Illness perceptions in adolescents with juvenile arthritis: applying the common sense self-regulatory model

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<th>Full Form</th>
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<tbody>
<tr>
<td>ANCOVA</td>
<td>Analysis of covariance</td>
</tr>
<tr>
<td>BIPQ</td>
<td>Brief illness perception questionnaire</td>
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<tr>
<td>$\beta$</td>
<td>Beta-coefficient</td>
</tr>
<tr>
<td>CAPS</td>
<td>Childhood arthritis prospective study</td>
</tr>
<tr>
<td>CBT</td>
<td>Cognitive behavioural therapy</td>
</tr>
<tr>
<td>CHAQ</td>
<td>Child health assessment questionnaire</td>
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<tr>
<td>CHQ</td>
<td>Child health questionnaire</td>
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<tr>
<td>CI</td>
<td>Confidence intervals</td>
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<tr>
<td>CS-SRM</td>
<td>Common sense self-regulatory model</td>
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<tr>
<td>DIRQ</td>
<td>Diabetes illness representations questionnaire</td>
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<tr>
<td>ESM</td>
<td>Experience sampling method</td>
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<tr>
<td>GCT</td>
<td>Gate control theory</td>
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<tr>
<td>GHQ</td>
<td>General health questionnaire</td>
</tr>
<tr>
<td>HAQ</td>
<td>Stanford health assessment questionnaire</td>
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<tr>
<td>HBA1c</td>
<td>Metabolic control</td>
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<tr>
<td>IASP</td>
<td>International association for the study of pain</td>
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<tr>
<td>ICC</td>
<td>Interclass correlations</td>
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<tr>
<td>ILAR</td>
<td>International league of associations of rheumatology</td>
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<tr>
<td>IMIQ</td>
<td>Implicit model of illness questionnaire</td>
</tr>
<tr>
<td>IMS</td>
<td>In my shoes</td>
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<tr>
<td>IPQ</td>
<td>Illness perception questionnaire</td>
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<tr>
<td>IPQ-C</td>
<td>Illness perception questionnaire child version</td>
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<tr>
<td>IPQ-R</td>
<td>Revised illness perception questionnaire</td>
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<td>IQR</td>
<td>Interquartile range</td>
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<tr>
<td>JIA</td>
<td>Juvenile Idiopathic Arthritis</td>
</tr>
<tr>
<td>JRA</td>
<td>Juvenile Rheumatoid Arthritis</td>
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<tr>
<td>LTC</td>
<td>Long term condition</td>
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<tr>
<td>MAR</td>
<td>Missing at random</td>
</tr>
<tr>
<td>MCAR</td>
<td>Missing completely at random</td>
</tr>
<tr>
<td>MNAR</td>
<td>Missing not at random</td>
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<td>OA</td>
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<td>Survey of pain attitudes for paediatrics</td>
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<td>Pain coping questionnaire</td>
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<td>Pain catastrophizing scale for children</td>
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<td>PPQ-YP</td>
<td>Pain perceptions questionnaire for young people</td>
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<tr>
<td>PsA</td>
<td>Psoriatic arthritis</td>
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<tr>
<td>$R^2$</td>
<td>Coefficient of determination</td>
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<td>RA</td>
<td>Rheumatoid arthritis</td>
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<td>RCT</td>
<td>Randomised control trial</td>
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<tr>
<td>RHO</td>
<td>Spearman’s rank correlation coefficient</td>
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<tr>
<td>Abbreviation</td>
<td>Description</td>
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<tr>
<td>SEM</td>
<td>Structure equation model</td>
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<td>SD</td>
<td>Standard deviation</td>
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<td>Type 1 diabetes</td>
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<tr>
<td>VAS</td>
<td>Visual analogue scale</td>
</tr>
<tr>
<td>WHO</td>
<td>World health organisation</td>
</tr>
<tr>
<td>ZPD</td>
<td>Zone of proximal development</td>
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</table>
Abstract

Background: Juvenile idiopathic arthritis (JIA) is a long-term inflammatory arthritis which starts before the age of 16; 60% of those with JIA continue to have symptoms into adulthood. There are wide variations in experiences of adolescents with JIA, including the effects of the condition on social and intellectual development, self-management, psychological and physical functioning. Individual differences may be due to differences in how adolescents conceptualise JIA. Leventhal’s Common Sense Self-regulatory Model (CS-SRM) has been used to theorise adults’ experiences of illness, however, this has rarely been used with adolescents. The CS-SRM has three components, mental representations (component 1) that drive coping procedures and illness behaviours (component 2) which are then evaluated and appraised (component 3). A review of studies with adolescent cohorts that applied the CS-SRM found no evidence-base with which to justify application of an adult theory with children or adolescents. Thus the applicability of the theory and the use of the associated questionnaire, the Revised Illness Perceptions Questionnaire (IPQ-R), with adolescents are problematic. Aims: The aims of this PhD were first to assess the suitability of the CS-SRM for adolescents with JIA by investigating the three components of the model, and second, to develop a questionnaire for use with this population. Methods: To investigate the first two components of the CS-SRM, twenty-one participants aged between 11-16 years were recruited from a national cohort of JIA patients. Data collection was undertaken using cognitive interviewing. Framework analysis of the data was used to identify domains used by adolescents to conceptualise their JIA and content analysis to further investigate the suitability of the IPQ-R to assess beliefs. Transcripts were analysed identifying problems or inconsistencies with IPQ-R use. Adolescents’ ways of coping with JIA were investigated using the somatic experience module of a computer-based interview, ‘In My Shoes’ (IMS). To evaluate the third component of the CS-SRM, quantitative data were used in a longitudinal mediation analysis to investigate the extent to which emotional representations and pain predicted physical behaviour (n= 50). To address the second aim of this PhD, Version 1 of the Pain Perception Questionnaire for Young People (PPQ-YP) was devised and sent to 18 healthy adolescents (11-16) to assess linguistic validity and face validity of the items using a recent pain to answer items and provide feedback on language and length. The psychometric properties of a revised version were tested with 76 adolescents with JIA. Results: Adolescents’ responses to having JIA were driven by their emotional and cognitive responses to symptoms (in this case pain) rather than illness beliefs per se. Thus, it is important to assess pain beliefs rather than broader illness representations. The need to preserve their social identity as ‘normal’ was a coping goal shared across the sample, however different strategies were identified, either to focus on maintaining normal activities or to attend to pain directly. Adolescents who focused on their pain held a more negative emotional representation, reported higher pain and lower functionality compared to adolescents who tried to maintain normality. Based on these results, longitudinal mediation models investigated the role of emotional representations and pain in predicting physical behaviour. Pain mediated 44% of the relationship between emotional representations and physical behaviour. Conclusions: Modifications to the model are recommended to take into account the role of social identity in the process of developing illness behaviours as well as the importance of a symptom driven conceptualisation of the condition.
Declaration

No portion of the work referred to in this thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

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I would like to express my gratitude to all the people who have made this work possible.

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And to my family, my sister, Kathiana, I would definitely not be here without you, thank you for the entire small to big and everything in the middle gestures of support that you have provided throughout my whole life. Thank you to my parents; my mother, for inspiring me to try and find my niche that can make a difference to the younger generations, and to my dad, for telling me to go for it no matter what. And lastly, to my partner Erik, for helping us build a place that felt like home, (and a computer to go in it), for having unwavering belief that I could get through this, and for reminding me how proud you were when I was too far in to see the light.

Most of all, I would like to acknowledge and thank deeply, the adolescents who took part in the research, the thirst for knowledge and to help is inspiring and I hope that this contribution to the literature can help with providing the empowering tools for you to take charge and accomplish great things! I am in awe of you all.
About the author

I studied my Bachelors of Psychology (Hons) at the University of Manchester. It was during this time I became interested in cognitive psychology and the translation of cognitive psychology in therapy, specifically for children and adolescents. Because of this interest, I began volunteering at ChildLine as a telephone counsellor. As a result of the training for this role I gained a Certificate of Professional Development in Listening to Children and Young People at Liverpool John Moores University. During my four years at ChildLine, I took up different roles, including a volunteer Shift Supporter, a Sessional Telephone Counsellor and ran workshops to help continue the training for other volunteers.

For my final year dissertation, I was fortunate to complete a project that was solely experimental cognitive psychology, examining how a moving protagonist’s temporal experience affects reader’s judgment of real time, supervised by Dr. Louise Connell. This introduced me to testing and examining mental representations and how people try to understand the world. However, knowing that I was interested in how these cognitive theories are then translated to a clinical world and to therapies, I undertook a Masters in Clinical and Health Psychology. During this time, I began to work as a teaching assistant in Statistics and Research Design. My post-graduate degree allowed me to gain a more in depth knowledge of methodology and the theoretical frameworks adapted to study health behaviours, coping with chronic illnesses and designing interventions. However, there were only a few that were related to children’s or adolescents’ experiences of chronic illness. It was at this point that I started to notice the gap with this group compared to adults.

Realising there was a strong pull towards research with children and adolescents; I applied for a project offered by the Childhood Arthritis Prospective Study. This project aimed to evaluate the assessment of adolescents’ experiences and the feasibility of utilising a computer assisted tool, In My Shoes (IMS), to communicate about pain. For this project, I conducted cognitive interviews with the Revised Illness Perception Questionnaire (modified for the younger sample), a questionnaire originally developed for adults from a theory also developed for adults, I tested the adolescents’ verbal ability, and I utilised the IMS tool to collect data on pain experiences and coping with pain while I also collected open-ended questionnaires from the parents that had similar questions to the interview schedule written for their child. For my MSc thesis, I analysed some of the data, to investigate how adolescents spoke about their experiences of control, both their own personal control, and treatment control as well as their emotional representation of the condition, and how this reflected their experiences of their condition. Furthermore, I reported the feasibility of using the IMS as a communication tool, using thematic analysis and content analysis to address these two aims.

During my immersion in the literature and the data, I began to realise that there was a gap in the literature of evaluating the applicability of a theoretical framework that was developed and tested with adults, to children and adolescents. There was also a lack of a coherent theory that accounts for how adolescents conceptualise a chronic illness. I was offered a PhD to continue examining the data to evaluate if a well-established theory developed for adults can address the question to how adolescents conceptualise a chronic illness. I realised this was an opportunity to immerse in the self-regulatory theory and its applicability to adolescents.

While completing my PhD, I continued to be involved in further work with the IMS tool, to develop a communication tool that assesses pain experiences. I became an international trainee at the Pain in Child Health organisation, and went for training on psycho-educational
interventions for chronic pain using technology at SickKids Research Institute, Toronto Canada. I also, analysed the data collected from the parents, having had 17 complete parent-child dyads that allowed an investigation of the role of the parent in the development of an understanding of a long-term condition.
Publications in Preparation

**Ghio D.**, *(accepted)* Adolescents: Missed opportunities in health psychology. Health Psychology Updates. (Related to Chapter 1)

**Ghio D.**, Calam, R., Thomson W., Cordingley L. *(in prep)* A Review of the Common Sense Self-Regulatory Model with Paediatric Samples (review from Chapter 2)

**Ghio D.**, Ulph F., Calam, R., Thomson W., Hyrich, K., Baildam E.M., Childhood Arthritis Prospective Study (CAPS), Cordingley L. *(in prep)* Parent-Child causal beliefs of Juvenile Idiopathic Arthritis. (related to results under subheading Cause in Chapter 5)

**Ghio D.**, Ulph F., Calam, R., Thomson W., Hyrich, K., Baildam E.M., Childhood Arthritis Prospective Study (CAPS), Cordingley L. *(under review)* “I just want to be normal”: A qualitative investigation of the coping goals of adolescents with pain related to arthritis and their parents. (Results from Chapter 6)

**Ghio D.**, Calam, R., Thomson W., Hyrich, K., Childhood Arthritis Prospective Study (CAPS), Cordingley L. Developing and Validating the Pain Perceptions Questionnaire for Young People. (Results from Chapter 9).
American College of Rheumatology ACR, Chicago, November 2011


UK Society of Behavioural Medicine, Stirling, December 2011


International Association for the Study of Pain (ISAP) 14th World Congress, Milan, August 2012


9th International Symposium on Pediatric Pain, Stockholm, June 2013

Ghio D., Ulph, F., Calam, R., Hyrich K., Baildam, E., Thomson W., Cordingley, L., Can we use the Illness Perception Questionnaire to assess adolescents’ pain beliefs?

Research and Innovation Conference, Manchester October, 2013

Ghio D., Ulph, F., Calam, R., Hyrich K., Baildam, E., Thomson W., Cordingley, L., Assessing adolescents’ illness beliefs - Is the adult illness perception questionnaire appropriate to use?

British Psychological Society: Division of Health Psychology (DHP), York, September, 2014

Ghio D., Calam, R., Ulph, F., Baildam, E., Hyrich K., Thomson W., Cordingley, L., Assessing adolescent’s illness perceptions – is the adult illness perceptions questionnaire appropriate to use?
IASP 15th World Congress, Buenos Aires, October, 2014

Ghio D., Calam, R., Hyrich K., Thomson W., Cordingley, L., Developing a Pain Perception Questionnaire for Young People with Juvenile Arthritis

British Society of Rheumatology, Manchester, April 2015

Ghio D., Calam, R., Hyrich K., Thomson W., Cordingley, L., Validating the Pain Perception Questionnaire for Young People with Juvenile Arthritis.

10th International Symposium on Pediatric Pain, May, Seattle, 2015

Ghio D., Calam, R., Hyrich K., Thomson W., Cordingley, L., Pain Perception Questionnaire for Young People with Juvenile Arthritis: Validation study.

10th International Forum on Pediatric Pain, Nova Scotia, October 2015

Ghio D., Calam, R., Hyrich K., Thomson W., Cordingley, L., Validating the Pain Perception Questionnaire for Young People with Juvenile Arthritis.
Chapter 1: Adolescents with long-term conditions

1.1. Overview

The following chapter provides contextual background to the understanding of the experiences of adolescents with juvenile idiopathic arthritis (JIA). It provides an overview of key psychological developments which occur during adolescence and how these may be affected by the presence of a long-term health condition.

In Sections 1.2 definition of adolescence is provided followed by Section 1.3 which provides contextual background to the study of adolescents with long-term conditions in the United Kingdom. Section 1.4 introduces developmental changes across the cognitive, social and physical domains occurring during adolescence, and how long-term conditions can influence these developmental changes. Section 1.5 describes specific processes involved in adjustment to a long-term condition, how they are influenced by age, and how poor adjustment can hinder achieving developmental milestones. In Section 1.6, the four frameworks that were used to understand how adolescents conceptualise illness are critically appraised. They elucidate the ways in which adolescents cope with and adjust to living with a long-term condition.

1.2. Defining adolescence

The World Health Organisation (WHO) defines adolescents in terms of chronological age as young people who are aged between 10 and 19 years (WHO, 2001). Adolescence is a critical transitional stage between childhood and adulthood during which rapid changes occur. Important health behaviours such as levels of physical activity and food choice behaviours are established or consolidated during adolescence (Kelder et al., 1994; Sacker & Cable, 2006). This means adolescence is a period of opportunity to affect and promote positive health behaviours resulting in well-being and better quality of life (Kleinert, 2007;
Sawyer et al., 2012; Resnick et al., 2012). The presence of a long-term condition during adolescence may diminish the opportunities to develop health preserving habits with implications for long term health outcomes, hospital and healthcare utilisation and self-management. As a result, there have been both international (UNICEF, 2011) and national (UK; Viner, 2012) calls to focus on adolescents with long-term conditions.

1.3. Adolescents with long-term conditions (LTCs)

The onset of certain long-term conditions (LTCs), such as juvenile idiopathic arthritis (JIA) or type 1 diabetes (T1D) often occurs during adolescence. In JIA disease onset occurs during adolescence in approximately a quarter of reported cases (Oen et al., 2002; Shaw et al., 2004) and onset of T1D occurs in adolescence in 70% of cases (UK data; (National Paediatric Diabetes Audit & Royal College of Paediatrics and Child Health, 2014). The audit also reported that adolescents have more emergency hospital admissions and have poorer symptom control than younger children or adults (National Paediatric Diabetes Audit & Royal College of Paediatrics and Child Health, 2014). The underlying reasons for the differences between adolescents compared to children and adults can only be speculative. For many, this period involves transition from paediatric to adult services, which can impact adolescents in many ways, for example, the expectations to transfer the responsibility and management of the LTC from parent to adolescent may impact the overall management of the condition.

In England, the prevalence of adolescents with an LTC was reported to be one in seven in those aged 11 to 15 years (Brooks et al., 2011). A survey collected in 2010 determined 15% of 4383 adolescents aged 11, 13, or 15 reported a diagnosis of an LTC, and more than half (66%) of those with an LTC took medication (Brooks et al., 2011); the study also stated that the proportion of girls aged 15 years reporting that their condition affects school attendance and participation was higher than boys of the same age (43% of girls, 28% boys).
In the UK, the most common conditions in adolescence are asthma, diabetes, epilepsy and cancer (Brooks et al., 2011). Disability is also prevalent within this age group; the term disability can be defined as a general impairment including physical and mental impairments that have long-term negative effects on everyday life and activities (Brooks et al., 2011). The highest reported underlying causes of disability are long-term health conditions, such as arthritis, and chronic pain (Vine et al., 2011). These two conditions, JIA and chronic pain, will be described further in Section 1.3.1 and 1.3.2 respectively. As shown in Figure 1, for adolescents aged between 11 and 15, disability and LTCs are possible barriers to everyday life as reported by parental proxy (Vine et al., 2011). These everyday activities are presented in Figure 2 showing that adolescents with a debilitating condition experience more restrictions than healthy adolescents.
1.3.1. Juvenile Idiopathic Arthritis (JIA)

Although healthy children and adolescents can experience musculoskeletal pain without associated disease (King et al., 2011), the most common cause of chronic musculoskeletal pain in adolescents is JIA (Schanberg et al., 2005). The incidence of JIA in the UK is approximately 1 in 10,000 per annum (Symmons et al., 1996).

JIA is an umbrella term for seven heterogeneous subtypes of childhood chronic inflammatory arthritis. Arthritis is defined as joint swelling, or limited movement due to joint pain, or tenderness, which persist for a minimum of six weeks (Jordan & McDonagh, 2006; Tattersall & Rangaraj, 2008). The current definition for JIA is derived from the International League of Associations for Rheumatology (ILAR; Petty et al., 2004). ILAR states that JIA is “arthritis of unknown etiology that begins before the 16th birthday and persists for at least 6 weeks; other known conditions are excluded” (Petty et al., 2004). Table 1 presents the classifications of the seven heterogeneous subtypes as identified by ILAR (Petty et al., 2004).
Table 1: Classification of the seven heterogeneous subtypes of JIA

Classification of the subtypes of JIA and the definitions adapted from Petty et al., 2004.

<table>
<thead>
<tr>
<th>Classification</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Systemic arthritis</td>
<td>• Arthritis in one or more joints</td>
</tr>
<tr>
<td></td>
<td>• might be accompanied with fever and rash</td>
</tr>
<tr>
<td>Oligoarthritis – persistent</td>
<td>• Arthritis affects 1-4 joints during first 6 months</td>
</tr>
<tr>
<td></td>
<td>• Continues with only up 4 for the course of disease</td>
</tr>
<tr>
<td>Oligoarthritis – extended</td>
<td>• Arthritis affects 1-4 joints during first 6 months</td>
</tr>
<tr>
<td></td>
<td>• Continues with more than 4 joints for the course of the disease</td>
</tr>
<tr>
<td>Polyarthritis – RF negative</td>
<td>• Arthritis affects 5 or more joints during first 6 months</td>
</tr>
<tr>
<td></td>
<td>• Test for RF is negative</td>
</tr>
<tr>
<td>Polyarthritis – RF positive</td>
<td>• Arthritis affects 5 or more joints during first 6 months</td>
</tr>
<tr>
<td></td>
<td>• 2 or more RF tests are positive</td>
</tr>
<tr>
<td>Enthesitis related arthritis</td>
<td>• Arthritis and enthesitis</td>
</tr>
<tr>
<td></td>
<td>• Or Enthesitis with at least 2 clinical signs of arthritis</td>
</tr>
<tr>
<td>Psoriatic arthritis</td>
<td>• Arthritis and psoriasis in patient</td>
</tr>
<tr>
<td></td>
<td>• or family history of psoriasis and/or clinical signs</td>
</tr>
</tbody>
</table>

Treatment for an LTC such as JIA involves managing and monitoring the symptoms of disease. The most common symptom in JIA is pain (Kimura & Walco, 2006). A recent thematic synthesis of qualitative studies exploring children’s experiences of living with JIA found that of the 542 participants one of the major themes to emerge was that there is unrelenting and unpredictable pain, and there is a strive for normality (Tong et al., 2012). In work exploring how children and adolescents express their own pain, the research reported that the young people utilise both behaviour and language to express their symptoms of pain. For example, in a study conducting interviews to investigate developmental differences between 6–11 year olds and 12–17 year olds, all participants used “aching” to describe their joints and described the pain as “burning” and/or “sharp” (Beales et al., 1983b). However, differences were noted between the two age groups with the older group more likely to rate their pain above the midpoint on a Visual Analogue Scale (VAS) and considered pain to be a reminder of the disease (Beales et al., 1983b).
Another study which used behavioural observational methods to assess pain behaviours (e.g. guarding, bracing or passive rubbing) demonstrated that children (aged between 6 and 17 years) exhibit similar pain behaviours as adults with rheumatoid arthritis (RA; Jaworski et al., 1995). This study indicates that children and adolescents express the symptom of pain and in a similar method as adults.

In order to validate reported pain in JIA patients, visual analogue scale (VAS) ratings (0-10 cm) were taken from 101 parents and children over 5 years old (Ross et al., 1989); the study found that the highest ratings for pain were in those with polyarticular disease with a mean score of 5.0, followed by oligoarthritis with a score of 2.1, and systemic disease with 1.8. Other symptoms, such as the presence of joint inflammation and morning stiffness, were associated with pain (Ross et al., 1989). The authors concluded that regardless of age, self-reported pain is a valid indicator of clinical disease severity (Ross et al., 1989). However, it is unclear whether the relationship between disease severity and pain is bidirectional. A study by Schanberg et al (2003) was the first to utilise daily diaries. For a two month period, 41 children, (aged 8-18 years, the majority (24 children) with polyarticular arthritis), completed a diary for daily symptoms (pain, fatigue, stiffness) and daily function. Out of the 2,095 diary days recorded, 1,533 of those days were recorded as pain days (73%; Schanberg et al., 2003). Sixteen participants reported experiencing pain every day (Schanberg et al., 2003). After examining the behavioural patterns documented in the daily diaries, the authors also observed a decrease in physical activity on days where participants experienced stiffness and/or pain (Schanberg et al., 2003). In a sample of 125 adolescents with JIA (aged between 10.3 and 17.8 years), pain rather than fatigue predicted a decrease in attending physical education classes (Sallfors et al., 2004). This link between pain which occurs on an almost daily basis, and decrease in physical activity may also account for research that reported that in inflammatory diseases such as JIA, pain has been described as the most likely cause of disability (Kimura & Walco, 2006).

Pain related to JIA is found to increase with disease activity index, which is a global assessment of the disease activity provided by the rheumatologist after a physical activity (i.e. moderate disease category had a mean pain intensity score of 4.81 while mild disease category had a mean pain intensity score of 3.90; bu-Saad & Uiterwijk, 1995). However, disease severity plays a minor role in accounting for pain variance (Stinson et al., 2012);
for example, a study investigating predictors of pain intensity found that the degree of joint inflammation accounted for only 10% of pain (Ilowite et al., 1992). This suggests that inflammation is not the only contributing factor to pain. One factor that could influence pain perception is disease duration. In an experimental pain study, there were differences in both pain coping strategies and pain responses in 16 patients with JIA and 14 healthy controls (Thastum et al., 2001); for example, a significant difference was found in participants using behavioural distraction as a pain coping strategy (patients 2.8 ± 1.0, healthy children 2.5 ± 0.7; p<0.02). Disease duration was strongly correlated with pain intensity (r = -0.70, p<0.01) and pain discomfort (r= -0.85, p<0.01) when controlling for age, and those patients who reported longer disease duration reported a lower degree of experimental pain (pain measured by a cold pressor pain test) and higher degree of clinical pain (pain related to their JIA; Thastum et al., 2001). Similar results demonstrated disease duration was positively correlated with self-reported pain (rho= -0.28 ; p< 0.05) in 37 participants aged between 7 to 17 years (Hagglund et al., 1995). These studies suggest that there is a need for a further understanding of pain mechanisms to map all contributing factors to pain perceptions.

1.3.2. Chronic Pain

Chronic pain, is usually defined as pain in the absence of diagnosed disease activity lasting for longer than three months and considered a disease in its own right, although the term also applied to pain which occurs in the context of a long-term condition, such as JIA. The International Association for the Study of Pain (IASP) currently define pain as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage or described in terms of such damage” (Loeser & Treede, 2008; Merskey & Bogduk, 1994). This definition of pain recognises pain as an individual experience, and includes the interpretation and an association of emotional and sensory information with the impaired tissue. This definition also encompasses pain, which may arise without any discernible underlying pathology (potential tissue damage). The following section outlines four models that have been applied with adolescent cohorts. These four models, which explore different pathways, have been used to study pain experiences.
1.3.2.1. Models of Pain

As suggested by the definition above pain is recognised as a subjective experience which results from the interaction between physiological and psychological processes as well as the psychological and social contexts in which the pain occurs. Taking a biopsychosocial approach improves our understanding of the interactions between these different components and how they influence perceived pain intensity (Rapoff & Lindsley, 2000).

1.3.2.1.1. Biopsychosocial Approaches

The following pain models incorporate physiological, psychological and social aspects of pain experiences and try to take account of the ways in which each of them interact with one another to create pain experiences (Gatchel, et al 2004; 2007). Biopsychosocial approaches recognise that potentially painful stimuli produce responses from a number of somatic systems (the autonomic, endocrine and immune systems). However, the main difference between the general biopsychosocial model proposed by advocates such as Gatchel et al, (2007) and earlier biomedical models of pain is the recognition of the influence of the psychological and social context in which the pain occurs (Gatchel et al, 2007). These influences act on both pain pathways and processes, as well as influencing emotional, cognitive and behavioural reactions to pain. Importantly, an individual’s genetic predisposition is seen as influencing all components. This multiplicity of influences helps to explain the wide variation in pain responses between as well as within individuals exposed to similar pain triggers.

Biopsychosocial pain models identify different biological processes which can contribute to the pain experience (Gatchel et al, 2007). The pain models explored below place slightly different emphasis on different biological mechanisms that contribute to either the pain sensation or the interpretation of pain processes, for example such as neurological emphasis in the neuromatrix model or a physiological emphasis in the fear-avoidance model.

What the biopsychosocial models have in common is the recognition that the stimulation of the nerves, the nociception, is only one process or dimension of the pain experience and biopsychosocial approaches aim to take account of the many additional dimensions.
including the emotional and behavioural responses (suffering) which may feedback directly to processes involved in the pain experience and the response (pain behaviour) (Loeser, 1982).

As indicated by the IASP pain definition (Loeser & Treede, 2008; Merskey & Bogduk, 1994), and by the current pain models in the literature both the cognitive and affective processes play an important role in the pain experience. Attention and vigilance to the threat that pain signals are key parts of pain processing (Eccleston and Crombez, 1999) and both are linked to levels of emotional distress. Cognitive processes such as hypervigilance, anticipation and attentional bias are related to worry and fear as indicated below in the diathesis-stress model. These processes are accounted for within cognitive and affective models such as the fear-avoidance model and described in more detail in the diathesis-stress model. A recent narrative review by Crombez and colleagues argued that motivational goals and values need to be taken into account in the fear-avoidance model (Crombez et al, 2012). A limitation of these cognitive and affective models is the omission of other cognitive processes such as personal control and self-efficacy. Research has found that these processes play an important role in procedural paediatric pain (Page & Blanchette, 2009) but are not assessed in paediatric patients with chronic pain.

There have been specific models developed for the particular context of children and adolescents with chronic pain. For example, Palermo and Chambers (2005) developed an integrative conceptual model which focused on psychological and social aspects, specifically immediate family factors such as operant-behavioural interactions in parent-child dyads. Palermo later (2012) expanded on the model to take account of other factors such as cultural norms or environmental contexts, and changing biology. Palermo identified important influences on adolescents pain experiences including physical health, sex, pubertal development, pain modulation, health habits such as sleep, smoking, physical activities and health beliefs as well as including additional social factors such as socioeconomic status, school environment and peer interactions. As with research undertaken with adults, there are bidirectional relationships postulated between all the relevant factors. The inclusion of health habits in the conceptual model is a key difference when compared to models developed for an adult populations. The reason why health habits are included in this model specific for adolescents, is because these health habits are
mainly established during adolescence and are predictive of behavioural responses to pain (Palermo 2012; Palermo, Valrie, Karlson, 2014) however components of the model have not been fully researched.

This new conceptual model developed by Palermo (2012) highlights key issues in chronic pain research that are specific to adolescents and help us to recognise that current models of chronic pain developed for adult populations are inadequate for addressing chronic and relapsing pain conditions in adolescents. The current cognitive and affective models (such as the diathesis-stress model described below) do not account for the cognitive developmental status of the child, nor for example, how in children and adolescents catastrophizing thoughts may be an extension of worry (Eccleston, Fisher, Vervoot et al, 2012; Eccleston and Crombez, 2007). The conceptual model developed by Palermo (2012) is the first model which attempts to integrate developmental factors in a biopsychosocial approach. As stated in the unified theory of development (Sameroff, 2010) biological, psychological and social factors are crucial at all stages of development but the impact of each differs across each phase. The manner in which these different processes may be influencing an adolescent’s pain perceptions has not yet been explored.

1.3.2.1.2. Gate Control Theory

The gate control theory (GCT) incorporates both the physiological and psychological aspects of pain and was the first theory to challenge early traditional models which focused on pathology solely driving pain, and which included the presence of a lesion and defined pain intensity by lesion size (Melzack & Wall, 1965). According to GCT, specific fibres and nerve endings are evoked through the stimulation of the skin. The fibres respond to different sensory modalities and carry different information of either intense sharp pain, (A-Delta fibres), or dull, throbbing pain, (C fibres). The GCT suggests a central processing centre inputs to brain areas processing attention, emotion, and memories/experiences of pain (Melzack & Casey, 1968). This influences the type and the level of input that reaches the higher processing centres which in turn directly affects the perception of an experience as ‘painful’ thereby modulating the overall pain experience. The GCT addressed many aspects of pain phenomena that were previously unaccounted for. This was ground-
breaking and subsequently led to the emergence of new approaches to research and management of pain.

1.3.2.1.3. Neuromatrix of Pain Theory

Whilst the GCT explained the dynamic nature and complex interrelationships between physiological and psychological pathways in pain experience, it did not account for all pain observations. For example, in the phantom limb phenomenon, there is no physiological input or an existing network, yet individuals still experience sensations as emerging from the missing body part. This phenomenon suggests that pain perceptions do not require a concrete sensory input to trigger a pain experience. This would also suggest that there are patterns in the neural networks in the brain that are being stimulated beyond the peripheral and spinal processes. These patterns in the neural networks bridge the connection between the bodily stimulation and pain perceptions (Melzack, 2001). Melzack (2001) proposed an adapted model, the neuromatrix (sometimes called the ‘pain matrix’; Iannetti and Mouraux, 2010) which is a large distributed brain network which responds to, inputs from the cognitive, sensory and emotions related brain areas or systems, and then results in outputs that produce pain perception, action programs, and stress-regulation. According to this way of thinking there is no ‘pain centre’ in the brain, but rather a complex system distributed over different areas of the brain.

Although the neuromatrix approach has been viewed as largely helpful in moving forward our approach to understanding pain, there have been a number of criticisms. For example, it is hard to determine precisely which brain centres and systems form the neuromatrix because of the large number of brain regions which may potentially be influencing pain perception (Tracey & Mantyh 2007). A recent commentary criticised the neuromatrix model because the processes described do not account for the most fundamental and primal aspect of pain, that is the basic feeling that something hurts, the “ouch”, referred to by the authors as a “pain switch” (Davis, Kucyi, & Moayedi, 2015). The authors who proposed the pain switch, argue that the switch is a response to perceived environmental demands, the perception of which is itself influenced by individual cognitive and emotional factors (Kucyi & Davis, 2015). This criticism reflects recent shifts in the literature toward models
which account for a salience detection mechanism involving multiple systems identified via brain imaging (Canavero and Bonicalzi, 2015; Legrain et al, 2011; Kucyi & Davis, 2015). This criticism also reflects the observation that different areas of the brain play different roles depending on the interplay of the dimensions underlying pain perception (Tracey & Mantyh, 2007).

These criticisms of the neuromatrix model suggest that there is a need to examine and assess the pain experience as a whole to find how a potentially painful stimulus does become salient. This will involve an exploration of cognitive and emotional factors involved but also the context and the environment in which the pain experience occurs. Therefore, when researching chronic pain experiences with adolescents there is a need to use developmentally appropriate measures of pain cognition and pain emotions.

1.3.2.1.4. Diathesis-stress model of chronic pain

A model that was developed to recognise the interactions of physiological, psychological and sociocultural factors is the Diathesis-stress model (Flor et al., 1990). The Diathesis-stress model is considered to be an integrated model of fear avoidance models (Vlaeyen & Linton, 2000) and biopsychosocial models to account for chronic pain behaviours (Asmundson & Wright, 2004). The basis of the Diathesis-stress model posits that people with an underlying vulnerability or predisposition to developing chronic pain can have reduced thresholds for nociceptive activation. This vulnerability can result from genetic susceptibility, previous trauma, social learning experiences, or possibly a combination of these three factors (Flor et al., 1990). Flor et al, (1990) suggest that cognitive processes which influence a state of vulnerability are sensitive to learning processes; these may result in fear of activity through conditioning due to reinforced pain behaviours and physiological responses (see Figure 3). An interaction between this vulnerability (such as reduced threshold or anxiety) and environmental stressors or triggers, leading to learnt and reinforced behaviour is the proposed pathway to chronic pain (Flor et al., 1990; Asmundson & Wright, 2004).
1.3.2.1.5. **Cognitive-Behavioural Models**

The cognitive-behavioural model emphasises the influence of cognitive factors, such as attention, memory, avoidance, and conditional reinforcement on chronic pain. This model proposes that people are active processors of information and they learn to anticipate and predict outcomes and prepare for them (Turk et al., 1983) Newton and Barbaree (1987) argued that thoughts, feelings and behaviour are all connected and can intensify the pain experience. This model incorporates the standpoint that a person’s response to an illness is driven by their representation of that illness and its symptoms (Nerenz & Leventhal, 1983; Turk et al., 1986).

1.3.2.2. **Translation of models into pain interventions**

Biopsychosocial approaches to pain that include social, physiological and psychological mechanisms to understanding pain have encouraged the development of holistic pain management programs. This requires treatment plans to include medication, physiotherapy and/or psychological treatments to empower the young person. The models presented
above, describe pain as a multidimensional concept that has translated into treating pain with both pharmacological and non-pharmacological approaches (Asmundson & Wright, 2004).

Although there are few non-pharmacological treatments for adolescents with pain related to JIA, two systematic reviews have suggested that children with chronic pain can benefit from Cognitive Behavioural Therapy (CBT; Eccleston et al., 2002; Eccleston et al., 2009). Children can benefit from CBT for juvenile chronic pain because the treatment entails the discussion of coping strategies to deal with pain. This can include relaxation techniques and psycho-educational aspects such as learning about alternatives to “all or nothing” behaviour, alternative behaviours such as pacing activities, problem solving, and distraction from the pain (Eccleston et al., 2003). A randomised control trial (RCT) with 48 adolescents aged between 11 to 17 year olds, showed that CBT for juvenile chronic pain patients significantly reduced pain intensity (Palermo et al., 2009).

Coping strategies are hypothesised to maintain pain and possibly account for pain variance (Thastum & Herlin, 2011). Research has highlighted that adolescents who experience pain and a negative impact of pain may be at risk of engaging in maladaptive coping strategies (Thastum et al., 2005). These maladaptive strategies may have implications for levels of pain and can result in higher disability (Varni et al., 1996; Meijer et al., 2002). However, despite attempts at defining adaptive and maladaptive coping strategies, there are still discrepancies found in the literature; for example, Varni (1996) argued that the coping strategy of ‘seeking help’ is maladaptive, whilst Meijer (2002) considered this strategy to be adaptive.

Similar concepts that are targeted by CBT treatment, such as psycho-education, relaxation and pacing activities, have been adapted for a self-management intervention for JIA patients (Stinson et al., 2010). The feasibility trial of this self-management intervention for JIA found that the sample of 13 adolescents aged between 9 and 18 years felt empowered by the information provided in the self-management programme. In a qualitative study preceding the feasibility trial, these adolescents were asked about what they would need to self-manage their JIA (Stinson et al., 2008c). For these adolescents, gaining control over their arthritis was a key aspect for them in self-managing their arthritis; they also felt they
required greater knowledge about and understanding of JIA in order to achieve a greater control over their arthritis (Stinson et al., 2008a). This finding was not a novel conclusion as similar findings were reported from a focus group study regarding psycho-educational interventions (Barlow et al., 1999). These studies highlight the importance of adolescents’ conceptualisation of their condition and the need to assess and provide information about the condition and self-management techniques that can empower the adolescents.

1.3.3. Adolescents’ self-management of long-term conditions

Successful self-management of an LCT includes both psychological and medical adjustment. Psychological adjustment is the ability to identify requirements to modify goals. This type of adjustment is dependent on cognitive development, as psychological adjustment requires both cognitive and behavioural attempts to deal with or manage external/internal demands that are appraised as a personal threat (Lazarus & Folkman, 1984). Definitions of successful functioning depend on how these concepts, both medical and psychological adjustments, are measured. One method of measuring adolescents’ function is by comparing an adolescent with an LCT with that of a healthy adolescent. An alternative approach to measuring adjustment is to undertake a comparison within the population of adolescents with the same chronic condition. This section describes and presents the literature about coping and the factors influencing adjustment to living with an LCT.

In JIA, indicators of positive outcomes include high physical activity, low disability index, and low pain intensity amongst self-management and treatment adherence. These indicators can be measured by the Child Health Assessment Questionnaire (CHAQ, Singh et al., 1994) which is now routinely used in paediatric rheumatology clinics in the UK. The CHAQ provides a disability index and pain intensity score. Behaviour and outcome have also been measured using the Child Health Questionnaire (CHQ), which provides two composite scores; one for physical state and one for psychosocial state (Landgraf et al., 1996). There are different versions of the CHQ dependent on who is completing the questionnaire; there is a version for parents based on their own observations of their child’s behaviour and a version for children completed as a self-report measure. However, the
parent’s version is biased towards the parents’ own experiences and beliefs and may not truly reflect that of the child (Landgraf et al., 1996).

These questionnaires aim to provide comparative scores to measure adjustment. For example, the CHQ is transformed into a Z-score to be compared to a standard group. The domains in the CHAQ are added together to provide a total score between 0 and 3. The CHAQ pain intensity score is less comparable between patients as this is measured using a Visual Analogue Scale (VAS). The VAS in the CHAQ is a 100 mm line along which a participant indicates the amount of pain they are experiencing, where 0 represents the verbal anchor point of no pain and 100 represents the anchor point of worst pain they have ever felt (Singh et al., 1994). The issues with utilising a VAS as a method of pain assessment are well documented, with one key issue being the anchor points. Anchor points make the VAS a subjective item to complete as it is dependent on an individual’s previous experience and this information is not captured. As such, a score for one person may not reflect the same experience in another person providing the same score. Therefore, an adolescent’s previous experience influences the levels of adjustment reported in the CHAQ or CHQ. The adolescents’ behaviour, and consequently their adjustment to an LTC, is dependent on their understanding or conceptualisation of their LTC.

1.3.3.1. Impact of a long-term condition

An LTC impacts many areas that contribute to development during adolescence, including education, cognitive function, emotional well-being, and relationships with peers and family. How much of an impact an LTC has on each of these areas varies between different LTCs and is reviewed in the following sections.

1.3.3.1.1. Education

The literature supports the theory that adolescents’ education can be affected by their LTC due to absence from school (Logan et al., 2008b); school attendance is lower in adolescents with LTCs (such as JIA or chronic pain) compared with healthy adolescents. Out of 20
days, 220 adolescents (aged between 12 and 17 years) had a mean of 4.5 full days absent from school (Logan et al., 2008b). Adolescents with an LTC are more likely to be absent from school due to illness severity, treatment adherence or psychological problems (Sturge et al., 1997). Therefore, it is possible that absenteeism may be the underlying cause of poorer educational qualifications in adolescents with an LTC rather than cognitive impairment.

1.3.3.1.2. Family and Peer Support

It is expected that the relationship in a parent-child dyad changes during adolescence (Sroufe & Cooper, 1988). However, for adolescents with an LTC, this change can be delayed (Palermo et al., 2014). A review showed that adolescents who experience chronic pain were more likely to depend on their parents or main carer for a longer period of time than healthy adolescents, and were also more likely to have tendencies to isolate themselves (Palermo, 2000). These adolescents consequently suffer from restricted opportunities for social development. One study of 110 adolescents (aged between 11 and 18 years) with chronic pain found that 90% of the sample perceived themselves as behind in at least one aspect of social development (Eccleston et al., 2008).

Adolescents experiencing severe pain can also experience isolation, negative self-perception, low self-esteem, and being dependent on their parents for longer. These factors could have implications for that young person’s emotional response to their health issue. Eccleston and colleagues (2008) found that pain intensity, depression and anxiety, predicted those adolescents who self-perceived themselves to be less independent, less emotionally adjusted and less developed in self-identity compared to their peers. These findings suggest that adolescents experiencing pain are not only a particularly vulnerable group in comparison to other adolescents with LTCs, but also have negative perceptions of self.

Negative impact on peer relationships is thought to occur due to missed opportunities to cultivate these relationships. However, work by Graetz and Shute (1995) showed that adolescents with an LTC, such as asthma, were not necessarily affected in terms of their
friendships at school; in this study, 21 students aged 8 to 18 years were matched with healthy control classmates, and found that there were no differences in the groups regarding the amount of friends, popularity, loneliness, or rates of rejection. These results might be dependent on the type of condition as the literature on the impact of LTCs on peer relationships provides mixed conclusions, particularly in adolescents with JIA. A quantitative study of 74 adolescents with JIA demonstrated that the severity of the disease did not have any impact on peer ratings; both measures were completed by the parents and by the children as a self-reported self-perception profile (Noll et al., 2000). However, in a qualitative exploration of this issue, Sallfors and colleagues (2002) found that adolescents with JIA felt they were met with disbelief at school, especially when the pain was fluctuating. This was apparent through their varied levels of activities, for example, ranging from joining in activities, to using crutches (Sallfors et al., 2004). However, this group were recruited through a JIA peer group, so it is unclear if this group and the relationships formed within it may have influenced the peer relationships at school. The key aspect of these findings relate to how the adolescents perceive their social support rather than the provision of support (Tak & McCubbin, 2002).

1.3.3.1.3. Cognitive Functioning

There is little research investigating the impact of LTCs on the development of abstract thinking capabilities and cognitive functioning in adolescence (Suris et al., 2004). However, there is research that has found that cognitive developmental naiveté impacts the management of an LTC (Suris et al., 2004). For example, cognitive developmental naiveté, such as lack of abstract thinking, can possibly lead to an inability to adhere to a treatment regime; this may be due to an inability to plan and imagine future consequences. An example of the impact of altered cognitive development on compliance was demonstrated in a study of 144 adolescents; beliefs that a treatment for diabetes was not necessary led to 25% of adolescents reporting not taking the insulin or providing the required blood tests to manage diabetes (Weissberg-Benchell et al., 1995). Arguably, an adolescent’s health beliefs or goals are not assessed regularly, but discrepancies between priorities and beliefs of adolescents and those of health professionals may account for impaired adherence (Suris et al., 2004). Furthermore, there is a hypothesis that adolescents who have a greater
understanding of their LTC will report better self-management and positive outcomes; there is evidence to suggest that adolescents with uncomplicated epilepsy who report a higher sense of coherence also report lower levels of illness severity (Räty et al., 2004). This suggests that beliefs and conceptualisation of an LTC is an important aspect to assess and address in treatment plans. These findings also suggest that the regular assessment of beliefs and goals related to the understanding of the conceptualisation of an LTC is vital.

1.3.3.1.4. Self-esteem and emotional wellbeing

Unlike the reported impact of an LTC on cognitive development, there is evidence to suggest there is an impact on identity, both in the development of a self-concept and self-esteem (Hauser et al., 1983). Research focusing on self-image has demonstrated that higher body dissatisfaction is reported in adolescents with chronic illnesses (Bucchianeri et al., 2013). Furthermore, the literature exploring pain and self-esteem shows that pain can interfere with an individual’s self-image; one study found that in 115 adolescents aged between 13 and 18 years, low self-esteem alongside negative self-perception may play a role in mediating pain intensity and functional disability (Guite et al., 2007). Research has yet to explain this relationship between pain intensity and self-esteem.

Issues with self-esteem and self-image may impact compliance with treatment, as demonstrated in adolescents with diabetes with treatment regimens requiring injections during lunchtime; the adolescents were not compliant as they prioritised their social identity over their treatment (Swift, 1997). The adolescents’ low self-esteem and negative perceptions of the self can lead to further isolation from peer groups impacting those adolescents’ social development (Kyngäs et al., 2000). In a study of adherence, those adolescents who viewed their LTC, such as diabetes or JIA, as a threat to their emotional wellbeing were more likely to report poorer adherence (Kyngäs et al., 2000).

Emotional wellbeing is reported to be poorer in those adolescents with an LTC compared to their healthy peers. This was evident in the Adolescent Health Survey of 1683 adolescents (grades 7 to 12), with those with an LTC reporting lower emotional wellbeing scores (Wolman et al., 1994); reporting of anxiety and depression were also higher in those
adolescents with an LTC. These results suggest that the struggle of establishing an identity that incorporates the LTC is emotionally taxing for adolescents. This emotional wellbeing appears to be related to the conceptualisation of the LTC; for example, research with adolescents with diabetes showed that those who reported a higher sense of coherence had higher self-esteem, better self-management and fewer emotional and social problems (Luyckx et al., 2014a).

1.4. Development during adolescence

LTCs and disabilities with the onset occurring during childhood or adolescence have the potential to impair the development of a young person. Pain and physical limitations can hinder or delay the achievement of the developmental milestones. Evidence of impaired development was highlighted in a recent meta-analysis of 195 studies that compared healthy young adults with young adults who had paediatric LTCs or disabilities (Pinquart, 2014). Lower numbers of those in the latter group reported achieving developmental milestones in comparison to the matched controls.

The development of autonomy from parents and social identity occurs during adolescence (Sroufe & Cooper, 1988); despite an increase in autonomy from parents being an expected outcome of adolescence in most cultures, it is unclear how individuality and autonomy occurs. One possibility is the theory of individuality and connectedness, where the transition and process of self-assertion occurs simultaneously and is in balance with connectedness of self with others (Grotevant & Cooper, 1986); this interplay between individuality and connectedness helps redefine the relationship between parents and the adolescent. Furthermore, this interplay is related to both the adolescent’s development of self-identity and the definition of their role (Grotevant & Cooper, 1985). This theory also proposes a continuity hypothesis; the development of autonomy and changes in interaction occurs within the context of other factors, such as parental styles (Baumrind, 1991) and attachment (Sroufe, 1996).

The earlier studies within this framework of individuality and connectedness found that individuality can be predicted by the interaction and communication styles within a parent-
child dyad (Grotevant & Cooper, 1986). Importantly, the parents hold expectations for their child to develop independence while also having their own perceptions and beliefs. This can influence or change the parent-child dyads’ interaction, while also providing the adolescent the opportunity to develop individuality (Grotevant & Cooper, 1986). A study utilising the Experience Sampling Method (ESM) recorded daily interactions and activities of 220 adolescents (Larson et al., 1996). Although there was a decrease in participation for overall family activities between the ages of 10 and 18, the amount of time spent one-to-one with the mother/father stays relatively the same (Larson et al., 1996). This supports the concept that the changes are occurring due to interaction within the parent-child dyad rather than the amount of time separated.

While adolescents are redefining their role with their parents, they are also continuing to further construct their social identity. This construction is dependent on the interaction with their peers, and the association with larger social groups (Brown et al., 1986). Interpersonal identity development is reliant on social relations and the identification of oneself with a group (Turner, 1982). For adolescents, inclusion in groups provides a context for developing attitudes and values without adult monitoring (Rubin et al., 2005). This allows for the change in the relationship with the parents (Dusek, 1991). Other aspects of an adolescent’s life are also influenced by peer relationships. For example, positive school adjustment (Ladd et al., 1997), lower levels of reported loneliness (Parker & Asher, 1993) and depression (Oldenburg & Kems, 1997) were all linked with higher levels of peer acceptance. Thereby, positive peer relationships are crucial to the development of emotional wellbeing and autonomy from the parents.

Changing goals from achieving the developmental milestones to goals about managing an illness can result in delayed independence from parents (Gledhill et al., 2000). One underlying reason for delayed independence is the shared responsibility in parent-child dyads in managing an LTC. The importance of the parents’ own goals and priorities about their child were demonstrated in a longitudinal study of adolescents with diabetes and their parents; the parents provided a list of general goals (e.g. “be their own person”) and diabetes-specific goals (e.g. “acquire the skills and ability to manage diabetes on own”) at baseline (Robinson et al., 2011). After 6 months, parent-child dyads completed questionnaires about who was responsible for the diabetes management activities (e.g.
“whose job is it in your family to see that it is done”). Those parents who prioritised diabetes-specific goals had greater involvement in and took more responsibility for the management of their child’s diabetes (Robinson et al., 2011). Therefore, this ‘power struggle’ for the management of an LTC, which is dependent on both the parent and the child’s beliefs and goals, can delay the expected parent-child dynamic change during adolescence.

1.4.1. Cognitive development

In the section below, three developmental approaches and perspectives of the cognitive developments in adolescence are reviewed; these are the Piagetian approach, the Vygotskian approach, and the Social Learning approach. This is followed by a description of how these approaches are applied to adolescents’ conceptualisations of LTCs. Both the Piagetian and Vygotskian approaches relate to the internal developments involved in developing understanding of illness, while, the Social Learning approach takes into account the external influences on understanding. However, to date there is no established theory on how adolescents conceptualise either LTCs or acute conditions.

1.4.1.1. Piagetian approach

The Piagetian theory is based on the idea that changes in children’s cognitive processes can be mapped to five different stages; sensorimotor, preconceptual, intuitive thought, concrete operations, and formal operations (Piaget, 1962). Each of these stages which can be characterised by specific types of thought processes. These five stages fall under what Piaget (1962) described as three overarching phases: sensorimotor development (0-2 years), concrete operations (2-11 years) and formal operations (11 - onwards). Development within these phases is viewed as continuous but Piaget argued that all children experience the same sequential progression. During adolescence there is a transition between two stages of cognitive growth; the concrete operational stage and the formal operation stage. According to Piaget (Piaget, 1969), cognitive development is a continuous process that begins from the interaction of cognitive processes with the environment. The premise of this process is that knowledge evolves from action. Development proceeds from previous learning; each new phase has roots from the previous phase and precursors for the
subsequent phase. This progression is completed once equilibrium is achieved. Equilibrium involves feedback, forecasting, and trying to achieve a coherent understanding. A new experience can introduce novel information, which requires fitting in with personal conception of events. This new information also needs to fit with previous information which then can lead to new thinking to achieve coherence of new and old information. This requires adaptation defined by Piaget (1971) as an ability to process the interaction between environment and personal experiences. This definition of adaptation involves two processes; internal which allows a child to understand new experiences through assimilation into existing cognitive schema, or external which requires existing schema be modified to incorporate new experiences (i.e. accommodation). These processes lead to a coherent understanding of the world to reach cognitive equilibrium. This sequence of development creates a hierarchy of cognitive experiences obtaining increasingly complex cognitive structures by which children, adolescents, and adults make sense of their experiences (Piaget, 1969).

The phase of formal operations is the last phase of cognitive development (ages 11-15). In these stages the focus is on acquiring a complex and comprehensive mental structure that is beyond a child’s own experiences and belief systems (Piaget, 1962). This phase has been described as a move to more abstract thinking. At this phase a child can begin to acquire the ability to understand ideas and the existence of different perspectives following from understanding a physical and a social world. Therefore, an adolescent begins to understand and conceptualise ideas that are built from propositions and hypothetical reasoning.

According to the Piagetian approach, adolescence is defined by the acquisition of the complex thinking that leads to equilibration (Piaget & Inhelder, 1958). This occurs once an adolescent accomplishes and acquires the ability to deduct from experience as well as thought. Thus, equilibration is also the state where an adolescent can utilise abstract and concrete cognitive constructions to envisage propositional operations (for example; if-then, either-or, or neither thoughts and constructs). Reason, logic and the ability to deduce from hypothetical situations are all associated with this developmental phase (Piaget, 1969).
1.4.1.1.1. Developmental approach to conceptualising illness

The developmental approach to theorising how children and adolescents conceptualise illness corresponds with Piaget’s stages of cognitive development described in Section 1.4.1.1 above. As described previously there are two stages during adolescence; the concrete operational stage where children begin to think logically but are unable to think or reason in abstract thought, and the formal operational stage at which the adolescents begin to develop the abstract thought. The corresponding stage approach to the conceptualisation of illness follows a similar pattern.

The underlying premise is that development of an understanding of illness occurs through stages and can be mapped to a child’s age and stage of cognitive development. From the ages of 7 to 11, a young person would begin to think about symptoms and illnesses in terms of internal mechanisms such as the immune system or germs (Bibace & Walsh, 1980). A young person can, at this stage, internalise illness and rely on internal mechanisms when trying to provide a causal explanation. This reliance on internal mechanisms is evident with earlier work with children and adolescents when asked how they get a common cold (Siegal, 1988; Williams & Binnie, 2002; Siegal et al., 2011). Participants in this age group provided explanations about germs and germ transmission from other people (Siegal, 1988; Siegal et al., 2011). According to the developmental approach, and in correspondence to the development of abstract thought in the formal operational stage, young people at the ages upward of 12 begin to think in concrete and abstract form. This two-level analysis mechanism allows adolescents to become more aware of control over developing and curing illness (Bibace & Walsh, 1980). Participants in this age group provided biological rationale in their explanation of the common cold (Siegal, 1988; Siegal et al., 2011).

One criticism of this approach is that most studies that have utilised the developmental approach to study conceptualisation of illness recruited healthy samples. These studies tended to explore the understanding of the common cold (Siegal, 1988) or causal beliefs related to eating unhealthy/healthy food (Springer & Ruckel, 1992). There are some studies that have studied children with LTCs. An example of this was by Berry (1993) with 54 children aged between 6.75 to 17.4 years with juvenile arthritis. This data was analysed
according to the stage categories developed by Bibace and Walsch (1979). These categories of cognitive development were a better predictor of accuracy than the age of the child, suggesting to the authors that the stage influenced the understanding of the illness. Although this is a plausible argument, the research has failed to describe how a child transitions between stages. Despite this, the authors argued from these results that communication to a child should be dependent on the child’s understanding and experiences rather than their age. This leads to a further key criticism of the stage model such that it does not consider experience or social context of the child. This was exemplified in a study that used tools developed to assess stages (illness causation task, (Bibace & Walsh, 1980) according to a Piagetian framework, to compare healthy controls (matched ages) with children experiencing chronic illness (cystic fibrosis and cancer). The results showed that the experience of being ill had a facilitating effect on children’s understanding of an illness. Although due to lower scores in the younger children, the authors also concluded that age has some effect (Crisp et al., 1996).

1.4.1.2. Vygotskian approach

In contrast to the Piagetian approach, the Vygotskian approach focuses on the development and emergence of higher mental functions. Vygotsky (1987) argued that a child approaching logical thinking requires the ability to utilise introspection. Introspection, the ability to examine and realise internal operations (mental and emotional processes), is developed through the influence of social factors and problem-solving. The development of mastering introspection and logical thinking is guided by an inability to solve increasingly complex problems. Vygotsky (1982; 1987) proposed that children have access to tools of the mind which once utilised, their mental abilities are developed and these abilities also influence physical, social and emotional behaviours.

According to the Vygotskian approach, adolescence is defined as a transition period between lower psychobiological processes to higher conscious psychological functions. This transition and development is dependent on learning and what Vygotsky refers to as the zone of proximal development (ZPD), which has later been referred to as scaffolding the mastering of a new skill or concept (Vygotsky, 1987).
1.4.1.2.1. Functionalist approach to conceptualising illness

To account for this development of a more complex conceptualisation of illness, researchers argued that a child is a “theory builder” such that children build their information (Nelson, 1986). This argument incorporated the importance of the social environment and provides an interpretation for the development of more elaborate scripts (Nelson, 1999) and concept building (Carey, 1985). This approach corresponds with the ‘scaffolding’ theory proposed by Vygotsky (1987).

The premise of the functionalist approach is that children’s knowledge is relative to their experiences and stage of life (this can be distinguished by age). For example, concept building would develop as the result of a shift from intuitive to biological knowledge. This idea was supported by research that asked children to describe a doctor’s visit. There were differences in the descriptions from 8 year olds to 5 year olds, where the older children provided elaborate descriptions and representations of the routines in comparison to 5 year old participants (Eiser, 1989). This approach supported the argument that age is only representative of amount of experience and experience is the driving factor of a more coherent and elaborates the conceptualisation of an illness. How this experience is transformed to the conceptualisation of an illness remains unclear.

1.4.1.3. Social Cognitive Theory

The interaction between behaviour, cognitive and environment contribute to learning and motivation for the overall behaviour. Humans are perceived as agents of their own experiences, thereby the biological systems are tools to achieve goals and tasks they set (Bandura, 1997). Bandura argued that learning occurs through a combination of exposure and regulated motivation and activities (Bandura, 1986). Cognitive processes exert determinative influence on behaviour (Bandura, 1986). Cognitive processes and behaviours are not simply a reactive consequence from the environment and experience (Bandura, 1986; Bandura, 2001). The environment through forethought, pro-action, self-appraisal, and self-reflection shapes these cognitive processes (Bandura, 1986; Bandura,
1997; Bandura, 2001). According to Bandura (1986; 2001) these are key underlying concepts in regulating learning and development.

Learning and development are not solely dependent on internal cognitive process, but rather the cognitive process and underlying concepts are tools to aid the learning and development. Self-efficacy is the belief an individual has that they can complete a task or are empowered to complete the task (Bandura, 1997). Self-efficacy alongside observational learning leads to further development of knowledge and behaviours.

Knowledge and behaviours are learnt by observing through modelling, imitation, reinforcement, vicarious reinforcement, and identification (Bandura, 1986). These methods of learning are dependent on a model’s (individual that an adolescent would observe) behaviour and the attitudes that are encoded by the adolescent. The encoded behaviours and attitudes are then imitated by the adolescent, which are either positively or negatively reinforced by the model. This can lead to either the adolescent to continue partaking in the behaviour or else to change their behaviour. Vicarious reinforcement is defined as when the young people also observe the consequences other people have when performing the same behaviours, which contributes to the decision whether to use or not use the behaviours. Lastly, identification involves the process of internalising and adapting those observed behaviours because the young people identifies with the model.

These models can change over time, and can be parent(s), TV characters, friends or teachers. The underlying premise of this approach to learning and behaviours are that there is a social interaction regarding behaviours and attitudes that are reinforced and internalised. If a child or adolescent wants approval from their parents or a group (peers) this would be external reinforcement and that motivation to seek approval (external positive reinforcement) would determine the child/adolescent’s behaviours (Bandura, 1986).

1.4.1.3.1. Social learning approach to conceptualising illness

One explanation of learning from experiences is provided by the Social Cognitive Theory approach. Aspects of the social learning theory such as self-efficacy and reinforcement
have been integrated into health psychology and specifically used with chronic pain. Social learning and experience is thought to have a vital role in pain (Loeser & Melzack, 1999).

Psychological treatments such as cognitive behavioural therapy (CBT) aim to target illness behaviour. When in pain or if in sickness, people engage in activities or behaviours known as illness behaviour (Fordyce, 1988). It is possible that within a family dynamic these behaviours are encouraged through positive reinforcement which can influence the child’s social function (Palermo, 2000).

The literature suggests that children imitate illness behaviours from a significant person in their lives whom they see as a role model. A study on this subject was conducted by Osborne and colleagues, who interviewed 20 children (mean age = 10.3 years) and found that these children dealt with their unexplained pain (pain without an organic cause) in the same way as their role model (Osborne et al., 1989). These 20 children described location, intensity and frequency of the unexplained pain and their coping style. The descriptions provided by the children were identical to the descriptions of pain given by the child’s role model. These results suggest that an adolescent modelled their pain as well as their illness behaviour to a significant person in their environment. These findings support the social learning theory about learning through imitation, but also suggest the important role environment and parental influence may have on pain and dealing with pain.

Further work within the social cognitive framework found that self-efficacy is required for managing JIA (Barlow et al., 2001; Barlow et al., 2000). An adolescent’s wellbeing was associated with their self-efficacy (Barlow et al., 2001). In a separate study with children with JIA, a child’s function was associated with the self-efficacy of their parent (Barlow et al., 2000). Together these findings emphasise the argument that an adolescent’s self-perception and the beliefs of their parents might account for variations in experiences of pain.

1.4.1.4. Emerging approach: Illness representations approach

There is a relatively new approach to how children and adolescents conceptualise illness by building a mental representation. This approach was adapted from the adult literature the Common Sense Self-Regulatory model (CS-SRM; Leventhal et al., 1984). The illness
representations approach utilises this CS-SRM, and the theory is that an individual builds a representation of an illness based on what makes the most sense to that individual. The strength of the illness representations approach is the flexibility which allows it to incorporate the concepts of all the other approaches. The CS-SRM allows for the role of social learning and experience, specifically, giving an individual the approach to how an adolescent understands their illness. By using the CS-SRM as a framework for understanding how an adolescent conceptualises illness and pain, it takes into account the developmental progress of a child from adolescence to adulthood. This approach can also map the role of the parent, the experiences, and individual differences between children show a relationship between behaviours and cognitions and due to the model being self-regulatory, it allows for the exploration of targets to implement interventions. However, the adaptability of this approach for the younger population has not been explored.

Work using Piagetian tools to interview younger children about illness found that children (aged between 4-6 years) discuss beliefs in all the domains proposed by the CS-SRM as found in the literature (Goldman et al., 1991). This work had found that younger children did not hold sophisticated attributions to the cause of illness. These results support development in information, which can be accounted for the CS-SRM. A qualitative study using the CS-SRM administered to an older group of adolescents (aged 7-14 years old) with asthma and matched aged controls found that adolescents with asthma had a more sophisticated conceptualisation of asthma, but again not all domains were found in this study, for example, no differences were found in consequences (Paterson et al., 1999). Paterson and colleagues (1999) discussed these results with consideration of the exposure of asthma information to children who do not have the condition. Thereby, these results support using this framework to investigate the individual mental representation and conceptualisation of illness and pain for adolescents. Apart from these studies, to date little work has been conducted to assess the extent to which this approach can be used to account for adolescents’ understanding of symptoms and if it accurately maps processes involved in adolescents’ conceptualisation of their illness.
1.5. Summary

The World Health Organisation (WHO) defined adolescence as a transitional period between childhood and adulthood, which occurs between the ages of 10 and 19 years old. This period is highly significant in human development, and international and national policies have identified this stage as crucial in developing health behaviours that are maintained in adulthood. Therefore, adolescence is an ideal stage for interventions for those adolescents with LTCs.

There is a high prevalence of adolescents with long-term conditions in the UK and high incidences of onset occurring during this period. LTCs and disabilities have been found to hinder the achievement of developmental milestones when comparing adolescents who have an LTC to an adolescent without. Literature has also provided evidence that dimensions of development (cognitive, social, physical) are affected by LTCs. There is a bidirectional relationship between positive outcomes in an LTC and developmental issues.

The highest reported causes of disability in adolescents in the UK are long-term health conditions such as arthritis and chronic pain. Juvenile Idiopathic arthritis (JIA) is the most common cause of musculoskeletal pain in adolescents and is defined as joint swelling or limited movement due to joint pain of unknown aetiology. Self-management of JIA symptoms is both demanding and complex, not least because of the relapsing-remitting nature of symptoms including pain. Furthermore, the relationship between pain and disease activity is not straightforward, with disease activity only accounting for a small percentage of pain variance. Whilst it is acknowledged that cognitive and affective processes are central to pain experience, to date there has been little research that focuses on the specific cognitive and affective processes experienced by young people with JIA.

Measuring and defining self-management is usually done by comparing individuals within a population of adolescents with long-term conditions. However, there are individual differences in achieving developmental milestones and self-management, and these are informed by how an adolescent conceptualises their condition. There is a strong need for the development of a framework that is suitable to account for how adolescents conceptualise illness. There are different approaches to understanding how an adolescent
would conceptualise illness. At present, each approach has had their limitations and one promising approach is utilising illness representations, a framework not yet validated with this age group.
Chapter 2: Illness Representations and Illness Behaviour

2.1. Overview

This chapter provides an overview of the theoretical approach that was evaluated in this PhD. This theory provides the framework used to study cognitive representations of illness (illness representations) and their influence on people’s responses to a health threat (illness behaviours). This framework provides a theoretical system of decision-making in health with the corresponding cognitive and emotional processes involved. These underlying cognitive and emotional processes motivate the behaviours that manage illness and improve health.

The chapter will begin with the theoretical background of the development of the Common Sense Self-Regulatory Model (CS-SRM; Leventhal et al., 1980). This is followed by a critical evaluation of the theory and model, which addresses each of the three components of the model in turn:

i) illness representations; how an individual builds an understanding of a health threat

ii) coping procedures and illness behaviours; which describe behavioural responses that are determined by the beliefs associated with specific illness representation,

iii) dynamic feedback; a key aspect of the model that indicates the self-regulatory nature of the proposed system

A separate evaluation of how illness representations are assessed and a review of empirical research about illness representations in adolescents are presented. The review identifies the need for further theoretical development, and empirical testing in order for the CS-SRM framework to be suitable for use with adolescents.
2.2. The development of the common sense self-regulatory model

The underlying drivers of illness behaviours and people’s responses to health information have been examined within the framework of the CS-SRM (Leventhal et al., 1980). The CS-SRM emerged from studies which showed that people still engage in risk-taking behaviour, for example smoking despite knowing the risks related to it (i.e. cancer) (Johnson & Leventhal, 1974). This cognitive theory is built on the premise that people build patterns of mental representation; mental representations are easily accessible structures of abstract thoughts and ideas that represent an external reality. The structure of mental representations can be viewed as organised patterns of thoughts or concepts known as schemas, or schemata. Schemata are considered to be the cognitive basis for both mental and physical action. A mental representation about illness, also known as an illness representation, is a construct linking abstract and concrete information regarding the health threat in a manner that would “make sense” to the individual (Leventhal et al., 1984).

Leventhal, Meyer, and Nerenz (1980) originally argued that illness behaviours (behaviours that manage illness and improve health) were motivated by the perception of danger. The concept of fear was thought to be the underlying motivation in forming action plans, and eventually, illness behaviours (Janis & Feshbach, 1953). However, experimental work found this motivation is reliant on other variables, including the conceptualisation of the health threat, individual differences, and the development of action plans (Leventhal et al., 2012).

Indicators of a health threat and fear were necessary in the processes involved in motivating behaviour, but were not sufficient for adherent and compliant behaviour (Johnson & Leventhal, 1974). Therefore, Leventhal and colleagues (1980) proposed to investigate and model the processes of activation of fear control and the perception of danger; they hypothesised that these processes interact to elicit coping responses. An early study with patients undergoing an endoscopic examination found that providing patients with a conceptual understanding of what to expect during the examination reported less gagging during the procedure leading to these patients demonstrating an adaptive fear response (Johnson & Leventhal, 1974). This illustrated the way in which individuals perceive a health threat, along with emotions associated with that threat, both of which
informed the response. This suggested that coping requires the formation of goals based upon the representation of a health threat and structuring action plans. These conclusions led to the development of the parallel process model, later named the Common Sense Model or the Self-Regulatory Model. Now, the model is commonly referred to as the CS-SRM (Leventhal et al., 1984).

2.3. **Common sense self-regulatory model**

The CS-SRM is a dynamic self-regulation model in which the behaviour of the individual is regulated. Leventhal and colleagues (2012) refer to the model as an ‘online system’; this means that when new information is introduced it is processed and evaluated, implying that beliefs and behaviours can change according to the new information. The model is constructed with three stages of information processing:

1. development of a mental representation, (of an illness, symptom or health threat),
2. coping response (plans to deal with the perceived threat based on specific representation), and
3. appraisal (evaluation of the coping response and how well this information “fits”).

Leventhal and colleagues argued that the information used to form the cognitive schema of an illness was gathered from three basic sources (Leventhal et al., 1980; Leventhal et al., 1984). The first source is bodily experience, or the current experiences of the illness; the second source is information gathered from previous experiences of that illness; and the third source is information from social contexts or authoritative sources (Leventhal et al., 1980; Leventhal et al., 1984). The identified symptoms were measured against previous experiences of those symptoms associated with an illness or health threat that activate a representation of that illness. By using open-ended questions, one study determined that five themes (or domains) were repeatedly mentioned when individuals talked about their understanding of an illness (Linz et al., 1982; Meyer et al., 1985); the five domains were timeline, cause, consequences, identity, and control/cure. Beliefs were found to be inter-
dependent of each other. A concrete somatic experience (along with the concrete somatic symptoms) enabled elaboration of abstract information to develop coherent mental representations, leading to the generation of an overall model of the illness or health threat (Leventhal et al., 1980).

Figure 4: Simplified model of the CS-SRM

(Adapted from Leventhal et al., 2011) There is an automatic process from the input of sensory stimuli that is evaluated against previous sensations functions and within the context of behaviour and cognitive social information. This information is checked against past experiences for patterns, location, onset and duration and this leads to coping procedures which are then appraised and evaluated.

This model links mental representations and coping strategies, implying a causal relationship (Leventhal et al., 1993). Research on coping strategies supports this; patients who felt they had control over their epilepsy engaged in problem-solving strategies to manage the epilepsy, while those who felt they had no control over their epilepsy denied or avoided addressing the epilepsy (Kemp et al., 1999).

Information can be processed through two parallel pathways: cognitive and emotional (Leventhal et al., 1984; Leventhal et al., 2012). Individuals can respond to an illness using both pathways simultaneously. It is possible for the pathways to inter-relate, which can have an influence on the information being processed, the coping strategies implemented,
and the appraisal of the information (Leventhal et al., 2012). In the final stage of the model, individuals evaluate the effectiveness of their success. The next stage is for them to choose (through automatic processes) either to continue with the beliefs they currently hold and their coping style, or else adapt an alternative. This evaluation results in both self-regulation of the behaviour(s) and reinforcement or modification of the illness representations and together help to explain the between- and within- individual differences in responses to coping with an LTC over time.

2.4. Illness Representations

Illness representations are structured on both abstract concepts and past experiences; the content of these concepts is developed from an individual’s perceptual and procedural knowledge (Squire, 2004). The structure and content of the illness representations provide the underlying mechanisms of goal identification and targets for action (Leventhal et al., 2012). The following section discusses and defines each of the domains that make up illness representations. These sections compare studies with adults identified by a meta-analysis by Hagger & Orbell (2003) and studies with children and adolescents under the age of 18 years old identified by a systematic review by Law, Tolgyesi and Howard (2012). The aim of these sections is to compare the relationship between illness representations with self-management in adults and young people with LTCs.

2.4.1. Identity

The identity construct is the label or symptoms that are associated with an illness (Lau et al., 1989; Meyer et al., 1985). Individuals attribute symptoms to a specific health threat and use this information to identify the threat. Therefore, this construct is built on knowledge about symptoms as well as illness labels (Nerenz & Leventhal, 1983).

Research with adults found that those who reported more symptoms also reported beliefs of more severe consequences, and the combination of these beliefs occurred in patients who were found to have poor self-management (Hagger & Orbell, 2003). Studies with
adolescents across different conditions mostly reported no significant relationship between identity and self-management (cystic fibrosis; Bucks et al., 2009; (Hypertension; Zugelj et al., 2010); Diabetes; (Law et al., 2002; Skinner et al., 2003). However, some studies identified that adolescents who reported better dietary self-care, reported fewer symptoms associated with their diabetes (Gaston et al., 2012; Griva et al., 2000). Further work is required to explore the role of the identity domain with other domains as shown in the Hagger & Orbell, meta-analysis (2003).

2.4.2. Cause

Causal attributions of a health threat can be external agents, internal susceptibilities, or behaviours. Within the CS-SRM framework, these attributions form the construct of the ‘cause’ domain (Leventhal et al., 2012). Factors such as stress or virus were considered as causal attributions that individuals believe are responsible for their health threat. As discussed in the previous chapter (Section 1.4.1.4), young children do not hold sophisticated causal attributions (Goldman et al., 1991). However, work with adolescents aged between 11 and 16 years with JIA found that the underlying causal attributions were related to adolescents’ health experiences, specifically the referral route (Cordingley et al., 2012). This suggests that adolescents are developing a more sophisticated and abstract conceptualisation of internal physiological changes that occur during an illness (Berry et al., 1993; Bibace & Walsh, 1980; Perrin & Shapiro, 1985). For adolescents, the link between causal attributions and self-management is unclear; in the systematic review with adolescents, there were five studies that included the measure of cause, however the results were not reported against self-management (Law et al., 2012). Similarly, within the adult literature, there are few studies that include the domain of cause within the analysis of self-management outcomes (Hagger & Orbell, 2003).

2.4.3. Timeline

Beliefs about the onset and duration of a health threat make up the construct of the timeline domains (Leventhal et al., 2012). The timeline domain also includes beliefs about the
course of the illness and disease symptoms, and comprises the constructs for two types of timeline beliefs (Moss-Morris et al., 2002).

The first construct concerns the chronicity of the health threat/illness; if patients perceive the threat/illness as acute, they would use concrete information from the symptoms to control the threat. Yet, when the threat/illness is perceived as chronic, the timeframe is abstract (Leventhal et al., 2012). In these cases, the patients would try to use other information to predict symptoms and regulate behaviour. The second timeline construct concerns the cyclical nature of the health threat/illness; this construct reflects an individual’s need to translate abstract concepts into concrete experiences by finding patterns in the health threat/illness (Quinn & Eimas, 1997).

Studies included in the meta-analysis found that adults’ belief that an illness is chronic is associated with psychological distress, and more likely to lead to coping behaviours of avoidance/denial and cognitive reappraisal (Hagger & Orbell, 2003). With adolescents, only two studies found a significant relationship between chronicity beliefs and self-management behaviours (cystic fibrosis; Bucks et al., 2009; diabetes; Gaston et al., 2012). See below for Table 2 of studies that included an assessment of chronicity beliefs). These two studies reported a discrepancy in the relationship between chronicity beliefs and behaviours with one study reporting better management (Bucks et al., 2009) the other poor management (Gaston et al., 2012).

2.4.4. Consequences

The construct of illness consequences is built on beliefs regarding the anticipated and experienced impact of a disease (Leventhal et al., 2012). Beliefs about illness consequences are those related to the impact the illness has on physical, social, and psychological functioning.

The association of beliefs about consequences with self-management and coping varies across studies and age groups. In adult populations, avoidance and/or denial coping strategies are found to be positively associated with beliefs that the illness has high impact
on an individual’s life (Hagger & Orbell, 2003). These beliefs regarding consequences are thought to be less likely to change over time (Weinman et al., 1996). However, beliefs on the impact have not been found to be associated with self-reported medication adherence in adolescents with chronic rheumatic diseases (Lawson et al., 2011).

2.4.5. Control/Curability

The construct of control and curability is defined by the perceived and expected illness response to treatment and self (Lau & Hartman, 1983). This domain is split into two constructs: beliefs about the cure, and beliefs about the controllability of the health threat or illness. The beliefs about the curability of the health threat or illness are linked to the treatment taken for the health threat. This includes the individual’s beliefs about the efficiency and efficacy of the treatment. The controllability construct refers to beliefs about the extent to which the individual is able to exert personal control over the health threat or illness.

The meta-analysis found that with adults, the general consensus in the research is that cure and control beliefs were negatively related to psychological distress; therefore, the stronger the sense of personal control and treatment control, the less psychologically distressed the individual (Hagger & Orbell, 2003). In two studies with adolescents who have strong beliefs regarding treatment control, individuals were more likely to engage in self-management behaviours (Skinner & Hampson, 2001; Skinner et al., 2000). However, in other studies there were no significant associations found between the adolescents’ personal control and the self-management behaviours (Fortenberry et al., 2011; Law et al., 2002; Zugelj et al., 2010). This supports the findings from other studies suggesting that the processes of perceiving control differ between adults and children or young people.

A qualitative study of the cure and controllability beliefs in 20 adolescents with JIA aged between 11 and 16 year olds found that there were two patterns of perception related to personal control (Ghio, 2011); the first pattern identified was that they felt they had no control over their arthritis, while the second pattern was that adolescents viewed their arthritis as an external entity and, therefore, the control was external. All adolescents in
this study were searching for predictable symptoms or indicators of flare ups in order to gain control, despite finding their arthritis was too unpredictable (Ghio, 2011). Adolescents attempted to anchor abstract concepts of control into a concrete experience by finding other ways of asserting personal control. For these adolescents, treatment was a strategy to control their arthritis. Treatment strategies included medication and exercise. However, the adolescents’ beliefs about treatment control were that treatment was limited and unreliable (Ghio, 2011).

2.5. Emotional representations

The CS-SRM describes two parallel systems: cognitive and emotional. The emotional system includes a pathway of a pre-existing emotion and coping procedures associated with it, which determine the emotional outcomes (Moss-Morris et al., 2002). Research into emotional representations has found there are common emotional responses, including depression, anger, and anxiety (Cameron, 2003). Depression and anxiety are the most commonly assessed responses in studies of emotional representations.

Emotional representations were not associated with self-management coping responses in adolescents in prior studies (Law et al., 2012; Bucks et al., 2009). Qualitative work in adolescents with JIA found that consequences, pain, causal thinking, judgment, and the unknown were the common underlying reasons for emotional representations (Ghio, 2011). The original work by Leventhal and colleagues (Leventhal et al., 1980; Leventhal et al., 1984) argued that cognitive pathways can influence emotional responses. Further work with adults confirmed that this relationship is bidirectional (Cameron, 2003). However, research with adolescents has not explored the inter-relationship between the emotional and cognitive pathways.

2.6. Coping procedures and illness behaviours

Intentions and coping behaviours are guided by an individual’s representation of a health threat or illness (Leventhal et al., 1980). Theoretically, a patient with a concrete and
detailed representation of a health threat or illness would also have a detailed coping plan (Leventhal et al., 2012). Drawing from the ideas of self-efficacy, it is hypothesised that if a patient focuses on self-regulating behaviour they will gain a sense of control over the emotional experience and improve their quality of life (Bandura, 1986); patients with abstract and remote goals would have increased distress and uncertainty due to the lack of cues that could be indicative of progress or failure. The early work conducted in order to develop this theory hypothesised that immediate and concrete goals facilitated coping (Leventhal et al., 1980). This hypothesis was supported by studies that found that preparatory information improved outcomes (Johnson et al., 1973; Johnson & Leventhal, 1974).

In the coping literature, illness representations were thought to characterise the first appraisal of a threat (Lazarus, 1966); this forms part of the coping process. The coping process involves two stages: primary appraisal of the threat and secondary appraisal of the capacity to cope with the threat (Lazarus, 1966). Internal and external demands that are perceived as exceeding personal resources require cognitive and behavioural efforts to manage the demands (Folkman & Lazarus, 1988). The cognitive and behavioural efforts are dependent on the first appraisal (illness representation). The functions of the cognitive and behavioural efforts can be classified by either addressing the threat (problem-focused coping), or by regulating the associated distress (emotion-focused coping).

A key premise of the CS-SRM is that the representations of coping procedures are determined by and reflect the illness representations (Leventhal et al., 2012). The choice of coping procedure is also dependent on the social context. Therefore, there are a variety of procedures with a range of objectives (Leventhal et al., 2012). When behaviour is selected, whether it is automatic or deliberative, it is informed by a coherent overall model of the illness (Leventhal et al., 2011). Objectives of the coping procedure are established from illness representations suggesting that the choices and decisions regarding response can be predicted by the CS-SRM.

Motivated reasoning, a concept in the process of decision-making, is thought to be influenced by “hot cognitions”, a term used to define cognitions that are inter-related with, or biased by, emotional state (Smith et al., 1993). Leventhal and colleagues (2001) argued
this construct as essential for the pathway to motivate change in behaviour. In reality, managing both the cognitive and emotional processing systems can create a dissonance (self-regulatory failure) in the pathways (Detweiler-Bedell et al., 2008). For example, effective self-management of physical symptoms of the disease (such as adhering to a demanding treatment schedule) can impact on everyday life (high perceived consequences), and reinforce a negative response to treatment and to the disease.

2.7. Self-regulation (Dynamic feedback stage)

The final stage of the CS-SRM is the assessment of the previous stages as identified in Figure 4. Specifically, the individual evaluates the content of the illness representations, the action plan, the interaction of the executive and automatic processes for selecting a coping procedure, and the illness outcome. These are dynamic processes occurring between the content and structure of a personal model. These processes are examined in linearity; but, by definition, the content and structure of an individual’s personal model is not static (Leventhal et al., 2012).

Appraisal of the cognitive and behavioural processes is identified within the coping literature as the second appraisal (Folkman & Lazarus, 1988). The aim of this phase of appraisal is to assess the failures, the progress of the health threat or illness, and the behaviours dealing with the health threat or illness. This evaluation leads to a change or adjustment in either illness representations or the behaviours, or else to reinforce the existing behaviour or representations (Leventhal et al., 1984). As argued by Leventhal and colleagues (Leventhal et al., 2012), these changes and reinforcements are difficult to capture in real time. However, following the theoretical framework, it is possible to develop a hypothesis to test, (for example, changes in either illness representations or illness behaviours derived from a theoretically-driven intervention) (Petrie et al., 2002). When new information is processed, it is matched with previous information and if it fits (provides equilibrium) the illness representations will change as well as the associated behaviour (Leventhal et al., 2011).
These hypotheses have been supported with interventions utilising the CS-SRM framework. The first of these interventions was by Petrie et al (2002). In this study they found that by providing a psychoeducational intervention to an experimental group of 31 patients to target the group’s illness representations, the patients returned to work after a myocardial infarction earlier (with fewer coronary related symptoms) in comparison to the 34 patients in the control group receiving standard care. The psychoeducational intervention was focused on challenging inaccurate beliefs, and there were differences in the groups’ timeline and control beliefs. Cameron and colleagues (2005) investigated the impact of negative affectivity on the intervention designed by Petrie and colleagues (2002). Negative affectivity was a moderator in the intervention group, where disability increased and adherence decreased in dietary and exercise behaviours compared to usual care. Interventions to improve emotional regulation strategies (such as decreased use of emotional suppression) in patients generated higher perceptions of personal control in those patients (2007).

The hypotheses regarding change in illness representations and the associated changes in behaviour have been supported by cross-sectional and longitudinal work. However, it should be noted that there are very few studies that have examined the impact of change in illness representations on outcomes and behaviours longitudinally. In a sample of patients with back pain (aged 18 – 60 years; mean age 43.9, SD 10.3), those who held beliefs that pain has serious consequences and reported a lack of personal controllability had poor clinical outcomes after 6 months (Foster et al., 2008). For those patients who reported positive clinical outcomes, there was a significant decrease in their associated symptoms (identity), as well as reductions in impact and emotional representations, while control (treatment and personal) increased (Foster et al., 2008). This study supports the theory that these beliefs are not static, and are related to outcomes.

Coping strategies may be mediators of the relationships between beliefs and outcomes (Leventhal et al., 2011). A recent study supporting this argument used a large longitudinal design with young adolescents with asthma (mean age 11.9) (Tiggelman et al., 2014). The study reported that in a longitudinal model, the indirect effect between illness perceptions at time 1 and emotional problems at time 3 was partially mediated by worry as a coping strategy at time point 2; this accounted for 63.2% of the variance in emotional problems,
but only 37.7% of the variance in asthma control (Tiggelman et al., 2014). One limitation of this study is that despite evidence of an association between asthma control and emotional problems, the authors did not present whether there was an association between the two outcomes. The cross-sectional data in this study had more associations; however, the authors did not explore these differences between cross-sectional and longitudinal data further.

The issue with study designs in which illness perceptions at ‘time point 1’ predict an outcome at ‘time point 3’ is that it does not account for possible changes in illness perceptions that may occur during the intervening period. One study monitored change between baseline and six-year follow-up for illness perceptions in adults with osteoarthritis (OA) (mean age 59.0, SD 7.5), and how change was associated with functional status (Kaptein et al., 2010). There were changes in the beliefs between the baseline and the six-year follow-up, with significant increased timeline chronicity, decreased personal control, increased coherence, reduced timeline cyclical beliefs, and reduced negative emotional representations (Kaptein et al., 2010). There were no significant differences in cause, treatment control, identity, and consequences between the two time points.

In this OA study, Kaptein and colleagues (2010) hypothesised that patterns in the illness perceptions (two cluster groups: positive and negative) were associated with functional status and pain intensity. There were differences in the outcomes of functional status and pain intensity in both the positive and negative illness perception groups (Kaptein et al., 2010). The positive cluster (decrease in identity, chronicity, consequences and emotional representations, with an increase in personal and treatment control) did not demonstrate significant associations with an increase in pain intensity, but there was a slight improvement in function. For the negative cluster group (increase in identity, chronicity, consequences, with a decrease in emotional representations, personal and treatment control), there were more negative outcomes (in function and pain) at both time points (Kaptein et al., 2010). These results support the idea that illness perceptions are not static over time, and even without an intervention, these perceptions may change.
2.8. Conclusions regarding utilising the CS-SRM with adolescents

Sections from 2.4 to 2.7 showed that the use of the CS-SRM with adolescents is limited especially when compared with adult research. The comparison of studies from a meta-analysis with adults (Hagger & Orbell, 2003) and the review with adolescents (Law et al., 2012) found important differences. This could either be due to problems with the assessment of illness representations in adolescents (reviewed below in Section 2.9) or because there are differences due to adolescents’ cognitive development that the CS-SRM does not take into account.

2.9. Assessment of illness representations

Different approaches have been used to assess and quantify illness representations in adults; the original work utilised open-ended descriptions (Bishop & Converse, 1986; Lau & Hartman, 1983) or unstructured interviews (Meyer et al., 1985). A modified version of the Implicit Model of Illness Questionnaire (IMIPQ; Turk et al., 1986) was developed to map the domains of the illness representations of patients with RA, multiple sclerosis, and HIV (Schiaffino et al., 1998). For example, the Personal Models of Diabetes Questionnaire (PMDQ; Skinner & Hampson, 1998; Skinner et al., 2000) was developed from the Personal Models of Diabetes Interview that assessed the constructs of treatment effectiveness, seriousness, and cause (Hampson et al., 1995). A theory-driven questionnaire assessing the CS-SRM constructs was later developed using patient-generated items that were elicited from interviews (Weinman et al., 1996). The next section describes this questionnaire and the revised version in more detail.

2.9.1. Illness Perceptions Questionnaire

The Illness Perception Questionnaire (IPQ; Weinman et al., 1996) and associated questionnaires (IPQ-R, Moss-Morris et al., 2002; Brief IPQ; Broadbent et al., 2006) aim to systematically assess beliefs theorised by the CS-SRM (Leventhal et al., 1984). The IPQ was adapted for different illnesses and patient populations to map the relationship between
illness perceptions and outcomes, outcomes such as functionality, coping, and adherence. However, reviews showed there were internal inconsistencies, namely in two subscales: cure/control and timeline. Therefore, a revised version of the IPQ was produced (Moss-Morris et al., 2002). The IPQ-Revised (IPQ-R) added three domains. Firstly, the authors separated the cure/control domain into two subscales: personal control and treatment control. Personal control was defined as an individual’s beliefs about having control over the illness (self-efficacy). Treatment control was defined as the beliefs about the treatments’ ability to cure/control the illness. Lastly, the authors of the IPQ-R highlighted that the CS-SRM includes both emotional and cognitive responses; however, emotional responses are not captured by the original IPQ. Therefore, the authors added a subscale to measure emotional representations. Five emotions emerged from prior research, these emotions were typical responses: depression, upset, anger, worry, and fear (Cameron et al., 1993). It is unclear whether any of these constructs are relevant to adolescents, as the development of the assessment of illness representations were with adult populations.

For the development of the IPQ-R, validation of the factor structure and internal reliability was investigated with 711 patients with different illnesses, including 76 RA patients. The test-retest reliability was assessed at two time points in one year where the RA patients filled out the IPQ-R. These results were all significant with a p-value higher than 0.5, except for timeline cyclicality with a value of 0.35. Predictive validity was tested with a sample of patients with multiple sclerosis using other questionnaires compared with assessment from a trained research assistant. To determine the questions for each domain, factor analysis and Principal Component Analysis (PCA) including Varimax rotation were used (Moss-Morris et al., 2002).

The study validating the IPQ-R recruited different adult illness groups to validate different features (such as retest reliability with an RA sample). However, the authors advocated adapting the questionnaire for appropriate use for different illness groups (Moss-Morris et al., 2002). However, French and Weinman (2008) argue that the work is limited due to not eliciting beliefs that are illness-specific. Some studies validating the IPQ-R within their illness group have suggested modifications to capture the specifics of an LTC. For example, a content analysis of illness perceptions of 36 adults with type 2 diabetes (mean age 67.77 years, SD 10) found that the proposed concepts such as cure/treatment control
could be further categorised within this population (McCorry et al., 2012). This study utilised a “think-aloud” methodology, which is a type of cognitive interviewing. This method allowed McCorry and colleagues (2012) to indicate which items were misunderstood (negative wording), and those items that were not applicable to the sample (‘my symptoms come and go in cycles’). This study highlighted an important issue. Even though the original authors of the IPQ-R advocated the modification and validation of the tool to make it more relevant to a variety of populations, this has not been evident in the majority of the studies using the IPQ-R.

This issue becomes even more problematic as the IPQ-R has been utilised in various samples of adolescents. These studies modified the IPQ-R by reviewing the language with the aim of distributing a questionnaire that is age-appropriate; however, these studies did not consider cognitive differences as well as illness-specific differences in their modifications (see review under section 2.8.2). For some of these studies, changes were piloted with a smaller sample to ensure comprehension. However, not all studies piloted the modified questionnaire (discussed further in the review under section 2.8.2). The diabetes illness representations questionnaire (DIRQ) developed for adolescents with T1D mellitus, found that there are distinct features to the constructs measured by the IPQ-R that are specific to T1D (Skinner et al., 2003). The proposed modifications to some constructs in the DIRQ, such as a multi-faceted approach to treatment control, should include the control of diabetes, as well as the treatment’s effectiveness in preventing complications. These suggestions were supported by the “think-aloud” study with adults by McCorry and colleagues (2012). Further work with adolescents with other LTCs will enable the development of appropriate measures of adolescents’ illness perceptions. A detailed review of the studies utilising the CS-SRM with adolescents is provided in the following section.

2.9.2. Research with adolescent cohorts

The research utilising the CS-SRM with adolescent cohorts is limited compared to the adult literature. Web of Science, Psycinfo and EMBASE were searched from inception to 31st July 2014. A search strategy was developed combining terms and keyword searches relevant to: illness representations (e.g. ‘illness representation$’, ‘illness perception$’,
illness ‘cognition$’), to the CS-SRM (e.g. ‘Common Sense Model’ ‘Self-Regulatory Common Sense Model’, ‘Leventhal AND Model’), and to adolescents (e.g. ‘adolescen$’, ‘child$’, ‘pediatric$’, ‘paediatric$’, ‘young’).

Titles and abstracts were screened to identify any study including adolescent cohorts (participants aged between 11 and 16 years). After the initial screening, full texts were accessed and assessed to review the usage of the model using adolescent cohorts. Twenty-six articles were identified as relevant and the methodology of identifying the articles is presented in the flow chart in Figure 5.

Of the 26 manuscripts included, 12 utilised the IPQ-R; however, of these 12 manuscripts, only two included modifications to the questionnaire (Law, 2002; Law et al., 2002). Only five of the 12 studies using the IPQ-R utilised all domains in the questionnaire (Gray & Rutter, 2007; Jones et al., 2014; Law et al., 2002; Law, 2002; Bucks et al., 2009). Five of the other 14 manuscripts that did not use the IPQ-R used the DIRQ (two studies; (Edgar & Skinner, 2003; Gaston et al., 2012) and the PMDQ (three studies; (Nouwen et al., 2009; Skinner & Hampson, 1998; Skinner et al., 2000). Four of the 14 studies used a qualitative approach to assess illness representations with either structured interviews (e.g. Paterson, Moss-Morris & Butler, 1999), or semi-structured interviews (Babooram et al., 2011; Meulenkamp et al., 2008; Veldtman et al., 2000). The remaining five manuscripts included three manuscripts using the Brief Illness Perceptions Questionnaire (BIPQ), which included nine of the domains (two of the manuscripts however, did not include the causal domain). The last two manuscripts used modified measures: one of the measures was modified from the original IPQ for children which does not include the new constructs from the IPQ-R (the CIPQ; Walker et al., 2006). Ratcliff, Blount and Mee (2010) developed the Perceived Adversity Scale, which utilises different items to measure the consequences of living with renal transplant.
Overall, these studies have shown support for utilisation of the CS-SRM with adolescent cohorts. The qualitative studies found that adolescents hold beliefs that map onto the domains of the CS-SRM, (Babooram et al., 2011; Meulenkamp et al., 2008; Veldtman et al., 2000) but one study found that age did not contribute to the beliefs held by adolescents regarding their illness despite age and intelligence being predictors of illness conceptualisation (Paterson et al., 1999). Studies with small samples also found that illness perceptions are associated with or were predictive of outcomes such as emotional wellbeing and/or coping. However, it is hard to draw strong overarching conclusions due to mixed results in each construct.
### Table 2: Characteristics of included studies

Review of 26 studies that utilise the CS-SRM as a theoretical framework to paediatric samples

<table>
<thead>
<tr>
<th>Reference (year)</th>
<th>Sample size (n)</th>
<th>Condition</th>
<th>Ages</th>
<th>CS-SRM Measure</th>
<th>Domains</th>
<th>Outcomes/other measures</th>
<th>Findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>(Babooram et al., 2011)</td>
<td>[Qualitative] n = 33</td>
<td>Obesity</td>
<td>7 – 12</td>
<td>Semi-structured interviews</td>
<td>Identity, cause, timeline, consequences, control/cure</td>
<td>To examine understandings of obesity</td>
<td>*All of the children had knowledge that fit within each domain. Consistent with adult research. *Children with normal weight were more aware of the consequences associated with overweight than those children who were overweight.</td>
</tr>
<tr>
<td>(Bucks et al., 2009)</td>
<td>[Cross-sectional] n = 38</td>
<td>Cystic fibrosis</td>
<td>11 – 17</td>
<td>IPQ-R</td>
<td>Identity, chronicity, consequences, cause, personal and treatment control, emotional representations</td>
<td>Reported adherence measured by Cystic Fibrosis Treatment Questionnaire (CFTQ) and non-adherence was measured by Medication adherence Report Scale (MARS)</td>
<td>*Treatment control and timeline chronicity were associated and predictive of antibiotic necessity and adherence. *Timeline mediated the relationship between age and adherence of antibiotics.</td>
</tr>
<tr>
<td>(Chong et al., 2012)</td>
<td>[Cross-sectional] n = 48</td>
<td>Cerebral palsy</td>
<td>8 – 18</td>
<td>BIPQ</td>
<td>Identity, treatment control, personal control, timeline, concern, coherence emotional impact</td>
<td>1 minute and 6 minute walk tests to measure/monitor progress of walking function. Gross Motor Function Classification System (GMFCS) Students’ Life Satisfaction Scale (SLSS)</td>
<td>*Relationship between GMFCS and age to perceptions regarding the efficacy the physiotherapy but not consequences. Those with highest function classification are the participants who perceived the CP as acute. *Higher SLSS was associated with lower concern, lower emotional impact, higher personal control but not with GMFCS or the walk test. *Concern (39% variance) and Consequences (46% variance) predicted highest SLSS scores.</td>
</tr>
<tr>
<td>(Chong et al., 2013)</td>
<td>[Cross-sectional] n = 52</td>
<td>Cerebral palsy</td>
<td>5-18</td>
<td>BIPQ</td>
<td>Identity, treatment and personal control, timeline, concern, coherence emotional impact consequences</td>
<td>A drawing of walking (size, activity portrayed, setting and inclusion of others), 1 minute and 6 minute walk tests. GMFCS, SLSS</td>
<td>*Only association found with BIPQ was emotional impact was lower in those participants who drew other figures as well as themselves (Mann-Whitney U = 79.00, r=.31, p&lt; .05)</td>
</tr>
<tr>
<td>(Cordingley et al., 2012)</td>
<td>[Cross-sectional] n = 122</td>
<td>JIA</td>
<td>11-15</td>
<td>IPQ-R</td>
<td>Cause</td>
<td>Characteristics: age, gender, Source of referral, JIA subtype, pain, CHAQ, active and limited joint count</td>
<td>*Genetics or immune system: more likely to be female or systemic arthritis *Accident or injury: more likely male or have orthopedic surgery as a source of referral *Infection: more likely systemic arthritis and shortest disease duration</td>
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<tr>
<td>Study</td>
<td>Population</td>
<td>Duration</td>
<td>Measures</td>
<td>Outcomes</td>
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<tr>
<td>(Croom et al., 2011)</td>
<td>Diabetes</td>
<td>10-14</td>
<td>IPQ-R</td>
<td>Perceived physician-patient relationships (PCC), competence (perceived competence for diabetes scale), efficacy (Self-efficacy for Diabetes Management Scale), Adherence (SCI)</td>
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<tr>
<td>Longitudinal</td>
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<td>PCC was associated with teen control and adherence (both parent and child report) and parent control, treatment control and self-efficacy from just adolescents’ reports.</td>
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<td></td>
<td>n = 190</td>
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<td>PCC predicted positive change in only the parent control reported by the parents.</td>
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<td>Only competence (teen) was a significant mediator between PCC and treatment outcomes.</td>
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<tr>
<td>(Edgar &amp; Skinner, 2003)</td>
<td>Diabetes</td>
<td>11-18</td>
<td>DIRQ</td>
<td>Emotional well-being (Well-being questionnaire), Coping strategies (Kidcope)</td>
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<tr>
<td>Cross-sectional</td>
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<td>Depression and anxiety is associated with both identity and perceived impact. Positive well-being is associated with identity, effectiveness to prevent and (−) association with perceived impact.</td>
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<td></td>
<td>n = 126</td>
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<td>Cognitive restructuring’s association with positive well-being is mediated by treatment effectiveness.</td>
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<td>High impact and high identity scores and using cognitive restructuring predicted lower depression scores.</td>
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<td>High anxiety was predicted by higher scores in impact and identity scores.</td>
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<tr>
<td>(Fortenberry et al., 2011)</td>
<td>Diabetes</td>
<td>10-15</td>
<td>IPQ-R</td>
<td>Personal control, treatment control, Negative affect (diary), blood glucose (daily), adherence (SCI), HbA1c</td>
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<tr>
<td>Cross-sectional</td>
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<td>Higher negative affect predicted more daily diabetes problems and this was moderated by perceptions of treatment control (and not personal control).</td>
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<td></td>
<td>n = 209</td>
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<td></td>
<td>Trajectories found that over time there were significant positive changes in chronicity, consequences, personal and treatment control and coherence. With significant negative changes in cyclacity and parent control.</td>
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<td></td>
<td>Adherence was associated with greater personal, parent, and treatment control, coherence and lower negative emotional representations.</td>
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<td>HbA1c is associated with less severe consequences, less parent control, coherent, and future negative consequences.</td>
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<td>QoL is associated with lower cyclical and lower consequences, higher coherence and higher treatment control and negative emotional representations.</td>
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<tr>
<td>(Fortenberry et al., 2014)</td>
<td>Diabetes</td>
<td>10-17</td>
<td>IPQ-R</td>
<td>Diabetes responsibility, cognitive functioning, diabetes quality of life, adherence (SCI), Metabolic control - HbA1c</td>
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<td>Longitudinal, baseline was not included</td>
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<td>Trajectories found that over time there were significant positive changes in chronicity, consequences, personal and treatment control and coherence. With significant negative changes in cyclacity and parent control.</td>
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<td>n = 213 (time 2)</td>
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<td>Adherence was associated with greater personal, parent, and treatment control, coherence and lower negative emotional representations.</td>
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<td>n = 196 (time 3)</td>
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<td>HbA1c is associated with less severe consequences, less parent control, coherent, and future negative consequences.</td>
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<td>n = 183 (time 4)</td>
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<td>QoL is associated with lower cyclical and lower consequences, higher coherence and higher treatment control and negative emotional representations.</td>
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<tr>
<td>Study (Year)</td>
<td>Condition</td>
<td>Sample Size</td>
<td>Measure</td>
<td>Focus</td>
<td>Findings</td>
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<tr>
<td>Gaston et al. (2012)</td>
<td>Diabetes 12 – 16</td>
<td>DIRQ</td>
<td>Identity, timeline, consequences, threat to health, control/cure-blood sugars, control/cure – prevent complications</td>
<td>Self-management of diet, exercise, blood glucose monitoring (BGM) and insulin administration</td>
<td>Significant differences in parent-child dyads in timeline, consequences, impact and threat, control/cure-blood sugars, prevention. Dissimilarity maximised threat and poorer BGM. Caregivers' beliefs about impact of diabetes and treatment prevention correlated with dietary self-management. Adolescents' beliefs about threat to health, timeline and prevention correlated with BGM.</td>
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<td>Gray &amp; Rutter (2007)</td>
<td>Chronic fatigue syndrome 8 – 25</td>
<td>IPQ-R</td>
<td>Identity, timeline, chronicity, consequences, personal control, treatment control, emotional representations, cause</td>
<td>Coping (Illness Management Questionnaire), Quality of life (QoL - Quality of Life Scale), Physical functioning (Functional Ability Scale)</td>
<td>QoL associated with (+) identity, (+) treatment control, (-) emotional representations. Physical function associated with (-) identity. Model: 40% variance; relationship between (-) chronicity with (+) QoL is mediated by accommodating. Illness coherence, emotional representations and (-) treatment control to (-) QoL mediated by focusing on symptoms. Model: 21% variance: Relationship with risk factor cause beliefs with physical functioning mediated by maintaining activity. Direct effect of (-) identity and (-) illness coherence.</td>
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<td>Hughes et al. (2012)</td>
<td>Diabetes 10 – 14</td>
<td>IPQ-R</td>
<td>Consequences, Emotional Representations</td>
<td>Self-control, emotional process (Emotional Approach Coping Scale), Self-Efficacy (Self-Efficacy for diabetes Management Scale), Adherence (Self Care Inventory), HbA1c</td>
<td>HbA1c is associated with (+) self-control, emotional processing, self-efficacy, adherence, and (-) negative affect. 22% variance in HbA1c – interaction of self-control and emotional processing. 23% variance - adherence. 24% variance – negative affect and also self-efficacy. Interaction of emotional processing and self-control better at predicting HbA1c.</td>
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<td>Study (Year)</td>
<td>Design</td>
<td>Sample Size</td>
<td>Condition</td>
<td>Measure/Questionnaire</td>
<td>Findings</td>
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<td>(Law, 2002)</td>
<td>Cross-sectional</td>
<td>n = 30 adolescents and 26 mothers</td>
<td>Diabetes 13–19</td>
<td>IPQ-R* modified</td>
<td>Well-being Questionnaire (measuring depression, positive well-being, perceived energy)</td>
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<td>• Significant differences between consequences, emotional representations in adolescent and maternal illness representations.</td>
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<td>• Dissimilarities (subtracting mothers from adolescents) correlated with both adolescent and mother score but none were significantly associated with psychological well-being.</td>
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<tr>
<td>(Law et al., 2002)</td>
<td>Cross-sectional</td>
<td>n = 30</td>
<td>Diabetes 13–19</td>
<td>IPQ-R* modified</td>
<td>Well-being Questionnaire, Diabetes self-management (Summary of Diabetes Self-Care Activities Questionnaire), HbA1c</td>
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<td>• Illness beliefs accounts for 52% of variance in anxiety and 32% variance in positive well-being. Anxiety was enhanced by the addition of illness beliefs.</td>
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<td>• Consequences and personal control were significant positive predictors of anxiety and positive well-being.</td>
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<tr>
<td>(Meulenkamp et al., 2008)</td>
<td>Qualitative</td>
<td>n = 33 families</td>
<td>Genetic testing for cardiovascular disease 8–18</td>
<td>Semi-structured interviews</td>
<td>Identity, cause, consequences, timeline, control, worries, frustration</td>
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<td>Interview topics: use of medication, lifestyle issues (sports, diet, smoking), concerns and coping</td>
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<td>• The children and adolescents were able to describe the disease they were tested for (identity), including the cause (hereditary), and the consequences (possible death). Reported to feeling worried and frustrated for being different.</td>
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<td>• Causal beliefs were found to distinguish groups; the group that explained a 'defect' in their heart or veins, the other group attributed the 'defect' in their gene. Those who attributed the defect to the gene had lower illness coherence, less controllable. This was also dependent on the type of disease.</td>
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<tr>
<td>(Munson et al., 2009)</td>
<td>Cross-sectional</td>
<td>n = 70</td>
<td>Mood disorders 12–17</td>
<td>IPQ-R</td>
<td>Chronicity, cyclical, Personal and treatment control, consequences, illness coherence, emotional representation</td>
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<td>Attitudes towards help-seeking, stigma, and psychological openness (IAS-MHS: Inventory of Attitudes toward Seeking Mental Health Services)</td>
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<td>• Emotional representations and treatment control were associated with attitudes towards mental health services.</td>
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<td>• Higher treatment control associated with seeking help. Higher emotional representations reported feeling stigmatization</td>
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<tr>
<td>(Munson et al., 2010)</td>
<td>Cross-sectional</td>
<td>n = 70</td>
<td>Mood disorders 12–17</td>
<td>IPQ-R</td>
<td>Chronicity, cyclical, Personal and treatment control, consequences, illness coherence, emotional representation</td>
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<td>Adherence to medication and mental health appointments collected from young person and their parent self-report Attitudes (IAS-MHS)</td>
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<td>• Illness perceptions were associated with adherence.</td>
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<td></td>
<td>• Full adherence to appointments were associated with higher scores in consequences and higher levels of emotional reactions.</td>
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<tr>
<td>Study (Year)</td>
<td>Condition</td>
<td>Age Range</td>
<td>Measure(s)</td>
<td>Description</td>
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<tr>
<td>Nouwen et al., 2009</td>
<td>Diabetes</td>
<td>12 – 18</td>
<td>PMDQ &amp; IPQ-R</td>
<td>Treatment effectiveness and treatment control, consequences; Dietary self-efficacy, measuring confidence to follow dietary recommendations, dietary sub-scale of the Summary of Self-Care Activities Scale, Diabetes distress (Problem areas in diabetes survey)</td>
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<tr>
<td>Paterson et al., 1999</td>
<td>Asthma, Colds</td>
<td>7 – 14</td>
<td>Structured interview</td>
<td>Identity, Cause, timeline, consequences; seriousness, affect the day, worst thing) control/cure, prevention; Demographic information (Age, socioeconomic status, amount of illnesses); Level of intelligence (Wechsler Intelligence Scale for Children – Third edition)</td>
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<tr>
<td>Ratcliff et al., 2010</td>
<td>Renal transplant</td>
<td>11 – 20</td>
<td>Perceived Adversity Scale</td>
<td>Consequences of living with renal transplant; Perceived adversity Coping (A-Cope) Adherence (Medical Adherence Measure &amp; serum immunosuppressant drug assay levels); Health care Provider (HCP) estimation of adversity</td>
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<tr>
<td>Skinner &amp; Hampson, 1998</td>
<td>Diabetes</td>
<td>12 – 18</td>
<td>PMDQ</td>
<td>Treatment efficacy, Self-management control, prevention, Consequences; worry and concern, impact on daily life; Depression &amp; anxiety, Self-management, Social support; Diabetes-specific support; Peer support</td>
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</table>

- Structural equation model with the best fit was with dietary self-efficacy and short-term treatment control (with age as a predictor of treatment control) were associated with diabetes distress and consequences was associated with distress.
- There are higher scores of conceptualisation cold to asthma except in the consequences domain.
- Age and intelligence were predictors of cold (36% variance) and asthma (43% variance).
- Asthma experience had no effect on the domains about colds but had significant effect on domains about cause, chronicity and controllability of asthma. Number of illnesses were positively correlated with cause, consequences, and control/cure of asthma.
- Females and those who were seeking spiritual support reported higher missing school.
- Those who received transplant at age 16 or above reported higher PA. Number of medications and seeking spiritual support was associated with PA for medical procedures and scared about what might happen.
- Taking medication was associated with HCP perception of both physical and psychosocial adversity.
- Seeking social support and family support was associated with feeling different from peers.
- Seeking diversions was associated with not feeling well.
- Consequences; seriousness and impact associated with depression and anxiety. Treatment; control and complications associated with diet.
- Treatment control beliefs partially mediate between family support and dietary self-management. Family support also has a direct effect.
- Social support and perceived impact both independently predicted depression.
(Skinner et al., 2000)

[longitudinal – 6 month follow up reported. Baseline (BL) and Follow up (FU)]
n = 52

Diabetes 12 – 18

Measure of Health Related Quality of Life (HRQL, Food allergy quality of life questionnaire; teenager and parent form adolescent version, food allergy independent measure teenager and parent form)

- There were significant differences between follow-up (6 months) and baseline measures of perceived seriousness [increased], treatment efficacy [increase], general family support [decrease] and diabetes-specific family support [decrease].
- Impact and change in impact were predictors of anxiety, depression, and general well-being. Baseline treatment control and (-) seriousness predicted dietary self-care.
- Treatment control (BL and FU) were predicted by general support and mediated the associations between social support and socioeconomic status and dietary self-management.
- Perceived impact (BL and FU) and social support (BL and FU) were independent predictors.
- Perceived impact partially mediating the relationship between gender and well-being.

(van der Velde et al., 2011)

[Cross-sectional]
n = 70

Food allergy 13 – 17

- There were significant differences in the BIPQ from the parents and the adolescents in the total score, concern and emotional representations.
- Poor adolescent-reported illness coherence, and increased age, higher perceived disease severity contributed to larger difference in parent-child reports of HRQL.
- Mean difference in HRQL reports were more strongly associated with adolescent-reported BIPQ (coherence, emotional and cognitive) than parent proxy reported BIPQ.

(Veldtman et al., 2000)

[Mixed methods]
n = 63

Congenital heart disease 7 – 18

- Participant explanations of their illness fell into three categories; wrong/poor explanation, partial explanation, or good explanation.
- Category of understanding was not related to age, gender, or complexity of disease.
- Understanding of the duration improved with age.
<table>
<thead>
<tr>
<th>Walker et al., 2004</th>
<th>Eczema, Asthma, Healthy controls</th>
<th>7-12</th>
<th>CIPQ</th>
<th>Timeline, consequences, cure/control, cause</th>
<th>Psychosocial factors (<em>Piers-Harris Children’s Self-Concept Scale</em>)</th>
</tr>
</thead>
</table>

- Children with eczema reported higher consequences than children with asthma (sig differences).
- For children with eczema, consequences beliefs were divided in high/low with significant differences in outcomes. High was associated with low behaviour, high anxiety, low popularity with peers, and less happiness.

*note: IPQ-R; Revised Illness Perceptions Questionnaire; BIPQ: Brief Illness Perception Questionnaire; DIPRQ: Diebetes Illness Representations Questionnaire; PMDQ: Personal Model Diebetes Questionnaire; CIPQ: Children’s Illness Perception Questionnaire*
2.9.2.1. Identity

The three qualitative studies in Table 2 included the construct of identity, and demonstrated that participants were able to describe their condition and provide knowledge about the label of their condition and their related symptoms (Babooram et al., 2011; Meulenkamp et al., 2008; Veldtman et al., 2000). In the study by Veldtman and colleagues (2000), the explanation of the heart condition provided to/by participants was not related to age, gender, or disease complexity. In the quantitative studies in Table 2, there were mixed results for the construct of identity on outcome measures. For example, Edgar & Skinner (2003), Gray & Rutter (2007), and Jones et al, (2014) found that identity was related to depression, lower quality of life, and adherence respectively. The remaining eight studies including an assessment of identity did not find a relationship with the primary outcome measures or with other variables (Bucks et al., 2009; Chong et al., 2012; Chong et al., 2013; Gaston et al., 2012; Law, 2002; Law et al., 2002; Paterson et al., 1999; van der Velde et al., 2011). However, these studies did not take into account the contribution that identity may have on the other domains, or how inter-correlation of these domains may be associated with the measured outcomes.

The results for identity suggest that symptom attribution as well as knowledge of the disease may not only be captured by the construct of identity. Those studies in Table 2 that utilised IPQ-R and the BIPQ had the opportunity to assess illness coherence, which would allow for better assessment of the clarity of knowledge about a condition. However, only one of the eight studies that included an assessment of illness coherence found that alongside other illness perceptions, coherence contributed to adherence, metabolic control as measured by HBA1c, and quality of life (Fortenberry et al., 2014). Contrary to the previous finding, van der Velde et al, (2011) found that lower scores in illness coherence contributed to a larger difference between parent-child reports of health-related quality of life of the adolescent. As these are currently the only two studies investigating these items, it is not possible to reach a conclusion about the assessment of knowledge and symptom attributions.
Theoretically, the identity and coherence of an illness would contribute to behaviours and/or other illness perceptions, for example, Edgar and Skinner (2003) found identity and coherence was associated with consequences. However, this hypothesis is not supported by the literature using adolescent cohorts aged between 11 and 16 years. This brings into question the suitability of either the theory or the assessment of the constructs within this age group.

2.9.2.2. Cause

Only 11 of the 26 studies in Table 2 identified included an assessment of cause. Studies such as Cordingley et al, (2012), Meulenkamp et al, (2008), and Paterson, Moss-Morris & Butler (1999) have highlighted the importance of assessing causal beliefs of conditions. It is evident from these studies that gender, disease duration, type and source of referral, (Cordingley et al., 2012), or experiences (Paterson et al., 1999), influence causal beliefs. Furthermore, causal beliefs can affect emotions, the adolescents’ coherence of their conceptualisation of the condition, and their sense of control over the condition (Meulenkamp et al., 2008). Gray & Rutter (2007) demonstrated that causal attributions were associated with both outcome and coping. They reported that the relationship between causal attributions of risk factors and physical functioning was mediated by maintaining activities (Gray & Rutter, 2007). As the other seven studies did not report any significant associations between causal attributions and outcomes it is hard to draw any conclusions on the role these beliefs have, or how they are assessed.

2.9.2.3. Timeline

The two constructs in the overarching domain of timeline were not measured consistently due to the different assessments of the domains. Of 19 studies in Table 2 that included a measure of timeline, only seven included both the chronicity and cyclical constructs. However, only two studies found significant results for cyclical
beliefs; Jones et al, (2014) demonstrated cyclical beliefs contributed to predicted adherence, while Fortenberry et al, (2014) showed that lower cyclical beliefs were associated with better quality of life. The cyclical construct demonstrates the ability to convert an abstract concept (such as time) into a concrete concept (such as a pattern or cycle with an identified trigger), which may reflect the cognitive developmental stage of the adolescents.

Taking into account the results linking chronicity and change over time, one possible conclusion regarding this construct is that an abstract concept might be better understood as the adolescents continue to develop. In an investigation of trajectories over time, the beliefs about chronicity increase over time for adolescents with diabetes and the beliefs about the cyclical nature of the diabetes scores decrease (Fortenberry et al., 2014). This modification in understanding beliefs alludes to knowledge about the condition increasing and learning that they have an underlying LTC. This proposition may also account for the significant difference in parent-child dyads’ beliefs regarding the duration of diabetes demonstrated by Gaston, Cottrell & Fullen (2012). Experience of time and of the condition may affect the perception of the duration of the condition. For example, the experience of having asthma had an effect on the beliefs regarding the duration of the disease (Paterson et al., 1999). Beliefs regarding duration of the cystic fibrosis mediated the relationship between age and adherence (Bucks et al., 2009). Finally, the understanding of the duration of congenital heart disease improved with age (Veldtman et al., 2000). The findings of these studies highlight the need to measure the interaction between the chronicity and cyclical concepts within the overarching domain of timeline, and evaluate how this might be relevant to this age group.

2.9.2.4. Consequences

There were only two studies in Table 2 that did not include an assessment of the construct of consequences in their measure of illness representations; however, this was because these studies had specific aims addressing cause (Cordingley et al.,
2012) and control (Fortenberry et al., 2011). The remaining 24 studies that included the consequences construct but reported mixed results in the role of this construct related to illness. Those studies reporting significant results provided evidence of the theory-driven hypotheses suggesting that there is a strong belief that the impact or severity of the condition will be related to poorer outcomes (Law et al., 2002; Edgar & Skinner, 2003; Fortenberry et al., 2014; Nouwen et al., 2009; Skinner & Hampson, 1998; Skinner et al., 2000; Walker et al., 2004). Despite this, the relationship between higher scores of beliefs about consequences and positive outcomes may be mediated by coping behaviours. This was evident in Edgar & Skinner’s (2003) study where the perceived impact was associated with depression. However, when using the coping strategy of cognitive restructuring, perceived greater impact predicted lower symptoms of depression. Another positive outcome associated with high scores in the beliefs regarding consequences was found where the full adherence to appointments of adolescents with mood disorders, and this was associated with higher scores of consequences (Munson et al., 2010). Beliefs about consequences have been identified as an important domain of the CS-SRM either through assessing illness severity, perceived impact, adversity, or beliefs regarding the threat to health. The different ways of defining and assessing beliefs regarding consequences make it more challenging to compare the results and evaluate this construct.

2.9.2.5. Control and curability

The control and curability construct was included in 24 of the studies, with 14 studies in Table 2 reporting significant results for either personal or treatment control. The studies including treatment control provided evidence that high treatment control is related to positive outcomes (Bucks et al., 2009; Croom et al., 2011; Fortenberry et al., 2011; Fortenberry et al., 2014; Gray & Rutter, 2007; Munson et al., 2009; Nouwen et al., 2009; Skinner & Hampson, 1998; Skinner et al., 2000). Despite including personal control and treatment control, 11 studies reported results for treatment control with only four studies reporting results for personal control (Chong et al., 2012; Croom et al., 2011; Law et al., 2002; Fortenberry et al., 2014). Higher
life satisfaction (Chong et al., 2012), perceived physician-patient relationship (Croom et al., 2011), and adherence (Fortenberry et al., 2014) were significantly associated with personal control. However, due to the lack of results with personal control, this relationship may not be assessed in a suitable manner and will require further research. The construct of personal control is similar to the concept of the self-efficacy. Hughes, Berg & Weibe, (2012) measured self-efficacy and found that it was associated with metabolic control and negative affect. Considering the importance of empowerment for self-management, this is an area that will require further development. In a recent review of the relationship between illness beliefs and self-management in adolescents and young people, there was little evidence of a relationship between personal control and self-management across the studies included in their review (Law et al., 2012). This review highlighted that there may be a difference in the construct of personal control for this age group compared with that of the adults. For those studies that did not find evidence supporting personal control, it is possible there is a requirement for a measure that is more sensitive to change across the age groups as this might be due to a developmental issue (Law et al., 2012; Nouwen et al., 2009). It is a possibility that treatment control beliefs are much more concrete or noticeable for adolescents (Fortenberry et al., 2011).

2.9.2.6. Emotional Representations

Emotional representation is a key pathway in the CS-SRM. Despite this, only 13 studies in Table 2 included an assessment of emotional representations. Only two of those studies did not report any significant results. Adolescents with cerebral palsy who drew illustrations of other people with them as they drew themselves walking also reported lower emotional impact suggesting a link between support and emotional representation (Chong et al., 2013). There were mixed results regarding the hypothesis that more negative emotional representations are associated with, or predictive of, negative outcomes. Lower emotional impact was associated with better life satisfaction (Chong et al., 2012), higher adherence (Fortenberry et al., 2014; Jones et al., 2014), higher metabolic control (Hughes et al., 2012) and higher quality
of life (Fortenberry et al., 2014; Gray & Rutter, 2007). Furthermore, the relationship between emotional impact and quality of life was mediated by the coping strategy of focusing on symptoms (Gray & Rutter, 2007). However, adolescents with mood disorders who reported higher emotional representation also reported full adherence to appointments but also reported feeling stigmatisation (Munson et al., 2009). These studies support the need to measure the emotional pathway in adolescents as this could be a potential target for interventions.

2.9.2.7. Conclusions from review

The studies presented in Table 2 that utilised the CS-SRM as a framework to study illness representations in adolescents aged between 11 and 16 years provided support in utilising the framework. Nonetheless, there were mixed results and limitations, suggesting there is a need for further research into the usability of this framework for adolescents. For example, Gray & Rutter’s (2007) study of children and young people with chronic fatigue syndrome scored the IPQ-R differently and restricted the possible item scores. This modification of the IPQ-R may have biased the results when measuring the internal consistency before removing the items. The longitudinal studies lost participants to follow-up, and did not adjust the baseline to keep the same participants throughout. As a result, it is unclear how illness representations change over time for individuals with an LTC (Fortenberry et al., 2014; Skinner et al., 2000). Additionally, a majority of the quantitative studies had small numbers, and this makes it difficult to determine the statistical significance of the results. Finally, as only five manuscripts utilised all the domains of the IPQ-R, it is difficult to conclude the validity of this assessment for this age group, or the transferability of the domains to the way adolescents conceptualise illnesses. There was a discrepancy between the qualitative work and the quantitative work in terms of how adolescents conceptualise their illness; this may be due to the assessment of the domains. The next step would be to conduct further empirical testing and theoretical development to ensure the suitability of utilising the CS-SRM framework for adolescents.
2.10. Summary

The CS-SRM draws upon cognitive theories and constructs to provide a framework for how individuals conceptualise their illness. Moreover, the CS-SRM provides a theoretical structure to explain how this conceptualisation (illness representations) drives illness behaviours and coping, which can affect clinical outcomes. The CS-SRM is supported by evidence from studies with adults who are living with different LTCs. The majority of studies utilise the IPQ-R to assess the nine proposed constructs constituting both illness and emotional representations. Research with adolescent cohorts that applies the CS-SRM is limited, and conclusions regarding the applicability of both the CS-SRM and the IPQ-R within this age group are unclear. Further work is required to address whether there are specific theoretical developments and modifications required to assess how adolescents conceptualise an LTC.
Chapter 3: Research Aims & Objectives

The arguments presented in the preceding chapters show that there is a need to investigate the conceptualisation of long term conditions for adolescents and how they influence the related illness behaviours. In adult populations the representations of long term conditions and behaviours have been studied within Leventhal’s Common Sense Self-regulatory model (CS-SRM). This model has been successfully applied and shown that illness representations predicted and explained significant proportions of the variation in disease outcomes. To date, however, this model has not been applied systematically to address illness representations and coping with adolescents with juvenile idiopathic arthritis.

3.1. Overarching research question

The overarching research question is to investigate whether Leventhal’s CS-SRM can be modified and applied for use with adolescents with a long term condition. To do this the work in this thesis addresses two broad aims. Thus the first aim is to examine the extent to which Leventhal’s Common Sense Self-Regulatory theory of illness representations effectively describes the ways in which adolescents understand their juvenile idiopathic arthritis and accounts for their illness behaviours.

The second aim is to apply the revised model to develop a new theory-based measure that can be used to assess the illness representations of adolescents with a long term condition.

3.2. Aim 1: Suitability of CS-SRM

To assess the suitability of the common sense self-regulatory model (CS-SRM) for use with adolescents with a long term condition, in this case juvenile idiopathic arthritis (JIA).
To accomplish this, the three components of the CS-SRM will be analysed individually as shown in Figure 6. These are illness representations, coping procedures, and appraisal stages. The objectives to be addressed within the first aim are listed below under each component.

(i) The specific objectives of the illness representation component are to:
   1. Identify whether or not the domains of emotional and cognitive representations identified by Leventhal et al (1984) in the CS-SRM are relevant to adolescents with JIA.
   2. Assess the suitability of the current measure of illness representations for adolescents with JIA.

(ii) The specific objectives of the coping procedures component are to:
   3. Identify adolescents’ goals and the coping strategies they employ to deal with pain related to JIA to determine the relationship between intention and behaviour.
   4. Identify the emotional and cognitive profiles associated with specific coping goals.

(iii) To investigate the self-regulatory component of the CS-SRM the specific objective is so to explore the dynamic relationship between illness beliefs and illness outcomes longitudinally by:
   5. Identifying the predictors of behaviour.
3.3. Aim 2: Assessment of representations in adolescents

The second aim is to develop the best method of assessing the illness representations of adolescents with a long term condition. The results of the first aim provide the basis for adapting the IPQ-R to create a form that can be used to assess adolescents’ illness representations. The specific objectives are to:

6. Develop an adolescent specific measure of illness representations.

7. Test and evaluate the psychometric properties of the new measure.

3.4. Assessment of Children’s Experiences – Sub-Study of the Childhood Arthritis Prospective Study (ACE-CAPS)

To address the aims and objectives reported in Section 3.1 and 3.2, two sets of data were analysed. One set was drawn from the national Childhood Arthritis Prospective Study (CAPS), a national cohort of observational survey and clinical data (Hyrich et al, 2010). CAPS was primarily designed as a prospective observational cohort to identify the predictors of outcome from childhood onset inflammatory arthritis. To
observe both short and long term predictors, including socio-demographic, clinical, psychological, laboratory, genetic, and treatment factors, baseline and follow-up data were collected annually from diagnosis to up to 10 years follow-up. A subset of the data collected by CAPS since 2003 was used to address Objective 5, specifically the questionnaires assessing the psychological predictors of disease outcomes. More information on the questionnaires is provided in Appendix 1.

The second set of data was drawn from a new study called ACE-CAPS. The acronym ACE stands for Assessment of Children’s Experience. The author (DG) designed this study and led the application to the NHS ethics committee for a substantial amendment to the original CAPS application. This was granted on the 14/12/2010 by the North West Research Ethics Committee (REC: 02/8/104). The aim of the ACE-CAPS sub-study was to explore the appropriateness of tools that measure adolescents’ experiences of JIA. The first phase of this sub-study required the design of a qualitative cross-sectional study of 21 young people aged between 11 and 16 years. Data collection commenced on 29 March 2011 and the last participant was interviewed on 24 August 2011. One component of analyses of the ACE-CAPS data was reported as part of the author’s Masters of Science dissertation. That analysis covered adolescents’ control perceptions and emotional representations of their JIA and findings are cited in Chapter 2 of this thesis. In this second phase of the ACE-CAPS study the author analysed the complete qualitative data she collected in 2011 to address objectives 1 to 4 of this thesis. In the next chapter there is a description of the measures (see section 4.2.2.4 and section 4.2.3.4) and procedures (see section 4.2.2.5 and section 4.2.3.5) used in the 21 interviews conducted. More details about this data set are contained in Appendix 1.
Chapter 4: Methods

4.1. Overview

The following chapter describes the methods selected to address each aspect of this thesis’ broad aim of applying Leventhal’s common sense self-regulatory theory to adolescents. This chapter discusses the mixed-methods employed to address the two sub-aims of this thesis. The rationale and theoretical underpinning of the methodology, analysis and design of each study is provided.

Sections 4.2.2 to 4.2.4 of this chapter describe the studies conducted to address the first aim of this thesis. These studies aim to investigate the suitability of the CS-SRM. The studies discussed in Sections 4.3.2 to 4.3.3 address the second aim of this thesis that is, to develop a tool for assessing illness representations of adolescents.

4.2. Suitability of the CS-SRM (Aim 1)

The first aim was to assess the extent to which each of the three components of the common sense self-regulatory model (CS-SRM) can be applied to adolescents with juvenile arthritis. Section 4.2.1 describes the research paradigm selected to address the application of the model. Qualitative studies were utilised to explore the illness representations and the illness behaviours of adolescents with JIA and quantitative methods were utilised to explore the dynamic relationships between illness beliefs and behaviours. In this section the methods selected for each objective are described under the particular component of the model.
4.2.1. Research Paradigm

To address the first aim of this PhD a triangulation strategy was applied by using both qualitative and quantitative data collection and analysis (mixed-methods) to investigate components of the CS-SRM model (see Figure 7). Mixed methods is defined as an approach to combine and integrate research at either the collection or analysis using methods of both quantitative and qualitative methods in either one single study or in related studies as proposed in this PhD. Findings from each methodological approach were given equal status in terms of their contributions to evaluate the application of the model to understand the experiences of adolescents’ with a long-term condition (Creswell, 2003; Tashakkori & Teddlie, 1998; Tashakkori and Creswell, 2007).

A key challenge to the idea of integrating quantitative and qualitative methods is the perceived difference in epistemological origin of the data. Whilst it can be argued that quantitative methods traditionally assume that knowledge gained from data collection is about the ‘real’ world which is separate to individuals’ perception of the world, qualitative methods assume that knowledge is made up of different understandings of the world. These different perspectives, one being a realist and one
being an interpretive perspective, have tended to drive different approaches to ‘knowledge’ and how to analyse the data. One key assumption of a realist approach is that knowledge is to be measured objectively to remove the bias and error, exposing the true knowledge. On the other hand, an interpretative approach would assume that knowledge is an outcome of novel understanding depending on socio-cultural context. Others have argued for a less clear-cut differentiation. Henwood and Pidgeon for example, argue that all analysis can only provide a ‘partial’ view of reality and view qualitative and quantitative research as residing on a continuum (Henwood and Pidgeon, 1992). The key issue that separates the methods is the point at which the ‘data-reduction’ process occurs. With quantitative analysis there is early data reduction in the form of reducing of complex phenomena into operationally defined variables. With qualitative approaches the reduction occurs at the end in the form of themes and codes.

Recently, the debate about the relative merits and roles of qualitative and quantitative methods has become less salient with researchers (particularly those in applied health settings) taking a more pragmatic approach by, recognising that the appropriate methods are required to address different research objectives (Yardley and Bishop, 2015). Yardley and Bishop (2008) have defined a pragmatic approach as one which acknowledges the epistemological differences but ultimately aims to produce knowledge that has valuable external consequences or value. For research in health psychology and health services research, the external consequences of research would include an improvement in public health services by targeting specific health behaviours or else an improvement in patients’ quality of life (Bishop, 2015). Within a pragmatic approach the researcher has the freedom to pick the design that is best suited to provide the data to answer the complex research questions (Bishop, 2015). These may include a range of qualitative and quantitative methods or techniques to develop and test a new questionnaire, testing hypotheses developed from qualitative data and theories, and testing feasibility or usability in a clinical trial (Bishop, 2015, Creswell, 2003). Taking this pragmatic approach has allowed mixed methods researchers to address the complex research questions which emerge in applied
health services research settings (O’Cathain et al, 2007). For this current work the use of both approaches was necessary to address the complex, multifaceted issues involved when evaluating the applicability of Leventhal’s CS-SRM to adolescents with a long-term condition. This included evaluating and modifying the theoretical approach to take account of developmental issues relevant to adolescents, and then using the new understanding to inform the design of a new measure.

Integrating different methods can occur at different points of interface, either at sampling, data collection, or at data analysis or at interpretation phases of research (Morse and Niehaus, 2009). The two overarching aims of this PhD are each addressed using two complementary sets of mixed-methods studies. To address Aim 1, two separate studies were utilised to collect data for objectives one to five of this thesis. For objectives one through to four a qualitative study was set up. The study was interview-based, utilising two different interview approaches. These interview approaches are described in detail in Section 4.2.2.2 and Section 4.2.3.2. To address the fifth objective of Aim 1, longitudinal quantitative data were utilised that were collected via the Childhood Arthritis Prospective Study (CAPS) a multisite national longitudinal inception cohort study of JIA. The point of convergence of the findings from objectives one through to five for this study occurred at interpretation stage to address the first Aim of the PhD.
4.2.2. CS-SRM: Illness Representations

The investigation into the illness representations of adolescents with JIA entails two objectives. The first objective was to assess whether or not the existing domains of emotional and mental representations are equally relevant to adolescents with JIA. The second objective was to assess whether the current measure of the existing domains, the Revised Illness Perceptions Questionnaire (IPQ-R; (Moss-Morris et al., 2002) was able to capture the illness representations of adolescents with JIA.

To achieve these objectives semi-structured interviews were conducted. Cognitive interviewing was selected as the method to elicit data about illness representations. In Section 4.2.2.2.1 the methodological justification for using cognitive interviewing with adolescents is described.

The first objective in this investigation of illness representations was to verify the relevance of existing domains. Thus framework analysis was applied to this dataset. The second objective was to assess the content validity of the questionnaire. For this, latent content analysis of the items and their endorsements was conducted. A rational for the use of these qualitative methodologies can be found in Section 4.2.2.7 for framework analysis and Section 4.2.2.8 for content analysis.

4.2.2.1. Design

For this analysis, data were collected through one-to-one interviews with adolescents with JIA using the cognitive interviewing approach (Willis, 2004). The data collected were used to develop the illness representation dataset.

Additional data collected during these one-to-one interviews were used to develop the pain dataset described in Section 4.2.3. Interviews lasted between 45 and 75 minutes.

4.2.2.2. Methodological Considerations – Interview

The approach utilised for this investigation of illness representations was cognitive interviewing. Cognitive interviewing is a method used to elicit and assess the
cognitive processes individuals are engaged in when they are answering a questionnaire (Willis, 2004; Willis et al., 1991). Using this approach enables an exploration and understanding of the reasoning and understanding an adolescent uses when answering the Revised Illness Perception Questionnaire (IPQ-R). The following Section 4.2.2.2.1 describes the theoretical underpinnings of this interview approach and provides a description of how this has been applied with adolescents.

4.2.2.2.1. Cognitive Interviewing

A number of cognitive processes are involved when answering a questionnaire. These cognitive processes include: the person’s comprehension of an item on a questionnaire (question intent and meaning of terms), their retrieval from memory of relevant information (recallability of information and recall strategy), the decision process involved in providing a response (taking into account an individual’s motivation and sensitivity/social desirability) and the response processes (mapping their own response to the responses available in the questionnaire). Responding to items in questionnaires may trigger some or all of these cognitive (mental) processes; this is highly dependent on the types of items. The aim of cognitive interviewing is to prompt an individual to reveal information about the types of mental processes they are utilising, and thus indicate the mental constructs they are drawing on in order to formulate their responses. There are two sub-types of cognitive interviewing methods: think-aloud and verbal probing (Willis, 2004).

Think-aloud is a very specific activity which requires the interviewer and interviewee to practice and train to understand the procedure. The aim of this method is to stop filtering in the process of answering. This allows the researcher to identify and analyse the cognitive processes mentioned above. There is a protocol to follow that begins with the following instructions:

“Try to imagine the place where you live, and think about how many windows there are in that place. As you count up the windows, tell me what you are seeing and thinking about.”
In the interview procedure, the interviewer can use generic probes to remind the interviewee to continue with the activity. Generic probes such as “What are you thinking about” are thought to reduce interviewer-imposed bias (Willis, 2004).

This method of interviewing has been derived from psychological procedures (Ericsson & Simon, 1980) in order to determine what people are able to report. People can only report certain components of high level mental processes that are temporally stored in their working memory (for example the sequence of steps that leads to the solution of a problem). Ericsson and Simon (1980) identified thinking aloud as an approach to map directly what an individual reports to that individual’s engaged mental processes. Directly mapping the reported thoughts to the mental process there is an indirect mapping of the internal stages of mental processing. This is one way of accessing a detailed breakdown of the mental processes. However this method also allocates the cognitive burden of interviewing to the participant (interviewee).

Verbal probing is an alternative to the think-aloud technique, where after the item is read the interviewer further “probes” to ask for specific information relevant to the item. The verbal probes can be directly after the item is addressed (concurrent probing) or they can be left for the end of the interview (retrospective probing). There are six categories of verbal probing: comprehension/interpretation (terms), paraphrasing, confidence judgement, recall specific, and general probes. Probes can be scripted or spontaneous, and there are advantages and disadvantages associated with each. One of the biggest disadvantages is introducing bias to the interviews; the interviewer has to ensure the verbal probes are not leading questions. Scripting verbal probes can allow for a structured protocol while spontaneous probes can develop throughout the interview that can allow an investigation in issues previously not anticipated. Willis (2004; 1991) maintains that the most effective cognitive interviews utilise verbal probing which incorporate combinations of the different types.
Cognitive interviewing with adolescents

There are a limited number of studies that have utilised cognitive interviewing with adolescents of the age group between 11 and 16 years. It has been assumed that the think-aloud approach is easy for children and adolescents but studies have shown some problems with the think-aloud method when applied to adolescent respondents (Strussman et al., 1993; Zukerberg & Hess, 1996). Those studies that utilised this method concluded that this age group are capable of the tasks in think-aloud and their language skills are sufficiently developed (de Leeuw et al., 2004). It is believed that from age 7 children can be introduced to structured questionnaires. By this age children’s reading skills are acquired, their language skills are expanding (Nelson, 1976) and they are able to conceptualise different points of views (Selman, 1980).

The changes occurring in cognitive development of children between ages of 7 and 11 imply improvements in tasks such as recall, reality monitoring, and improved effectiveness of memory strategies. These are all skills required in cognitive interviewing. From the age of 11, there is an increase in the memory capacity in adolescents and they are capable of manipulating ideas about hypothetical situations (Conger & Galambos, 1996). However, at this stage an adolescent is particularly context sensitive and this has been shown to make an adolescent within the age groups between 11 and 18 vulnerable to peer pressure and pressure to conform to social norms (Gray, 2002). This has implications for emphasising and explaining confidentiality to an adolescent in an interview setting. When adolescents take part in cognitive interviewing they require more extensive probing than adult participants (Strussman et al., 1993; Zukerberg & Hess, 1996). Therefore, detailed protocols are required to include probing questions in combination with the think-aloud approach. A mixed-approach of both probing questions and think-aloud method was applied to the current study. A description of the interview schedule is provided in Section 4.2.2.4 and is presented in Appendix 2.
4.2.2.3. Participants

The participants were recruited from a concurrent study, the Childhood Arthritis Prospective Study (CAPS). CAPS is a multi-centre study set up in 2001, designed as a prospective, longitudinal observational cohort to identify predictors of outcomes from childhood-onset inflammatory arthritis. Baseline and follow-up data are collected over ten years from onset in order to observe both short and long-term predictors, including socio-demographic, clinical, psychological, laboratory, genetic, and treatment factors (Hyrich et al., 2010).

Participants were eligible for the current study if they were aged between 11 and 16 years and had a clinic appointment at the participating centre in the North West of the UK. A research nurse identified the eligible patients and initiated the recruitment of interviewees. Out of the eligible participants that had a clinic appointment between March 2011 and May 2011 that were contacted by the research nurse, twenty-five contact forms were returned. A purposive sampling strategy was used to recruit equal numbers of adolescents aged between 11 and 13 years and adolescents aged between 14 and 16 years. The aim was to ensure representation of both older and younger adolescents. Twenty-one interviews were organised by the author during this time period because the other four participants cancelled their clinic appointment and consequently cancelled their interview. One participant completed only the cognitive interviewing of the IPQ-C because of an eye inflammation and one participant could not complete the cognitive interviewing task because of time constraints. Therefore, in total 20 cognitive interviews took place.

4.2.2.4. Materials

*Interview Schedule-cognitive interview*

The author was trained in interviewing children via a training programme “Listening to Children and Young People using a Person-Centred Approach” (Rogers, 1951) prior to the interviews. Further training took place in using think-aloud and verbal probing approaches. The method adopted for this qualitative study was a combination of think-aloud with inserted generic probes. The aim was to encourage
most of the interview to be think-aloud but still have an assortment of verbal probes available should they be needed. Before the interviews commenced, the protocol of think-aloud was explained to the participant and the method was practised. Further description of the procedure is provided in Section 4.2.2.5. The items of the Illness Perception Questionnaire Child Version (IPQ-C) were read out and the participant provided their endorsement of the item while also undertaking the thinking-aloud task.

**Illness Perceptions Questionnaire – Child Version**

The Illness Perception Questionnaire Child version (IPQ-C) is an adapted version of the Revised Illness Perception Questionnaire (IPQ-R) devised by Moss-Morris and colleagues (2002) (discussed in Section 2.9). The IPQ-C version was modified for use in the CAPS in 2004, and patients aged between 11 and 16 years have been completing this version of the IPQ-R. The IPQ-C version was modified with consideration to the reading ages of 11 to 16 year olds (for the table of the changes with respect to the IPQ-R see Appendix 3). The items in the IPQ-C are similar to the IPQ-R in that the items are statements of beliefs and the responses to these statements are endorsement scales from strongly disagree to strongly agree. As described in Chapter 2, the IPQ-R has nine domains: identity, timeline chronicity and cyclical, treatment and personal control, illness coherence, consequences, emotional representation, and cause.

**Clinical Characteristics**

To report the clinical characteristics of the participants interviewed, the author gained access to the CAPS database. The information had been collected within six months before or after the interview date. The clinical characteristics included: disease duration, subtype of condition using the ILAR classification of JIA, disability index, and pain score. The disease duration for the participants of the interview was calculated by subtracting the baseline year from the interview year. CAPS also collect copies of their participants’ medical notes completed by a consultant. JIA
subtypes were assigned by paediatric rheumatologists according to ILAR classifications.

**Disability Index**

For the purposes of this study, disability was defined according to the patient’s perceptions of their daily life using the Childhood Health Assessment Questionnaire (CHAQ; (Singh et al., 1994). This assessment was developed as a disease specific, self-report, disability measure for children with JIA. The CHAQ was an adaptation of the Stanford Health Assessment Questionnaire (HAQ; (Fries et al., 1980) that is extensively used within adult arthritis studies. Singh and colleagues’ (1994) rationale for adapting the HAQ was to enable the study of function longitudinally from childhood into adulthood.

The CHAQ (Singh et al, 1994) was adapted and validated in a British cohort, to change the original American-English to British-English (Nugent et al., 2001). The British version was validated for a UK population with 219 JIA patients and 221 healthy children. It is this British version of the CHAQ that is used in this study.

Like the HAQ, the CHAQ has different domains that measure function in 8 areas of daily life: dressing and grooming, arising, eating, walking, hygiene, reach, grip and activities. There are three questions in each domain: ratings of degrees of difficulties from zero (without difficulty) to three (unable to do); reported use of aids or devices; and if they require assistance from other people. The total Disability Index is calculated by adding together the highest score of each of the eight domains and then dividing the sum by eight to give an average score on a scale between zero and three.

Twenty health professionals (7 rheumatologists, 3 child psychologists, 2 developmental paediatricians with the remainder being nurses, social workers, occupational and physical therapists) and parents of 22 healthy children were recruited to evaluate the face validity of the first version of the adapted HAQ for children. After modifications the CHAQ’s validity and reliability was tested with
completed CHAQ from 72 children aged between 1 and 19 years with JIA (Singh et al., 1994).

**Pain Score- Visual Analogue Scale**

A pain score using a visual analogue scale (VAS) is collected to measure discomfort in CAPS participants. The VAS is a 100 mm to measure pain severity where 0 represents no pain and 100 represents very severe pain as shown in Figure 8. The Pain VAS is found at the end of the CHAQ and a copy of the score was kept in the medical records.

![Figure 8: A pain VAS as included in the CHAQ.](image)

4.2.2.5. Procedure

Eligible participants were identified and then sent information about the interview study (see Appendix 4). Potential participants were contacted and the interview for each was arranged to take place before or after a clinic appointment. At the clinic, introductions to the adolescent and their parent/guardian were made by one of the research nurses, CL or OL. This was a crucial stage for building a rapport with the adolescent and essential for successful cognitive interviewing. An empty clinic room was used for the interviews. Most of the parents/guardians remained in the clinic room during the interviews and completed a questionnaire while their child was interviewed.

The cognitive interviews began with clear instructions about think-aloud and the adolescent was allowed a practice run through. The cognitive interviews had a combination of think-aloud and verbal probing. For the interviews, a probing protocol was developed. However, probes were also inserted to encourage think-aloud and to clarify issues (Levine & Huberman, 2002). After the tasks were
completed, there was a chance to debrief the adolescent by asking if they had any questions or anything to add. The adolescent was reminded that they have the contact information on their sheets for any future questions.

_Ethical Considerations_

The information sheets were sent out to the eligible participants, but were also presented at the beginning of the interview session. This was an opportunity to ensure the adolescent understood the tasks and had the opportunity to ask questions. This also allowed the adolescent to complete the assent and/or consent forms. Parents signed consent forms for those adolescents who were under 16 years. All interviews were recorded and these recordings were deleted from the recorder once transferred to a password-protected computer. For confidentiality reasons the participants were provided with a pseudonym.

The North West Research Ethics Committee (REC: 02/08/104) granted a substantial amendment application for these interviews on the 13/01/2011. This amendment included a sub-study for a group of adolescents aged between 11 and 16 years.

4.2.2.6. Methodological Considerations – Analysis

Two qualitative methodologies were selected to analyse the illness representation dataset. In this current Section, these qualitative methodologies of framework and content analysis are described.

4.2.2.6.1. Framework Analysis

In order to address the first objective of this thesis, framework analysis was used to analyse the data collected from the cognitive interviews described above (Section 4.3.5). Framework Analysis is a flexible analysis that uses deductive and inductive approaches simultaneously to complex data (Ritchie & Spencer, 1994). Qualitative approaches may differ across key features, including the status of the data and how it is retained in context as well as how the data is accessed and displayed, the primary
focus of the analysis, the kind of concepts generated and how they are applied, the choice of the level of abstraction, the treatment and status of the categories and lastly the place of the researcher in the analytical account (Ritchie and Lewis, 2003). Framework analysis is a matrix based method which aims to order the data to facilitate interpretation of the complex datasets. This involves two linked stages: data management and sequential data interpretation. This type of research using framework analysis was first developed to use in large-scaled policy research, but has been applied to health research (Gale et al, 2013).

There are five phases in framework analysis that are presented in Table 3. These are not linear phases and this process may require repeating phases until there is a thematic framework that includes all data. This “back and forth” through the phases would allow access to different levels of abstraction without losing access to the original or ‘raw’ data for example the transcripts and the participants’ own words. This flexibility allows both deductive and inductive approaches to analysing the data. The thematic framework is the means by which the data are organised in a matrix where the columns are the themes and the rows are each participant. Organising the data in this manner allows for integration at an individual level but also at the level of the themes overall. After the construction of a conceptual framework the rest of the data is indexed with it. The matrix framework allows for a systematic reduction of the data to analyse by the theme as well as by the case/participant. Both ‘top-down’ and ‘bottom-up’ approaches to data interpretation were undertaken. Top-down refers to the use of the a priori theory (in this case CM-SRM) to interpret the data, and a bottom-up analysis allows the researcher to engage with data which may not fit within the theory but may still be relevant. In this way the approach is sufficiently flexible to include novel themes or unanticipated issues in the analysis.

There are three major levels of interpretation (Kvale, 1996), and in each of these levels of abstractions the analyst should be aware of their own experiences and biases. The first level is the context of self-understanding, where the analyst formulates what each participant means by the speech and words selected. The second level is placing these words and speech in a more general context using
general knowledge using critical common-sense to understand the context of the data. The third level is the theoretical understanding, placing the data and the context into a theoretical perspective, using theoretical work to understanding the speech and data from the participants. Distinction between these levels can be difficult, as the process of theory development can be both through the choice of interpretive approach and the selection of meaning and interpretation (Ritche and Lewis, 2003). This analytical approach is most commonly used to analyse data collected through semi-structured interviews, which allows the data to be categorised and to be compared and contrasted. While a highly systematic method is used to organise the data, this qualitative approach still requires the rigour and reflexivity expected in qualitative analysis to interpret the data.

This approach was applied to the data collected using the cognitive interviews because of the flexibility it allowed in utilising an established theoretical framework in this case the CS-SRM. The data collected were organised and indexed according to the nine domains that were assessed by the questionnaire. This allowed for an exploration and identification of the pre-determined illness representation constructs and their applicability to adolescents. Theoretically, the domains or themes in the conceptual framework of the CS-SRM are interdependent (Leventhal et al, 2012; Moss-Morris et al, 2002). Framework analysis has the ability to accommodate homogeneous data, which allows for the categorization of the data within thematic frameworks (Gale et al, 2013; Ritche and Lewis, 2003) for this reason, framework analysis can be helpful in applied health services research. This ability to accommodate homogeneous data also makes this analytical approach ideal for this analysis in this PhD for developing a framework based on the conceptual framework of the CS-SRM. One of the strengths of using framework analysis is the ability to use the analysis to undertake appropriate theory modification by either adding to or challenging existing components of the theory. This flexibility allows using the matrix as a tool for a generation of themes from the data.
Table 3: Phases of framework analysis and the definitions of each phase.

<table>
<thead>
<tr>
<th>Phases and definitions</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Familiarisation</strong></td>
</tr>
<tr>
<td>Analyst becomes immersed in the data. Reviews and lists key ideas and recurrent themes</td>
</tr>
<tr>
<td><strong>Identifying a thematic framework</strong></td>
</tr>
<tr>
<td>Analyst uses the indexes and priori issues to identify analytical themes and categories.</td>
</tr>
<tr>
<td><strong>Indexing</strong></td>
</tr>
<tr>
<td>Systematically applying the categorical themes to the data.</td>
</tr>
<tr>
<td><strong>Charting</strong></td>
</tr>
<tr>
<td>Organising the data (cut and paste) from the original transcripts into the categories.</td>
</tr>
<tr>
<td><strong>Mapping and interpretation</strong></td>
</tr>
<tr>
<td>Analyst identifies key characteristics of the charted data reporting ‘patterns’ or ‘typologies’ in the categories.</td>
</tr>
</tbody>
</table>

4.2.2.6.2. Content Analysis

To address the second objective of this thesis (see section 3.2), cognitive interview data were analysed to determine the content validity of the Revised Illness Perception Questionnaire (child version) when used with adolescents. As such, a content analysis methodology was selected to frame a comparative analysis of the endorsements provided by the participants and their think-aloud data. To utilise this approach successfully, the researcher needs to specify the content characteristics (codes) and apply explicit rules for identifying and recording these codes (coding manual). As with a framework analysis, this type of analysis is used to develop a coding manual in order to define the codes to index and chart the data. A sample of the data is used to develop the coding manual in an iterative process to then quantify the frequency of the identified codes. A coding manual was developed for this illness representation dataset and is described in Table 4.

Content analysis has been defined as a “research technique for making replicable and valid inferences from texts (or other meaningful matter) to the contexts of their use.”
(Krippendorff, 2013, page 104). It is a qualitative method that aims to identify the frequency of codes systematically in order to analyse and make inferences about a specific dataset (Krippendorff, 1980). This definition of content analysis does not specify what the content to be analysed may be. According to the literature it can be categorised into three categories, first, content which is contained in a text (manifest), second, content which is dependent on the source (underlying aim of the text; e.g. aim of a questionnaire) third, content that emerges from an analysis of the text (latent) (Krippendorff, 2013).

The category of content identified has implications for the type of analysis undertaken with the available data. Manifest content analysis focuses on the nature of the content within the actual text; the assumption taken in this approach is that what is said maps directly to what is implied. For this type of content, analysts identify words or phrases present in the text and use these as the codes. In contrast, latent content analysis approach goes ‘beyond the text’ and incorporates a number of important assumptions or features to inform the analysis. These include specific assumptions that are salient to the analysis undertaken in this thesis, namely that meanings are dependent on perspectives of the reader or the giver, that texts or data can evoke meanings that go beyond the words provided. In this sense it is recognised that qualitative data have implicit representations. Researchers using latent content analysis often recognise that meaning of data can be dependent on context and intent of both the informant and the analyst. Therefore, the analyst aims to make systematic inferences of the underlying meanings from the data collected.

Taking these assumptions into consideration, latent content analysis was identified as appropriate in order to address the second objective of this thesis, to investigate the suitability of the current measure of illness representations for adolescents with JIA. Latent content analysis was used to code and classify the difficulties or challenges experienced by adolescents completing the measure. This approach allowed sufficient flexibility to infer meaning, for example, gaps in the think-aloud process, and the need for verbal probes. The approach allowed for a systematic but flexible approach to data reduction (Schreirer, 2012). For this study the process included the
endorsement of items, the item itself and the context of the endorsement provided by
the adolescent. Thus codes developed for this analysis were driven by the data
(Schreirer, 2012).

4.2.2.7. Framework Analysis for Illness Representations Dataset

For the illness representations dataset, framework analysis was utilised due to using a
pre-existing framework outlined by the Common Sense Self-Regulatory Model (CS-
SRM; Leventhal et al., 1984). For the illness representations interviews data
management consisted of charting data according to concepts and domains outlined
by the CS-SRM using the software NVivo 10. This CS-SRM framework
incorporated the domains of identity, timeline, consequences, coherence, and cause.
These domains were the ‘parent themes’ of this analysis. During the first charting
phase, the CS-SRM framework was applied to the data. As well, the transcripts were
indexed according to the IPQ-R items of each domain. While the charted data were
being mapped and interpreted, patterns were identified and detected in the CS-SRM
framework categories (major-themes) that were coded to sub-themes. An iterative
process in reorganising the data within the parent themes led to a coherent
description of each construct. A narrative account of the data was produced which
described the data supporting the sub-themes within each parent theme. The narrative
account allowed a comparison of the constructs with the literature supporting the CS-
SRM domains and led to the development of a theoretical framework specific for
adolescents.

4.2.2.8. Content Analysis for Illness Representations Dataset

Analysis of the dataset containing the endorsements of IPQ-C items followed a latent
content analysis approach. Three random participants’ transcripts were taken to
develop a coding manual. The codes were generated from issues that participants
encountered with the items of the IPQ-C. These issues were identified from any
discrepancies in the transcripts of participants’ cognitive interviews and the written
endorsements. The final code manual consisted of five codes found in Table 4.
All of the items were coded either because there was evidence of a problem or no problem in the cognitive interviewing transcripts. Each participant’s transcripts of each item on the IPQ-C and their endorsements were coded according to the coding manual.

Table 4: Code manual developed for the content analysis

<table>
<thead>
<tr>
<th>Code</th>
<th>Definition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Not relevant</td>
<td>the participant claimed the item was not relevant to them</td>
</tr>
<tr>
<td>Confusion</td>
<td>the participant was confused, asked to repeat the question or had to skip it because they did not understand the item</td>
</tr>
<tr>
<td>Incongruent endorsement</td>
<td>the endorsement the participant gave was different to what they were saying to the answer</td>
</tr>
<tr>
<td>Incongruent answer</td>
<td>the answer did not match the question</td>
</tr>
<tr>
<td>No problem</td>
<td>the participant had no problems with the item</td>
</tr>
</tbody>
</table>

4.2.3. CS-SRM: Coping Procedures and Illness Behaviours

In order to investigate the coping procedures and illness behaviours component of the CS-SRM there were two specific research objectives. The first objective was to determine the relationship between intention and behaviour by identifying the goals of that particular coping behaviour, that is, what was the individual trying to ultimately achieve. The second objective was to identify the cognitive and emotional profiles associated with those goals. To collect these data, an interview approach was utilised to explore the adolescent’s pain experiences and how they attempted to cope with that pain episode. Further description of this interview approach is presented below in Section 4.2.3.2 and a description of the interview tool is provided in Section 4.2.3.4.
This was a cross-sectional investigation into coping procedures and illness behaviours and utilised the qualitative methodology of framework analysis. This analytical method was selected to provide a framework created by coping behaviours. This framework was used to organise and categorise the participants’ illness representations data. Further information about this analysis is provided below in Section 4.2.4.6.

4.2.3.1. Design

To investigate coping behaviours the participants that took part in the interviews described in Section 4.2.2.1 completed another task in the interviews. The participants were randomised to the order of the two interview tasks. The adolescents took 20 to 30 minutes to complete this particular interview task.

4.2.3.2. Methodological Considerations – Interview

Qualitative tools have been developed to enable adolescents to discuss experiences that might otherwise be difficult for them to discuss. The use of computers has been found to increase engagement and compliance with assessment (Stinson et al., 2008b; Watson et al., 2002). This medium has been found to empower a child (Calam et al., 2000b). A computer-assisted interview called In My Shoes (IMS) was originally designed in conjunction with the Department of Health to help children communicate different aspects of their experiences (Calam et al., 2000a). The interview tool IMS was utilised in this study to elicit discussion about pain experiences.

4.2.3.3. Participants

The same participants described in Section 4.2.2.3 completed the pain interviews.

4.2.3.4. Materials

For this interview, the data was collected with the IMS computer-assisted interview tool. Overall, IMS is a modular interview that has 10 modules. Since IMS was
designed to provide flexibility in an open-ended process, questions can be developed based on a child’s response. Studies have demonstrated that one of the modules can be utilized in hospital settings during post-operation periods to discuss pain (Watson et al., 2002). Before undertaking the interviews in the clinic, the author (DG) received specific training in utilising three of the modules of the IMS interview tool.

4.2.3.5. Procedure

For this task, the participants completed three of the IMS modules on a laptop: the introduction, the emotion palette, and the somatic experience module (for the screen shot of the somatic experience module which also includes the emotion palette see Appendix 5). The introductory module is used to provide descriptive information about the adolescent. The emotions module is used to provide a discussion about different emotions and how facial expressions should be labelled. Therefore, the aim of this module is for the participants to label a palette of nine faces representing different emotions. The somatic experience module was used to collect information about an adolescent’s pain experiences. An adolescent would describe their pain experiences using key descriptors: location of pain, type of pain, size, and degree of throb. In addition, the participants ascribed a facial expression label to a face that had already been labelled. The interview schedule for this task is in Appendix 2. The questions were designed to use the adolescents’ own language to describe their pain and their pain experiences.

4.2.3.6. Analysis

The coping dataset from the transcripts of the IMS interviews was analysed using a framework analysis. The aim of this analysis was to develop a coping framework. In the first charting phase the coping behaviours that were used were previously identified (Ghio, 2011) to organise the coping data. The coping behaviours were indexed according to the intent for that coping and two initial themes of coping goals were identified. To generate the final thematic framework, the coping goal themes were reviewed for major themes (these are usually labelled as parent themes in the
literature (Ritchie & Spencer, 1994); however, it will continue to be referred to as *major themes* in this work. The final thematic framework identified consisted of two *sub-themes* of coping goals and one *major theme* as a higher order coping goal.
4.2.4. CS-SRM: Dynamic Feedback

The final stage of the CS-SRM is the feedback component. The aim was to examine this component by identifying predictors of behaviour over time. Therefore, a longitudinal model was used to investigate the dynamic relationship between illness perceptions and illness outcomes. This study represented the self-regulatory component of the CS-SRM that proposed the illness perceptions and outcomes would change and inter-correrate over time. The specific hypotheses for this study can be found in Chapter 7 section 7.3.

4.2.4.1. Design

Longitudinal data were used to explore relationships between illness perceptions and behaviours collected by CAPS over three time points: baseline, year 1, and 2. The use of longitudinal data enabled an investigation of the direction of the relationship, as well as the role of mental representations and pain in predicting the physical activity. The variables of emotional representations as measured by the IPQ-C were modelled with physical activity as measured by the Child Health Questionnaire (CHQ-PF50; Landgraf et al., 1996). The CHQ is completed by parent/guardian therefore it can be considered a perhaps more objective observational measure of aspects of their child’s behaviours as opposed to the child’s self-report of their behaviour. Two covariates were included. These were the pain score on the VAS as filled out by the adolescent, and the General Health Questionnaire score (GHQ; Goldberg & Williams, 1988), a measure of parents psychological wellbeing filled out by the parents about their own health and perceptions.

4.2.4.2. Methodological Consideration – Data Management

*Missing Value*

Missing data is a common problem in any dataset; the issues of bias, power and recoverability arise when managing the missing values. These issues are dependent
upon the mechanisms used to capture and generate data that fail to do so in certain cases. There are different statistical methods for imputing values for missing data. The choice of statistical test utilised to impute values for missing data is determined by these mechanisms. The following section describes and defines those mechanisms used to capture data and statistical methods for missing-value imputation.

**Definition of Missing Data**

The missing-data mechanism is the probability that data are missing (M) given the values of the observed and missing data (Rubin, 1976). These mechanisms according to Rubin (1976) can be classified in three categories, 1) missing completely at random (MCAR) 2) missing at random (MAR) 3) missing not at random (MNAR).

1) **Missing completely at random (MCAR)**
   If missing data are missing completely at random then the probability that data are missing does not depend on the observed or missing values.

2) **Missing at Random (MAR)**
   If missing data are missing at random then the probability that data are missing depends on the observed values but not dependent on the missing data values.

3) **Missing not at Random (MNAR)**
   Data that are categorised as missing not at random; the missing data is informative missing data. Therefore, the probability that data are missing depends on the values of the missing data. These missing data are expected through logical inferences about the missing data.

**Missing Value Analysis**

The missing values were explored in STATA 13 software by identifying missing value patterns and the available complete cases when all potential confounders and variables for the longitudinal model are included. For some missing values, the missing data were not missing at random, for example some baseline data were
missing because questionnaires like the IPQ-C were completed by the parents as a proxy for the adolescent age 11, rather than by the adolescent. The questionnaires completed by proxy were not included in the dataset as this might have biased the relationship with the outcome measure of physical activity that was completed by the parent. As shown in Table 5 there were missing values at each time point and these were not always the same participant. Therefore, there were different samples available at different time points. In each time point there was more than half of the data missing. To reduce the sample to only include participants with complete-cases which have data at baseline, time point 1, and time point 2, meant a reduction from 264 participants to 55 participants this meant that there were around 79% missing data.

<table>
<thead>
<tr>
<th></th>
<th>Baseline</th>
<th>Year 1</th>
<th>Year 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total</td>
<td>136</td>
<td>134</td>
<td>148</td>
</tr>
<tr>
<td>Missing</td>
<td>81</td>
<td>79</td>
<td>93</td>
</tr>
<tr>
<td>% Missing</td>
<td>59.6%</td>
<td>59%</td>
<td>63%</td>
</tr>
</tbody>
</table>

*Table 5: Missing Value Patterns and Percentages*

Dealing with Missing Data

There are major issues to consider when there is such an extensive amount of missing values. There are different methods of imputation to handle missing data. One method would be to use only the complete-case method which is the most conventional method. A limitation of this method is that it can exclude a large amount of the original sample (Little & Rubin, 2002), and this leads to issues with possible bias estimates and that the sample may be unrepresentative of the original sample. Other methods may include different types of imputation, including simple mean imputation or regression mean imputation, however, these methods are not recommended with when the proportion of missing to complete cases is so high and can introduce bias (Jones, 1996). A much more complex method using multiple
imputation is thought to address the limitations of these other imputation methods as well as the issues that might arise from complete-case methods. Although there is no inherent reason why multiple imputation is not possible to use with a small sample, the more missing values to impute the longer the analysis will run. Especially as the imputation becomes a much more complex analysis and there is more space for issues or bias and would require ad-hoc analysis as this may result in more mistakes. A multiple imputation analysis rerun can produce different estimates due to the nature of the analysis (Nakai & Weiming, 2011), therefore the comparison would be between an analysis of complete-case and the multiple imputation to explore if similar results are being identified in the larger imputed sample. This requires time which was beyond the scope of the PhD timeline, as the longitudinal model had to be built after the qualitative work was completed (Chapter 6). Complete-case method was used in this study to deal with missing data, after consideration of other methods to handling missing values, and after reviewing the sample numbers.
4.2.4.3. Participants

The participants whose data were included in this study reported in Chapter 7 were those recruited by CAPS from 2004 onwards (when IPQ-C introduced) and were aged between 11 and 13 years at the baseline time point. The following flow diagram (Figure 9) shows the final sample of 264 participants from CAPS that were included in this study.

![Flow diagram of participants recruited in Sub-Study](image)

*Figure 9: Flow diagram of participants recruited in Sub-Study*  
Participants that were included and the exclusion criteria to reach the final sample of participants included in the CAPS sub-study for the dynamic feedback study of this thesis.
4.2.4.4. Variables and Materials

Illness Perceptions – emotional representations

The IPQ-R assesses different domains separately and allows an analysis to include different domains. For this longitudinal model one of the domains was fitted into the model, the domain of emotional representations. As introduced in Chapter 2, the CS-SRM proposes a separate but interlinking pathway for emotional representations. This construct is theorised to inform about coping behaviours and derive specific illness behaviours. For this domain the questionnaire assess different emotional states of upset, anger depression, worry, nervous, and anxiety.

Pain Score

The hypothesis is that pain would influence the beliefs about the impact of the arthritis, the emotional representations and also the adolescent’s behaviour. Therefore, the model will explore the role of pain in these two factors. Pain intensity as measured by a visual analogue scale (VAS) was utilised in this model.

Child Health Questionnaire (CHQ) – outcome

The Child Health Questionnaire (CHQ, Landgraf et al., 1996) was developed as a generic quality-of-life of children and adolescents and is filled out by parents/guardians. This measure aimed to produce an assessment of quality of life as an outcome measure. The CHQ was adapted and validated for a British cohort involving a sample of patients with Juvenile Idiopathic Arthritis (Nugent et al., 2001). In the British cohort the CHQ was successful in discriminating between healthy controls and JIA patients (Nugent et al., 2001). The adapted British version of the CHQ-PF50 is the one that CAPS collects.

The CHQ-PF50 has fourteen concepts and two summary component scores measured with twelve multi-item scales and two single items. The items of the CHQ-PF50 were developed through qualitative techniques: comprehensive literature reviews, interviews, and focus groups with children and parents. The concepts and component scores are presented in Table 6, listing items and the descriptions of the each
concept. Internal reliability of the CHQ was found in a clinical sample of 71 children with Juvenile Rheumatoid Arthritis (JRA; Landgraf et al., 1996). The scoring of the CHQ-PF50 is standardized. There are algorithms for scoring the overall scales and this is to ensure the reliability and validity and comparisons across studies (Landgraf et al., 1996).

**Table 6: The Child Health Questionnaire (CHQ-PF50)**

<table>
<thead>
<tr>
<th>Concept scale/item</th>
<th>Number of items</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Global health (GGH)</td>
<td>1</td>
<td>Measure of a subjective assessment of overall health and illness</td>
</tr>
<tr>
<td>Physical functioning (PF)</td>
<td>6</td>
<td>Measure of self-care, mobility, limiting activities to assess the presence and extent of physical limitations.</td>
</tr>
<tr>
<td>Role/social limitations – emotional/behavioural (REB)</td>
<td>3</td>
<td>Measure of limitations in social role (school/friends) due to emotional and behavioural difficulties.</td>
</tr>
<tr>
<td>Role/Social Limitations – Physical (RP)</td>
<td>2</td>
<td>Measure of limitations in social role due to physical limitations.</td>
</tr>
<tr>
<td>Bodily Pain/Discomfort (BP)</td>
<td>2</td>
<td>Measure of degree (intensity) and frequency of general pain and discomfort.</td>
</tr>
<tr>
<td>Behaviour (BE)</td>
<td>5</td>
<td>Measure of frequency of behaviour problems and ability to get along with others.</td>
</tr>
<tr>
<td>Global Behaviour Item (GBE)</td>
<td>1</td>
<td>Measure of overall overt behaviour as a component of mental health.</td>
</tr>
<tr>
<td>Mental Health (MH)</td>
<td>5</td>
<td>Measure of the frequencies of both negative and positive states.</td>
</tr>
<tr>
<td>Self Esteem (SE)</td>
<td>6</td>
<td>Measure of self-esteem, satisfaction with different aspects and ability to get along with others.</td>
</tr>
<tr>
<td>General Health Perceptions (GH)</td>
<td>5</td>
<td>Measure of the parent’s perceptions of overall health including past, present and future health.</td>
</tr>
<tr>
<td>Change in Health (CH)</td>
<td>1</td>
<td>Measure of reported change in health status compared to one year ago.</td>
</tr>
<tr>
<td>Parental Impact – Emotional (PE)</td>
<td>3</td>
<td>Measure of the amount parental distress related to their child’s health, emotions and behaviours</td>
</tr>
<tr>
<td>Parental Impact – Time (PT)</td>
<td>3</td>
<td>Measures of parent’s personal time in relation to child’s health, emotions and behaviours.</td>
</tr>
<tr>
<td>Family Activities (FA)</td>
<td>6</td>
<td>Measure of the frequency of disruption due to the child’s health or behaviour in the usual family activities</td>
</tr>
<tr>
<td>Family Cohesion (FC)</td>
<td>1</td>
<td>Measure of the family’s ability to get along with each other.</td>
</tr>
</tbody>
</table>

**General Health Questionnaire (Covariate)**

The General Health Questionnaire (GHQ, Goldberg & Williams, 1988) was developed for a community population in order to detect psychiatric disorders. The self-administrated screening questionnaire does not measure traits but rather tests for patterns that increase an inability to engage in healthy behaviours. In CAPS, the parents of adolescents with JIA complete the GHQ about their own experiences and
health. The GHQ is not intended to measure attribution styles; the tool was designed
to measure a present state (Goldberg & Williams, 1988). The results of this
questionnaire were included in the model because the state of the parent, at the time
of the testing, might have an effect on how the parent perceives their child’s function
and behaviours.

The GHQ utilised in this study was the GHQ-30, which consists of 30 items. Each of
these items includes a four point Likert scales for the responses. There are two
methods of scoring these responses. The GHQ method is by assigning a 0 or a 1 to
the four points of the Likert scale. Thereby, the two lower scores are both recorded as
0 and the two upper scores are recorded as a 1. The other scoring method is the usual
method of scoring Likert scales. Each point on the scale is assigned a number from 1
to 4. An overall sum of the responses is calculated for either scoring method. The
GHQ-30 was completed at every time point by the parents. For this study, the Likert
scale scoring method was utilised to allow for sensitivity to change in scores.

4.2.4.5. Procedure

On recruitment into the CAPS study participants and their parent/guardian are
provided with questionnaires to fill out (baseline). The baseline date is the date first
seen by a paediatric rheumatology consultant. At that point the following data are
collected; family history, education, treatment/medication, and previous medical
history of the child. At this time, the patient and/or their parent/guardian complete a
series of questionnaires; which included the CHQ-PF50, CHAQ, and GHQ-30. The
follow-up periods are yearly after baseline, or the initial consultations, for 5 years
and then again at 7th and 10th year. The follow-up questionnaires are sent out to the
patient and a parent/guardian using the mailing database.

4.2.4.6. Analysis

Descriptive statistics were used to summarise variables with the median and inter-
quartile range (IQR) for non-normal continuous variables. Spearman’s correlations
were used to determine associations between physical activity, emotional
representations and pain at baseline, year 1, and 2. Cross-sectional linear regressions
were used to determine whether pain and emotional representations significantly predicted the physical activity (dependent variable) the same year. Backward stepwise regressions were used to determine whether physical activity (dependent variable at year 1) from baseline predictors; emotional representations, pain, parent’s emotional distress GHQ-30, time to first referral (disease duration), and gender. Longitudinal mediation models were used to determine the role of pain and emotional representations in predicting physical activity. A likelihood ratio test was used to compare the fit of the two models to provide a statistical rationale to reject or accept a model. The best fitted model was taken forward for mediation analysis, to determine whether the associations between emotional representations and physical activity were explained to some extent by pain. A Sobel-Goodman test was used to determine the extent of mediation in the total effect being mediated in the model.
4.3. Assessment of Representations – Adaptation and modification of the IPQ-R for use with adolescents with JIA (Aim 2)

The second aim was to identify the best method of assessing the illness representations of adolescents who have a long-term condition. Therefore, based on the results from the first aim and the studies described above, the second aim of thesis was to develop a new version of the IPQ-R to assess adolescents’ illness representations. The first objective was to develop an adolescent specific measure of illness representations. The second objective was to test and evaluate the psychometric properties of the new measure.

4.3.1. Research Paradigm

This second part aimed to develop an adaption of the assessment of illness representations based on the interpretation of the previous work which addressed the first aim of this thesis (Section 3.2). To develop an adapted questionnaire, two studies were conducted utilising mixed-methods over two phases. The research paradigm to address this second aim is a sequential exploratory design with a qualitative and quantitative sequence (Creswell, 2003). For the first phase of this research paradigm, the objective was to develop the questionnaire deriving items from the previous qualitative work of Section 4.2.2 and Section 4.2.3 and adapting the revised illness perceptions questionnaire (IPQ-R). The first phase of this research paradigm as shown in Figure 10, was to test the face validity of the new items in the adapted questionnaire. The data were collected qualitatively through an open-ended feedback form (pre-test phase). The next phase was a quantitative study using statistical methods with the objective to test the validity and reliability of the new questionnaire (field-testing phase).
The research paradigm utilised in the development and adaptation of a new measure is presented above in Figure 10. The questionnaire was sent out to adolescents in the Childhood Arthritis Prospective Study (CAPS). Description of the recruitment and the statistical tests utilised can be found under Section 4.3.2 and Section 4.3.3.
4.3.2. Assessment: Developing Questionnaire

An adapted questionnaire was developed from the results collected through the cognitive interviews. Before field-testing the modified questionnaire, there was a pre-test (pilot) study that aimed to identify any ambiguous or difficult phrasing or responses. The following sections in 4.3.2.1 describe this pre-test study.

4.3.2.1. Design

The purpose of this initial study was to identify and then address or pre-empt potential problems with the modifications before sending it out to the patient population. Therefore, the objective of this pre-test study was to establish face validity of the changes made to the questionnaire. To establish face validity and to test the linguistic validity, the questionnaire was sent to adolescents without JIA to answer the questionnaire and provide feedback on the language and length (found under Section 4.3.2.4).

4.3.2.2. Methodological Considerations – Content Validity

Validation is a process of assessing if the tool measures what it intends to measure. These processes involve different stages in providing evidence in the adequacy of the tool in measuring the intended theorised constructs. Validity can be further elaborated into three main categories of validation these are: content validity, construct validity, and criterion validity. For the pre-test study described in this current section (Section 4.3.2) content validity was tested. Construct validity was tested in the subsequent field-testing study and is defined in Section 4.3.3.2.

Content validity is the process of testing of the relevance of the items of the questionnaire and assessing the development of those items. Item coverage and relevance along with face validity are aspects that are closely related to the category of content validity. One key aspect of content validity is the comprehensiveness of the items and the process of content coverage and relevance should be the first to be tested when developing a questionnaire. Testing item coverage and relevance can entail different approaches that ensure all items are relevant to the theorised
construct. For the adaptation of the IPQ-R in this PhD, the item coverage and relevance were assessed in the content analysis described in Section 4.2.2.8. After the new and adapted items were generated, the new items in the adapted questionnaire were assessed by the research team before testing the face validity in the pre-testing study. Following construction of the items, face validity was tested using processes that ensured the validity and lack of ambiguity of the language used. Face validity was thus defined for this study by having validated the items at face value; the tool appeared to be valid through these rigorous developmental procedures and approvals.

4.3.2.3. Participants

Participants were recruited through an opportunity sample of adolescents aged between 11 and 16 years willing to take part in testing the questionnaire and answering further questions about the new items.

4.3.2.4. Materials

First version of new questionnaire

The questionnaire was developed through rigorous analysis of the results from the work described in Section 4.2 above. There were two stages of development before sending the new questionnaire to adolescents for assessment of the comprehension of the items. The first stage development of the new questionnaire was deriving the items and changes using the synthesis of the results of the studies examining the CS-SRM (from results in Chapter 5, 6, and 7). This was followed by a consensus validation exercise undertaken by members of the wider CAPS team. Further modification of items resulted from this exercise. After consensus was reached the questionnaire was distributed to adolescents for the final face validity.

Feedback questionnaire

A feedback questionnaire was developed to provide the space for adolescents to give specific and general feedback on the version of the IPQ-R they completed. Fayers and Machin (2007) recommend asking participants to review the instrument in
general as well as asking for specific information regarding items. The questions that were completed from the feedback questionnaire are as follows and the full questionnaire is found in Appendix 6.

a) How long did it take for you to finish the questionnaire?
b) Were the instructions clear?
c) Were there any words or terms you found difficult?
d) Were there any questions you did not understand?
e) Have you any other comments?

4.3.2.5. Procedure

The participants were provided a cover letter with instructions that asked them to think of a recent occurrence of pain when answering the questionnaire. Furthermore, they were asked to provide feedback on each of the four sections of the new questionnaire; asking specifically if the instructions were clear, if there were any words or terms that were difficult and if there were questions they did not understand. Lastly, there was an open-ended question for the adolescents to write any other comments. Feedback was collected and changes were made to clarify the items in the new questionnaire. For the complete pack of what was sent to the participants see Appendix 7.

4.3.2.6. Analysis

The purpose of the pre-test study was to examine the face validity of the adapted questionnaire and the new items. Language validity was established by testing if the items are comprehensive to adolescents aged between 11 and 16. This was completed qualitatively through synthesising the answers from the feedback questionnaire. If there were any issues derived from the synthesis of the answers of the completed feedback questionnaires, changes were made for the version to be sent out with the field-testing study described in Section 4.3.3.
4.3.3. Assessment: Validating Questionnaire

The second objective was to validate the adapted questionnaire as part of the work of developing an adaptation of an assessment tool of the IPQ-R. To validate a questionnaire, the psychometric properties were evaluated and tested. For the scope of this thesis, re-test reliability and construct validity were tested. These psychometric properties are described in Section 4.3.3.2 and 4.3.3.6.

4.3.3.1. Design

This was a survey-based questionnaire validation. This study used the postal survey recruitment system from CAPS to recruit a clinical population. This field-testing study was set up with two phases; the first phase was to complete the questionnaire alongside the other CAPS questionnaires and in the second phase to complete the new questionnaire again to assess the re-test reliability.

4.3.3.2. Methodological Consideration – Validation and Reliability

One key aspect of validation is assessing how well the items are measuring the construct as intended. Construct Validity is an assessment based on a hypothetical model of the relationships and concepts being measured by the tool (Fayers & Machin, 2007). There are three questions to address when testing the construct validity: the dimensionality, homogeneity and the overlap of the items and proposed constructs.

To test the dimensionality of the proposed constructs, multivariate analyses can be utilised. Analyses such as factor analysis, or latent variable analysis, can distinguish if all items in a proposed construct/domain load to a single latent variable. To test the homogeneity of those items, a Cronbach’s Alpha test (Cronbach, 1951) determined if the items are internally consistent. Lastly, correlating all the items to assess and test overlap in the latent variables.
While validation testing is to ensure that the tool is measuring the intended constructs and hypothesised models, \textit{reliability} testing is to ensure that the tool has consistency and reproducibility. One method of testing the reproducibility of a measure is to distribute the tool repeatedly, assuming that the patient is stable over the testing time period. A measure of the level of agreement would determine the reliability.

4.3.3.3. Participants

The participants that were recruited for this study were included if they met two conditions. The first condition is that potential recruits were already enrolled on CAPS, thereby meeting the inclusion criteria for CAPS. The second condition was that they are aged between 11 and 16 years at the time of recruitment into this study. There were 816 adolescents in CAPS who met these recruitment conditions.

4.3.3.4. Materials

\textit{Adapted questionnaire}

The questionnaire (see Appendix 9) developed in the pre-test study previously described in Section 4.3.2.4 was sent out to complete. A full description of the questionnaire is provided in the results Chapter 9.

\textit{Pain VAS}

A pain visual analogue scale (VAS) was added to the questionnaire \textit{(as described in Section 4.2.2.4)}.

4.3.3.5. Procedure

Potential participants for the field-test study were identified from the database of CAPS participants. Those who were eligible were sent an additional pack alongside their usual CAPS questionnaire packs. Within the additional pack was included a letter introducing the field-test study and the study team. As part of the recruitment process, a five-minute film was created and posted on YouTube.
and this film was sent out through the CAPS newsletter and Facebook to provide more information on the study and the questionnaire.

**Phase 1**

As shown in Figure 11 in Phase 1 of the field-testing study, the new questionnaire was sent with the questionnaire pack according to the CAPS protocol. A complete questionnaire pack was sent, that included: a cover letter, the consent/assent forms, and a copy of a questionnaire with pre-paid envelope in accordance to the CAPS protocol.

**Follow-up Phase**

If the participants consented to be contacted for the follow-up phase they were sent the questionnaire again to complete two weeks after the initial questionnaire. The data collected in the follow-up phase will assess the test-retest reliability.
Ethical Considerations

A substantial amendment to the CAPS study was submitted to the REC in August 2013. The amendment to add the adapted questionnaire with the questionnaires CAPS send every month was accepted September 2013. The questionnaire has been incorporated into the study mailings and the questionnaires were posted in October 2013. All the data were anonymised and entered into an Access database by the study coordinator. A secure database was established and the usual CAPS protocols were followed with storing the data separate from personal records. The databases are password protected.

The North West Research Ethics Committee (REC: 02/8/104) granted a substantial amendment to include the adapted questionnaire in the CAPS questionnaire packs that are sent monthly. This amendment specified that this new adapted questionnaire would only be included for the participants who are eligible.
4.3.3.6. Analysis

There were three analyses completed to test the construct validity with this data set as shown in the flowchart in Figure 12. The first analysis was calculating the Cronbach’s alpha (1951) statistics as well as assessing the homogeneity of the items within each domain assessed the internal consistency. The factor structure was tested using factor analysis to assess whether the items within each domain loads onto the theoretically derived factors.

The data collected by administrating the same questionnaire over a brief period of time (in this case 2 weeks) can be tested to evaluate the stability of the measure over time. The data from phase 1 and the data from phase 2 were compared using pairwise t-tests to determine, within subject, if there was a significant difference over time. The pain scores completed from the pain VAS were utilised to identify those participants who have not experienced any changes in their pain experiences and may influence how they completed the questionnaire in the follow-up phase.
Chapter 5: Illness Representations – findings and discussion

5.1. Overview

This chapter presents the results of the study that address objective 1 and 2, which both aimed to evaluate if the illness representation domains as identified by the CS-SRM are relevant to adolescents. Objective 1 was to assess the degree to which the domains identified by Leventhal et al (1984) in the CS-SRM are relevant to adolescents with JIA and objective 2 was to assess the suitability of the IPQ-R (Changed to IPQ-C in the study) for adolescents with JIA. To answer the questions raised in objectives 1 and 2, two separate analyses, content and framework, were conducted on the cognitive interview data (see Section 4.2.2.7 and 4.2.2.8). Section 5.2 presents demographic information about the participants who completed the cognitive interviewing with the IPQ-C questions. Results of the content analysis and the framework analysis of each domain assessed by the IPQ-C are reported within the individual sections (found in Section 5.3 to 5.8). For ease, and to report the items of the questionnaire that elicited the think-aloud data, the results of the content analysis which addresses objective 2 are presented first, and these are followed by the findings and discussions which address objective 1.

The coding manual for the content analysis was generated from three random transcripts and was presented in Chapter 4, Table 4. There were five codes: “no problem” (the participant had had no difficulties responding to this questionnaire item), “not relevant” (the item did not apply to them), “incongruent endorsement” (the endorsement did not match their ‘think aloud’ statements), “incongruent answer” (the answer did not match the item), and “confusion” (asked for repetition, took longer to answer or had to skip it). This classification was applied to each of the participant’s responses to each item of the IPQ-C in order to allow a comparative
analysis of the endorsements and the underlying reasoning. The results of this analysis are presented separately for each domain of illness representations.

Cognitive interviewing aimed to elicit the cognitive processes underlying a participant’s response to each of the items of the IPQ-C. Further analysis of the think-aloud data was then used to ascertain a more in-depth understanding of the underlying mental constructs of the adolescents. This framework analysis facilitated determining a range of beliefs held within each construct by the adolescents and if these beliefs map to the original proposed constructs.

5.2. Participants

Twenty-one adolescents were recruited. The content analysis was undertaken on 21 sets of data, and the framework analysis on 20 datasets because one participant did not complete the cognitive interviewing task due to time constraints at the clinic. Of the final sample used in this analysis, 70% were aged between 11 and 13, with the average age of this group being 12.4 years. The remaining 30% were aged between 14 and 16, with an average of 15.4. Descriptive demographics can be found in Table 7.

Due to the nature of the interviews and the setting, some adolescents took longer to complete the think-aloud stage of the interview, therefore not all adolescents completed the section of the IPQ-C that assessed the Identity domain. Participants who completed the Identity domain were the adolescents who agreed to continue with the interview despite going over the timeframe suggested. Only half of the participants who completed the cognitive interviews also completed the cognitive interviewing of the identity domain. These participants are highlighted in Table 7. A mean age for this group of participants is 13.4 years, with mean disease duration of 5.1, and a mean pain and CHAQ score of 0.59 and 26.6 respectively.
Table 7: Description demographics of the participants who took part in the qualitative study.

<table>
<thead>
<tr>
<th>Participant Alias</th>
<th>Age (Years and months)</th>
<th>JIA Category</th>
<th>Pain Score (0-100)</th>
<th>CHAQ Score (0-3)</th>
<th>Disease Duration (Years)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Faye†</td>
<td>11.2</td>
<td>2</td>
<td>0</td>
<td>0.13</td>
<td>8</td>
</tr>
<tr>
<td>Kevin†</td>
<td>11.2</td>
<td>4</td>
<td>68</td>
<td>0.38</td>
<td>3</td>
</tr>
<tr>
<td>Lucy</td>
<td>11.7</td>
<td>1</td>
<td>18</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Neil†</td>
<td>11.7</td>
<td>2</td>
<td>13</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>Bianca</td>
<td>11.8</td>
<td>2</td>
<td>10</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Gwen†</td>
<td>11.11</td>
<td>4</td>
<td>9</td>
<td>0.63</td>
<td>6</td>
</tr>
<tr>
<td>Vincent</td>
<td>12.0</td>
<td>4</td>
<td>83</td>
<td>1.3</td>
<td>7</td>
</tr>
<tr>
<td>Jodie†</td>
<td>12.1</td>
<td>2</td>
<td>26</td>
<td>0.63</td>
<td>2</td>
</tr>
<tr>
<td>Carrie</td>
<td>12.7</td>
<td>2</td>
<td>41</td>
<td>0.50</td>
<td>3</td>
</tr>
<tr>
<td>Wyatt†</td>
<td>13.3</td>
<td>1</td>
<td>7</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>Adam</td>
<td>13.4</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Daisy</td>
<td>13.8</td>
<td>5</td>
<td>6</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Rachel†</td>
<td>13.8</td>
<td>1</td>
<td>4</td>
<td>0.38</td>
<td>4</td>
</tr>
<tr>
<td>Sara</td>
<td>13.11</td>
<td>2</td>
<td>53</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Eleanor</td>
<td>14.3</td>
<td>2</td>
<td>48</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Zahra²</td>
<td>14.5</td>
<td>4</td>
<td>1</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Ian†</td>
<td>15.5</td>
<td>3</td>
<td>49</td>
<td>1.38</td>
<td>5</td>
</tr>
<tr>
<td>Taya†</td>
<td>15.6</td>
<td>3</td>
<td>11</td>
<td>0.13</td>
<td>8</td>
</tr>
<tr>
<td>Paige</td>
<td>15.6</td>
<td>2</td>
<td>6</td>
<td>1.75</td>
<td>11</td>
</tr>
<tr>
<td>Mia†</td>
<td>16.5</td>
<td>1</td>
<td>61</td>
<td>1.50</td>
<td>1</td>
</tr>
<tr>
<td>Harrison†</td>
<td>16.10</td>
<td>2</td>
<td>22</td>
<td>0</td>
<td>5</td>
</tr>
<tr>
<td>Mean</td>
<td>13.38</td>
<td>36.3</td>
<td>0.80</td>
<td>4.76</td>
<td></td>
</tr>
</tbody>
</table>

Note: † those participants who completed the Identity task. ‡ participant who only completed the pain interview. * the participant who only completed the cognitive interview of the IPQ-C. JIA category: 1. Systematic arthritis, 2. Oligoarthritis- persistent, 3. Oligoarthritis – extended, 4. Polyarthritis – RF negative, 5. Polyarthritis – RF positive, 6. Enthesitis related arthritis, 7. Psoriatic arthritis, 8. Undifferentiated arthritis

5.3. I

Illness Identity

The construct of illness identity assesses the knowledge about both the ‘label’ and the related symptoms that are attributed to the condition. The following provides an analysis of the IPQ-C items that assess identity (symptom attribution) and illness coherence (comprehensible understanding of the condition). The domain of identity is assessed by a list of 14 symptoms that a participant would attribute to arthritis. The
domain of illness coherence is assessed with five items asking about the adolescents’ beliefs regarding how well they understand arthritis.

5.3.1. Illness Identity – Content Analysis

The items assessed in the domain of Identity take a different form to the other domains in the questionnaire. There is a list of symptoms with two columns. If the participant assesses they experienced the symptom they then answer if they believed this symptom was because of their arthritis. The frequency of problems with these symptoms is shown in Table 8.

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total number of problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>Feeling Pain</td>
<td>9</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Sore mouth/throat</td>
<td>9</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Felt like vomiting</td>
<td>8</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Couldn’t breathe well</td>
<td>8</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Lost or put on weight</td>
<td>7</td>
<td>1</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Feeling tired</td>
<td>7</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>My joints felt stiff</td>
<td>8</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Sore eyes/not see well</td>
<td>8</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Feeling unwell</td>
<td>6</td>
<td>1</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Getting headaches</td>
<td>10</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Could not sleep well</td>
<td>9</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Upset tummy</td>
<td>7</td>
<td>1</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Felt dizzy</td>
<td>9</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Felt weak</td>
<td>9</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>TOTAL</td>
<td>114</td>
<td>14</td>
<td>9</td>
<td>2</td>
<td>1</td>
<td>26</td>
</tr>
</tbody>
</table>
The most frequent problem the participants had with these items was that the attribution was not directly due to the arthritis but rather a side effect of the treatment. The most common of these symptoms were *lost or put on weight, feeling unwell, upset tummy*. For these items, participants like Daisy, aged 13, wanted to circle both *yes* and *no* to the question whether these symptoms are related to her arthritis but were due to the treatment for her arthritis. The other problems were found with the item *my joints felt stiff*, as stiffness did not encompass other symptoms such as sore joints. For some adolescents the relevance of the symptoms differed, like Wyatt, aged 13, the two weeks duration period to report symptoms was not relevant to Wyatt, and for Taya, aged 15, the item *couldn’t breathe well* was not specific enough as it was *shortness of breath* that was related to her arthritis.

5.3.2. Illness Identity – Framework Analysis

The adolescents who completed the cognitive interviewing for the identity domain of the questionnaire circled similar symptoms. However, the attribution of the symptom was both attributed with the arthritis and the treatment related to their arthritis. The framework analysis found that individuals attributed symptoms according to what they believed caused that specific symptom. This is a higher order of appraising the symptoms, as it follows the primary appraisal of decoding and converting sensations into symptoms (Henderson et al., 2007). For these participants, they believed only some symptoms were caused by arthritis and the other symptoms were caused by aspects related to their arthritis (as shown in Figure 13).
5.3.2.1. Arthritis

The two symptoms that were consistently identified as associated with arthritis were *feeling pain* and *my joints felt stiff*. The underlying cause of these symptoms was attributed to arthritis and participants substituted these symptoms as indicators for arthritis. For example, Harrison, aged 16, only picked these symptoms as they were the only ones caused by his arthritis. Berry and colleagues (1993) found that children (6 to 17 years) would describe what juvenile arthritis is either by specifying that arthritis is a joint disease (i.e. “swollen, sore, stiff joints”), or else that it is pain without causal attribution (i.e. “when your leg hurts and makes you stop walking”). This association between pain and joints stiffness caused by arthritis seems to be common amongst children and adolescents with JIA.

5.3.2.2. Related to Arthritis

Other participants viewed pain as an indication of ‘doing too much’. For example, Gwen, aged 11, said that she feels pain “because I’ve done too much” and later she attributed *feeling tired* to arthritis because “when walking feet hurt because of arthritis.” It seemed for adolescents like Gwen, pain had a purpose. For Gwen, pain
was an indicator of the personal limits set by her condition. However, the cause of
the pain was not arthritis *per se*, but rather the behavioural limitations, which
occurred after doing too much.

There were other symptoms that participants attributed to aspects related to arthritis
such as medication or in one case, uveitis. Symptoms such as *lost or gain weight*,
*unwell*, and *felt like vomiting* were all perceived as caused by side effects from the
medication. The adolescents map the causal attributions to the symptoms. Ian, aged
15, said that *feeling unwell* was “*because of medication.*” Similarly, Wyatt, aged 13,
and Ian, attributed *gaining or losing weight* to medication.

5.3.2.3. Other cause

Similar to adults, adolescents tried to make sense of symptoms by trying to attribute
a cause. For some symptoms that were selected, the cause was not linked to arthritis
or anything related to arthritis. As Ian, aged 15, said about the symptom *sore
mouth/throat;* “*arthriti* *s wouldn’t cause a sore throat.*” Other chosen symptoms
were mapped to other illness as Tanya and Gwen, aged 15 and 11 respectively,
pointed out for the symptom *felt like vomiting* was linked to being “*generally ill*”. 
This is despite the fact that other adolescents attributed the symptom *felt like
vomiting* to the side effects of medication. The difference in attribution can be
mapped back to the adolescents’ experiences of those symptoms; which provide a
framework for what would make sense to them. Therefore, those adolescents who
were taking medication that would have a side effect of nausea would have attributed
the nausea to their medication. For those who had other illnesses for which nausea
was a symptom, nausea would then be attributed to the other illnesses, suggesting
that adults and adolescents carry out symptom attribution in a similar fashion to each
other.
5.3.3. Identity – Conclusions

The content analysis showed that the full range of symptom attributions was not captured as there are symptoms also attributed to medication and treatment. While the framework analysis showed how adolescents tried to make sense of coded sensations, by mapping the causes of the symptom, research argued that symptoms are a starting point to establishing if there is an illness (Pennebaker & Skelton, 1978). One method of establishing a condition is attributing symptom causes. The original work by Leventhal, Meyer and Nerenz (1980) theorised that symptomatic representations guides coping as well as both the regulation and the evaluation of the treatment. The relationship between symptomatic representations, coping or treatment, can exist because of how adolescents are making sense of abstract sensations into concrete representations.

5.3.4. Illness Coherence – Content Analysis

The frequency of problems for the items assessing illness coherence is found in Table 9. The highest accumulation of problems was for the item my symptoms are puzzling to me with the participants providing mismatched answers to the item by not answering in relation to symptoms but rather in relation to the arthritis. This is reflective of a problem with comprehension and perhaps improper definition of symptoms. The adolescents were able to define puzzling, (many like Bianca, aged 11 described puzzling as “it’s like I don’t understand how it works”), but were not providing an answer regarding symptoms but rather answered the item referring to their arthritis. Bianca answered the item with “I know what my arthritis is about and I can sort of explain it but can’t find the right words at times.” For the items I understand my arthritis clearly and I don’t understand my arthritis it would have been expected that the answers would be mirrored; however this was not always the case. This mismatch led to incongruent endorsements, as there were still aspects of arthritis about which the adolescents were unclear. This may also be a reflection of the use of a negative statement in the item I don’t understand my arthritis; as Ian,
aged 15 points out “they could make it a bit more clear, cause when you asked me it, I had to think if I say disagree, does that mean I didn’t know anything about it or did it mean I did know something about it.”

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total number of problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>My symptoms are puzzling to me</td>
<td>5</td>
<td>1</td>
<td>0</td>
<td>10</td>
<td>3</td>
<td>14</td>
</tr>
<tr>
<td>My arthritis is a mystery to me</td>
<td>14</td>
<td>2</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>I understand my arthritis clearly</td>
<td>12</td>
<td>0</td>
<td>5</td>
<td>1</td>
<td></td>
<td>6</td>
</tr>
<tr>
<td>My arthritis doesn’t make any sense to me</td>
<td>11</td>
<td>0</td>
<td>4</td>
<td>2</td>
<td>1</td>
<td>7</td>
</tr>
<tr>
<td>I don’t understand my arthritis</td>
<td>9</td>
<td>0</td>
<td>5</td>
<td>3</td>
<td>1</td>
<td>9</td>
</tr>
<tr>
<td>TOTAL</td>
<td>51</td>
<td>3</td>
<td>14</td>
<td>17</td>
<td>6</td>
<td>40</td>
</tr>
</tbody>
</table>

5.3.5. Illness Coherence – Framework Analysis

The responses indicated that the items assessing illness coherence were too vague for the adolescents. Framework analysis found patterns in the aspects that the adolescents reported not to understand, and these aspects were not assessed individually. The adolescents had a general understanding of arthritis; however the understanding was not coherent as there were specific gaps in their knowledge. None of the participants provided a coherent explanation of the internal processes involved in arthritis. The gaps, as shown in Figure 14, included the cause of both the arthritis and the related pain, how the treatment works, and if there is still more to know about their arthritis.
5.3.5.1. Uncertainty

Some of the participants reported that despite having information about their condition, there were still gaps to their knowledge. These adolescents spoke with uncertainty regarding the extent of their knowledge. As Harrison, aged 16, said, “I know a bit about it, I don’t know everything, I know its inflammation around the joint that’s it I guess.” These participants could not completely disagree or agree with items such as I understand my arthritis clearly because they felt there may be gaps in their knowledge. As Ian, aged 15, pointed out, “I’ve been told that it happens, I know what happens. I don’t know if anything else is progressing or if gets worse I don’t know what I could do to control.” The belief adolescents held that they had limited understanding of their condition; it may be an indication that these adolescents were not provided with a coherent explanation of their condition. Just as Adam, aged 13, specified that for him he felt that “a little bit is missing.”

5.3.5.2. Cause

For other participants, the gaps in knowledge were identified. The questions the participants had about their condition were how and why they had the condition.
Some of the participants claimed they understood arthritis but not the cause. There were different definitions of cause, including the cause of the arthritis, as Daisy, aged 13, said “I don’t know what brought it on, it just happened there’s not really a cause it just happened.”, or the cause of the pain or the flare-ups, as reported by as Eleanor, aged 14, who said “I know how I’ve got it because it runs in the family and but like when it come up I was like what does that mean like when I’ve got it and why I got it.” The unknown cause of the flare-ups also meant that the adolescents were unable to understand the connection between the arthritis and the related symptoms. As Eleanor later said “It’s like I know it’s there and I know I got it and I know how but like I don’t understand it clearly because I don’t know why it flares up and why it gives me pain even though its joints.”

5.3.5.3. Treatment

For some participants, the gap in their knowledge was about treatment. These adolescents wanted to know more about the treatment and how treatment worked. This may suggest that there are difficulties with communications between physicians and adolescents as Faye, aged 11, highlights, “I know what it is and what they are talking [about] sometimes but not all the time. Sometimes they can do stuff for my ankles when I know what they are talking about but if they do it for a different [location] like my neck sometimes if it’s new I don’t know what they are talking about.” Another issue regarding the treatment is side effects of a particular treatment as Lucy, aged 11, said, “I don’t understand like the effects of the medication but I know about my arthritis.” This again may be a reflection of a lack of communication regarding different treatment plans and the possible side effects or even how the treatment will work.

5.3.6. Illness Coherence – Conclusions

Research with adults suggests that a diagnosis and a label for a condition leads to the acceptance and integration of the condition into self-identity (Whitehead, 2006). However, the current study showed that even with a diagnosis there were still uncertainties in understanding the condition despite having a label for it. In this case,
the label of idiopathic itself infers that the cause is unknown. This identification and definition of JIA might be why the participants felt there were gaps in their knowledge, and that they were not provided with a coherent account of their condition. These gaps that the adolescents either identified or felt existed in their knowledge requires careful assessment, as this may identify what information or communication is required to consolidate their understanding of the condition. This closer assessment may be achieved through modifications of the items to include more specific questions about different aspects of the condition.

5.4. Cause

The domain assessing causal attributions had two sections. The first section followed a similar pattern to a Likert scale, representing an endorsement of how much the participant agrees with the item being a cause of their arthritis. The list of items included different possible causes that are thought to be possible causal attributions to a health threat. The second section of the cause domain required the participants to list the three causes they attributed to their condition from the items of the first section. The content analysis reported below is an analysis of the first section of the cause domain. The framework analysis included both sections of the cause domain in the analysis.

5.4.1. Cause – Content Analysis

For most of the adolescents, they had one key theory regarding what caused their arthritis. For some of the adolescents, they also had theories on the cause of the flare-ups they experienced as well as the cause of the arthritis. For example, Bianca, aged 11 said, “Well there are other people in the family like my granddad and my mum’s cousin and other people who have got arthritis. So I think it does run in the family and I have got it from relatives.” However, Bianca also believed that her own behaviour and the type of person that she is caused the flare-ups, “because I know that I got it from the family, but it’s when I do sport and dance, that’s my own behaviour and that is what I did and that’s when it hurts and when it [pain] is caused the most.” Later on in the interview, Bianca explained that “I’d probably do too
much to be able to control my arthritis and that affects it quite strongly, I don’t know, I stop but then I would carry on doing all the sports and activities straight afterwards after it calms down a bit I go straight into it but that could be a problem because it hurts afterwards.” Causal attributions such as doing too much were perceived as an episodic cause because it was inconsistent as Neil, aged 11, pointed out, “sometimes it might not be doing too much it might come, and it might not come.”

As shown in Table 10, the causal attributions of drinking alcohol and smoking were considered irrelevant to the adolescents as they understood the question as if they were drinking alcohol or smoking prior to the onset of the arthritis. For example, Paige aged 15, referred to age of onset, “because at 3 and a half I wouldn’t have drunk any alcohol” (Paige). For some adolescents, the causal attribution was not relevant to them because they currently do not smoke or drink alcohol just as Bianca, aged 11, said “I wouldn’t know” or Carrie, aged 12, said, “I don’t smoke.”

For many of the adolescents, the item chance or bad luck included two different concepts in one item. For some adolescents, chance and bad luck were similar. For example Lucy, aged 11, who said, “It’s something [I] was born with.” However, most adolescents like Neil, aged 11, said they would have answered differently if chance and bad luck were separate items. For instance, Neil would have put “agree” as “it’s not like it was certain I would have gotten it or would not have gotten it’s just like chance that I got it.” Neil would have put strongly disagree for bad luck as “’cause a chance is just something that might happen but bad luck is when everything is wrong.” The belief that bad luck and chance are different led to incongruent endorsements.
Table 10: Content Analysis with cause

Frequency of problems with the items related to cause

<table>
<thead>
<tr>
<th>Cause</th>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total number of problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stress or worry</td>
<td>3</td>
<td>7</td>
<td>0</td>
<td>4</td>
<td>5</td>
<td>16</td>
<td></td>
</tr>
<tr>
<td>It runs in the family</td>
<td>18</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>A germ or virus</td>
<td>12</td>
<td>2</td>
<td>2</td>
<td>3</td>
<td>1</td>
<td>8</td>
<td></td>
</tr>
<tr>
<td>Diet or eating habits</td>
<td>8</td>
<td>11</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>12</td>
<td></td>
</tr>
<tr>
<td>Chance or bad luck</td>
<td>7</td>
<td>2</td>
<td>8</td>
<td>3</td>
<td>0</td>
<td>13</td>
<td></td>
</tr>
<tr>
<td>Poor health and bad medical care in my past</td>
<td>13</td>
<td>4</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>7</td>
<td></td>
</tr>
<tr>
<td>Pollution (like fumes, dirty water, and like) or toxins in the environment</td>
<td>11</td>
<td>6</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>My own behaviour</td>
<td>11</td>
<td>6</td>
<td>1</td>
<td>0</td>
<td>2</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>My attitude (for example thinking negatively) about life</td>
<td>14</td>
<td>4</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>6</td>
<td></td>
</tr>
<tr>
<td>Family problems, family worries</td>
<td>11</td>
<td>8</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>Doing too much</td>
<td>12</td>
<td>1</td>
<td>2</td>
<td>5</td>
<td>0</td>
<td>8</td>
<td></td>
</tr>
<tr>
<td>Feeling down, lonely, nervous or empty</td>
<td>11</td>
<td>8</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>Getting older</td>
<td>7</td>
<td>10</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>13</td>
<td></td>
</tr>
<tr>
<td>Drinking alcohol</td>
<td>6</td>
<td>12</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>14</td>
<td></td>
</tr>
<tr>
<td>Smoking</td>
<td>7</td>
<td>12</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>13</td>
<td></td>
</tr>
<tr>
<td>Accident or injury</td>
<td>13</td>
<td>0</td>
<td>4</td>
<td>3</td>
<td>0</td>
<td>7</td>
<td></td>
</tr>
<tr>
<td>Type of person that I am</td>
<td>11</td>
<td>5</td>
<td>0</td>
<td>2</td>
<td>2</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>Immune system in my body</td>
<td>7</td>
<td>0</td>
<td>6</td>
<td>2</td>
<td>5</td>
<td>13</td>
<td></td>
</tr>
<tr>
<td><strong>TOTAL</strong></td>
<td><strong>182</strong></td>
<td><strong>98</strong></td>
<td><strong>25</strong></td>
<td><strong>32</strong></td>
<td><strong>22</strong></td>
<td><strong>177</strong></td>
<td></td>
</tr>
</tbody>
</table>
The items related to psychological processes created confusion in how those items can contribute to the cause of arthritis. For example, Ian aged 15, could not understand the relationship between psychological processes, such as my attitude (for example thinking negatively) about life and the item family problems, family worries. He said, “I don’t think thinking about thinking down would cause it. I don’t think if I think negatively would just cause it” followed by, “I don’t think there is something going on in the family that caused it to me. I am not sure how to explain it, if there was something going on in the family I don’t think worries about it would have caused arthritis.” The link between psychological attributions such as feeling down, lonely, nervous or empty and an onset of a physical disease was hard to grasp, as Wyatt, age 13, said, “arthritis is more of a physical illness so I don’t think my emotions had any effect on it at all.” However, to some adolescents, psychological attributions may be linked to triggering flare-ups and this led for some of these psychological attributions to be endorsed with a different answer.

5.4.2. Cause – Framework Analysis

The participants’ causal attributions fell into three thematic categories of processes that they believed were the underlying cause of their arthritis as shown in Figure 15. The participants created plausible causes based on their experiences. As highlighted by the content analysis, for some adolescents there was one overall theory of the cause of arthritis, and then another theory of the cause of the flare-ups associated with their arthritis. For the framework analysis, the ratings of the causes (the second section of the cause domain in the questionnaire) were taken into account. Furthermore, how these leading causal beliefs mapped onto the other causal attributions was also taken into account to assess the coherence of their causal attributions.

The adolescents’ understanding of the underlying cause of their arthritis was mapped back to the information that was available to them. This was either from what they know about how the body works or from their experiences. This finding supported
the cognitive psychology theories of how an individual reaches equilibrium through the processes of both assimilation of that experience or the accommodation of previous schemas (Piaget, 1969), and they keep information that can help build a mental representation (Leventhal et al., 1980). Evidence for these theories was highlighted by Ian, aged 15, who could not make sense of the connection between stress or worry to the onset of the arthritis, but there may also be a relationship between stress, worry, and the pain episodes.

“I don’t think there is a logical information that stress or worry had caused it I got [it] when I was younger and I was never stressed or worried when I was younger. I am not sure to be honest, it’s just it could be now, it could be involved. Stress or worry could be involved in triggering it so when I get stressed a lot it’s just, it could be painful.”

Figure 15: Framework analysis with Cause
Themes identified through the framework analysis for the construct of cause.
5.4.2.1. Internal Processes

The adolescents who reported that the mostly likely cause of their condition was internal processes, such as their immune system, a virus or germ, or hereditary were those adolescents with family members who were diagnosed with arthritis. These adolescents named the different relatives who also has arthritis and as Eleanor, aged 14, concluded, “so it carries on and I’ve got it.” Adolescents like Eleanor had a theory for the underlying cause and in addition believed that there was a separate cause for symptomatic episodes, that is, something else that “sets it off” for example, doing too much, growing older, or external factors such as damp or going from hot to cold. Experiences of being ill before onset led adolescents like Harrison, aged 16, to believe the cause was a combination of a germ or virus and immune system as he recalled, “I think I was quite sick a few weeks before I got it I think that might have trigged it…I think the way it [immune system] reacted to the symptom erm could have affected it triggered.”

Earlier qualitative work reported that children and adolescents do believe that internal processes such as heredity, infection and aging can account for how children get arthritis (Berry et al., 1993). However, the participants in the current study did not believe these processes were mutually exclusive, and they spoke of underlying illness triggers and of episodic triggers. Participants such as Eleanor, Lucy, and Bianca had beliefs about separate underlying causal belief and a belief about episodic cause of the painflare-ups of arthritis. Participants such as Mia, aged 16, also believed the cause was by internal processes; a combination of hereditary and immune system, with chance:

“because I think it’s by chance I got it ‘cause my sister could have got it cause its all the women in my Nan’s family that have it so my sister could have had it but I got it instead but then she could develop it in future like my mum and her sister has so I don’t know.”
5.4.2.2. External Processes

There were participants who emphasised an external trigger as an underlying cause of their arthritis. When discussing environmental triggers, the participants took into account the exposure to the environment. For example, if they were exposed to similar environments as their family, and they were the only ones in the family who had arthritis, they reasoned that it could not have been an external trigger such as diet or eating habits, exemplified by Bianca, aged 11, “it just doesn’t depend on what I eat if I get arthritis, like if I eat the same food as my brother and sister and my friends and they haven’t got arthritis.” However, adolescents such as Daisy, aged 13, pointed out both her and her mum had arthritis, so it was a combination of hereditary and environmental factors because they lived behind a train track “the pollution cause like where we live right behind a train track but it’s just a theory so I don’t know if it’s true or not.” Similar to Daisy, there were adolescents who believed that external processes triggered internal processes, and so it was a combination of both internal and external processes. For example, Wyatt, aged 13, stated, “sort of my environment and pollution because I chose that one over my immune system because it’s things like that would have to override my immune system to make a difference to my body.”

5.4.2.3. Unknown

There were adolescents who were unclear about the cause of the arthritis onset. There were adolescents like Faye, aged 11, who were not confident with committing to causal attributions due to feeling uncertain because “I don’t know what did it”. For a cause such as it runs in the family, Faye said, “my nan and my nanna have got it but I wouldn’t say it was. I don’t think it would, I disagree.” While, for the cause my behaviour: Faye said, “I really don’t think it was my behaviour, it might have been because I don’t know.” Faye said this about other causal attributions, including accident or injury and the type of person that I am. There were adolescents who were unsure about the cause of their condition. These adolescents were more likely to pick causal attributions such as bad luck as their main underlying cause, and they also had the longest disease duration. This meant that these adolescents were less than 5 years
old at the onset of their condition. As Paige, aged 15, recollected, “I didn’t know what arthritis was. I thought it was a piece of paper. I didn’t understand it. It’s just like an arm or a leg it’s me just part of me.” This relationship between age of onset and causal beliefs has not been investigated, however these findings suggest that there might be significant relationship, and this might be indicative of either the child’s memories of experiences prior to the onset of the arthritis or the information provided to the child about the cause of arthritis.

5.4.3. Cause – Conclusions

Causal beliefs and attributions were mapped onto their experiences, both before each episodic flare-up and their memories of onset. If the adolescents knew of a family history, they strongly believed that arthritis was hereditary. If they were asked for environmental triggers, they used their experiences of having similar environments as siblings. Those who had experienced an accident, injury or illness prior to the onset of arthritis were more likely to believe that event to be a trigger. This was not dependent on age, but rather the experience prior to onset. These findings account for previous work that found a relationship between referral route and causal beliefs (Cordingley et al., 2012). As highlighted by the content analysis, there were differences in flare-up triggers and onset triggers which are not supported or measured in the IPQ-C. Finally, the relationship between age of onset and causal beliefs should be investigated further.

5.5. Timeline

The timeline domain includes two constructs, and the IPQ-C measures two different concepts. Firstly, the IPQ-C measures beliefs about the chronicity, and secondly it measures beliefs about the cyclicality of the condition. Overall, there are 10 items with six items comprising an assessment of chronicity, and four items comprising an assessment of cyclicality. Both of these constructs were evaluated through content and framework analyses as detailed in the following sections.
5.5.1. Chronicity – Content Analysis

The frequency and type of problems classified with the chronicity items are provided in Table 11. Highest cumulative problem provided a different endorsement to what was reasoned out loud when the participants were asked about the item (incongruent endorsement category). The item with the highest frequency of incongruent endorsement was \[\text{my arthritis will be with me for the rest of my life.}\] This item’s endorsement should match the item “Will last forever” as logically, those who believe that the condition will last forever will also believe it will last a lifetime as forever is an arbitrary timeframe for a lifetime. However, this was not evident with all participants, and there may be some reasons behind this discrepancy. Firstly, the information that was provided to the adolescents regarding the duration of the arthritis may have been unclear, and this may have led to the adolescents being unable to have a concrete idea or belief regarding chronicity. Therefore, this may be a reflection of developmental issues where low experience of long-term conditions meant that they do not have any past experiences or information to form beliefs about duration. Otherwise, this might be a methodological issue; for example, similar items in questionnaires provided a method of testable reliability, but this can be counterintuitive, as the adolescents may have assumed that they were expected to provide different answers. As Lucy, aged 11, pointed out, “very similar, they are more about how long my arthritis will last. A bit harder to answer with different answers, because if I put agree agree agree agree it would be erm like I don’t know really.”
Table 11: Content Analysis with timeline chronicity
Frequency of problems with the items related to Timeline Chronicity

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>My arthritis will go away soon</td>
<td>10</td>
<td>0</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>9</td>
</tr>
<tr>
<td>Will be with me for the rest of my life</td>
<td>12</td>
<td>0</td>
<td>6</td>
<td>0</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>Will last a long time</td>
<td>15</td>
<td>0</td>
<td>2</td>
<td>1</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Will soon get better</td>
<td>9</td>
<td>0</td>
<td>4</td>
<td>4</td>
<td>1</td>
<td>9</td>
</tr>
<tr>
<td>Will improve in time</td>
<td>11</td>
<td>0</td>
<td>3</td>
<td>4</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>Will last forever</td>
<td>15</td>
<td>0</td>
<td>3</td>
<td>1</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>TOTAL</td>
<td>72</td>
<td>0</td>
<td>21</td>
<td>13</td>
<td>5</td>
<td>39</td>
</tr>
</tbody>
</table>

There were some participants like Mia who gave an incongruent answer to the items assessing acute timeline perceptions, such as *my arthritis: will go away soon, will soon get better* and *will improve in time.* For example, Mia, aged 16 years old, strongly believed that she would have an underlying condition that could get better or worse at any point: “*strongly disagree because it’s like forever, it will get better and it will get worse but it will always be there.*” However, she then endorsed the item “*will improve in time*” with “*neither agree nor disagree because it’s just random, sometimes it will be ok and other times it won’t.*” What was evident in Mia’s reasoning is that she used her experiences of an intermittent condition, as did other participants, to formulate her beliefs that her condition is long-term, but the symptoms such as pain were not constant and these beliefs were not captured by the current items.
5.5.2. Chronicity – Framework Analysis

There was a range of beliefs regarding the chronicity of arthritis, which fell into three thematic categories as shown in Figure 16. These categories were that the arthritis is improving, that the adolescent hopes to ‘grow out’ of the arthritis, or that the arthritis will last a lifetime. Previous pain experiences dictated which belief the adolescent held. These thematic categories and the relevant underlying pain experiences are described below under each respective heading.

5.5.2.1. Improvement

Some adolescents reported an agreement that their condition would go away soon and will not last a lifetime or forever. These conclusive beliefs were reached through the thought process that their pain was occurring less often and that they felt a general improvement. For example, Adam, aged 13 stated, “I strongly agree with that [will improve in time] cause it’s already proven it has and still doing it.” For some adolescents like Wyatt, also aged 13, this seemingly spontaneous improvement led to beliefs that the improvement may not be permanent:

Figure 16: Framework analysis with Timeline chronicity
Themes identified through the framework analysis for the construct of timeline chronicity.
“Agree it has improved in the last few years and I’ve not really done much to make that happen it’s just gone away and I don’t know what but it has [on why not picking strongly agree] because it does not necessarily continue just keeps on improving. It could come back. But I don’t think it will, sort of, not as badly. I can’t really say what is going to happen but if it does then there.”

For some adolescents, they were unable to agree or disagree regarding a short-term improvement because they felt uncertain of how long an improvement lasts, implying the episodic nature of the condition. Furthermore, for the adolescents, still experiencing pain supported this uncertainty. For example, Carrie, aged 12, described how she chose neither agree nor disagree “because I got a lot of pain and I don’t how to cope with it and I don’t know how long it will stay” and later on she admitted “I don’t know when it will stop or if it will stop or if it will just stay with me.” For adolescents like Carrie, the previous experiences of the pain and the arthritis did not provide enough evidence to support a concrete belief about the timeline of the condition.

5.5.2.2. Hope to grow out of it

It was evident that for the adolescents to make sense of the uncertainty and the lack of concrete information regarding how long JIA may last, they hoped that, as Harrison, aged 16, described, “maybe when I’m fully grown or something maybe it might go away.” This expression of hope was developed from two different sources; firstly, from what the doctors say, as Vincent portrayed “most doctors say that it goes when you start getting older.” Secondly, since the condition is a childhood disease, there is an expectation that growing out of childhood also means growing out of the condition. The hope and uncertainty of Lucy, aged 12, was developed from both sources, “agree, doctor said it might go away but it might not so I’m not going to strongly agree because I am not really sure.” Later on Lucy articulated that “I don’t think it would last forever it’s like a child disease.” This provided evidence that adolescents try to make sense of a health threat. When previous experiences
provide mixed ideas, the adolescents also utilised what doctors say and the condition’s label. Furthermore, for participants such as Eleanor, aged 14 years, who was uncertain about the chronicity also expressed hope that she will grow out of the condition because of what she expects when she is older: “I was picturing myself getting old, seeing myself getting better. Erm, I don’t know if it will go away but I am hoping it will be soon.”

5.5.2.3. Lifetime

The final thematic category identified in the data was that the condition would last a lifetime. The participants utilised their experiences and previous information that led them to believe that their condition will last their whole life. For example, Gwen, aged 11, believed her condition would be long-term, “because my mum has told me that nothing can truly be done, I’ve [been] taken off medication once. But I had to come back on because it’s not doing well off it. I got pain.” However, Gwen also believed she would be experiencing improvement in the symptoms associated with the condition, “because when you get older I will start doing other activities and I shouldn’t be in a lot of pain then because I’m getting older.” Therefore, from Gwen’s experience of being told by her mum nothing can truly be done for her condition, and her still experiencing pain once off medication, combined with her perception that with getting older comes change, Gwen believed her condition is long-term and with intermittent but decreasing pain episodes. This was evident in the other adolescents who believed their condition was chronic, and similar to Gwen, this belief was based on pain experiences such as the fact that the disease had already been chronic. This belief in chronicity was not distinguishable by age. As Ian, aged 15, reasoned, “I don’t think after having it for so long it would just disappear.”

5.5.3. Chronicity – Conclusions

The framework analysis provided evidence of a translation of experience to the duration beliefs. This translation consolidated the usability of the theory put forward
by the CS-SRM. As highlighted in the literature, chronicity provides an abstract timeframe (Leventhal et al., 2012), and the adolescents were struggling with being certain about the abstract timeframe. The adolescents dealt with uncertainty by utilising different experiences or information from the doctors or parents to formulate their answers regarding the chronicity of their condition.

What emerged from the data is that the construct of chronicity required an additional assessment of beliefs regarding the episodic nature of the reoccurring pain. The adolescents’ focus on the pain experiences was not a reflection of their age, but rather how they developed their beliefs regarding the duration. This may be how these adolescents dealt with abstract concepts such as chronicity. This might also be highlighting a problem with measuring chronicity beliefs about a long-term condition. Similar issues were found in a study by McCorry et al, (2012) where there was a discrepancy in the scoring of chronicity beliefs because participants considered diabetes was an acute (indicating that they believed diabetes is an acute condition) despite verbally acknowledging diabetes as a chronic condition. The content analysis highlighted specific methodological issues that have to be addressed. Specifically, those items included for validation reasons (similar items) and those that included arbitrary timelines has to be reviewed.

5.5.4. Cyclical – Content Analysis

The item most cited as being not relevant was *my arthritis comes and goes in cycles*, as presented in Table 12. The reason for this item lacking relevance to a majority of the adolescents is the concept of a cycle. Cycle implied the ability to identify an exact pattern the condition became either better or worse, at the same time or on the same day. As Carrie, aged 12, disclosed, “*I’ve never known it come in cycles, only if I push myself in the gym or if I just erm if I just it never comes and goes in cycles.*” Carrie was unable to think of a time when pain related to the arthritis came as a cycle. For some participants, to be able to answer this question they adjusted the meaning of the word ‘cycles’ to mean patterns. Other participants focused on the phrase ‘comes and goes’ as that was more relevant to their experiences and beliefs.
Furthermore, the items in this construct were not capturing the nature of the episodic flare-ups. For example the use of the term *everyday* led to some adolescents to disagree with the item because change was not an everyday occurrence as Neil, aged 11, pointed out “it doesn’t change everyday.”

### Table 12: Content Analysis with timeline cyclical

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>My arthritis changes everyday</td>
<td>11</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>1</td>
<td>7</td>
</tr>
<tr>
<td>My arthritis comes and goes in cycles</td>
<td>9</td>
<td>6</td>
<td>0</td>
<td>2</td>
<td>4</td>
<td>12</td>
</tr>
<tr>
<td>It is hard to tell what my arthritis will do next</td>
<td>17</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>My arthritis gets better and worse all the time</td>
<td>10</td>
<td>2</td>
<td>1</td>
<td>5</td>
<td>0</td>
<td>8</td>
</tr>
<tr>
<td>TOTAL</td>
<td>47</td>
<td>10</td>
<td>3</td>
<td>10</td>
<td>5</td>
<td>28</td>
</tr>
</tbody>
</table>

### 5.5.5. Cyclical – Framework Analysis

All of the participants described their condition as episodic, referring to ‘flare-ups.’ These flare-ups were times when they felt their condition worsen, and when they experienced symptoms such as pain. There were two thematic categories under the cyclical domain as shown in Figure 17. All the adolescents attempted to identify a pattern in their painful episodes. Specifically, they were trying to identify triggers of the pain related to their condition. This led to two thematic categories: the expected flare-up and the unpredictable flare-up.
5.5.5.1. Expecting Flare-up

For those participants who were looking for triggers, it was possible to map some flare-ups that followed certain events such as, after a day of shopping with friends, or after a class of physical education, or walking. However, this was not always consistent as Bianca recounted, “I know it doesn’t always come, like, every certain times like in P.E. It does come but doesn’t always come all of the times.” The association between P.E and a painful flare-up had been established for Bianca because she thought of times when her arthritis got worse and she thought of pain and the triggers: “I was trying to think of times it does do that [gets worse]. Walking home from school and P.E.” For other adolescents, they looked for signs or other symptoms that would be associated with pain for example Vincent, aged 12 said, “I could know a little bit by the swelling and all that. Like I can recognise if my knees will be sore ‘cause of the swelling.”
5.5.5.2. Unpredictable Flare-up

For most of the adolescents, the search for patterns and known triggers for pain was futile. Pain was unpredictable and described as random. Adolescents like Daisy, aged 13, described the pain as “at the moment it like all the swelling is going down and I’m not in pain but then in like two days it’s won’t so it stays the same. It’s just the same but then just randomly stuff happens but it’s usually like little stuff like the pain. ‘Cause like sometimes nothing is hurt and I don’t know why it happens or I don’t have any signs it just does it.” Although, Daisy also pointed out that even if the pain was not predictable and was random, the pain itself was the same. Therefore, she knew what to expect when pain occurred: “it’s just the same really but it’s like, sometimes stuff happens and then it goes. But it’s basically the same.” So what is key is not the changes in the pain, but rather the inability to identify triggers of the flare-ups.

5.5.6. Cyclical – Conclusions

It was evident that all of the adolescents were looking for patterns when they were answering the cyclical items. The adolescents referred to changes that ranged from pain-free to having episodic flare-ups. Their focus was on identifying signs or triggers that a pain episode would occur. For some, this was successful through association of other symptoms such as swelling, or else activities such as P.E class or walking. For other adolescents, the flare-ups were random and impossible to predict.

5.6. Consequences

The domain of consequences included items that assess different aspects of everyday life and that may have been impacted by the condition. The construct was assessed with six items that measured the impact on their life, the perceptions of others, financial costs, and the impact on their family.
5.6.1. Consequences – Content Analysis

As shown in Table 13, there was a high frequency of ‘confusion’ in the domain of consequences. Some of these items were abstract, and it was evident that the adolescents attempted to answer them in as literal a way as possible. For example, Eleanor, aged 14, said that, “Erm I am not too sure about that one. [Affects what other people think of me] Because I don’t really know whether it does because I don’t ask people about it so I don’t know whether it does or does not.” Another example of this literal response was with the item asking about the financial costs when the adolescents rationalised there must be a cost, but they were unable to answer this with certainty. The adolescents could not know the full amount, claiming that the cost was different for the family and the hospital. Many of the adolescents tried listing the possible costs as Lucy, aged 11, said, “I don’t know if it costs a bit of money because they have to buy the equipment and pay to get over here and they have to like pay for the medication but erm.”

Table 13: Content Analysis with consequences

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>My arthritis is serious</td>
<td>12</td>
<td>1</td>
<td>3</td>
<td>2</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>Has a large effect on my life</td>
<td>11</td>
<td>0</td>
<td>6</td>
<td>0</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>Does not have much effect on my life</td>
<td>17</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Affects what other people think of me</td>
<td>8</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>4</td>
<td>10</td>
</tr>
<tr>
<td>Costs a lot of money to my family or the hospitals</td>
<td>7</td>
<td>6</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>12</td>
</tr>
<tr>
<td>Makes difficulties for my family</td>
<td>15</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>TOTAL</td>
<td>70</td>
<td>10</td>
<td>13</td>
<td>8</td>
<td>8</td>
<td>39</td>
</tr>
</tbody>
</table>
A key problem occurred with endorsing these items is that adolescents were endorsing *neither disagree nor agree* when they were comparing pain-free and painful situations as they reasoned it does not always interfere. As Mia, aged 16, said, “*neither agree nor disagree because when I’m well it does not bother me and when I’m not [well], it does, so it is like a bit of both.*” The endorsement did not reflect that when Mia was in pain or bothered by her condition there was an effect on her life.

5.6.2. Consequences – Framework Analysis

When analysing the beliefs related to the consequences of JIA, a key finding was that pain was the reason for the highest impact on the adolescents’ lives. As Ian, aged 15, concluded, “*It’s more the pain that stops me from doing things than the actual arthritis if it wasn’t for the pain I could do a lot more than if there wasn’t any arthritis and pain.*” The impact of pain ranged across two thematic categories and it was hard to plan for and predict painful flare-ups resulting in interference in their lives (as shown in the thematic diagram Figure 18). The domain of consequences combined a lot of the adolescents’ beliefs regarding the cyclical and recurring nature of pain, their personal control, and the emotional impact.

![Figure 18: Framework analysis with consequences](image)

Themes identified through the framework analysis for the construct of Consequences.
5.6.2.1. Not affected when not in pain

A key impact the arthritis had on the adolescents’ lives was that the unpredictable pain occasionally made it difficult to plan activities. When they were not in pain, it was not affecting them to the same extent. For example, Adam, aged 13, said, “it’s not as much serious as it was when it was affecting me.” So when an adolescent was not in pain, the degree of impact was less as Eleanor, age 14, stated, “when it’s all settled as Dr [---] calls it. It doesn’t affect my life.” Therefore, there was a difference in the impact on the adolescents’ lives between being in pain and being pain-free. When the adolescents were pain-free, they were capable of doing everything and anything like their peers.

5.6.2.2. Interference when in pain

Many of the adolescents described the pain as interfering with their life. As found in the content analysis, the endorsement was not capturing the full extent of this interference. This was because pain was not a constant intrusion in their lives. As Zahra, aged 14, reasoned “because it doesn’t affect all my life. But it affects [me] when it flares up sometimes, and sometimes it doesn’t. I can’t go to school ‘cause of it.” The pain interfered with different aspects of an adolescent’s life including feeling different to their peers, especially when in pain. Wyatt, aged 13, implied that not experiencing pain as often meant it did not interfere with his life, and said, “it does not really limit me physically anymore it used to when I was in primary school but now it’s ok and it doesn’t make me any different I don’t think.” This fear of being different to peers or being embarrassed had been the reason underlying the results in a study where children (aged between 8 to 12 years) suppressed pain behaviours in front of their peers (Larochette et al., 2006).
5.6.3. Consequences – Conclusions

A key issue is that an item such as impa{ts my life} is vague, and there is a need to capture how it may impact differently on each area of their lives. However, there were variations in the adolescents’ beliefs from when they were not in pain to when they were in pain. A systematic review of qualitative studies of children with JIA identified similar themes and showed that there is an aversion to being different and a striving for normality (Tong et al., 2012). Pain interfered with their sense of normality, and that makes them feel different. It is at those times that the disease and related pain has a larger effect on their lives. These conclusions were not isolated to adolescents with JIA. This has been found in adolescents (aged between 10 and 17) with chronic pain which led to isolation and limitations to activities (Meldrum et al., 2008). Therefore, the results of the framework analysis of the consequences domain suggested that the adolescents with JIA experienced similar consequences to those adolescents with chronic pain.

5.7. Control/Curability

The control/curability domain overarched the personal control and treatment control constructs and these were included together in the IPQ-C. Overall, there were 11 items in this domain, with six items assessing the adolescents’ beliefs about their own personal control, and five items assessing the adolescents’ beliefs about treatment control. Previous work has shown that adolescents hold a range of personal control beliefs where they felt they had a limited amount of control, considered pain to be its own entity, and they may feel some control over their pain through taking their treatment (Ghio, 2011). However, these adolescents also believed that their treatment had limitations and ranged from being reliable to unreliable which led to them feeling uncertain about the future (Ghio, 2011). The following section presents the results of the content analysis of the items assessing personal and treatment control.
5.7.1. Personal Control – Content Analysis

The content analysis of the items shown in Table 14 assessing personal control items highlighted that adolescents had the highest issue with this construct in comparison to the other constructs measured by the IPQ-C. There were two reasons underlying why this construct led to the most reported problems. Firstly, there were methodological problems with the use of double negative items such as; *nothing I do will have any effect on my arthritis*. This item led to the highest reporting of incongruent endorsements. This confusion with incongruent endorsements was not unique to just adolescents. McCorry et al, (2012) found through content analysis of cognitive interviews that negative items were also confusing for adults with diabetes.

There is evidence to suggest that the constructs of the personal and treatment control may overlap. In the validation study of the IPQ-R, the principal component analysis loaded the items “*there is a lot which I can do to control my symptoms*” and “*the course of my illness depends on me*” on both personal and treatment control (Moss-Morris et al., 2002). When the adolescents endorsed these items as having personal control, they used their experiences of treatment helping control their arthritis, and the related pain to rationalise their endorsement. As mentioned above, adolescents answered items assessing their personal control by considering taking or seeking treatment for pain as a method of gaining control (Ghio, 2011). French and Weinman (2008) argued that one criticism of this construct was the inability to distinguish what a low score might actually mean, specifically, distinguishing if a low score reflected beliefs about low self-efficacy or beliefs that controlling a condition was not dependant on behaviour. For these adolescents, the latter seemed true since when attempting to control pain, the adolescents reported their personal control was limited by pain. As Bianca, aged 11, said “*I don’t know if I haven’t tried anything to make it better. So I don’t know if it would make it better or worse, but I know there is nothing I can do to control my arthritis because if it hurts it hurts and I can’t stop it from hurting.*”
5.7.2. Personal Control – Conclusions

Individuals develop their understanding by trying to develop a coherent representation that reaches equilibrium (Piaget, 1969; Leventhal et al., 1984). For adolescents with JIA, their presentation of pain dictates their sense of control over their arthritis. Therefore, one way of making sense of personal control is through treatment control. From the work reported in this thesis, new items are proposed to bridge that gap in the assessment of personal control. As treatment-taking was classified as a behaviour that the adolescents felt they controlled, this would need to be assessed and distinguished. Furthermore, there is a need for modifications to assessment in order to separate low self-efficacy from beliefs that behaviour has no impact on the condition (French & Weinman, 2008).

5.7.3. Treatment Control – Content Analysis

A majority of the adolescents claimed that the item “my treatment will cure my arthritis” was irrelevant because there was no available cure, as shown in Table 15.
For some adolescents like Zahra, aged 14, cure was only temporary: “sometimes it doesn’t, it does, I mean just for that time but sometimes it doesn’t. When it does flare up then the treatment does help but it still does [flare-up].” There was an observation that the pain can be resolved by treatment, however the cyclical nature of the disease lead the adolescents to believe that the underlying disease will always be an issue.

The adolescents believed that treatment efficiency was short-term. A criticism of the construct of treatment control is that the word “treatment” was associated with only medication and not physical activity or diet (French & Weinman, 2008). However, the adolescents referred to different types of treatment therefore, they understood the term “treatment”.

The other item that accumulated the most problems was my treatment will prevent the bad effects of arthritis on me. The rationale for the adolescents was that it was not possible to prevent pain; a symptom that is unpredictable. Therefore, it was unclear for them whether the treatment helped or can prevent a trigger of pain. Therefore, the term “prevent” may not be applicable in this instance. As Faye, aged 11 said, “Medicine wouldn’t stop it, but like I said it would help make sure it doesn’t get worse but I wouldn’t say it would prevent it.”

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>Treatment Control</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>My treatment will cure my arthritis</td>
<td>3</td>
<td>14</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>16</td>
</tr>
<tr>
<td>There is very little that can be done to make my arthritis better</td>
<td>16</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>My treatment will prevent the bad effects of arthritis on me</td>
<td>6</td>
<td>1</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>13</td>
</tr>
<tr>
<td>Can help control my arthritis</td>
<td>14</td>
<td>1</td>
<td>2</td>
<td>1</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Nothing can help my arthritis</td>
<td>18</td>
<td></td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>TOTAL</td>
<td>57</td>
<td>16</td>
<td>6</td>
<td>8</td>
<td>6</td>
<td>36</td>
</tr>
</tbody>
</table>
5.7.4. Treatment Control – Conclusions

The relevance of the term “cure” with a long-term condition has been highlighted with the study with adults with diabetes (McCorry et al., 2012). There is a need to ensure the items regarding treatment reflect the capabilities of the treatment. This includes removing cure and prevention when referring to pain related to JIA. Furthermore, to map on how the adolescents represented their condition (as episodic with flare-ups), the controllability of the treatment could be measured by how much it helps an adolescent to continue with activities. This will aid in highlighting those adolescents who believe their treatment has short-term benefits as well as long-term (pain gets better over time).

5.8. Emotional Representations

The construct of emotional representations was assessed with six different negative emotions. The items of the IPQ-C assessed emotional representation towards the long-term condition. There were different reasons underlying the emotional representations that were related to arthritis such as the unpredictable nature and high impact of pain, and not necessarily the condition itself (Ghio, 2011). The following section provides the results of the content analysis of the six items assessing the emotional representations.

5.8.1. Emotional Representations – Content Analysis

In comparison to the other constructs, the construct of emotional representations has the lowest frequency of problems as shown in Table 16. There was a pattern in the underlying reasoning for how the adolescents addressed the emotional representations items. None of the participants had difficulties understanding or answering the items about their emotional states. However, the difficulty was that neither the items nor the responses given reflected what the adolescents defined as their emotional representations. Many of the adolescents used the endorsements to explain how often they felt that emotion. For example, they justified choosing
“disagree” over “strongly disagree” if the emotion in question was not felt as often. As Bianca, aged 11, pointed out, “I just thought well I know that I can’t well I don’t think about it as always a bad thing because I can move on from it and doesn’t always make me upset or worry me.”

Table 16: Content Analysis with emotional representations

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>No problem</th>
<th>Not relevant</th>
<th>Incongruent endorsement</th>
<th>Incongruent answer</th>
<th>Confusion</th>
<th>Total problems</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel down and sad thinking about my arthritis</td>
<td>15</td>
<td>2</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>My arthritis makes me angry</td>
<td>15</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>My arthritis makes me upset</td>
<td>12</td>
<td>1</td>
<td>3</td>
<td>2</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>My arthritis makes me feel afraid</td>
<td>16</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>My arthritis does not worry me</td>
<td>12</td>
<td>0</td>
<td>2</td>
<td>4</td>
<td>1</td>
<td>7</td>
</tr>
<tr>
<td>My arthritis makes me nervous and anxious</td>
<td>14</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>TOTAL</td>
<td>84</td>
<td>3</td>
<td>9</td>
<td>11</td>
<td>2</td>
<td>25</td>
</tr>
</tbody>
</table>

5.8.2. Emotional Representations – Conclusions

The items and responses available in the current measure did not adequately capture the emotional representations as described by the adolescents. Emotional representation of a condition was defined as emotional responses elicited and associated by the health threat (Leventhal et al., 2012). As they are measured by the IPQ-C, the phrasing of the items indicated that the authors might have assumed that the emotional response towards the condition is continuous and static. However, for the adolescents, emotional representations were triggered by the pain experiences and were context dependent. This may be a reflection of the relapsing and remitting nature of the condition. Changing the questions’ stems to how often and how much of an emotion the adolescents were experiencing may allow for greater precision in determining the emotional representations.
5.9. Summary

The two different analyses above found that certain aspects should be addressed by means of a modified questionnaire. The most critical change is the need to move from the assessment of illness beliefs to more specific foci: pain beliefs. Pain experiences and the beliefs related to those experiences were driving the underlying reasoning of the participants’ endorsements. Therefore, by assessing beliefs specific to pain, the accuracy of how adolescents are conceptualising their JIA can be improved in relation to subsequent behaviours. There were also methodological issues to be addressed such as the removal of negative questions and multiple questions regarding the same topic. There were also particular terms that, according to the adolescents, were not relevant to the condition such as “cure” or in the case of how often change occurs “everyday.”

A conceptual issue was evident due to the remitting nature of the condition. This meant that there was a need for additional items in different domains to map the experiences of adolescents with JIA. For the domain of timeline, there was also a requirement to assess the episodic nature of the pain and the duration of that episode, as well as an overall assessment of chronicity. For the domain of consequences, it arose that their pain behaviours changed in different areas of their lives, for example school in comparison to home. Therefore, there is a need to add more items to this domain to assess accurately the impact of pain. In the domain of control/curability, there is a need to assess the overlap of the constructs of personal and treatment control since the adolescents felt taking treatment helped them feel a sense of control. For the domain of emotional representations, the extent of emotions was not being captured. It was not possible to distinguish between those who ruminate about their condition and those who are integrating pain in their lives. The extent of emotional burden needs to be assessed in a more fluid manner as it is currently being treated as a static state. Currently, emotional representations scores were not able to distinguish between those who elicit an emotional response more often than others. Therefore, there is a proposition to change the responses to the items in the emotional representations domain. Overall, the results of the content analysis highlighted the
questionnaire’s methodological issues, and the framework analysis highlighted the need to assess pain beliefs.
Chapter 6: Coping Procedures and Illness behaviours: Findings and Discussion

6.1. Overview

In the previous chapter the first component of the Common Sense Self-Regulatory Model (CS-SRM; Leventhal et al., 1980) was evaluated. That chapter addressed the first two objectives of this thesis, first to identify the relevant domains of emotional and mental representations and secondly, the suitability of the current assessment of these domains. This current chapter investigates the second component of the CS-SRM, which outlines the coping procedures and illness behaviours, influenced by the individuals’ emotional and mental representations. Therefore, the current chapter addresses the thesis’ third and fourth objectives, to identify the goals in the adolescents’ coping strategies and to identify the profiles that are driving and influencing these coping goals.

As mentioned in Chapter 4, to evaluate the coping procedures of adolescents with JIA, the methodological approach of framework analysis, was applied to data collected in the ‘In My Shoes’ (IMS) interviews about the adolescent’s pain experiences. The IMS interviews were completed with 19 participants. These participants’ coping behaviours have been previously reported (Ghio, 2011). For this current work, these coping behaviours were analysed to create a coping framework. This coping framework was developed by identifying core psychological concepts that drive the adolescents’ illness behaviours (the previously identified coping behaviours). The analysed data are reported and organised according to the coping framework and reported alongside these identified core psychological concepts. As is common practice in qualitative work, some discussion is included amidst the
findings of the analysis. So the findings of Chapter 6 are reported within the context of the literature and theories.

The coping framework, (Presented in Figure 19 below) has three hierarchal stages, with one overall major-theme and two sub-themes labelled as maintenance category and attending category, and lastly coping strategies specific to these sub-themes. The illness representations presented in Chapter 5 were organised within these categories (sub-themes) to identify if there was a mental representation (patterns of cognitions and emotions) that were particular to these coping categories and driving these behaviours. These results are presented in Section 6.3, as cognitive and emotional profiles. Unique patterns in the illness representations data were found across the two coping categories, the maintenance and attending category.

6.2. Coping with Pain – Creating the Coping Framework

As described in Chapter 4, the Methods Section 4.2.3.6, the data were indexed and charted (according to framework analysis methodology) to identify the function driving the nine coping behaviours (previously identified in Ghio, 2011). These coping behaviours were synthesised under four categories of coping, these are summarised in Table 17.

<table>
<thead>
<tr>
<th>Coping Category</th>
<th>Coping Behaviours</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mental Process</td>
<td>- Get on with it</td>
</tr>
<tr>
<td>Physical Activity</td>
<td>- Rest and relax</td>
</tr>
<tr>
<td></td>
<td>- Staying still</td>
</tr>
<tr>
<td></td>
<td>- Exercising and movement</td>
</tr>
<tr>
<td>Seeking Support</td>
<td>- Not telling at all</td>
</tr>
<tr>
<td></td>
<td>- Telling mum but not school or friends</td>
</tr>
<tr>
<td></td>
<td>- Telling anyone</td>
</tr>
<tr>
<td>Seeking Medical Care</td>
<td>- Hospital or doctors</td>
</tr>
<tr>
<td></td>
<td>- Taking medication</td>
</tr>
</tbody>
</table>
The indexed coping behaviours from Table 17 lead to the development of the coping framework. The coping framework was developed through the theoretical hypothesis that actions and action plans are organised through personal motives or reasons. The answer to the question “Why?” would provide higher-level goals which can be highly abstract ideals (Kruglanski, 1996). For the participants in the current study, this is the desire for preserving a social identity of a normal adolescent. This has been labelled as the overarching major-theme. The answer to the question “How?” would provide lower-level goals which can be more concrete in the methodology (Kruglanski, 1996). For these participants in the current study, there were two types of coping sub-themes, or sub-goals. Therefore, as shown in the Figure 19, the structure of the coping framework consisted of one major-theme of desiring to preserve a social identity of a normal life and two sub-themes of immediate coping sub-goals.

The Sections (6.2.1 – 6.2.3) address different coping goal themes. The coping goals are presented alongside the core psychological theories of developing a sense of self, self-concept and social identity. Section 6.2.1 examines in detail the overarching goal of the adolescents attempting to preserve their social identity. Following this, Section 6.2.2 and Section 6.2.3 review the adolescents’ immediate coping sub-goals. For
these immediate sub-goals, two categories were identified: focus on maintaining normality and focus on attending to the pain. These sections (6.2.2 – 6.2.3) also present other data collected alongside these interviews. These other data address differences in pain intensity, function, disease duration and age between the two categories of adolescents. Profiles were synthesised to discern the key characteristics underpinning the coping strategies employed by the two categories, found in Section 6.3. The cognitive and emotional profiles created a unique pain representation that underlay the coping strategies and coping goals of the adolescents as presented in Figure 19.

6.2.1. Overarching Goal

The hypothesised goal to developing these coping behaviours was to create a social identity of a normal adolescent. Social identification is the association of people with social categories that are used to define people (Tajfel, 1974; Tajfel & Turner, 1979; Turner, 1982). According to social identity theory (Tajfel & Turner, 1979), the process of developing an in-group identity begins with social categorisation, followed by social identification and social comparison. The participants in the current study viewed themselves as adolescent in terms of social categorisation. The participants aimed to adopt the identity of the adolescent (social identification). The participants defined this ‘adolescent’ social category by comparing their own current behaviours with what is expected of adolescents their age (social comparison). Expectations were developed from using social comparison and social adolescence standards. Social comparison allowed for the establishment of a normative standard for the adolescent social category. The participants engaged in a drive-upward approach (Festinger, 1954) which meant they were comparing themselves with other adolescents within their age group who were leading perceived normal lives. This is demonstrated by Eleanor (age 14): “Erm, it stopped me from doing things that I would have liked. Like my cousins were going into the pool and I was like I am too sore to and I couldn’t do what I would have liked to do.” Elsewhere Eleanor also said “I am hoping that I can go be normal and be arthritis free.”
As was evident with the participants of this study, social comparison facilitated self-evaluation and drove goal formations (Mussweiler, 2001). Self-evaluation indicated that the adolescents’ coping strategies were linked to their self-concept. Social identity is one aspect of their development of a self-concept. Self-concept is hypothesized to be a multifaceted system that aims to be coherent and which is developed by the interaction of two cognitive structures of a social identity and a personal identity (Gregen, 1971; Turner, 1982). For the participants in the current study, they aimed to preserve their social identification in their peer group. Previous literature found that adults with chronic pain report changes to their self-identification and social identity (Richardson et al., 2006; Osborn & Smith, 2006). These changes also have implications for social interactions. Adults report that their chronic pain is disruptive to their everyday life and damaging to their sense of self-worth (Osborn & Smith, 2006). This disruption to everyday life and social interactions is a barrier to developing social-identity but also developing self-concept for adolescents with recurring pain.

The participants coped directly with pain but also indirectly with the implications to their social life, peers and school. The participants engaged in two types of coping strategies to achieve the goal of preserving their social identity of a normal adolescent. The coping strategy they engaged in depended on which immediate sub-goal they were attempting to pursue. The following two sections report these two different sub-goals and the coping strategies the participants engaged in to pursue achieving the sub-goals and the overall coping goal. Furthermore, the following sections will report that these coping strategies were dependent on the participants’ pain representations.

6.2.2. Sub-Goals: Maintaining a Sense of Normality

To achieve the goal of preserving a social identity, a group of the participants engaged in adjustments to attain this goal. They used accommodative coping strategies as defined by the dual-process framework (Brandtstädter & Rothermund, 2002) to reduce the impact pain had on their lives. Their focus was on maintaining
their sense of normality. Therefore, their behaviours had a functional purpose to reduce being treated or perceived differently from their peers. This behaviour stemmed from this group of participants’ perception of pain.

6.2.2.1. Pain Representation: Challenge

The pain representation for this group was that pain was a challenge to their social image. Therefore, pain is a challenge to overcome in order to be treated or perceived similar to their peers. To account for this challenge this group of adolescents reappraised the methods used to attain their overarching goal. This pain representation is inferred from the emotional profiles of this group of participants (Section 6.3.3). These adolescents acknowledged pain caused barriers to their overarching goal of leading normal lives in their activities, school and social interactions. These participants illustrated that they accepted pain as part of their norm and adjusted their goals to include pain. McCraken (2010) defined acceptance of pain as a pathway between behaviours and experiences. The adolescents who aimed to maintain their normality described how they adjusted their behaviours to achieve their goals. This adjustment originated from accepting pain as a normal experience and resulted in an overall adjustment to their definition of normal.

6.2.2.2. Coping Strategies: Accommodative and Goal Adjustment

Accommodative coping strategies stem from adjusting and reappraising an unattainable (blocked) goal (Brandtstädter & Rothermund, 2002). To achieve goal adjustment an individual revises the normative standards. For this group of participants, pain was seen as a challenge to how they perceived themselves and their social identity as well as a challenge to their overarching goal. Adaptive flexibility is the ability to disengage from blocked goals. For these participants to disengage from their overarching blocked goal of being perceived the same as their peers, they engaged with functional behaviours with the strategy to normalise the pain. Table 18 below, shows the coping behaviours previously identified and the strategy and function underlying those behaviours. The behaviours had a specific function and strategy driven by the pain representation challenge. These behaviours aimed to
achieve the lower-level goal maintain normality and the higher-level goal preserve social identity of a normal adolescent. These coping strategies are listed in Table 18 and the function is defined underneath.

Under the coping category of mental processes, there was only one coping strategy of get on with it and for some adolescents this was the best strategy, to carry on as normal as possible. These adolescents re-evaluated their ‘self’ and their everyday ‘normal’. They experienced changes to their self-identity due to the disruptions pain had on the activities they could engage in and their social interactions.

Under the category of physical activity there were three coping strategies; exercise, slowing down an activity, or resting and relaxing. There was a range of activities in which these adolescents engaged. If the adolescents who perceived their pain as a challenge were in pain they would either try to slow down or attempt their usual activities or else they would change the activity for less impact. If the pain was overbearing the adolescents mentioned techniques to bring the pain down to a level that was manageable again (rest and relax). By using these strategies of physical activity these participants demonstrated their adaptive flexibility in still engaging in physical activity but adjusting how they can engage. Although the adolescents in this group acknowledged that pain placed limitations on their physical activities, the continuation of some form of activities was an opportunity to develop their self-identity with prosocial behaviours. Longitudinal studies following the 1958 British Cohort and the 1970 British Cohort from age 16 to 33 years found that those individuals engaging in physical activities were more likely to exhibit long-term psychological wellbeing (Sacker & Cable, 2006).

Within the coping category of seeking support, this group of adolescents used a variety of coping strategies that ranged from ‘not telling anyone’ to ‘telling anyone who was around.’ Each of these strategies had an underlying function. Those adolescents who did not tell anyone about their pain explained that doing so would bring attention to their differences from their peers and the expectation that they themselves would be treated differently. This would endanger their efforts to be
perceived as one of their peers and undermine their goal of maintaining normality. For other adolescents, ‘telling anyone’ held the purpose of providing justifications for certain behaviours that may be perceived as out of the norm. The example provided in Table 18 was Carrie (age 14) who described that by telling her peers that she is in pain avoided any misunderstanding of her behaviours, which could have been construed as not socially accepted behaviour.

In the group of adolescents in the maintenance category, there were no adolescents that reported going to hospital as a way of managing their pain. However, they did report taking medication such as painkillers. Medication was not viewed as a coping strategy on its own but rather as part of regime of managing the pain in order to carry on with activities. This coping strategy of seeking medical care was only a technique to bring down the pain to a level that keeps them maintaining their normal standards of activities.
Table 18: Underlying function and intent of coping when perceiving pain as a challenge
Coping categories and coping intent with examples from the adolescents who perceived pain as a challenge.

<table>
<thead>
<tr>
<th>Coping Category</th>
<th>Mental Process</th>
<th>Physical Activity</th>
<th>Seeking Support</th>
<th>Seek medical care</th>
<th>Function</th>
</tr>
</thead>
<tbody>
<tr>
<td>Get on with it</td>
<td>Slow down the activities</td>
<td>Rest and relax</td>
<td>Exercise</td>
<td>Do not tell anyone</td>
<td>Tell mum but not school</td>
</tr>
<tr>
<td>Physical Activity</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>This was a mental process coping strategy to maintain the normalisation of the pain. Using this strategy, the normal standards have been adapted.</td>
<td>Still attempting to do activities however slower or else less of. They recognise that pain can be limiting their activities. They are comparing themselves to their usual self.</td>
<td>When the pain was too overwhelming to manage this strategy allowed to ease the pain back to a manageable level.</td>
<td>Meant making some changes (such as swapping football to swimming) it meant that they could still exercise just in a low-impact sport.</td>
<td>Did not tell because of the chance of being viewed or treated differently from their peers. This would interfere with their maintaining an image and also a sense of normality in their lives.</td>
<td>Not tell people at school because this would interfere with carrying on as a normal student like their peers. Mums understood the pain and kept tabs.</td>
</tr>
</tbody>
</table>

| Example | "I just carried on as normal." Kevin, age: 11 | "I just tried not to move it, tried to keep it still. I did not do much because I was in pain. I couldn’t do many things because I was in pain.” Gwen, age: 12 | "Get on with it, try not to think about it and if it’s really sore put something on it that helps like cool it down or stop it from feeling as much pain." Daisy, age: 14 | "The worst is when I try to do sports, but good thing is swimming that’s the thing they told us to do is swimming because you are weightless.” Adam, age: 13 | "Well I didn’t want to be treated like sort of erm I don’t know make a big deal about it ‘cause they would probably say do you want someone to write for you and I didn’t really want anyone to write for me.” Daisy, age: 14 | "I don’t if I get pain at school I won’t tell anyone because I just get on with it but I don’t usually get pain often at school because I am either sat down in lessons or at break and break doesn’t last long so I don’t walk round long to get pain.” Ian, age 15 | "Talking about it makes other people understand, so I do tell a lot of people when I am in pain so that they know that I am in pain, so that they don’t think I am ignoring them or not talking to them as much.” Carrie, age: 12 | "Take a tablet and then it normally dies down.” Harrison, age: 16 |
6.2.3. Sub-Goals: Attending to the pain

To achieve the goal of ‘preserving a social identity’, a group of the participants continued to pursue their goal and engaged in assimilative coping strategies as defined by the dual-process framework (Brandstädter & Rothermund, 2002). They utilised their coping strategies to attend to their pain and focused on managing the pain. This group were committed to being perceived as a normal adolescent. To achieve this higher-level overarching goal, they engaged in compensatory means by coping with the pain episodically and waiting for the episode to pass. Some of these adolescents went to extremes to uphold a normal social identity. These strategies also included planning for pain episodes and including times to rest and stop activities to manage the impact pain would have on their lives. These strategies and behaviours were informed from this group’s pain representation.

6.2.3.1. Pain Representation: Threat

The participants in this group represented their pain as a threat to their social identity as a normal adolescent and being treated and perceived as similar to their peers. This pain representation was inferred from the emotional profiles of this group of participants (Section 6.3.3). To account for this pain representation these participants considered themselves in two states, first an adolescent with pain waiting for it to pass and be managed and second an adolescent without pain that can be normal.

6.2.3.2. Coping Strategies: Assimilative and goal pursuit

For those adolescents who perceived their pain as a threat to their social identity (as a normal adolescent), assimilative coping strategies were used. Assimilative coping strategies are defined as focusing on changing a situation so that the situation becomes compatible with a desired goal (Brandstädter & Lerner, 1999). These adolescents pursued goals that were to achieve and maintain normative standards defined by their
peers’ social identity. Assimilative coping strategies are employed when an individual is committed to achieving a goal that is perceived as a normative standard or ideal (Brandtstädter & Rothermund, 2002). Table 19 presents how the coping strategies were identified in the coping behaviours. The pain representation ‘threat’ to an adolescent’s social identity determined the choice of the adolescent’s specific coping behaviours. These behaviours had an underlying function and a strategy to manage pain with the overall aim and goal of preserving a social identity of a normal adolescent.

Under the coping category of mental processes these adolescents reported using the strategy of ‘ignoring the existence of pain’. They expressed that there were two instances when they have tried to ignore pain. The first instance was when pain flared up in a situation that would expose them as being vulnerable such as in class at school or with friends. On those occasions, they would be exposed as different or limited in comparison to their peers. The second instance that adolescents ‘ignored the existence of pain’ was when they were experiencing no flare-ups thereby feeling pain-free. During these pain-free occasions the adolescents expressed a desire not to think about pain so that they can feel normal.

Under the coping category of physical activity there were only two coping strategies reported. None of the adolescents reported utilising exercise as a coping strategy. Rather, these adolescents described ‘stopping activities’ when pain flares-up. These participants shared that limitations are accentuated when they are comparing themselves to their peers. The pain acted as a barrier to their engagement with physical activity. Table 19 below shows how participants like Bianca (age 11), can feel unequal to their peers as a result of pain. The other coping strategy these adolescents utilised was ‘resting and relaxing’ as part of the process of waiting for the pain episode to pass. Engagement in sport or physical education contributes to opportunities to the development of self-identity (Coatsworth et al., 2005). For this group of adolescents, resting and relaxing was a method of waiting, which can be classified as an avoidance strategy.
Being perceived as an equal to their peers was a method of preserving their social identity and this was evident in the coping strategies within the category of *seeking support*. To be equal to their peers, these participants focused on managing their pain. A method of sustaining the image of being equal to their peers is by ‘not telling anyone’ about their pain. This was a key strategy in avoiding publically dealing with pain. Furthermore, there were some adolescents that acknowledged the fear of not being believed. For these adolescents they disclosed ‘telling their mother but not their friends or anyone at school.’ The underlying reason for confiding in their mother but not school was the fear of being unable to prove the pain. Their mothers were more likely to believe them thereby legitimising their pain experiences.

The underlying function of *seeking medical care* was to aid in the management of the pain. Medication was perceived as accelerating the process of waiting for the flare-up to disappear. If medication did not achieve this goal, adolescents disclosed their beliefs that the hospital would be a last resort to dealing with the pain. Medical care was part of the process of their ‘sick role’, which occurred in between their pain-free moments. The pain-free moments were when the adolescents felt normal and medication aided in achieving that state of normality.
Table 19: Underlying function and intent of coping when perceiving pain as a threat

Coping categories and coping intent with an example from the adolescents who perceived pain as a threat.

<table>
<thead>
<tr>
<th>Coping Category</th>
<th>Mental Process</th>
<th>Physical Activity</th>
<th>Seeking Support</th>
<th>Seek medical care</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Coping Strategy</strong></td>
<td>Ignore existence of pain</td>
<td>Stop Activities / Stay still</td>
<td>Rest and relax</td>
<td>Do not tell anyone</td>
</tr>
<tr>
<td><strong>Function</strong></td>
<td>The mental process of getting on with it meant to avoid thinking about the pain. Trying to get on with it meant not dealing or telling others about it.</td>
<td>Stop doing activities when experiencing a pain episode. Measure ability by comparing peers’ ability.</td>
<td>Resting and relaxing was a strategy to stay comfortable while waiting for the pain episode to pass.</td>
<td>Telling someone meant having to deal with the pain risking being perceived as different from peers and as someone unable to cope with the pain.</td>
</tr>
<tr>
<td><strong>Example</strong></td>
<td>“I just don’t like to think about it I like to get on with it”</td>
<td>“I also had P.E and it had started to hurt then as well so I couldn’t run as fast as everyone. I just had to stop and just say that I couldn’t like do it so I didn’t go as far as everyone else”</td>
<td>I don’t move I get comfortable and just stay there.</td>
<td>“I don’t know I just don’t like to complain so I just keep it to myself”</td>
</tr>
</tbody>
</table>
6.3. Profiles – Applying the coping framework

Applying the coping framework presented in Figure 19 above, to the illness representation data meant that the illness representation data were charted against the two identified groups of coping goals. Cognitive and emotional profiles were synthesised from the illness representations data. As shown in Figure 20 below, there were clear similarities in the cognitive profiles across the two groups of coping goals and there were interesting differences in their emotional profiles. Sections 6.3.1 and 6.3.2 below present the cognitive and emotional profiles respectively and these sections provide more detail through in-depth considerations of the similar and dissimilar beliefs in the two groups of coping goals. These profiles created the pain representation corresponding to the different coping strategies (See Figure 20).

![Figure 20: Coping framework with Pain Representations and Profiles](image)

Identified from the data, with cognitive and emotional profiles that are corresponding to the different pain representations.
6.3.1. Clinical characteristics

As reported in Section 4.2.2.4 clinical characteristics were collected from the participants +/-6 months of the interview and were calculated within each category. Disability index and pain intensity scores were self-reported. The nine adolescents charted in the maintenance category had a mean age of 12.98 years with the median being 12 and mean disease duration of 3.89 years with the median being 3. The range of pain scores was from 0 to 68 on a scale of 0 to 100 with a mean of 24.56 and a median score of 22. CHAQ scores ranged from 0 to 1.375 within a possible range of 0 to 3, and had a mean of 0.40 with a median of 0.39 (See Table 20).

### Table 20: Clinical characteristics of the maintenance category

(Adolescents who perceived pain as a challenge)

<table>
<thead>
<tr>
<th></th>
<th>Age</th>
<th>Disease Duration (years)</th>
<th>Pain VAS (0 – 100)</th>
<th>CHAQ (0 -3)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Faye</td>
<td>11</td>
<td>8</td>
<td>0</td>
<td>0.125</td>
</tr>
<tr>
<td>Kevin</td>
<td>11</td>
<td>3</td>
<td>68</td>
<td>0.375</td>
</tr>
<tr>
<td>Gwen</td>
<td>12</td>
<td>6</td>
<td>9</td>
<td>0.625</td>
</tr>
<tr>
<td>Jodie</td>
<td>12</td>
<td>2</td>
<td>26</td>
<td>0.625</td>
</tr>
<tr>
<td>Carrie</td>
<td>12</td>
<td>3</td>
<td>41</td>
<td>0.5</td>
</tr>
<tr>
<td>Adam</td>
<td>13</td>
<td>3</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Daisy</td>
<td>14</td>
<td>3</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td>Ian</td>
<td>15</td>
<td>5</td>
<td>49</td>
<td>1.375</td>
</tr>
<tr>
<td>Harrison</td>
<td>16</td>
<td>2</td>
<td>22</td>
<td>0</td>
</tr>
</tbody>
</table>

(9) **MEAN** 12.89 3.89 24.56 0.40

The ten adolescents whose data were charted in the attend category had a mean age of 13.3 with a median age of 13.5 years (Table 211). For disease duration the mean was 5.4 years and the median duration was 5.5 years. Pain scores ranged from 0 to 83.
on a 0 to 100 scale with a mean of 31 and the median being 18. The CHAQ scores for this group ranged from 0 to 1.75 on a scale of 0 to 3 and the mean was 0.86 and the median was 1.

Table 21: Clinical characteristics of the attend category
(Adolescents who perceived pain as a threat)

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Disease Duration (Years)</th>
<th>Pain VAS (0 – 100)</th>
<th>CHAQ (0-3)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lucy</td>
<td>11</td>
<td>5</td>
<td>18</td>
<td>1</td>
</tr>
<tr>
<td>Neil</td>
<td>11</td>
<td>7</td>
<td>13</td>
<td>0</td>
</tr>
<tr>
<td>Bianca</td>
<td>12</td>
<td>4</td>
<td>10</td>
<td>0</td>
</tr>
<tr>
<td>Vincent</td>
<td>12</td>
<td>7</td>
<td>83</td>
<td>1.25</td>
</tr>
<tr>
<td>Wyatt</td>
<td>13</td>
<td>6</td>
<td>7</td>
<td>0</td>
</tr>
<tr>
<td>Sara</td>
<td>14</td>
<td>2</td>
<td>53</td>
<td>2</td>
</tr>
<tr>
<td>Eleanor</td>
<td>14</td>
<td>3</td>
<td>48</td>
<td>1</td>
</tr>
<tr>
<td>Taya</td>
<td>15</td>
<td>8</td>
<td>11</td>
<td>0.125</td>
</tr>
<tr>
<td>Paige</td>
<td>15</td>
<td>11</td>
<td>6</td>
<td>1.75</td>
</tr>
<tr>
<td>Mia</td>
<td>16</td>
<td>1</td>
<td>61</td>
<td>1.5</td>
</tr>
<tr>
<td><strong>(10) mean</strong></td>
<td><strong>13.3</strong></td>
<td><strong>5.4</strong></td>
<td><strong>31</strong></td>
<td><strong>0.86</strong></td>
</tr>
</tbody>
</table>

6.3.2. Cognitive profiles

Despite the similarities in the cognitive profiles, there were some underlying trends that were different across the two groups. In Table 22, a breakdown of each domain is provided and in the following section each domain is reported highlighting the differences and the similarities in each coping goal category: maintenance and attend. What differed between the two groups was how they interpreted their similar beliefs; this is portrayed further below under each domain.
Table 22: Comparisons of the cognitive profiles

Cognitive profiles of the adolescents in the maintenance category and attend category.

<table>
<thead>
<tr>
<th></th>
<th>Maintenance Category</th>
<th>Attend Category</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Timeline- Chronicity</strong></td>
<td>Expect to get better soon. Hoping that since it is a childhood disease it will be getting better as they grow older.</td>
<td>Believe they will continue having some form of pain when they are older. Some hoped they will grow out of it and not last forever.</td>
</tr>
<tr>
<td><strong>Timeline- Cyclical</strong></td>
<td>Pain is unpredictable. The arthritis has some expected patterns but not cyclical.</td>
<td>Unable to predict pain from doing physical activities. Cannot predict painful joints or when it will hurt.</td>
</tr>
<tr>
<td><strong>Consequences</strong></td>
<td>Pain and disease impacts their lives and affects their family</td>
<td>Pain has a high impact on their lives and creates obstacles for their family.</td>
</tr>
<tr>
<td><strong>Treatment- Control</strong></td>
<td>Medication is a method of gaining temporary control over arthritis.</td>
<td>Treatment is a short-term rather than a permanent approach to gaining control over pain and arthritis.</td>
</tr>
<tr>
<td><strong>Personal Control</strong></td>
<td>Limited control over pain but can use medication to try to control the pain thereby gaining control over the arthritis.</td>
<td>No control over pain, especially increase and decrease of their pain. Medication can help with feeling limited control.</td>
</tr>
</tbody>
</table>

**Timeline- Chronicity**

There was a difference in the two categories in the domain of beliefs about the chronicity of the arthritis. While some adolescents disclosed believing they will be
pain-free soon the underlying reason was different between the two groups. For those adolescents in the maintenance category, their expectancy stemmed from their experiences of their pain episodes, noting that there fewer instances in which pain interfered with their lives as time passed. These adolescents identified that recently there was less of a need to adjust their life to preserve their social identity. In the attend category, the adolescents believed the frequency of their pain episodes may lessen over time and they expressed hope that their arthritis will go away soon. In both categories, there were adolescents who still expected to experience pain when they are older.

*The way that I’m actually going I think it’s going to go away soon. From what I’ve seen so far because it’s progressed a lot from what it was.*”

Adam, age: 13 *(Maintenance Category)*

*“I think I will have it till I die but I think it will gradually start to go away but I will still have it.”*

Lucy, age: 11 *(Attend Category)*

Hope was reflected in both categories as there were adolescents that hoped to grow out of the disease. This belief was based on their perception that since juvenile arthritis is a childhood disease, it should only occur in their childhood. Furthermore, this belief was reinforced by doctors telling them that this disease can go into remission.
“When I first got diagnosed they said that sometimes it can go away with children so hopefully it will go.”

Daisy, Age: 14 (Maintenance Category)

“It’s called juvenile arthritis so I don’t think it will affect me forever and it doesn’t really affect me now so it must be going away.”

Wyatt, Age: 13 (Attend Category)

Timeline- cyclical

In both categories of coping, the adolescents perceived their pain as unpredictable. This is illustrated by Harrison and Bianca below, in the maintenance and attend category respectively. For both Harrison and Bianca, pain can change in a day or weeks.

“It changes, from days, so like one week it would be gone by the day it’s normally just the same it’s always there but it’s worse on some days than others. If I wake and it’s really hurting and then I wake up and its fine.”

Harrison, age: 16 (Maintenance Category)

“Some days I will be fine and the next I can be in pain in quite a lot of pain and then like it can go on for weeks when I am fine but then it can appear out of nowhere.”

Bianca, age: 11 (Attend Category)
Due to the lack of predictability, it was difficult for any of the adolescents to plan for the pain. Therefore, it was not possible to find a pattern in what triggers pain. There was a key difference in how this affected the adolescents in the two categories. For those adolescents in the *maintenance* category knew what to expect and had a strategy in place despite not being able to predict when a flare-up might occur. For the adolescents in the *attend* category they anticipated the pain, as in the case of Paige (age 15), who would plan for a rest day thereby engaging in all-or-nothing behaviour. Like other adolescents in the *attend* category, Paige would engage in bursts of activity and would anticipate that such activity might trigger pain and so would also plan rest time. This behaviour would counteract how pain impacted on their perceptions of normality.

**Consequences**

The adolescents in both the maintenance and the attend categories reported that their pain impacted their lives and their families. All of the adolescents perceived the pain as having more consequences in their lives regardless of the strategy they employed. The adolescents had to adjust when dealing with the pain episodes, especially if they were aiming to preserve their social identity of normality.

“In PE, sometimes I have to sit out and I can’t do many things I could do before. I still can do everything, just not as much as I used to before.

*They [family] have to do certain things for me like I can’t eat with my fork so we have to get special things for me to use.*”

Gwen, age: 11 (*Maintenance Category*)

“When it’s really really bad and I can’t move or anything but when the medication is working then its fine and I could do anything. I can’t do certain things when its bad and I have to go to hospital every two weeks so it does have an effect.”

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My sister always undresses me and things and helps me have a bath and my mum has to do certain things for me and it stops them from doing things they want to do.”

Mia, age: 16 (Attend Category)

Personal control

All of the adolescents felt that they had limited control over their pain. Most of the adolescents believed they had no personal control over pain and their disease. This was mostly due to their perceptions of the unpredictability of the disease. The struggle to gain control over the pain was evident in the adolescents of both categories. The inability to identify triggers to pain episodes was an obstacle to preserving the social identity of normality.

“It could be randomly happening without me doing anything.”
Daisy, Age: 14 (Maintenance Category)

“There is nothing I can do to control my arthritis. If it hurts it hurts I can’t stop it from hurting.”
Bianca, Age: 11 (Attend Category)

Treatment Control

The adolescents in both categories believed that by taking medication they have limited personal control. However, they also perceived that their medication had limited control over pain.

“I can’t control it fully. It will always be there but I can just help.”
Ian, Age: 15 (Maintenance Category)

“’Cause there are some things that can be done, but it can’t just go away completely it can get better but not disappear.”
Mia, Age: 16 (Maintenance Category)
They could achieve their overall coping goal by attempting to have control over their pain with either treatment or their own actions. Nonetheless, treatment was not perceived as a cure for their pain. This meant they still required working towards preserving their social identity when the treatment failed to control their pain.

“It might cure it for a bit but it might not keep being good and it might come back.”

Jodie, Age: 12 (Maintenance Category)

“Treatment makes it go down but I don’t think it can make it fully go away.”

Wyatt, Age: 13 (Attend Category)

6.3.3. Emotional profiles

Data on the emotional representations came from two sources; the cognitive interview and the pain interview. The data were matched to synthesis of the range of emotions adolescents reported when experiencing pain. There was an evident difference in the two groups of adolescents as shown in Table 23.

<table>
<thead>
<tr>
<th>Emotional Profile</th>
<th>Maintenance Category</th>
<th>Attend Category</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normalized the pain; described feelings like ok, fine, normal because they are attempting to carry on with activities and life.</td>
<td>Feelings of anger, really sad, emotional, annoyed or confused because of having to stop activities completely and attend to the pain.</td>
<td></td>
</tr>
<tr>
<td>If they reported feeling angry or sad and upset it was because it was inconvenient having to adjust for pain.</td>
<td>Angry because the pain has a high impact on their lives.</td>
<td></td>
</tr>
</tbody>
</table>
**Maintenance category**

The only adolescents who had normalised their pain were in the maintenance category. These adolescents described their feelings as “ok”, “fine”, “normal” because the pain became a typical experience for them and they were focused on maintaining their sense of normality.

“Normal because it is always there.”

Harrison, age: 16 (Maintenance Category)

In both categories, there were adolescents who labelled their feelings as “angry”, “sad”, or “upset”. However, they reported different reasons for these emotions in each coping category. In the maintenance category the adolescents felt these emotions when they felt that the pain was interfering with their activities and counteracting their preservation of normality. Therefore, they had to make adjustments to their lives. Pain was a challenge to their being perceived as a normal adolescent by their peers.

“Asthritis [is] trying to bring me down.”

Carrie, age: 12 (Maintenance Category)

Despite the adolescents reporting these feelings, they also reported feeling “fine” because they identified that they can tolerate the pain. For example, Ian (age 15) expressed feeling “worried” about the pain reoccurring, however, he also added feeling “fine” because “If I think about it, I can think about ways of dealing with it...”
and it can help.” This meant that these adolescents felt they could still engage in their coping behaviours to maintain their normality. The negative feelings were only reported because of the additional challenge pain was to this sense of normality and having to find ways of overcoming this new challenge.

**Attend category**

In the attend category, the adolescents reported emotions such as “angry”, “sad”, or “upset” because they stopped activities completely to attend to the pain episode. However, these emotions existed during the pain episodes and not when they were identifying themselves as a normal adolescent (pain-free). Therefore, the pain was a threat to this social identity and their belonging to the same group as their peers.

“it makes me upset when it’s hurting but it doesn’t make me upset all the time like when I haven’t got pain I’m fine about it but when I have it hurts.”

Lucy, Age; 11 (Attend Category)

There were adolescents that explicitly explained that their emotional response was due to having pain. They were angry at simply having to deal with a pain episode. Pain became perceived as an entity that was inconvenient and what defined them as a different to their peers. When these adolescents were asked why they felt the emotion they were reporting, they replied that it was due to their pain. For example, Sara (age 14) acknowledged that it was because of “the way I had the pain” and Taya (age 15) disclosed her anger was “because it [pain] was there.”

Most of the adolescents in the attend category also reported feeling angry because of the impact and interference the pain could have on their lives. Therefore the pain
interfered with their goal of preserving their social identity of normality. This was only evident in the attend category. Pain was limiting them and affecting the identity they wished to have and these reasons triggered feeling angry, sad or upset.

“It does make me angry if it’s painful and I can’t play out.”

Vincent, age: 12 (Attend Category)

“Day is ruined.”

Paige, Age: 15 (Attend Category)

“I couldn’t do what I would have liked to.”

Eleanor, Age: 14 (Attend Category)

6.4. Summary

In the coping framework developed from the coping behaviours previously identified, one overarching coping goal emerged. This overarching goal of the coping framework was that the adolescents tried to preserve their social identity, namely to be seen as ‘normal’. Normality was defined as what their peers did in everyday life. Two immediate coping sub-goals were identified and defined as maintenance category and attend category. These categories described the function underlying the coping strategies of the adolescents. Which sub-goal an adolescent tried to achieve was determined by their pain representations. In the maintenance category, the adolescents attempted to maintain a sense of normality by decreasing
activities. The adolescents in the attend category, focused and attended to the pain to continue with normal life when the pain episode is over. The cognitive and emotional profiles were synthesised for both categories.

What emerged from the analysis was that there were no clear differences in the cognitive profiles in both categories. However, the emotional profiles were different. Normalising the pain was an emotional endeavour; occasionally the adolescents in maintenance category felt anger or sadness at having to adjust for the pain but all had managed to normalise the pain. In the attend category, the adolescents clearly define the pain episodes, thereby distinguishing the moments when they are not in pain and when they are. Therefore, this group of adolescents reported negative emotions towards the pain because the pain existed, interfered with their lives and would not allow them to feel normal like their peers. These patterns suggest that there are links between the coping goals and behaviours and their emotional representation of pain. Furthermore, these data signified that the emotional pathways were critical to an adolescent’s coping behaviours. The way the adolescents perceived their pain was rooted in their emotional profiles. One group of adolescents viewed their pain as a normal occurrence and a challenge they adjusted and adapted to. The other group viewed their pain as a threat and dealt with the pain as episodic.
Chapter 7: Self-Regulatory (Feedback Dynamic Component) - Analysis and Findings

7.1. Overview

The investigation of the self-regulatory component of the CS-SRM requires an exploration of the longitudinal and dynamic relationships between mental representations and illness outcomes, such as physical activity. Therefore, the current chapter addresses the thesis’ fifth objective to predict adolescent’s behaviour. The qualitative work presented in Chapter 6 concluded that adolescents’ coping behaviours differed and this difference depended upon the adolescents’ emotional representations. In particular, those adolescents with negative emotional representations (i.e. frustration, anger) were the adolescents who stopped activities to manage their pain; this led to the hypothesis that emotional representations would predict physical activity. To address this research hypothesis sequential analyses including Pearson’s’ correlations, linear regressions, and stepwise regressions and a longitudinal mediation analysis were performed to determine the role of emotional representations and pain in predicting physical activity. This chapter reports the results of each analysis, which precedes building the longitudinal models also presented later in this chapter. The longitudinal models were compared to identify the strongest model predicting physical activity.

7.2. Research Rationale

The main conclusion drawn from the qualitative work presented in Chapter 6 was that a relationship may exist between emotional representations, pain, and physical activity; this finding is congruent with the CS-SRM theoretical framework. In summary, the adolescents who represented their pain as a threat and who coped by focusing on managing the pain (including stopping activities) had higher self-reported pain and held a more negative emotional profile compared to the adolescents who represented their pain as a challenge. In contrast, those adolescents
who represented their pain as a challenge reported lower pain and held a more neutral emotional profile compared to the adolescents who represented their pain as a threat. The last component of the CS-SRM proposes an individual evaluates and appraises the effects of their coping and illness behaviour and as a result, adjusts either their representations or their coping response. Therefore, to evaluate the last component of the CS-SRM, it was hypothesised that repeated measures of the emotional representations, pain and behaviour would predict future behaviour.

7.3. Hypothesis

There is a direct relationship between emotional representations and physical activity and effected by pain, specifically:

I. If emotional representations increase then physical activity score will decrease. 
   \((\uparrow\text{emotions} \rightarrow \downarrow\text{physical activity})\)

II. If pain increases, then emotional representations increase and then physical activity scores decreases. \((\text{pain} \rightarrow \text{emotions} \rightarrow \text{physical activity})\)

OR

III. If emotional representations increases then pain increases and physical activity decreases \((\text{emotions} \rightarrow \text{pain} \rightarrow \text{physical activity})\)

![Figure 21: Hypothesised Models to test](image)

(a) emotional representations mediates the relationship between pain and physical activity
(b) pain mediates the relationship between emotional representations and physical activity.
7.4. Participants

As the flowchart in Figure 4, Chapter 4 (methods), Section 4.2.4.3 demonstrates, the final sample consisted of 264 participants from CAPS aged 11 to 13 years at baseline who each had IPQ-C data at four time points. Table 24 presents the demographic and clinical characteristics of the individuals at baseline of the complete-cases of the sample, compared with the final sub-study sample and the same baseline characteristics for all 1,492 CAPS participants aged between 4 and 16 years of age. Key differences in the sub-study and the CAPS cohort were the proportions of patients in each of the ILAR subtypes; Polyarthritis RF positive (2.7%, 4.1% respectively), PsA (5.9% and 10.6% respectively), and Enthesitis-related arthritis (4.2% and 11.7% respectively).

For the longitudinal models in Section 7.7, only cases with complete data (complete-case) were included for baseline, follow-up years 1 and 2. This reduced the dataset from 264 participants to 55 participants. Table 24 reports the demographic and disease characteristics for this group of 55 participants were summarised to see if they are representative of the larger dataset. There were differences in the complete-cases and the CAPS sub-group, with the subtypes oligoarthritis (38% and 29.5% respectively), polyarthritis RF negative (20% and 12.1% respectively) and there were also a lower numbers of 12 year olds in the complete case compared to the sub-group (22% and 30.3% respectively).
Table 24: Demographic and disease characteristics of the participants in the CAPS

Characteristics of complete-case (n=55) sub-group (n = 264) compared with the CAPS cohort (N = 1,492)

<table>
<thead>
<tr>
<th>Demographic and disease characteristics</th>
<th>Complete-case N (%)</th>
<th>Sub-group N (%)</th>
<th>Total N (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>26 (52 %)</td>
<td>160 (60.6%)</td>
<td>956 (64.1%)</td>
</tr>
<tr>
<td>Male</td>
<td>29 (53%)</td>
<td>104 (39.4%)</td>
<td>536 (35.9%)</td>
</tr>
<tr>
<td>Ethnicity</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>44 (88%)</td>
<td>228 (86.4%)</td>
<td>1082 (72.5%)</td>
</tr>
<tr>
<td>Other</td>
<td>6 (12%)</td>
<td>35 (13.3%)</td>
<td>282 (18.9%)</td>
</tr>
<tr>
<td>Missing</td>
<td>0</td>
<td>1 (0.3%)</td>
<td>128 (8.6%)</td>
</tr>
<tr>
<td>JIA sub-type</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Systemic</td>
<td>2 (4%)</td>
<td>11 (4.2%)</td>
<td>85 (5.7%)</td>
</tr>
<tr>
<td>Oligoarthritis</td>
<td>19 (38%)</td>
<td>78 (29.5%)</td>
<td>602 (40.3%)</td>
</tr>
<tr>
<td>Extended oligoarthritis</td>
<td>2 (4%)</td>
<td>4 (1.5%)</td>
<td>28 (1.9%)</td>
</tr>
<tr>
<td>Polyarthritis RF negative</td>
<td>10 (20%)</td>
<td>32 (12.1%)</td>
<td>216 (14.5%)</td>
</tr>
<tr>
<td>Polyarthritis RF positive</td>
<td>2 (4%)</td>
<td>11 (4.1%)</td>
<td>41 (2.7%)</td>
</tr>
<tr>
<td>Enthesitis-related arthritis</td>
<td>7 (14%)</td>
<td>31 (11.7%)</td>
<td>63 (4.2%)</td>
</tr>
<tr>
<td>Psoriatic arthritis</td>
<td>4 (8%)</td>
<td>28 (10.6%)</td>
<td>88 (5.9%)</td>
</tr>
<tr>
<td>Unclassifiable</td>
<td>2 (4%)</td>
<td>23 (8.7%)</td>
<td>264 (17.7%)</td>
</tr>
<tr>
<td>Other</td>
<td>1 (2%)</td>
<td>8 (3.0%)</td>
<td>54 (3.6%)</td>
</tr>
<tr>
<td>Missing</td>
<td>0</td>
<td>38 (14.4%)</td>
<td>51 (3.4%)</td>
</tr>
<tr>
<td>Age at baseline</td>
<td>11 12 13</td>
<td>18 11 21</td>
<td>82 80 102</td>
</tr>
<tr>
<td></td>
<td>(36%) (22%) (42%)</td>
<td>(31.1%) (30.3%) (38.6%)</td>
<td></td>
</tr>
</tbody>
</table>
7.5. Pain, Emotional Representations and Physical activity over 3 time points

Table 25 gives a description and the score ranges for emotional representations, pain and physical activity, which are included in the analysis. There was a choice of physical activity measures, one that is completed by the adolescents (the Childhood Health Assessment Questionnaire; CHAQ (Nugent et al., 2001) and the other by the parents (the Child Health Questionnaire; CHQ-PF50; (Landgraf et al., 1996). Utilising the CHQ data completed by the parents reduced self-reporting bias of their physical activity. The CHQ provides two composite scores and for this study the physical activity score alone was included. The IPQ-C domain assessed emotional representations.
Table 25: Characteristics of Emotional Representations, Pain, and Physical Activity
Range, median, and interquartile range at Baseline (n = 55), Year 1 (n = 55), and Year 2 (n = 55)

<table>
<thead>
<tr>
<th>Variables</th>
<th>Baseline</th>
<th>Year 1</th>
<th>Year 2</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Median</td>
<td>IQR</td>
<td>Median</td>
</tr>
<tr>
<td><strong>Emotional Representations</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Measured by IPQ-C at every time point, high scores mean more negative emotions.</td>
<td>17</td>
<td>13.5 to 22</td>
<td>16</td>
</tr>
<tr>
<td>Possible range: 0-30</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Pain</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Measured in the CHAQ as a self-reported VAS, high scores are worse.</td>
<td>32</td>
<td>10 to 59</td>
<td>15</td>
</tr>
<tr>
<td>Possible range: 0-100</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Physical Activity</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CHQ outcome, high scores are better scores. Completed by the parents.</td>
<td>32.5</td>
<td>15.2 to 47.3</td>
<td>43.6</td>
</tr>
<tr>
<td>Possible range: 0-100</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note: IPR: Interquartile range, IPQ-C: Illness Perception Questionnaire modified for children, CHAQ: Children’s Health Assessment Questionnaire, VAS: Visual Analogue Scale, CHQ, Child Health Questionnaire.
7.5.1. Interrelationships between pain, emotional representations and physical activity

The relationships between variables were explored using Spearman’s rho to determine the presence of significant associations between physical activity and emotional representations and with pain. Table 26 presents the correlation matrix with analyses performed for each time point. A p value of 0.05 was considered to demonstrate a significant relationship between two variables. Correlations above 0.7 are considered to be strong correlations, while correlations lower than 0.3 are considered weak correlations (Field, 2009). There were strong correlations between pain and physical activity cross-sectionally at year 1 ($r_s = -0.78, p<0.001$), and cross-sectionally at year 2 ($r_s = -0.77, p<0.001$). At baseline, pain and physical activity had a weak correlation ($r_s = -0.36, p<0.001$), but at the same time point emotional representations has shown relationship with physical activity ($r_s = -0.62, p<0.001$) as well as pain ($r_s = 0.62, p<0.001$).

Pain at baseline correlated with emotional representations at year 1 ($r_s = 0.58, p<0.001$) but emotional representations at year 1 also correlated with pain at year 1 ($r_s = 0.60, p<0.001$). Pain at baseline still correlated with emotional representations at year 2 ($r_s = 0.63, p<0.001$). There were also longitudinal correlations between pain at year 1 and physical activity at year 2 ($r_s = -0.78, p<0.001$) and pain at year 2 with physical activity at year 1 ($r_s = -0.61, p<0.001$). From the Table 26, it is clear there are correlations across the time points as well as cross-sectional associations between self-reported pain intensity, physical activity as reported by the parents, and emotional representations, all of which were significant correlations ($p< 0.001$). Emotional representations and pain were both negatively correlated with physical activity which meant that higher emotional representations or pain scores were associated with decreased levels of physical activity.
Table 26: Correlation matrix between physical activity, pain and emotional representations
(Spearman’s Rho) n = 55

<table>
<thead>
<tr>
<th></th>
<th>1.</th>
<th>2.</th>
<th>3.</th>
<th>4.</th>
<th>5.</th>
<th>6.</th>
<th>7.</th>
<th>8.</th>
<th>9.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>BL Physical Activity</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2.</td>
<td>BL Emotional Rep</td>
<td>-0.62**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3.</td>
<td>BL Pain</td>
<td>-0.36*</td>
<td>0.62**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4.</td>
<td>Year 1 Physical Activity</td>
<td>0.40*</td>
<td>-0.38**</td>
<td>-0.46**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5.</td>
<td>Year 1 Emotional Rep</td>
<td>-0.45**</td>
<td>0.52**</td>
<td>0.58**</td>
<td>-0.55**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6.</td>
<td>Year 1 Pain</td>
<td>-0.25</td>
<td>0.31</td>
<td>0.62**</td>
<td>-0.78**</td>
<td>0.60**</td>
<td>1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>7.</td>
<td>Year 2 Physical Activity</td>
<td>0.36*</td>
<td>-0.50**</td>
<td>-0.62**</td>
<td>0.84**</td>
<td>-0.67**</td>
<td>-0.78**</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>8.</td>
<td>Year 2 Emotional Rep</td>
<td>-0.44*</td>
<td>0.59**</td>
<td>0.63**</td>
<td>-0.45**</td>
<td>0.73**</td>
<td>0.52**</td>
<td>-0.61**</td>
<td>1</td>
</tr>
<tr>
<td>9.</td>
<td>Year 2 Pain</td>
<td>-0.19</td>
<td>0.31</td>
<td>0.66**</td>
<td>-0.61**</td>
<td>0.55**</td>
<td>0.83**</td>
<td>-0.77**</td>
<td>0.46**</td>
</tr>
</tbody>
</table>
7.6. Factors affecting physical activity

Regression analyses, both bivariate and multivariate, were used to test longitudinal relationships. Linear regressions were used to describe the relationships between physical activity, the dependent variable, with emotional representations and with pain, the independent variables a simple univariate linear regression model is a statistical equation. The model assumes that:

\[ y = \beta_1 X + \epsilon \]

Where the \( X \) is the observed independent variable, multiplied by the \( \beta \)-coefficient (\( \beta_1 \)) which represents the expected change in the dependent variable (\( y \) added with a normally distributed error \( \epsilon \) that is distributed with mean 0 is added to the equation. The \( \beta \)-coefficient and the 95% confidence interval (95% CI) are reported for each univariate analysis.

As this study has two independent variables, pain and emotional representations, multiple linear regressions were used. This is an extension of the linear regression model equation reported above to model and include covariates. A multiple linear model assumes that:

\[ y = \beta_1 X_1 + \beta_2 X_k + \ldots + \beta_n X_n + \epsilon \]

The difference in multiple linear regressions is that it takes the effect of an independent variable (i.e. emotional representations) on the dependent variable (physical activity) and adjusts for the effect of other variable (pain). When reporting the \( \beta \)-coefficient, this represents the amount the dependent variable changes \( \pm 1 \) unit while the covariate is kept at a constant. Figure 22 represents the variables available for the longitudinal models of multiple linear regressions. The following section reports testing the variables to include in the final model with the aim to identify predictors of physical activity.
7.6.1. Predictors of physical behaviour activity: cross-sectional

As a further investigation of the inter-relationships between pain, emotional representations, and physical activity, cross-sectional relationships were tested through multiple linear regression models at each time point. The aim of this analysis was to investigate the predictors of physical activity cross-sectionally (specifically investigating the relationship between pain, emotional representations and physical activity at each time point; baseline, year 1 and year 2) by testing the effect of the two independent variables, when adjusting for one of the variables in predicting the outcome.

Figure 22: Predictors of physical activity at year 2
As shown in Table 27, the β-coefficient for the baseline model, of physical activity with pain and emotional representations as covariates (β = -0.35; 95% CI [-0.45, -0.25]), is similar to those for year 1 (β = -0.31; 95% CI [-0.39, -0.22]) and year 2 (β = -0.32; 95% CI [-1.39, -0.25]). While, the β-coefficient for the baseline model, of physical activity with emotional representations and pain as covariates (β = -0.57; 95% CI [-1.12, -0.02]), is different to those for year 1 (β = -1.30; 95% CI [-1.76, -0.85]), and year 2 (β = -0.99; 95% CI [-1.39, -0.60]). The adjusted R² values for each model are 0.35, 0.52, and 0.49 respectively, with years 1 and 2 having the best fit.

7.6.2. Determining the roles of parents’ emotional distress, sex, and disease onset as covariates

To address the objective of identifying the confounding role of sex, disease onset and parents at baseline in predicting physical behaviour at year 2, the aim was to build statistical models as hypothesised in (a) and (b). There are three types of variable in a statistical model: the ‘independent’, ‘dependent’, and ‘other’ variables. A confounding variable (U) is defined as an extraneous variable that is correlated with both the independent (X) and dependent variables (Y), as depicted in Figure 23.
Parents’ emotional distress

The CHQ-PF50 is a self-report measure completed by the parents of the adolescents in CAPS, therefore, it might be influenced by the parents’ own state of mind when both observing the behaviour and completing the measure. To account for this source of bias, the General Health Questionnaire (GHQ-30; Goldberg & Williams, 1988) was included to determine the emotional distress in the parent at the time of completing the CHQ-PF50. Table 28 reports the spearman rho’s correlations, between the parent’s emotional distress (GHQ scores) with three main variables. Although the associations were significant, they were all weak correlations.

Table 28: Correlations with parent’s emotional distress

Parent’s emotional distress’ correlations with emotional representations, pain and physical activity n = 84

<table>
<thead>
<tr>
<th></th>
<th>Emotional Rep</th>
<th>Pain</th>
<th>Physical Activity</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Baseline</strong></td>
<td>Parent’s emotional distress</td>
<td>0.30*</td>
<td>0.27*</td>
</tr>
<tr>
<td><strong>Year 1</strong></td>
<td>Parent’s Emotional distress</td>
<td>0.27*</td>
<td>0.22*</td>
</tr>
<tr>
<td><strong>Year 2</strong></td>
<td>Parent’s Emotional distress</td>
<td>0.33*</td>
<td>0.27*</td>
</tr>
</tbody>
</table>

* p < 0.05

Figure 23: Statistical model of the types of variables
where X is the independent variable, Y is the dependent variable, and U is the confounder.

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7.6.2.2. Duration from onset to diagnosis

The time between onsets of symptoms and first presentation to a paediatric rheumatologist (what this study defined as baseline) was included to test as a potential confounder. Time to diagnosis was calculated by subtracting the date of onset from the date of baseline. Table 29 demonstrates the Spearman’s rho correlations between time to diagnosis and emotional representations, pain, and physical activity at baseline, year 1 and year 2. All of the associations were very weak and none reached significant $p$ values.

Table 29: Correlations with time to diagnosis

<table>
<thead>
<tr>
<th></th>
<th>Emotional Rep</th>
<th>Pain</th>
<th>Physical Activity</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Baseline</strong></td>
<td>Time to diagnosis</td>
<td>-0.09</td>
<td>0.05</td>
</tr>
<tr>
<td><strong>Year 1</strong></td>
<td>Time to diagnosis</td>
<td>-0.06</td>
<td>0.15</td>
</tr>
<tr>
<td><strong>Year 2</strong></td>
<td>Time to diagnosis</td>
<td>0.07</td>
<td>0.06</td>
</tr>
</tbody>
</table>

7.6.2.3. Sex

Table 30 reports the Spearman’s Rho correlations between sex and the main variables at baseline, year 1 and year 2. None of the correlations were significant.

Table 30: Correlations with sex

<table>
<thead>
<tr>
<th></th>
<th>Emotional Rep</th>
<th>Pain</th>
<th>Physical Activity</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Baseline</strong></td>
<td>Sex</td>
<td>-0.04</td>
<td>-0.04</td>
</tr>
<tr>
<td><strong>Year 1</strong></td>
<td>Sex</td>
<td>-0.17</td>
<td>-0.12</td>
</tr>
<tr>
<td><strong>Year 2</strong></td>
<td>Sex</td>
<td>-0.14</td>
<td>-0.11</td>
</tr>
</tbody>
</table>
7.6.2.4. Entering variables into a stepwise regression

A stepwise regression model was performed to identify the predictors of physical activity. This was an exploratory technique to identify which variables were independent and statistically significant predictors. Variables were excluded if they were not statistically significant predictors (p>0.1). Table 31 presents the results of the stepwise regression model that included all the possible covariates; emotional representations, pain, parents’ emotional distress, time to diagnosis, and sex as predictors. All of the variables included in the model were from Year 1 to determine those which significantly predicted physical activity at the following year (Year 2). The significant predictors in this model were pain and emotional representations (Table 31). Therefore, only these variables were taken forward for further analysis.

Table 31: Results of the backward stepwise regression

<table>
<thead>
<tr>
<th>Variables</th>
<th>β</th>
<th>95% CI</th>
<th>(Adj) R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>Emotional Representations</td>
<td>-1.21</td>
<td>-1.78, -0.63</td>
<td>0.49</td>
</tr>
<tr>
<td>Pain</td>
<td>-0.27</td>
<td>-0.37, -0.17</td>
<td></td>
</tr>
</tbody>
</table>

Removed

<table>
<thead>
<tr>
<th>Variables</th>
<th>P</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Parent’s emotional distress</td>
<td>0.92</td>
<td>&gt; 0.10</td>
</tr>
<tr>
<td>Time to diagnosis</td>
<td>0.88</td>
<td>&gt; 0.10</td>
</tr>
<tr>
<td>Sex</td>
<td>0.52</td>
<td>&gt; 0.10</td>
</tr>
</tbody>
</table>
7.7. Determining the role of pain in the relationship between emotional representations and physical activity

Cross sectional models are limited in their ability to identify causal relationships. Thus longitudinal models were constructed to determine the role of pain and emotional representations in predicting physical activity.

A complete case analysis of participants with data for emotional representations and pain at baseline and year 1, and for physical activity at year 2, were included in the two models, shown in Figure 24. There were two goodness of fit criteria used to assess which model to take forward, the coefficient of determination ($R^2$) reports the proportion of the total variance in physical activity scores explained by the covariates of self-reported pain and emotional representations, and the likelihood-ratio test, which allows a comparison of the fit with the null model (Figure A) and the alternative model (Figure B). The likelihood-ratio test provided statistical rationale for rejecting the null model in favour of the alternative model.

![Figure 24: Hypothesised Models to test over time](image)

- (a) emotional representations at year 1 mediates the relationship between pain at baseline and physical activity at year 2 or (b) pain at year 1 mediates the relationship between emotional representations at baseline and physical activity at year 2.
7.7.1. Comparing the goodness of fit of Model A: Emotional representations at baseline and Model B: Pain at baseline

As shown in Figure 24, Model A included emotional representations at baseline, pain at year 1 and physical activity at year 2. The adjusted $R^2$ for the linear regression model constructed with these variables was 0.44 (Table 32), meaning Model A explains 44% of the variance in the physical activity scores. Emotional representations at baseline were not statistically significant. This suggested that the pain pathway at year 1 might better explain the relationship between emotional representations and physical activity (mediation). However, this would have to be examined in a mediation analysis. Model B, baseline pain and year 1 emotional representations were used to predict physical activity at year 2 (see Figure 24). This model, as shown in Table 33, explained 33% of the variance in physical activity at year 2. The baseline variable of pain was not significant, while emotional representations at year 1 were a significant predictor.

<table>
<thead>
<tr>
<th>Table 32: Multiple Linear Regression of Figure A</th>
</tr>
</thead>
<tbody>
<tr>
<td>Regression predicting Physical Behaviour at Year 2, ($n = 55$)</td>
</tr>
<tr>
<td>Variables</td>
</tr>
<tr>
<td>Pain (Year 1)</td>
</tr>
<tr>
<td>Emotional Rep (Baseline)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table 33: Multiple Linear Regression of Figure B</th>
</tr>
</thead>
<tbody>
<tr>
<td>Regression predicting Physical Behaviour at Year 2, ($n = 55$)</td>
</tr>
<tr>
<td>Variables</td>
</tr>
<tr>
<td>Pain Baseline</td>
</tr>
<tr>
<td>Emotional Rep Year 1</td>
</tr>
</tbody>
</table>
The likelihood-ratio analysis tested the assumption that Figure A, as the null model was a better fit compared to Figure B as the alternative model, LR chi$^2$ (2) = 9.18, (p=0.01). Figure A, was kept for further mediation analysis.

7.8. Pain mediates the relationship between emotional representations and physical activity

The interaction and effect between the covariates with the independent variable can be assessed by mediation analysis. As shown by the analysis, to determine the role of pain, the regression models are directional, where the mediator variable is associated with both the independent variable and the dependent variable. An example of a mediation model is presented in Figure 25 (Baron & Kenny, 1986). Mediation analysis can be used to determine whether or not the indirect path between the independent and dependent variables through the mediator (path a, and path b, Figure 25) is stronger than the direct path (path c, Figure 25). This is the simple model of mediation (Hayes, 2009) where indirect and direct effect are calculated by simple algebraic models, indirect effect is the difference between the total and direct effect. Direct effect (pathway c in Figure 25) is interpreted as independent pathway of the effect of Z in figure 25. Taking this step by step approach to calculate each of the paths separately is known as the “causal steps approach” (Baron and Kenny, 1986).

To test the mediator the conditions outlined below need to be met.

The statistical strategies for testing mediating effects can either be by multiple linear regressions as recommended and reviewed by Baron and Kenny (1986), or the most recent development is through Structural Equation Modelling (SEM) to estimate using multiple regressions simultaneously. SEM is the preferred method as it is possible to control for measurement error and includes latent variables. However, in other work in paediatric psychology, working with small samples and due to power considerations, the use of multiple regressions was proven necessary (Holmbeck,
In keeping with these studies, and due to the small number of complete cases with longitudinal data, the statistical strategy chosen was the regression approach to test the mediated effect of pain on the relationship between emotional representations and physical activity.

Baron and Kenny (1986) outlined four conditions that need to be met for a variable to be considered a mediator. These were tested by linear regressions and are (letters refer to variables in Figure 25):

(i) The predictor variable (X) must be significantly associated with the dependent (Y)

(ii) the dependent variable (Y) is significantly associated with hypothesised mediator (Z)

(iii) The predictor variable (X) should also be significantly associated with the hypothesised mediator (Z)

(iv) When adjusting for the mediator variable (Z), the impact of predictor variable (X) is less on the dependent variable (Y).

These four conditions can be tested with three multiple regression analysis models. Condition (i) is examined by controlling for the mediator variable (Z). Condition (ii) is examined by controlling for the predictor variable and provides an indirect effect. Condition (iii) and (iv) are tested by a third regression model to compare the change in β-coefficient. The β-coefficient for the total effect is the sum of the β-coefficient for the direct effect and the product of the β-coefficients forming the indirect effect. The total effect mediated was calculated by dividing the product of the β-coefficients
forming the indirect effect (path a x path b) by the $\beta$-coefficient for the total effect to determine the proportion of the total effect that was explained by the inclusion of the mediating variable in the model (last column in Table 34). Indirect effects that are significantly greater than zero were considered to have mediation via the covariate entered into that model. It should be noted that Baron and Kenny (1986) argued that in psychology, it would be unusual for the direct effect to be reduced completely to zero. However, the change in the $\beta$-coefficient should indicate the extent of mediation that the variable Z has on the relationship between X and Y.

More contemporary schools of thought have criticised the Baron and Kenny approach since it is not necessary to meet all conditions such as the first and fourth conditions (Kenny, Kashy and Bolger, 1998; MacKinnon, Fairchild and Fritz, 2007). Other criticisms of the approach are that in comparison to other methods in simulation studies the causal steps approach has the lowest power (Fritz and MacKinnon, 2007) and does not provide a quantifiable indirect effect (Hayes, 2009). Therefore, there has been a move to focus on different methods to measure the indirect effect which is a combination of paths a and b in Figure 25 and is considered the measure of the amount of mediation.

One method of measuring the indirect effect is by measuring estimations of the standard error. The parametric method is using the Sobel Test, and the non-parametric test uses a bootstrapping method. The Sobel test can be limiting because of the constraints of the assumptions and is far more conservative thereby having a low statistical power. To detect a large effect a sample size of 50 would be sufficient, but to detect a small effect a sample size of 1000 would be required (MacKinnon et al, 2002). These approaches are thought to be more accurate than the Baron and Kenny approach. Measuring estimations of the indirect effect by obtaining standard errors can be completed using the bootstrap method which creates an empirical representation of the data by resampling the dataset over 1000 times (Shrout & Bolger, 2002). Whilst this approach has been successfully implemented with cross-sectional designs, some researchers have made the criticism that the bootstrapping approach does not consider the time variable that occurs in longitudinal data in the
mediation analysis and therefore, it is unclear whether estimating standard errors is appropriate in longitudinal designs (Cole and Maxwell, 2003; MacKinnon, 2008).

There are three types of longitudinal mediation models, which differ on how time and change variables are treated, which would fall under different types of SEMs (Cheong et al, 2003). One type is known as cross-lagged panel model which is a type of autoregressive model. The second type is known as a latent-growth mediation model, and the third type is known as latent difference score model (McArdle 2001, Selig and Preacher, 2009). The latent-growth and latent difference scores are both observing change in the time waves. The latent growth model change is observed through trajectories, where change in variable Z (mediator) is correlated with change in variable Y (outcome) at the same time point. Whilst latent different score models utilise the difference between the mediator and dependent variables scores and then applied as though cross-sectional model. Autoregressive model is a model of variables at different waves, and these variables become covariates of each other at later wave. For example, X1 and Y1 would be related and predictive of X2 as well as Y2, allowing observation of stability in variables over time. The step-by-step approach can examine these relationships individually over three waves of data collection, which due to the small sample in this current study, is the proposed approach.

7.8.1. Mediation model

In step 1 of the mediation model the regression of physical activity (year 2) on emotional representations (baseline), ignoring the mediator, was significant, $\beta = -1.39$, $t(49) = -2.96$, $p=0.005$. Step 2 showed that the regression of physical activity on the mediator, pain (year 1), was also significant $\beta = .39$, $t(49) = -5.90$, $p<0.001$. Step 3 of the mediation process showed that the mediator, pain (year 1), controlling for emotional representations (baseline) was significant $\beta = 1.77$, $t(49) = 2.18$, $p=0.035$. Step 4 of the analyses revealed that, controlling for the mediator (pain at year 1), emotional representations (at baseline) were no longer
a significant predictor of physical activity (at year 2), $\beta = -0.77$, $t(49) = -1.95$, $p=0.06$. A Sobel test was conducted and found partial mediation in the model ($z = -2.00$, $p = 0.045$) with approximately 44% of the total effect (of emotional representations and physical activity) being mediated. The indirect effects shown in Table 34 had stronger associations with the dependent variable than the association observed for the direct effect. It is possible that a proportion of the total mediated effect provided an indication of the strength of the indirect effect. The ratio of indirect to direct effect is 0.80, while ratio of total to direct effect is 1.80.

The role of the cross-sectional relationships between pain and physical activity is unclear and it is possible that the effect found through another pathway of physical activity (year 1) to physical activity (year 2). To account for this possible pathway a multiple regression was run to include physical activity at year 1 as well as pain year 1. The first step was to test the relationships between pain at year 1, physical activity at year 1 and physical activity at year 2 through simple linear relationships. The regression of physical activity (year 2) on physical activity (year 1) was significant $\beta = 0.62$, $t(49) = 6.16$, $p<0.001$ (Adj $R^2 = 0.43$). As reported above in Section 7.8.1, the regression of physical activity (year 2) on pain (year 1) was significant, $\beta = -0.39$, $t(49) = -5.90$, $p<0.001$ (Adj $R^2 = 0.44$). Cross-sectionally, the regression of physical activity (year 1) on pain (year 1) was also significant $\beta = -0.55$, $t(49) = -12.15$, $p<0.001$ (Adj $R^2 = 0.75$). When all these variables were included in a multiple
regression model, as shown in Table 35, regression of physical activity at year 2 on emotional representations, while adjusting for pain at year 1 and adjusting for physical activity at year 1 none of the predictor variables remained significant.

<table>
<thead>
<tr>
<th>Variables</th>
<th>β</th>
<th>95% CI</th>
<th>R²</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain (Year 1)</td>
<td>-.17</td>
<td>-.42, .08</td>
<td></td>
</tr>
<tr>
<td>Physical activity (Year 1)</td>
<td>.33</td>
<td>-.07, .73</td>
<td>0.46</td>
</tr>
<tr>
<td>Emotional Rep (Baseline)</td>
<td>-.66</td>
<td>-1.45, .13</td>
<td></td>
</tr>
</tbody>
</table>

7.9. Summary

Based on the findings from chapter 6, the analyses presented in this chapter demonstrated there were significant associations between emotional representations, pain and physical activity. Correlations performed at three time points showed that these relationships have strong correlations (>0.6) both cross-sectionally and longitudinally. However, the role pain and emotional representations play in predicting physical activity was unclear. To address this, longitudinal meditational models were tested. To develop the models, covariates and confounders were tested through a stepwise regression; however, only emotional representations and pain remained in the model as significant predictors of future physical activity.

Two mediation models were compared to identify the roles of emotional representations and pain for the final mediation model. To compare these models, only participants with complete data for the variables of interest at each time point, were included in the analysis. Model A(Figure 24) included emotional representations at baseline as the independent variable with pain at year 1 as the mediating variable while model B (Figure 24) included pain at baseline as the independent variable and emotional representations at year 1 as the mediating variable; physical activity at year 2 was the dependent variable for both models.
(Figure 24). Through comparing the adjusted $R^2$ and performing a likelihood ratio test, Figure A was taken forward for the mediation analysis as this model explained more variance and the likelihood ratio was significantly different. Baron and Kenny’s steps of mediation analysis were performed; pain was identified as a mediator of the relationship between emotional representations and physical activity. A Sobel test was performed and determined that pain mediated 44% of the total effect. Due to the strong cross-sectional relationships between pain and physical activity, another model was tested adjusting for physical activity at year 1, alongside pain at year 1, and emotional representations at baseline, to predict physical activity at year 2. By adjusting for physical activity at year 1, none of the predictors remained significant, suggesting that there may be other pathways through which pain is predicting physical activity.
Chapter 8: Theoretical Synthesis

8.1. Overview

The overarching aim of this thesis is to investigate if Leventhal’s CS-SRM and the associated assessment can be modified and applied for use with adolescents with JIA. This current chapter aims to synthesise the results from Chapters 5 to 7 which investigated the applicability of the CS-SRM with adolescents. This chapter presents the recommended developments of the CS-SRM, to address the implications of the findings from the qualitative work presented in Chapters 5 and 6 and quantitative work in Chapter 7. This chapter concludes with a revised model based upon these recommendations (See Figure 26).

As observed in the review in Chapter 2, questionnaires assessing adolescents’ illness representations are limited to four types of questionnaire; these include the IPQ-R, BIPQ, a modified IPQ and the original questionnaire designed specifically for diabetes. The review in Chapter 2 concluded that there is a need to assess the content validity of the IPQ-R with adolescents’ illness representations (in the case of this thesis; illness representations of JIA). The work presented in Chapter 5 assessed the content validity of a version of the IPQ-R and these results highlighted methodological issues and themes which inform the item development presented in Chapter 9. Chapter 6 and 7 indicated that there needs to be a focus on capturing emotional representations, as these may be potential targets for interventions. The development of a modified assessment of mental representations is presented in Chapter 9.

8.2. Creation and development of mental representations in adolescents- recommended modifications

The key finding of the framework analysis (Chapter 5) was the discovery that, in the case of JIA adolescents’ *pain experiences* and *pain beliefs* were driving their
conceptualisation of their experiences of arthritis (see Figure 26) rather than broader perceptions about the illness. Specifically this means that the main symptoms of the condition play a stronger role than broader illness representations of the condition per se.

How perceptions, actions, concepts and language are used in the management of a health threat is described in the Common Sense Self-Regulatory Model (CS-SRM; Leventhal & Diefenbach, 1991). This model describes that the input of sensory function (i.e. chest pains) raises concepts (i.e. stroke/heart attack) and is checked against the underlying abstractions that are formed from the individual’s experience. These abstractions are also known as prototypes. Prototypes are acquired over time and are subjected to change from new information and/or from observation (Leventhal et al., 2011). The results from this thesis highlight the important role of social context. For example, in Chapter 5, the adolescents’ perceived consequences of pain was dependent on where it occurred (i.e. home or school), this was even more evident in Chapter 6, when the context (home or school) helped determine whether or not they sought social support (told someone or not). These results lead to the recommendation that a behaviour check is added to the model (See Figure 26). A behaviour check would address the underlying reason for the behaviour. This recommended development of the CS-SRM accounts for the interaction of cognitive and behavioural processes with the behavioural environment.

Another recommendation is to add another process: checking whether the patterns of symptom occurrence fit with existing knowledge and previous experiences of pain. Figure 26 indicates how these checks are informed by conceptualisation of arthritis as well as conceptualisation of pain. According to the most recent depiction of the CS-SRM, the sensory function is checked against the information of an elicited prototype (pattern, location, onset and duration) to determine if there is a deviation (Leventhal et al., 2012). This ‘search and match’ of previous information provides the framework for action (Leventhal et al., 2012). This was evident in the results of Chapter 6, the adolescents’ pain representations were dependent on whether pain was prototyped (perceived) as episodic or constant. Those adolescents
who perceived pain as a threat prototyped their pain episodes as a series of acute pain. The others redefined their prototypes and normalised the reoccurring pain condition.

This process of *normalisation*, that is, changing the perception of an experience previously considered abnormal (i.e. pain for the adolescents) to normal, is not a new concept in the literature. An experience elicits both the retrieval and constructions of knowledge and these processes are thought to be constrained by context (Kahneman & Miller, 1986). The experience would be more likely to be perceived as normal if it raises memories of similar experiences, and if there are fewer or no counterfactual alternatives (Kahneman & Miller, 1986). While Kahneman and Miller (1986) are referring to how individuals store knowledge and experiences this is still applicable to how adolescents are conceptualising their experiences of an LTC.

As outlined in Chapter 2, cognitive science indicates that people build mental representations of reality, and within these mental representations, there are expectations and constructed knowledge (Kahneman & Tversky, 1982; Kahneman & Miller, 1986; Leventhal et al., 2011; Leventhal et al., 2012). When discrepancies occur between reality and mental representations, emotional responses are elicited (Kahneman & Tversky, 1982; Kahneman & Miller, 1986). Discrepancies between reality and mental representations were found in the participants in Chapter 6. Adolescents define “normal adolescents” by generalising from what their peers do, and this constructed the mental representations. This involves social processes such as *social comparison* (Chapter 6 results). The adolescents in the current study who felt that their reality did not match their idea of normal (they held counterfactual alternatives) were those who expressed the negative emotional representations (i.e. frustrations and anger). The second section of Chapter 6, demonstrated how the coping framework mapped back to the mental representations, and why there were apparent differences between adolescents who perceived pain *either* as a threat *or* as a challenge. The fundamental difference distinguishing between these two groups of adolescents was through their *emotional representation of the symptom* indicating there may be a discrepancy between reality and their mental representations.
This discrepancy between mental representations and reality may also be influencing the perceptions of pain intensity as well as predicting physical activity. Findings reported in Chapter 7 indicated that there may be internal processes linking between emotional representations and perceived pain intensity. Together, emotional representations and self-reported pain intensity predicted physical activity at the same time point and also future physical activity. This suggests that if there is a discrepancy between the expected and constructed mental representations and reality, causing emotional responses such as frustrations, depression or anxiety, this may also be influencing how the adolescents are reporting and experiencing pain while also decreasing the adolescents’ physical activity.

What emerged from the data is that the understanding of arthritis and the emotional representations were reliant on the adolescents’ experiences (related to pain) and information they have about the condition (Chapter 5) and the expectations they have for themselves (Chapter 6). The social context and self-concept were not only considered during action plans as proposed by the CS-SRM (see below), but self-concept also has a bidirectional process with schemata and prototypes (see Figure 26).

The results from Chapter 5 also suggested that causal attributions of pain are the first step in the process from identity to eliciting prototypes about control, treatment and behaviours. All this contributes to how adolescents make sense of their pain. This leads to another proposed modification to CS-SRM for adolescents; that is to include a prototype check of cause as one of the first steps of the process (Figure 26).

The results regarding causal beliefs and identity in Chapter 5 implied that adolescents search for a coherent explanation, one that makes sense to them. The adolescents search for triggers or experiences that can account for a cause-effect process. This is supported by data from those adolescents who had an early onset of JIA, and were unclear or uncertain about the cause of the condition. Their uncertainty and inability to explain a causal process is not based on inability to think in abstract terms but rather highlights a different disease course to other adolescents and is
dependent on their experiences. This highlights an issue with stratifying by cognitive stages, which makes the assumption that age reflects a cognitive stage. However, social determinants play such a vital role in cognitive development that age alone cannot account for coherence of a condition (Bandura, 1986). Furthermore, this assumption that age equates to a cognitive developmental stage, could be creating perceptions that there would be developmental issues which may be influencing the communication and information in clinic. Theoretically, moving into a modified CS-SRM framework that takes into account experiences and is reflective of how knowledge is stored and created, could improve this communication.

8.3. Function and action plans of adolescents – recommended modifications

Mental representations create the framework and the underlying reason for action (Leventhal et al., 2011). The first section of Chapter 6 provided a new coping framework which showed that individuals have different coping goals underlying their behaviours (the represented set target) in order to deal with pain (Section 6.2). The results of this analysis suggested that the role of social comparison and social identity are underplayed in the current CS-SRM theoretical framework. However, Leventhal and colleagues (Leventhal et al., 2012) acknowledge there are social constraints on the variance of behaviour. This variance is accounted for in the action plan which takes into consideration the environment for behaviour, linking treatment to everyday life.

According to Leventhal and colleagues (2011) the IPQ and related questionnaires are not sufficiently sensitive to predict variations in behaviour because they are not able to differentiate between representations of patients with the same condition, especially if the condition elicits homogenous representations. This shift from illness perceptions to symptom perceptions can assess heterogeneous representations. This shift may also capture changes in the representations that lead to changes in behaviour (Leventhal et al., 2011). Furthermore, Leventhal and colleagues (2012) argue that when a condition is chronic with an abstract time frame, individuals with
an LTC attempt to use cues and indicators to regulate and evaluate behaviour. This may account for findings in this study of a relationship between adolescents’ pain experience, pain beliefs and their conceptualisation of their LTC. Therefore, adolescents cognitively attempt to associate abstract concepts in their concrete experiences, which is arguably part of cognitive processes and systems (Leventhal et al., 2012; Quinn & Eimas, 1997).

In this approach, actions or behaviours are the result of the combination of mental representations of health threats and action plans, informed by treatment representations and procedures that are driven by mental representations (Leventhal et al., 2012). One of the proposed modifications to CS-SRM for adolescents is highlighting the need to identify goals, and identify the ultimate objective of the behaviour. In Chapter 6, goal adjustment and goal pursuit emerged as major drivers of behaviours, in this case, with a goal of achieving their definition of normality. In 2012, Leventhal and colleagues, proposed CS-SRM included representation of set targets that could have been both concept and experience and either objective or subjective as part of the model that led to an output function (Leventhal et al., 2012). Leventhal and colleagues (2011) maintained that investigating coping strategies such as problem-solving coping or seeking social support to predict behaviour is inconsistent with the CS-SRM. The type of coping strategy employed is not indicative of what behaviour an individual will engage in nor is the coping strategy indicative of the reason underlying the behaviour. The results of Chapter 6 found that coping strategies such as seeking social support have underlying intent and goal setting which differ across the adolescents. The procedures are elicited to target specific outcomes. For example, adolescents can choose not to tell anyone about their pain, however, there were different outcomes. For some adolescents not telling was aiming to reduce emotional cost (i.e. embarrassment in class), for other adolescents not telling was to maintain a similar status as their peers (i.e. not being treated differently or given special treatment). For this reason, the modified model includes behaviour check as shown in Figure 26. This addition of an environment and behaviour check is not novel as the CS-SRM is considered to be a situated theory.
where cognitive and behavioural processes interact within the behavioural environment (Greeno, 1998).

Lastly, the CS-SRM is considered to involve a feed-forward effect, where represented set target and outcome expectations are established and associated with a cue, and so the procedure can activate the somatic stimuli (Cameron et al., 1993; Leventhal et al., 2011; Leventhal et al., 2012). This is an important association, especially with pain, and can account for cognitive processes such as anticipatory processing (anticipating pain) and attentional bias (looking for cues and triggers of pain onset). Therefore, an association between the output and the input of the model is included in Figure 26. An example of this feed-forward effect is when the adolescents in Chapter 6 reported allowing for a pain day (a day with no activities to allow for anticipatory pain) after a day of activities.

8.4. Summary of recommendations

The diagram (Figure 26) summarises the proposed modifications, highlighting new concepts and processes including:

i. a separation of identity and cause prototype checks at the beginning of the process of eliciting the relevant mental representations

ii. the schemata of both condition and symptom in this case arthritis and pain

iii. self-concept which also informs the prototypes and the selected behaviour

iv. a behaviour check that evaluates the impact a behaviour would have on the environment and to self

v. a feedback loop to the schemata for reinforcement or change and to assess the impact on self-concept.

vi. an association between the output and input to the model. To account for when behaviours can induce somatic stimuli.
Figure 26: Modified CS-SRM for adolescents
Proposed modifications based on original model Levental et al (2012) including the new information form the qualitative work previously reported.
Chapter 9: Development and validation of the new questionnaire – Analysis and Findings

9.1. Overview

The results of the content analysis and framework analysis reported in Chapter 5 showed that:

i) adolescents’ beliefs about symptoms rather than illness representations play a stronger role in their responses to living with JIA

ii) there was a need to modify existing items of the IPQ-R and develop new items.

Section 9.2 of this chapter outlines the development of the new items and modifications to the domains driven by the qualitative work presented in Chapters 5 and Chapter 6. This addresses Aim 2, objective 6 of this thesis: to develop an adolescent specific measure of illness representations. Section 9.3 to Section 9.4 presents the results of the validity and reliability testing of the new questionnaire. This addresses, Aim 2: objective 7 of this thesis: to test and evaluate the psychometric properties of the new measure.

9.2. Developing new questionnaire

As mentioned in Chapter 4, Methods, Section 4.3.2, questionnaire development follows a systematic process, which is made up of a sequence of stages to ensure that the new measure is able to capture and assess the constructs required. Figure 27 shows the stages required to support the claim that the questionnaires have content validity, that is, how well the items assess the concepts they intend to measure. The name Pain Perceptions Questionnaire for Young People (PPQ-YP) was chosen to
reflect both the aim of the questionnaire and to maintain the theoretical link with other measures of illness perceptions (see Appendix 9). The aim of the PPQ-YP is to assess the pain representations of adolescents (aged between 11 to 16 years) with a relapsing remitting, long-term condition, in this case, JIA.

The following chapter is organised according to the phases presented in Figure 27. The results from the interviews (developing the questionnaire) are presented first. The pre-testing phase and the field-testing (validation) phase, follows the item generation section. Criterion validity is usually tested by comparing a new questionnaire to a gold standard that measures the same constructs (Streiner &
Norman, 2008; Fayers & Machin, 2007) however, criterion validity assessment was not undertaken because PPQ-YP is a novel questionnaire, and therefore, there is no pre-existing gold standard to test against.

Results from the qualitative studies reported in Chapter 5 were the data sources used for item generation and to design the response scales and response options for the PPQ-YP. For the pre-testing phase, participants were adolescents without JIA, whereas the field-testing (validation phase) involved adolescents with JIA.

The validity testing in both pre-testing and field-testing phases refers to different categories of validity; content, face, criterion, and construct (Streiner & Norman, 2008; Fayers & Machin, 2007). Content validity is one of the preliminary stages of questionnaire development and refers to the process of assessing the degree to which all possible domains of the measured construct are included. The initial item pool of the PPQ-YP was expanded using data from the cognitive interviewing study to ensure coverage of modified or new constructs. Starting with a large initial item pool allows for the possibility of dropping the items after statistical testing.

Face validity includes assessing the comprehensiveness of the items and this was tested with a sample of adolescents without JIA aged between 11 to 16 years. Construct validity of the PPQ-YP was assessed using principal component analysis (PCA) and testing hypotheses regarding anticipated subscale inter-relationships.

9.2.1. Development of the Pain Perceptions Questionnaire for Young People

The development of the PPQ-YP spanned two areas. The first was the selection of the items and the second addressed response formats and scoring. The following section describes the development of the items and the justifications for modifying existing items of the IPQ-C. This is followed by a description of the proposed changes to the response formats as well as the scoring of the domains.
9.2.1.1. Item generation for the PPQ-YP

New items and constructs were included in the PPQ-YP to take into account the changes to the conceptual model. There were changes to the assessment of emotional representation, by separating the items into an assessment of the amount of time and extent of an emotion. Changes to the domain of timeline items included a new measure of episodic pain timelines.

Changes to the assessment of causal attribution items now address episodic pain rather than underlying cause of the condition. The identity subscale now also included a measure of symptoms associated with treatment. The consequences domain now included more contextual items regarding the pain experiences.

Summaries of the changes to each of the original and existing IPQ-C construct are presented in Table 36.

Table 36: Comparision of IPQ-R to the PPQ-YP

<table>
<thead>
<tr>
<th>Existing Construct</th>
<th>Changes in the construct</th>
</tr>
</thead>
<tbody>
<tr>
<td>Identity</td>
<td>This construct now includes symptoms associated with treatment as an additional column. This has been done previously with modifications of the IPQ-R to measure hypertension.</td>
</tr>
<tr>
<td>Illness coherence</td>
<td>This construct now assesses the degree to which the adolescent understands aspects of pain including; cause, treatment, and if they have any questions. These are aspects of the condition (such as cause and treatment) that the adolescents said they did not fully understand.</td>
</tr>
<tr>
<td>Cause</td>
<td>This construct now assesses beliefs about the causes of the most recent episode of pain rather than overall cause.</td>
</tr>
<tr>
<td>Timeline chronicity</td>
<td>This construct now measures two types of chronicity beliefs of pain: how long they will continue getting pain in their lifetime and how long they believe individual episodes of pain will last.</td>
</tr>
<tr>
<td>Construct</td>
<td>Description</td>
</tr>
<tr>
<td>----------------------</td>
<td>-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------</td>
</tr>
<tr>
<td>Timeline Cyclical</td>
<td>This construct aims to measure the patterns of episodes looking at cyclical timeline beliefs.</td>
</tr>
<tr>
<td>Consequences</td>
<td>This construct aims to measure impact of the condition on their lives includes costs (financial) relationships and 'life'</td>
</tr>
<tr>
<td>Personal control</td>
<td>This construct aims to measure if participants hold beliefs about their controllability over disease. This construct is associated with coping.</td>
</tr>
<tr>
<td>Treatment Control</td>
<td>This construct aims to assess beliefs in treatment or recommended advice. While also measuring the expectancy of treatment curing/controlling illness.</td>
</tr>
<tr>
<td>Emotional Representation</td>
<td>The aim of this construct is not a proxy indicator of patients’ general mood but to provide an assessment of the emotional responses generated by illness. The assumption is these emotional responses are associated with emotion-focused coping procedures.</td>
</tr>
</tbody>
</table>

**Modification of items**

Modification of individual items was only made if it could be justified by the results of the analyses. Table 37 to Table 42 present the changes. There were four categories of change; new item (N), old item removed (R), item reworded to be about pain (REP) or item reworded because of comprehension (REC). Each item was categorised into one of the four as shown in the last column of Table 37 to Table 42.
Table 37: Identity and Illness Coherence items of the IPQ-C compared with PPQ-YP
Differences in Identity and Illness coherence items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Feeling pain</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>Sore mouth/throat</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>Joints feel sore</td>
<td></td>
<td>N</td>
</tr>
<tr>
<td>Tightness</td>
<td></td>
<td>N</td>
</tr>
<tr>
<td>Change in my mood</td>
<td></td>
<td>N</td>
</tr>
<tr>
<td>Added column: Because of my treatment</td>
<td></td>
<td></td>
</tr>
<tr>
<td>My symptoms are puzzling to me</td>
<td>I feel confused about why I get pain</td>
<td>REC</td>
</tr>
<tr>
<td>I understand how my treatment for pain works</td>
<td></td>
<td>N</td>
</tr>
<tr>
<td>My arthritis is a mystery to me</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>My arthritis doesn’t make any sense to me</td>
<td></td>
<td>REC</td>
</tr>
<tr>
<td>I don’t understand my arthritis</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>When I have pain I understand what causes my pain</td>
<td></td>
<td>N</td>
</tr>
</tbody>
</table>

Table 38: Cause items of the IPQ-C compared with PPQ-YP
Differences in the cause items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td>List of Causes with Likert Scales</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>Circle three causes you believe cased your most recent pain [Changed instructions]</td>
<td>N</td>
<td></td>
</tr>
</tbody>
</table>

Table 39: Timeline items chronicity and cyclical of the IPQ-C compared with PPQ-YP
Differences in timeline items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td>My arthritis will go away soon</td>
<td>I believe I will stop getting pain soon</td>
<td>REC</td>
</tr>
<tr>
<td>Will be with my for the rest of my life</td>
<td>I believe I will keep having pain when I am an adult</td>
<td>REC</td>
</tr>
<tr>
<td>Will last a long time</td>
<td>When I get pain, it lasts a long time</td>
<td>REP</td>
</tr>
<tr>
<td>Will soon get better</td>
<td>Over time I am getting pain more often</td>
<td>N</td>
</tr>
<tr>
<td>Will improve in time</td>
<td>When I get pain I think my pain will improve in an hour, a day or less, a week or less, two weeks or less, more than two weeks</td>
<td>R</td>
</tr>
<tr>
<td>My arthritis changes everyday</td>
<td>My pain changes everyday</td>
<td>REP</td>
</tr>
<tr>
<td>My arthritis comes and goes in cycles</td>
<td>My pain comes and goes</td>
<td>REC</td>
</tr>
<tr>
<td>It is hard to tell what my arthritis will do next</td>
<td>I can predict when I get pain</td>
<td>REC</td>
</tr>
</tbody>
</table>
Table 40: Consequences items of the IPQ-C compared with PPQ-YP
Differences in consequences items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Consequences</td>
<td></td>
<td></td>
</tr>
<tr>
<td>My arthritis is serious</td>
<td>When I get pain it makes me think my pain is: not serious at all, a bit serious, serious, somewhat serious, Is very serious, very serious</td>
<td>REC</td>
</tr>
<tr>
<td>Does not have much effect on my life</td>
<td>When I have pain, it stops me from taking part in activities such as PE such as school work, school friends</td>
<td>N</td>
</tr>
<tr>
<td>Affects what other people think of me</td>
<td>When I have pain, it affects me at school such as school work, school friends</td>
<td>N</td>
</tr>
<tr>
<td>Costs a lot of money to my family or the hospitals</td>
<td>When I have pain, it affects how I am at home</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 41: Personal and Treatment control items of the IPQ-C compared with PPQ-YP
Differences in control items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Personal Control</td>
<td></td>
<td></td>
</tr>
<tr>
<td>There is a lot I can do to control my arthritis</td>
<td>I can do a lot to control my pain</td>
<td>REP</td>
</tr>
<tr>
<td>What I do makes my arthritis better or worse</td>
<td>This is the amount of control I feel I have over my pain: no control, some control, quite a bit of control, a lot of control full control</td>
<td>N</td>
</tr>
<tr>
<td>The way my arthritis acts depends on me</td>
<td>There are things I can do to make my pain better</td>
<td>REP</td>
</tr>
<tr>
<td>What I do now will not change how arthritis affects me in future</td>
<td>[Item removed]</td>
<td>R</td>
</tr>
<tr>
<td>Nothing I do will have any effect on my arthritis</td>
<td>There are things I do now which can affect future pain</td>
<td>REC</td>
</tr>
<tr>
<td>Treatment control</td>
<td></td>
<td></td>
</tr>
<tr>
<td>My treatment will cure my arthritis</td>
<td>I am in control of my treatment for my pain (personal control)</td>
<td>N</td>
</tr>
<tr>
<td>There is very little that can be done to make my arthritis better</td>
<td>My treatment helps my pain get better</td>
<td>R</td>
</tr>
<tr>
<td>My treatment will prevent the bad effects of arthritis on me</td>
<td>My treatment protects me from pain</td>
<td>REC</td>
</tr>
<tr>
<td>Nothing helps my arthritis</td>
<td>I can continue with my activities because of my treatment</td>
<td>N</td>
</tr>
</tbody>
</table>

Table 42: Emotional representations items of the IPQ-C compared with PPQ-YP
Differences in emotional representation items are coded

<table>
<thead>
<tr>
<th>IPQ-C Items</th>
<th>PPQ-YP Items</th>
<th>CODE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
The PPQ-YP was separated into sections of similar style questions. This means that specific instructions explaining the response formats can be provided before each section. Instructions were placed on a front page with a sentence emphasising that these questions are about their views. The PPQ-YP items were separated into sections labelled, Section A, Section B, and so on, each with different instructions and response formats. The decision to separate the questionnaire into sections was made to ensure that instructions were adjacent to the relevant items. This was to keep the questionnaire as clear and easy to complete as possible.

9.2.2. Forming response scales

There are a variety of scale formats for questionnaires, which can classified as either binary format (yes/no) or ordinal scales (rank categories). Walker and colleagues (2006) argued that a binary format is more appropriate response style for younger children and used a true/false response format for the CIPQ. However, the participants of the cognitive interviews were able to answer the Likert scales and understood that the scale represented a continuum.
**Likert scales**

Likert scales were originally designed to include a neutral middle option along with four other category responses (Likert, 1932). There has been debate in the literature about the use of a neutral middle option such as **neither agree nor disagree**, and whether adding a neutral option might introduce bias as respondents are more likely to choose a neutral option if offered (Schuman & Presser, 1981; McColl et al., 2001). Another argument against including a neutral option is that the respondents that are less motivated to exert cognitive effort to pick a direction are then more likely to be a ‘satisficing’ respondent (Krosnick, 1991). However, a recent uptake on this debate is that the neutral option is a proxy for ‘I do not know’ and some have argued that respondents select this option to avoid social embarrassment of explicitly saying this (Sturgis et al., 2014). On the other hand, removing a neutral option might also increase the likelihood of meaningless responses. In the cognitive interviewing study, described in Chapter 5, a verbal probe was included to explicitly ask the individuals who selected **neither agree nor disagree** what they believed this option meant. In response to this verbal probe these participants indicated that when it was picked this was a proxy for ‘sometimes’ rather than a substitute for ‘I do not know’ (as reported in Chapter 5). For these items it was clear that the responses verbal anchor labels did not match the intended responses.

**Visual analogue scales**

Visual analogue scales (VAS) as described in Chapter 4, are lines with verbal anchor labels of extreme states at either end. In health psychology the use of VAS has been less frequent. The reason for this may be because of the higher demand on resources to measure the outcome (i.e. having to use a ruler to measure where the participant marked the VAS). One method of reducing this demand on resources is computerised data-collection. McCormack et al, (1988) found that despite the VAS being more sensitive and easier for patients to complete (in comparison to categorical scales such as Likert scales) there are clusters of responses in either low, middle or high parts of the scale. A possible explanation of these clusters is the lack of reference points along the scale with only the extreme anchor points available. Questionnaires such as
the PPQ-YP are measuring beliefs and concepts, such as duration and control, which lie on a continuum. To treat these concepts as a continuum and also address the problem of no reference points, the PPQ-YP was designed to have response scales that use a combination of VAS and ordinal categories for reference points for the adolescents to complete. The scoring of the response format is calculated by measuring millimetres of the distance along the scale, the scores range would be between 0 and 100.

*Verbal anchor labels*

The choice of which verbal anchor labels to use is an important decision when using Likert scales. Research into the relationship between anchor labels and re-test reliability found that more specific labels resulted in better re-test reliability (Weng, 2004). The IPQ-C utilises a 5 point rating scale that were labelled as Strongly Disagree, Disagree, Neither Agree nor Disagree, Agree and Strongly Agree. For 21 of the PPQ-YP items this was a relevant response and so these category labels were kept as shown in Figure 28.

![Figure 28: Verbal anchors of level of agreements on a visual analogue scale.](image)

The literature on questionnaire development advocates using labels, which are specific to the questions being asked (Streiner & Norman, 2008; Fayers & Machin, 2007). For that reason, the items where the cognitive interview data indicated that a continuum was a more appropriate approach, the response formats and the reference points for these items were modified. Figure 29 gives an example of the different verbal anchor labels used, in this example the item was asking if the adolescents believe their condition was serious.
9.3. Pre-testing the questionnaire

The face validity of the PPQ-YP was assessed by asking adolescents without JIA to read the PPQ-YP items and answer a feedback questionnaire (Appendix 6). The adolescents provided feedback on each of the four sections of the PPQ-YP. They were asked to comment on the following aspects: clarity of the instructions; identification of specific words or terms that were difficult to understand; and if there were items they did not understand. These questions allowed for analysis of the comprehensibility of the items as well as the layout. Finally, there was an open-ended question for the young person to write any other comments. These data were collected and relevant changes were made to the first version of the PPQ-YP to address the issues that were identified.

9.3.1. Participants in the pre-testing

Eighteen adolescents without JIA reviewed the PPQ-YP. As this was an opportunity sample, there was not a gender match and the group only consisted of females with a majority (nine) aged 12 years old. There were three 11 year olds, three 13 year olds, two 15 year olds, and one 16 year old.

9.3.2. Proposed changes

There were some words and concepts that were difficult for the adolescents. Table 43 summarises those items that required further clarification. All of the adolescents reported completing the questionnaire within 10 minutes; this suggests that its length was not a burden. Furthermore, none of the adolescents reported difficulties in understanding or using the response formats. Once the outcomes reported in the last
column in Table 43 were included in the second version of the PPQ-YP, the questionnaire was ready to be sent to the broader sample of adolescents with JIA aged between 11 to 16 years old.

Table 43: Changes for PPQ-YP version 2

<table>
<thead>
<tr>
<th>Item</th>
<th>Issue</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Instructions – on the first page</td>
<td>Include ‘how you feel’ instead of just saying ‘your views’</td>
<td>We are interested in your views and how you feel about pain you may have relating to your arthritis</td>
</tr>
<tr>
<td>Tightness (symptom)</td>
<td>Not understood</td>
<td>‘Feelings of tightness in my body’</td>
</tr>
<tr>
<td>I believe having pain makes my family spend more money</td>
<td>Unclear</td>
<td>I believe my family spend more money because I have pain</td>
</tr>
<tr>
<td>Things I do now can affect my future pain</td>
<td>Unclear</td>
<td>There are things I do now which can affect whether I have pain in the future</td>
</tr>
<tr>
<td>Smoking/ drinking causal attribute</td>
<td>Not relevant</td>
<td>Any other cause that you think of</td>
</tr>
<tr>
<td>Give an example to section D</td>
<td>Hard to follow instructions</td>
<td>Provided example to section D</td>
</tr>
</tbody>
</table>

9.4. Field-testing the PPQ-YP

The seventh objective of this thesis was to test the psychometric properties of the modified questionnaire; this included identifying dimensions, the internal reliability of these dimensions, and the re-test reliability of the PPQ-YP. Adolescents with JIA completed the modified PPQ-YP to collect initial data to test the questionnaire. The results are reported to address each psychometric objective, aiming to test this questionnaire (identifying dimensions, internal reliability and re-test reliability).

9.4.1. Participants in the field-testing

Eighty participants recruited from the CAPS cohort took part in field-testing the questionnaire. This represented a response rate of 38% from 221 adolescents aged between 11 to 16 years, who were invited to complete the questionnaire alongside other CAPS questionnaires. However, four participants had to be removed from the PPQ-YP sample, one adolescent consented but did not send the questionnaires, another adolescent was given a different diagnosis and was no longer eligible for CAPS, another sent the questionnaire empty twice, and another adolescent’s PPQ-YP
was completed by the parent and not the adolescent. This brought the total of adolescents included to 76 participants. Table 44 provides clinical and demographic descriptive information regarding the 76 participants. The mean age of participants was 13.93 years (SD 1.56) with disease duration (calculated from onset of disease) ranging from 0.96 to 12.42 years with mean of 5.89 (SD 3.08).

<table>
<thead>
<tr>
<th>Characteristics of participants (n = 76) compared with the total CAPS cohort (N = 1,492)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Demographic and disease characteristics</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Gender</td>
</tr>
<tr>
<td>Female</td>
</tr>
<tr>
<td>Ethnicity</td>
</tr>
<tr>
<td>White</td>
</tr>
<tr>
<td>Other</td>
</tr>
<tr>
<td>Missing</td>
</tr>
<tr>
<td>JIA subtype</td>
</tr>
<tr>
<td>Systemic</td>
</tr>
<tr>
<td>Oligoarthritis</td>
</tr>
<tr>
<td>Extended oligoarthritis</td>
</tr>
<tr>
<td>Polyarthritis RF -</td>
</tr>
<tr>
<td>Polyarthritis RF +</td>
</tr>
<tr>
<td>Enthesitis related arthritis</td>
</tr>
<tr>
<td>PsA</td>
</tr>
<tr>
<td>Unclassifiable</td>
</tr>
<tr>
<td>Other</td>
</tr>
<tr>
<td>Missing</td>
</tr>
</tbody>
</table>

9.4.2. Results for objective 1: to identify the dimensions of the PPQ-YP

As mentioned above, the items of the PPQ-YP were developed under the domains that the items were related to (i.e. the item ‘I can do a lot to control my pain’ was related to personal control) and items are organised in the questionnaire according to
these domains as well as by similar scoring, which is why there are four sections in the questionnaire. Dimensionality is the process of searching for patterns of correlations among items’ scores. Theoretically, correlated items are assumed to measure a single dimension, independent of other sets of correlated items. However, for the items of the PPQ-YP, there are strong interrelationships between the items, not necessarily just within the previously identified domains. To identify the factor structure of the PPQ-YP, a PCA was conducted to determine how the items loaded on dimensions and to determine if there were any items that should be removed.

9.4.2.1. Determining dimensions with items related to timeline, consequences, cure/control, illness coherence

All of the 32 items related to Timeline, Consequences, Cure/Control and Illness Coherence (see Table 45), were subjected to PCA using SPSS version 22. Prior to performing PCA, the suitability of data for factor analysis was assessed. Inspection of the correlation matrix revealed the presence of many coefficients of 0.3 and above. The Kaiser-Mayer-Olkin value was 0.79, exceeding the recommended value of 0.6 (Kaiser, 1974) and Bartlett’s Test of Sphericity (Bartlett, 1954) reached statistical significance (p< .001), supporting the factorability of the correlation matrix.

Table 45: Reference of cognitive representations item

<table>
<thead>
<tr>
<th>Item</th>
<th>Code Name</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. I believe I will stop getting pain soon</td>
<td>PainSoon</td>
</tr>
<tr>
<td>2. I believe I will keep having pain when I am an adult</td>
<td>PainAdult</td>
</tr>
<tr>
<td>3. When I get pain, it lasts a long time</td>
<td>PainLong</td>
</tr>
<tr>
<td>4. Over time I am getting pain more often</td>
<td>PainOften</td>
</tr>
<tr>
<td>5. When I get pain I think my pain will improve in (an hour, a day or less, a week or less, two weeks or less, more than two weeks)</td>
<td>PainImprove</td>
</tr>
<tr>
<td>6. When I get pain it makes me think my pain is (not serious at all, a bit serious, serious, somewhat serious, Is very serious, very serious)</td>
<td>PainSerious</td>
</tr>
<tr>
<td>7. I believe my pain affects what other people think of me</td>
<td>PainOthers</td>
</tr>
</tbody>
</table>
8. I believe having pain makes my family spend more money

9. I believe my pain makes hospital spend a lot of money

10. I believe my pain causes problems for my family

11. When I have pain, it stops me from taking part in activities such as PE

12. When I have pain, it affects me at school such as school work, school friends

13. When I have pain, it affects how I am at home

14. I can do a lot to control my pain

15. This is the amount of control I feel I have over my pain (no control, some control, quite a bit of control, a lot of control, full control)

16. There are things I can do to make my pain better

17. My behaviour can affect how much pain I have

18. There are things I do now which can affect future pain

19. I feel confused about why I get pain

20. I understand how my treatment for pain works

21. I understand my pain clearly

22. I don’t have any questions about my pain

23. When I have pain I understand what causes my pain

24. My pain changes everyday

25. My pain comes and goes

26. I can predict when I get pain

27. I can see a pattern in how and when I get pain

28. I am in control of my treatment for my pain

29. My treatment helps my pain get better

30. My treatment protects me from pain

31. I can continue with my activities because of my treatment

32. Taking my treatment means I have (no control over my pain, some control over my pain, quite a bit of control over my pain, a lot of control over my pain, full control over my pain)
Determining number of components – Analysis 1

PCA revealed the presence of eight components with eigenvalues exceeding 1, explaining a cumulative 73.5% of the variance. An inspection of the screeplot revealed a break after the third and again after fourth component. Using Catell’s (1966) scree test, it was decided to retain four components for further investigation. However, the results of Parallel Analysis (using the statistical program Monte Carlo PCA for Parallel Analysis developed by (Watkins, 2000), showed only three components with eigenvalues exceeding the corresponding criterion values for a randomly generated data matrix of the same size. According to the Parallel analysis as shown in Table 46, after the third component the criterion value from the Parallel analysis was larger than the actual eigenvalue from the PCA.

For a final judgement on how many components to keep, the component matrix from the PCA was consulted; this matrix showed that most items were loading on the first three components. However, there were more than three items on loading with factors above 0.3 on the fourth component. So the final decision, taking into account all the checks, was to keep four components rather than three components.

Table 46: Results from the parallel analysis with Monte Carlo PCA program

<table>
<thead>
<tr>
<th>Component number</th>
<th>Actual eigenvalue from PCA</th>
<th>Criterion value from Parallel analysis</th>
<th>Decision</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>11.351</td>
<td>2.4924</td>
<td>Accept</td>
</tr>
<tr>
<td>2</td>
<td>3.191</td>
<td>2.2579</td>
<td>Accept</td>
</tr>
<tr>
<td>3</td>
<td>2.347</td>
<td>2.0849</td>
<td>Accept</td>
</tr>
<tr>
<td>4</td>
<td>1.911</td>
<td>1.9507</td>
<td>Reject</td>
</tr>
<tr>
<td>5</td>
<td>1.345</td>
<td>1.8130</td>
<td>Reject</td>
</tr>
<tr>
<td>6</td>
<td>1.176</td>
<td>1.7098</td>
<td>Reject</td>
</tr>
<tr>
<td>7</td>
<td>1.111</td>
<td>1.6032</td>
<td>Reject</td>
</tr>
<tr>
<td>8</td>
<td>1.077</td>
<td>1.5045</td>
<td>Reject</td>
</tr>
</tbody>
</table>
The PCA was run again forcing four components. The four component solution explained a total of 58.8% of the variance with Component 1 contributing 35.5% and Component 2 contributing almost 10%, component 3 contributed 7.3% and finally Component 6 contributed almost 6%. To aid in the interpretation of these components, oblimin rotation was performed. The rotation solution revealed the presence of a simple structure (Thurstone, 1947), with four components showing a number of strong loadings. There were weak correlations between the components, with correlations under 0.5. The relationships between the components one and two (r = -0.306), one and three (r = -0.319), one and four (r = -0.022), two and three (r = 0.222), two and four (r = -0.137), three and four (r = -0.036), ranged from low (r = -0.036) to more strongly correlated (r = -0.319) so Oblimin Rotation for Factors Solution, Structure Matrix as well as the Pattern Matrix are reported in the Appendix 8.

**Rerunning PCA with removed item – Analysis 2**

The Item ‘My behaviour can affect how much pain I have’ (code: BehaviourPain) was not loading strongly on any factor, and had a low communality score. This led to rerunning the PCA without the item. This second analysis, had a Kaiser-Mayer-Olkin value of 0.80, still exceeding the recommended value of 0.6 (Kaiser, 1974) and Bartlett’s Test of Sphericity (Bartlett, 1954) reached statistical significance (p< .001), again supporting the factorability of the correlation matrix.

The PCA was run with four components again, and for this second analysis the four component solution explained a total of 60% of the variance. The first component contributed 36%, the second contributed almost 10%, and the third 7.5% with the last component contributed 6.2%. The correlations between the four components stayed quite similar and weak, one and two (r = -0.360), one and three (r = -0.314), one and four (r = 0.000), two and three (r = 0.235), two and four (r = -0.098), three and four (r = -0.031), so Oblimin Rotation for Factors Solution was used again and Structure Matrix as well as the Pattern Matrix are reported in Table 47.
The final 31 items loaded on four factors that resembled the original domains. The first column of the first component, was similar to the original consequences domain, which included items originally designed to assess beliefs about the impact of a health threat, but this domain in the PPQ-YP also included beliefs about the chronicity of pain (i.e. ‘I believe I will keep having pain when I am an adult’, ‘When I get pain, it lasts a long time’, ‘Over time I am getting pain more often’). The second component has items loading that were aiming to assess personal and treatment control, for the PPQ-YP, these items loaded strongly on the same component, suggesting that the items are assessing the same domain of control. The third component had all the items related to illness coherence loading strongly. The last component, the fourth included items from what previously was identified as cyclical, however, it also includes items about future pain and finding patterns in the pain episodes, and is more about the recurring nature of the pain condition, therefore this domain was renamed as recurrence.
Table 47: Principal component analysis of the PPQ-YP cognitive items

<table>
<thead>
<tr>
<th>Item</th>
<th>Pattern Coefficients</th>
<th>Structure Coefficients</th>
<th>Communalities</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>I</td>
<td>II</td>
<td>III</td>
</tr>
<tr>
<td>PainHome</td>
<td>.824</td>
<td>-.140</td>
<td>.008</td>
</tr>
<tr>
<td>PainLong</td>
<td>.805</td>
<td>-.097</td>
<td>-.181</td>
</tr>
<tr>
<td>PainOthers</td>
<td>.815</td>
<td>.339</td>
<td>.209</td>
</tr>
<tr>
<td>PainSchool</td>
<td>.768</td>
<td>-.222</td>
<td>-.002</td>
</tr>
<tr>
<td>PainActivities</td>
<td>.760</td>
<td>-.125</td>
<td>-.056</td>
</tr>
<tr>
<td>PainImprove</td>
<td>.750</td>
<td>-.284</td>
<td>-.356</td>
</tr>
<tr>
<td>PainOften</td>
<td>.760</td>
<td>-.080</td>
<td>.085</td>
</tr>
<tr>
<td>FamilyProblem</td>
<td>.744</td>
<td>-.011</td>
<td>.139</td>
</tr>
<tr>
<td>PainSerious</td>
<td>.694</td>
<td>-.187</td>
<td>-.247</td>
</tr>
<tr>
<td>FamilyMoney</td>
<td>.678</td>
<td>-.006</td>
<td>.223</td>
</tr>
<tr>
<td>HospitalMoney</td>
<td>.601</td>
<td>.084</td>
<td>.246</td>
</tr>
<tr>
<td>PainAdult</td>
<td>.445</td>
<td>-.130</td>
<td>.111</td>
</tr>
<tr>
<td>StopSoon</td>
<td>-.367</td>
<td>.201</td>
<td>-.259</td>
</tr>
<tr>
<td>TreatmentBetter</td>
<td>.086</td>
<td>.766</td>
<td>-.237</td>
</tr>
<tr>
<td>TreatmentProtect</td>
<td>.029</td>
<td>.726</td>
<td>-.047</td>
</tr>
<tr>
<td>TreatmentActivities</td>
<td>-.163</td>
<td>.717</td>
<td>-.104</td>
</tr>
<tr>
<td>CandoBetter</td>
<td>-.177</td>
<td>.657</td>
<td>-.056</td>
</tr>
<tr>
<td>ControlPain</td>
<td>-.197</td>
<td>.626</td>
<td>-.126</td>
</tr>
<tr>
<td>TreatAmountControl</td>
<td>-.171</td>
<td>.642</td>
<td>-.171</td>
</tr>
<tr>
<td>ControlofTreat</td>
<td>-.141</td>
<td>.629</td>
<td>.008</td>
</tr>
<tr>
<td>AmountControl</td>
<td>-.204</td>
<td>.616</td>
<td>-.220</td>
</tr>
<tr>
<td>UnderstandClearly</td>
<td>.059</td>
<td>.264</td>
<td>-.822</td>
</tr>
<tr>
<td>UnderstandCause</td>
<td>.038</td>
<td>.240</td>
<td>-.711</td>
</tr>
<tr>
<td>UnderstandTreat</td>
<td>.022</td>
<td>.185</td>
<td>-.683</td>
</tr>
<tr>
<td>QuestionsPain</td>
<td>.026</td>
<td>.117</td>
<td>-.651</td>
</tr>
<tr>
<td>WhyPain</td>
<td>.267</td>
<td>-.102</td>
<td>.630</td>
</tr>
<tr>
<td>PredictPain</td>
<td>.204</td>
<td>.261</td>
<td>-.017</td>
</tr>
<tr>
<td>PatternPain</td>
<td>.208</td>
<td>.164</td>
<td>.248</td>
</tr>
<tr>
<td>ComesandGoes</td>
<td>.158</td>
<td>.254</td>
<td>.365</td>
</tr>
<tr>
<td>ChangesEveryday</td>
<td>.379</td>
<td>.059</td>
<td>.287</td>
</tr>
<tr>
<td>FuturePain</td>
<td>.029</td>
<td>-.051</td>
<td>.098</td>
</tr>
</tbody>
</table>
9.4.2.2. Emotional representations subscales

Emotional representations are conceptually different to cognitive representations, and therefore a separate PCA was run with the items assessing emotional representations. The 14 items related to emotional representations were subjected to PCA (shown in Table 48). Inspection of the correlation matrix revealed the presence of many coefficients of 0.5 and above. The Kaiser-Mayer-Olkin value was 0.863, exceeding the recommended value of 0.6 (Kaiser, 1974) and Bartlett’s Test of Sphericity (Bartlett, 1954) reached statistical significance (p< .001), supporting the factorability of the correlation matrix.

Table 48: Reference of each emotional representations item

<table>
<thead>
<tr>
<th>Item</th>
<th>Code Name</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. When I am in pain I feel down and sad (how often)</td>
<td>DownSadOften</td>
</tr>
<tr>
<td>2. When I am in pain I feel down and sad (how much)</td>
<td>DownSadMuch</td>
</tr>
<tr>
<td>3. When I am in pain I feel upset (how often)</td>
<td>UpsetOften</td>
</tr>
<tr>
<td>4. When I am in pain I feel upset (how much)</td>
<td>UpsetMuch</td>
</tr>
<tr>
<td>5. When I am in pain I feel afraid (how often)</td>
<td>AfraidOften</td>
</tr>
<tr>
<td>6. When I am in pain I feel afraid (how much)</td>
<td>AfraidMuch</td>
</tr>
<tr>
<td>7. When I am in pain I feel angry (how often)</td>
<td>AngryOften</td>
</tr>
<tr>
<td>8. When I am in pain I feel angry (how much)</td>
<td>AngryMuch</td>
</tr>
<tr>
<td>9. When I am in pain I feel frustrated (how often)</td>
<td>FrustratedOften</td>
</tr>
<tr>
<td>10. When I am in pain I feel frustrated (how much)</td>
<td>FrustratedMuch</td>
</tr>
<tr>
<td>11. When I am in pain I feel worried (how often)</td>
<td>WorriedOften</td>
</tr>
<tr>
<td>12. When I am in pain I feel worried (how much)</td>
<td>WorriedMuch</td>
</tr>
<tr>
<td>13. When I am in pain I feel anxious (how often)</td>
<td>AnxiousOften</td>
</tr>
<tr>
<td>14. When I am in pain I feel anxious (how much)</td>
<td>AnxiousMuch</td>
</tr>
</tbody>
</table>
The PCA on the emotional representations items revealed the presence of two components with eigenvalues exceeding 1, explaining a cumulative 81.2% of the variance. A decision was made to continue with two components, as these would separate the emotions into emotions related to anger/sadness and emotions related to anxiety. The first component explained almost 72%, while the second component explained 9.5%. To aid in the interpretations of these components Oblimin Rotation was performed. The two components are strongly correlated ($r = -.723$) and this is because together these two factors do measure emotional representations. Oblimin Rotation for Factors Solution, Structure Matrix as well as the Pattern Matrix are reported in Table 49.

Table 49: Principal component analysis of the PPQ-YP emotional representations items

<table>
<thead>
<tr>
<th>Item</th>
<th>Pattern Coefficients</th>
<th>Structure Coefficients</th>
<th>Communalities</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>I</td>
<td>II</td>
<td>I</td>
</tr>
<tr>
<td>AngryMuch</td>
<td>1.008</td>
<td>.115</td>
<td>.925</td>
</tr>
<tr>
<td>FrustratedOften</td>
<td>.838</td>
<td>-.117</td>
<td>.923</td>
</tr>
<tr>
<td>UpsetOften</td>
<td>.764</td>
<td>-.189</td>
<td>.901</td>
</tr>
<tr>
<td>AngryOften</td>
<td>.977</td>
<td>.117</td>
<td>.893</td>
</tr>
<tr>
<td>FrustratedMuch</td>
<td>.917</td>
<td>.065</td>
<td>.870</td>
</tr>
<tr>
<td>UpsetMuch</td>
<td>.650</td>
<td>-.301</td>
<td>.868</td>
</tr>
<tr>
<td>DownSadMuch</td>
<td>.707</td>
<td>-.216</td>
<td>.863</td>
</tr>
<tr>
<td>DownSadOften</td>
<td>.761</td>
<td>-.124</td>
<td>.851</td>
</tr>
<tr>
<td>AfraidOften</td>
<td>.016</td>
<td>-.916</td>
<td>.678</td>
</tr>
<tr>
<td>AfraidMuch</td>
<td>-.063</td>
<td>-.967</td>
<td>.636</td>
</tr>
<tr>
<td>AnxiousOften</td>
<td>-.004</td>
<td>-.922</td>
<td>.663</td>
</tr>
<tr>
<td>WorriedOften</td>
<td>.060</td>
<td>-.878</td>
<td>.684</td>
</tr>
<tr>
<td>WorriedMuch</td>
<td>.034</td>
<td>-.863</td>
<td>.668</td>
</tr>
<tr>
<td>AnxiousMuch</td>
<td>.110</td>
<td>-.795</td>
<td>.684</td>
</tr>
</tbody>
</table>

The emotions of anger, frustration and down/sad were grouped in the first component. This component was considered to be encompassing more responsive emotions. While emotions related to anxiety and fear were grouped together in the
second component. Therefore, these emotions were considered to be defining as anticipatory emotions.

9.4.2.3. Identity subscale

The identity subscale had two components, symptoms associated with illness and symptoms associated with treatment, but the score is similar to the original questionnaire. Firstly the frequencies of the symptoms endorsed were calculated. As shown in Table 50 all the symptoms were endorsed, confirming the validity of the range of symptoms included in the PPQ-YP, although not all were associated with either pain or treatment. Joints stiff and joints sore were the most frequently endorsed symptom with 60.5%, and 48.7% respectively. “Can not Breathe well” was only endorsed by 7.9% and was not endorsed as associated with either pain or treatment.

Table 50: Identity subscale frequencies

Frequency of symptoms endorsed and frequency of symptoms associated with and those symptoms associated with treatment. n = 73.

<table>
<thead>
<tr>
<th>Freq of Symptom</th>
<th>Freq Associated with Pain</th>
<th>Freq Associated with Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Vomiting</td>
<td>25</td>
<td>46</td>
</tr>
<tr>
<td>Breathe Well</td>
<td>7</td>
<td>64</td>
</tr>
<tr>
<td>Weight Change</td>
<td>22</td>
<td>52</td>
</tr>
<tr>
<td>Tired</td>
<td>38</td>
<td>33</td>
</tr>
<tr>
<td>Joints Stiff</td>
<td>46</td>
<td>24</td>
</tr>
<tr>
<td>Joints Sore</td>
<td>37</td>
<td>34</td>
</tr>
<tr>
<td>Sore Eyes</td>
<td>17</td>
<td>55</td>
</tr>
<tr>
<td>Feeling Unwell</td>
<td>25</td>
<td>46</td>
</tr>
<tr>
<td>Headaches</td>
<td>26</td>
<td>45</td>
</tr>
<tr>
<td>Not sleep well</td>
<td>24</td>
<td>47</td>
</tr>
<tr>
<td>Upset tummy</td>
<td>20</td>
<td>51</td>
</tr>
<tr>
<td>Felt Dizzy</td>
<td>13</td>
<td>57</td>
</tr>
<tr>
<td>Felt Weak</td>
<td>21</td>
<td>50</td>
</tr>
<tr>
<td>Feel tightness</td>
<td>14</td>
<td>58</td>
</tr>
<tr>
<td>Change Mood</td>
<td>26</td>
<td>42</td>
</tr>
</tbody>
</table>
To test the validity of the identity subscale, a paired samples t-tests was conducted to test the difference between the experienced symptoms and the association with pain and then repeated with symptoms associated with treatment. This analysis showed there was a significant difference between the symptoms patients experience compared with the symptoms associated with pain (t(7.20), p<0.001) and with treatment (t(7.96), p<0.001). There was also a significant difference between symptoms associated with pain and those associated with treatment (t(3.728), p<0.001). These results support the fact that there are a conceptual differences between somatisation, association with pain and association with treatment.

9.4.2.4. Cause subscale

The cause subscale of the PPQ-YP was a new method of assessing causal beliefs. To validate this subscale, the frequency of a cause according to importance (endorsed 1, 2 or 3) and overall frequency of endorsement of cause regardless of rating was calculated. All of the causes were endorsed, mostly rated as a second or third cause. As shown in Table 51 the mostly frequently endorsed causes were immune (63%), genetics (34%), luck (40%) and doing too much (26%). There were a lot of missing in this subscale (42%) and this proportion of missing may indicate that there is a need to evaluate how this subscale is completed and if there is a need to make this subscale easier to complete. Psychological causes were the least endorsed causes, especially rated as the least important cause.
9.4.3. Objective 2: To assess the internal reliability of the PPQ-YP

Following the PCA to establish the subscales, the internal reliability of these subscales was tested by calculating the Cronbach’s alpha. Those items that are negatively related to other items such as stopsoon, whypain, comesandgoes and changeseveryday were reversed prior to the calculation of the internal consistency (Field, 2009). The alpha represents how they measure the same underlying construct. Values of 0.7 to 0.8 are considered acceptable for psychological constructs (Field, 2009). Table 52 shows that all subscales demonstrated good internal consistency with scores ranging from 0.585 to 0.962. The lowest internal consistency is found in the Recurrence subscale, and if the item futurepain is removed the internal consistency goes up to 0.618.

Table 51: Frequency of causal beliefs

<table>
<thead>
<tr>
<th>Cause 1</th>
<th>Cause 2</th>
<th>Cause 3</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stress or Worry</td>
<td>2</td>
<td>5</td>
<td>4</td>
</tr>
<tr>
<td>Genetics</td>
<td>14</td>
<td>8</td>
<td>4</td>
</tr>
<tr>
<td>A germ or virus</td>
<td>4</td>
<td>10</td>
<td>3</td>
</tr>
<tr>
<td>Diet</td>
<td>-</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>luck</td>
<td>9</td>
<td>9</td>
<td>13</td>
</tr>
<tr>
<td>Poor health</td>
<td>-</td>
<td>3</td>
<td>-</td>
</tr>
<tr>
<td>Pollution</td>
<td>-</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Behaviour</td>
<td>-</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Attitude</td>
<td>-</td>
<td>2</td>
<td>-</td>
</tr>
<tr>
<td>Family problems</td>
<td>1</td>
<td>3</td>
<td>-</td>
</tr>
<tr>
<td>Doing Too much</td>
<td>8</td>
<td>6</td>
<td>6</td>
</tr>
<tr>
<td>Feeling Down</td>
<td>-</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Getting Older</td>
<td>2</td>
<td>2</td>
<td>5</td>
</tr>
<tr>
<td>Accident</td>
<td>3</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>Type of Person</td>
<td>-</td>
<td>-</td>
<td>1</td>
</tr>
<tr>
<td>Immune</td>
<td>25</td>
<td>9</td>
<td>14</td>
</tr>
<tr>
<td>Other</td>
<td>3</td>
<td>-</td>
<td>1</td>
</tr>
<tr>
<td>Missing</td>
<td>5</td>
<td>11</td>
<td>16</td>
</tr>
</tbody>
</table>
Table 52: Internal consistency of the PPQ-YP Subscales

<table>
<thead>
<tr>
<th>Subscale</th>
<th>n</th>
<th>α</th>
</tr>
</thead>
<tbody>
<tr>
<td>Consequences</td>
<td>69</td>
<td>.934</td>
</tr>
<tr>
<td>Control</td>
<td>71</td>
<td>.901</td>
</tr>
<tr>
<td>Illness coherence</td>
<td>75</td>
<td>.860</td>
</tr>
<tr>
<td>Recurrence (timeline)</td>
<td>74</td>
<td>.585</td>
</tr>
<tr>
<td>Responsive (Emotional Representations)</td>
<td>71</td>
<td>.962</td>
</tr>
<tr>
<td>Anticipatory (Emotional Representations)</td>
<td>73</td>
<td>.957</td>
</tr>
</tbody>
</table>

9.4.4. Objective 3: To assess test-retest reliability of the PPQ-YP

After two weeks from initial completion of the PPQ-YP, adolescents who consented to be sent the questionnaire again were asked to complete the PPQ-YP as second time. Only four of the adolescents did not consent to complete the second PPQ-YP but out of the 72 adolescents who consented, 43 returned the second questionnaire. The mean age of these 43 participants was 14.28 years (SD 1.35), so the mean age was higher for the re-test reliability than the mean age of the initial 76 participants.

A two-week timeframe is a usual timeframe for test-retest reliability, and was chosen to avoid large changes in pain which may impact on the results, but also a two-week timeframe allowed enough time to pass to minimise answers being repeated through practice effects and recall (Fayers & Machin, 2007; Streiner & Norman, 2008). To account for changes in pain, a pain VAS was included at both time points (see Table 53), and while time two was correlated with time one ($r_s = .688, p < 0.001$) a Wilcoxon Signed-Ranks test indicated that time 2 was not statistically higher than time 1 ($Z = -1.1, p=0.274$), when the pain score at two-weeks was subtracted from the pain score at baseline, the median difference was 0 (IQR: -17.50 to 2), with the score differences ranging from -71 to 43.
Table 53: Test-retest reliability over two weeks

Interclass correlations, Spearman’s rho correlation, Wilcoxon test significance n = 43 to assess the test-retest reliability over two week period

<table>
<thead>
<tr>
<th>PPQ-YP Subscale</th>
<th>ICC</th>
<th>Spearman’s Rho Correlation</th>
<th>Wilcoxon test Significance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Consequences</td>
<td>.857</td>
<td>.708**</td>
<td>.701</td>
</tr>
<tr>
<td>Control</td>
<td>.736</td>
<td>.674**</td>
<td>.592</td>
</tr>
<tr>
<td>Coherence</td>
<td>.701</td>
<td>.545**</td>
<td>.604</td>
</tr>
<tr>
<td>Recurrence</td>
<td>.477</td>
<td>.369*</td>
<td>.971</td>
</tr>
<tr>
<td>Reactive</td>
<td>.820</td>
<td>.687**</td>
<td>.769</td>
</tr>
<tr>
<td>Apprehensive</td>
<td>.679</td>
<td>.649**</td>
<td>.343</td>
</tr>
<tr>
<td>Symptoms (Identity)</td>
<td>.835</td>
<td>.654**</td>
<td>.057</td>
</tr>
<tr>
<td>Pain Identity</td>
<td>.799</td>
<td>.602**</td>
<td>.005*</td>
</tr>
<tr>
<td>Treatment Identity</td>
<td>.757</td>
<td>.638**</td>
<td>.413</td>
</tr>
<tr>
<td>Pain VAS</td>
<td>.789</td>
<td>.688**</td>
<td>.274</td>
</tr>
</tbody>
</table>

Note: * p < 0.05, ** p < 0.001

The test-retest reliability was calculated through two recommended methods, first by calculating the Interclass Correlation (ICC) between the PPQ-YP subscales completed at the two time points. Test-retest reliability scores of 0.5 have been described as a reasonable minimum (Streiner & Norman, 2008). The ICC scores ranged from 0.477 to 0.857, this range of scores showed reasonable test-retest reliability. The second method was to test if there was significance difference between the two time points and this was assessed through related-samples Wilcoxon test. There were no significant differences between subscales total scores at each time point except for the scores on the symptoms associated with pain (pain identity subscale where Z = -2.79, p=0.005; See Table 53). Spearman’s Rho correlations between the PPQ-YP at each time point was included and found that there was good stability over this period with most of the correlations being greater than 0.5 ranging from .369 to .708 (See Table 53). Only one subscale was less than 0.5 and that was the recurrence subscale.
9.5. Summary

The previous results reported in Chapter 5 led to the development of the Pain Perceptions Questionnaire for young people with JIA (PPQ-YP). The development of the PPQ-YP was divided into two phases, the pre-testing and the field-testing phases. In the first phase, the pre-testing of the PPQ-YP, adolescents without JIA completed the questionnaire and provided feedback regarding the language and comprehension of the PPQ-YP. Modifications to the PPQ-YP were made according to the results of the pre-testing phase. The second version of the PPQ-YP was piloted with adolescents with JIA to test the validity and reliability of the PPQ-YP in the field-testing phase.

To test the psychometric properties in the field-testing phase, the factor structure of the PPQ-YP was tested by using Principal Component Analysis (PCA) on items related to timeline, consequences, personal/treatment control, illness coherence and a second PCA was conducted with items related to emotional representations. In total, there are eight domains in the PPQ-YP. These eight domains or sub-scales had internal reliability, Cronbach alpha that ranged from 0.585 to 0.962. The PPQ-YP had test re-test reliability, ICC ranges from 0.477 to 0.857, and the two time points are correlated, Spearman’s rho was ranging from 0.369 to 0.708 with no significant differences after two weeks except for the pain identity subscale ($Z = -2.79$, $p=0.005$).
Chapter 10: Discussion

10.1. Overview

This chapter discusses the overall contribution of this thesis to understanding adolescents’ conceptualisation of illness. The main findings of the current work are reported, followed by an evaluation of how the research question and objectives of the thesis were addressed. The three key findings of this thesis are discussed separately. First the observation of the shift in what constitutes a health threat for adolescents from illness perceptions to symptom perceptions; second that for adolescents the emotional representations appear to be strong drivers of behaviour, and finally the value of having a ‘normal’ identity. After the discussion of the three key findings of this thesis, the subsequent section describes the strengths and limitations. This section includes a consideration of the thesis as a whole and provides discussions relating to each study. This section also addresses issues relating to the methodological and the theoretical approaches, followed by consideration of the generalisability of the findings. Finally, the implications of the results for the field of pain beliefs research and for clinical practice are discussed with additional suggestions for the future direction of the research.

10.2. Summary of main findings

The literature review (see Chapter 1) of research into how adolescents conceptualise illness found that there is a significant theoretical gap accounting for how experiences and knowledge are converted into an understanding of a long-term condition (LTC).

The ways in which adults conceptualise illness or a health threat, have been researched within the theoretical framework of Leventhal’s Common Sense Self-
regulatory Model (CS-SRM). The systematic review identified 26 studies of adolescents (aged between 11 to 16 years) which used the CS-SRM framework. The review found that none of these studies investigated the application of the framework or the assessment of the illness perceptions to this age group (Chapter 2). Furthermore, with only five manuscripts in the review that utilise the Revised Illness Perceptions Questionnaire (Moss-Morris et al., 2002), it is difficult to justify the use of the IPQ-R with this age group. No study had formally evaluated the use of the IPQ-R with adolescents. Therefore, the primary aim of this thesis was to investigate whether the CS-SRM can be applied with adolescents to understand how they conceptualise an LTC such as Juvenile Idiopathic Arthritis (JIA). The secondary aim was to identify the best ways to systematically assess illness perceptions of adolescents with an LTC.

10.2.1. Addressing the research question and objectives

To address the overarching research question, the three components of the CS-SRM were investigated in turn to identify the degree to which they can account for adolescents’ experience of an LTC. Table 54 summarises the findings according to each objective to address the overarching research question. The first component of the CS-SRM addresses the mental representation of the condition (JIA). The first two objectives were to evaluate whether domains identified in the CS-SRM and assessed by the IPQ-R were relevant to the adolescents. The second component of the CS-SRM addresses how the mental representations informed the goals and function of illness behaviours. Objectives three and four mapped to this second component of the CS-SRM. Objectives one to four of this thesis were addressed by a qualitative study and analyses (framework and content analyses). Finally, the last component of the CS-SRM is the self-regulation of cognitions, emotions (both make up the mental representations) and behaviours by appraisal and evaluation of cognitions, emotions and behaviours. To evaluate this component, the fifth objective was to investigate the relationship between the mental representation and the behaviour using a quantitative approach.
The qualitative findings reported in Chapters 5 and 6 relating to objectives one to four (See Table 54 below), led to modifications proposed to the CS-SRM for adolescents. The proposed modified model is described in Chapter 8. Modifications to the IPQ-R are also recommended, the main recommendation is shifting from the assessment of perceptions about JIA to the assessment of pain perceptions. Therefore, objectives six and seven of this thesis were to develop and validate the Pain Perceptions Questionnaire for Young People (PPQ-YP; Chapter 9).

Table 54: Summary of Findings

Each of the objectives from the aims presented in Chapter 3 with a summary of the findings and which Chapter the results are presented.

**Research Question:** Can the Common Sense Self-Regulatory Model be applied to adolescents’ conceptualisation of illness? In this instance, Juvenile Idiopathic Arthritis (JIA).

<table>
<thead>
<tr>
<th>Objectives</th>
<th>Summary of Findings</th>
<th>Chapter</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Identify whether or not the domains of emotional and mental representations identified in the CS-SRM are relevant to adolescents with JIA.</td>
<td>• The domains proposed were relevant but there were some disease specific modifications required (such as taking into account flare-ups and the episodic nature of the chronic condition). o Experience and not age determined their level of understanding, for example those who were too young to remember pre-JIA, were unclear about the cause of the condition.</td>
<td>5</td>
</tr>
<tr>
<td>2 Assess the suitability of the current measure of illness representations for adolescents with JIA.</td>
<td>• The current measure was not suitable for the adolescents: o There were methodological issues such as negative wording/items.</td>
<td>5</td>
</tr>
<tr>
<td>3 Identify adolescents’ goals and the coping strategies they employ to deal with pain related to JIA to determine the relationship between intention and behaviour.</td>
<td>• Behaviour $\rightarrow$ intention: maintain normality vs. Behaviour $\rightarrow$ intention: focus on pain. o There was an overarching goal to be socially identified as ‘normal’ when compared to their peers. To achieve this, the function of coping behaviours was to either: o Cognitively adjust the meaning of normality (perceived pain as a challenge). o To focus on overcoming pain to get back to normal (perceived pain as a threat).</td>
<td>6</td>
</tr>
</tbody>
</table>
4 Identify the emotional and cognitive profiles associated with specific coping goals.

- There were no differences in the cognitive profiles of the two groups.
- The pain representations of ‘Challenge’ and ‘Threat’ stemmed from the emotional representations. This was where the adolescents differed.

5 Identifying the predictors of behaviour.

- Associations between emotional representations, pain and physical behaviour get stronger over time and have strong relationships cross-sectionally.
- Emotional representations and self-reported pain intensity are the best predictors of future physical behaviour (a year later).
- Pain mediating the relationship between emotional representations and physical behaviour was a robust model.
- The relationship between emotional representations and physical behaviour two years later was strongly influenced by levels of pain.

6 Developing an adolescent specific measure of illness representations.

- Modified the original questionnaire to develop the Pain Perceptions Questionnaire for Young People (PPQ-YP)
- Modified the items, verbal anchor labels, and response formats based on the results from the cognitive interviews.

7 Testing and evaluating the psychometric properties of the new measure.

- The PPQ-YP assesses the following 8 domains:
  - Consequences
  - Control
  - Coherence
  - Recurrence
  - Reactive
  - Apprehensive
  - Pain Identity
  - Treatment Identity

Sections 10.3 to 10.5 present the conclusions and interpretations of the findings which contributed to the modifications of the CS-SRM to adolescents with an LTC. Three significant modifications are proposed: a shift in emphasis from illness perceptions to symptom perceptions (Section 10.3), emotional representations as
more influential in driving behaviours (Section 10.4), and incorporating the value of a normal self-concept (Section 10.5).

10.3. Shift from illness perceptions to symptom perceptions

The results presented in this thesis provide justification for a shift in the focus from illness to symptom perceptions as key drivers of behaviour in adolescents with JIA. There are potentially three arguments supporting this. The first argument is that the recurring nature of the condition and the experiences of unpredictable flare-ups mean that adolescents focus on the immediate health threat, in this context it is pain. The second argument relates to developmental immaturity affecting the understanding of the complexity of having an LTC. In this case the adolescents’ tendency to focus on symptoms reflects a stage of cognitive ability. The third argument is that this shift reflects a limitation of the CS-SRM theory and the associated assessment of a ‘health threat.’ Once a conceptualisation of an LTC is established, the focus moves from the LTC to the symptoms. The established schemata for the underlying condition do not change as frequently, but the schemata for the symptoms would change more frequently.

Argument 1: shift from illness to symptoms is due to the condition

The first argument is that this shift from illness to symptom is due to the relapsing and remitting nature of the condition. The condition itself is complex, with different treatment routes, and with an unknown disease prognosis. In the 1980s children and adolescents with JIA participating in a qualitative study defined arthritis as pain in the joints (Beales et al., 1983b), demonstrating that for them pain was not simply a symptom of JIA but an integral part of the illness. More recently a synthesis of 27 qualitative studies, Tong and colleagues (2012) concluded that the unpredictable nature, the invisibility and subjectivity of pain was driving the children and adolescents’ experiences of their arthritis. The current results from this thesis would
suggest that the relationship between pain and the understanding of arthritis is far more integrated.

**Argument 2: shift from illness to symptoms is due to the cognitive abilities of adolescents**

A second argument accounting for this shift from illness to symptoms is based upon the cognitive abilities of adolescents. Understanding and making sense of living with an LTC requires abstract constructs which makes it a complex task for adolescents. As argued in Chapter 8, one method of dealing with abstract information and experience is to anchor the abstractions into concrete experience (Quinn & Eimas, 1997; Leventhal et al., 2012).

According to the Piagetian approach, adolescents are still developing the ability to move from concrete to abstract thinking. However, the evidence about adolescents’ abilities to conceptualise abstract constructs of illness is mixed. For example, research with a pain perspectives inventory found that there was a move from concrete to more abstract conceptualisation of pain (Crow, 1997; Esteve & Marquina-Aponte, 2012). While, other studies with found no Piagetian developmental trends or age-related trends (Ross & Ross, 1984; Gaffney & Dunne, 1986). The current work in this thesis provides evidence that adolescents are dealing with abstract concepts by anchoring into their concrete experience. In the case of the adolescents with JIA, to deal with the abstract information of JIA, they are anchoring the abstractions in their everyday pain experiences. This argument is supported by the work by Paterson, Moss-Morris and Butler (1999) and Crisp (1996) that found that experiences and not age contributed to the conceptualisation of illness.

**Argument 3: shift in the definition of health threat**

The shift from illness perceptions to symptoms perceptions may reflect a limitation of the theory and in the assessment of health beliefs related to an LTC. The theory was developed to account for the conceptualisation of acute health threats. The shift between the two perceptions could result from what would be considered a health threat for an individual with an LTC, which may differ to those people without an
LTC. At the onset of an LTC, the illness and its associated symptoms would be perceived as health threats. However, over time the LTC would become a normalised experience, as described in Chapter 8 (Kahneman & Miller, 1986). Theoretically, over time a mental representation is established until an individual no longer experiences deviations or acquires new information from the LTC; therefore, the need to make sense of a health threat is longer required (Leventhal et al., 2011). The shift from illness to symptom perceptions signals a narrowing of an individuals’ focus, moving from the overarching label to the more immediate health threat of the symptoms. In the current work, it is possible that the adolescents shifted their focus from JIA to a more immediate health threat; the associated pain. This argument would be relevant to work with adult cohorts as well as adolescents.

10.4. Emotional representations – drivers of behaviour

One of the key findings of this thesis was the increased emphasis on the role of emotional representations of JIA in influencing the responses of adolescents, particularly the levels of physical activity. The qualitative findings in Chapter 6 support the hypothesis that the emotional pathway is important in distinguishing between adolescents who engage in different procedures, including those who continue with physical activity (adolescents who focused on maintaining a newly defined normal) and those who would stop activity (adolescents who focused on managing their pain).

Findings from the qualitative coping study (Chapter 6) showed that the adolescents who normalised their emotional response (i.e. reported feeling fine, normal and happy) still took part in their activities; the activities were reduced but not stopped. Unlike those adolescents who had negative emotional responses who focused on managing their pain and stopped their activities. This finding about adolescents with negative emotional responses has been supported by previous research where Schanberg and colleagues (2005) found that adolescents with JIA stopped activities at school, and also reported higher pain intensity when experiencing high levels of
negative emotions. This relationship may have been influenced by pain or else mediated by pain, and this relationship with pain was explored in Chapter 7.

Connelly and colleagues (2012), found through an electronic diary study with children and adolescents (8 to 17 years old) with JIA, that adolescents with variability in positive emotions had higher reported pain. However, only variable negative emotions were related to activity limitations, not positive emotions (Connelly et al., 2012). While this electronic diary study suggests that stability of emotions is important to measure (Connelly et al., 2012) emotions and their variability are not currently measured. The items about emotional representations in the PPQ-YP can measure the variability of negative emotions.

The work reported in this thesis explored the relationships between pain, emotions and behaviour (physical activity). This work provided evidence that emotional representations at baseline predicted pain and physical activity at a later time point. This finding indicated that there may be both a direct and an indirect pathway in how emotions, pain and behaviours work at a cross-sectional time point and in longitudinal relationships where pain mediates emotional representations and physical activity.

The results in this current study have added to the literature included in the review in Chapter 2 which suggested that emotional representation is an important pathway in predicting clinical outcomes (Chong et al., 2012; Chong et al., 2013; Fortenberry et al., 2014; Jones et al., 2014; Hughes et al., 2012; Gray & Rutter, 2007; Munson et al., 2009; Munson et al., 2010). Chapter 7 reported findings that negative emotions predicted higher reported pain as well as lower physical activity.

The identification of a relationship between emotional distress and pain in adolescents is not novel. Previous work with 80 adolescents with chronic pain (aged between 11 and 17 years) found that these adolescents report high anxiety, with 70% of them reporting depression (Eccleston et al., 2004). Anxiety was predicted by pain duration, coping strategies, specifically internalizing/catastrophizing, and by a
decrease of seeking social support. Internalising/catastrophizing, seeking social support and behavioural distraction predicted depression (Eccleston et al., 2004). Coping strategies are not the only factors associated with negative emotions. Children with unsophisticated conceptualisation of their JIA have higher emotional reactions and fear (Beales et al., 1983a).

Self-regulation of emotions is a key dimension of the CS-SRM (Leventhal et al., 2003). Emotion regulation interventions are designed to address the self-regulation of emotions to promote wellbeing and adjustment (Cameron & Jago, 2008). The emotional pathway regulates anxiety and fear, and this pathway is also a motivational system that has reciprocal relationships with cognitions (Cameron, 2003; Cameron & Jago, 2008). This investigation of a reciprocal relationship is important because an intervention or emotional regulation can influence the course of the illness, and efforts to manage the illness can influence emotions (Cameron & Jago, 2008). Reid, Gilbert and McGrath (1998) found that the function of emotion-focused avoidance coping is to deal with negative emotions by avoidance behaviour. The authors found that this type of emotion-focused avoidance coping was reported to be higher in older adolescents aged between 13 to 18 years compared to children aged 8 to 12 years. Furthermore, those adolescents who did cope through emotion-focused avoidance also reported high pain intensity and longer pain duration. This suggested that the relationship between pain and emotions is particularly strong in adolescence and justifies an increased focus on the role of emotional representations in adolescents.

10.5. The value of “normal”

Previous research has provided evidence that the meaning of pain is pivotal in understanding reported pain intensity (Leventhal, 1993). Findings from the current work demonstrate that the meaning adolescents attached to their pain was linked to their evaluation and development of their self-concept. The emphasis was on preserving a sense of being or feeling ‘normal’. There were two types of pain
representations; either that pain is a threat or pain is a challenge to achieving that goal.

Pain perceptions were thus influenced by the extent to which they viewed having JIA as a threat to a positive sense of a normal identity. Adolescents either developed a coherent sense of self, where JIA is just one aspect of their lives, or they develop alternative accounts of themselves whereby they contrast an identity with JIA with an identity without JIA. In other conditions it has been found that the degree to which individuals incorporate their illness into their sense of self predicts outcomes or severity. A study of adolescents with uncomplicated epilepsy found that a higher sense of coherence led to lower self-reported illness severity (Räty et al., 2004). Similar results were found in a study of adolescents with T1D. Those who integrated having diabetes with their sense of self (higher sense of coherence) had higher self-esteem. They also reported better self-management and fewer emotional and social problems (Luyckx et al., 2014b).

In the current study, the group of adolescents who perceived pain as a challenge (focused on maintaining normal) associated the meaning of pain with a normal occurrence and integrated the chronic condition in their self-concept. This group also reported less pain severity and higher function. In comparison, the group of adolescents who perceived pain as a threat (focused on managing pain) associated their pain with episodes of flare-ups. This association led to the adolescents in the management category to treat and perceive their condition as a series of separate episodes of acute pain which triggered heightened emotional and cognitive threat responses rather than a recurrence of an underlying chronic pain condition which generated adaptive coping responses. This can be mapped back to differences in the underlying beliefs about the nature of the JIA, where some adolescents perceive JIA as just one relapsing remitting condition whereas, other adolescents experience re-occurrences of the condition. Consequently, these adolescents had two conflicting identities and had higher emotional distress as well as reporting higher pain severity and lower function. Therefore, for these adolescents they had a lower sense of coherence and possibly lower self-worth. Associations between self-perceptions and
pain and function have been reported previously (Guite et al., 2007) where higher pain was reported in those adolescents with lower self-worth. However, further evidence of differences in these underlying beliefs is required.

Findings from the current study have highlighted the value that adolescents place on feeling ‘normal’. It is the underlying motivator of some coping behaviours (Chapter 6). Striving for normality is a theme identified in the synthesis of the qualitative analysis of the experiences of children and adolescents in JIA (Tong et al., 2012). Tong and colleagues (2012) defined the theme striving for normality as the attitudes and behaviours which adolescents intended to use to deal with feeling different, and this was either by preserving social identity, by being resourcefulness, by finding a sense of community having a focus on remission or lastly gaining a mastery over body. Results in Chapter 6 provided a framework for understanding how adolescents attempted to achieve a sense of normality and this is the implicit and explicit goal of coping behaviours. The coping framework developed in Chapter 6 could be the basis for future research and the modified model presented in Chapter 8 theorises how striving for normality is a motivational pathway for behaviour and behaviour change.

10.6. Strengths and limitations

The following sections outline the strengths and limitations of the thesis, addressing the methodological considerations, the theoretical approach, and the generalisability of the work. The strengths and limitations of each of the studies are considered under separate headings.

10.6.1. Methodological considerations

One of the main strengths of this work was the use of mixed methods. Mixed methods can complement and balance the methodological strengths and weaknesses of each approach. New insights can be derived from the synthesis of qualitative and quantitative work. The research paradigm applied in this work meant the quantitative work was not only underpinned by existing theory but also informed by new insights.
and theoretical modifications derived from the qualitative work. This research paradigm was deliberately chosen as an approach to cross-validate both the qualitative and quantitative findings (Creswell, 2003). For example, the model tested in Chapter 7 was informed by the qualitative work (in Chapter 6) that explored the relationships between mental representations and coping procedures. Similarly, the questionnaire development and validation (Chapter 9) was directly informed by the qualitative results in Chapter 5. However, there are strengths and limitations for each methodological consideration as discussed below.

Cognitive interviewing is a useful method because it reveals the cognitive processes that an individual utilises when answering a questionnaire (Willis, 2004; Willis et al., 1991). This interview approach of thinking aloud was derived from psychological procedures to map the mental processes and the indirect mapping of the stages. The strengths of utilising both sub-types of cognitive methods, think-aloud and verbal probing, reduces the cognitive burden of the interview on the interviewee. Using both methods is also recommended with adolescents as they require more extensive probing than adults (Strussman et al., 1993; Zukerberg & Hess, 1996). Using concurrent protocols of the think-aloud approach and verbal probing counteracted this limitation of relying on the respondents’ cognitive processes. The use of both protocols is the same approach employed and validated by previous work with adolescents (Zukerberg & Hess, 1996; Willis, 2004).

However, a limitation of the use of cognitive interviewing may be the dependency on the abilities of the respondent; in particular verbalization is dependent upon problem-solving, language, and comprehension (Czaja, 1998; Taylor, 2000; Schuwirth et al., 2001). In the earlier work with these participants, Ghio (2011) found all but two participants’ verbal ability and their scores were within the normal range.

Another potential problem arising in cognitive interviewing is specific to the think-aloud approach, namely the artificial nature of the task (Drennan, 2003). This may be a data collection limitation. Conversely, one of the strengths of this thesis is that the same participants completed the two interviews; the data collected from each
interview corresponded with each other. Specifically, the same participants were interviewed about emotions in both the IMS interviews (about pain reported in Chapter 6) and about emotional representations of their JIA (reported in Chapter 5). The emotional response corresponded with the data from the emotional representations in the cognitive interviews. This correspondence of the data increases the confidence in the data collected through the cognitive interviewing.

To assess the coping procedures, a framework approach to the data was considered appropriate and led to the development of a framework to elicit adolescents’ coping behaviours and goals. Throughout the development of the framework there was a need for reflexivity and transparency. Therefore, the findings and the coping framework were grounded in the data as well as developed through the theoretical mapping of psychological theories and the consultation of the research team.

The coping framework developed for this thesis relied on cross-sectional data and it is difficult to infer if the identified groups (maintaining normality versus focused on pain) were interchangeable. As the interviews only captured the coping behaviours related to one pain episode, it was difficult to determine how typical the behaviour was in terms of the adolescents’ usual responses to pain episodes. Further work is required to identify if these behaviours are related more to the context of the pain than the pain episode, and if their coping goals (maintaining normality versus focusing on pain) stay consistent. The data reported in Chapter 6, regarding if adolescents sought social support by telling someone about their pain, identified that communication about pain was situation dependent (school or home) and the emotional cost was considered (if they were going to be believed or treated differently).

One of the strengths of Chapter 6 was that the method of analysis resulted in clearly identified differences between the two groups, those who strove to maintain normality and those who focused on pain. The adolescents who coped by focusing on pain management had negative emotional representations, higher self-reported pain intensity, and higher scores of function measured by the CHAQ. Due to the cross-
sectional and qualitative nature of the study it was difficult to infer whether members of the group focussing on managing their pain and engaging in behaviours such as stopping activities were doing so because of their emotional representations and remembering the impact of pain having previously experienced more pain. It may also be because they were experiencing more pain, leading to more negative emotional representations and then being more likely to stop activities. This subsequently became a question of which came first; the pain or the emotional representations, which then influenced the behaviour. This limitation was addressed by the longitudinal modelling reported in Chapter 7, which found that negative emotional representations lead to reporting higher pain intensity and lower physical activity. This link between the qualitative study and quantitative study was one of the strengths of the mediation analysis.

The statistical approach implemented in Chapter 7 had separate limitations. First, statistical power was an issue which meant that Structural Equation Modelling (SEM) could not be used. SEM would have allowed an investigation all the variables which measured each concept by loading them onto latent variables, which would have given a more accurate estimate of measurement error (Hayes, 2009). The analysis in Chapter 7, failed to take into account the measurement errors. Furthermore, SEM would have allowed a test of the multiple dependent variables simultaneously, however, the current study was limited to small numbers such that the mediation analysis had to be completed in separate steps. The strength of the longitudinal design was the greater confidence in identifying relationships since the predictor variables preceded the modelled outcome variables in time (Kenny, 1979).

A strength of the results in Chapter 9 was that the items developed for the PPQ-YP, were informed by the underlying theory and the subsequent qualitative analysis of the adolescents’ experiences and their understanding of their illness. To validate these items, the questionnaire was sent to the adolescents with JIA who were already taking part in CAPS. However, with only a 37% response rate there may be sample bias, mostly with female participants. The small numbers may be due to the recruitment process involved in postal questionnaires. This low response rate and
small sample size resulted in 2-3 participants per item for the first PCA and 5-6 participants per item for the second PCA for emotional representations. The emotional representations PCA met the minimum requirements for a PCA, but the first PCA did not. Therefore, it was unclear if those items that did not load on the factor, such as the item ‘There are things I do now which can affect future pain’ would load more strongly with more power.

10.6.2. Theoretical approach

The aim of this thesis was to assess the CS-SRM, and the links to cognitive psychology literature (Leventhal et al., 2011) for adolescents with JIA. Two strengths were a) the use of the same adolescents to map the underlying mental representations from coping procedures and goals allowed an exploration of the interaction between the components of the CS-SRM and b) the use of a longitudinal dataset.

The work presented in this thesis aimed to investigate the underlying reasons and thought processes elicited from the items of the IPQ-R. The focus of this thesis was on the underlying internal processes. Illness schemata are developed from three sources of information, one of which is the social contexts or authoritative sources (Leventhal et al., 1980; Leventhal et al., 1984). This source of information has not been explored within the scope of this thesis. Therefore, the thesis had limited discussion of how these processes interacted in social contexts, and the influence of the family and society (Cameron, 2003).

Most studies which adapt a CS-SRM framework have a common weakness; providing only a snapshot of the variables at specific time points (Leventhal et al., 2012). In the current study yearly time points were used. The cross-sectional relationships between the variables in Chapter 7 were strongly associated, which may explain how the inclusion of physical behaviour at year 1 in the longitudinal mediation model reduced the significance of pain at year 1 for predicting physical
behaviour at year 2. The yearly time points may be too long in duration to distinguish the process underlying these relationships between emotional representations, self-reported pain and physical behaviour as observed by the parents.

10.6.3. Generalisability

Testing the CS-SRM with every condition that occurs during adolescence was out of the scope of this thesis, the investigation of each component of the CS-SRM has facilitated the development of a recommended conceptual model for the CS-SRM for adolescents. This conceptual model has the potential to be applied to other adolescent cohorts. The work in this thesis has limitations with regards to the generalisability of the results. Considering the applicability of the CS-SRM to adolescents, the focus on JIA may be perceived as a limitation. The investigation of the applicability of the model to one condition may not be generalisable to adolescents with other LTCs.

The collection of self-report measures has its own limitations in providing generalisable results to other adolescents. There is an argument that the use of self-report measures in questionnaires generates new cognitions and emotions rather than eliciting the existing state (Ogden, 2003). This self-report nature may also elicit responses that are socially desirable (Logan et al., 2008a) not just new cognitions or emotions. Not all the parents left their child during the interviews, but the parents were provided with their own task to complete. This was considered throughout the use of the data, which explains why the outcome variable was completed by the parents and the parents’ mental state was taken into account for the longitudinal modelling in Chapter 7. The outcome variable was also included as a possible confounder. As mentioned previously, the overlap in the two tasks of the interviews, both the cognitive interviewing and pain interviews of emotional response and representations, were mapped. Due to the random order of the tasks, this method validated that the emotions were elicited. Additionally, the test-retest reliability in Chapter 9 for the PPQ-YP suggested that the beliefs they were holding were
consistent, which further suggests that despite the data being collected through self-report, it was eliciting the existing states.

10.7. Implications

The results from this thesis have implications for the CS-SRM theory (summarised in Chapter 8) as well as implications for future research into adolescents’ pain beliefs and for clinical practice. The following sections outline the implications of the work for each of these areas. Finally recommendations and suggestions for future direction are presented in section 10.8.

10.7.1. Adolescents’ pain beliefs research

The literature review in Chapter 2 did not find any manuscripts that included the assessment of adolescents’ pain beliefs within a CS-SRM framework. Therefore, the key finding of a shift from illness perceptions to pain perceptions, represents a novel result in this research area. The different models of pain presented in Chapter 1 all suggest that cognitions play a role in the pain experience. Despite the importance of cognitions in a biopsychosocial approach to pain, there are only two limited tools available to measure pain perceptions in adolescents.

Currently, the most popular tool in measuring pain perceptions is the Pain Catastrophizing Scale for children (PCS-C; Crombez et al., 2003). The PCS-C has three constructs: rumination, magnification and helplessness. These three constructs are associated with displaying communicative pain behaviours when there are observers for the behaviours (Sullivan et al., 2004). Pain catastrophizing is part of the fear-avoidance model where it feeds into pain-related fear and increases behaviours of avoidance and hypervigilance in a vicious cycle (Vlaeyen & Linton, 2000). Pain catastrophizing is a cognitive process in that how an individual perceives pain underlies fear beliefs. However, the original work with the CS-SRM found that fear
alone cannot motivate behaviour or behaviour changes (Leventhal et al., 1980), and motivation of illness behaviours is dependent on the conceptualisation of the health threat and developing action plans (Leventhal et al., 2012). These include key attributes about the nature of the threat such as controllability, chronicity etc. Therefore, the PCS-C is very limited in the role of developing behaviour change interventions while also limited in the range of beliefs that does motivate behaviour. The PPQ-YP provides an assessment of the conceptualisation of pain and a measure of a range of theory-driven beliefs, an assessment that is missing in the PCS-C. Overall, the aim of the PPQ-YP is to provide an approach to assessing pain cognitions as well as a variable emotional representation; an area that is important for the psychological interventions for adolescents with chronic pain.

The fear-avoidance model (Vlaeyen & Linton, 2000) for chronic pain has provided an underlying mechanism for the role of fear and avoidance in perceiving a pain experience. However, there are limitations with this approach. The applicability of the model and the associated assessment the PCS-C (Crombez et al., 2003) to adolescents with pain related to LTCs has not been tested systematically. The fear-avoidance approach does not take into consideration the role of how young people, both children and adolescents are making sense of information or evaluating the contextual information of their pain experiences. The assumption of the fear-avoidance model is that if the injury and perceived pain experience is associated with no fear then the individual will experience a recovery after confronting the injury/pain. Such a framework does not allow a study of the more complex cognitions adolescents have when “confronting” a pain experience and cannot account for the findings in this thesis. The findings of the current work have shown that the perception of a pain experience is complex and does involve a consideration of previous experiences, previous knowledge, and is situational dependent with attention towards the emotional and social impact.

One of the main limitations of the PCS-C (Crombez et al., 2003) is the move from adults to adolescents without considering if the items are capturing the cognitive processes of an adolescent. Similarly, the Survey of Pain Attitudes for Pediatrics
(Ped-SOPA; Engel et al., 2012) was developed by modifying the measure of items developed for adults with chronic pain (Turner et al., 2000; Jensen et al., 2007). The 37 items of the PED-SOPA were chosen by the authors and parents of young people with physical disabilities but not the young people themselves and neither content validity or face validity were assessed (Engel et al., 2012; Miró et al., 2014). In contrast, the PPQ-YP underwent a number of processes which included adolescents in all phases. This method of questionnaire development brought to light the importance of consulting adolescents. The findings of this thesis highlighted important conceptual and methodological differences in items used with adults and those used with adolescents. Thus, the PPQ-YP provides researchers and clinicians a tool which is appropriate for adolescents that can now be utilised as targets for interventions as discussed below.

One of the main findings of this thesis is the motivational role of emotions in driving behaviours. Emotions are not measured by the PCS-C and the PED-SOPA. Rather, the PED-SOPA includes items measuring emotions as a cause of pain such as bad feelings can make my pain worse and being scared makes me hurt more (Engel et al., 2012). The results from the adolescents participating in the current study suggest that they did not consider emotions as a cause of pain (Chapter 5 and 9). The authors of the PED-SOPA did not take steps to determine any difficulties adolescents may have in conceptualising the links between psychological and physical systems; conceptualising psychoneuroimmunological links are likely to involve more complex cognitive processing. For this reason the PPQ-YP measures the frequency and intensity of emotions. Changes in either of these (emotional variability), demonstrates the impact pain has on the adolescents’ lives. Importantly, the emotional representation subscales of the PPQ-YP can differentiate between those adolescents who are experiencing a responsive emotional representation and those who are experiencing an anticipatory emotional representation. Furthermore, the emotional representation subscales can provide an indication of the interventions, as mentioned in the next section, have been successful, as the subscales would track the amount and frequency of an emotion over multiple completion of the PPQ-YP.
10.7.2. Clinical practice

The results of this thesis demonstrate the importance of incorporating illness representations in the management of adolescents with chronic pain conditions. The two key areas that this should influence are assessment and communication of adolescents’ understanding of their condition within routine clinical consultations and second the requirement psychological treatments for adolescents coping with LTCs. As outlined in Chapter 8, communication in clinic may have a bidirectional relationship with adolescents’ conceptualisation of a condition. An influential study of a CS-SRM-based intervention with adult patients provided evidence that communication in clinical consultations in which an adaptive understanding was provided meant that the patient felt understood, and adhered to prescribed treatment (Phillips et al., 2012). The intervention group included the physicians discussing the meaning of symptoms. This group provided a specific response with underlying reasons for management and expected treatment outcomes and possible alternatives when the treatment expectations were not met (Phillips et al., 2012). As discussed in Chapter 1, for adolescents the management of an LTC can be shared with the parents, and this dynamic between the clinician, parents and the adolescent patient has not been examined. However, with a CS-SRM approach, taking into consideration both the social context and the individual experiences, an establishment of independence and autonomy can be achieved. This independence can aid in the transition from paediatric services to adult services and the transition from shared management of the condition to self-management. The CS-SRM and the developments suggested in Chapter 8, do not just have implications on the type of information provided in clinic but also provided an underlying framework to study, understand and anticipate preferences, decisions and behaviours (Leventhal et al., 2012) to better manage the adolescent’s condition.

Leventhal and colleagues (Leventhal et al., 2012) argued that one approach to implement the CS-SRM in clinical settings is by using cognitive behavioural techniques. Tailored CBT treatment has been developed to encourage reappraisal and acceptance by promoting coping behaviours. These coping behaviours include
problem solving, relaxing, and sleep hygiene (Moss-Morris et al., 2013). There is an argument in pain research that for better management of complex regional pain syndrome there is a need to reconceptualise pain and what pain is (Moseley, 2007). At a biological level, pain is complex but what is evident is that the conceptualisation of pain provides the notion that there is damage to the tissue. However, pain is not a measure of the state and is multidimensional. Somatic, psychological, and social factors are implicated in pain. Therefore, there is a need to reconceptualise pain (Moseley, 2007). The barriers to reconceptualisation of pain; the shift from conceptualisation of a structure-pathology model to a conceptualisation of a neurophysiological model of pain, were investigated. This investigation found that both health professionals and patients accurately understood the neurophysiological model of pain however, the health professionals underestimated the ability of patients to understand the neurophysiology of pain (Moseley, 2003).

The PPQ-YP developed in this thesis would provide the justification for a psychoeducational intervention to reconceptualise pain. Furthermore, mood and emotions might be used as an indicator for goal conflict as well as an indicator for the pain representation. The goal orientation used in CBT, the process of identifying goals and the hierarchical model of goals (Powers, 1973), could be beneficial for clinic and for the process of reconceptualising pain for adolescents.

A more personalised and relevant approach is important, and the individual experiences were highlighted in this thesis. The results of a recent RCT which compared a book of metaphors to a pain management booklet based on cognitive-behavioural principles found that metaphor and stories reduced the patients’ catastrophising (Gallagher et al., 2013). There is a need for further work to extrapolate results of this thesis to further develop the CS-SRM driven tools to make them relevant to adolescents. Especially since most of the published work regarding reconceptualisation of pain has solely been with adults.
10.8. Future work

Whilst this thesis has made important contributions to exploring adolescents’ conceptualisation of an LTC these findings need to be expanded on. Research questions arose from this thesis, which require further work. For example, there is a need for further validation of the PPQ-YP to confirm the underlying factors. Considering the limitations described in Section 10.6.1 and Section 10.6.3, further studies are needed to determine if the results presented in Chapter 9 can be replicated in this sample and with other samples.

Following further validation of the PPQ-YP with other samples, it may be possible to investigate whether the shift from illness to symptom perceptions is due to the condition, to the adolescents’ cognitive development, or due to a development requirement of the CS-SRM and the IPQ-R. Comparisons with other adolescents’ relapsing and remitting pain conditions, adolescents’ with other LTCs, and adults with relapsing and remitting pain conditions is one method of contributing to further investigating this shift from illness to symptoms perceptions identified in this thesis.

Research within a CS-SRM framework examining symptom reporting found that patients with chronic illnesses and also with negative affect reported more symptom reporting compared to those with low negative affect (McAndrew et al., 2014). Further work is required to investigate the role of emotional representations in reporting symptoms in adolescents. Additional work should further investigate the perception of the impact of the condition, the adjustment to an LTC, and the perception of pain experience. Especially since emotions may make an individual more vigilant, internally focused, and increase pain-sensitivity (Watson & Pennebaker, 1989). This thesis provides interesting results regarding the relationship between the self-reported pain, emotions, and physical activity. However, taking into account the limitations described in Section 10.6.1, further work with advanced statistical models such as a non-recursive model to map the CS-SRM is required.

The advantage of utilising the CS-SRM with adolescents was that the development of the model can also include the social context of how the parents’ mental
representations impacted the adolescents’ development of mental representations. The CS-SRM also included the impact the parents may have on the adolescents’ illness behaviours. Contextual factors such as earlier experiences of illness influence the representation of the health threat as well as the coping response (Leventhal et al., 1998). Chronic pain itself interferes with developmental changes such as relationships with parents (Palermo et al., 2014) and peers (Forgeron et al., 2011). Pain in children is a family issue affecting many aspects of family functioning (Palermo, 2000). Research shows that parents’ experiences, beliefs, emotional wellbeing, and responses to pain are associated with their child’s pain outcomes (Connelly et al., 2012; Cohen et al., 2010; Palermo & Eccleston, 2009; Sieberg et al., 2011), and their children’s engagement in activities (Connelly et al., 2010). However, the underlying mechanisms are unclear. The parents of adolescents with high functioning, regardless of pain severity, expressed lower levels of anxiety and stress (Cohen et al., 2010). Furthermore, parents and children are more likely to stop activities when parents catastrophise about their child’s pain (Caes et al., 2012). In an observational study, Reid and colleagues explored the interactions between children with JIA and their parents during three exercise tasks. Parents’ behaviours, both encouraging and discouraging, affected the child’s task behaviour, and determined whether they stayed or discontinued the task (Reid et al., 2005). However, the dynamic and complex nature of parent-child interactions makes it difficult to identify causal pathways or specific targets for intervention.

10.9. Conclusions

The CS-SRM can be modified and applied for use with adolescents with an LTC. The three components of the CS-SRM were tested individually. Framework analysis found that the domains of the emotional and cognitive representations were relevant to the adolescents; their conceptualisation of JIA was built on their pain beliefs and experiences. The existing measure assessing illness representations required some modifications, both methodological issues related to comprehension (such as vague
or negative items), and conceptual issues related to the relevance to the condition (such as the assessment of the flare-ups). These modifications and developments were used to create a new tool, the PPQ-YP which was validated with a small group of adolescents with JIA.

Adolescents strived for normality as defined by social comparisons, and did so by either goal adjustment or goal pursuit to achieve a sense of normality. Adjustment or pursuit of their goal was dependent on whether the adolescent perceived their pain as a challenge or a threat to their normality resulting in differences in emotional profiles. Emotional representations were predictive of physical activity and this relationship was mediated by pain over time. The relationships between emotional representations and physical activity changed over time supporting the appraisal component of the CS-SRM.

This thesis has advanced the literature into how adolescents conceptualise an LTC. Given that the mental representation of a health threat is developed from current experiences, previous experiences, and information from their social context, future studies should consider if the results from this thesis to examine adolescents’ conceptualisation can be applied for other conditions.

This thesis is the first to examine the applicability of each component of the CS-SRM in adolescents, and to develop a theory and model for adolescents. The approaches and theory presented in this thesis provide a framework and the platform on which to build a better understanding of the conceptualisation and illness behaviours of adolescents with long-term conditions. Understanding adolescents’ conceptualisation and illness behaviours may help to address the challenges of self-management and inform health care interventions optimising quality of life and empowering this population.
Reference List


Henwood KL, Pidgeon NF Qualitative research and psychological theorizing..Br J Psychol. 1992 Feb;83 ( Pt 1):97-111.


**Appendix 1 - Aims and Objectives of the PhD and the Data sources**

**Research Question**: Can the Common Sense Self-Regulatory Model be applied to adolescents’ conceptualisation of illness? In this instance, Juvenile Idiopathic Arthritis (JIA).

<table>
<thead>
<tr>
<th>Objectives</th>
<th>Data Collection Instruments/tools/measure</th>
<th>Sample</th>
<th>Data Source</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Identify whether or not the domains of emotional and mental representations identified in the CS-SRM are relevant to adolescents with JIA.</td>
<td>Cognitive Interviewing with the Revised Illness Perceptions Questionnaire (IPQ-R). During the interview collected age and consent to access the CAPS data.</td>
<td>ACE-CAPS sub-study sample. 21 participants interviewed.</td>
</tr>
<tr>
<td>2</td>
<td>Assess the suitability of the current measure of illness representations for adolescents with JIA.</td>
<td>The cognitive interviewing data and the endorsements (answers) to the IPQ-R.</td>
<td>ACE-CAPS sub-study sample. 21 participants interviewed.</td>
</tr>
<tr>
<td>3</td>
<td>Identify adolescents’ goals and the coping strategies they employ to deal with pain related to JIA to determine the relationship between intention and behaviour.</td>
<td>Interviews about pain experiences using the In My Shoes (IMS) computer assisted interview tool.</td>
<td>ACE-CAPS sub-study sample. 21 participants interviewed.</td>
</tr>
<tr>
<td>4</td>
<td>Identify the emotional and cognitive profiles associated with specific coping goals.</td>
<td>Using the analysed cognitive interviewing data in combination to the analysed data from the IMS interviews.</td>
<td>ACE-CAPS sub-study sample. 21 participants interviewed.</td>
</tr>
<tr>
<td>5</td>
<td>Identifying the predictors of behaviour.</td>
<td>CAPS Questionnaires: -Emotional representations: section from IPQ-R -Pain: Visual Analogue Scale from</td>
<td>CAPS database. 55 participants.</td>
</tr>
<tr>
<td></td>
<td>6 Developing an adolescent specific measure of illness representations.</td>
<td>Feedback questionnaire developed specifically for the face validity study.</td>
<td>Opportunity sample for face validity study. 18 participants.</td>
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<td>---</td>
<td>-----------------------------------------------------------------------------</td>
<td>-----------------------------------------------------------------------------</td>
<td>-----------------------------------------------------------------------------</td>
</tr>
<tr>
<td></td>
<td>7 Testing and evaluating the psychometric properties of the new measure.</td>
<td>The Pain Perception Questionnaire for Young People developed in this PhD.</td>
<td>CAPS participants who consented to complete the new questionnaire e. 80 participants.</td>
</tr>
</tbody>
</table>

<sup>1</sup> Data collected in 2011 by author (DG) part of prior work.  
<sup>2</sup> Data collected between 2003 and 2014 by CAPS by members of the CAPS team.  
<sup>3</sup> Data collected between 2013 and 2014 by DG during PhD.
Appendix 2 – Interview Schedule for interviews

Cognitive Interview - examples of questions that the researcher will be asking

Adolescents will be asked to ‘think aloud’ as they answer it with ‘prompts’ given by the researcher. For example:

Written Item : IP1 - My illness will last a short time  Strongly agree to strongly disagree
Researcher: “You have just said disagree to the question. Could you tell me what made you pick disagree over strongly disagree?
Questions related to causes of arthritis symptoms.
Written Item : C2 - My illness runs in my family  Strongly agree to strongly disagree
Researcher: “You have just said disagree to the question. Could you tell me what made you pick disagree over strongly disagree?” “Is this something you have thought about before answering this questionnaire?” “How did this idea come to you”?

At the end of the questionnaire participants will be asked the following questions:

For any of these questions:

Were they difficult for you to answer?
What made them difficult?
Were there any questions you particularly liked?
What did you like?
What about the responses – were there any questions where you wanted to respond differently to the options given?

The aim of the questionnaire was to see how you think about your condition:

Is there anything that has been missed out?
What?

Is there anything listed there which you think could be dropped?
What?
What are the reasons?
If you could meet the person who designed this, what would you tell them?
What do you like about this questionnaire?
What do you think needs changing?

This example has been written around the CIPQ measure to illustrate the nature of the questions to be asked of participants. Similar questions will be asked of the IMS measure with adaptations reflecting the different response modalities in the measures.
<table>
<thead>
<tr>
<th>Question</th>
<th>Justification</th>
</tr>
</thead>
<tbody>
<tr>
<td>I’m going to ask you about a recent time when you found yourself feeling pain that had to do with arthritis. Can you think of a time in the past week?</td>
<td>The CHAQ uses the past week to discuss quality of life, and to discuss pain specifically due to illness. <em>(Singh, Atten, Tria &amp; Goldsmith, 1994)</em></td>
</tr>
<tr>
<td>Could you take the mouse and click on where it was?</td>
<td>This would choose the place on the body, describing where the pain was.</td>
</tr>
<tr>
<td>What kind of pain was it? Pick a picture that describes that pain.</td>
<td>This is using the palettes of IMS; gives YP the opportunity to use their own adjectives to describe pain to be used throughout the interview.</td>
</tr>
<tr>
<td>What does that picture mean to you?</td>
<td>This would be utilizing the palettes of IMS, speaking of the size of the pain, to see if size of pain would also determine the following answers.</td>
</tr>
<tr>
<td>Now chose the size of the pain, was it a small pain, like this or was it a big pain like this one? Or somewhere in the middle? Ok. Click on that one.</td>
<td>This would be just giving the YP further opportunity to use their own words to describe the pain, individualised to what they feel. Using the palette of IMS.</td>
</tr>
<tr>
<td>Now what was that pain like, was it constant stayed the same like that picture, or was it throbbing coming and going like that one there. Or somewhere in the middle. Ok. Click on that one.</td>
<td>This would reflect back how YP feels about pain and illness. Beliefs of illness be able to determine extent of negativity towards illness.</td>
</tr>
<tr>
<td>And how did you feel when you had that pain? Pick a face. What made you pick ...?</td>
<td>Based on CHAQ, where parents/YP is asked about the ability to do everyday things with illness. This would give YP the opportunity to give information relevant to them. Reflects about control.</td>
</tr>
<tr>
<td>Can you describe for me, what the day was like when you had that pain?</td>
<td>Timeline</td>
</tr>
<tr>
<td>How long did it last?</td>
<td></td>
</tr>
<tr>
<td>Tell me more about what it was like to have that pain?</td>
<td></td>
</tr>
<tr>
<td>Who did you tell about this pain?</td>
<td></td>
</tr>
<tr>
<td>What was it like telling someone? What happened? How did you feel?</td>
<td></td>
</tr>
<tr>
<td>OR. What made you not tell anyone about this pain?</td>
<td></td>
</tr>
<tr>
<td>Ok, so we can write here that it was ....</td>
<td></td>
</tr>
<tr>
<td>And here we are going to write; something about that pain that you felt, describe what it meant to have that pain.</td>
<td></td>
</tr>
<tr>
<td>Ok, so what would you do when you felt ...what do you usually want to do?</td>
<td></td>
</tr>
<tr>
<td>Did you feel any other pain at the same time as...?</td>
<td></td>
</tr>
<tr>
<td>At the end: What did you think of that?</td>
<td>Comments of opinion.</td>
</tr>
</tbody>
</table>
### Appendix 3 – Table of differences from IPQ-R to IPQ-C items

<table>
<thead>
<tr>
<th>New</th>
<th>Old</th>
</tr>
</thead>
<tbody>
<tr>
<td>Felt like vomiting</td>
<td>No question</td>
</tr>
<tr>
<td>Could not sleep well</td>
<td>No question</td>
</tr>
<tr>
<td>Sore eyes/not see well</td>
<td>Couldn’t see well</td>
</tr>
<tr>
<td>Feeling unwell</td>
<td>Not feeling well</td>
</tr>
<tr>
<td>Will be with me for the rest of my life</td>
<td>Will always have an effect on my life</td>
</tr>
<tr>
<td>Has large effect on my life</td>
<td>Stops me doing what I want</td>
</tr>
<tr>
<td>Does not have much effect on my life</td>
<td>Has little effect on what I do</td>
</tr>
<tr>
<td>Affects what other people think of me</td>
<td>Affects how other people treat me</td>
</tr>
<tr>
<td>Costs a lot of money to my family, or the hospitals</td>
<td>Costs a lot of money</td>
</tr>
<tr>
<td>Makes difficulties for my family</td>
<td>Upsets my family</td>
</tr>
<tr>
<td>There is a lot I can do to control my arthritis</td>
<td>Can be helped by what I do</td>
</tr>
<tr>
<td>No question</td>
<td>Is under my control</td>
</tr>
</tbody>
</table>
Appendix 4 – Information Sheets for interviews

Childhood Arthritis Prospective Study (CAPS)

Title of Project: Childhood Arthritis Prospective Study - Identifying the best ways to Assess Children's Experiences of arthritis within the CAPS study (ACE-CAPS)

Name of Main Researcher: Dr Wendy Thomson

1. We would like you to take part in a research study.
2. Please take time to read this leaflet carefully and discuss it with others if you wish.
3. Ask us if anything is unclear, or if you would like more information.
4. Take time to decide if you wish to take part.

Thanks for reading this!

Information of Study

We would like to ask you and your parent/guardian/s to take part in a sub-study. This sub-study will tell doctors about the best way to measure young people’s experience of arthritis and will be run at your clinic. We have provided more information about the sub-study below. Please read through this information before you decide whether you would like to take part. Please feel free to discuss this sub-study with your parent/guardian/s and doctors before making a choice. If you have any questions, the researcher will be in the clinic to answer them or you can contact one of the team using the contact information below.

Part 1

Why me?

We are asking all 11-16 year olds who attend clinic and are enrolled in the CAPS study if they would like to take part.

Do I have to take part?

No. It is up to you and your parent/guardian/s. If you would like to take part, the researcher will ask you if you have any questions. They will then ask you for your assent (agreement to take part) and then ask if you would sign the form. We will give you a copy this information sheet and your signed assent form to keep. You are free to stop taking part at any time during the research without having to give a reason. If you decide to stop, this will not affect the care you receive.
**What will happen if I take part?**
The researcher will be at the clinic when you attend your next check-up. If you would like to take part we ask that you and your parent/guardian/s are at the clinic 45 minutes earlier so you do the sub-study before your appointment. The sub-study will take place in a private room (not the waiting room). You and your parents will be given a chance to ask questions before the researcher asks you to take part in three tasks.

**What will I be asked to do?**
The three tasks that you will be asked to do are as follows:

1. You will be asked to fill in a questionnaire called the Illness Perception Questionnaire (CIPQ for short). You may recognise this questionnaire as everyone in the CAPS study is asked to fill this out starting from the age of 11. This time, the researcher will ask you to talk to them as you fill it out so you can tell them what you think of the questions and why you gave the answer you did on the questionnaire.

2. You will be asked to use a computer programme called ‘In my Shoes’. This has been designed to help young people talk about their experience of pain in relation to their condition. We want to know your thoughts on how useful this is for describing the pain you feel with arthritis. Again, the researcher will ask you to talk to them during this task so you can tell them what you think of ‘In My Shoes’ and explain to them why you gave the answer you did.

   It is really important for you to know that we simply want to know what you think about these measures: there are no right or wrong answers. We will be recording what you say during these tasks so the researcher can remember your feedback after the study has finished.

3. You will be asked to do a simple task of matching pictures which are shown to you with a word that the researcher will give you.

   Your parent/guardian/s will be in the room with you and the researcher for all of the activities. They will be given some tasks to do at the same time.
What if I have any questions?
You will be given a chance to ask questions before the study starts, so please feel free to bring a list of questions with you. You will be able to ask questions at any time during the study. If you have any questions after the study or any problems that you would like to discuss.

Please contact the researcher at:
Name: 
Email: 

Or you can contact Dr. Lis Cordingley, Lead Researcher on this sub-study:
Email: Lis.Cordingley@manchester.ac.uk
Telephone: 0161 275 7799

Or you can contact Prof. Wendy Thomson, CAPS Chief Investigator, at the Arthritis Research UK Epidemiology Unit:
Email: wendy.thomson@manchester.ac.uk
Telephone: 0161 275 5641

Thank you for reading so far. If you are still interested please continue reading part 2.
Part 2

Will anyone else know I'm doing this?
We will keep your information in confidence. This means we will only tell those who have a need or right to know. Wherever possible, we will only send out information that has your name and address removed.

What happens to the recordings of what I say and the forms I complete?
The researcher will write up the feedback you gave her when you were completing the measures. Anything that would identify you in the tapes will be removed your name will not be mentioned anywhere. The feedback and the forms you completed will only be accessed by staff working on this study.

Although we will be reporting our study findings back to the doctors you will not be identified in any way.

Who is organising and funding the research?
This research is being organized by the CAPS study and the University of Manchester.

Who has reviewed the study?
Before any research goes ahead it has to be checked by a Research Ethics Committee. They make sure that the research is fair. Your project has been checked by the Northwest Multi Centre Research Ethics Committee

Thank you for reading this - please ask any questions if you need
CONSENT FORM FOR PARENTS OR GUARDIANS OF CHILDREN TAKING PART IN RESEARCH STUDIES
(Version 1, 3rd December 2010)

Title of Project: Childhood Adversity Perspectives Study - Identifying the best ways to assess children's experiences of adversity within the CAPS study. (ACE-CAPS)

Name of Researcher:

1. I confirm that I have read and understand the information sheet dated 12/12/20 (Version 1) for the above study and have had the opportunity to ask questions.

2. I understand that my child's participation in this study is voluntary and the child is free to withdraw at any time, without giving any reason, without my child's medical care or development being affected.

3. I agree to my child taking part in the above sub-study.

4. I agree to my child being audio recorded while taking part in the above study and understand that this is confidential information, e.g. names, places, will be removed from the written records of the recording.

5. I agree to take part in the sub-study.

Name of Parent: ___________________ Date: ___________ Signature: ___________________

Name of Parent/Guardian: ___________________ Date: ___________ Signature: ___________________

Phone: ___________________ Date: ___________ Signature: ___________________

I have signed this form in the presence of: ___________________ Date: ___________ Signature: ___________________

This patient: 1 for race/ethnicity, 2 to be female, 3 to be male.

ASSENT FORM FOR CHILDREN TAKING PART IN RESEARCH STUDIES
(Version 1, 3rd December 2010)

Title of Project: Childhood Adversity Perspectives Study - Identifying the best ways to assess children's experiences of adversity within the CAPS study. (ACE-CAPS)

Name of Researcher: ___________________

1. I have read and understood the information sheet dated 12/12/20 (Version 1).

2. I have been asked the questions that I need to ask.

3. I understand that I do not need to take part in the sub-study and that I can stop taking part at any time without taking any action.

4. I understand that my medical notes may be kept by people doing the research.

5. I understand that what I did during the sub-study will be recorded and not written

6. I understand that the names of people or places I may be referred to will be changed to make the notes and these things.

7. I agree to take part in the sub-study.

Name of Patient: ___________________ Age: _______ Date: ___________ Signature: ___________________

Name of Parent/Guardian: ___________________ Date: ___________ Signature: ___________________

Phone: ___________________ Date: ___________ Signature: ___________________

This patient: 1 for race/ethnicity, 2 to be female, 3 to be male.
Appendix 5 – Screenshot of the Somatic Experiences Module from the In My Shoes Interview Tool

- Emotion Palette: Pick a face that described how they felt.
- Back/Front Body View: Here the young people choose the location of the pain; it can either be a boy or a girl depending on the young person.
- Description: Either of the day, emotion, or coping strategy.
- Picture of Pain: Pictures to describe a type of pain.
- Size of pain
- Throbbing
- Label: Description of the pain.
Appendix 6 – Feedback question for PPQ-YP

Feedback Questions

We created a new questionnaire, the Pain Perception Questionnaire for young people (PPQ-YP) to assess pain beliefs of young people who have pain because they have arthritis. We want to check if the instructions and questions make sense and we need your help.

We would like to ask you to think about a time you felt pain and fill in the questionnaire based on your experiences of pain. After you finish the PPQ-YP we would like you to answer the questions on the next page about the PPQ-YP.

But before you answer the questions on the next page please can you tell us your age [years] and gender: male female ?

And how long did it take for you to finish the questionnaire (PPQ-YP)?
Feedback Questions

1. For questions in Section A: were the instructions clear?
   YES □   NO □   if NO why not?

2. For questions in Section B: were the instructions clear?
   YES □   NO □   if NO why not?

3. For questions in Section C: were the instructions clear?
   YES □   NO □   if NO why not?

4. For questions in Section D: were the instructions clear?
   YES □   NO □   if NO why not?

5. Were there any words or terms you found difficult?
   YES □   NO □   If YES which words or terms and in which questions?

6. Were there any questions you did not understand?
   YES □   NO □   If YES which question and why?

Have you any other comments?
Appendix 7 – Pack sent to potential participants for recruitment

Dear Participant

Developing and Validating the Pain Perceptions Questionnaire for Young People (PPQ - YP)

Thank you for your participation in the Childhood Arthritis Prospective Study (CAPS). You may still be under regular follow-up with a paediatric rheumatologist or you may have been discharged from their care.

We are writing to you because we would like to ask if you would be willing to participate in a sub-study, ‘Developing and Validating the Pain Perceptions Questionnaire for Young People (PPQ - YP)’. The aim of this sub-study is to test a new questionnaire we are developing which measures young people’s beliefs about the pain that they experience. For your participation in this sub-study we would ask you to fill in an additional questionnaire that is included in this package. Once completed it is returned with the other questionnaires as usual in the pre-paid envelope provided.

We would very much appreciate it if you could take the time to read the included information sheets to find out more about this sub-study.

If you have any queries about this letter, please feel free to contact the CAPS team in at the University of Manchester (Daniela Ghio at daniela.ghio@postgrad.manchester.ac.uk or by telephone on 0161 275 7506).

If you do not have any further questions please complete and sign one consent form and return it with the completed questionnaires to the Arthritis Research UK Epidemiology Unit in the pre-paid return envelope.

Many thanks for your help.

Peter Ward
Study Coordinator

Telephone: 0161 275 1055
Email: Peter.Ward@manchester.ac.uk

Patient Invitation Letter – Version 1, 23rd June 2013
Title of Project: Childhood Arthritis Prospective Study - Developing and Validating the Pain Perception Questionnaire for Young People (PPQ-YP)

Name of Main Researcher: Dr Wendy Thomson

We would like you to take part in a research study.
Please take time to read this leaflet carefully and discuss it with others if you wish.
Ask us if anything is unclear, or if you would like more information.
Take time to decide if you wish to take part.

Thanks for reading this!

Information of Sub-Study
We would like to ask you and your parent/guardian/s to take part in a sub-study. This sub-study will be able to help us make sure a new questionnaire works properly. We have provided more information about the sub-study below. Please read through this information before you decide whether you would like to take part. Feel free to discuss this sub-study with your parent/guardian/s and doctors before making a choice. If you have any questions, you can contact one of the team using the contact information below.

Part 1

Why me?
We are asking all 11-16 year olds who are enrolled in the CAPS study if they would like to take part.

Do I have to take part?
No, it is up to you and your parent/guardian/s. If you would like to take part, we will ask you for your assent (agreement to take part) in the assent form. We will give you a copy this information sheet and your signed assent form to keep. You are free to stop taking part at any time during the research without having to give a reason. If you decide to stop, this will not affect the care you receive.
What will happen if I take part?
If you would like to take part please sign the assent form that is in this pack. You can choose to take part in this sub-study just once, or if you want to take part again you can say so on the assent form. The second time we will ask you to only fill out just the new sub-study questionnaire.

If you have any questions about the new sub-study questionnaire you can contact the researcher, the contact details are in Part 2 of this information sheet.

What will I be asked to do?
You have just received your usual pack of questionnaires from CAPS, included in this pack is the new sub-study questionnaire the 'Pain Perceptions Questionnaire for Young People (PPQ-YP)'. What you will be asked to do is to fill it out just like the other questionnaires and send it back with the rest of the CAPS questionnaires as normal.

If you are interested in taking part in the second stage to this sub-study then you will be contacted again 3 months after filling out the first questionnaire. In the second stage you will only be given the new sub-study questionnaire the 'Pain Perceptions Questionnaire for Young People (PPQ-YP).
What if I have any questions?
If you have questions or problems before the start or at any time during the sub-study then please feel free to discuss with our research team:

Please contact the researcher at:

Daniela Ghio
Email: daniela.ghio@postgrad.manchester.ac.uk
Telephone: 0161 275 7506

Or you can contact Dr. Lis Cordingley, Lead Researcher on this sub-study:

Email: Lis.Cordingley@manchester.ac.uk
Telephone: 0161 275 7799

Or Professor Wendy Thomson, CAPS Chief Investigator, at the Arthritis Research UK Epidemiology Unit:

Email: wendy.thomson@manchester.ac.uk
Telephone: 0161 275 5641

Thank you for reading so far.
If you are still interested please continue reading part 2.
Part 2

Will anyone else know I’m doing this?
We will keep your information in confidence. This means we will only tell those who have a need or right to know. Wherever possible, we will only send out information that has your name and address removed.

What happens to the forms I complete?
The researcher will write up the feedback you gave her when you were completing the measures. The feedback and the forms you completed will only be accessed by staff working on this study.

Although we will be reporting our study findings back to the doctors you will not be identified in any way.

Who is organising and funding the research?
This research is being organized by the CAPS study and the University of Manchester.

Who has reviewed the study?
Before any research goes ahead it has to be checked by a Research Ethics Committee. They make sure that the research is fair. Your project has been checked by the Northwest Multi Centre Research Ethics Committee

Thanks for reading this information leaflet. If you do decide to take part in the study, you will be given a copy of this leaflet to keep and will be asked to sign a consent form.

This research is funded by the Arthritis Research UK and is based at the University of Manchester.
ASSENT FORM FOR CHILDREN TAKING PART IN
RESEARCH STUDIES
(Version 1, 20th June 2013)

Title of Project: Developing and Validating the Pain Perceptions Questionnaire for Young People (PPQ-YP)

Name of Researcher: 

Please initial box

1. I have read and understood the information sheet dated 28.06.13 (Version 1).

2. I have asked all the questions that I want to ask.

3. I understand that I do not have to take part in the sub-study and that I can stop taking part at any time without saying why.

4. I understand that parts of my medical notes may be looked at by people doing the research.

5. I understand that I might be contacted to fill out the questionnaire again.

6. I agree to take part in the sub-study.

7. I agree to being contacted again in three months.

Name of Patient:_________________________  Age:________ Date:________  Signature:_____________________

1 for patient 1 for researcher; 1 to be kept with hospital notes
Appendix 8 – Matrix for the results of the first PCA including all items

<table>
<thead>
<tr>
<th>Item</th>
<th>Pattern Coefficients</th>
<th>Structure Coefficients</th>
<th>Communalities</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>I</td>
<td>II</td>
<td>III</td>
</tr>
<tr>
<td>PainHome</td>
<td>.819</td>
<td>-.164</td>
<td>-.015</td>
</tr>
<tr>
<td>PainLong</td>
<td>.808</td>
<td>-.115</td>
<td>.175</td>
</tr>
<tr>
<td>PainOthers</td>
<td>.788</td>
<td>.299</td>
<td>-.202</td>
</tr>
<tr>
<td>PainSchool</td>
<td>.750</td>
<td>-.258</td>
<td>-.002</td>
</tr>
<tr>
<td>PainActivities</td>
<td>.749</td>
<td>-.156</td>
<td>.052</td>
</tr>
<tr>
<td>PainImprove</td>
<td>.749</td>
<td>-.313</td>
<td>.349</td>
</tr>
<tr>
<td>PainOften</td>
<td>.738</td>
<td>-.116</td>
<td>-.086</td>
</tr>
<tr>
<td>FamilyProblem</td>
<td>.733</td>
<td>-.041</td>
<td>-.144</td>
</tr>
<tr>
<td>PainSerious</td>
<td>.693</td>
<td>-.209</td>
<td>-.144</td>
</tr>
<tr>
<td>FamilyMoney</td>
<td>.673</td>
<td>-.023</td>
<td>-.230</td>
</tr>
<tr>
<td>HospitalMoney</td>
<td>.587</td>
<td>.062</td>
<td>-.248</td>
</tr>
<tr>
<td>PainAdult</td>
<td>.443</td>
<td>-.129</td>
<td>-.117</td>
</tr>
<tr>
<td>BehaviourPain</td>
<td>.387</td>
<td>.211</td>
<td>.053</td>
</tr>
<tr>
<td>StopSoon</td>
<td>-.361</td>
<td>.203</td>
<td>.265</td>
</tr>
<tr>
<td>TreatmentBetter</td>
<td>.077</td>
<td>.758</td>
<td>.256</td>
</tr>
<tr>
<td>TreatmentProtect</td>
<td>.021</td>
<td>.717</td>
<td>.064</td>
</tr>
<tr>
<td>Treatment Activities</td>
<td>-.177</td>
<td>.709</td>
<td>.126</td>
</tr>
<tr>
<td>CandoBetter</td>
<td>-.177</td>
<td>.666</td>
<td>.071</td>
</tr>
<tr>
<td>ControlPain</td>
<td>-.199</td>
<td>.636</td>
<td>.142</td>
</tr>
<tr>
<td>TreatAmountControl</td>
<td>-.190</td>
<td>.620</td>
<td>.194</td>
</tr>
<tr>
<td>ControloTreat</td>
<td>-.155</td>
<td>.615</td>
<td>.099</td>
</tr>
<tr>
<td>AmountControl</td>
<td>-.219</td>
<td>.600</td>
<td>.240</td>
</tr>
<tr>
<td>UnderstandClearly</td>
<td>.064</td>
<td>.255</td>
<td>.833</td>
</tr>
<tr>
<td>UnderstandCause</td>
<td>.044</td>
<td>.235</td>
<td>.720</td>
</tr>
<tr>
<td>UnderstandTreat</td>
<td>.027</td>
<td>.172</td>
<td>.692</td>
</tr>
<tr>
<td>QuestionsPain</td>
<td>.045</td>
<td>.118</td>
<td>.653</td>
</tr>
<tr>
<td>WhyPain</td>
<td>.271</td>
<td>-.090</td>
<td>-.642</td>
</tr>
<tr>
<td>PredictPain</td>
<td>.184</td>
<td>.214</td>
<td>.028</td>
</tr>
<tr>
<td>PatternPain</td>
<td>.184</td>
<td>.118</td>
<td>.240</td>
</tr>
<tr>
<td>ComesandGoes</td>
<td>.162</td>
<td>.284</td>
<td>-.365</td>
</tr>
<tr>
<td>ChangesEveryday</td>
<td>.374</td>
<td>.067</td>
<td>-.288</td>
</tr>
<tr>
<td>FuturePain</td>
<td>.043</td>
<td>-.042</td>
<td>-.108</td>
</tr>
</tbody>
</table>
Appendix 9- PPQ-YP

CAPS ID:

Pain Perceptions Questionnaire
for Young People

PPQ-YP

- We are interested in your views and how you feel about pain you may have relating to your arthritis.

- These are statements other people have made about their pain.

- For section A, please show how much you agree or disagree with each of the following statements about your pain by marking it on the scale. Like this:

  ![Scale Image]

  - Strongly disagree
  - Disagree
  - Neither agree nor disagree
  - Agree
  - Strongly agree

Please tell us the date on which you completed this form

Date: ______________
Section A.

A1. I believe I will stop getting pain soon

A2. I believe I will keep having pain when I am an adult

A3. When I get pain, it lasts a long time

A4. Over time I am getting pain more often

2 Version 2, 11th November 2013
A5. When I get pain I think my pain will improve in

- An hour or less
- A day or less
- A week or less
- Two weeks or less
- More than two weeks

A6. When I get pain it makes me think my pain is

- Not serious at all
- A bit serious
- Serious
- Somewhat serious
- Very serious

A7. When I have pain it stops me from taking part in activities such as PE

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

A8. When I have pain, it affects me at school such as school work, school friends

- Strongly disagree
- Disagree
- Neither agree nor disagree
- Agree
- Strongly agree

Version 2, 11th November 2013
A9. When I have pain, it affects how I am at home

A10. I believe my pain affects what other people think of me

A11. I believe my family spend more money because I have pain

A12. I believe my pain makes hospitals spend a lot of money
A13. I believe my pain causes problems for my family

A14. I can do a lot to control my pain

A15. This is the amount of control I feel I have over my pain

A16. There are things I can do to make my pain better
A17. Things I do now can affect whether I have pain in the future

Strongly disagree  Disagree  Neither agree nor disagree  Agree  Strongly agree

A18. My behaviour can affect how much pain I have

Strongly disagree  Disagree  Neither agree nor disagree  Agree  Strongly agree

A19. I feel confused about why I get pain

Strongly disagree  Disagree  Neither agree nor disagree  Agree  Strongly agree

A20. I understand how my treatment for pain works

Strongly disagree  Disagree  Neither agree nor disagree  Agree  Strongly agree
A21. I understand my pain clearly

A22. I don't have any questions about my pain

A23. When I have pain I understand what causes my pain

A24. My pain changes everyday
A25. My pain comes and goes

A26. I can predict when I will get pain

A27. I can see a pattern in how and when I get pain

A28. I am in control of my treatment for my pain
A29. My treatment helps my pain get better

A30. My treatment protects me from pain

A31. I can continue with my activities because of my treatment

A32. Taking my treatment means I have

Version 2, 11th November 2013
We would like to ask you about how you FEEL when you are in pain.

Show how often you feel this way by marking on the scale.

And when do you feel these emotions, how much do you feel them?

Section B.

B1. When I am in pain I feel down and sad

How often?

How much?

B2. When I am in pain I feel angry

How often?

How much?

B3. When I am in pain I feel upset

How often?

How much?
B4. When I am in pain I feel afraid

How often?

Never  Rarely  Sometimes  Most of the time  Always

How much?

None  A little  Quite a bit  A fair amount  A lot

B5. When I am in pain I feel frustrated

How often?

Never  Rarely  Sometimes  Most of the time  Always

How much?

None  A little  Quite a bit  A fair amount  A lot

B6. When I am in pain I feel worried

How often?

Never  Rarely  Sometimes  Most of the time  Always

How much?

None  A little  Quite a bit  A fair amount  A lot

B7. When I am in pain I feel anxious

How often?

Never  Rarely  Sometimes  Most of the time  Always

How much?

None  A little  Quite a bit  A fair amount  A lot
We would like to ask you about any SYMPTOMS you may have experienced since you have been having pain.

- Please show whether you have experienced each of the following symptoms in the PAST WEEK by circling YES
- For each please show whether you believe it is related to your pain by circling YES and whether you believe it is related to your treatment by circling YES
- If you have not felt the symptoms please circle NO and move on to the next symptom

<table>
<thead>
<tr>
<th>Symptom</th>
<th>I had this recently</th>
<th>If answer is YES</th>
<th>This symptom is related to my Pain</th>
<th>This symptom is related to my treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>C1 Fast like vomiting</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C2 Couldn’t breathe well</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C3 Lost or put on weight</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C4 Feeling tired</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C5 Joints feel soft</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C6 Joints feel sore</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C7 Sore eyes /nose/ear</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C8 Feeling unwell</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C9 Getting headache</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C10 Could not sleep well</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C11 Upset tummy</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C12 Fast dizzy</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C13 Felt weak</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C14 Feelings of tightness in my body</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
<tr>
<td>C15 Change in my mood</td>
<td>NO</td>
<td>YES</td>
<td>YES</td>
<td>NO</td>
</tr>
</tbody>
</table>
For example: **stress or worry**

- **THEN** Place a number near the circled causes in the order of importance with 1 meaning the most important.

For example: If you think that runs in the family is the most important cause of your pain, then you would number this 1. Like this:

<table>
<thead>
<tr>
<th>Stress or worry</th>
<th>runs in the family</th>
<th>immune system</th>
</tr>
</thead>
<tbody>
<tr>
<td>3.</td>
<td>1.</td>
<td>2.</td>
</tr>
</tbody>
</table>

### Section D.

<p>| | |</p>
<table>
<thead>
<tr>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>D1</td>
<td>Stress or worry</td>
</tr>
<tr>
<td>D2</td>
<td>It runs in the family (genetics)</td>
</tr>
<tr>
<td>D3</td>
<td>A germ or virus</td>
</tr>
<tr>
<td>D4</td>
<td>Diet or eating habits</td>
</tr>
<tr>
<td>D5</td>
<td>Chance or bad luck</td>
</tr>
<tr>
<td>D6</td>
<td>Poor health and bad medical care in my past</td>
</tr>
<tr>
<td>D7</td>
<td>Pollution (like fumes, dirty water) or toxins in the environment</td>
</tr>
<tr>
<td>D8</td>
<td>My own behaviour</td>
</tr>
<tr>
<td>D9</td>
<td>My attitude (for example thinking negatively) about life</td>
</tr>
<tr>
<td>D10</td>
<td>Family problems, family worries</td>
</tr>
<tr>
<td>D11</td>
<td>Doing too much</td>
</tr>
<tr>
<td>D12</td>
<td>Feeling down, lonely, nervous or empty</td>
</tr>
<tr>
<td>D13</td>
<td>Getting older</td>
</tr>
<tr>
<td>D14</td>
<td>Accident or injury</td>
</tr>
<tr>
<td>D15</td>
<td>The type of person that I am</td>
</tr>
<tr>
<td>D16</td>
<td>My immune system</td>
</tr>
<tr>
<td>D17</td>
<td><em>Any other cause that you think of</em></td>
</tr>
</tbody>
</table>
In the PAST WEEK how much pain have you had because of your arthritis? Place a mark on the line below, to indicate the severity of the pain.

0  
No pain

100  
Very severe pain