mentors, nominated by HCPs, screened, and trained in a 2-day structured programme *Delivery:* Individual *Recipient:* CYP *Format:* Online *Setting:* Remote *Duration:* 2-3 months, 10 sessions	Trained peer mentors matched to CYP to provide peer support and education for effective self-management.	Education Enablement Environmental restructuring Modelling Training	Peer mentors found the experience positive and rewarding, supportive of their own self-management, social connection, and personal growth. They deemed formal training as a necessity and stressed the importance of aligning mentor-mentee characteristics to enable a positive relationship to ensue. Use of video also facilitated connection. There were a few instances where parents insisted on staying in the room with mentees during the call, and mentors found that this interfered with the depth of social connection.
Ammerlaan (2017)	Self-efficacy theory	*Name:* Challenge your arthritis *Type:* Internet self-guided self-management intervention with social support *Deliverer:* Self-guided; Peer trainers with JIA aged 20-30 years *Delivery:* Individual, group *Recipient:* CYP *Format:* Online *Setting:* Remote *Duration:* 6 weeks, ~12 hours	Three components: Chat section; home exercises; discussion board. Once a week, six CYP and two trainers had a planned grouped chat for a maximum of 90 minutes, working through a weekly theme (are you a self-manager; friends, family, and communication; feeling blue; sport and exercises; relations and intimacy; having control over your life and JIA) by setting goals, practicing, asking questions, giving/receiving feedback, playing a game, or watching a real-life story. After the chat, participants worked through the intervention and completed the exercises (~1 hour per week).	Education Enablement Environmental restructuring Modelling Persuasion Training	Although the intervention was regarded to influence an activate coping style by sharing experiences, enhancing social support, increasing autonomy, and increasing goal-setting behaviour, the intervention did not significantly improve self-efficacy, self-management, or QoL.
Arman (2019)	Canadian occupational performance measure role model	*Name:* TOAT *Type:* Video games-based TOAT *Deliverer:* Physiotherapist, pedagogical agent *Delivery:* Individual *Recipient:* CYP	An individualised rehabilitation approach including intensive training, variable practices, and intermittent feedback. There were 2 programmes: training in daily living conditions (using items such as clothes, pencils, exercise bands), and video games-based training (using Kinect™ for Xbox 360®).	Enablement Environmental restructuring Incentivisation Persuasion Training	Video games-based TOAT, developed by physiotherapists, can be effective treatment options, by increasing motivation and decreasing fear avoidance around physical activity, which is an important component of self-management.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Format: Face-to-face using technology *Setting:* Hospital, home *Duration:* 8 weeks, ~1 hour 3 times weekly			
Armbrust (2015)†	Pender's health promotion model	*Name:* Rheumates@Work *Type:* Internet- and group-based cognitive behavioural programme *Deliverer:* Self-guided with pedagogical agent, staff including physiotherapist, student psychologist, paediatric rheumatologist *Delivery:* Individual, group *Recipient:* CYP *Format:* Online, face-to-face *Setting:* Home, hospital *Duration:* 14 weeks, ~1 hour per week	A combined internet-based and in-person instruction model, guided by a pedagogical agent called Buddy, to increase physical activity in 8-13-year-olds. The topics included: health education; emotions and affect; barriers and benefits of being physically active; self-efficacy and perceived effect of physical activity; peer support; smart goals; setbacks; keep it up. Four group sessions contained the following elements: health education related to JIA and physical activity; information on barriers preventing physical activity; explanation of the benefits of physical activity; and self-efficacy towards becoming more physically active. CYP have to set attainable goals based on current physical activity. Information is provided through cartoons, puzzles, and brain teasers.	Education Enablement Environmental restructuring Incentivisation Training	Rheumates@Work is a way to educate and stimulate CYP to become more active, develop self-management skills, and seek peer support. Commitment and satisfaction with the intervention were high, and costs involved were low.
Armbrust (2017)†					Rheumates@Work provided a small improvement in moderate-to-vigorous physical activity levels and increased exercise capacity without exacerbating disease status. Physical activity levels continued to improve for 3 months and lasted for 12 months, though HRQoL was not affected. Participation ins school and physical education classes increased, and those who started the programme in Winter benefitted the most.
Békési (2011)	Therapeutic Recreation	*Name:* Bátor Tábor program *Type:* Therapeutic recreation summer camp programme *Deliverer:* Volunteer staff *Delivery:* Group *Recipient:* CYP *Format:* Face-to-face *Setting:* Camping centre *Duration:* 8 days	Campers attend free of charge. Non-categorical approach is used for commonalities across conditions. The camp aims to: i) provide a fun-filled, age-appropriate experience where they can acquire activity-related skills (*e.g.*, archery, boating, crafts, team games); ii) encourage development of a self-sufficient attitude; iii) enhance self-esteem; iv) provide opportunities for a sense of mastery and efficacy in peer relationships (*e.g.*, campfire, beach party, casino, and talent show); v) provide disease education through formal education and informal peer interactions. Activities offer a challenge,	Education Enablement Environmental restructuring Incentivisation Training	Participation in the therapeutic recreation camping programme has significant positive impact on HRQoL, in particular on enhancing their self-perception and reducing the autonomy of children under 14 years.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
			encouraging campers to work through their perceived limitations.		
Brinkman (2017)	Breakthrough series model; Plan-do-study-act cycles	*Name:* JIA medication choice issue cards *Type:* Decision aid for medication choices *Deliverer:* HCP *Delivery:* Dyadic, triadic *Recipient:* Parents, CYP *Format:* Face-to-face *Setting:* Hospital *Duration:* 18 weeks	Six JIA medication choice cards using plain language and pictorial representations. Each card covered a key issue on how medications differ. These included: dose frequency, effect time, side effects, cost, length of time to be taken, and other things to be considered before starting or while taking each medication. The cards were designed to enable HCPs and CYP/parents discuss medications that are reasonable options in a given clinical context. They were accompanied by a 1-page instruction sheet with pictures depicting the cards intended use, a short training video, and pamphlet containing the content of the cards. This was made available as an interactive electronic portable document format.	Education Enablement	Although user acceptability of the decision aid was high, its reliable implementation into routine clinical care proved challenging. No difference in outcomes were identified with and without the decision aid.
Burbage (2015)	NR	*Name:* NR *Type:* Family retreat weekend *Deliverer:* Volunteer staff *Delivery:* Group *Recipient:* CYP, parents, siblings *Format:* Face-to-face *Setting:* Camp-based *Duration:* Up to 2 weekend days	The family retreat intervention consisted of lectures and discussions to boost knowledge, treatment, symptom management, and family coping strategies; small group activities developing family plans to enhance child and family coping; camp-like activities (*e.g.,* ropes course, hiking, cycling) to build self-esteem and foster group cohesiveness.	Education Environmental restructuring Training	A family retreat weekend benefited CYP with JIA, as well as parents, and siblings. Providing support to CYP and their families was important, as assisting in improvements in family functioning and coping strategies. The group setting aided in families supporting each other.
Connelly (2019)	NR	*Name:* TTC *Type:* Internet self-guided self-management programme *Deliverer:* Self-guided *Delivery:* Individual *Recipient:* CYP, parents *Format:* Online, telephone	Twelve multimedia-based modules (20-30 content pages, taking ~30 minutes per module), comprising psychoeducation, training in cognitive-behavioural coping skills and stress management; and other self-management topics. No more than 2 modules could be completed per week. Brief monthly telephone support calls (<30 minutes) from	Education Enablement Environmental restructuring Modelling Training	The internet self-guided SMI modestly improved pain and HRQoL in CYP with JIA, comparable to accessing disease education online.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Setting: Remote *Duration:* 12 weeks, with 12 months follow-up	health coaches were provided for three months. Two modules were also aimed at parents about facilitating their child's self-management skills.		
El Miedany (2019)	International Patient Decisions Aids framework	*Name:* NR *Type:* SDM aid *Deliverer:* Paediatric Rheumatologist *Delivery:* Individual *Recipient:* CYP *Format:* Face-to-face *Setting:* Hospital *Duration:* ~30 minutes	There were 2 tools, 1 for csDMARDs and 1 for bDMARDs. Five visual aids were utilised: i) emojis to make numerical information easier to understand; ii) illustrated visual aids to figure out treatment targets; iii) virtual risk tools for the possibility of side effects; iv) visual progressometer to provide some room to customise treatment options; and v) therapeutic visual aids to estimate the chances of developing side effects as well as the rate of halting joint damage.	Enablement	The SDM aid offered CYP evidence-based information about the positives and negatives of different treatment options. It improved their disease understanding and ability to make informed decisions, which was reflected by improved adherence and patient-reported outcomes, and reduced absenteeism.
Hanghøj (2018)	NR	*Name:* Transition for Unge i Børnereumatologisk *Type:* Transition programme/ clinic *Deliverer:* HCPs (doctor and nurse), trained in adolescent medicine *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents *Format:* Face-to-face *Setting:* Hospital *Duration:* 3 years	Two annual consultations in the transition clinic were offered, conducted in parallel with and supplemental to usual outpatient clinic appointments. The consultations, held with the same doctor and nurse pair, involved 60 minute afternoon (2-6pm) split appointments, with parents participating only in the second half; address of topics such as school, friends, relationships, sex, alcohol and drugs, as well as any issues CYP wanted to raise; and an opportunity to meet other CYP in groups if desired.	Enablement Environmental restructuring	The most common reasons for declining to participate where unspecified and practical issues, with most dropouts because CYP didn't wish to continue. Reasons for no-shows were forgetting and being too busy. CYP felt that the advantages of a transition intervention clinic were participation without parents, being able to set the agenda, greater trust and confidentiality, and responsiveness from the HCP pair. Disadvantages included a lack poor expectations of what was happening with the clinic, the thought of meeting others with JIA in a peer support setting, too infrequent conversations with HCPs, and transport issues to clinic.
Haverman (2013)	NR	*Name:* ePROfile *Type:* Internet-based application (ePRO) to monitor HRQoL *Deliverer:* Self-guided, paediatric rheumatologist	CYP/parents completed online questionnaires at home via a website before 2 consultations. The answers were then automatically converted to an ePROfile, keyed to colours, which could be used by the paediatric rheumatologist on screen during the	Enablement	The ePROfile increased discussion of psychosocial topics, contributing to an increased satisfaction of the paediatric rheumatologist with the care provided during consultation. ePROs in clinic could aid the systematic monitoring of HRQoL.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Delivery: Individual, dyadic, triadic *Recipient:* CYP, parents, paediatric rheumatologist *Format:* Online, face-to-face *Setting:* Home, hospital *Duration:* 12 months	consultation, to focus on identifying, monitoring and discussing HRQoL problems.		
Hilderson (2013)‡	MRC Framework	*Name:* Devices for the Optimisation of Transfer and Transition of Adolescents with Rheumatic Disorders *Type:* Transition programme *Deliverer:* Transition Co-ordinator *Delivery:* Individual, group *Recipient:* CYP, parents *Format:* Face-to-face *Setting:* Hospital *Duration:* 2 years	A 5-step brief intervention consisting of eight components: i) a transition co-ordinator; ii) providing information and education about JIA and medication management (including adherence), health behaviour, dealing with fatigue, school, and friends; iii) availability by telephone; iv) information about and contact with the adult rheumatology programme; v) guidance for parents; vi) meeting with peers; vii) a transfer plan; and viii) actual transfer to the adult rheumatology programme. The 5 steps are as follows: i) outpatient appointment with transition co-ordinator; ii) outpatient appointment with transition co-ordinator; iii) information day for CYP and parents; iv) individualised transfer plan; and v) actual transfer.	Education Enablement Environmental restructuring	Implementation of a transition programme like this may be more feasible, because it is estimated that one full-time equivalent employee of the entire team could take on a case load of 250-300 transitioning patients. This brief intervention focused on counselling and education, rather than skills training, starting in the pretransfer period.
Hilderson (2016)‡					Implementation of a transition programme (or a transfer programme given the timeframe) like this can improve perceived health (physical, psychosocial and JIA-specific status) and QoL of CYP with JIA during the transition process, as well as parenting behaviours.
Lalloo (2021)	NR	*Name:* iCanCope *Type:* Self-management smartphone application *Deliverer:* Self-guided *Delivery:* Individual *Recipient:* CYP *Format:* Smartphone application *Setting:* Remote *Duration:* 55 days	The application features symptom tracking (including check ins, historical viewing, pain map and inflammation areas), goal setting, library of disease education and pain coping strategies, and peer-based social support. Only symptom tracking was available in one group for evaluation.	Education Enablement	The iCanCope application was deployed with a high success rate and was rated highly acceptance. CYP demonstrated moderate-to-high adherence. A clinically meaningful reduction in pain intensity was observed.
Mendelson (2017)	NR	*Name:* Neta and the Medikidz Explain JIA *Type:* Comic book	A comic book about a young girl with JIA, who struggles to participate in physical activity at school and her friends do not understand.	Education Persuasion	JIA knowledge increased significantly after reading the comic book, which lasted after 1 year. There was no impact on

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Deliverer: HCPs *Delivery:* Individual *Recipient:* CYP *Format:* Comic book *Setting:* Home *Duration:* Follow-up at 1 month and 1 year	Using text and illustrations, the Medikidz help her to understand the facts about JIA in a way that is not delivered in the clinic.		adherence, though initial adherence was reported to be high.
Ramelet (2017)	The Cox's Interaction Model of Client Health Behaviour	*Name:* NR *Type:* Telenursing intervention *Deliverer:* CNS *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents *Format:* Face-to-face, telephone *Setting:* Hospital, home *Duration:* 12 months	The intervention began with a face-to-face medical and nursing consultation to allow the nurse to introduce themselves, outline the intervention, and familiarise themselves with CYP and their family. For the following 12 months, they would receive a monthly telephone call, each following a two-part standardised form of telephone interviewing: the first part including descriptions of the call, action/decision taken, and a brief summary of the conversation and planned action. The second part related to the intervention itself and included 8 questions on: i) everyday life, school and social; ii) treatment; iii) physiotherapy; iv) occupational therapy; v) pain; vi) schedule; vii) administrative issues; and viii) additional topics that CYP and families wanted to discuss. CYP and families were also given a telephone number to contact when needed.	Enablement Environmental restructuring	The telenursing intervention combined affective support, health information, and assistance in decision making. CYP and families were satisfied, and CYP tended to have less morning stiffness and pain. It has the potential to reduce health problems while increasing satisfaction with clinical management, particularly during the newly-diagnosis period.
Stinson (2010a)[#]	Hermeneutical circle	*Name:* TTC: Managing Arthritis Online *Type:* Internet self-guided SMI *Deliverer:* Self-guided *Delivery:* Individual *Recipient:* CYP *Format:* Online *Setting:* NR *Duration:* NR	JIA-specific information; self-management strategies; social support; two parent-focused modules to promote healthy behaviours.	Education Enablement Environmental restructuring Training	Usability testing is an important part of developing complex interventions the one described, to optimise its completeness, understandability, quality, credibility, and relevance for future CYP using the intervention.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
Stinson (2010b)[#]	NR	*Name:* TTC: Managing Arthritis Online *Type:* Internet self-guided SMI with individual telephone/email support *Deliverer:* Self-guided, trained health coach (non-HCP) *Delivery:* Individual *Recipient:* CYP *Format:* Online, telephone *Setting:* Remote *Duration:* 12 weeks	JIA-specific information; self-management strategies; social support with weekly telephone calls; two parent-focused modules to promote healthy behaviours.	Education Enablement Environmental restructuring Modelling Training	An internet-based education SMI with social support is acceptable for CYP and parents, and indicates efficacy in improving knowledge and decreasing pain, although longer-term benefits would require longitudinal evaluation. Contact with the health coach was integral for improving motivation and adherence. This may be a useful component of a stepped-care approach, moving from an online management intervention to more intense (face-to-face) interventions if required.
Stinson (2016)[^]	NR	*Name:* iPeer2Peer *Type:* Internet peer mentoring programme *Deliverer:* Trained peer mentors *Delivery:* Individual *Recipient:* CYP *Format:* Online *Setting:* Remote *Duration:* 8 weeks, 10 sessions (~20-30 minutes); 2 per week for 2 weeks, then weekly for 6 weeks	Trained peer mentors (16-25 years, successfully managing their JIA) matched to CYP with JIA to provide peer support and education for effective self-management.	Education Enablement Environmental restructuring Modelling Training	Peer support is valued by CYP, and iPeer2Peer is a promising SMI which demonstrated significant improvements in self-management skills. However, the intervention requires greater flexibility and individualisation to suit mentee needs, while additional training for mentors would facilitate optimal interactions.
Stinson (2020)[#]	Teens Taking Charge Conceptual Model	*Name:* TTC: Managing Arthritis Online *Type:* Internet self-guided self-management programme with individual telephone/email support *Deliverer:* Self-guided, trained health coach (non-HCP) *Delivery:* Individual *Recipient:* CYP	A 12-module website consisting of JIA-specific information, self-management strategies, and social support; telephone health coaches; 2-module website for parents, consisting of education about the impact of JIA, and strategies to support CYP in self-management.	Education Enablement Environmental restructuring Modelling Training	An internet-based education SMI with social support can improve HRQoL in CYP with JIA, while reducing pain intensity and pain interference.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Format: Online, telephone *Setting:* Remote *Duration:* 12 weeks, with assessment over 12 months			
Svavarsadottir (2020)	Wright and Leahey's Calgary Family Assessment and Intervention Models; Illness Beliefs Model	*Name:* Family Strengths Orientated Therapeutic Conversation *Type:* Therapeutic family nursing conversations *Deliverer:* Advance practice nurses *Delivery:* Individual *Recipient:* Parents (mothers) *Format:* Face-to-face *Setting:* Hospital *Duration:* 2 sessions ~4-10 weeks apart, starting 1-week post-diagnosis	Establishing therapeutic relationship with parent; drawing a family genogram; exploring quality of family relationships; encouraging family illness story narrative; asking therapeutic questions; identifying strengths, resilience, and resources; offering evidence-based information and recommendations; identifying facilitating or helpful illness beliefs or challenging constraining/hindering illness beliefs.	Education Enablement	A brief, therapeutic family nursing conversation can be beneficial and can improve outcomes for CYP and families. Advanced practice nurses in the clinical setting play an important role in supporting CYP and families with a new diagnosis, as well as those undergoing treatment, by assisting families to adjust to a new reality, develop resilience, and subsequently improve family outcomes.
Toupin-April (2020)	International Patient Decision Aid Standards, Ottawa Decision Support Framework	*Name:* JIA Option Map *Type:* Patient decision aid *Deliverer:* NR *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, families *Format:* NR *Setting:* NR *Duration:* NR	JIA-specific information; patient decision aid goal; pain self-assessment; pain management self-report; values clarification exercise; treatment options; decisional prompt.	Education Enablement	A patient decision aid can be a useful tool for CYP and families, to aid SDM about pain management options. Web-based options were favoured over paper, but should be tailored to individual preferences, and closely followed by a discussion with HCPs.
White (2012)[#]	Bordin's Theory	*Name:* TTC: Managing Arthritis Online *Type:* Internet self-guided SMI with individual telephone/email support *Deliverer:* Self-guided, trained health coach (non-HCP psychology graduate)	JIA-specific information; self-management strategies; social support with a weekly telephone call/supplemental emailing.	Education Enablement Environmental restructuring Modelling Training	A therapeutic alliance between CYP and a health coach in the absence of visual cues can be achieved and correlates positively with reduced symptoms. Remote connection may enhance perceived sense of anonymity, meaning that CYP felt comfortable sharing with their coach. Findings were comparable with existing studies.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
		Delivery: Individual *Recipient:* CYP *Format:* Online, telephone *Setting:* Remote *Duration:* 12-21 weeks (average 14.7 weeks)			
Systematic reviews					
Lindsay (2014)	*Key findings*: Most SMIs ran several sessions for at least three months by a trained interventionist or HCP, had one-to-one sessions and meetings, homework activities and parental involvement. Although outcomes varied, all SMIs reported at least one significant improvement in either overall self-management skills or a specific health behaviour.				
Lavigne (1992)	Thermal biofeedback and cognitive behaviour training	*Name:* NR *Type:* Psychological treatment package for pain management *Deliverer:* Paediatric psychologist *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents (usually mothers) *Format:* Face-to-face *Setting:* Hospital *Duration:* 3 months	Six individual sessions scheduled biweekly over a 3-month period; the first session lasted 90 minutes and the remainder 1 hour. Time was spent with CYP and parents, with homework assignments given after each session. Session content was as follows: i), ii) progressive muscle relaxation; iii), iv) electromyogram biofeedback to enhance general relaxation, plus operant pain management techniques with parents, including a functional analysis of CYP's pain behaviour at home; v), vi) thermal biofeedback, with autogenic exercises.	Education Enablement Training	The package modestly reduced self-reported pain among CYP, with stronger evidence from maternal pain diaries. Physiotherapists reported a reduction in pain, but this was not significant.
McDonagh (2007)	NR	*Name:* NR *Type:* Programme of transitional care *Deliverer:* Local programme co-ordinator, local paediatric rheumatologist *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents *Format:* Face-to-face *Setting:* Hospital *Duration:* 12 months	The programme centred on templates for individualised transition plans, designed to reflect adolescence development stages. These were reviewed at each clinic visit and/or every 6 months. Once the template was complete, CYP would move onto the next plan. Other key components included a local programme co-ordinator, age- and developmentally appropriate information resources, and a departmental transition policy.	Enablement Environmental restructuring	The structured programme of transitional care had a positive impact on CYP's HRQoL, knowledge, satisfaction, and vocational readiness, irrespective of age. Continuous improvement was observed for CYP and parents' knowledge, with significantly greater improvement in the younger age groups at 12 months.
Rapoff (2002)	Applied behaviour analytic theory	*Name:* NR	Educational and behavioural strategies for enhancing adherence. CYP and parents	Education Enablement	The intervention improved medication adherence, though improved adherence

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
	(contingency-shaped behaviour)	*Type:* Adherence improvement strategy planning plus telenursing *Deliverer:* Nurse practitioner *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents *Format:* Face-to-face, telephone *Setting:* Hospital, home *Duration:* 12 months	viewed a 10-minute audiovisual programme and received a booklet describing adherence-enhancement strategies (*e.g.* cueing and pairing medication with another routine), monitoring (using a calendar to track medication), positive reinforcement (praise and rewards exchanged for privileges), and discipline (using time-out for defiant refusals). The nurse reviewed these strategies with CYP and parents. The control group received a general educational video. The intervention took place during a 30-minute clinic visit, followed by a nurse phone call every 2 weeks for 2 months, and then monthly for 10 months. For the intervention group, the nurse reviewed and problem-solved about adherence improvement strategies.	Environmental restructuring	had no significant effect on disease activity or functional limitations.
Sheng (2019)	*Key findings*: SMIs mainly involved disease education, skills training, environmental change, psychological intervention, physical exercise, experience sharing, monitoring, or social support. Caregiver-involved interventions significantly improved caregiver HRQoL, particularly face-to-face.				
Lomholt (2015)	CBT principles	*Name:* NR *Type:* CBT programme *Deliverer:* CBT-trained psychologist and psychology research assistants *Delivery:* Individual, dyadic, triadic *Recipient:* CYP, parents *Format:* Face-to-face *Setting:* Local venues *Duration:* 6 weeks	Six weekly sessions lasting 2 hours (the last 2 sessions were biweekly). The intervention was manualised, with CYP and parents receiving a workbook, worksheets, and guides for home practice. The intervention focused on psychoeducation CYP and parents on pain mechanisms, restructuring pain-related negative automatic thinking, and gradually confronting pain-related avoided situations. Each session followed the same structure: i) evaluation of homework; ii) discussion of session theme; iii) separation of CYP and parents; iv) therapist working with CYP / parents given topics to discuss regarding homework; v) CYP have a break / therapist works with parents; vi) CYP and parents re-join together for next homework briefing. Topics discussed per session were as follows: i) Gate-control theory,	Education Enablement Training	The CBT programme was acceptable to those who participated; however, feasibility in general was unclear due to a low participation rate. A reduction in pain was not seen; however, disease severity increased during the study. An increase in QoL, reduction in pain catastrophising, and improvement in adaptive pain cognitions were seen.

First author (year)	Theory, framework	Intervention description	Intervention content	Intervention functions	Key findings
			biopsychosocial model of pain, setting treatment goals; ii) cognitive restructuring; iii) per previous session plus rewards / parenting a child with JIA; iv) distraction exercise, exposure ladder; v) social skills and assertiveness training with role-play; vi) painful situation strategies, and re-evaluation of family's goals.		

[^]Studies describing iPeer2Peer. [†]Studies describing Rheumates@Work. [‡]Studies describing the brief transition programme. [#]Studies describing the Canadian TTC. CBT: Cognitive behavioural therapy; CNS: Clinical nurse specialist; cs/bDMARD: Conventional synthetic/biological/biosimilar disease modifying antirheumatic drug; CYP: Children and young people; ePRO: Electronic patient-reported outcome; HCP: Healthcare professional; HRQoL: Health-related quality of life; JIA: Juvenile idiopathic arthritis; MRC: Medical Research Council; NR: Not reported; QoL: Quality of life; SDM: Shared decision making; SMI: Self-management intervention; TOAT: Task-orientated activity training; TTC: Teens Taking Charge.

Appendix 9. Exemplar participant information sheet

INFORMATION FOR CHARITIES AND PATIENT ORGANISATIONS ABOUT iSMART

iSMART: Innovative approaches to the Self- and shared-Management of ARThritis by children, their families and professionals: A realist approach

This study is funded as part of Simon Stones' PhD within the School of Healthcare at the University of Leeds.

Simon Stones is a PhD student from the University of Leeds. For his PhD, he has designed a study called 'iSMART', which is looking to improve the way children and young people with juvenile idiopathic arthritis (JIA) manage their condition independently and with the support provided by organisations like you, as well as their family members, healthcare professionals and teachers.

It would be great if you wanted to take part in the study! But before you decide, it's important for you to understand what the study is about, and what it will involve. Please read this leaflet carefully and talk to others about it if you wish. You can also ask me anything that is unclear before making a decision.

What is the purpose of the study?

We want to understand what it's like for children and young people living with JIA, and how you as a patient organisation want to be supported, so that you can continue to support children, young people and families in learning to manage JIA, with support from other family members, teachers, and healthcare professionals. As part of the study, we will also be talking with children and young people with JIA, as well as families, teachers, healthcare professionals and other organisations who support children and young people with JIA.

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Why have I been invited?

You are a representative of your organisation, who provides information and/or support services to children and young people with JIA, as well as their families. Given your experience of working with children, young people and families with JIA, we would welcome the opportunity to discuss how to better support children and young people in developing their self-management skills.

Do I have to take part?

No, you do not. It is entirely up to you! If you decide not to take part, it won't affect the way that you are treated. If you do decide to take part, we would like you to sign a consent form (included in this pack). You can still change your mind later though. If you decide that you don't want to take part at any point in the study, please let Simon know - it won't be a problem. Should you change your mind after the interview, you will have two weeks after the interview to withdraw from the study. After this point, it won't be possible to delete your data because it will have been anonymised.

What will happen if I take part?

Simon will contact you to arrange to meet you at a time and date that is convenient for you. Simon will then meet you either at a location agreed in advance, such as the University of Leeds or a local library. Alternatively, you can take part over the telephone or Skype.

Simon will gather your views about what support is needed to enable children and young people with JIA to self-manage their health. The discussion is expected to take between 45 minutes and 1 hour. There may be an opportunity for a second discussion in the future for you to share your comments about our ideas that have been developed during the study.

If it's okay with you, Simon will use an audio recorder during the discussion to facilitate the analysis process. However, you can ask him to switch the recorder off at any time.

What will happen with my information?

Any information we gather from you will be kept private and handled in strict confidence. We will not disclose your contributions to anyone else, unless there was a concern about your safety or wellbeing. In that case, Simon would speak to his supervisors at the University of Leeds, following guidance from the University of Leeds. Any information that could identify you will be removed, and a number or a false name will be used instead of your real name when we write up the findings of the study, so that you won't be able to be identified from what you say. Direct quotes may be used, but they will be anonymised.

The University of Leeds is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. All information will be stored on the University of Leeds computer network or in a locked cabinet accessible only to Simon, or his PhD supervisors (Veronica Swallow and Linda Milnes). Any personal, identifiable features within the audio recordings of discussions will be downloaded and anonymised by Simon before being accessible to anybody else, including agencies (approved by the University of Leeds) who may type up transcripts of the discussion.

The University of Leeds will only keep identifiable information (names and contact details) until the study finishes (no later than 30 September 2020), and scanned consent forms for six years after the end of the study. Your rights to access, change or move your information are limited, as we need to

manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible, which will be your name, contact details and position, e.g. Chairperson. The information you give to us during the discussion will be anonymised, and you will have two weeks after the discussions to request for anything you said to not be used. After this point, it will be impossible to remove your information. Anonymised information, analysed with other participants' information, will be published in Simon's PhD thesis, medical journals, presentations, on websites, and on social media.

The team may carry out further analysis of the information and/or share it with other researchers in future studies through an online University of Leeds repository where it will be available for 10 years. This information will not identify you and will not be combined with other information in a way that could identify you. The information will only be used for the purpose of health and care research and cannot be used to contact you or to affect your care. It will not be used to make decisions about future services available to you, such as insurance.

You can find out more about how we use your information by contacting the University of Leeds Data Protection Officer, David Wardle, by emailing DPO@leeds.ac.uk

What positives may emerge from this study?

What we learn may help us know more about how to better support children and young people with JIA. We may also learn how to better support teachers, families, healthcare professionals and organisations supporting young people with JIA. You may also discover approaches or existing initiatives which could be useful to your organisation.

We can't promise that the study will help you, but many people enjoy taking part in research because they can share their views and experiences! It is unlikely that there will be any risks in taking part in this study, but if you feel upset in any way, we would stop the discussion.

What if there is a problem?

If you are concerned about any aspect of this study, you should speak to Simon who will do his best to answer your questions. You can also speak to one of Simon's supervisors too.

Who has reviewed this study?

Before any study is allowed to happen, it has to be checked by various people, including an independent group of people sitting on a 'Research Ethics Committee', who protect the interests of those taking part in research. The London - Surrey NHS Research Ethics Committee (REC) have reviewed and approved this study. The REC reference is 18/LO/1087.

How can I find out more?

If you would like to speak to someone before deciding to take part or not, please contact Simon or one of Simon's supervisors (Veronica and Linda) who are supporting his PhD study. It is advised that you contact Simon in the first instance, either by telephone or email:

Appendix 10. Exemplar consent and assent form

CONSENT TO TAKE PART

iSMART
Innovative approaches to the Self- and shared-Management of ARThritis by children, their families and professionals

in the iSMART research project,
looking at how to support children with juvenile idiopathic arthritis (JIA) to self-manage their health with the support of their families, healthcare professionals, teachers and charities.

If you agree with the statements below, please add your initials in the box to the right of each statement. Please also tell us if you would like a summary of the findings at the end of the project.

FOR EXAMPLE...	
I understand that if I do not wish to answer any particular question, I am free to decline.	JG
I have read and understand the iSMART information leaflet (iSMART Information for Parents, Carers and Grandparents (Version 1.2, 09 August 2018)) and I have had the opportunity to ask questions about the project.	
I understand that my participation is voluntary and that I am free to withdraw at any time during the discussion, without giving any reason and without there being any negative consequences. Anonymous information collected before this point will be kept.	
I understand that if I do not wish to answer any particular question, I am free to decline.	
I understand that audio recordings will be made as part of this project to help the researcher write up the findings from our discussion. These will remain confidential.	
I give permission for members of the research team to have access to my anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in any of the articles or reports that result from the research.	
I understand that direct quotes may be used and published with the findings from the project, but any personal identifiers would be removed (for example, names, hospitals and location).	
I agree for the anonymised information collected from me to be stored and used in relevant future research, and I agree for anonymous information I provide to be archived in the Research Data Leeds Repository.	
I agree to take part in the above research project and will inform the lead researcher should my contact details change during the project and, if necessary, afterwards.	
I would like to receive a summary of the findings at the end of the project. (PLEASE CIRCLE YES OR NO)	YES NO
I would be happy to be contacted in the future to potentially take part in a second interview. (PLEASE CIRCLE YES OR NO)	YES NO

If you are happy with all of the above points and still want to take part, please write your name, signature and today's date below!

YOUR NAME _____ YOUR SIGNATURE _____ TODAY'S DATE _____

RESEARCHER'S NAME _____ RESEARCHER'S SIGNATURE _____ TODAY'S DATE _____

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PARTICIPANT COPY / RESEARCHER COPY

iSMART Consent Form for Parents, Carers and Grandparents (Version 1.3, 09 August 2018); IRAS Number 234475

MY PERMISSION FORM

iSMART
Innovative approaches to the Self- and shared-Management of ARThritis by children, their families and professionals

for taking part in the iSMART research project,
looking at how children with juvenile idiopathic arthritis (JIA) look after their health with the help of their family, people at the hospital, teachers and other groups.

If you are happy with the sentences below, please tick the circle at the end of each sentence (like this!)

- Have you read (or had read to you) the information about the project (iSMART Information for Children 8 to 12 (Version 1.1, 26 July 2018)?
- Has somebody explained this project to you?
- Do you understand what this project is about?
- Have you asked all of the questions that you have?
- Do you understand that it's okay to stop taking part at any time?
- Are you happy for what you say to be recorded?
- Are you happy for the researcher to use some of the things you say to write stories? Your name will not be used.
- Are you happy to take part in this project?
- Would you be happy to take part in another chat in the future?

If you have **ticked** all of the circles above and do want to take part, please write your name and today's date below!

YOUR NAME _____ TODAY'S DATE _____

RESEARCHER'S NAME/SIGNATURE _____

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iSMART Assent Form for Children 8 to 12 (Version 1.2, 01 August 2018); IRAS Number 234475

PARTICIPANT COPY / RESEARCHER COPY

Appendix 11. Criteria to assess conceptual richness

'Conceptually rich' (Ritzer, 1991)	**'Thicker description' (Roen et al., 2006), but lacking conceptual richness**	**'Thinner description' (Roen et al., 2006) –**
Theoretical concepts are explicit and described in sufficient detail.	The programme theory is described in enough detail for additional information to be 'surfaced'.	Insufficient information can be identified to enable the programme theory to be 'surfaced'.
Relationships between and amongst concepts are clearly described.	The context in which the programme took place is considered.	The context in which the programme took place has limited or no consideration.
Concepts discussed have been sufficiently developed and defined to enable a reader without first-hand experience of SSM.	There is some discussion between programme theory and implementation in practice.	There is limited or no discussion of the difference between programme theory and implementation in practice.
Concepts discussed are grounded strongly in a cited body of literature.	There is some recognition and discussion of the strengths and weaknesses of the programme as implemented.	There is limited or no recognition and discussion of the strengths and weaknesses of the programme as implemented.
Concepts are parsimonious, in that they provide the simplest but not an over-simplified explanation.	There are some attempts to explain anomalous results and findings with reference to context and data.	There are limited or no attempts to explain anomalous results and findings regarding the context and data.
There is a clear description of the factors affecting implementation.	There is some description of the factors affecting implementation.	There is limited or no description of the factors affecting implementation.
	Documents are typified by terms, *e.g.*, 'model', 'process' or 'function'; verbs, *e.g.*, 'investigate', 'describes' or 'explains'; and topics, *e.g.*, 'experiences'.	Documents are typified by mentioning only an 'association' between variables.

SSM: Self- and shared management.

Appendix 12. Initial question theory document review mapping

First author (year)	Title	Source	Conceptual richness*
ARMA (2010)	Standards of care for children and young people with juvenile idiopathic arthritis	Medical society publication	3
Bandura (1977)	Self-efficacy: toward a unifying theory of behavioural change	Journal article	1
Bandura (1977)	Social learning theory	Book chapter	1
Bandura (1999)	Social cognitive theory: an agentic perspective	Conceptual article	1
Bandura (2001)	Social cognitive theory: an agentic perspective	Conceptual article	1
Bate (2014)	Perspectives on context: a collection of essays considering the role of context in successful quality improvement	Report/guide	2
Beresford (2010)	Comment on: developing standards of care for patients with juvenile idiopathic arthritis	Editorial	3
Bosworth (2020)	The NRAS New2RA right start service – a comprehensive and tailored support service for people newly diagnosed with rheumatoid arthritis	Practice/guideline	3
Bridges (2020)	Self-management consultancy	Webpage	3
Bronfenbrenner (1979)	The ecology of human development	Book chapter	2
BSPAR (2009)	Standards of care for children and young people with juvenile idiopathic arthritis	Medical society publication	3
Burd (2016)	Supporting self-management. A guide to enabling behaviour change for health and wellbeing using person- and community-centred approaches	Report/guide	2
Burd (2016)	Making the change: behavioural factors in person-and community centred approaches for health and wellbeing	Report/guide	2
Burd (2016)	Spreading change: a guide to enabling the spread of person- and community-centred approaches for health and wellbeing	Report/guide	2
De Silva (2011)	Helping people help themselves: a review of the evidence considering whether it is worthwhile to support self-management	Report/guide	2
Erickson (2005)	Brief interventions and motivational interviewing with children, adolescents, and their parents in pediatric health care settings	Journal article	2
Finnis (2016)	Realising the value: ten key actions to put people and communities at the heart of health and wellbeing	Report/guide	2
Grey (2006)	A framework for the study of self- and family management of chronic conditions	Journal article	2
Grey (2015)	A revised self- and family management framework	Journal article	2
HCSA (2015)	Individual healthcare plan	VCSE publication	3
Kavirayani (2013)	Paediatric rheumatology practice in the UK benchmarked against the British Society for Paediatric and Adolescent Rheumatology/Arthritis and Musculoskeletal Alliance standards of care for juvenile idiopathic arthritis	Journal article	3
Kazak (2006)	Pediatric psychosocial preventative health model: research, practice, and collaboration in pediatric family systems medicine	Journal article	2
Kieckhefer (2000)	Supporting development of children with chronic conditions: from compliance toward shared management	Journal article	3
Kirk (2010)	Evaluating self-care support for children and young people with long-term conditions	Report/guide	1
Kirk (2012)	Perceptions of effective self-care support for children and young people with long-term conditions	Journal article	2

First author (year)	Title	Source	Conceptual richness*
Kirk (2012)	The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review	Journal article	2
Knafl (1990)	Family management style: concept analysis and development	Journal article	2
Knafl (2008)	The interplay of concepts, data, and methods in the development of the family management style framework	Journal article	2
LTHT (2018)	Children's rheumatology	Webpage	3
LTHT (2018)	Yorkshire children and young people's rheumatology network	Webpage	3
LGA (2016)	Just what the doctor ordered. Social prescribing-a guide for local authorities	Report/guide	3
Lorig (2003)	Self-management education: history, definition, outcomes, and mechanisms	Conceptual article	1
Lozano (2018)	Supporting self-management in children and adolescents with complex chronic conditions	Journal article	2
Michie (2011)	The behaviour change wheel: a new method for characterising and designing behaviour change interventions	Journal article	1
Modi (2012)	Pediatric self-management: a framework for research, practice, and policy	Journal article	2
NRAS (2020)	New2ra right start service	VCSE publication	3
Paterson (2001)	The shifting perspectives model of chronic illness	Journal article	2
Perry (2011)	Behavioural insights in health care: nudging to reduce inefficiency and waste	Report/guide	1
Rapoff (2018)	Assessing barriers to therapeutic regimens for young people with juvenile idiopathic arthritis	Editorial	3
RCPCH (2017)	Paediatric rheumatology level 3 paediatrics sub-specialty syllabus	Medical society publication	3
Ryan (2008)	Facilitating health behaviour change and its maintenance: interventions based on self-determination theory	Conceptual article	1
Ryan (2009)	The individual and family self-management theory: background and perspectives on context, process, and outcomes	Conceptual article	1
Sattoe (2015)	Self-management interventions for young people with chronic conditions: a systematic overview	Journal article	2
Sattoe (2015)	Growing up with a chronic condition: challenges for self-management and self-management support	Thesis	1
Service (2014)	EAST - four simple ways to apply behavioural insights	Report/guide	2
Stinson (2016)	The ipeer2peer program: a pilot randomized controlled trial in adolescents with juvenile idiopathic arthritis	Journal article	2
Stones (2021)	Summary of patient/parent organisation services promoting self- and shared-management of JIA in the UK and Ireland	Conference proceeding	3
Tong (2012)	Children's experiences of living with juvenile idiopathic arthritis: a thematic synthesis of qualitative studies	Journal article	3
Van Dongen (2014)	The patient segmentation model	Webpage	3
Versus Arthritis (2019)	Individual healthcare plan	VCSE publication	3
Wallace (2010)	Developing standards of care for patients with juvenile idiopathic arthritis	Editorial	3
Wilson (2018)	Good and bad help - how purpose and confidence transform lives	Report/guide	2
Wood (2016)	At the heart of health: realising the value of people and communities	Report/guide	2

*1=Conceptually rich, 2=Thicker description, 3=Thinner description. ARMA: Arthritis and Musculoskeletal Alliance; BSPAR: British Society for Paediatric and Adolescent Rheumatology; EAST: Easy, attractive, social, timely; HCSA: Health Conditions in Schools Alliance; LGA: Local Government Association; LTHT: The Leeds Teaching Hospitals Trust; NRAS: National Rheumatoid Arthritis Society; RCPCH: Royal College of Paediatrics and Child Health; VCSE: Voluntary, community, and social enterprise.

Appendix 13. Exemplar interview discussion guide

Researcher introduction	• Welcome and thank you for participating • Introduce as Simon Stones, PhD student, patient advocate with lived experience), University of Leeds, supervised by Professor Swallow and Dr. Milnes • Revisit information sheet, time to complete the interview, and audio recording consent
Exploring individual (CYP) context	• What factors do you think contribute to a CYP's willingness to self-manage their JIA? • From your experience, what things do they typically do (or not do), and why?
Exploring individual/ interpersonal (family) context	• What roles do families play in managing JIA? What has been your experience? • Do you have any experience of how siblings cope, and whether they are supported? • Beyond parents and siblings, who else do you think/do you know contributes to supporting CYP?
Exploring interpersonal (peer) context	• Do you think that JIA impacts on CYP's relationships with peers? • Do you have any examples of how JIA influences CYP's social and family life? • Do CYP and their families have many opportunities to speak to peers with JIA? Does the clinic provide opportunities for this, and if so, what impact does this have?
Exploring individual/ interpersonal (HCP) context	• What are your experiences of how CYP communicate with you? • Do CYP tend to take an active or passive roles? • Do parents tend to take more of an active role, and if so, at what point does this tend to shift towards CYP? • What is the typical consultation dynamic like? • How do you try to engage and involve CYP with JIA in shared decision making? • Do CYP and their families differ in their communication/personalities depending on the healthcare professional (*i.e.*, doctor, nurse, physio) they are with? • Do you think you provide enough information and support (*i.e.*, to VCSEs)? • What works well from your current practice? • What do you feel could be improved in terms of information provision and self-management support?
Exploring institutional/ infrastructural (education) context	• How does JIA affect CYP's education and development? • What have been your experiences of schools in terms of supporting CYP and families? Have schools ever contacted you? • Do you find that schools are proactive in supporting CYP (*i.e.*, use of individual healthcare plans)? • What needs to be improved from the education perspective?
Exploring institutional/ infrastructural (health/VCSE) context	• How do you think the healthcare system could become more efficient for CYP families (*i.e.*, joint clinics, digital health)? • What role do you feel that VCSEs play in supporting CYP and families?
Exploring mechanisms	• What strategies do you believe can motivate/empower CYP to take control? • What are your thoughts on individual and group learning/activities? • How do you think interventions need to be developed/delivered? • What are your thoughts on peer support and peer-delivered information? Do you feel that CYP and families are more likely to listen and act if information and support is delivered by other children/young people and families? • Do you find that CYP alter how they manage their condition during different times in their lives? If so, how and why? • Do you think that CYP have enough of a say in how their JIA is managed? • Do you think that families have enough of a say? • Do you feel that CYP and families know enough about JIA and how to manage? • Where do the families you work with tend to find most information about JIA? • How do you think information and support should best be delivered?
Exploring outcomes	• Do you think that anything or anyone may influence whether CYP and/or families have a say about JIA? • How do you balance the CYP-family relationship/dynamic? • Education has been an important part of other condition self-management programmes, but they have not necessarily led to CYP developing skills to learn to take control and manage their JIA. Why do you think that is the case? • What is different about paediatric self-management from adult self-management? • What do you think we need to consider when we design and develop resources for CYP with JIA and their families?

CYP: Children and young people; HCP: Healthcare professional; JIA: Juvenile idiopathic arthritis; PhD: Doctor of Philosophy; VCSE: Voluntary, community, and social enterprise.

Appendix 14. Ethical approval

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Mr Simon Stones
Postgraduate/Doctoral Researcher
University of Leeds
2.30 Baines Wing
University of Leeds
Leeds
LS2 9JT

Email: hra.approval@nhs.net
Research-permissions@wales.nhs.uk

10 August 2018

Dear Mr Stones

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Innovative approaches to the Self- and shared-Management of ARThritis by children, their families and professionals: A realist approach.**
IRAS project ID:	**234475**
Protocol number:	**N/A**
REC reference:	**18/LO/1087**
Sponsor	**University of Leeds**

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.).

IRAS project ID	234475

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?
The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?
You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: The Faculty NHS Research Ethics Officer
Tel: 01133437587
Email: governance-ethics@leeds.ac.uk

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **234475**. Please quote this on all correspondence.

Yours sincerely

Laura Greenfield
Assessor

Email: hra.approval@nhs.net

Copy to: *Faculty NHS Research Ethics Officer [Sponsor Contact on behalf of Leeds University]*
Ms Anne Gowing [Lead NHS R&D Contact on behalf of Leeds Teaching Hospital NHS Trust]

Appendix 15. Question theory comparisons

IQT1	RQT1
If self-management support opportunities for CYP with JIA are introduced, easily accessed, attractive, socially-orientated and consistently endorsed from the point of diagnosis in an age, developmentally-appropriate and personally meaningful way for each CYP, within a trusted and familiar network across healthcare, education and VCSE organisations, **then** CYP will have the opportunity over time to become increasingly competent at the medical, role and emotional management of their H&W, by improving their knowledge, beliefs, self-regulation skills and abilities during childhood, in anticipation for adulthood.	**If** self-management opportunities for CYP with JIA are introduced from the point of diagnosis and consistently over time in age, developmentally-appropriate, meaningful, attractive, and individualised ways that focuses on underlying behaviours, within a trusted, and credible network of professional and peer support with the support of their families, **then** CYP will be more likely to become increasingly competent at the role, emotional, and medical management of their H&W, by accepting their condition, improving their knowledge, beliefs, confidence, self-regulation skills and abilities during the receptive and adaptive period of childhood and adolescence, in anticipation for adulthood.
Outcome (distal) • Influence the medical, role, and emotion management of JIA. • Readiness for adulthood.	**Outcome (distal)** • Improved emotional management, including emotional domain of HRQoL, psychological outcomes, attitudes towards JIA, acceptance, coping, self-appreciation, self-confidence, and self-esteem. • Improved medical management, including health outcomes, physical domain of HRQoL, knowledge of JIA and treatment, condition-related self-efficacy, condition-specific self-management behaviours, symptom management, adherence with treatment, relationships with HCPs, family involvement in condition-related tasks, and problem solving.
Outcome (proximal) • Increasing self-management competence.	**Outcome (proximal)** • Improved role management, including health outcomes, social domain of HRQoL, general self-efficacy and sense of control, autonomy, social participation, vocational participation, coping, disclosure, psychosocial functioning, behavioural compliance with parental instructions, family functioning, problem solving, peer support, conflict resolution and lifestyle modifications.
Mechanism (response) • Knowledge and beliefs. • Self-regulation skills and abilities.	**Mechanism (response)** • Self-regulation skills and abilities, including problem solving, critical appraisal, decision making, planning and action, goal setting, pacing, self-monitoring, reflective thinking, risk assessment, self-evaluation, including relative comparison, emotional control, level of expectation, and communication. • Knowledge and beliefs, including seeking information, taking ownership, goal congruence, outcome expectancy, empowerment, self-efficacy, self-assurance, self-confidence, and repetitive behaviours, such as internalisation and introjection; resolving cognitive dissonance. • Faith and trust in professionals and families who make them feel valued and safe. • Receptiveness to new information and support, including modelling the behaviours of others, in particularly peer role models. • Willingness to accept their condition and responsibility from their families.
Mechanism (resource)	**Mechanism (resource)**

• Self-management support from the point of diagnosis in an easily accessed, attractive, socially-orientated, and timely manner. • Age- and developmentally-appropriate. • Meaningful to each individual.	• Timely access to relevant and engaging information and support, including social prescribing, delivered consistently during childhood and adolescence. • Multichannel approach to education, catering for different learning styles and preferences, including the use of familiar and emerging technologies and environments which make learning attractive, engaging, and pleasant. • Experiential learning including mastery experiences and a space for negotiated collaboration within a non-judgemental, informal, and familiar environment. • Emotional, information and instrumental support, including positive reinforcement. • Social influence, peer dynamic, and role model motivators and supporters. • Honesty, respect, and encouragement to be involved, framed by reality. • Reassurance related to the gradual transfer of responsibility as part of future planning.
Context • Non-modifiable demographics. • Behaviours. • Condition-specific factors. • Family structure, function, and relationship. • Interest in self- and shared-management. • Trusted and familiar network. • Receptiveness for change. • Shifting of responsibilities. • Cultural norms and stigmatisation.	**Context** • Non-modifiable demographics, such as age, gender identity, development stage, cognitive functioning, information processing, learning style, communication preferences, and cultural norms. • Stigmatisation, disabling attitudes, inferiority complex, public understanding and social empathy. • Condition-specific factors including severity, trajectory, volatility, temporal disease activity, multimorbidity, and treatment complexity. • Family factors, including family structure and function, parental anxiety and coping styles, socioeconomic status, and pre-existing adaptations. • Relationship with families and professionals supporting their health and wellbeing. • Growth mindset, including acceptance, receptiveness and intention to change behaviour, motivations, risk preference, and perseverance. • Personality, maturity, perspective, resilience, fears (including phobias and anticipatory behaviours), coping style, cognitive dissonance, behaviour and temperament, including feelings of grief, guilt, punishment, anger, anxiety, and depression. • Health literacy and health numeracy. • Proximal versus distal outcome focus and prevention versus reaction focus. • Competing priorities.
IQT2	**RQT2**
If shared-management support opportunities for parents, siblings and other family members supporting CYP with JIA are available and accessible over time from the point of diagnosis, with an emphasis on the gradual transfer of responsibility at appropriate timepoints to CYP, **then** families will generate their own strategies to aid CYP and the entire family to more competently manage the medical, role and emotional aspects of JIA now and in the future.	**If** parents, siblings and other family members are recognised, and holistically supported to enhance their influential shared-management role of JIA, from the point of diagnosis, while acknowledging the shifting responsibility of management to CYP over time, **then** families will be equipped with the knowledge, beliefs, skills, abilities, and support infrastructure to positively influence the role, emotional and medical aspects of JIA for their child and the entire family, while supporting their child's to become increasing autonomous.
Outcome (distal)	**Outcome (distal)**

• Influence the medical, role, and emotional management of JIA.	• Improved emotional management, including psychological outcomes, attitudes towards JIA, acceptance, coping, self-appreciation, self-confidence, and self-esteem. • Improved medical management, including health outcomes, HRQoL, knowledge of JIA and treatment, symptom management, and relationship with HCPs.
Outcome (proximal)	**Outcome (proximal)** • Improved role management, including general self-efficacy, coping, psychosocial functioning, family involvement in tasks, family functioning, problem solving, peer support, conflict resolution and lifestyle modifications. • Influence on CYP's self-management behaviours and autonomy.
Mechanism (response) • Develop own strategies.	**Mechanism (response)** • Self-regulation skills and abilities, including problem solving, critical appraisal, decision making, planning and action, goal setting, self-monitoring, reflective thinking, risk assessment (including managing protectiveness and catastrophisation), self-evaluation (as proxy), emotional control, level of expectation, and communication. • Knowledge and beliefs, including seeking information, taking ownership, goal congruence, outcome expectancy, empowerment, self-efficacy, self-assurance, and repetitive behaviours (including projection). • Faith and trust in professionals supporting them and their child who make them feel valued. • Receptiveness to accept and access support, including proactive steps and measures. • Willingness to transfer responsibility to their child.
Mechanism (resource) • SSM support available and accessible over time from the point of diagnosis. • Emphasis on gradual transfer of responsibility.	**Mechanism (resource)** • Timely access to relevant and engaging information, education and support, including social prescribing, delivered strategically at regular touchpoints. • Multichannel approach to education, catering for different learning styles and preferences, including use of familiar technology. • Experiential learning including mastery experiences. • Emotional, information and instrumental support for parents and siblings, including registration as carers with the local authority. • Space for discussion and negotiated collaboration within a non-judgemental atmosphere. • Social influence, peer dynamic and coerced friendships. • Bridging roles and continuity of interaction with professionals providing support. • Emphasis and reassurance on gradual transfer of responsibility.
Context • Non-modifiable demographics. • Behaviours. • Condition-specific factors. • Family structure, function, and relationship. • Interest in self- and shared-management. • Receptiveness for change. • Shifting of responsibilities. • Cultural norms and stigmatisation.	**Context** • Non-modifiable demographics of families and their child. • Growth mindset, including receptiveness and intention to change behaviour, motivations, and risk preference. • Health literacy and health numeracy. • Shifting responsibilities related to the family structure and function, and increasing management beyond the clinical environment. • Relationship with professionals supporting their child's health and wellbeing. • Perspective, parental anxiety, fear, coping style and behaviour/temperament, including feelings of grief, guilt, anxiety, and depression, heightened during periods of acceptance and adjustment.

	• Proximal versus distal outcome focus, and seeing the value of self- and shared-management in the short- and long-term. • Condition severity, trajectory, temporal disease activity, multimorbidity, and treatment complexity. • Competing priorities. • Level of disruption previously experienced. • Availability of pre-existing adaptations and assistive equipment. • Income and socioeconomic status. • Cultural norms, stigmatisation and inferiority complex, reaching unseen and unheard families. • Social capital.
IQT3	**RQT3**
If IHPs are made available to all CYP with JIA and their families from the point of diagnosis, are unfailingly used and regularly updated over time to reflect on the specific needs and preferences of CYP with JIA, **then** this will facilitate how key stakeholders (such as school staff, other HCPs and VCSE professionals) support the shared-management of JIA for CYP under their professional care, in a manner which reflects the medical, psychological and social needs of every CYP with JIA.	**If** IHPs are uniformly recognised as a shared management communication tool and are made available to CYP with JIA and their families from the point of diagnosis, maintained and updated over time to reflect on the individual needs and circumstances of CYP, **then** CYP and families will be enabled to play a bigger role in articulating the support they need, facilitating SDM between relevant professional stakeholders involved in the shared-management of JIA in order to improve the manner in which holistic care and support is provided.
Outcome (distal) • Influence medical and psychosocial needs of CYP.	**Outcome (distal)** • Improved role management of JIA. • Impact on emotional and medical management of JIA.
Outcome (proximal) • Involvement in shared-management of CYP's condition.	**Outcome (proximal)** • SDM between CYP, families, and other relevant stakeholders. • Enhanced engagement/involvement with care, treatment, and support.
Mechanism (response)	**Mechanism (response)** • Self-regulation skills and abilities, including problem solving, decision making, planning and action, goal setting, self-monitoring, reflective thinking, risk assessment, level of expectation, communication, and behaviour. • Knowledge and beliefs, including taking ownership, goal congruence, outcome expectancy, and empowerment. • Articulation of needs and preferences. • Appropriate interpretation and consistent application of recommendations.
Mechanism (resource) • IHPs made available from point of diagnosis. • Regularly used and updated. • Reflect needs and preferences.	**Mechanism (resource)** • Templated IHPs in desired formats utilised from the point of diagnosis with guided completion to facilitate communication. • Maintained and regularly updated by all key stakeholders, with CYP and families involved at all times. • Reflect the unique needs and preferences of CYP and families, including contingent planning.
Context	**Context**

• Relationship between CYP/families and other stakeholders.	• Healthcare and education professionals' awareness, value and utility of IHPs, in contrast to EHCPs, and comparison to other disease areas. • Designated responsibility of professional stakeholder to initiate IHP development and dissemination. • Influence of active/refractory disease versus minimal disease activity/remission on the role of IHPs. • Reactive versus proactive outlook of healthcare and education professionals. • Implementation of legislation and guidance within the education setting.
IQT4	**RQT4**
If all members of the paediatric rheumatology MDT (internally and externally) recognise the value of SSM support, are aware of all available support opportunities for CYP with JIA and their families, and consistently refer and/or signpost them to such support from the point of diagnosis and throughout long-term follow-up, **then** it is more likely that CYP and families will secure access to trusted information and support from the outset, in order to aid the medical, role and emotional management of JIA in a more consistent manner.	**If** all members of the paediatric rheumatology MDT and other professionals involved in JIA care and support recognise, value, and consistently encourage SSM support for CYP with JIA and their families, from diagnosis and throughout follow-up in a standardised and transparent manner that is holistic, proactive, and inclusive, **then** CYP and their families are more likely to access and trust the information and support that they require from the outset, to improve their relationship with HCPs and develop their skills to better manage the, role, emotional, and medical aspects of JIA, enhancing their experience of care in the process.
Outcome (distal) • Influence the medical, role, and emotional management of JIA.	**Outcome (distal)** • Improved role, emotional, and medical management of JIA.
Outcome (proximal) • Access trusted information and support.	**Outcome (proximal)** • Enhanced experience of care. • Enhanced skillset amongst CYP and families. • Improved relationship between CYP, families, and HCPs. • More honest and transparent conversations. • Increased emphasis on CYP's care which more accurately reflects their lives. • CYP and families are more likely to access trusted information and support from the outset.
Mechanism (response)	**Mechanism (response)** • CYP and families feel valued, respected, and part of the conversation. • CYP and families, develop a mutual understanding, level of familiarity and trust with HCPs, reducing the likelihood of misinterpretation. • Knowledge and beliefs, including taking ownership, goal congruence, outcome expectancy, empowerment, self-efficacy, and self-assurance. • Feelings of guilt are minimised.
Mechanism (resource) • From diagnosis through long-term follow-up.	**Mechanism (resource)** • Clear, consistent and honest communication. • Transparency of health data. • SDM, including aligned goals, involvement in clinical conversations, power sharing, co-creation, and informed consent/assent.

	• Engagement of CYP through repeated questioning, direct addressing, negotiation, provision of time, recognising their expertise and that of their family. • Pre-empting and documenting specific needs and preferences from diagnosis through long-term follow-up. • Co-ordinated and continuous care by trusted, credible, and familiar professionals working in partnership with CYP and families. • Incentivised HCPs.
Context • Interaction and relationship between CYP, families, and HCP. • MDT collaboration (internal and external). • Recognise the value of self- and shared-management support. • Consistent referral/signposting.	**Context** • Compassionate and empathetic interaction and relationship between CYP, families, HCPs, and other professional stakeholders, including adequate provision of time. • Consistent and meaningful signposting and referral to bespoke and individualised opportunities. • Understanding and valuing the need to promote SSM across the lifecourse. • Collaboration and aligned perceptions (social capital) among the paediatric rheumatology MDT and other professional stakeholders. • Appropriate training, social skills, and administrative support, including allocation of roles and responsibilities. • Receptiveness to change. • Consideration of biopsychosocial model of H&W and application to H&SC, beyond conventional clinical focus. • Medical hierarchy and influence of omnipotent consultant. • Increasing presence of patient and parent advocates changing the patient-HCP relationship. • Professional membership, affiliation, and research-focus. • Audit and peer review of services offered.
IQT5/6	**RQT5**
If SSM support services are commissioned in a way that are integrated with statutory services (*e.g.*, VCSE-delivered services within H&SC services), so as to be available and accessible in a timely manner for all CYP with JIA and their families as part of routine care, **then** those CYP and families will have quick and easy access to the resources required for them to be able to more competently manage all aspects of JIA on a more equitable and informed level, while reducing their reliance on constrained H&SC services that could otherwise be avoidable. **If** paediatric rheumatology H&SC services are designed to facilitate access to, and navigation of services in a CYP- and family-centred way, including an holistic focus on integrated SSM support within clinical practice, shared care across specialities and settings, continuity of care, seamless transition and involvement of CYP and families in all decision-making processes, **then** the H&W of CYP with JIA, as well as their families, is likely to be improved.	**If** paediatric rheumatology services are designed and commissioned in a CYP-, family-focused, and co-ordinated manner, including a biopsychosocial focus on integrated SSM services, that are readily available and accessible, **then** CYP and their families will have the ability and desire to access the necessary information and support that they require and trust, to feel informed, empowered, and better supported to improve their overall H&W, and readiness for the future, while reducing the pressures on statutory H&SC services that could potentially be avoided by optimal access of the services that best suit their situational needs.

Outcome (distal) • Health status (*e.g.*, remission) • Quality of life • Cost of health (*e.g.*, direct and indirect costs)	**Outcome (distal)** • Improvement to longer-term overall H&W of CYP and families, principally minimal disease activity or remission. • Improved adherence to prescribed treatment. • Reduction to direct and indirect costs of health. • Prioritisation of integrated services between statutory H&SC and VCSEs. • Reduction in overall pressure on statutory H&SC services through optimal access and service utilisation, including emergency department visits, hospitalisation, and other service-related costs.
Outcome (proximal) • Healthcare service utilisation (*e.g.*, hospitalisation). • VCSE service utilisation (*e.g.*, attendance at events).	**Outcome (proximal)** • Utilisation of VCSE-provided services. • CYP's readiness for transition to adult H&SC services.
Mechanism (response) • CYP feel more informed.	**Mechanism (response)** • CYP and families feel more informed, empowered, and better supported. • Increased involvement in, and desire for, SDM. • Increased familiarity and trust with key stakeholders, settings, and services.
Mechanism (resource) • Individualised, person- and family-focused care and support from H&SC and VCSE sectors. • Timely access and availability.	**Mechanism (resource)** • Transparency of health data and consistent reporting. • A multi-disciplinary, biopsychosocial, CYP-friendly focus. • Consistent and meaningful signposting to bespoke and individualised opportunities. • Readily available and accessible information, care, and support. • Co-ordinated care by key professionals advocating for CYP and families.
Context • H&SC access and dynamic. • Integration of SSM (including VCSE services) with clinical practice. • Patient-centredness. • VCSE funding, location and coverage.	**Context** • Valuing the need to promote SSM across the lifecourse. • Politicised culture inhibiting collaboration and sharing of best practices. • Implementation and audit of relevant standards and guidelines. • Integrated interdisciplinary services between statutory H&SC, local authorities, education providers, and VCSEs. • Physical and social environmental factors, including social incentives/disincentives, material and community resources, culture and cultural norms, transportation, access to care, models of care, continuity of care, co-ordinated care, system navigation, and MDT provision, in addition to VCSE funding, location, and coverage. • Stigmatisation, general awareness of JIA and overall musculoskeletal health priority in all settings.
IQT7	**RQT6**
If schools are inclusive for all CYP with SEND (including LTCs like JIA) with appropriate policies and provisions in place, and if relevant education professionals within schools (and the local authority) are involved in the shared-management of JIA by working with CYP, families, HCPs and VCSEs in identifying and communicating specific support needs for each CYP, **then** CYP with JIA will be more likely to enjoy school and have a	**If** schools proactively provide bespoke, co-ordinated, and inclusive support for CYP with JIA, guided by appropriate SEND policies and procedures, within a culture of open and regular dialogue between CYP, their families and other external professionals where appropriate and necessary, **then** CYP are more likely to have a positive experience in education that fulfils their academic potential and social development needs, akin to their peers, while feeling safe, supported and reassured that their H&W will not be jeopardised as a result of them attending their place of education.

positive experience, akin to their peers, so as to not be at a disadvantage in terms of educational attainment and social development.	
Outcome (distal) • CYP experience an equivalent educational attainment and social development akin to peers.	**Outcome (distal)** • CYP experience a fulfilling education (meeting academic potential/social development needs), akin to peers. • Improved overall attendance, if CYP feel better supported and cared for in school, and families are more confident about school's ability to support their child.
Outcome (proximal) • School is a positive and supportive environment.	**Outcome (proximal)** • School becomes a safe, supportive, and inclusive environment which CYP want to attend. • CYP, families, schools, and other key stakeholders work proactively and collaboratively maintaining an open and regular dialogue.
Mechanism (response)	**Mechanism (response)** • All stakeholders set expectations and goals. • CYP and families having faith and trust in their school. • CYP and families feel reassured that their health and wellbeing is a priority.
Mechanism (resource) • Individualised, person- and family-focused care and support from education sector. • Involvement of relevant professional stakeholders.	**Mechanism (resource)** • Schools co-ordinate the involvement of relevant professional stakeholders and peers. • Needs, preferences, and plans are transparently documented and regularly reviewed, including evidence generation for examination access arrangements. • Contingency plans are made to account for every eventuality, including remote and flexible learning. • Schools actively involve CYP in prioritising bespoke arrangements that meet their needs and preferences. • CYP and families are proactively made aware of, and encouraged to access support that is available, and adjustments which can be made.
Context • A supportive and inclusive school setting. • Relevant SEND provision and policies. • Regular and consistent communication between school and families.	**Context** • School foster an inclusive and supportive culture, that is empathetic, flexible, and sensitive to the unique needs and preferences of CYP and their families. • Schools consistently and correctly implement relevant legislation, evidenced through internal policies and provision for CYP with JIA, audited by the governing body. • Education professionals are proactive and responsive, including communicating with CYP, families and other stakeholders. • Schools minimise the general pressure felt by CYP regarding attendance by ending attendance rewards. • Ringfenced budget and resource, including appropriate staffing, should be in place to support all CYP with additional H&W needs.

CYP: Children and young people; EHCP: Education, health, and care plan; H&W: Health and wellbeing; H&SC: Health and social care; HCP: Healthcare professional; HRQoL: Health-related quality of life; IHP: Individual healthcare plan; IQT: Initial question theory; LTC: Long-term condition; MDT: Multi-disciplinary team; RQT: Refined question theory; SDM: Shared decision making; SEND: Special educational needs and disabilities; SSM: Self- and shared-management; VCSE: Voluntary, community, and social enterprise.