JUST GETTING ON WITH IT: FAMILY EXPERIENCE OF JUVENILE IDIOPATHIC ARTHRITIS

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ABSTRACT

Introduction: A chronic condition does not just affect the individual diagnosed, but also their families. The process of adaptation, following the onset of symptoms, can be complex requiring flexibility from the family. This may be especially pertinent with Juvenile Idiopathic Arthritis (JIA) which is characterised by unpredictable flare-ups and an uncertain disease trajectory. Families negotiating JIA may be at an increased vulnerability of distress as a result of additional demands placed upon their resources. This may have implications for health services. It is therefore important to understand family experiences of living with a chronic condition in order to support families throughout the adaptation process. To date, the majority of studies have investigated individual family members' reports in order to assess family functioning, but these investigations have neglected to study the family as a unit.

Method: This study utilised a multiple-perspective case study design in order to explore family experiences of JIA. Two families were recruited from a paediatric rheumatology service in Leeds. Family group interviews were conducted and five of the seven participating family members completed follow-up individual interviews, which used a semi-structured interviewing format. Interviews were transcribed and an interpretative phenomenological approach was used to analyse each case study. A synthesis of the results was also conducted.

Results: Four master themes were identified from the first family interviews. These were: *negotiating power, not letting go: managing transitions, when the invisible becomes visible* and *just getting on with it*. Four master themes were also identified from the second family: *a positive outlook, being 'normal', power and empowerment* and *medications: friend or foe.* Analysis also focused upon how both families negotiated their understandings of JIA. Five themes were identified following a synthesis of the case study data. These were: *Just getting on with it and maintaining a sense of normality*, *battling, fighting and the negotiation of power*, *transitioning*, *JIA as a hidden condition* and *negotiating understandings*.

Discussion: The themes relating most significantly to the research aims: *just getting on with it and maintaining a sense of normality, battling, fighting and the negotiation of power* and *negotiating understandings,* from the synthesised data were discussed within the context of the existing chronic health conditions and family communication literature. The study's methodological strengths and limitations were also presented following the discussion of the themes. Clinical implications relating to families experiencing JIA and services providing care to these families was discussed throughout the discussion chapter, and finally, recommendations for further research were outlined.

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ABBREVIATIONS

CF: Cystic Fibrosis

DoH: Department of Health

IPA: Interpretative Phenomenological Analysis

JIA: Juvenile Idiopathic Arthritis

LGI: Leeds General Infirmary

NHS: National Health Service

CHAPTER ONE

Literature Review

This chapter will outline the current literature and evidence-base regarding childhood chronic health conditions, with a specific focus upon Juvenile Idiopathic Arthritis (JIA) and its impact upon family functioning. There is value in taking a general approach to understanding chronic health conditions, as it is likely that there will be commonalities between conditions with regards to psychological response and impact upon the young person and family. There is equal value, however, in remaining disease-specific, as there may also be unique challenges posed to families as a consequence of particular health conditions. This study focused specifically upon JIA and the literature reviewed in this chapter will cover the specific and unique challenges individuals and families face when diagnosed with this health condition. However, to gain a rich understanding of the impact a chronic health condition has upon young individuals and their families, this review also includes research of other health conditions.

Due to the need to take broad approach to outline the literature, only a limited number of papers can be presented. Where possible, papers of a high quality and meta-analyses were used to highlight the themes emerging from the current evidence-base. In addition, due to the large volumes of research studying the medication effectiveness of JIA and other health conditions, medical focused databases, such as Medline, were not utilised. A fuller range of research databases were utilised to search for research most relating to the present study to identify gaps within the evidence-base (see Appendix 1 for table of search terms).

Childhood chronic health conditions

It is estimated that 15% of all children will experience a chronic health condition at some stage in their development (Newacheck & Stoddard, 1994). This indicates that a significant proportion of individuals and families are faced with the prospect of managing and adjusting to a chronic condition. A chronic health condition, by definition, is a complex and multi-faceted concept (Stanton, Revenson, & Tennen, 2007). Definitions of chronic conditions vary throughout the literature resulting in difficulties in determining reliable estimates of their prevalence. Furthermore, what is defined as being 'chronic' is also determined by the definitions presented in the literature (Perrin, et al., 1993; van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). Perrin et al. (1993) highlight the difficulty in categorising chronic conditions, due to considerations regarding: the duration of a condition[1], terminology use[2], whether the impact is

[1] Intermittent or continuous trajectory

physical or psychological, the severity of a condition and prognosis. Historically, definitions have excluded the psychological and emotional impact of conditions, although more current descriptions have incorporated these concepts. Eiser (1997) suggests that a condition should be considered 'chronic' if it lasts more than a year, limits at least one aspect of physical, social, cognitive and/or emotional functioning and the individual requires the assistance of either psychological, medical and/or educational services. Furthermore, Eiser (1997) proposes that 'chronicity' also results in the individual displaying disproportionate levels of dependency upon others (for example, parents) compared to expected developmental trajectories. With regards to childhood chronic conditions, Perrin et al. (1993) more generally define a chronic health condition as lasting more than three months whereby there is some observed limitations, such as the young person's level of physical or psychological functioning, and a level of dependency that is greater than expected for a young person of that age. Perrin et al. (1993) suggest that definitions of a chronic health condition need to be general and flexible in order to encompass the many dimensions, such as severity, impact and idiosyncratic presentations. A consensus across all of these definitions is that a chronic condition will place added demands upon the young person diagnosed and their family, however, the demands and the impact may vary both within and between conditions (Eiser, 1997).

Juvenile Idiopathic Arthritis

JIA is the most commonly diagnosed arthritic condition in children (Prince, Otten, & van Suijlekom-Smit, 2010) and according to definitions outlined earlier, is considered to be a chronic health condition. In the UK approximately one in every 10,000 young people are diagnosed with JIA every year (Symmons, Jones, Osborn, Sills, Southwood, & Woo, 1996) and approximately one-third of these young people continue to report symptoms into adulthood (Packham, 2008), indicating the longevity and magnitude of the condition.

Arthritis is an auto-immune condition defined as a swelling and tenderness of the joints, invariably accompanied by a restriction in joint movement and permanent joint damage. Physical mobility, joint deterioration, tissue damage, fatigue, disfigurement and chronic pain are also common symptoms or secondary consequences of the condition (Payne & Norfleet, 1986; Petty, et al., 2004; Prince et al., 2010). The aetiology of JIA is unknown, although it is generally assumed that both genetic and environmental factors are important contributors (Prince et al., 2010), with developing hypotheses of viral infections as causal factors (Ellis, Munro & Ponsonby, 2010).

A young person will receive a diagnosis of JIA if both their physical symptoms have persisted for at least six weeks, and if he or she is aged 16 years or younger (Petty, et al., 2004).

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[2] For example, hay fever can be classified as a chronic condition but not as a disease

JIA is the umbrella term for a number of sub-types of JIA which have a range of different symptoms and treatments and can also be defined at the age at which the symptoms become present (Prince et al., 2010). The JIA subtypes can be viewed in *Table 1*on the following page.

Diagnosis and treatment can be both emotionally and physically invasive and thus distressing for both child and family. Diagnosis comprises: a history taking of the problems and symptoms, physical examinations[3], observations and functional assessments (Prince et al., 2010). Treatment is dependent on the type of arthritis diagnosed; however, at present there is no specific cure and disease remission is the best possible outcome. Treatment aims to control the inflammation to improve joint movement and to minimise permanent joint damage, which may result in osteoporosis (Prince et al., 2010). Enduring raised levels of inflammation are therefore correlated to poorer rates of physical functioning (Packham, Hall, & Pimm, 2002) and so early aggressive treatment is essential. Typically, medication and physiotherapy are the prioritised medical interventions for JIA (Prince et al., 2010).

JIA can also be particularly distressing for a young person as adherence to prescribed treatment regimens does not necessarily predict a positive prognosis. In addition, intermittent periods of symptom-free episodes and periods of exacerbation can render the condition as unstable and unpredictable (Boekaerts & Röder, 1999). At least one-third of young people with the condition continue to have active symptoms into adulthood (Packham et al., 2002). Due to the nature of JIA, delays in physical, emotional and social development in the growing young person are possible, including social isolation as a result of missed schooling (Suris, Michaud, & Viner, 2004). These factors may continue to impact upon the individual into adulthood (McDonagh & White, 2008).

[3] For example, haematology tests and bone scans

Table 1. **Subtypes of JIA.**

Category	Characteristics	Age of onset	sex ratio (F:M)
Systemic JIA	Arthritis and daily fever for more than 3 days, accompanied by at least one of the following: erythematous rash, generalised lymph node enlargement, hepatomegaly or splenomegaly, serositis	Throughout childhood	1:1
Oligoarthritis	Arthritis affecting 1-4 joints during the first 6 months of the disease	Early childhood (peak 2-4 years)	5:1
Persistent	Arthritis affecting less than 4 joints throughout disease course		
Extended	Arthritis affecting more than 4 joints after the first 6 months of the disease		
Polyarthritis	Arthritis affecting more than 5 joints during the first 6 months of the disease		
Rheumatoid factor positive	Two or more positive tests for rheumatoid factor at least 3 months apart	Late childhood or adolescence	3:1
Rheumatoid factor negative	Tests for rheumatoid factor negative	Early peak 2-4 years and late peak 6-12 years	3:1
Psoriatic arthritis	Arthritis and psoriasis, or arthritis and at least 2 of the following: dactylitis, nail pitting, psoriasis in a first degree relative	Late childhood or adolescence	1:0.95
Enthesitis related arthritis	Arthritis and enthesitis, or arthritis or enthesitis with at least 2 of the following: sacroiliac joint tenderness or inflammatory lumbosacral pain (or both), HLA-B27 antigen positive, onset in male over 6 years old, acute anterior uveitis, HLA-B27 associated disease in first degree relative	Early peak 2-4 years and late peak 6-12 years	1:7
Undifferentiated arthritis	Arthritis that fulfils criteria in no specific category or meets criteria for more than one category		

Table from Prince et al. (2010).

Adaptation and adjustment to a chronic health condition

The literature indicates that individuals recently diagnosed with a chronic condition must manage and negotiate a number of stressors and new regimes quickly, in order to gain control over the condition and to minimise its physical and psychological impact. This may result in increased levels of psychological distress if an individual struggles to effectively manage these added demands (Eiser, 1990; Wallander & Varni, 1998). The individual with the chronic condition, together with their family, may need to integrate complex physical and medical regimes into daily life such as: negotiating time for hospital appointments, adjusting to new roles and identities, living with new restrictions and learning to tolerate uncertainty (Boekaerts & Röder, 1999; Christie & Khatun, 2012). A chronic health condition may also impact upon self-esteem, social relationships and education, which may place further strain on the psychological well-being of the young person (Yeo & Sawyer, 2005). The degree to which an individual adjusts to disease related features can be dependent upon biopsychosocial factors, that is, biological and disease factors, psychological factors and social factors (Walker, Jackson, & Littlejohn, 2004). The biological aspects of a chronic condition that could impact on adjustment may be: disease symptoms, the degree of disability, delayed physical development, invasive treatment, chronic pain and the side effects of medication (Eiser, 1990). The psychological factors may include: personal resources, coping strategies and cognitive appraisal, loci of control (Sperry, 2009) and illness representations (Leventhal, Brissette, & Leventhal, 2003). The social factors may include: the impact the family's copying styles have upon the individual with the chronic condition, resources, access to medical facilities and the amount and quality of social support available (Suris et al., 2004).

The two terms 'adjustment' and 'adaptation' are used interchangeably throughout the chronic health literature, with limited distinction between these two concepts. Within this literature review, research that has employed both these two concepts as measures or processes will be utilised in order to better comprehend individuals' and families' responses to a diagnosis of a chronic condition. Adaptation can be regarded as a continuously developing and changing process, in which individuals must make modifications across a number of life domains. Adaptation, therefore is a pathway to adjusting to change (Livneh, 1997; Miller Smedema, Bakken-Gillen, & Dalton, 2009). This is idiosyncratic to the individual experiencing a chronic condition, and the degree to which an individual 'adjusts' to significant life events is not linear and will therefore fluctuate over time (Christie & Khatun, 2012; Walker et al., 2004).

Stanton, Collins and Sworowski (2001) identified five domains of positive adjustment to a chronic condition: mastery of disease-related tasks, preserving optimal functioning and quality of life, an absence of significant psychological difficulties and low levels of negative affect. These above authors additionally synthesised past literature on adjustment and proposed several adaptive

tasks to aid adjustment to a serious health condition, including: regulating distress, restoring relationships, pursuing optimal physical functioning (including that of managing symptoms), enhancing or restoring self-esteem and searching for meaning from the experience. Adjustment, in the literature, is often measured by examining levels of psychological well-being in a young person or the family close to that individual. Young people who exhibit positive psychological functioning following the onset of a chronic condition, will demonstrate healthy, age appropriate behaviours and normative social interactions that follow a trajectory towards positive adult functioning (Wallander, Thompson, & Alriksson-Schmidt, 2003). Research suggests that the way a young person adapts to a chronic health condition will have consequences for long-term functioning and well-being (LeBovidge, Lavigne, Donenberg, & Miller, 2003).

The evidence-base demonstrates mixed results with regards to psychological well-being following a diagnosis of a chronic health condition. For example, some studies have observed that young people exhibit greater psychological distress, such as anxiety and depression, when comparisons are made against matched controls (Billings, Moos, Miller, & Gottleib, 1987). A meta-analysis of adjustment to chronic conditions also concluded that affected young people report higher rates of somatic complaints, social withdrawal, together with anxiety and depression (Lavigne & Faier-Routman, 1992) and young people experiencing JIA were included in this study. Furthermore, this meta-analysis identified that self-concept was poorer in children and adolescents with a chronic condition than individuals without, as measured by: behaviour, intellect, appearance, popularity, happiness and physical competence. These presentations could be seen as evidence for poor adjustment to a significant life change. Conversely, Ding, Hall, Jacobs and David (2008) found that young people diagnosed with JIA were no more likely to experience psychological difficulties than the 'normal' population unless their mobility was particularly affected.

The number of variables that contribute to how an individual adjusts is substantial; however, a number of risk factors for poor adjustment have been identified. For example, disease severity and progression have been correlated with poorer psychological well-being (LeBovidge et al., 2003; Billings et al., 1987). Billings et al. (1987) identified that greater levels of disease severity were correlated with higher levels of anxiety and depression in a sample of 43 young people with rheumatic diseases. Yeo and Sawyer (2005) posit that level of functional independence, and the degree to which individuals have contact with peer groups, may also impact on the adaptation process following the onset of a chronic condition. Following a meta-analysis of 38 child adjustment studies, conclusions were made as to what factors impact upon functioning and well-being. These included an uncertain prognosis, being diagnosed with an invisible and/or unpredictable condition and chronic pain (Patterson & Blum, 1996). All these elements are

characteristic of JIA symptoms, perhaps indicating that young people may be at increased risk of psychological difficulties. In addition to Patterson and Blum's (1996) suggested factors, other research looking at JIA populations also indicates that the levels of functional ability may also pose a particular risk factor for psychological distress, with more severe restrictions correlating with lower levels of psychological well-being (Ding et al., 2008; Timko, Stovel, Moos, & Miller, 1992).

Alternatively, not all young people diagnosed with a chronic condition report maladjustment and thus, poor adjustment is not inevitable. For example, LeBovidge, Lavigne and Miller (2005) found that for individuals who demonstrated a positive attitude towards JIA, levels of anxiety and depression were lower, and self-concept higher than the young people who did not demonstrate a positive attitude towards the condition. This could imply that individuals can draw upon their resiliencies at times of high stress. Coping styles for example, may be a protective factor for poorer levels of adjustment. Lazarsus and Folkman's (1984; Folkman, 1984) transactional theory of stress and coping has been widely used to describe adjustment and functioning following the diagnosis of a chronic condition. It is proposed that adjustment as an outcome is dependent upon primary and secondary appraisals and coping. Appraisals can be viewed as an individual's physical, social, psychological and material resources that can be drawn upon to manage a threat, such as that of a chronic condition. These appraisals then have implications for how one utilises relevant coping strategies (Lazarus & Folkman, 1984). Cognitive appraisals relate to the individual's appraisal of the stressor, their perceived control over the stressor and the degree to which they believe they have the resources to address or adapt to it (Lazarus & Folkman, 1984).

While it is important to understand adaptation and adjustment to a chronic health condition, the current literature exploring these concepts has its limitations, and this may lead to inconsistent findings. Firstly, many of the studies addressing this construct in young people do not use matched controls, nevertheless, when control groups are used as a comparison against the chronically unwell sample, fewer significant differences in well-being are demonstrated (LeBovidge et al., 2003). It is therefore difficult to determine if the relationship between a chronic condition and psychological well-being is determined by the chronic condition, or by other biopsychosocial factors unrelated to the chronic condition (Garstein, Short, Vannatta, & Noll, 1999). Secondly, the interchangeable and ambiguous definitions of adjustment and adaptation within the literature results in the use of a number of different measures to gather data. This can make it challenging to develop adequate comparisons of results and develop reasonable conclusions from the data (Boekaerts & Röder, 1999). Finally, a large number of individual adaptation and adjustment studies are based upon an indirect approach of reporting a young

person's well-being (for example, parental reports), as opposed to direct measures. It may be that children and adolescents have different thoughts on their adaptation process and can imply that young people's experiences are not as valid or as reliable as their parent's. In addition, the quality of the parent-child dyad might also impact upon what sense parent's make of their child's difficulties.

Chronic health conditions and the family

It is evident that the impact of a chronic condition does not occur in isolation (Robinson, Gerhardt, Vanatta, & Noll, 2007). For the individual with a chronic condition, adjustment, adherence and coping with new stressors, in part, is mediated by the family and other systems surrounding the young person in question (Robinson et al., 2007). Thus, the difficulties adapting to a chronic health condition, as experienced by each family member, or the family as a whole, could have a negative impact on the well-being of the individual with the chronic condition. The literature has consistently demonstrated that the family plays a critical role for the well-being of the young person (Varni, Wilcox, & Hanson, 1988). Specifically, family functioning is a significant predictor of a young person's adjustment to a chronic condition (Varni et al., 1988), condition management and illness-related quality of life (Botello-Harbaum, Nansel, Haiyne, Iannotti, & Simons-Morton, 2008). Drotar (1997) reviewed the literature from 57 studies addressing the adjustment of young people diagnosed with a chronic condition. Drotar (1997) found that in all but four of the studies reviewed, parental or family functioning related significantly to the young person's psychological well-being. Family cohesion and supportive familial relationships predicted better adjustment for young people, than families who reported low cohesiveness. Lower levels of maternal adjustment was most frequently identified as a predictor of poor child or adolescent adjustment, and is likely to be because of the reduced levels of social support, which has been found to buffer against stress (Varni, et al., 1988).

Conversely, Robinson et al. (2007), compared the functioning of 95 families of young people diagnosed with cancer, against matched controls. The authors found that the well-being of both mothers' and fathers' impacted upon the level of distress experienced by their child; that is, parental distress correlated with reduced well-being in the individual with cancer. This correlation however, was noticeable for both families experiencing a chronic condition and matched controls, suggesting that chronic illness may not always precipitate reduced family well-being, and other factors unrelated to the condition, may be involved.

It is evident that the family of a young person with a chronic condition can have some influence on how that young person adjusts, appraises and copes with a chronic condition; but this also means that a chronic health condition will inevitably impact upon other family members and

the family as a unit. As a result of the diagnosis of a chronic health condition, families may find their available resources depleted, thus rendering them vulnerable to poor adjustment (Patterson & McCubbin, 1983). Families may experience financial hardship should a parent need to take a leave of absence from work, to pay for medical equipment or have to fund home modifications. Families may also be affected by being unable to plan for the future, to be spontaneous in their everyday activities, or miss normative developmental milestones (Cohen, 1999).

Family members may also have different concerns about the condition which may impact on how they are affected by this. This could be as a result of the positions the family members hold within the unit and how they individually experience the condition. Konkol, Lineberry, Gottlieb, Shelby and Miller (1989) gave open-ended questionnaires to 50 families experiencing JIA. From these responses, they found that young people with JIA focused on the physical limitations of the condition, being different from their peers and their pain experience. Healthy siblings commented on how their parents treated them differently, the concerns they had for their siblings' condition, their relationship with their sibling and how this differed from their other relationships. Finally, parents focused mostly upon: their feelings of helplessness, concerns for the future, schooling, the stress of the condition, not knowing sufficiently about JIA, and the impact JIA had on family life. This study indicates that each individual within the family views the impact of a chronic health condition differently to one another, which may have implications as to how they adjust and communicate their fears.

Impact of a chronic health condition on parents and siblings

Parents

When a young person has been diagnosed with a chronic condition, it is often the parent's responsibility to manage treatment regimes and hospital appointments resulting in increased demands being placed upon them (Barlow et al., 1998; Wallander & Varni, 1988). Literature indicates that parents experience fundamental changes to their relationships with their children following the onset of a chronic condition. For example, Jordan, Eccleston and Osborn (2007) identified that a chronic condition can have a positive impact on the parent-child dyad, such as spending more time together to bonding and building stronger relationships; but can also have a negative impact, such as sensing the development of an overly enmeshed relationship with their child (Britton, 2006; Britton & Moore, 2002a, 2002b). This could result in reduced autonomy of the individual and perceived neglect of other family members.

Predominantly, parental literature has reported negative effects on parental well-being including: depression (Timko, Stovel, & Moos, 1992), marital dysfunction (Jordan et al., 2007),

grief (Jordan et al., 2007) and avoidant coping strategies (Jerrett, 1994). The distress reported may result from a variety of combined risk factors that occur when a family member has been diagnosed with a chronic condition. Wallander and Varni (1998) synthesised parental literature and identified factors that placed parents at particular risk of poor functioning, as measured by rating psychological well-being. Results identified correlations between parents' distress levels and the degree to which their child was physically impaired by a condition, the number of daily condition-related stressors experienced by the parents, the degree to which parents are able to problem-solve and levels of family support. More current research has also found that parents experience guilt, self-blame and frustration with medical regimes and treatment, impacting upon well-being (Barlow et al., 1998; Tong, Lowe, Sainsbury, & Craig, 2008). Parents with a child experiencing JIA report finding that they have to refocus their lives as their role shifts from a parent to a carer (Britton, 2006; Britton & Moore 2002a). Furthermore, mothers report becoming more serious due to their constant worries about JIA and its unpredictability. Mothers also found themselves becoming over-protective of their children and grieving as they experienced a sense of loss and sadness (Britton, 2006; Britton & Moore, 2002a).

Not all parents demonstrate poor adjustment to a childhood chronic condition. Horton and Wallander (2001) studied 111 mothers of children with chronic illness finding that a hopeful and a positive attitude buffered the impact of increased illness-related stress on the mothers, and prevented maladjustment. This may suggest that levels of resiliency, in the face of a chronic health condition, vary in mothers and might act as a moderating factor to heightened distress. Equally, these discrepancies across the parent experiential literature could also be due to the constructs that are used to measure well-being, for example, problem-focused versus resiliency-focused measures, or as a result differing samples, such as recruiting parents of children with the same or different health conditions.

Siblings

In addition to the impact a chronic condition can have upon parents, research suggests that siblings of children with a chronic illness may also experience difficulties as a consequence of the condition (Barlow & Ellard, 2006; Waite-Jones & Madill, 2008a). Factors as to why siblings may be vulnerable to psychological distress and reduced levels of well-being have been investigated. Siblings may experience a change in the family unit structure and may be required to undertake tasks that are disproportionate to their age or development (Coffey, 2006). For example, siblings may be required to take more responsibility within the household, or become carers for their brothers and sisters (Coffey, 2006). They may experience differential treatment from parents, in

comparison to their chronically unwell sibling, or be directly impacted by their parents' distress (Vermaes, van Susante, & van Bakel, 2012).

Research has been relatively consistent with regards to the psychological reaction of siblings to chronic health conditions. The majority of siblings exhibit psychological resilience and adjust well to chronic illness in the long-term (Houtzager, Oort, Hoekstra-Weebers, Caron, Grootenhui,s & Last, 2004), but, there is a minority population that may struggle to adjust (Houtzager et al., 2004; Sharpe & Rossiter, 2002). Vermaes et al. (2012) recently conducted a meta-analysis of 52 sibling-related adjustment studies. This synthesis, reported that siblings who demonstrate poorer adjustment were more likely to internalise or suppress their problems, as opposed to externalise them. This may be due to feeling that their parents were not able to attend to, or meet their needs (Vermaes et al., 2012). Siblings may therefore experience anxiety, depression, loneliness and low self-esteem (Houtzager et al., 2004). Furthermore, Vermaes et al. (2012) proposed that reduced parental interaction with the non-diagnosed sibling could result in the development of negative self-attributes, further impacting on their self-esteem. The age of the sibling and the severity of the condition may also impact upon adjustment, with both younger children, and siblings of individuals with more severe illness-related symptoms reporting more psychological distress than older siblings experiencing a less intrusive chronic condition. These results are contrary to those found by Houtzager et al. (2004) who found that older siblings reported more internalising behaviours and higher levels of anxiety as a result of both having a higher involvement in the illness process and having more knowledge of the condition than younger siblings. These discrepancies may be a result of the group of participants utilised in these studies. Vermaes et al. (2012) synthesised all chronic condition papers whilst Houtzager et al. (2004) focused only on cancer.

While it is important to acknowledge the impact a chronic health condition has on individuals, the condition will also have an impact upon the family in its entirety. Research on the impact a chronic condition has on the family will now be presented.

Impact of a chronic health condition on the family

In a review of the literature considering families experiencing chronic ill-health, Cohen (1999) identified several key areas in which a chronic condition impacts upon the family system. Firstly, families commonly experience higher levels of stress and distress after symptom onset, due to the added strain imposed on them by the condition. These stressors may impact upon the family structure and organisation (for example, alterations in boundaries and relationships between members) and generate a vulnerability to interpersonal conflict and psychological distress. Conflicts, for example, can arise between parents regarding caregiving and siblings may feel

excluded as a result of increased parental attention to their chronically unwell child. Secondly, depleted or strained family resources can impact both the chronically unwell individual and other family members but also upon interactions between family members. Cohen (1999) suggests that a combination of increased illness demands and reduced family resources can cause an imbalance to the family's equilibrium, impacting on their well-being. Finally, families may need to reconstruct meanings as a unit, in order to help them adjust to the condition collectively. Patterson and Garwick (1994, as cited in Cohen, 1999) propose that families utilise three different processes in order to develop meaning in their experiences that facilitates their response to a chronic condition. Initially, families construct and share their meanings regarding the stressors they experience and then they will attempt to build a new family identity that incorporates the chronic condition. The third level attends to wider cultural and systemic values that influence how the family manage the condition, such as to whom and how the condition will be communicated, who should be involved in decision making and who should care for the chronically ill individual.

The literature described above depicts just a few of the responses families have when faced with a diagnosis of a chronic condition. It seems that families' resources and relationships may become strained after the onset of a chronic condition although increasingly evidence also demonstrates that some families show great resiliency during adversity and work together to create new meanings for the family (Patterson, 2002a). In order to assist understanding about how families are impacted by a chronic condition and then how they begin to adjust to a new reality, it is useful to draw upon theoretical models.

Models and theories that can help explain family impact and adjustment to a chronic health condition

Bronfenbrenner (1986) argues family systems are bi-directional and multi-faceted, meaning that family level coping and adjustment at a time of instability is mediated by the other individuals within, and outside, the family. Based on this understanding, it may be important that clinicians are informed of family responses and interactions that may contribute to family level adaptation and maladjustment. Ideas that may be useful in understanding adjustment are the 'systems theory' (Von Bertalanffy, 1968 as cited in Kazak, 1992) and the 'resiliency model' (McCubbin, Balling, Possi, Frierdich, & Bryne, 2002; Patterson, 2002b).

The systems theory focuses on the context in which the child is currently socialised, which is influential in the adjustment process. Therefore, this theory encapsulates a dynamic quality within familial systems. Central to the theory, is a belief that systems are comprised of interrelated parts and so when a change occurs in one family member the entire system will be affected. The quality of relationships, the family's resources, prior understandings and experiences of illness,

roles and communication patterns both prior to, and after, the stressor all contribute to the adaptation process. The family's ability to alter these patterns in response to the stressor is critical to maintain the family's status quo (Kazak, 1997). This indicates that, in general, families and the wider systems around them continually construct and comprehend their experiences to find collective meaning and to restore a sense of homeostasis (Branstetter, Domian, Williams, Graff, & Piamjariyakul., 2008; Kazak, 1989). The systems theory also recognises that seemingly 'maladaptive' patterns observed in families, where a chronic condition is present, could be a strategy for maintaining this homeostatic stance (Kazak, 1989).

Relating to the systems theory, the resiliency model places emphasis and value on family strengths as opposed to family deficits (Patterson, 2002a; Patterson 2002b). The model illustrates that it is how family systems react under stress, that determines adjustment and adaptation of its members (especially the individual with the chronic condition), and the speed to which this is achieved. Families with high levels of resiliency adapt at a faster pace than those with low levels of resiliency. The model proposes that the more stressors and demands the family experiences, the more likely that adjustment will take a negative course. Moreover, families with successful histories of coping under stressful conditions, with low levels of anxiety, fare better than families without accessible resources (McCubbin, et al., 2002). This model also suggests that a family will seek a shared perspective between members, to give the family a meaning and purpose to move forwards. These family meanings interact with the demands and capabilities of the family to reach a level of adjustment (Patterson, 2002b). Families' capabilities include psychosocial resources the family possesses and what they do to cope with the demands; these arise from individual family members, the family as a unit and wider systems (Patterson, 2002b).

These theories and models demonstrate that the family in its entirety will be affected by a diagnosis of a chronic condition in a young family member. The resiliency model may help clarify why it is that families experiencing chronic a chronic health condition may be more vulnerable to poor functioning, that is, because they are negotiating a number of added stressors in addition to normative stressors that other families may experience. The impact of a diagnosis may not, however, be a negative one if families feel that they have good resources prior to the onset of a chronic condition and that they have the abilities to quickly adapt themselves in uncertain situations. This will consequently impact on the long-term adjustment of a chronic condition. Systems theories however have been criticised for negating individual experience (Kazak, 1989).

Impact of a chronic health condition upon family functioning

As discussed, a chronic condition will impact upon the family as a unit and the extent of the impact can be dependent on the family's resources and methods of coping with added stressors and demands (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010). This inevitably means that how a family functions and consequently adjusts post diagnosis will alter as a result of a change in its system. Cohen (1999) posits that the family system as a *whole* is the most significant factor in adjustment to a chronic condition. Lewandowski et al. (2010) reports that family functioning encompasses the social and structural properties of the family environment, that is: the interactions and relationships within the family[4], levels of conflict and cohesion, adaptability, flexibility, organisation and the quality of familial communication. All or some of these facets can be affected in families experiencing chronic illness (Patterson, 2002a; Lewandowski et al., 2010).

There is a lack of consistency of findings across the research literature as to whether families are prone to maladjustment and poorer functioning following a diagnosis of a chronic condition in a young person (Reisine, 1995). Lewandowski et al. (2010) for example, reviewed 16 studies that have investigated the functioning of families of children and adolescents experiencing chronic pain[5]. The review yielded mixed results; and four of the seven studies measuring family functioning in families with an adolescent diagnosed with a chronic condition (as opposed to children) found that these families demonstrated poorer functioning than comparative health controls. Furthermore, family functioning was poorer if the families were coping with higher levels of pain related disabilities. However, the cause-effect relationship cannot be made clear from this cross-sectional analysis; it could be the case that pain related disability resulted in poorer family functioning as opposed to poorer family functioning impacting upon a young person's level of functional ability. Conversely, one study in this review of studies demonstrated that greater experiences of pain in the chronically unwell sample correlated with *better* family functioning. The authors suggest that higher levels of experienced pain may bring families closer together and enable parents to be more responsive to their child thus increasing reports of positive well-being (Lewandowski et al., 2010).

Knafl and Gilliss (2002) also found mixed results when synthesising 73 studies of family functioning research. Results ranged from families that continued to function as they did prior to the onset of symptoms, to families who reported that chronic health condition had a negative impact on them and their functioning. Families reported reduced levels of functioning during times of transition, for example, at the diagnostic stage or moving from hospital to home. In support of

[4] Especially that of parent-child dyads.
[5] Pain is frequently a significant and disabling symptom of JIA.

the family resiliency model, these authors found that the greater the number of stressors the families experienced, the more likely that these families were to report reduced levels of well-being. This may provide an explanation as to why some families do not appear to be adversely affected by a chronic condition and others do. The reviewed studies also indicated that over time, families may learn to master treatment regimens and incorporate these into their everyday lives resulting in better family functioning in the long-term. Jerrett (1994) proposed that families experiencing JIA move through stages of initial shock and confusion, to taking charge of the situation and becoming experts in managing the condition. This may also support the 'systems theories' of family functioning in that families will adapt to their circumstances to regain a sense of balance in their lives.

Similar to research on the impact of a chronic health condition on an individual, it appears that families may also be vulnerable to poor adjustment during a time where there are added burdens or stressors to contend with. It seems however, that families may have protective factors or unique qualities that can reduce their vulnerability to the negative impact of a chronic health condition. Furthermore, there is much heterogeneity of disease factors and family factors that may also contribute to a lack of consistency within the literature, and it may be that more in-depth studies studying the idiosyncrasy of experiences can further enhance the family functioning and adjustment evidence-base (Varni, Katz, Colegrove, & Dolgin, 1996).

Factors that affect family functioning

Research is emerging as to what factors may contribute to families reporting positive or poor adjustment following the diagnosis of a chronic condition in a child or adolescent (Varni et al., 1996). In line with the family resiliency model, factors such as high family cohesion and expressiveness (including high levels of intra-familial communication) have been found to be good predictors of family functioning, positive adjustment (Varni et al., 1996) and reduce the family's vulnerability to the negative impact of a chronic health condition.

Olson's (1993) 'circumplex model' proposes that flexibility and family cohesion are essential to family functioning. Flexibility refers to families' ability to adapt to changing roles, relationships and structure during times of stress. Families who are high in flexibility respond quickly and easily to environmental demands. Families who struggle to adapt, their behaviours remain fixed and constant despite environmental changes. Olson (1993) argued that flexibility is on a continuum, with very low levels of flexibility (i.e. rigidity) and very high levels of flexibility (i.e. chaotic) being least functional, as these families are unable to change behaviours to adequately manage demands. Cohesion, relates to the emotional bonds within the family, and how much time members spend with one another. Very high levels of cohesiveness (i.e. enmeshed) and

very low levels (i.e. disengaged) are considered to be least functional for families and thus, a moderate level is considered to be optimal for family functioning. Positive communication patterns between family members is said to facilitate flexibility and cohesion during these times of stress, such as that of managing a chronic health condition; additionally it can facilitate adaptation and adjustment. Attentive listening, empathy towards other family members and willingness to self-disclose are examples of positive communication skills. Less positive communication may involve criticism, excessive conflict and failure to listen (Olsen, 1993). The two constructs of cohesion and communication, which have yet to be outlined, will subsequently be discussed in relation to the chronic health literature

Family Cohesion and functioning

The literature indicates that family cohesiveness and the level of support care-givers and families provide to those affected by a chronic condition, can mediate the psychosocial adjustment of an individual to the chronic condition, regardless of disease severity or symptoms (Wallander & Varni, 1988). Adolescents with a chronic condition have reported that family cohesion is the most salient factor in well-being and maintaining low levels of condition-related stress (Salewski, 2003). The psychosocial well-being of the other family members can also be affected by levels of family cohesion (Gerhardt et al., 2003). Family cohesion can be measured using constructs such as: family support, affection, levels of interaction and nurturance (Helgeson, Janicki, Lerner, & Barbarin, 2003) and it can be employed as a method for measuring family adjustment to a chronic condition.

Research addressing family adjustment and psychological functioning, following diagnoses of childhood chronic conditions, makes comparisons with healthy control groups (for example, McClellan & Cohen, 2007). McCellan and Cohen (2007) conducted a critical review of family functioning studies, making comparisons across diagnoses in order to determine variables that promote positive family functioning. They identified that families with a member diagnosed with Cystic Fibrosis (CF) experienced low levels of familial cohesion and communication, which resulted in low rates of affect management, higher levels of stress, when compared against a control group. This study also reviewed families experiencing diabetes, and parents reported spending less time with their healthy children, which could have an impact on the levels of cohesion within the family. Families with a child diagnosed with JIA were not dissimilar in their functioning to those with healthy children in this study. However, one significant difference within the JIA sample was the greater levels of cohesion within families with children (as opposed to adolescents) with JIA. This difference may reflect the developmental trajectory of a young person,

and it appears that more conflict and less cohesion arises as children become adolescents, where autonomy and individuation from parents are important milestones (Grotevant & Cooper, 1986).

Reisine (1995) synthesised family functioning research with particular attention to studies focusing on juvenile arthritis. The author found that when controlling for disease severity, families experiencing higher rates of cohesion and fewer stressful events, reported lower levels of behaviour and adjustment problems and also reported fewer condition-related symptoms. Moreover, parents and siblings reported that disrupted family relationships and emotional distress were stated as being the most significant condition related burden, indicating that for this particular sample, it was not the condition that directly impacted on the well-being of individuals within the family, but rather it was how the condition changed intra-family interactions.

Strong intra-familial bonds are also likely to increase the amount of positive and supportive communication between family members, and this may encourage families to be able to share and construct the meanings created by their experiences as a family (Miller, 2009). Theoretically, it has been argued that social support buffers individuals from potentially stressful and negative life events ('the buffering effect' model; Cohen & Wills, 1985) and creates an environment in which individuals will draw on his or her resources to promote coping in order to manage the stressor (Varni et al., 1988).

Equally, a lack of family cohesion has been correlated with poorer levels of family functioning in families experiencing JIA (Helgeson et al., 2003). Reduced levels of cohesion may encompass family expression of anger, hostility, criticism and conflict (Helgeson et al., 2003). In a sample of 94 children and adolescents with JIA, family conflict was related to greater illness worry, more worries about physical appearance and lower self-esteem (Helgeson et al., 2003). Furthermore, age appears to be a mediating factor for reported family cohesion. In this study, Helgeson et al. (2003) found that older individuals with JIA (adolescents as opposed to children) reported less family cohesion. These are similar results to those found by McCellan and Cohen (2007). The authors suggest that adolescents, as they develop, rely less on parents for support and rely increasingly on their peers, potentially reducing levels of familial cohesion. These results indicate that it may be important to make distinctions between children and adolescents when understanding family resilience and risk factors relating to adjustment to a chronic condition. The sibling research presented earlier also indicated age related adjustment differences (for example Vermaes et al., 2012).

Family communication and family functioning

Research investigating the positive effects of good quality family communication following a diagnosis of a chronic condition, appears to be more consistent than studies measuring a wide array of adjustment and adaptation constructs. Poor communication within families is correlated with greater psychological distress (Wallander & Varni, 1988) and poorer pharmacological adherence (Wiebe et al., 2005). Parents and chronically unwell children who report positive collaboration in condition management also report better psychological well-being and closer familial relationships, as compared to families where parents take a more controlling stance to management (Miller, 2009; Wiebe et al., 2005). This indicates the importance of establishing a sense of collaboration and expressiveness within the parent-child dyad, rather than an authoritarian relationship, where the young person is passive to their care. Waite-Jones and Madill (2008b), in a qualitative study found that family members can find communicating their feelings about the condition to other family members difficult. For example, fathers of children with JIA reported concealing their distress from their families which they felt constrained levels of communication between them and other family members. In addition, these authors found that healthy siblings reported that they did not want to express their emotional needs to their families, resulting in feelings of isolation (Waite-Jones & Madill, 2008a).

Orbuch, Parry, Chesler, Fritz and Repetto (2005) offered parent-child relationship questionnaires and quality of life questionnaires to 190, 16-28 year olds who were long-term survivors of cancer. They found that survivors who rated themselves as having a more positive relationship with their parents also rated themselves as having a better current quality of life in psychological (but not physical) domains, than those who rated lower on the relationship questionnaire. The authors conclude that positive parent-child dyads during a period of chronic illness can foster a greater number of positive outcomes for adolescents who may otherwise be susceptible to psychological distress. Positive mother-child dyads were related to an increase in overall quality of life; and positive father-child dyads were more highly correlated with psychological and spiritual well-being. This may suggest that family members contribute something unique to their relationship with the adolescent with a chronic condition that promotes adaptation and a better quality of life. These results are in accordance with the family 'resiliency model', which postulates that family strengths can promote positive adjustment to chronic conditions. There are, however, limitations to this study. Firstly, 493 participants were originally contacted to take part in the study and of these, 158 participants were either deceased or had moved house and 145 did not return the questionnaires. It may be that the sample was biased towards reporting only positive outcomes or, participants who had better relationships with their parents or a higher quality of life were more willing to participate in the study. Secondly, the

authors do not report on their participant demographics. It is possible that families with higher socio-economic status, who potentially have more resources available to them, demonstrate higher levels of familial cohesion or reduced stress (Hagan & Smail, 1997).

Miller (2009) studied shared and collaborative decision making in families with children experiencing diabetes, asthma and CF. Shared decision-making may foster independence from the family, as those with chronic conditions may develop the confidence and self-esteem to make their own treatment decisions which might improve or maintain positive family relationships. Miller (2009) conducted individual interviews and focus groups with parents and young people with a chronic health condition. Children and adolescents stated they were less likely to be actively involved in future decision-making and felt less in control of their condition when parents neglected to involve them in condition related decisions. Furthermore, children and adolescents reported they were less likely to share information regarding the condition if they believed their parents would respond in an anxious manner. Miller (2009) proposed that this may communicate to children that their opinions are not valid or that they do not have a meaningful voice within the family. Parents reported benefits in shared decision-making as both parties would be better informed of each other's understanding of the condition and that they were offering their children a sense of freedom that may already be restricted by the condition. Parents also felt they were providing the opportunity for their children to learn effective problem-solving behaviours and gain knowledge about decision-making. Miller (2009) suggested that decision-making of this kind offered the family opportunities to interact around chronic condition management and higher levels of interaction could mean that all family members have the most recent information about the condition to provide the most effective treatment. This research suggests that family members can see positive benefits from shared decision-making, however this may not always happen if members predict negative consequences from doing so.

Family shared understandings and developing meanings

Within the chronic health field, developing understandings or meanings about a health condition may include beliefs about its consequences (the impact of the condition upon everyday life) and beliefs about the significance of the condition (what the condition means to the self and others) (Bury, 1991). High levels of familial cohesion and communication may enable individual family members to attain a greater understanding of one another's experiences and beliefs about a chronic condition. Integration may encourage the development of shared understandings as part of the adaptation process. This idea was proposed by Blumer (1969, as cited in Segrin & Flora, 2005) as part of the 'symbolic interaction theory' which argues that shared meaning is generated and modified through interaction with other family members. Evidence of families' shared

understandings is limited and mixed within the family literature. Olsen, Berg and Wiebe (2008) investigated the beliefs that mothers and their adolescent children had about diabetes and found that mothers and adolescents did not view the chronic condition in the same way. Mothers believed the condition to be more chronic and having a greater degree of emotional impact than adolescents. Differing understandings of diabetes did not impact upon adolescent adjustment but had some impact upon maternal well-being. Mothers who believed they had a greater understanding of diabetes than their children reported negative adjustment. No further beliefs about diabetes predicted negative adjustment for either the adolescents or mothers. It may be that different perspectives and unique experiences of the condition may result in discrepant understandings between family members; or, that it is less about sharing perspectives of the chronic condition, but how that sharing of information is communicated.

Salewski (2003) employed quantitative measures with 30 family members of children diagnosed with a chronic skin disorder and assessed the extent to which family members had similar 'illness beliefs' (timeline, causation, curability, control, identity and treatment) about the skin condition. The author found that if parents and their children shared similar beliefs about the condition, then adolescents reported higher levels of well-being than adolescents who do not share similarities with their parents. Adolescents however did not demonstrate lower levels of illness-related stress despite similar illness beliefs to their parents. Furthermore, adolescents rated families as cohesive if their parents shared similar beliefs about the condition. Salewski (2003) suggested that shared family beliefs are important for adolescents, despite the growing autonomy.

Critique of family research

Within the family adjustment and functioning research, family outcome data demonstrates huge variability in reactions to a chronic health condition, both within and between conditions. Research is inconsistent and conclusions vary from families demonstrating poor adjustment to chronic health conditions, to families reporting no impact and family functioning at levels comparable to families not experiencing chronic health condition. It may be tentatively concluded that families are vulnerable to psychological distress and maladjustment, which may, in turn, have an impact upon the overall functioning of the family. Inconsistencies in the data are perhaps a result of the idiosyncrasy of family interactions and dynamics, but also as a result of methodological limitations (Resine, 1995). Limitations include: non-representative or small samples of participants, broad range of ages, assessing more than one condition in any one study and using a wide range of assessments to evaluate and measure psychological distress. Additionally, it is not possible to account for all variables that contribute to family functioning in response to a chronic condition due to the heterogeneity of conditions and families, and thus studying smaller samples may yield

results that can elicit these idiosyncrasies. For a better understanding of presenting behaviours, researchers would need to account for: developmental stages of the families they are studying, all variables in relation to the condition, family demographics, the families' history of stress and coping, cultural and socio-economic variables and levels of social support (Mussatto, 2006). It is unlikely that a study will be able to control for all these variables, so more in-depth investigations into families and understanding not only the similarities in experiences, but differences as well, may offer support to some of the findings presented in this review.

Kazak (1989) has also stressed the importance of taking into account the advances in medical treatment when assessing adjustment and functioning of individuals or families. The author asserts that caution should be taken when interpreting the results of this evidence-base, especially when making comparisons between older and more recent studies. For example, Kazak (1989) hypothesised that access to more advanced treatment, could reduce the impact of a chronic health condition on daily life, resulting in a larger proportion of more recent research reporting non-significant findings between these individuals or families and matched controls. This may be especially pertinent with JIA where early aggressive treatment is essential to prevent long-term disability and pain. Furthermore, better access to psychological treatment in recent years may also aid the adjustment of families (Vermaes et al., 2012). This could result in older studies becoming less relevant to the literature base. Conversely, Vermaes et al. (2012) argue that more recent medical and psychological treatment is now more intrusive to families than it was in the past, for example, families are often expected to adhere to strict daily regimes and are regularly monitored. This may have consequences for family resources and work implications for parents.

In addition, research that has incorporated matched control groups into their design has often yielded data that indicate that families who demonstrate poorer levels of cohesion, function less well regardless of whether there is a child in the family who has a chronic condition or not (Patterson, 2000a). It may be the case that families who do struggle to adapt to a chronic condition may have had particular risk characteristics prior to the onset of a chronic condition that predisposed them to be susceptible to the added stressors (Mussatto, 2006). Moreover, studies only tend to report on 'positive' or 'negative' cohesion, without attention to the finer details of family interactions. Whilst family cohesion is generally considered to be a good indicator of positive family functioning, there is a paucity of research regarding family interactions within the chronic health literature.

Finally, there is a paucity of research looking at how families negotiate and develop shared meanings following the onset of JIA. Past research, therefore, has tended to address family functioning as an outcome as opposed to a process. Outcome measures of family functioning have been reliant on parental, especially maternal, reports (McCellan & Cohen, 2006), with fathers

often under-represented (Garhardt et al., 2003). While parental reports add value to the understanding of the adaptation processes, neglecting other sources of information may result in uncertainty about how families as a unit adapt to diagnoses of a childhood chronic condition (Garhardt et al., 2003). Moreover, utilising parental reports alone may also neglect the complexities and multifaceted units of family interactions, losing important information about families.

Justification for the present study

There is still much to learn about the family in its response to adapting to a chronic condition as a process as opposed to an outcome. To the best of the researcher's knowledge, only three qualitative studies to date have addressed family experiences of JIA utilising more than one family member's perspective (Waite-Jones & Madhill, 2008a, 2008b; Britton, 2006; Britton & Moore, 2002a, 2000b; and Rossato, Angelo, & Silva, 2007)[6]. All three studies utilised a grounded theory approach to their methodology and developed themes relating to family experience following the onset of JIA in a family member. The papers published demonstrate a degree of quality and rigour, for example, Britton utilised a number of different methods of collecting family experiential data, such as observation, diaries and semi-structured interviewing. These studies provide in-depth information from a number of sources within the same family, however, at the same time, these papers fail to acknowledge that families' experiences as a unit, and how these are talked about, could be different from the individual's perspective within the family, which may be important to gaining further in-depth understanding of family experiences as opposed to individuals within families. As outlined within the literature review, discrepancies in beliefs or perspectives between family members may cause a breakdown in family functioning which may impact upon psychological well-being of family members. Rossato et al. (2007) interviewed some of their participating family members together to elicit family experiences. These authors identified some interesting experiential themes such as: struggling with the symptoms of juvenile arthritis and trying to lead a normal life. However, despite these joint interviews, Rossato et al. (2007) fail to analyse shared and unshared narratives within the family and between family members. An additional point is that Rossito et al. (2007) were not explicit about how they analysed the data from their group interviews and how this different from their individual interviews. This may have implications for the validity and quality of the study. To date, and to the best of the researcher's knowledge, no research has both looked at family experiences of a chronic condition from a multi-perspective viewpoint and whilst also paying attention to the processes by which families negotiate their experiences as a family and with one another.

[6] Waite-Jones &Madhill and Britton published multiple papers from the same participant data.

Furthermore, the majority of studies aimed at further understanding family responses to a chronic condition in a member, have been quantitative in nature. This means that large samples are utilised and the uniqueness of families can be lost. In addition, due to the small numbers of young people experiencing JIA samples are often heterogeneous which may have contributed to mixed results obtained from family adjustment studies. It is therefore worth paying attention to the complexities of inter-relational interactions that may both contribute to families' adjustment processes and further understand the unique ways in which families will make these adjustments.

Based on the current JIA literature, it is hoped that studying the family as a unit (as opposed to individual members) should offer something unique to the chronic health condition literature. This is especially the case for studying how families negotiate and share their understandings which has not yet been investigated in families experiencing JIA. The results of this present study may also be of interest to paediatric rheumatology services that have regular contact with families adjusting to JIA. If family experiences are better understood, then this may have implications for recognising if families are struggling with their adjustment to a chronic health condition. Early recognition may also have resource implications for health services.

Summary

In summary, there appears to be some evidence that poor psychological adjustment in family members results from a multitude of stressors associated with diagnoses of chronic health condition (McClellan & Cohen, 2007). Factors hypothesised to aid positive adjustment are family cohesion, family communication and sharing their understandings about the condition. Increased cohesiveness may be as a result of positive and supportive communication between family members; which in turn, may result in families sharing their experiences and understandings of the chronic condition with other family members. As a consequence it could be that this experience of sharing may act as a buffer to increased rates of distress and negative affect (Salewski, 2003). Family functioning is often used as a measurement for adjustment, however, little is known about how families as a unit experience a chronic health condition, and about the processes by which families negotiate their experiences and understandings of JIA. Research in this area is useful for clinical application; especially when endeavouring to understand more about families who exhibit poorer levels of communication and discord. This research may help contribute to the growing literature aimed at developing family interventions for those experiencing difficulties adjusting to a chronic health condition.

Aims of the present study

The aim of this study is to:

1. Explore familial understandings of JIA following a diagnosis of the condition in a young family member.

2. Explore how these understandings are negotiated and communicated among family members.

CHAPTER TWO

Design and Methodology

This chapter will outline the rationale for the methods and procedures employed to gather and analyse the obtained data. This chapter will also provide a brief summary of the quality checks undertaken to enhance the validity of the study and a reflexive statement from the researcher.

Research design

This study has utilised a qualitative multiple case study research design, employing a mixed method approach to data collection and analysis. Face-to-face semi-structured interviews were conducted with two families, using both family group interviews and interviews on a one-to-one basis. This was with an attempt to elicit family experiences and understandings of living with JIA, from a multi-perspective view-point. This was thought to be the most suitable approach to elicit family experiences of a chronic health condition and to respond to the study's research aims.

Methodological considerations

A qualitative research approach offers researchers an opportunity to explore how people manage and experience a phenomenon by obtaining rich detailed idiographic accounts (Willig, 2008). In addition, qualitative research is less concerned with obtaining high volumes of participants in order to determine causal relationships (Willig, 2008) but instead aims to explore the how and the why. In psychology, qualitative research is concerned with study of how people make sense of their unique lived experiences and the meanings that they attribute to these experiences (Willig, 2008).

Case study research

Case study research offers the opportunity to understand a phenomenon in-depth and often uses more than one source for evidence (Willig, 2008). A case study is defined as a focus upon a particular unit of analysis, as opposed to the method used to collect data (Willig, 2008). Common elements to a case-study design are that they are idiographic, they pay attention to context and interactions, they use triangulation therefore integrating different sources of information and/or analysis, they focus on change and they facilitate theory generation (Willig, 2008). An obvious limitation of a case study approach is its limited ability to generalise results to wider populations; however, case studies still retain the ability to generalise by influencing existing theory and can also be utilised to complement experimental data (Yin, 2009). A further limitation is that, historically, case studies have not always demonstrated rigour, making it difficult to utilise the findings. The present study, however, can demonstrate rigour, by utilising methodological

triangulation and quality checks. Within the present study, multiple cases were analysed (i.e. two families) and then a single set of cross-case themes was identified in order to draw conclusions.

Mixing methods

A triangulation approach was utilised in the present study, mixing (qualitative) methods of data collection that increased the breadth and depth of the generated research data (Moran-Ellis et al., 2006). In the present study, family group interviews and subsequent individual interviews with the family members were conducted. It is argued that a triangulation of both group and individual interview data allows for a greater understanding of the phenomenon under investigation (Lambert & Loiselle, 2008). In addition, it has been argued that increasing the breadth and depth of the data through multiple ways of data collection and from multiple perspectives, may enable the researcher to draw more reliable or valid conclusions of the data obtained due to approaching the phenomena from a number of different angles (Eisikovits & Koren, 2010; Lambert & Loiselle, 2008; Larkin, Flowers, & Shaw, 2013). A rationale for using these two approaches will be discussed at a later stage.

Family Group Interview

The present study is concerned with understanding the recruited families' experiences of JIA as a unit, and therefore it was deemed appropriate to interview the individuals within each family together. The use of group interviewing is increasing in popularity in qualitative research, especially within the health psychology research field due to their utility in providing multiple perspectives at one time (Willig, 2008). In addition, group interviews are an opportunity to bring together experiences, thoughts, feelings and behaviours about a given phenomenon (Morgan, 1998; Fern, 2001). Furthermore, a group interview approach was utilised in this present study as it offered the opportunity of eliciting rich data about the complex interactional processes between family members (Willig, 2008). This was particularly important in order to respond to the research aims.

The role of the researcher in a group interview is to ask questions from a pre-prepared topic guide, to elicit information from the members, but the researcher will also act as a facilitator who will guide participants' discussions and encourage interaction between group members. In doing so, important information regarding differences and similarities between individual family members' experiences can be elicited from the process (Lambert & Loiselle, 2008). This was particularly important for meeting the aims of this present study. Koeing-Kellas and Trees (2006) argue that eliciting data about familial narratives can produce important information regarding

family norms, identities, and functions, which may be difficult to draw out from other forms of interviewing.

A further benefit to utilising a group interview approach is that more than one point of view can be explored at one time. A chronic condition does not merely affect the individual diagnosed, but also impacts on family, friends and wider systems. It is therefore important not to negate other people's perspectives that will impact on how sense is made of experiences, especially when these experiences have been shared (Eisikovits & Koren, 2010). As discussed in the previous chapter, research exploring families as a unit within the chronic health field has yet to be thoroughly investigated.

A further advantage of group interviewing can be linked to observing different perspectives as they operate within a particular context, and therefore, members are able to investigate how their thoughts are similar and different from one another (Kitzinger & Barbour, 1999). However, Eggenberger and Nelms (2007) propose that family group interviews are most effective if the researcher takes care to include all family members, demonstrating an absence of bias towards particular members or narratives; particularly when disagreements between family members occur.

While a group interview design was utilised, it is not without its limitations. It can be argued that some accounts may not be spoken about, or may become lost among the stories offered by more dominant, extroverted or powerful group members (Barbour & Kitzinger, 1999; Michelle, 1999). Furthermore, participants may be more likely to offer socially desirable descriptions if they feel they will be judged by the other group members (Morgan, 1998). These limitations are particularly pertinent in the present study, as the group interview consisted of members who belonged to a pre-existing group. Pre-existing groups will tend to have naturally occurring power hierarchies which may silence some members, resulting in a bias within accounts offered. It could be argued that parents are considered powerful members within the family unit and, therefore, younger members may feel they are less able to voice their thoughts. This may be especially the case for siblings or the youngest child.

As a final point, there is debate in the literature regarding the suitability of using group interviewing techniques for investigating sensitive topics. The prevailing argument might suggest that a group situation could lead to higher levels of distress for those who may be asked to disclose sensitive information about themselves in the presence of others (Willig, 2008). This could have potential ethical implications, or the silencing of some accounts. Conversely, other researchers have argued that sharing experiences can be empowering for group members, which, as a consequence, may lead to a cathartic process for the members concerned (Farquhar, 1999).

Semi-structured interviewing

To produce qualitative research, it is important that the data obtained is comprehensive and sufficiently in-depth to gain a sense of how people attribute meanings to their experiences. Semi-structured interviewing is a popular method in which to obtain this in-depth data as it offers sufficient flexibility in order for the researcher to be guided by the participant's storytelling (Smith, Flowers, & Larkin, 2009; Willig, 2008). In addition, semi-structured interviewing enables sufficient flexibility so that further questions can be formulated during the interview process, based on information offered by the participant. Interviewing techniques such as prompting and probing are methods used by the researcher, in semi-structured interviewing, which are designed to guide the participant to further descriptions of their experiences (Smith et al., 2009).

Individual Interviews

In order to address some of the limitations for utilising a group interview approach, individual interviews were also conducted. Individual interviews have the benefit of obtaining rich in-depth information from one key person via one-to-one exploration of experiences (Lambert & Loiselle, 2008; Willig, 2008). The individual's viewpoint, beliefs and experiences, about the phenomenon under investigation, can be investigated in relative isolation from the influence of other family members, however, it is still recognised that individuals may provide socially desirable responses. The individual may also have more opportunity to describe their accounts without having to negotiate other conversations. While the researcher will inevitably have an influence on the interview process, as a result of the increased levels of interaction from the researcher, a participant may feel less restricted in what they discuss, in a one-to-one context (Willig, 2008).

Data collection sequence

In the present study, individual family member interviews were conducted following the family group interview. There were several reasons as to why this was considered the most appropriate sequence for data collection, Firstly, the research aims were to explore the family experiences of JIA and therefore the family experience, as opposed to individual experience was privileged by interviewing the unit first. Secondly, the individual interviews were then used to supplement and further explore accounts and stories that were discussed in the family interview and to allow individuals to discuss any experiences that had not arisen in the family interview. Lastly, it was thought to be more ethical to conduct individual interviews last, so that the researcher did not have access to any individual accounts during the family interview; this may have resulted in uneasiness for both the participants and the researcher

Methodological considerations to data analysis

There are a number of ways to analyse the data obtained from utilising a qualitative approach to data collection. The method chosen to analyse data can be dependent upon factors such as this study's research aims, however, qualitative data analyses share some salient features. These features include the researcher needing to interpret and make sense of the data and the fact that most approaches are bottom-up process of analysis (Miles & Huberman, 1994). Qualitative methods include thematic analysis (Braun & Clark, 2006), grounded theory (Glaser & Strauss, 1967), narrative analysis (for example, Crossley, 2007) and IPA (Smith et al., 2009). It was felt that IPA was the most suitable approach to use to answer the research aims of this study. The IPA approach will be outlined in the following section.

Interpretative Phenomenological Analysis

This study has utilised an Interpretative Phenomenological approach to data analysis (Smith & Etough, 2007). Interpretative Phenomenological Analysis (IPA) uses an inductive approach to data analysis which generates summaries, patterns and themes from the available raw data (Thomas, 2006). It is a method of analysis that is concerned with utilising in-depth explorations of subjective personal accounts of phenomena, in an attempt to understand lived experiences (Smith & Eatough, 2007). IPA views participants as experts in their own experiences and the approach can help to identify how people make sense of their personal and social world.

IPA has a number of theoretical underpinnings: phenomenology, hermeneutics and idiography (Smith et al., 2009). IPA is phenomenological as it aims to explore the subjective experience of a person or group of people as the experience is lived, and how sense has been made from their experience (Smith et al., 2009). IPA therefore, is interested in what aspects of experiences are particularly important to an individual, which then in turn, influences how such individuals make sense of this experience (Smith et al., 2009). Husserl (1927) argued that the way in which experiences are interpreted, for example, by researchers, can be influenced by their own pre-existing knowledge or understandings of that given phenomena, modifying that subjective experience. To prevent modifying or misinterpreting the lived experience, Husserl indicated that phenomenology involved bracketing off pre-existing assumptions in order to look at a phenomenon as it is in its own right. Pre-existing assumptions for a researcher may include prior knowledge of a phenomenon through the reading of relevant literature, personal experience and personal values and/or knowledge of theories or models relating to the area of interest. To minimise this effect, Husserl (1927) endorsed adopting a reflexive stance whereby attention is focused inwardly to identify those assumptions that we are then able to set aside. Bracketing is currently considered an important component to the analysis process; however, Heidegger (1962)

implied that prior fore-structures cannot be bracketed off. Instead, Heidegger suggested that those attempting to study another's subjective experience should acknowledge assumptions and not attempt to disregard this knowledge all together. Heidegger (1962) also discussed the concept of *intersubjectivity* which refers to the inability to step out of an experience due to the 'shared, overlapping and relational nature of our engagement in the world' (Smith et al., 2009, pp. 17). For a researcher, this means that it is important to acknowledge that the research cannot be completely removed from the data or participants with which she or he is studying.

IPA is interpretative and draws upon the theory of hermeneutics. As individuals we are constantly and actively making sense of our experiences through interpretation of events and actions. In qualitative data analysis this results in a double-hermeneutic whereby the researcher's central role in the analysis process is to make an interpretation of the participant's interpretations of their lived experience. In other words, the researcher attempts to make sense of their participants' world as accurately as possible as to how the participant had perceived and made sense of it, at the same time as making sense of their own experiences (Smith, 2004). Therefore the interpretative nature of IPA embroils a subjective and reflective process (Reid, Flowers & Larkin, 2005). It is also worth noting that, in the present study, there is the additional hermeneutic of participants making sense of one another's experiences during the interview group interview.

IPA is idiographic and places emphasis on individual and unique experiences; it is less concerned with hypothesis testing and moves away from the nomothetic (Reid et al., 2005). An idiographic approach additionally views an individual as set apart from other individuals. This means that during analysis, the data is looked at on a case-by-case basis, and only when analysis has been conducted, will a synthesis of the data occur. In the present study, a 'case' equated to the family as a unit and therefore, both a within analysis and a between analysis was conducted.

Rationale for IPA

IPA (Smith & Etough, 2007) was considered to be the most appropriate method of analysis. This is because the way families develop, share and communicate their understandings of JIA will be shaped by how individual family members have made sense of their experiences.

IPA is considered to be well suited to gaining access to people's experiences in health, social and clinical fields where there is a need to understand how people make sense of significant events (Smith & Eatough, 2007). Additionally, a primary aim of IPA is to build on existing psychological research (Smith, 2004). Utilising an IPA approach is of increasing interest to the NHS, as in recent years the NHS has placed emphasis on hearing service users' idiosyncratic experiences including experiences of well-being and resilience as opposed to purely illness and deficit. This is in order to move away from a top-down model of care and utilise more patient-

centred informed practices (Reid et al., 2005). IPA can be used to gain an understanding of the commonalities and differences between a specific group of individuals with similar experiences through the integration of themes, but also preserving the narratives of the participants at the same time. IPA is well suited to small sample sizes and places emphasis on unique experiences and idiosyncrasies (Smith et al., 2009).

Furthermore, in recent years IPA has also been used to analyse data gathered from group interviews and from multiple methods of data collection (Smith et al., 2009). Due to its flexible approach to analysis, IPA is suited to analysing data of this kind. Smith et al. (2009) also stress that IPA must be used flexibly in order to get the most from the data. However, they do recommend some caution using IPA to analyse group data because individual meanings and sense making can be lost among multiple voices. The aim of the present study was to address understanding and making sense of JIA within the family, and so the family as a unit was be the focus of analysis, as opposed to looking at individual experiences. Palmer, Larkin, de Visser and Fadden (2010) argue that IPA can support this approach because *relatedness'* is a central concept to the model.

Alternative methodological approaches

Grounded theory (Glaser & Strauss, 1967) is used as an approach to developing theory that has generated from the data. The research aims of the present study were to examine family's experiences of JIA and how they have made sense of their experiences as a family and so grounded theory was not considered the most suitable approach to address the study's aims. A narrative analysis approach could have been considered for this study. IPA and some forms of narrative analysis share similar ideas and theoretical underpinnings (Smith et al., 2009), for example, IPA is fundamentally concerned with meaning-making and a potential process for this is to construct narratives (Smith et al., 2009). Narratives are accounts of people's experiences over time and how a person describes their accounts relates to their process of sense-making (Smith et al., 2009). Looking for narratives within the data was part of the analysis process for the present study, but essentially the study was concerned with the lived experience of JIA, and so IPA was felt to be best suited to address the research's aims.

Sampling in IPA

Samples are purposefully selected for IPA studies as participants are able to offer a unique insight into a particular phenomenon, and thus the focus for the research is to learn about perspective and experience as opposed to obtaining large numbers for studies addressing efficacy, for example (Smith et al., 2009). IPA has been largely used to analyse how *individuals* make sense of their

experiences. Whilst no specific minimum numbers for studies are referenced, it is generally thought that a sample size of between three and eight is sufficient for an IPA study (Smith et al., 2009). However, due to the strong idiographic element to IPA, it is also recognised that very small sample sizes and even case studies, are suited to the IPA approach which can yield important and significant findings (Smith, 2004). It is therefore important that the chosen sample be homogenous in order to be able to study variability within the group (Smith et al., 2009). In recent years, IPA has been used to analyse case study material and group data. Like many other qualitative approaches, IPA can be used flexibly to meet the needs of the raw data obtained (Smith et al., 2009).

Analysing using IPA

Four common stages of analysing individual interviews using IPA have been proposed (Smith et al., 2009) although there is no one method for working with the data (Palmer et al., 2010). The first stage of analysis is the initial reading and re-reading of the transcript in order to immerse oneself in the data. Initial thoughts and reflections are noted in order to encourage bracketing-off and reflexive thinking. The second stage involves initial noting of observations or anything of interest and these are recorded within the left-hand margin. Comments can be descriptive, linguistic or conceptual and similarities, differences, amplifications and contradictions are also important in the initial noting phase. The third stage involves the development of emergent themes based on small sections of the transcript but also on the overall feel of the data. The aim is reduce the volume of data and begin to think conceptually and psychologically about the data. The fourth stage is to cluster themes together based on conceptual relatedness and also to also discard themes that are irrelevant to the research aims. Themes are clustered into super-ordinate themes and these stages are conducted for each participant. Patterns across super-ordinate themes are identified and then grouped and given a master theme title.

In order to accommodate the more challenging and complex data from the family group interview, such as looking at interactional processes, some additional stages to the analysis were added. Palmer et al. (2010) outlined eight stages to aid the analysis of group data. The additional stages were added as a method of acknowledging the interactional processes that occurs within groups. This in turn, can help the researcher to further make-sense of participants' understandings and how these may have been jointly constructed as part of the interviewing process (Palmer et al., 2010). The additional stages of analysis were drawn from discursive and narrative approaches to qualitative data analysis, paying close attention to the process of the interview as well as content (Palmer et al., 2010). These stages were utilised to analyse the family transcripts in the present study. The stages are outlined in the *Table 2*.

Table 2. **Stages in IPA group analysis.**

Stage	Description of stage
1. Object of concern and experiential claim	This involves the researcher reading the transcript in depth, extracting participants' experiences and objects of concern and organising the material into emerging themes.
2. Positionality	Analysis of the how the group members and the facilitator position themselves within the group is also important. This involves looking at the perspectives people take in relation to their experiences and what they hope their response to the questions achieves. This aims to help the research gain an understanding of *how* the groups work together and *how* the data emerges during the interviews.
3. Roles and relationships	This stage involves examining the references to other people, including what relationships are described and how, what are their understandings and expectations/consequences of the relationships. This is with the intention to find meanings in participant's accounts and to understand them within the context (familial and/or organisational) from which they arise.
4. Organisations and systems	This stage is concerned with participant's views and references to organisations and systems such as; positive and negative experiences, how they systems are described, how they believe the system works and what are the consequences of their relationships with the system or organisation.
5. Stories	This is a narrative approach to the analysis which focuses on examining the structure, the tone, the imagery and the genre of the stories told. The analysis incorporates what stories are elaborated on or hampered by, other members of the group. Furthermore how one talks about their experience will be shaped their experience of the world, so this is also an important aspect to consider.

6. Language	As with traditional IPA, language use is also monitored throughout the analysis. The categories recommend to consider are *patterns* of discourse (such as repetition, turn taking, emphasis and jargon), the *context* in which that discourse is used (such as the impact of the language and the descriptions used), and finally the *function* of the discourse (such as why was certain language used).
7. Adaptation of emergent themes	Earlier themes may need adaptation based on the information gathered from latter stages. It may be useful for the researcher to address what experiences are being shared, what are the individuals doing by sharing their experiences, what consensus/conflict is there within the group and is anyone marginalised. This means an overall picture of what is happening within each group will emerge.
8. Integration of multiple cases	Integrating themes from the groups builds up an overall picture of the experience under investigation. Similarities and discrepancies can be analysed and superordinate themes identified.

Table adapted from Palmer et al. (2010).

Methodology and Procedure

Ethical Considerations

The study was originally reviewed by an independent academic panel at The University of Leeds in November 2011 and March 2012. NHS ethical clearance was then obtained through Newcastle and North Tyneside 1 Proportionate Review Research Ethics Committee **(**see Appendix 2) and the project was registered with the research and development centre in Leeds. Three main ethical principles were considered for this study.

Principles of safety and well-being

For some participants, discussing experiences and understandings of JIA had the potential to become distressing. Participants were made aware of their rights to withdraw from the study, pause or terminate the interview if they felt distressed. It was also considered that the researcher had a sufficient level of training to be able to facilitate conversations sensitively and remained attentive to any distress in the participants. The researcher was also aware that families could be referred to a clinical psychologist within the Leeds Teaching Hospitals Trust if she felt there was continued or significant distress. Participants were fully debriefed at the end of the interviews and

offered the opportunity to ask any questions and to discuss their experiences of the interview process. Interviews took place in the family homes and so the researcher worked according to the University of Leeds, lone worker policy.

Principles of consent

This research is concerned with *family* understandings of JIA which meant that young people of any age could have opted to participate in the research. It was important therefore that every family member taking part gave informed consent, despite an opt-in approach to recruitment. Comprehensive information sheets were sent to families prior to any contact with the researcher, via the paediatric rheumatology consultant. Two versions were sent in each pack, designed to be age appropriate and to facilitate informed consent. Families were also encouraged to ask any questions about the study during initial contact and throughout the data collection process in order to ensure continued consent. Consent was assessed throughout the process, and for participants under the age of 16 years, Gillick Principles were utilised. To ensure full consent, four consent procedures were conducted. These will be outlined later in the chapter.

Principles of anonymity, confidentiality and data protection

In accordance with the Data Protection Act (1998) the researcher did not have access to any participant information prior to them opting in to be contacted. In order to protect the identity of the participating families, each participant was given a pseudo first and last name and these were used throughout the transcripts. Identifiable names or places were also removed. In addition, families were made aware of the small sample size and were informed, during debrief, that they could opt for any of the data to be removed from the transcripts. Due to the group nature of the family interview, participants were also asked to respect the confidentiality of the other family members' responses. Quotes from participants were not used if they contained any identifiable information that could not be changed. All audio recordings and transcripts were dealt with in compliance with the Data Protection Act (1998) and also in line with the University of Leeds policy.

Recruitment

Participants

Participants were families recruited from an NHS paediatric rheumatology service at the Leeds General Infirmary (LGI). Families were eligible to participate if the young person was aged between 12 and 19 years old, had been diagnosed with JIA for at least 18 months, and all family

members could speak fluent English. Families were excluded from the study if they did not meet the above criteria.

Recruitment procedure

Following ethical approval, a list of potential families who met the inclusion criteria was compiled by a paediatric rheumatology consultant and a senior clinical psychologist at the LGI. Information packs were sent out, by the consultant, to potential families explaining the nature of the study. Information sheets were devised for young people aged 12 and under and 13 years to adults, which used age-appropriate language (see Appendices 3-5 for example cover letter information sheets). Included in the information pack was a reply slip in which the family were required to fill in and return, stating whether they would be interested in hearing more about the research (see Appendix 6). If this reply slip was not returned, follow-up phone calls were made by the paediatric rheumatology team in order to gauge interest in the study. Participating families were contacted by telephone and any questions they had about the research were answered. Interview dates were arranged during this telephone conversation and both families requested the interviews be conducted in their homes.

Recruitment for the individual interviews occurred after the completion of the family interviews. Participants were asked to take part in an individual interview immediately after the family interview to gauge interest, and then asked again in a later telephone call.

Procedure

Consent Procedures

Four consent procedures were utilised in the present study (see Appendix 7 for example consent form). Firstly, family consent was initially sought. This was signed by parents to indicate that the family had given their permission to participate in the research. This was designed to include members who did not themselves, wish to participate in the research, but consented to be discussed as part of the family's experiences. Secondly, an adult consent form was given to all participating family members aged 16 years and above. Thirdly a young person's consent form was signed by family members aged 15 years and below giving assent to be interviewed. Finally, parents were also required to sign a form giving their consent for their children under the age of 16 years to take part in the study. The latter three consent procedures were also followed for individual interviews.

Devising a topic guide

An initial topic guide for the family interview was designed by the researcher and influenced by the literature in relation to families' responses and experience of chronic health conditions, and guided by the research aims. The schedule was guided by Smith et al.'s (2009) principles on semi-structured interviewing. Early questions were designed to elicit descriptive information with the hope to build rapport and engagement with the participants. Later questions focused on gathering in-depth experiential accounts, and were framed in a way that enabled minimal input from the researcher. The later questions were aimed at eliciting accounts about the families' understandings of JIA and how they have negotiated their sense-making processes with one another. Smith et al. (2009) recommends a guide that includes open-ended questions which allow room for further probing and prompts (see Appendix 8 for topic guide). Questions were designed to be jargon free and inclusive of all family members. The topic guide was devised over a number of weeks and questions were revised and re-drafted through the use of supervision and the use of a qualitative support group on the Leeds Doctorate in Clinical Psychology course. Additionally, a 90 minute pilot interview was conducted with a family who was experiencing early onset arthritis in a member. The feedback from this interview also contributed to the topic guide. Whilst the topic guide had a structure, the questions were used flexibly throughout the interviewing in order to maintain a participant-centred interview process. Furthermore, questions were amended during the course of the interviews based on feedback from participating families.

Interview schedules for the individual interviews were idiosyncratic to each family member, although followed similar principles to those outlined by Smith et al. (2009). Following the family interview, some preliminary analysis was conducted on the interview material in order to construct further questions and probes that had the potential to elicit further in-depth experiential data. Questions generally related back to conversations and accounts that had been discussed in the family interview that had not been fully explored. The questions also designed to check that the researcher had understood the participant's correctly and to determine any inconsistencies in the conversations (see Appendix 9 for example schedule).

Data collection

All interviews were held at the participating families' homes and were facilitated by the researcher. Prior to starting the interviews, any additional questions the participants had been answered and consent forms signed. All interviews were audio recorded using a digital recording device.

Family Interview

Both family interviews followed the same procedure. Prior to starting the interview, some basic ground rules were set out by the researcher, such as respecting one another's responses to questions and maintaining confidentiality. Participants were also asked their names and a piece of information about themselves for voice recognition. The researcher then used the topic guide as guidance throughout the interviews. Once questions had been exhausted the researcher brought the interview to a close. Families were debriefed and then asked if they were willing to take part in a subsequent individual interview. Interviews lasted between 90 minutes and128 minutes.

Individual Interview

Individual interviews were conducted several weeks following the family interview in order for the researcher to transcribe and begin some preliminary analysis the family data to develop a second topic guide. Participants were contacted via telephone and via email and the similar consent procedures to the family interview were followed. Five participants were interviewed on an individual basis. All interviews were audio recorded and all interviews were conducted within the family home. Families were again, debriefed after their interview and offered the opportunity to feedback on their experiences. At this point, the researcher also reminded participants about the use of quotes and how the interview data would be used. Individual interviews lasted between 40 minutes and 67 minutes.

Transcribing

The first family interview was transcribed by the researcher and the remaining six interviews were transcribed by an external and university approved transcriber. Recordings were transcribed verbatim using pseudonyms and non-verbal communications were included (for example, laughter). The first interview was transcribed by the researcher so there was some familiarisation with the material to construct individual interview schedules. However, due to times restrictions and the complexity of the family interviews, it was decided that an external transcriber would transcribe the remaining transcripts. These were all thoroughly quality checked by the researcher prior to analysis.

Data Analysis

The analysis of multiple perspectives has been conducted using IPA, in an attempt to a move away from simplified case-effect models (Dancyger, Smith, Jacobs, Wallace, & Michie, 2010; Glasscoe & Smith, 2011; Larkin, et al., 2013; Rostill-Brookes, Larkin, Toms, & Churchman, 2011). The implication of multiple perspectives is a more complex analysis process. Flowers (2008) proposed

three possible methods of integrating multiple interview data. Firstly, Flowers (2008) suggests analysing all interview data as one interview, which has the advantage of maintaining a simple and clean analytic process. However this process ignores the continued relationship with the researcher and the different contexts within which the interviews were conducted. A second method of synthesising data proposed by Flowers (2008) is to conduct a preliminary superficial analysis of the first interview in order to inform the subsequent interview. The advantage of this is to crudely quality check aspects of the first interview analysis but the disadvantage is that the second interview can become researcher-led as opposed to participant-centred. The third possibility is fully analysing the first interview and taking themes back to the participants (Flowers, 2008). This allows for fully informed quality checks however, the analysis becomes complex using this method. The present research used the second proposed analysis due to time constraints on the researcher to complete a full comprehensive analysis prior to second interviews. This means that family interviews were analysed separately to individual interviews, then themes were integrated to develop one set of themes per family.

In keeping with IPA focus on idiography, each case study was analysed in its own terms. This meant as much as possible, bracketing off the fore-structures developed from the previous case study (Smith et al., 2009).

Analysis of family interviews

The analysis of the transcript was informed by the eight stages outlined earlier (Palmer et al., 2010). After in-depth reading of the transcript, notes were written in the left hand margin of the transcript, closely paying attention to experiences, thoughts and feelings. Initial emergent themes were noted by the researcher and the research supervisor. Later stages of the analysis involved identifying the interactional patterns between participants, paying attention to nuances of agreement or divergence, contradictions and how the family positioned themselves in relation to JIA (see also Åstedt-Kurki, Hopia, & Vuori, 1999). In addition, notes were made about how the families described their relationships, both between family members and descriptions about people outside of the family. With whom the family members positioned themselves was also recorded, such as the use of ‘I’ and ‘we’, and noting agreements and disagreements within the family. Analysis of interactions helped the researcher to develop an understanding of how the family worked together throughout the interview, who constrained or enabled conversation, how conversations were shaped or changed as a result of the stories told and which participants contributed to which accounts (see Appendix 10 for example of analysed transcript). Field notes made by the researcher, and audio recordings were also used at this stage to inform the analysis. Careful consideration was made in interpreting covert interactional data. For example, disparities

in individual understandings were not necessarily indicative of strained family relationships and shared understandings were also not necessarily indicative of close familial relationships (Eisikovits & Koren, 2010).

Finally, initial themes were revisited and revised based on all the additional notes made on the interactional notations. Themes were organised into super-ordinate themes, however, master themes were not identified until after the analysis of the individual data. This process was repeated for the second family group interview (see Appendices 12 and 13 for examples of the analysis process).

Analysis of individual interviews

The analysis process of the individual interviews followed the four stages outlined earlier, by Smith et al. (2009) however the themes identified in the family interview were used as a structure of analysing the individual interview. This is because the individual interviews acted as supplementary data for the family interview, as opposed to developing a distinct set of themes (Butt & Chelsa, 2007; Rostill-Brookes et al., 2011). Special care was made to ensure that the researcher was not committed to specific themes identified at the family interview and was observant to new themes that emerged from the data. Furthermore, particular attention was paid to positionality, reference to the family interview, contradictions in accounts, and reference to relationships (see Appendix 11 for an example of analysed transcript).

Integration of family group data and individual interview data

Once the individual interview data had been fully analysed, any new emergent themes were then integrated into the family interview data. In relation to both families' data, the themes identified from the individual interviews either enriched or added further concepts to the themes identified in the family interview, or contributed to a new understanding of an existing theme. As a consequence, the researcher's understanding of the experience developed as a result of integrating the individual data and themes either took on new meanings or meanings were strengthened as a result of integration. For example, a theme of *'transitions'* was identified in the individual interviews of the young person with JIA, but was not identified in the family interviews. This enhanced and added a new dimension to existing themes from the family interview. Any contradictions and opposing narratives that emerged from the individual interviews were also noted and integrated into the existing themes. Particular attention was paid to shared narratives within the family and when these narratives were identified as divergent at the individual level.

Themes were re-examined across all the data, in order determine patterns of relatedness. As a result, clusters of themes were identified to determine super-ordinate themes and for one

super-ordinate theme, sub-themes. Each super-ordinate theme was labelled which described the group of themes. These super-ordinate themes were then clustered conceptually, and labelled to develop master themes. This process was repeated for the second case study, resulting in two distinct case studies.

The development of master and superordinate themes was completed in conjunction with the researcher's supervisor within supervision. Potential themes were discussed, clustered and re-clustered following these conversations in order to develop a thematic map whereby all concepts and ideas that had emerged from the families' accounts could be clustered into an appropriate super-ordinate and master theme. The themes identified within supervision meetings were also discussed within a qualitative research support group. The conversations in supervision were also reflective in nature to encourage distance from the data in order to provide a more objective view of the emerging themes and minimise researcher bias. The selected themes were decided upon if they most closely represented and captured the phenomenological aspects of the families' accounts, if participant quotes clearly supported the emergent themes and the identified themes related to the aims of the research.

Synthesis of case studies

In order to synthesis the case studies, commonalities and differences between the families themes needed to be identified. It has been suggested that often, the synthesis of data requires renaming and reconfiguring existing themes (Smith et al., 2009). Synthesis of the themes involved looking for shared concepts within the master and super-ordinate themes that represented all family members and both families. Assurance was made not to favour the results from one case-study over the other and to represent both case studies equally. Similarly, contrasts between the cases were also identified within the themes in order to preserve the idiosyncrasy of the data.

Quality Checking: validity and credibility

It is widely accepted that qualitative research should undergo the same degree of rigor-checking that occurs with quantitative research. There are several ways in which the validity of qualitative research can be enhanced (Mays & Pope, 2000; Yardley, 2000). Suggested methods of enhancing the quality and validity of a study are: triangulation, respondent validation, reflexivity, attention to discrepant accounts, questioning the relevance of the research and transparency and coherence of data collection and analysis, such as, demonstrating audit trails and using quotes to support concepts (Mays & Pope, 2000; Yardley, 2000). The following procedures were used to improve the quality of the study:

- *Research and field supervision:* Regular meetings were held which involved on-going communication and also sections of transcripts were read and themes identified and verified during supervision. Supervision was important during the analysis stage of the study, for example, with the arrangement of codes and themes.
- *Independent coding:* Throughout the data analysis stage, the research supervisor and researcher read sections of a transcript and comparisons were made between emerging themes. This was an important stage in order to determine any biases towards certain themes or participants' stories and highlighting any of the researcher's assumptions or fore-structures that impacted upon data analysis.
- *Transparency:* An overt and clear description of the process of data analysis and example of data analysis has been included in this write-up. Furthermore, quotes have been used in order to support identified themes.
- *Triangulation:* This study utilised a number of different perspectives and two methods of data collection to increase the validity of the study and its findings (Moran-Ellis et al., 2006).
- *Respondent validation:* This is thought to be a method used to increase the validity of research studies (Smith, 2008); however, due to constraints (discussed in Chapter Four) this method of quality checking was not conducted. However, individual interview were utilised to follow up and further explore some of the initial thoughts the researcher had of the data during the preliminary reading of the family interview transcripts.
- *Reflexivity:* Reflective journals and memo writing was used in order to enhance self-reflection and recognise researcher biases during data collection and analysis (Smith et al., 2009).
- *Attention to divergent narratives:* Attention was paid to divergent participant accounts during the analysis and discussion stage of the study. This was also integral to respond to the research's aims.
- *Training in methods:* The researcher attended a one-day IPA workshop led by a member of the core IPA team, which gave the researcher the opportunity to discuss and gain recommendations on the complex analysis procedure and become more skilled in the IPA approach.
- *Peer group validation:* The use of a qualitative support group enabled the research to utilise peer-coding opportunities and gain advice on data analysis and clustering themes. This group was run by an experienced researcher with a particular interest in qualitative analysis approaches.

Reflexivity

It is important to openly reflect on how my experiences, assumptions and biases may have influenced the research and analytic process (Smith et al., 2009; Yardley, 2000). As stated earlier in the chapter, the researcher's own perspectives and assumptions inevitably influences how accounts are interpreted and analysed, however, engaging in reflexivity can help the researcher to become aware of these assumptions and biases. There are two particular methods of reflexivity I utilised, especially throughout the interview and analysis process of the study, and these were maintaining a reflective journal (including memo writing) and reflective conversations during supervision meeting and support groups. I have also included a reflexive statement which outlines some entries of my reflective journal early on in the research process. Keeping a reflexive journal was particularly useful, and was primarily used to note down thoughts, ideas, my impressions of the participants and to identify potential interview questions for individual interviews. The journal was key to developing pen portraits and linking concepts and ideas together during the theme development and analysis stage. Memo-writing also aided thoughts regarding clustering themes and was useful as a memory aid during the analysis process. Minutes from meetings were also written within the journal to reflect on in-between appointments. Reflective conversations with supervisors and peers also helped elicit my assumptions, distance myself from the transcripts and data and to observe the accounts from a different perspective.

Reflexive Statement

Whilst I have had no direct experience of early onset arthritis or indeed a chronic health condition that requires regular medical input, I have had significant personal and professional experience of adults and young people diagnosed with chronic health conditions. Professionally, I have had an interest in the field of health psychology and I completed a Masters in health psychology, post-graduation, and as part of my doctorate in clinical psychology, I am currently completing my year-long elective placement in the field of adult and paediatric health. My experiences of working closely with people who have struggled to adjust to a health condition could bias my thinking toward looking for accounts that corroborate with my professional experience.

For many years, I have also worked for a charity providing therapeutic respite care for young people and families experiencing chronic and life-limited conditions. My role within this charity is to provide emotional and/or physical support to families and to especially facilitate self-efficacy and self-esteem. During this work, I have been particularly interested at observing family interactions during these occasions and how each family differed in how they spoke and coped with negotiating a health condition. Furthermore, I also noticed that within some families,

members coped in different ways to one another. This was a significant influence in my motivation for this research.

Another motivation for my research is that I have a family member who was diagnosed with early-onset arthritis, when in her early twenties. This is a particularly aggressive sub-type, and as a result she now experiences significant limitations in her mobility. However, whilst this is the case, I have also observed her strength, determination and resiliency in facing the many problems that come with the condition. I believe that this personal experience biased my understanding that arthritis in a young person will be aggressive and limiting, resulting in the need to make significant life adjustments.

Finally, during the course of my clinical training, I have been 'diagnosed' with dyslexia. This means that throughout the process of my thesis, I have needed to make adaptations in the way I write, read, study, learn and negotiate a 'condition' where there is no cure. This at times has led to frustration throughout the thesis process and a need to 'get it right'. I have also needed to negotiate associated social and cultural narratives of 'deficit', 'disability' and 'inability' that come with dyslexia. These are perhaps also labels that are associated with chronic health conditions and I am aware that these parallels (including my own experiences of adjustment) could have impacted upon my analysis.

CHAPTER THREE

RESULTS

This chapter will present the results of the analysis conducted, using IPA. The analysis was focussed on addressing the following research aims:

1. Explore familial understandings of JIA following the diagnoses of the condition in a young family member.
2. Explore how these understandings are negotiated and communicated among family members.

This chapter will be split into the two case studies. For each case study I will present the family pen portraits, a thematic map of the master, super-ordinate and sub-themes themes and then outline and describe the identified themes. In addition, a synthesis of the case study data will also be presented at the end of this chapter. Please refer to Appendices nine and ten for example transcript extract which demonstrates an element of the analysis process. Quotes from the case study transcripts will be used to provide examples for each super-ordinate and sub-theme in order to illustrate the identified concepts. To protect the confidentiality of the two families who participated in the study, each participant was given a pseudonym which was used throughout the results and discussion chapter.

Sample

A total of 18 information packs were posted to families, over a number of weeks. From these 18 packs, seven reply slips were returned, of which three families reported that they would be interested in hearing more about the study. Following telephone conversations, two families agreed to take part in the study. The third family felt they would not be able to contribute because they felt they had not been affected sufficiently by JIA to contribute to the study.

Case Study 1: The Hunter family

Hunter family pen portrait

The Hunter family were the first family interviewed and this was a single parent household. The family consisted of three members who were British in ethnicity: Annie, the young person experiencing arthritis, Robert, the father of Annie, and Emily the younger sister. All family members lived within the same household at the point of interview and knew of no other family members experiencing early-onset arthritis. For the purpose of the family's anonymity, there will be no information presented in relation to Annie and Emily's mother.

The Hunter family have lived with JIA for approximately eight years and all members had contributed to the management of the condition. Since the onset of JIA, no medication had been effective in managing Annie's symptoms for any significant period of time and so much of the family's focus, with regards to JIA, was centred around managing flare-ups, pain management and finding new combinations of medications which could control the condition.

Annie was 17 at the time of the initial family interview and turned 18 just prior to the second individual interview. She was diagnosed with JIA at the age of 10 years with an initial diagnosis of psoriatic arthritis, which was subsequently retracted and replaced with the general diagnosis term of JIA. At the point of interview, Annie was experiencing active symptoms of JIA in many of her joints, although her hands, feet and shoulders have, historically, caused her the most difficulty with pain and mobility. Typical of young people experiencing JIA, Annie experiences periods of significant pain and mobility restriction which has, on occasion, led to the use of crutches. Due to the severity of her symptoms Annie has undergone several hospital trips to have steroid injections into the joints, as a way of controlling the symptoms and relieving the pain. In one appointment she described having sixty injections under general anaesthetic. Annie is also on weekly medication which she administers at home.

The family described Annie as having a difficult relationship with her regular prescribed medication. Annie also experienced significant anxiety around giving herself injections, which are part of her medical regimes, and this resulted in a period of choosing not to continue with her medication for approximately a year. At the point of interview, Annie was in transition from child into adult rheumatology services. Despite these experiences she continues to lead a busy life. Annie described herself as disliking having any association with 'impairment' or 'disability' and would hide the JIA in order to prevent any negative judgements or stereotypes from others.

Robert has had full involvement in the care of Annie since she was diagnosed at the age of nine years. Robert had no prior knowledge of juvenile arthritis and had always associated arthritis with the older generations. From the onset of symptoms, Robert attended the majority of medical

appointments with Annie and so viewed himself as being knowledgeable about JIA and its treatments. Since Annie started transitioning into adult services, Robert no longer attends most the appointments. Despite Annie's anxiety around medication and their limited effectiveness, Robert remained fully supportive of the medications, but he did however express feeling immensely frustrated that no intervention had provided Annie with any relief. Robert had also found it difficult to divide his time between parenting two children, along with the additional resources that the JIA requires.

Emily is the younger sibling of Annie and was 12 years old at the time of the interviews. Emily was four years old when Annie was first diagnosed and has no memories of this time. However, she described having clear memories of the time when the JIA first started to demand more of the family's resources. Emily has had no involvement in any of Annie's medical routines or appointments, but does help with the care of Annie on days when she is struggling with mobility or in pain. Emily described enjoying caring for Annie when Annie struggled most with JIA. Emily did not feel as if she knew much about JIA in a medical sense, however, she had learnt a lot through observation. When Emily first noticed the impact JIA had on the family, she described finding the changes within the family difficult and unsettling but she is now more accepting of it. Emily is also involved in the scouts and leads a very active life. Emily is a regular church goer and described herself as having a spiritual outlook to life which had contributed to how she felt about the JIA. Emily believes in fate and that ultimately challenging life experiences has made the family stronger.

The family interview lasted for 90 minutes. All three family members consented to an individual interview, Annie's lasted for 41 minutes, Robert's interview lasted 49 minutes and Emily's interview lasted for 45 minutes. All individual interviews were conducted approximately six weeks after the initial family interview.

Reflections from the interviews

My impressions of this family were that they quickly engaged in the interview process and it appeared that Annie and Robert were comfortable discussing their experiences together. It appeared that many of the stories told, especially those around the subject of medication, were well rehearsed between Annie and Robert and this led to few discrepancies in their accounts. Due to the difficulties the family had experienced with the medication, a large focus of the interview was medication management and, at times, I found it difficult to steer the family onto other topics.

Annie appeared shy during the interviews and she did comment that she had not often spoken about the JIA in detail with people before. Annie had also had some negative experiences following the disclosure of the JIA to her peers so I was mindful that perhaps she may have

struggled to disclose some of her experiences to me. In addition, I was also mindful that Annie may have found some experiences hard to articulate if she had not often expressed these to others. In the family interview Robert offered very factual responses and I found it difficult to elicit his thoughts and feelings about events. In contrast, Robert offered more personal reflections about his experiences in the individual interview, with some focus on sometimes feeling helpless to help Annie.

What struck me most about Emily was the vast difference in her presentation across the two interviews. In the family interview, I observed that she was very quiet and spoke little, although it was clear that she was engaged in the conversations. Predominately, Emily spoke to prompt Annie and Robert about certain past events or to verbalise that she did not know much about the topic area. My impression was that Emily was uncertain about discussing her experiences with Robert and Annie present, and was unsure of the response she would receive if she revealed her thoughts. In the individual interview Emily opened up and offered more information about her feelings towards the arthritis. Emily was very reflective and honest about her family and she also talked about other family events that had impacted upon the family.

Results of analysis

This case study explored the Hunter family's experiences of JIA by way of four master themes: *Negotiating power, not letting go: managing transitions, when the invisible becomes visible* and *just getting on with it* (see Figure 1 for thematic map). Additionally a section on negotiating understandings based on family process and storytelling will also be presented with example quotes to illustrate the concepts.

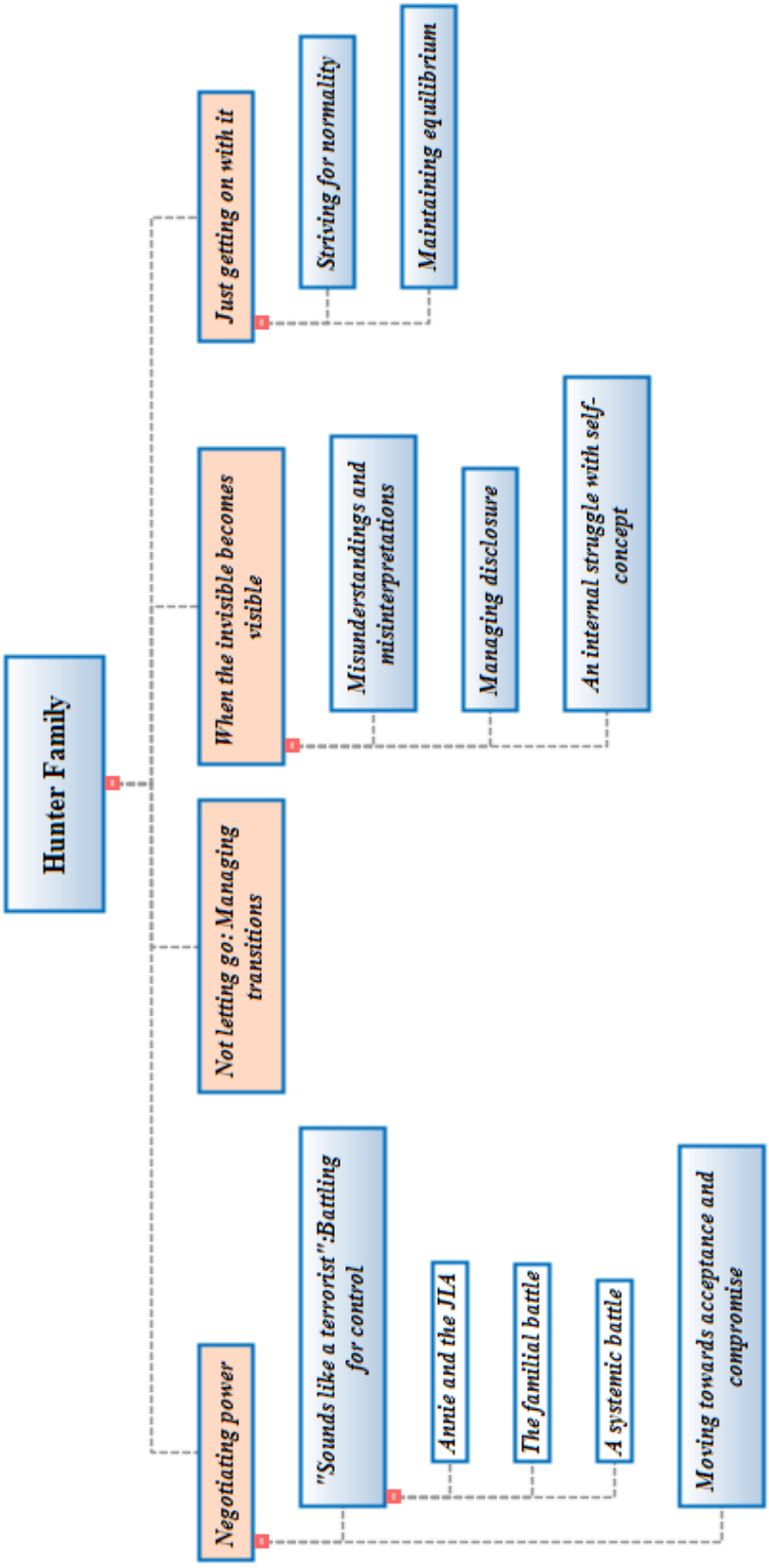

***Figure 1*. Hunter family thematic map.**

Negotiating power

This master theme describes the family's experiences of JIA that was related to their perceived power and control over the condition and the experienced consequences during times when the family was unable to negotiate with the JIA. This master theme encompasses two super-ordinate themes: *It sounds like a terrorist: Battling for control* and *moving towards acceptance and compromise.*

"It sounds like a terrorist": Battling for control:

This super-ordinate theme captured the family's experience that JIA, and its associated elements, could be challenging to control, potentially leaving the family feeling powerless. The family described that some of these JIA related aspects were non-negotiable for them as individual family members, and some were negotiable. For those elements that the family felt were non-negotiable, they fought to retain a perceived sense of being in control. This theme is divided into three subthemes incorporating three layers of struggle for the family: *Annie and the JIA, the familial battle* and *a systemic battle.*

Annie and the JIA:

This sub theme relates to the family's experience that their relationship with the JIA and the medication is a battle. Annie described the medication as a powerful force that evoked fear and disempowerment:

> '*It sounds like a terrorist'* (Annie, family interview: line 342).

There was awareness that Annie's battle was both physical and psychological in nature and bi-directional. This can be explained by the following extract relating to one of her medical interventions:

> *'I went onto the Methotrexate injections which I had a nightmare with and they made me really ill and really sort of depressed and things like that and fought forever well it felt like I was fighting for ages anyway to get off it and eventually I did'* (Annie, family interview, line 295).

This quote highlights that the family experienced a continued battle with finding a medication that would be effective enough to relieve the symptoms of JIA for Annie, and enable to family to regain a sense of control. In addition, Annie felt that 'giving in' to pain was non-negotiable, and described pushing herself as far as she was able to in order to prevent a sense that JIA could 'get the better of her'.

> *'they let me set off with them and they said "ok you'll do a bit" and then when they get a bit higher you can come back down and go for a coffee or something…and I went as far as I could and I kept pushing them to let me go further I got about half way and they were like 'look you're going to have to go back down now'* (Annie, family interview, line 1529).

The family also described that Annie's battle with the JIA is, at times, lost. Annie reflected on her experiences of pushing herself and often questioned whether this was a something that was worth the effort of battling. Annie explained a residual feeling of exasperation and frustration with herself which ultimately left her feeling more powerless than was intended: '*It's frustrating and…I normally just end up doing it and hurting myself and then I think was it really worth doing that'* (Annie, individual interview, line 305).

Finally, the losing battle and the lack of control the family had over the medication was described by Robert:

> *'on the face of it's a relatively straightforward disease… cannot just be managed …at the moment the Adalimumab is still not working you that that she's rejecting it so so now she's on the Methotrexate and Adalimumab...and because of the Methotrexate she's on anti-sickness and folic acid and she's also on oral steroids at the moment as well 'cos her hands are bad'* (Robert, individual interview, line 40).

Robert described a sense of relentlessness to Annie's medical regimes as he listed Annie's medications, and the attempts made to find a drug combination that would prevent this physical rejection. There was a sense that the medications which were designed to make Annie feel better, were causing the opposite to happen. This further perpetuated Annie's struggle with the JIA.

The familial struggle:

This second sub-theme addressed family-level differences in viewpoints between Robert and Annie which initiated a lengthy disagreement and highlighted their divergent values in relation to JIA. This theme additionally reflects the differences in opinion between the family members as to what was negotiable and what was non-negotiable and how this was subsequently managed:

> '*I was a lot of the time refusing medication erm dad did not agree with this decision…and voiced it quite clearly that I shouldn't be rejecting medication…he wanted the best and he wanted me not to have the symptoms but he didn't understand what it was like to be on the medication'* (Annie, family interview, line 1343).

Annie explained understanding Robert's reasons for his limited support of her decisions, however she felt misunderstood and isolated. Whilst Annie had believed she had solved her own personal battle with JIA by refusing the medication, this 'resolution' had then facilitated the development of

this additional struggle within the family. In addition Robert feared that Annie's unwillingness to negotiate would result in a much longer, irreversible battle, whereby the arthritis would gain significant control: '*I believe your line was "you'll be crippled by eighteen if you don't do it*"' (Annie, family interview, line 1377). The resulting experience was that these battles for control had left the family with an overall sense of feeling out of control and powerless:

> '*when she was in that period where she was off the Methotrexate and symptoms were getting bad she struggled to get dressed sometimes you know and it was difficult watching her suffer*' (Robert, individual interview, line 1132).

This theme also reflects the family's joint efforts in their battle against JIA and their unwillingness to stop fighting for a solution that would offer Annie some relief from the JIA. However their preferred solution, as described, highlights their divergent viewpoints.

Robert described the longevity of their battle and an anticipation that they will continue to be challenged for some time to come. This can be demonstrated by the following two quotes from Robert: '*buckle up, it's going to be a long one'* and '*eight years later and we're still trying'* (Robert, individual interview, lines, 874 and 395 respectively).

A systemic battle:

This sub-theme relates to the Annie's experience of her battles with the professionals and involved in her care in relation to the JIA, for example: '*no matter what he told me I'd made my decision as soon as he mentioned that it was an injection I was not having this medication'* (Annie, family interview, line 1390). Annie explained that within the context of her usual appeasing character; a conversation such as this would be rare, indicating the importance she had placed upon winning this battle and what it had meant to her to assert her opinions. However she described that these efforts caused a contradiction of feeling empowered because on one hand she had won her battle but on the other hand she experienced substantial distress and a sense of powerlessness after disagreeing with what had been recommended to her:

> '*I was just so sick of it and so upset that I'd been on it for so much longer than I wanted 'cos I remember one time I had an appointment I'd gone in thinking it's ok I'm going to be off it after today it was a Friday so I'd had I'd have to do it that evening so it's ok I don't have to do it they kept me on it and I'd gone into school I just burst into tears I was so upset'* (Annie, individual interview, line 692).

This feeling of powerlessness was accentuated by the fact that the medical professionals and Robert believed that this was not the right battle for Annie to try and win. Robert described

divergent views within the family as he battled Annie alongside the professionals, meaning these battles developed within the family, as described in the previous sub-theme.

Moving towards acceptance and compromise:

This super-ordinate theme captures elements of the family's JIA related experiences in which they felt they were resolved to accept and view these aspects as more negotiable. The family described this as a method of maintaining a sense of empowerment and control in relation to their circumstances. Whilst there was a family-level understanding of acceptance of the JIA, each family member identified their individual ways of how they reached the point of acceptance. Emily described her acceptance process as a recognition that arthritis was part of the family and so there was little point in fighting something that may never change, but that did not mean that it should be welcomed: '*I don't like it it's not a good thing but you can't change it'* (Emily, individual interview, line 761). The importance of changing the attitude towards the arthritis as opposed to changing the circumstances was also highlighted by Emily '*I feel it's important to put your trust in God and God has a path set for you you just have to choose which way to go down it'* (Emily, individual interview, 609). This indicates that paradoxically, relinquishing control resulted in Emily feeling more in control.

In contrast, Robert reflected on his own personal struggles to accept their circumstances. He described drawing upon his experiences by comparing them with his understanding of cultural and social expectations regarding adjustment '*People say you know time's a great healer and it's it's not things don't get better you just learn to live with them'* (Robert, individual interview, line 619). Robert identified that acceptance comes with time, but similar to Emily's outlook, he did not feel his opinion of JIA had shifted alongside the acceptance process.

Annie described her experience of acceptance as arising from perceived lost battles, and the process of acceptance ran parallel to that of thoughts of '*admitting defeat'* and '*feeling disappointed'*:

> *'like one camp we went on I was freezing cold and we were we were swimming in a lake or something and we were doing rafting which I am perfectly capable of doing all the lashing and the knots and everything but my hands I just couldn't do it'* (Annie, family interview, line 1594).

Annie explained that, in some instances, she was able to accept that there were times when she was not able to manage some activities that she would have liked to partake in, but this acceptance came after a 'failed' attempt. This form of acceptance was also described by Robert. The lengthy

process of being unable to manage the JIA symptoms meant that the family were coming to terms with the fact that there may not be a cure for Annie:

> *'in the end…we're not sure if there's another one after Adalimumab …but we're getting into the experimental round now so I'm not sure how much further there is to take this'* (Robert, individual interview, lines 825-831).

However, the process of acceptance was made more challenging by the family holding on to past successes, enabling them to believe that they could have control over the JIA again:

> '*she couldn't hold a pen and then within six weeks of starting the new Adalimumab she scampered up [mountain name] with the scouts' (Robert)…'Yeah it was really good for the first six months or so and then it it's deteriorated'* (Annie. Family interview, line 354).

The family also reflected on their process of acceptance by thinking flexibly and demonstrating preparedness to compromise with JIA. This can be shown by a quote from Annie regarding taking her medication:

> *'in the end I figured well I may as well just try it erm…I still hate doing it and now I'd still much rather not do it and I'm not comfortable with doing it but I just kind of get it over with'* (Annie, family interview, line 1461).

Furthermore, Robert also identified that flexibility was the key to moving alongside the arthritis by re-establishing and re-negotiating the family's boundaries in order to gain a new perspective '*you just have to adjust parameters and take a different view'* (Robert family interview, line 1295). Robert and Emily described that them being able to move parameters, such as accepting that that Annie required help with tasks she had previously mastered as a young child, meant the family could better accommodate the JIA:

> *'if like you try getting a top on or something on one way and then she goes "ow no" then you like stop and do it a different way or something like start with the other arm or do something like that'* (Emily, family interview, line 1324).

Not letting go: negotiating transitions

This stand-alone master theme, named *not letting go: negotiating transitions,* identifies the family's experience of Annie growing up and negotiating adolescence alongside the negotiation of a chronic health condition and additional support that this requires.

This theme encapsulates Robert's internal conflict of wanting to encourage Annie to gain a sense of independence and experience life as an 'average' teenager would, but at the same time, still experiencing a desire to remain involved in her care. On one hand, Robert acknowledged Annie's age and expressed his beliefs of what people of Annie's age should be managing independently from their families, at her stage of development: '*I keep an eye on it from arm's length now she's a er young adult it's up to her to deal with the jabs'* (Robert, individual interview, line 77). However, on the other hand, Robert also expressed that he had not always felt able to remain at '*arm's length'*:

> '*we've had an episode a few weeks back where she had a problem with the injections…and we worked out a system and it was working fine and then I think she became a bit blasé about it…I came home one Friday and she was in floods of tears and it had all gone wrong and she'd scared herself and then…she was really really upset so I ended up doing the jabs for a few weeks'* (Robert, individual interview line 80).

Robert described wanting to protect Annie from distress but found it difficult to achieve a balance between encouraging her independence and ensuring her long-term health.

In addition, Robert further demonstrated his struggle to 'let go' during a point where decisions needed to be made about JIA. Robert felt it was important that his opinion should be heard and that his opinion was the 'right' one, negating Annie's ability to make independent decisions '*I guess it comes down to whatever we think is best for [Annie]*' (Robert, individual interview, line 235).

This theme also describes a conflict that Annie experienced in negotiating her own transition into adulthood. On one had Annie describes her independence as expected and necessary: '*there's not really any need for my dad to be there because obviously I'm not young I don't need a chaperone'* (Annie, individual interview, line 645), yet the additional support required for her to negotiate JIA when the symptoms are particularly active was essential:

> '*I'm going to have to keep giving myself injections I'm going to have to remember which week I'm doing which won't be an issue 'cos I'll work it out but I'm going to have to be the one to make myself do it rather than dad leaving it all on the counter…he gets it out*

> *before I get back form school so that I when I get in it's on the counter'* (Annie, individual interview, line 553).

Furthermore, whist Annie found this additional support helpful, there was ambivalence about how she perceived this help:

> *'holding the cutlery well enough to be able to cut up food when your hands are sore is really hard so that's another little thing I can get people to help me with even though it's a bit of a childish thing'* (Annie, family interview, line 743).

Annie viewed this event as being childish and incongruent with what is expected of someone who is culturally considered a young adult and competent of this task, however, seeing this as something which could not be avoided.

The theme *not letting go* also encompasses Annie's experience of transition from paediatric services into adult rheumatology services. The imminent move to adult services left her feeling uncertain and ambivalent about this particular transition:

> *'I'm currently moving up into the adult clinic so I don't know what that's going to do because I'm not going to see the same doctor every time I'm just going to see any doctor so they're not gonna know me they're not gonna know everything whereas my current doctors do'* (Annie, individual interview, line 511).

Annie expressed not wanting 'to let go' of the existing, trusting relationship she had with her previous consultants, and the prospect of new and inconsistent relationships resulted in a reluctance in her approach to emerging adulthood. Furthermore the transition into adult services also highlighted Annie being 'different' which resulted in feeling isolated in her experiences and changing identity:

> *'I have had one in the adult clinic it was an…emergency one when I got really bad and actually my doctor made an effort to make sure he was the one that came to see me so it was it was fine…it was just odd 'cos I walked into this room and erm I think I must have been the only one under 70 (laughs) just me…it was just kind of weird…it was just the fact that there wasn't a single person there that was remotely young'* (Annie, individual interview, line 753).

When the invisible becomes visible

This master theme relates to aspects of the family's positive and negative experience of JIA being an '*invisible*' condition that, at times, can become observable to others '*it's just the little things like the strength of the grip and stuff that show it up'* (Robert, individual interview, line 958). This

master theme encompasses three super-ordinate themes of: *misunderstandings and misinterpretations, managing disclosure* and *an internal struggle with self-concept*.

Misunderstandings and misinterpretations:

This super-ordinate theme reflects the family's observations that Annie wants the condition to remain hidden so that she does not have to defend herself, or the condition, from people who are unaware of the JIA, or who do not fully understand it. However, despite wanting the JIA to remain hidden, it was not always possible:

> '*when I was on crutches and things people presumed I had an injury and so then when I was on and off them they were like "you don't need them do you?" and I was like 'well I do' it's not 'cos I've not got an injury it's just 'cos I don't need them every day'* (Annie, individual interview line 234).

Annie's determination to hide the condition paradoxically resulted in increased feelings of distress and upset when other's failed to make allowances and accommodate the JIA:

> *'I remember one lesson where we had to do sprints and we had been running for an hour and I thought I really don't want to do this last sprint…I'm really sore and my teacher made me do this sprint and I finished and just burst into tears at the end'* (Annie, family interview, line 568).

Annie keeping the JIA well hidden meant that others may have not known to accommodate the JIA. It was only until Annie demonstrated her distress that people were able understand her experience. Robert also reflected on the fact that the family's reactions, when JIA became visible to others, had to be interpreted within the context of people's awareness of Annie's diagnosis:

> *'…trying to unscrew the sparkler and she couldn't…and the lad who was working with her was laughing at her you know it's just little things like the strength of the grip and stuff that show it up…it depends on whether it is malicious or not I know [name of colleague] and he wouldn't if he knew he wouldn't so when it's done through ignorance it's not a problem if it was malicious then I would have a problem'* (Robert, individual interview, line 938).

Misunderstandings and misinterpretations also relates to times when the family have also struggled to accommodate the JIA and understand the extent to which Annie is affected by the condition. JIA as a hidden condition led to strained family relationships on occasion, for example, the following quote from Emily described her reflections of Annie's behaviour as Annie attempted to 'cover up' the condition:

> *'I just thought she was too tired…I just thought she was being lazy and like erm "well my fingers hurt" I thought that was just an excuse but it wasn't…excuse for not wanting to go…being lazy'* (Emily, individual interview, line 351).

Emily explains how her limited understanding of Annie's experience resulted in an understanding that Annie did not want to spend time with her.

Managing disclosure:

This super-ordinate theme reflects Annie's experience of managing the disclosure of JIA to others. It also described the uncertainty she felt during this process, in relation to how much information about the condition she offered to share with others. This was partly due to the fact that disclosure was often as a result of the JIA becoming inadvertently visible and therefore it became unavoidable to acknowledge the JIA and disclose to others:

> *'a lot of the time it's just people noticing things like…if I have a swollen finger or something they'd be like "oh what did you do?" and it's like "well I haven't done anything"'* (Annie, individual interview, line 230).

In contrast, the family also described how disclosure can be a positive experience if it is within the context of long-term trusting relationships, such as the scouts group that Annie was involved in:

> '*they've always been really good to me though they've given me exactly what I need but then they've been encouraging and…they've just been really good'* (Annie, family interview, line 1572).

> ' *'cos the normal team obviously know her (.) and they you know know if it's a good day or a bad day'* (Robert, family interview, line 1517).

Annie described that disclosure can encourage supportive and beneficial relationships which ultimately enabled her to feel positive about allowing others to 'see' the JIA.

In addition, this theme *managing disclosure* also encapsulates Annie's experience of post-disclosure management and her negotiation of how much help she was willing to accept from others. Annie described developing boundaries as to when it is acceptable for family and friends to offer their support:

> *'it's kind of nice when you come out of hospital after I've had loads of steroid injections into my joints erm I've had a couple of friends come round before while I was kind of lying on the sofa and it was quite nice then 'cos they knew obviously they just…that was*

> *kind of nice but the rest of the time I just don't like 'cos then I really do need it when I'm like that but they know'* (Annie, individual interview, line 184).

Additional reflections were made as to when disclosure caused friction in Annie's peer relationships and what responses to the JIA she preferred and did not prefer, as demonstrated by the following extract:

> *'it's just when people start treating you…they're trying to be nice but when they treat you different and it's like "no come on I'm fine"…and that annoys me'* (Annie)… '*I still make you give me a piggyback'* (Emily)…*'which is fine that's how that's how I'd much rather it be'* (Annie, Family interview, line 596).

Annie explained that the disclosure process meant that she perceived others to treat her as delicate and fragile. However, for Annie, the aim of disclosure was not for differential or 'special' treatment, especially when it was not needed. Annie and Emily described her preferred response from others was to be treated like every other teenager unless it was unavoidable. Those who knew when it was appropriate to negotiate a different kind of treatment were those who had been informed about the JIA for a long period of time. However Emily described that understanding about JIA comes with time:

> *'when I saw her doing it for the first time when I was actually in there with her…she kind of scrunched up her face in pain and I didn't really know what to do'* (Emily, individual interview, line 161).

Annie also describe the consequences of having undesired responses to disclosure which then, in turn, made her more reluctant to disclose again: *'I think I got more hesitant in the way that people reacted'* (Annie, individual interview, line 927).

An internal struggle with self-concept:

This theme encapsulates the family's experience of how the JIA has threatened Annie's sense of self and identity. Annie explained feeling an internal conflict between who she wanted to be and what she wanted her identity to 'look like' and facing the reality of her circumstances. For Annie, the reality of the situation often arose when the JIA became visible to others, which resulted in a change in the way people related to her and, in turn, forced Annie to question who she was.

Annie explained that her first experience of a threat to her sense of self occurred at the same time the JIA became quite severe and her mobility became restricted. For example: *'my memory of it is being sat at school being captain of the rounders team but sitting out'* (Annie, individual interview, line 427). Annie identified this event as being very poignant for her as this

was her first memory of when JIA started to become a problem for her. This quote depicted Annie's perceived her prior identity as being a leader, being competent in sports and being good enough to be singled out as captain. She then described a sharp contrast of then still holding the title of 'captain' but not being able to identify with her previous self-concept of being 'sporty'.

The theme *an internal struggle with self-concept* also described Annie's longitudinal struggle to integrate these two identities of an 'ideal' self and a self which includes the JIA. This left her with a sense of a longing for, and holding on to an identity she could have had if she did not have a chronic health condition:

> *'I just always have wanted to be the person that was helping other people instead of being helped I don't know it's the same as I don't tell people about it when they meet me'* (Annie, individual interview, line 135).

Robert also reflected upon a loss of promise as JIA put a halt to Annie's previous identity as being 'sporty' and active.

> *'she just discovered she was that she was quite good at er jogging distance running…so she was always keen on PE lots of time spent doing sport and obviously that tailed off''* (Robert, family interview, line 100).

Annie described that disclosing the condition to others, encouraged others to observe, and act on, the parts of Annie's identity that she did not want to be made visible, such as relying on others or being vulnerable. For Annie, being able to keep the JIA hidden was seen as positive as she could preserve an identity that was more congruent with how she wanted to be seen by others, and therefore some elements could remain unchallenged:

> *'they look at me and they're like "oh" and then they're like "ok mental note made" and it's like "no you don't have to do anything"... it's like people are looking at you and thinking "oh I didn't know like there was something wrong with her" …which there's not'* (Annie, individual interview, line 156).

Annie described here that external events relating to the JIA, such as her being unable to manage some activities and how people then relate to her contradicted who she wanted to be:

> *'I was not happy there was a wheelchair I just thought it was so embarrassing…everyone knew and then everyone else question and it was just I didn't want people to view me as a person who was in a wheelchair'* (Annie)… '*like people thinking you're disabled'* (Emily)*... 'yeah'* (Annie)… '*and like you have problems'* (Emily. Family interview, line 1892).

Just getting on with it

This master theme captures the family's pursuit to prevent the JIA from dominating their family life and the way the family managed this was to keep moving forwards. This master theme encompasses two super-ordinate themes of *striving for normality* and *maintaining equilibrium*.

Striving for normality:

This super-ordinate theme encapsulates the family's experience that they are willing to acknowledge JIA as a condition that is present within the family, but they have made an active attempt to try and keep any disruption at bay by continuing with family life as normally as they can, without an excessive focus on the disruptions: '*you know in the grand scheme of things…it's an inconvenience and it's uncomfortable but it's not a showstopper'* (Robert, individual interview, line 716). Emily described her belief that the JIA demands respect from the family because it is not something that will just go away, however, the family can manoeuvre their way around it in a diplomatic and peaceful way:

> *'we haven't forgotten about it we know it's there and we we're not ignoring it but we've kind of just gone past it and carried on but we still know it's there and we're not ignoring it'* (Emily, individual interview, line 647).

The family described an understanding that JIA hasn't blocked or prevented the family from focusing on aspects of life beyond that of the arthritis.

In order to diminish or lessen the impact that JIA had on their lives and to maintain their sense of normality, the family described attempting to gain perspective by comparing their circumstances with other more significant life events they have experienced and with what they considered as more serious health conditions: '*it's just a condition that needs to be acknowledged and managed it's not she's got cancer you know it's just arthritis'* (Robert, individual interview, line 1047). Furthermore the family reflected that it was also Annie's wish that the family did not emphasise the arthritis which could prevent the family from maintaining that sense of normality and enabling JIA to dominate: *'she doesn't want us to make a big thing of it…she's quite a…private person'* (Robert, individual interview, line 1070).

In addition, Annie and Emily described what they believed the consequences would be if the family focussed excessively on the arthritis. Annie described that there would be a sense of loss or hopelessness if they did not look forwards and beyond the JIA. Emily explained that

normality was not always possible so in order to minimise these consequences the family were required to relinquish their sense of normality for short periods of time before 'setting off' again:

> *'we don't make a big fuss like we make a fuss but not a big one that it affects us in a big way and we can't set off and carry on again it's like we are almost doing little pit stops but then we set off and carry on again'* (Emily, individual interview, line 676).

The family described that there were times when normality was harder to sustain and therefore the family had no choice but to utilise the '*pit stops'* and allow JIA to temporarily disrupt family life. Thus, what was predominantly important to the family was how they dealt with the disruptions in order to return to normality as opposed to focussing energy on trying to prevent the disruptions occurring altogether, demonstrating the family's *striving* for that sense of normality.

Maintaining equilibrium:

This super-ordinate theme identified the family's active and deliberate attempts to maintain a stable and manageable family life alongside the turbulent nature of the JIA and its associated elements. *Maintaining equilibrium* was viewed as a method of shared family coping with JIA-related demands in order to minimise the impact JIA had on the family, to be able to get on with family life. Robert described his willingness to be flexible and open to new ideas in order to try and make JIA-related difficulties more manageable and less stressful: *'if there's something that can be done to ease the process and help…then we'll do it'* (Robert, individual interview, line 600). *Maintaining equilibrium* was made difficult by the family's experience that the JIA was largely unpredictable, meaning that the family found it difficult to envisage when flare-ups would occur and therefore more difficult for the family to continue as normal:

> *'during the bad times…our activities were restricted by have to bear in mind what [Annie's] capabilities at that particular time are…some days she might be fine and dandy you know and we can go off and yomp over the hills or whatever and other days…walking to the end of the car park would be a problem so…I mean it's not black and white it's not that variable but there are periods when she's fine and then periods when she's really not fine'* (Robert, family interview, line 553).

Emily described her participation in the process of maintaining equilibrium, which involved her and Robert staying strong for Annie. Emily explained that staying strong helped minimise their sense that the family could fall apart and suffer as a result of these adverse circumstances:

> '*person with arthritis need the support from friends and family to get through it all and the person and the friends will all need to stay strong for the person's sake so that they don't all like collapse in tears in front of them like "oh I want you to get better" and stuff" they all need to stay strong'* (Emily, individual interview, line 625).

Maintaining equilibrium was also described as being something that the family had attempted early on, following diagnosis. Robert described his experience of endeavouring to maintain family stability, during the time when the JIA became most disruptive, as the demands placed upon him was impacting upon other family relationships. The following quote from Robert describes the point at which he could identify that JIA was beginning to impinge on family life and coming to the conclusion that it was the right time to disclose to Emily:

> '*we need to do things gently and calmly you know hands need to be warmed up and you know she needs extra help doing buttons and bows and stuff so it's not that we're ignoring you it's just that at the moment [Annie] needs additional emphasis on this and this you know and sorry we can't go out for a family bike ride 'cos her knees are bad'* (Robert, family interview line 1082).

The theme *maintaining equilibrium* also reflected the family's experience of looking at the positive outcomes that have occurred as a direct result of JIA restoring the family's sense of balance and calmness. Emily described that the disruption in their normal routine and family balance offered them the opportunity to develop closer familial relationships, diminishing any sense that the family were held back by the arthritis. Paradoxically, this imbalance restored the balance and strengthened familial relationships. The following quote from Emily describes her experiences of times when Annie struggled with her mobility following her steroid injections:

> '*it's fun [Annie] can't run away I don't do anything bad (laughs) erm we spend more time together I like helping her'* (Emily, individual interview, line 727).

Negotiating understandings of JIA

This section outlines the results relating to the analysis of how the family negotiated their understandings of JIA. The family demonstrated that many of their experiences and understandings of JIA were shared and constructed at a family level. For example, *just getting on with it* was described as a coping strategy that worked for them as a family. All family members explained that this is how the family preferred to manage the JIA. However, while some experiences of JIA were shared and negotiated at a family level, this was not always the case at the individual level, leading to a tension between family member's accounts. For example, Emily described her experiences from her vantage point of being a sibling who had felt excluded from

some elements of Annie's experience of JIA. This meant that aspects of JIA had not been negotiated with her resulting in an unshared understanding of JIA and uncertainty and annoyance about this exclusion:

> *'I used to always try and peek in the kitchen door to see what it's like 'cos I didn't know what she was doing…I was always told to leave 'cos [Annie] didn't want me there so I was a bit annoyed about it but I understood so I left'* (Emily, individual interview, line 172).

Additionally the non-negotiated understandings of JIA led Emily to feel fear and anxiety about the changes that were occurring at the family level:

> '*I was a bit like erm I don't know scared that something serious was wrong with her…it was like scary that something could be wrong that I could lose her yeah…I was scared for her as well'* (Emily, individual interview, line 210).

On this individual level, Emily explained feeling that the family had experienced disruption with a sense that this would create family relationships were being torn apart*: 'felt like we were all kind of like tearing away'* (Emily, individual interview, line 340). Whereas Annie and Robert, who had negotiated their understandings with one another, understood more about the nature of JIA and that Annie's condition was not life-threatening. Emily described wanting to have more communication about JIA between family members to better able to negotiate JIA:

> *'I'm not really told much' (Emily)…'mmm would you like to be told more?' (Researcher)…'I don't know I think so…like it would help me understand what kind of state she is in not state but like how she is and stuff'* (Emily, individual interview, line 191).

Furthermore, Robert also described times when a shared construction and negotiation of JIA was more difficult at the family level. Robert described that him being the parent within the household, meant that he felt it was his responsibility to maintain the family's equilibrium, however, at times, he did not feel he had done enough in his negotiation with JIA to manage this: *'as a parent…you think why can't I do something about this you know there must be something that we haven't done yet or that's where the frustration comes'* (Robert individual interview, line 1163). This also described a breakdown of the family's strategy of *just getting on with it* which appeared to occur when individual concerns and worries about the JIA obstructed attempts to maintain that sense of normality.

The tension between the familial level and individual level of negotiation was also identified in the construction of stories communicated in the family interview. Within the

described accounts, the family expressed shared constructions of stories as they spontaneously negotiated their understandings of JIA with one another. This can be demonstrated by the family sharing storytelling, taking turns and completing one another's sentences to elicit and prompt accounts. For example:

> *'haven't done anything at all and by that time she was on crutches she'* (Robert)… '*I was on crutches at school'* (Annie)… '*she couldn't she couldn't walk around home let alone to school'* (Robert*)… 'and I couldn't write so I had a laptop for my lesson's* (Annie) … '*couldn't hold a pen and then within six or eight weeks of starting the new Adalimumab she scampered up [mountain name] with the scouts'* (Robert)… *'laughs…yeah it was really good for the first six months or so and then its deter deteriorated erm after that'* (Annie)… *'it's becoming less effective isn't it?'* (Robert)… *'Yeah'* (Annie. Family interview, line 347).

It can be demonstrated here that the family had previously negotiated some aspects of JIA which contributed to the shared storytelling. In contrast, there were times when the family did not have a shared understanding of a particular account and this was constructed and negotiated during the interview, which, in turn, spontaneously changed the meaning of that experience for individual family members:

> '*I was talking to the guy who was organising the group…and unbeknownst to [Annie] we'd arranged for a wheelchair to go out for all the gear'* (Robert)… *'did you allow that!? I hated that I was absolutely gutted when I arrived and there was a wheelchair for me'* (Annie)… *'but the camp was massive…and it was miles of rough terrain some of it wasn't it?'* (Robert)… *'yeah…but there was no way that I would…let myself be put in a wheelchair'* (Annie)… '*just as a back-up plan'* (Robert. Family interview, 1617).
>
> '*I wish he'd told me the the amount of times I've sat and complained about it and blaming them and he's just not said anything why*?' (Annie, individual interview, line 18).

For Annie, this event in the family interview also challenged her held belief that most aspects about the JIA had been shared between her and Robert, therefore potentially leading to a renegotiation of her own understanding of what is shared and not shared in relation to JIA.

Moreover, the process by which the family spoke of their experiences also identified stories that had been negotiated and shared at a family level, and those which were not. For example, there were times when the family used the term 'we' as opposed to 'I', which indicated that the family were 'in it together' as a unit:

> *'that's part of the problem…some of the drugs work for some people and they don't for others…that's why we have to go through this process'* (Robert, individual interview, line 860).

The following extract from Robert describes that he and Emily perceive the JIA to be part of the whole family's experience as opposed to just Annie. The family also utilised 'I' terms which were communicated within stories when there was an understanding that some aspects or views of their JIA experiences felt independent from other members:

> *'well obviously he wanted the best and he wanted me not to have the symptoms but he didn't understand what it was like to be on the medication urm yeah I mean obviously my opinion was the one that counted'* (Annie, family interview, 1354).

In this extract, Annie specifically discussed the individual positions held, which demonstrated a tension between the individual level experiences.

Equally, Robert shifted his experiential position on occasion, in which he moved from a 'we' that included family shared understandings, to that of a 'we' which removed him from the family to align himself with the medical professions: '*because she was reasonable active we all assumed she had tweaked something somewhere'* (Robert, family interview, line 192) and '*so I guess it comes down to whatever we think is best for for [Annie]'* (Robert, individual interview, line 235).

Case study 2: The Aitkin Family

Aitkin family pen portrait

The Aitkin family consisted of four members: Carly, the young person experiencing arthritis, Oliver, the elder brother of Carly, Michelle, the mother of Carly and Oliver, and finally Simon, the father of Carly and Oliver. All family members were white British in ethnicity and lived within the same household at the point of interview. Simon and Michelle knew of no other cases of JIA in the family and the family stated that they were not experiencing any other health-related difficulties at the point of interview.

The Aitkin family have lived with JIA for approximately 12 years and, as a result, saw it as part of 'normal' family life. The family described JIA as having minimal impact on them, with the exception of when Carly was younger and struggled to take her medication, which caused disruption to the family every week.

Carly was 17 years old at the time of being interviewed and was diagnosed with systemic onset arthritis at the age of five years. She is currently at college completing a course in media

make-up. Carly was suddenly taken ill one afternoon and became unwell for several weeks during the school summer holidays. She developed a rash and became virtually immobile due to severe pain in her joints. At the time of interview, Carly was prescribed weekly medication and mild non-prescriptive pain-relief when necessary. Historically, she also had steroid injections into the joints on average once every six months, which have now ceased due to her steady recovery Carly has not experienced a flare up where additional intervention is required for approximately one year. She has experienced some minimal amounts of pain, usually triggered by a cold or an infection. Carly described herself as being a 'normal' teenager whereby JIA has caused minimal disruption, but no more than this. Carly also explained that she strongly believes that within a year she will be discharged from the rheumatology service due to disease inactivity. She is currently in transition into adult services.

Oliver was 18 years old at the point of data collection and he is currently completing an apprenticeship in building and service engineering. Oliver explained that he had not been adversely affected by JIA and had could not recall a time when it had not been present in the family. He reported knowing very little about arthritis and he agreed with other family members concerning the belief that JIA was not an integral or central feature of the family's identity. Oliver had some input into the management of Carly's JIA but this has been minimal. Oliver's input was largely as a consequence of either Michelle or Simon being unavailable to support Carly, or when 'everyone else got fed up' with the management of JIA. He additionally helped Carly when she was less mobile by carrying her or driving her to college. During both the family interview and Carly's individual interview, it was mentioned that Carly often could not attend Oliver's rugby matches due to having to stand out in the cold, and Michelle would stay behind to look after her. While the family explained that they believed this had not adversely impacted on Oliver, there was an acknowledgement that this is something that Oliver had missed out on.

Simon, Carly's father, was in his late forties at the time of interview and works in engineering. Simon was not present for the first 40 minutes of the family interview. Simon explained that he had little input into any arthritis related care, and that his wife, Michelle, predominantly attended appointments and took a lead in the management of JIA. Simon described that most of his understandings of JIA was as a result of being informed by either Michelle or Carly. Simon explained that he was happy not to be involved in the management of JIA but would assist when necessary, such as taking Carly for some of her hospital appointments and encouraging her to adhere to her medication. It was Simon who also most frequently commented on JIA being on the periphery of family life and lacking importance. He frequently commented on the family continuing as 'normal' and had little doubt into JIA's trajectory of a complete absence of symptoms in the future.

Michelle, Carly's mother, is in her late forties and is a beauty therapist. Michelle has had full involvement in the care and management of the JIA from its onset. She attended the majority of the appointments with Carly, up until the age of 14, when Carly began attending appointments alone as part of the transition into adult services. Michelle described feeling shocked at the diagnosis as it challenged her previous understandings of the age at which people could develop arthritis. Michelle offered the most detailed stories around onset, diagnosis and treatment which she explained was due to her being more involved in JIA-related care than any other family member. Like the other family members, Michelle did not feel like JIA had any long term negative or adverse effects on the family although she reflected that the onset of the JIA and the quick deteriorating in Carly's health was very distressing for her. She recognised the disruptive nature of JIA, such as some impact on family holidays if JIA became active, and struggling with helping Carly to take her medication. However, she did not feel that these events had shaped their view of how they had managed living with a chronic health condition.

The family interview lasted for 120 minutes and every family member agreed to the use of all the acquired data for analysis. Due to receiving a limited amount of experiential data from Oliver, I did invite him to participate in an individual interview. Oliver declined the opportunity of this second interview and my sense is that he did not feel he could contribute any more information about JIA, as opposed to experiencing the interview as a negative or challenging process. Due to Michelle's extensive participation in the family interview, it was thought appropriate to invite the other family members for a second interview. This was to allow all experiences to be fully explored, and to minimise any bias in analysing one account over another. Carly's individual interview lasted for 65 minutes and Simon's for 45 minutes.

Reflections on the interviews

While waiting for Simon to return to the family home prior to starting the family interview, I spent a significant amount of time building a rapport with the other family members present. However, once the interview had started, I found it difficult to elicit accounts from Carly and Oliver. Oliver said very little during the family interview despite efforts to include him in the interview process. His responses were often brief which he explained was due to his limited involvement with the JIA. For most part, Oliver inputted by correcting others' accounts which demonstrated there were points when he was actively listening to the conversations. There were also points when Oliver appeared less engaged in the process, for example, at one point he walked out of the interview room.

Michelle was engaged in the interview process and made a significant number of contributions throughout. Michelle often seemed to mediate the conversations between other

family members to find a middle ground between discrepant stories and between the medical professionals and another family member's stories about those professionals. She also spoke about the more adverse events relating to JIA than other family members. Michelle often provided a richer amount of detail than any other family member. As a result, I found myself directing questions to Michelle during the family interview because of her greater recall of early events and her involvement in the management of the JIA. I was aware that this approach may have biased the data by representing only Michelle's perspective or experiences therefore representing an individual level of understanding as opposed to the family's. For this reason I did not invite Michelle to complete an additional individual interview.

Carly offered more details of her experiences during the individual interview and appeared more engaged with this interview process. One of the topics touched upon was her fear and disgust with her medication which aroused some anxiety during the interview and at one point she struggled to speak about her experiences. Furthermore, during both interviews Carly mentioned feeling unheard by professionals, so I was aware that this may have influenced the interview process through a power imbalance between us. During the interviews I made sure that I demonstrated active listening and asked Carly questions based on previous answers to demonstrate that I was listening to her.

Throughout the family and individual interview Simon was consistent in his belief that JIA had not had a major impact upon the family. I got the sense that he thought I was wanting something different from him such as a 'declaration' of any negative experiences. At one point he mentioned 'this might be an angle for you' indicating that he had his own preconceptions about what I needed from him as an interviewee. At the beginning of the individual interview, Simon also made a reference to the short time it may take to complete, believing that he did not know enough about JIA or that his stories would not take much time to tell.

The family had a dominant narrative of JIA not impacting significantly on family life and despite the family describing some periods when JIA did impact, I found it difficult to encourage them to elaborate on these particular stories. Additionally, due to difficulties pertaining to memory recall, often it was only Michelle and Simon who were able to offer detailed descriptions of the family's experiences.

Results of analysis

This case study explored the Aitkin family's experiences of JIA by way of four master themes: *A positive outlook, being 'normal', power and empowerment* and *medications: friend or foe?* (see Figure 2 for thematic map). A section entitled *negotiating understandings* will also be presented using example quotes to illustrate this concept.

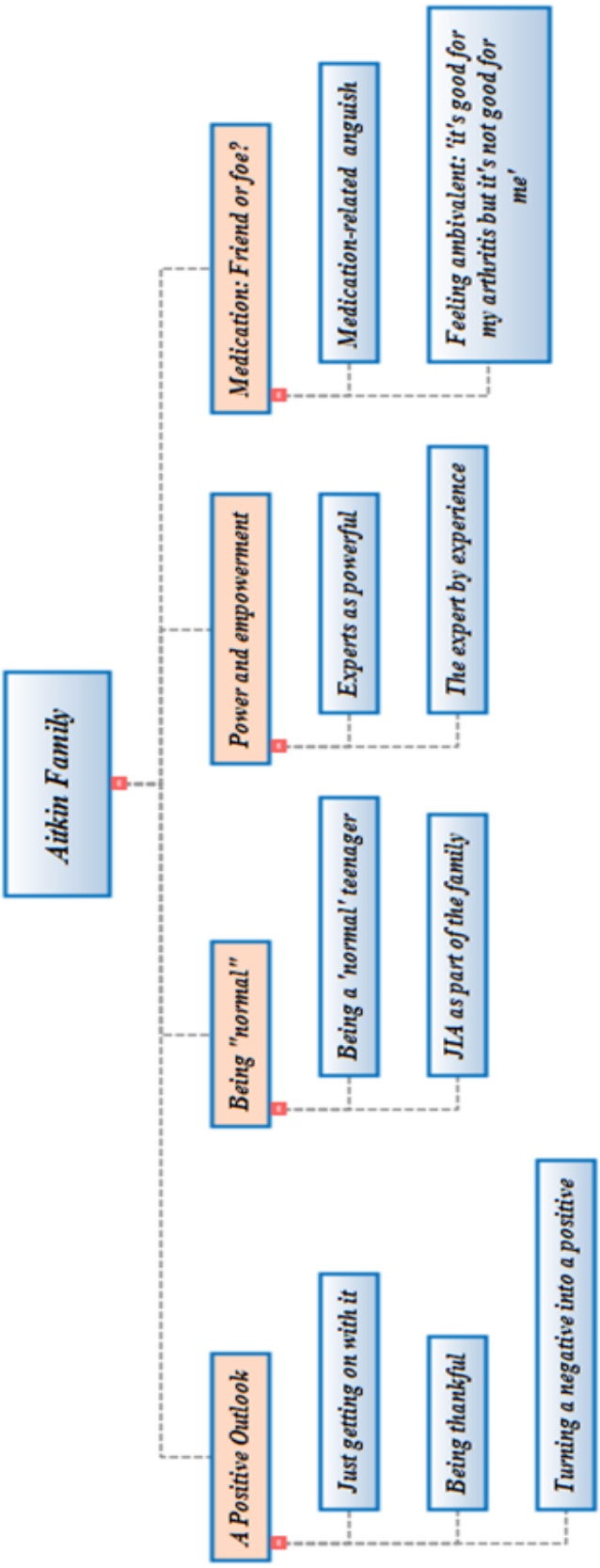

***Figure 2*. Aitkin family thematic map.**

A positive outlook

This master theme *'a positive outlook'* relates to the family's method of coping with JIA related experiences by a described optimistic as opposed to pessimistic stance. The super-ordinate themes that encompass this master theme include: *Just getting on with it, being thankful,* and *turning the negative into a positive.*

Just getting on with it:

This super-ordinate theme *just getting on with it* addresses the family's approach to JIA in which they described their experiences of the condition as not dominating family life. Their reported approach was that one cannot be passive to JIA and allow it to dominate:

> *'from my point of view…I think some people might just let it it rule their lives a little bit and just err just go on about it you know but you've got to sort of rise above it a little bit and err just get on with things you know'* (Simon, individual interview, line 670).

The family outlined *'getting on with it'* as both an established outlook for them as a family, but also, on occasion, there was a need for them to make an active effort to maintain a sense of normality in order to not allow JIA to overrun family life. The former concept was described by every family member:

> *'you didn't really need that sort of support did you? I don't think or we didn't think that we did…we were just getting on we were getting on happily enough anyway'* (Simon, family interview, line 2773).

The family felt they did not focus on the JIA and indicated that the severity of the condition was not enough to warrant any dramatic family alterations. The family had a well-established routine prepared if a flare-up did occur, contributing to their experience that they would *just get on with it.* This is demonstrated by Carly:

> *'say my knee was hurting…and it had been all day I'd just go to bed and I'd take some ibuprofen…then tomorrow if it was still hurting I'd tell my mum and then I'd start taking it every four hours and then if it still carried on hurting we'd probably get in touch with the hospital…and then they'd see how it was by the end of the week, if it's still unbearable then…I'd go in and they'd give me a…joint injection'* (Carly, individual interview, line 256).

The routines set in place at times when flare-ups occurred, helped the family accommodate the JIA, which in turn, facilitated a view that JIA flare-ups were part of 'normal' family life. The family described always perceiving that any JIA-related flare-up would cause a temporary

disruption to the family but always had an assumption that 'normal' family life would shortly resume.

> *'yeah that's like I say it's been beauty of it…whenever it's got bad we've been able to take her to hospital and they've within like I say within sort of I would say probably a day of her getting home again she's been right which you know everything's back to normal err like I say there's been trauma just once a week of err having to take this medicine you know but you couldn't really say that it's been err unbearable or anything like that could you'* (Simon, family interview, line 2378).

In contrast, there were times when these routines did not always work in achieving a sense of normality. The family described that these routines failed when the symptoms were at their worst:

> *'we didn't really do anything that summer because she were too poorly we tried to do things the odd time but we had to give up 'cos she couldn't manage'* (Michelle, family interview, line 268).

This quote describes the family's efforts to intersperse some typical activities during a time in the year when the family would usually be more active. Michelle described that if they were unable to maintain a sense of normality then the family made a conscious effort not to focus or dwell on these changes to maintain focus on the times when JIA did not disrupt family life: *'I think you just have to take it in your stride really and not erm…not dwell on it…happening'* (Michelle, family interview, line 2403).

Being thankful:

This super-ordinate theme encompasses the family's beliefs that they all view themselves as being in a fortunate position despite of some of the limitations and difficulties that have arisen as a result of JIA. For example, the family recognised that they were fortunate that the medication had always worked for Carly and the family had not needed to change their lifestyle or choices in any way:

> *'we haven't ever stopped anything really have we?...no plans have changed 'cos err…these drugs have done so well'* (Simon, family interview, line 2501).

The family described that having a *positive outlook* encouraged them to maintain perspective and see the bigger picture regarding their circumstances. This enabled the family to view JIA-related disruptions as being minimal, manageable and bearable, and therefore, not enabling JIA to dominate the family:

> *'there's been trauma for just once a week of err having to take this medicine…you know but you couldn't really say that it's been unbearable…it's just one of those things that you have to do once a week'* (Simon, family interview, line 2384).

In addition, the family described that they are able to *'count our blessings'* by making comparisons with other people and families who they consider as being worse off than them, including other people experiencing early onset arthritis and other life-threatening conditions. Thus, saw their position as favourable in comparison:

> *'when you go to a children's hospital where…you wouldn't go otherwise and you see some of these kids…and you think "bloody hell" you know there's there's a lot worse isn't there'* (Simon, family interview, line 2827).

The family reminded themselves that Carly's condition is chronic and not terminal, further strengthening their beliefs that they thought themselves as being lucky. A further consequence of thinking from this perspective was that the family felt that that any additional emphasis they placed upon the JIA would be unfair and unjust:

> *'when I was like six I'd go into hospital and see kids that have totally 10 times worse than what I had…that's just what's made me not…care that I'm missing out on things I've just thought oh well next week I'll be able to do it so it's all going to be alright'* (Carly, individual interview, line 110).

Turning a negative into a positive:

This super-ordinate theme of *turning a negative into a positive* encompasses a family strategy of reframing and thinking optimistically about their experiences, even when they have acknowledged that they have faced some challenging circumstances. This can be described by a quote from Michelle:

> *'she did have to go back in a pushchair for a little while…between five and six which was a little bit, a little bit embarrassing for you I think…luckily because she's so small…I think it didn't look out of place because she were always on the small side'* (Michelle, family interview, line 436).

Furthermore, the family also turned more challenging events into a positive experience by viewing them as comedic and humorous, which encouraged the family to think of past events in a light-hearted way. This can be demonstrated using an extract form the family interview during a

conversation about Carly fainting before undergoing general anaesthetic to have steroid injections into her joints:

> *'I think you woke up and we all had…your legs in the air didn't we'* (Michelle)…*(family laugh)… 'yeah it were really funny and then you started reading me "Where's Wally" we were playing "Where's Wally" when I was going to sleep 'cos it were so childish and we were laughing about how childish it was'* (Carly. Family interview, line 1813).

Carly explained that the family had always accommodated JIA in a light-hearted way:

> *'we've always just it's always just been like a joke hasn't it "oh go on carry her carry her t' car" so he's [Oliver] like try and carry me t' car and stuff we haven't really been like it's not really been serious has it?...it's just been like kind of a joke like carry her to the car 'cos it's funny'* (Carly, family interview, line 3383).

In contrast, *turning a negative into a positive* was more difficult for Michelle and Simon at the point in which there was a limited amount of certainty regarding disease remission. They described that this resulted in a preoccupation with Carly's future and a worry about permanent mobility difficulties.

> *'when we had to have a wheelchair it were…a worrying part for you know because it sort of gets worse and worse and worse then the next things she's getting pushed about in a wheelchair you think "oh I hope this isn't a sign of things to come" and…having to be pushed around in a wheelchair for the rest of her life…it did start to become a concern you start thinking about it a little bit'* (Simon, individual interview, line 237).

Simon described the invasion of doubt and having his hoped dashed as the disease remission process slowed, leaving him questioning if the disease activity would conclude altogether or continue into Carly's adulthood. Furthermore while the family identified *a positive outlook* and a positive reframe was a useful strategy for them at a family level, at an individual level Simon identified that this strategy was sometimes harder to use. This was especially the case when there was uncertainty about their future. Hence turning a negative into a positive was not always an established narrative for Simon and Michelle as an individual level.

Being 'normal'

This master theme encapsulates the family's sense that they are an 'average' family whose identity has not altered or is different from any other family as a result of their experiences with JIA. The super-ordinate themes within this master theme are: *being a 'normal' teenager* and *JIA as part of the family*.

Being a 'normal' teenager:

This super-ordinate theme reflects the family's experience that Carly has capabilities and a quality of life comparable to that of her peers; and has not principally being 'disadvantaged' as a result of being diagnosed with a chronic health condition:

> *'she's even been skiing with school so you know we…can't really complain…it's not been that bad has it? ..you did sport didn't you? …that's the beauty of these drugs that she takes…when she's right she's like everybody else which is good'* (Simon, family interview, line 2352).

The family discussed the importance of not '*wrapping Carly in cotton wool'* allowing her to take part in the hobbies, holidays and activities that she enjoyed, as a way of supporting her to have a 'normal' teenage life. This was encouraged by Michelle and Simon, even on occasions when they believed Carly could struggle with her choice of activity.

Being a normal teenager also encapsulates Carly's sense of being her 'normal' self at times when JIA imposed on her mobility or had the potential to threaten acceptance amongst her peers:

> *'I've not just sat there and thought I wish I would be doing it 'cos they've always gave me something else to do so I've always felt a part'* (Carly, individual interview, line 89).

Carly described finding alternative ways to be 'normal' that meant she did not feel different or defined by her health condition.

The family also described how the periods in between experiencing active symptoms had increased exponentially over the previous two years which additionally contributed to the family feeling as if Carly was like any other teenager. Simon and Oliver described forgetting that she has JIA, and indeed, Carly also overlooked the condition, as described by the following quote from Carly discussing her medication regimes:

> *'…pretty slack at it it's usually like the Sunday but I have it once a week just whenever, it's not like every single Wednesday it's like some Wednesdays some Mondays…just when I can remember in the week'* (Carly, family interview, line 1611).

Indeed, the extended periods between active symptoms has led Carly to believe that in two years' time she would no longer have a diagnosis of JIA.

In contrast, Michelle described a divergent narrative that differed to that of the family level narrative of Carly *being a normal teenager*. She explained that she sometimes viewed Carly

as being more vulnerable than, and treating her differently to, Oliver and her peers as a direct result of the arthritis:

> *'when I say to [Oliver] like the other day he went out in his tee-shirt I were like "put a jumper on you'll be cold" when I'm saying it to [Carly] I'm thinking "if you get cold you're going to suffer you know" I really don't care if [Oliver] gets cold but yeah I I'd probably say the same thing but there's a different reason behind it…[Carly] shouldn't be going out …without being properly prepared for the weather…because of her illness so that's the difference'* (Michelle, family interview, line 3191).

The effect of this perceived vulnerability was to treat Carly differently to Oliver. While at a family level a narrative about Carly being a normal teenager was well established, this was not always the case at an individual level, whereby family members, including Carly, acknowledged that there were allowances made as a result of the JIA.

JIA as part of the family:

This super-ordinate theme refers to the family's experience of being unable to separate JIA from their other family experiences, Michelle and Simon described JIA as something they assimilated into their family life many years ago and JIA-related care became 'normal' for them as opposed to it defining them as a family:

> *'just getting toothpaste ready for her dressing her sometimes if her wrists and elbows were swollen you know I had to dress her erm can't think it's become so normal really'* (Michelle, family interview, line 389).

In this quote, Michelle described that JIA management had become a part of routine family life as the family engaged in normalising processes such as by finding ways to accommodate the JIA and making JIA related care part of the family's routine.

In comparison Carly and Oliver do not distinguish between the two experiences that Michelle and Simon described, but rather they reflected upon the fact that they did not have different experiences to draw upon. For example: *'it just doesn't feel any different and I can't remember it being bad so it hasn't really changed or owt'* (Oliver, family interview, line 2548). Both Carly and Oliver described that they have only ever experienced family life with the JIA therefore they were unable to make comparisons of their experiences. Michelle and Simon described that, historically, there were times when Carly did struggle and was admitted into hospital, however, Oliver reported not recalling a time when it had been different, indicating that the assimilation strategies perhaps were effective at a family and individual level.

Power and empowerment

This master theme relates to the family's experience of feeling in control over the JIA which contributed to their sense of feeling empowered. This theme also describes when threats to empowerment resulted in an impact on well-being. This master theme encapsulates three super-ordinate themes entitled: *experts as powerful, expert by experience* and *the trusting relationships.*

Experts as powerful:

This super-ordinate theme relates to Michelle and Simon's relationship to the 'powerful' professionals involved in Carly's care, and meanings they attributed to their 'expert' position. Michelle and Simon expressed their view that Carly's remission from JIA and her recovery was fully attributable to the medical professionals involved in her care. They placed a significant amount of importance to the influence and power of the professionals and did not want to deviate from their advice for fear of precipitating arthritis flare-ups or making 'bad' decisions that might have consequences for future well-being. They also described feeling contained by the professionals and comfortable with relinquishing decisions about treatment, which meant the family were content not to try and exert any control. The parents felt they did not have the knowledge or educational attainments to question or contradict the advice given to them:

> *'there's people go spend six years at university learning about it so they're going to know a lot more about things than you do aren't they?...I think erm just go with the advice you're given and make sure you give the medication that they ask you to give them erm and just go with the flow really…that's the way to do it'* (Simon, individual interview, line 1081).

This theme also relates to Simon's fears about other possible consequences if the family deviated from expert advice. He described the potential for the onset of familial and wider system disputes concerning who would believe they knew what was best for the young person with arthritis:

> *'go with what they tell you and…I mean I think maybe…that's where it might go off the rail if some people decide that they know better and that's…where the parents will fall out with each other and they'll fall out with the doctors and it won't go right for the kid'* (Simon, individual interview, line 1070).

Simon described that it is easier for the family if the professionals make the decisions in relation to treatment, identifying that the family is better off relinquishing decision making because it maintains equilibrium within the family and avoids blame if the any member makes a 'wrong'

decision which will impact upon the young person with the chronic condition. Michelle described not always agreeing with the professions when it came to decision making but aligned herself with the narrative about trusting their decisions:

> *'I didn't really want to stop going with her I did think she was slightly too young but they decided…she decided with the clinicians that that's what…they wanted so sort of guessed it were nothing to do with me anymore really...she was starting to grow up and know her own mind so so long as they were instigating it then…we've always been happy to be led by the clinicians…I can't say that I would have thought of it this early but you know I knew I knew it would come eventually'* (Michelle, family interview, line 3285).

The *power of professionals* also relates to the long-term trusting relationships the family had with the medical professionals that contributed to their experience that the professional should always be listened to. Michelle and Simon felt complete trust and respect with the arthritis specialists as they had proved to the family that their advice was correct and accurate. This led the family to put complete faith in the professionals making the decisions: *'we have always trusted put our faith into what the doctors say if they said it was ok then it was ok'* (Simon, family interview, line 2160). They also identified how the trusting relationships resulted in the family feeling supported and spoke about the contributions to the development of these relationships which included: a quick response to the flare-ups, the longevity of the relationships, giving advice and the effectiveness of the medication prescribed to Carly, for example:

> *'they've been good have the nurses you know they ring you back straight away and give you…advice if you need it really supportive I think aren't they? and if they has to go in for an injection they've always come up on the ward*' (Michelle, family interview, line 2630).

The expert by experience:

In contrast to *the power of the professionals,* Carly described that her experience of having a chronic health condition also made her an expert. This was an experience divergent to the family level strategy that the professionals are the experts and should always be listened to. *The expert by experience* encapsulates Carly's understanding that she is also an expert on her own body's idiosyncratic response to JIA, which over time, she had learnt to listen and respond to her body's needs. Moreover, Carly's understanding of her body, on occasion, contributed to discrepant views to that of the medical professionals, leaving her with a sense of being unheard and the potential to feel disempowered:

> *'they'd seen my blood and it was high...my blood had like high dose of…inflammation…so erm I was poorly and I told them when I got that blood test I was poorly and it always sets me off when I'm poorly and it was just I was absolutely fine and they were like "oh we're going to have to up your medicine" and I was like "no 'cos I don't need my medicine" and they wouldn't believe me so I was like "well do another blood test and I bet it will come back normal" and it did'* (Carly, family interview, line 2184).

Carly identified that despite the professionals having medical knowledge, she was able to exert her own expert knowledge of herself, which maintained her sense of control over the situation and her body. Carly also described managing the power imbalances that existed with the professionals by needing to provide proof that she was able to manage the condition. The family identified this as being more difficult when the disease activity was not always visible and when no-one else can share her experience:

> *'I thought…listen to the expert 'cos you do don't you but erm none of us have got arthritis so we don't know…we haven't had to live with it have we?'* (Michelle, family interview, line 2251).

Michelle additionally recognised that potentially, Carly not listening to the professionals could cause disruption because her views sometimes contradicted those of the professionals. Michelle, however, also acknowledged that the family's strategy of perceiving the professional as powerful does not fit with Carly's experiences.

This theme, *expert by experience,* also encompasses Carly's experience of transition into adult services whereby she expressed receiving conflicting messages between being required to take more control over the JIA management, which was considered appropriate for her age, and not being listened to or trusted to manage the JIA. Carly explained that the newly developing relationships with the adult professionals made her feeling less in control:

> *'I went from Dr [name] one week and then like the next month Dr [name] but still in the same building and…in between switching to Dr [name] I had random other doctors as well ..I didn't even know they were changing me until like now I'm getting moved on to this one I realised that's what they were doing'* (Carly, individual interview, line 829).

This changing dynamics of relationships and lack of control of decision making therefore, on occasion, made it difficult for Carly to take charge of the JIA to become an 'expert'. Furthermore, the wavering trust also diverged from the family narrative that the professionals were always trustworthy and reliable.

Medication: friend or foe

This master theme describes the family's experience of medication as having both a positive and negative impact on their well-being, which resulted in the family having mixed views about Carly's treatment. This master theme was split into two super-ordinate themes: *medication-related anguish* and *feeling ambivalent: 'it's good for my body but it's not good for me'*.

Medication-related anguish:

This super-ordinate theme relates to the family's distress before and during the point at which Carly was required to take her weekly medication. The family experienced an impending dread as the time for Carly to take her medication drew closer:

> *'yeah it took hours didn't it? Absolutely hours every Sunday we all dreaded Sunday night. who's turn is it this week… we all dreaded it as much as she did'* (Michelle, family interview, line 2079).

The family described that the dread would lead to frustration and a struggle to empathise with Carly because they knew the medication would ultimately help her but Carly would not see this logic while feeling so distressed:

> *'I remember erm when watching her doing it once and she tried to trick me 'cos she had the tablet in her hand and she's erm go like that then drink water and pull a funny face and I knew she still had it in her hand'* (Oliver, family interview, line 2041).

The family additionally described finding means to defuse any pre-empted, and actual, distress by bartering and bribing Carly into taking her medication in order to positively reinforce her efforts for managing the medication. On occasion, the family would force Carly to take her medication leaving the family feeling 'traumatised': *'we ended up having…we had to like squeeze her cheeks didn't I and push it in that were quite traumatic wasn't it'* (Michelle, family interview, line 1375).

The family also described feeling like the *'bad one'* for attempting to encourage Carly to take her medication resulting in further anguish and distress: *'we ended up having to sort of get pin her down and then open her mouth and pour this yoghurt in down her throat'* (Michelle, individual interview, line 1396). Michelle described this as being *'traumatic'* drawing upon her experience of feeling torn between trying to comply with what had been recommended by the professionals, but also wanting to better support and understand Carly: *'you had to take it to make you better…and that's what you couldn't sort of seem to understand wasn't it'* (Michelle, family interview, line 2112). Michelle observed a further battle of wanting Carly to adhere to the

medication and abide by the advice from professionals, but perceived Carly to not recognise the importance of this advice.

Feeling ambivalent: 'it's good for my arthritis but it's not good for me':

This super-ordinate theme comprises the family's experience of ambivalence toward medical interventions as medication is a required part of disease control for Carly, and was always effective in inflammation suppression; however, there was also an understanding amongst members that it did not facilitate or encourage well-being.

The family experienced a shared viewpoint that the medication as mostly effective in managing the JIA symptoms. This also contributed to the belief that JIA was a temporary problem, was fixable and a controllable condition. This can be demonstrated by the following quote from Simon:

> *'as soon as she's come out of the [hospital name] like and she's been alright again you know they've…fixed they've sorted it out haven't they…she's never had to go back again afterwards…it's been sorted for the next six months a year hasn't it'* (Simon, family interview, line 1238).

Simon offered the perspective that the medication offered some sense of predictability and control over the arthritis. He also commented that his expectations for the medication had always been met and so reinforcing his belief that the medication was beneficial for Carly.

However, while the outcomes of taking the medication contributed towards the family's sense of feeling contained and safe, the process of Carly taking her medication and receiving treatment, in contrast, created a sense of unease within the family. The family identified these 'paddies' as having an impact upon each member as they coaxed and persuaded Carly to take her medication.

> *'it got bad at one point didn't it with big screaming paddies and all sorts wan't it'* (Simon*)…'yeah….so we thought that maybe if we crushed it up and gave it to her in a yoghurt she wouldn't know (laughs)'* (Michelle. Family interview, line 1387).

In addition, Michelle and Simon described themselves as feeling like helpless observers during hospital procedures when Carly required general anaesthetic, which rendered them feeling distressed: *'I've been through with her a few times watching them put her to sleep that were traumatic'* (Michelle, family interview, line 1824).

The theme *feeling ambivalent* also reflects Carly's ambivalence towards her treatment to which she felt this would not change due to the longevity of her experienced anxiety and 'failed' attempts:

> *'it's awful it's I just can't do it… I've tried with like tic tacs with stuff like this and even medicine but I just can't do it like I've just but it's all built up and I've just mentally just come this like…I just can't take them'* (Carly, individual interview, line 346).

This left Carly reflecting on feeling torn between having to take the medication as it was needed and necessary for her recovery, but also knowing it caused her significant distress and anxiety. Carly described an internal struggle between what was good for her physically and how to accept this in light of the distress it causes her:

> *'I take this one that's like completely ruined like makes me feel sick makes me like it still does it now...it makes my mouth really phlegmy so I have to spit like constantly….Oh I don't know…but it was good for my arthritis but it wasn't good for me…made me feel sick got me wound up and I just don't not liked it ever since'* (Carly, individual interview, line 421).

Negotiating understandings

This section will outline the observed family processes that occurred during the family interview that contributed towards how the family spoke about and negotiated their experiences of JIA. Firstly, negotiating understandings for the family describes how each of the individual member's experiences and recall of events in relation to the JIA created divergent narratives that were difficult to negotiate at a family level. The following extract provides an example of this concept:

> *'do you remember losing your hair?'* (Michelle)… *'I remember getting a teddy for losing my hair, I don't remember losing the hair'* (Carly)… '*there's photographs of you isn't there at the zoo and you've got this like horrible hairstyle'* (Michelle)… *'mmm but no I just remember getting the teddy 'cos I lost my hair'* (Carly)… *'I think you were a bit upset about the hair loss 'cos she had really long dark hair'* (Michelle)… '*umm I just remember getting the teddy…it were a dog with a bandage on it'* (Carly)… '*I can't remember'* (Michelle. Family interview, line 192).

This extract demonstrates that, on occasion, the family's divergent accounts were difficult to negotiate due to the different meanings attributed to their experiences, in this case, of Carly losing her hair. Carly's recall of events was related to the significance of her receiving a gift at around the time of her losing her hair. Carly states 'the hair' indicating a distancing of the experience Michelle is attempting to share with her. In contrast, Michelle does not recall the teddy, but recalls

Carly being upset and a haircut that made her stand out. Potentially this reflected Michelle's distress at Carly's hair loss as opposed to Carly's distress.

In contrast, divergent family accounts discussed during the joint interview also meant the family could renegotiate and reconstruct their individual meanings to develop a shared family meaning of their experiences: This can be demonstrated by the following extract:

> *'they said give her it by liquid if she won't take her tablet but…the first few weeks she were really good and then that got like…you had to chase her round the house' (Michelle)… 'but that's probably why she had to keep going back to hospital every sort of month'…(Simon)… 'Yeah' (Michelle)… ''cos she probably wasn't taking it…'cos even half of the time when you thought she was taking it she probably wasn't was she?' (Simon)… 'she wasn't no if you didn't shut close her mouth in time she'd just go bleugh' (Michelle)… 'so really in the long run you were probably better off with the injections aren't you'* (Simon. Family interview, line 1436).

During the interview the family attempted to draw together their experiences of JIA and develop formulations and hypotheses of their understandings, in order to better understand the condition. Based on information from other family members' accounts, Simon attempted to find some sense in the experience in order to try and understand why there were occasions that Carly's medication was not always effective.

Synthesis of Results

This section outlines the synthesis of the case study data. , Four themes emerged from the synthesis of the data: *just getting on with it and maintaining a sense of normality, battling, fighting and the negotiation of power, transitioning* and *JIA as a hidden condition*. The negotiation of understandings were also synthesised (see *Figure 3* for thematic map).

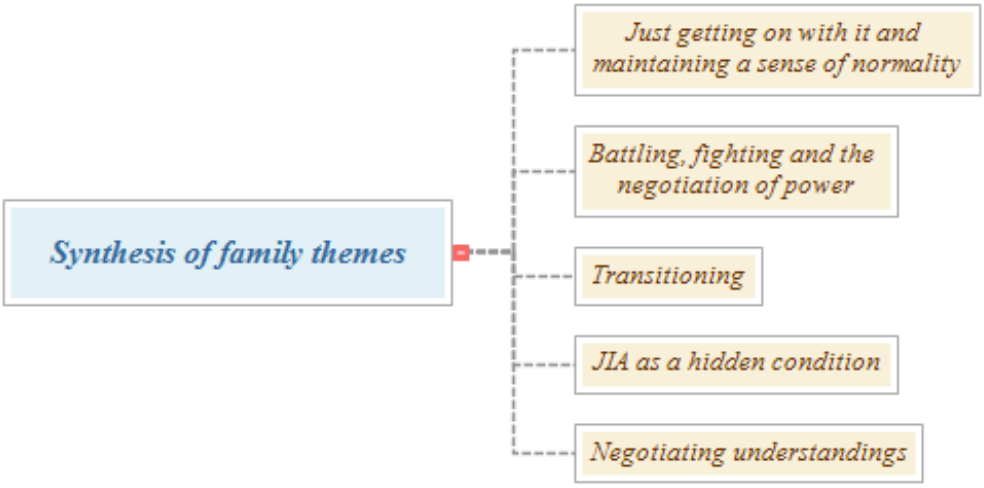

Figure 3. **Synthesis of family data.**

Just getting on with it and maintaining a sense of normality:

Just getting on with it was a theme that was identified in both families. Both families stressed the importance of not allowing JIA to dominate family life and made active attempts not to dwell on anything JIA related:

> *'we've just sort of like I say we've really taken it in our stride and not you don't dwell on it you don't been upset a few times you know seeing her in pain but you know wishing that she didn't have to cope with it but I've always thought to myself like I said earlier you know some families really you know'* (Michelle)… '*got it a lot worse haven't they?*' (Simon)… '*got it a lot worse yeah so you sort of count your blessings really erm you know she's got a chronic illness not a terminal illness so that's how I've always tried to look at it really you know*' (Michelle. Family interview, line 2796).

An important part of this was so the families could avoid being defined by the JIA and in that way, maintain a sense of normality. Both families additionally made comparisons against other conditions or families they viewed to be worse off than them, which was a strategy they seemed to use in order to maintain perspective of their circumstances.

> *'it's because I don't know it's it's just a condition that needs to be acknowledged and managed it it's not you know it's not she's got cancer you know it's just she's got arthritis'* (Robert, individual interview, line 1046).

While this was identified as a family level method of coping with living with a chronic health condition, the families also demonstrated that *just getting on with it* was more difficult at an individual level whereby fears and worries would creep up on them, making it more difficult to use this strategy, and think positively about their situation. This meant that there were divergent narratives at a family level and at an individual level. On an individual level the families described being more conscious and worrying more about the well-being of the family member with the chronic health condition. Examples quotes which demonstrate that just getting on with it was a less visible at an individual level are illustrated below:

> *'when her hands are bad you know you know she's got naturally curly hair…so it's hard for her to hold…the brushes you know when her hands are bad erm when she was in that period where…symptoms were getting bad she struggled to get dressed sometimes you know and it was difficult watching her suffer…in fact it was very very frustrating as as we probably mentioned last time we had disagreements…'* (Robert, individual interview, line 1123).

> *'just a bit, again sort of a bit disappointing 'cos that's the I think err that's the err thing err with it being junior arthritis you're always hoping that it's going to go away, then you're thinking when is it going to go away and then they take her off medication and you think ahh maybe it's going to go away and then when she's back on the medication then oh no it's not going to go away'* (Simon, individual interview, line 623).

The families also described a joint narrative of times when it was acceptable that JIA could disrupt family life, and therefore just getting on with things and maintaining normality was still difficult, but tolerable. The families were aware that flare-ups could happen at any time as JIA was not always predictable and so maintaining normality became difficult. However, following steroid injection treatment or more severe flare-ups, the families were prepared for a period of family disruption and enabled JIA to govern the family temporarily. Emily described enjoying these predicted family disruptions because it meant a time for strengthening family bonds:

> *'we don't make a big fuss like we make a fuss but not a big one that it affects us in a big way and we can't set off and carry on again it's like we are almost doing little pit stops but then we set off and carry on again'* (Emily, individual interview, line 676).

> '*it's fun [Annie] can't run away I don't do anything bad (laughs) erm we spend more time together I like helping her'* (Emily, individual interview, line 727).

From prior experience, the Aitkin family, in particular, were aware that these more significant disruptions were temporary and therefore it was easier for them to relinquish 'normality' until it again became unacceptable for the family to negotiate with JIA in this way:

> *'like I say now it's just a matter of knowing when she's she's just a bit under the weather and it will blow over or when when it's no this isn't just 'cos you're under the weather it's ah it's another sort of blow up so you have to go to the hospital* (Simon)… *'no she just we just she just rests you know she's she'll stay in bed for a few days'* (Michelle. Family interview, 2304).

There were also discrepant views amongst the families with their experiences of maintaining normality. While it was evident that both the families had made active attempts to not allow JIA to dominate family life, the Hunter family described experiencing more significant disruptions than the Aitkin family did:

> *'I think the most…after I've had 20-odd steroid injections because it just takes so long to be able to walk around and just things like that because in..'* (Annie)… *'pick up a glass of water'* (Emily)… *'yeah because if you can't hold a glass…it's just it takes so long to get out of bed and then be able to move your hands properly'* (Annie)… *'we hold the glass and she drinks through a '* (Emily)… *'but it's things like you brought me a bun back from school and I was like "oh thanks" I can't take the wrapper off or anything…I couldn't hold it'* (Annie. Family interview, line 1236).

The Aitkin family described for most part, JIA impacted very little on the family. This was especially the case in the recent past because Carly had experienced few flare-ups, therefore the family, in turn, experienced fewer disruptions:

> *'sometimes if they have been more poorly you have to make decisions on which way you go haven't you? It's not affected our work, it's not affected our holidays, it's not affected our day to day business just every now and again it's been a little trip to the hospital but sort of few and far between really'* (Simon, family interview, line 3125).

Battling, fighting and the negotiation of power:

Both the Hunter and the Aitkin family described that much of the distress relating to JIA was a result of the physical and/or psychological difficulties they had experienced with medication. Both families described individual internal battles and intra-familial battles directly relating to the process of taking medication:

> *'we ended up having to…pin her down and then …open her mouth and pour this yoghurt…down her throat…'* (Michelle)… *'even the, even the injections made me feel sick'* (Carly)… *'I know I know it's nasty stuff isn't it…it were awful weren't it it was about 2 hours wasn't it every Sunday…she wouldn't she didn't want anyone else doing it but she didn't want me doing it either…awful awful I think we had to like pin her down then didn't we she would be kicking and screaming and "don't you come near me help help"'* (Michelle. Family interview, line 1397).

Both families also shared a view that the medications helped relieve, at least some, of the JIA symptoms but the young people with JIA described divergent beliefs that quality of life also incorporated psychological well-being. Each family member had an awareness of these divergent views and had been discussed within the families:

> *'she was definitely much happier when she was symptom free but that's understandable she's frustrated now that the symptoms are back…so I think her relationship with it is definitely up and down you know depending on the success of the treatment at that particular time…I think it's, it's perfectly normal really…you know she's bound to be upset with it when it's err not going well'* (Robert, individual interview, line 692).

Annie and Carly both described ambivalence towards the medication as they were aware it was an intervention that would improve their symptoms, but felt the psychological distress of taking it outweighed the benefits of the medication outcomes. Annie and Carly additionally described having adverse physical responses to taking the medication. Annie explained that her body was fighting against the medication as a result of its side effects. Carly, on the other hand, explained having a physical reaction which resulted from her psychological rejection of the medication. Their reactions to the medication resulted in both Annie and Carly undergoing a period of time whereby medication was not prescribed for the JIA:

> *'well obviously he wanted the best and he wanted me not to have the symptoms but he didn't understand what it was like to be on the medication urm yeah I mean obviously my opinion was the one that counted (laughs) which was lucky because obviously the doctors wouldn't put me on something I didn't want'* (Annie)… *'I mean we spent probably 12 months in and out of clinics and going over to [Annie] I mean for 12 mon no it wasn't it was two years wasn't it that you were off it completely'* (Robert. Family interview, line 1354).

As seen in the quote above, the families described their battles with Annie and Carly over the procedures, which was the most significant divergent narrative within the families between parent and child. In both families, the parents described the importance they placed upon the medication.

Both Robert and Simon described their worries for Annie and Carly's future and felt that taking medication protected them against permanent mobility problems:

> *'well it's it's quite depressing really because the longer the condition goes on obviously the the greater the chances of permanent damage within the joints are you know we were always told if they can get on top of it and stop it then there's every chance that there is no damage in the joints'* (Simon, individual interview, line 400).

Annie and Carly placed a greater precedence on their psychological well-being, as opposed to physical well-being, in the moment of taking the medication, which meant that during those situations, these divergent narratives were most evident and an internal struggle for Annie and Carly:

> *'I would take it only if I absolutely have to like with my methotrexate I do take that. I don't like it but like I don't know, I can, but I don't, I don't like the thought of it at all. And I don't think I'd be able to take any other tablet, I just know I have to take that one'* (Carly, individual interview, line 388).

A divergent experience between the families was their expressed hope about the effectiveness of the medications. The Aitkin family shared a narrative that the treatments were successful and a 'cure' and therefore, held the belief that Carly would have a stable and unaffected future, resulting in a sense that their battle had been won:

> *'generally wherever whenever there has been a sort of poorly time…'*(Simon)… 'o*h yeah they're straight…'* (Michelle)… *'as as soon and she's come out of the [hospital name] like and she's been alright again you know they've err they've fixed, they've sorted it out haven't they'* (Simon)… 'y*eah they're really good… as soon as you've got a problem you're sorted out aren't you within a….'* (Michelle)… 's*he's never had to go back again afterwards is really…it's been sorted out for the next 6 months a year hasn't it'* (Simon. Family interview, line 400).

Conversely, Robert expressed hopelessness about the future of the medications as a result of the family experiencing a number of failed attempts:

> *'erm but in the end you know I mean we're on we're not sure if there's another one after Adalimumab I'm I'm not sure it I'm I'm I'm sure he mentioned a another drug but we're getting into the experimental round now so I'm not sure how much further there is to to take this'* (Robert, individual interview, line 825).

The Hunter family described a losing battle and spending time renegotiating their own parameters about what is 'good enough'. The Hunter family had a shared belief of their losing battle with managing the symptoms of JIA and had become accepting of some elements of the JIA.

In addition, both Annie and Carly experienced a wider systemic battle with the medical professionals. Both perceived a sense of disempowerment as the professional were viewed as the 'experts', which meant that there was an expectation that Annie and Carly would be concordant and agree with the decisions made on their behalf. This lead Annie and Carly to assert themselves to be heard:

> *''cos I don't argue with the doctors like even if I feel really strongly about something I will I'll just I maybe say it but I wouldn't push it kind of thing like the only thing I've put my foot down the only time I've ever put my foot down was when I didn't take anything'* (Annie, individual interview, line 650).

Conversely, the parents described aligning themselves with the professionals placing importance upon following the advice for a positive outcome in relation to the JIA.

> *'just don't panic erm and rely you know trust in the doctors erm and just get on with things'* (Simon, family interview, line 3093).

Transitioning:

Both Annie and Carly described adverse experiences with transitioning into adult rheumatology services. Both participants explained feeling a loss of the relationships they had had with their medical teams and found it difficult renegotiating new relationships with the professionals in the adult service. Both Annie and Carly described the number of doctors they had seen who were not aware of their medical history which impacted upon their trust of professionals. Carly described feeling unheard, confused and uninformed by the professionals and Annie experienced a sense of isolation and a threat to her identity. An example quote from Carly demonstrates these concepts:

> *'erm so like yeah, he'd, he, Dr [name] would have seen like oh this has happened before where she's had a cold and it's shown up in her blood that her arthritis is hurting her where actually it hasn't been whereas she just jumped to the conclusion that I hadn't been telling my mum that it's been hurting just so I wouldn't move my medicine up…so yeah, I don't like this whole change thing'* (Carly, individual interview, line 865).

Robert and Michelle also described experiencing a sense of uneasiness during this transition period. Both described a struggle to 'let go' of their children and had to redefine and negotiate their role as parents. Robert talked of an internal struggle of still wanting to be involved in Annie's care and Michelle described feeling left out of decision making:

> *'I er mean I didn't really want to stop going with her I did think she was slightly too young but they decided you know she decided with the clinicians that that's what you know that's what they wanted so sort of guessed it were nothing to do with me any more really…she was starting to grow up and know her own mind*' (Michelle, family interview, line 3285).

However, both parents believed that their children needed to manage the condition independently from them. Again, the family had negotiated a shared understanding of the age at which it is appropriate to establish independence from parents, and how this should be done, but at an individual level, the family members described a struggle with these transitions.

JIA as a hidden condition:

Both families, although largely the Hunter family, described their experiences of JIA being a predominantly hidden condition and this often made it difficult to negotiate reactions from others who are unaware or ignorant to JIA. Both Annie and Carly expressed the difficulty explaining why they were using crutches and wheelchairs when there was no obvious sign of injury or that something was 'wrong':

> *'I don't know 'cos err it weren't like…there wasn't anything to show that I'd done something wrong…and I think like say if I'd had a pot on my leg obviously you can see that there's something wrong with me but just saying "oh I've got bad arthritis my knee hurts" it's like "oh your knee hurts like so what"…there's no visual like I don't know it's just like I was sitting in a I don't know I just didn't like it…I felt a bit embarrassed'* (Carly, family interview, line 495).

Annie described the difficulty negotiating disclosure as she explained having a preference for keeping the JIA hidden. She also described that when the JIA became visible to others, that this created difficulties in her wanting to be seen as 'normal' but the condition threatened this preferred identity:

> *'I just always have I've always wanted to be the person that was helping other people instead of being helped I don't know it's the same as I don't tell people about it when they meet me…just 'cos I don't want to be be the one that they feel like they have to help or err things like that it's like if I offer er…I offered somebody a piggy back and*

> *they wouldn't take it I was like well I wouldn't have offered if I hadn't'* (Annie, individual interview, line 135).

Michelle and Carly also described an embarrassment when the JIA became visible, demonstrating an awareness of what others may think. Such as, Michelle noticing that Carly would feel embarrassed after losing her hair or needing to use a pushchair to move around. The 'visibility' of the JIA had the potential to threaten the family's shared sense that they were a 'normal' family:

> *'she did go back in a pushchair for a little while about yeah between 5 and 6 which was a little bit, a little bit embarrassing for you I think wasn't it'* (Michelle, family interview, line 436).

Negotiating understandings:

The families displayed a number of shared family narratives, such as that of wanting to maintain a sense of normality; however, it seemed that these shared narratives did not always work for the individual. In some instances, divergent narratives could be identified whereby individual family members struggled to maintain the family viewpoint, such as not being able to positively reframe some of their experiences relating to Annie and Carly' future in relation to JIA (see quotes above). There were also times when a family member more overtly relinquished and challenged some elements of a family narrative, for example, in relation to Carly describing herself as an 'expert'. Carly explained that she wanted to assert her right to be considered an expert; however, this moved against her parent's beliefs that the health care professional is the expert and should be listened to:

> *'yeah we've always gone along with them haven't we…ever been a time when we we haven't gone along with them (.) and they've always been right err or seem to have been always right'* (Simon, family interview, line 3093).

> *'I was poorly and I told them when I got that blood test I was poorly and it always sets me off when I'm poorly and it was just that and I was absolutely fine and then they were like oh we're going to have to do your medicine and I was like "no 'cos I don't need my medicine" and they wouldn't believe me so I was like "well do another blood test and I bet it will come back normal" and it did so that that was pretty much it…you told me to take more and I told you'* (Carly)… *'that it's…she says it's my body and I know I know how I how I react but she says because they're…sort of expert…they just say what they know this is what right this is happening so you need to do this'* (Michelle. Family interview, line 2192).

Negotiating understandings also encompasses the importance that the families placed upon having shared beliefs about the JIA, in order to maintain a sense of the family togetherness which would limit family imbalance or disequilibrium. For example, Simon described that disputes between family members could lead to parents thinking each other knows what is best for the young person with JIA, which could result in poorer outcomes for the young person:

> *'I say just go with what they tell you and you know what I mean I think maybe I think that's where it might go of the rails if some people decide that they know better…and that's when that's probably where the parents will fall out with each other and they'll fall out with the doctors and it won't go right for the kid'* (Simon, individual interview, line 1072).

CHAPTER FOUR

DISCUSSION

The aim of this chapter is to relate findings from the two case studies to the existing research literature, some of which was outlined in Chapter One. This chapter will first provide a brief outline of the aims of this study and then continue to relate the synthesis of the case study findings to the wider literature. The chapter will then provide a critical evaluation of the study, including both strengths and weaknesses. Finally, recommendations for clinical practice and future research will be proposed.

Revisiting the study's aims

The study was designed to respond to the following research aims

1. To explore family understandings of JIA following a diagnosis of the condition in a young family member.
2. To explore how these understandings are negotiated within the family.

Discussion of the main research findings

A synthesis of themes from the two case studies yielded four themes, two of which will be discussed in the present chapter, and which are most salient the aims of the study. These will be *just getting on with it and maintaining a sense of normality* and *battling, fighting and the negotiation of power*. In addition, a further synthesis of family communication and negotiation strategies will also be outlined, which relates to the second aim of this study.

Just getting on with it and maintaining a sense of normality

Overall, both families described themselves as functioning well and significant distress or relational problems were neither described by the family nor observed by the researcher. The families described a shared family narrative in which it was important for them to move their focus away from JIA, in order for it not to dominate family life. The families described employing active coping strategies in order to maintain a sense of normality and family equilibrium. Strategies included: finding ways not to dwell on their situation, getting on with other aspects of family life that did not involve JIA, positively reframing potentially adverse events, assimilating JIA and adaptive strategies into family life so they become 'normal' everyday activities, and comparing themselves against people who were worse off than them. Stanton et al. (2001) argued that minimising the impact of the condition by preserving quality of life is an important process to positive adaptation and adjustment.

The strategies employed by the families may be seen as shared family coping strategies, which relates to how people adapt to adverse circumstances (Biesecker & Erby, 2008) and effectively maintain a sense of stability and equilibrium. For example, Lazarus and Folkman's (1984; also Folkman, 1984) transactional theory of stress and coping has been widely used to describe adaptation and functioning following the diagnosis of a chronic condition. It is proposed that adjustment, as an outcome, is dependent upon primary and secondary appraisals and coping. Coping and adjustment to chronic health conditions has been extensively researched, and it has often been found that the coping strategies utilised by individuals and families impact upon adjustment and family functioning (for example, Thompson, Gustafson, George, & Spock, 1994).

While adjustment and subsequent family functioning was not objectively measured in the present study, the families described how the JIA had become normal, as a family experience. The families described how JIA related care had been assimilated into the families' experiences and largely become more familiar and less stressful. This potentially relates to the transactional theory of stress and coping, in that over time, the families may have appraised the stressors associated with JIA differently and as less threatening than they did at the time of onset. This could be as a result of increasing familiarity with the condition, discovering what works for them and an increased sense of self-efficacy. In the present study, the families adopted action plans in anticipation of a flare-up, which helped create a sense of routine and 'normality' during times of disruption. In support of this, Rotter (1975) described that increased ambiguity about a stressor may lead to raised levels of uncertainty and may influence levels of perceived control. Therefore, reducing the ambiguity could result in higher levels of being in control and *getting on with it*. The families described experiencing high levels of ambiguity and uncertainty at the time of diagnosis, but reported a reduction in this uncertainty after a number of years of experience managing the JIA. Utilising the learnt routines could have meant that the family perceived a sense of proficiency and minimising the disruption enough to resume normal life quickly and efficiently.

The Hunter family described that striving for a sense of normality and getting on with other aspects of family life was important to prevent a sense of stagnation, as they would not be able to get on with other aspects of family life, unrelated to the JIA. It was also noted, from both families, that this sense of moving forward was made difficult by the unpredictable and uncertain nature of JIA; however, striving for normality promoted a perceived sense of achieved adjustment. Further processes described by the families to encourage normality involved continuing with life despite the condition and continuing with activities that were congruent with other families who were not experiencing a chronic health condition, such as going on school or scouts trips and partaking in further education. Both families reported that regaining a sense of normality was experienced as a process that required continual adjustment. They had also arrived at an

understanding that JIA did not need to dominate family life but rather they could have an acceptance of its presence. Normalising experiences has frequently been evidenced in the qualitative chronic health literature, which can be described as an integral part of the sense-making process (Barlow, Shaw, & Harrison, 1999; Guell, 2007; Robinson, 1993; Sanderson, Calnan, Morris, Richards, & Hewlett, 2011). In addition, studies have found that families report the importance of maintaining normality as a way of being able to identify themselves as 'normal' regardless of living with a chronic health condition (for example, Knafl, & Gilliss, 2002). This corroborates with the results of the present study, in that the families could have felt a 'threat' to their identity of being 'normal' when the JIA became 'visible' to others.

Moreover, the families attempted to shift their focus towards their abilities (as opposed to disabilities) and the parts of family life which remain undisrupted. This process has been found in other qualitative chronic health literature (Robinson, 1993) and the family resiliency theory would propose that maintaining a positive outlook is important to successful adaptation and adjustment. Studies demonstrated mixed results with regards to positive reframing and the impact upon adjustment. Some literature states that the coping strategies used by adults experiencing rheumatoid arthritis do not impact upon long-term adjustment (for example, Ramjeet, Smith, & Adams, 2008). However, other studies demonstrate that positively reframing may buffer against the stress associated with a chronic condition and thus contribute towards a greater perceived quality of life (Folkman & Moskowitz, 2000; Mahat, 1997). Mahat (1997) found that optimism in adults diagnosed with rheumatoid arthritis was the most effective strategy to cope with the stressors of the condition. A positive attitude towards JIA has also been found to buffer against anxiety and stress, which resulted in better adjustment for young people than those who did not have this attitude (LeBovidge et al., 2005) and parents (Horton & Wallander, 2001). In addition, studies have also demonstrated that 'hope' is important in helping caregivers manage their experience of caring for someone with a chronic health condition (Duggleby, Holtstander, Kylma, Duncan, Hammond, & Williams, 2010).

It is generally found that avoidant coping strategies result in poorer adjustment and approach-focused coping strategies (including optimism and reframing) facilitating better adjustment when faced with a chronic health condition (Compas et al., 2006; Lazarus & Folkman, 1984; Treharne, Lyons, Booth, & Kitas, 2007; Williamson, Walters, & Shaffer, 2002). Conversely, emotion-focused coping strategies such as wishful thinking have been associated with poorer outcomes, such as lower levels of functioning in adults with arthritis (Bombardeir, D'Amico, & Jordan, 1990; Felton, Revenson, & Hinrichsen, 1984). It may therefore be important that families retain a sense of positivity and optimism about their experiences for an optimal outcome; however,

it may also be vital that families demonstrate a degree of acceptance of the JIA in order to utilise approach-focused coping strategies in order to accommodate it into family life.

Positive reframing and *just getting on with it* could be seen as a family shared narrative of 'resilience' and 'bouncing back' from potentially adverse events. Drawing upon the family resiliency model, which is derived from the systems theory (Patterson, 2002a), maintaining a sense of normality could be seen as the family's ability to adapt and return to a sense of equilibrium, or, at least, create a new sense of normality that then felt 'normal' to them. The resiliency model (Patterson, 2002a) emphasises three levels of meaning: situation meanings, family identity and family world view. Situational meanings relate to the family's appraisal of the demands and their perceived abilities to cope with the demands. Family identity refers to how the family views themselves as a unit, including shared beliefs, and family world view is associated with how the family situates themselves within the wider systems (Patterson, 2002b).

Patterson (2002a) proposed that families experience the process of adjustment to a chronic health condition when they perceive themselves to be successfully managing the demands of the condition alongside their capabilities. This model proposes that the adaptation of families to life changes requires some negotiation at one or more of the levels outlined above. At the situational level, the Hunter and Aitkin family both described a sense of disruption during flare-ups, which increased the demands placed upon the family. The families described active attempts to reduce these demands, such as increasing medication intake and employing learnt coping strategies, to enable the families to return to a sense of normality, and thus balance. The families also described that when normality could not be negotiated, expectations were reduced and disequilibrium was temporarily permitted. Yet during these periods, the families had expectations and strategies in place regarding how this disequilibrium would be managed; therefore, conveying a sense of control and containment at a family level. In support of the above discussion, research investigating the psychological impact that arthritis has on families has demonstrated that families report utilising more coping strategies than those of normative controls (Harris, Newcomb, & Gewanter, 1991). This may potentially indicate that some families experiencing a chronic condition learn to be flexible and highly adaptable, and are, therefore, able to adjust more efficiently and rapidly to stressors that may disrupt their equilibrium (Harris et al., 1991). This may be especially the case with JIA as it is characterised by unpredictable flare-ups (Boekaerts & Röder, 1999), therefore requiring families to be alert and ready for disruption.

The families described their experiences of JIA related care as becoming part of everyday life, so it could be considered that they redefined or 'moved parameters' in order to identify a 'new' sense of normality. Researchers (for example, Sanderson et al., 2011) have labelled normalisation as a 'biographical repair' that can be defined as a re-establishment and renegotiation

of norms. Biographical repair is achieved through an acknowledgement of a change or difference and defining life as largely normal, thus minimising the consequences of the condition and engaging in behaviour that demonstrates normalcy to others (Knafl & Deatrick, 1986). The families in the present study demonstrated these four factors, and few variations amongst the family member's accounts were noted. This could indicate a shared understanding of acceptance and adaptation in order to achieve restoration.

Two further strategies the families identified in an attempt to minimise the dominance of JIA was positively reframing experiences and comparing their family to other families experiencing chronic health conditions. In relation to the latter aspect, both families attempted to strive for a sense of normality in the form of making comparisons against other families they perceived to be 'worse off' than them. Within the Aitkin family, comparisons were made as a way of attempting to remain positive about their situation, and in the Hunter family, comparisons were primarily made to maintain perspective. The families collectively negotiated a shared identity that fitted with their experiences of being a 'normal' family as opposed to a family with a 'disability' or 'problem'. This response to living with a chronic condition relates to the social comparison theory, first described by Fetsinger (1954). Festinger (1954) proposed that individuals are motivated to evaluate abilities and opinions in order to achieve a sense of normalcy. The process by which individuals appraise themselves is to make comparisons against people who are considered similar to themselves in some way, such as another family unit experiencing a chronic health condition. Two forms of social comparison have been described: 'upwards' and 'downwards'. 'Upwards' comparison relates to comparisons made with individuals or other groups who are considered as 'better off', which gives rise to a sense of hope, motivation and self-improvement. 'Downwards' comparison relates to a comparison made with individuals or groups who are considered 'worse off' and this can give rise to feelings of achievement and increased self-esteem (Salzer, 2002). The findings from the present study suggest that families described downwards comparisons by comparing both the severity of the JIA against more severe conditions and comparing their coping strategies against families who did 'dwell' over their circumstances. The families also described upward comparisons in their aspirations to be seen as being 'normal'. These comparisons, along with positively reframing experiences, may have created a shared culture of hope, optimism and self-esteem, which may have buffered the family against disequilibrium and a perceived sense of poor adaptability (Patterson, 2002a).

At an individual level, family members described that maintaining a positive outlook and viewing the family as 'normal' was, on occasion, difficult to sustain. This was a divergent narrative to the family shared view of being 'normal', with the exception of only intermittent periods of disruption during flare-ups. Both Robert and Simon described times when they would

become preoccupied with doubt about their futures and questioned if Annie and Carly would have permanent mobility difficulties throughout adulthood. Michelle described being more protective of Carly in comparison to Oliver, as a result of the JIA. In relation to the social comparison theory (Fetzinger, 1954) it may have been the case that at a family level, downwards comparisons create optimism and hope, and upwards comparisons facilitate motivation. However during times of uncertainty and stress, at an individual level, the upwards comparisons may facilitate worry and hopelessness, as the gap between reality and their 'ideal' family life becomes wider. This may further develop a heightened focus upon the negative impact that JIA had upon the family. It could also be the case that the family level narrative of being 'normal' and coping, and the support this preferred narrative had for family members, buffered against feelings of uncertainty and worry that was felt at an individual level. Linking with this, the family members may have also negotiated and accepted the family level approach because being 'normal' is a more culturally accepted and valued identity than being 'different' (Burry, 1988). Striving for normality could, therefore, have been an impression management strategy utilised by the family, in order to prevent being labelled or negatively appraised (Bury, 1988).

Impression management could have occurred between family members in order to present to one another that they were coping sufficiently. Waite-Jones and Madill (2008b) found that fathers concealed their worries from other family members which may mean that some family members mask how they are feeling to protect the family. This impression management strategy could also have been operationalized by the families during the interviews in order to present a desirable family identity. The implications of impression management and divergent views at the two levels may be that some family members struggle to discuss their worries with one another for fear of being negatively appraised, disrupting the 'shared' family identity, or the family's balance.

Battling, fighting and the negotiation of power

The Hunter and the Aitkin family described episodes of battling for control over both the JIA and one another's beliefs and opinions about JIA. Both families shared an understanding that they wanted control over the JIA and as a result worked together to battle against the condition in order to remain a sense of 'normality', as discussed above. In contrast, the process by which the families battled for a sense of control, in relation to the JIA, demonstrated divergent views between family members. Unlike *just getting on with it and maintaining a sense of normality*, which largely demonstrated a shared family narrative, the battling for control was more indicative of divergent understandings and opinions of JIA within the families. The most significant disagreement found within both families was largely identified between the young persons with JIA and their parents with regards to treatment regimes. Annie and Carly described that their anxiety around taking the

medication induced feelings of being out of control and battled against taking the medication to regain control. Both Annie and Carly described the importance of being in control over their bodies and Carly, especially, described that she was an expert about her own body and should be included in the decisions made in relation to the JIA. Essentially, Annie and Carly identified that their psychological needs were as important, if not more important, than their physical needs. Feeling powerless has been linked with lower levels of psychological well-being (Hagen & Smail, 1997). While on the other hand, the parents described feeling that the family had more control over the JIA when their daughters were on the medication, and the parents described feeling more out of control when they were not taking it. The parents described a strong belief that they should follow the advice of the doctors and utilised the professional's views in order to attempt to regain a sense of control. In essence, the battles therefore related to whose view would be most influential within each family in their battle for control that would result in reduced discord between family members. This also relates to Festinger's (1962) ideas around cognitive dissonance within groups. Festinger (1962) proposed that individuals are often influenced by those within the same social group (in this case the family) and that divergent views held within the group create tension. This tension drives members to reduce dissonance by changing their views to 'fit' with other members. Questions then arise as to whose views are most influential within the family and how the families should negotiate these. These questions move beyond the scope of the present study; however, both Annie and Carly presented with a sense of ambivalence about the medication, for example *'it's good for my body but not good for me'.* Both eventually resumed their medication regimes, indicating that perhaps their views converged with those of their parents and medical professionals, in this example.

Evidence from the chronic health literature indicates that parent-child discrepancies, or divergent viewpoints, may result in reduced levels of well-being for family members. For example, Olsen et al. (2008) found that young people with a chronic health condition demonstrated more difficulties with emotional adjustment if they had different illness beliefs to their parents. In addition Konkol et al. (1989) found that each family member experiencing JIA had different concerns depending upon the position they held within the family, which may indicate that how family members make sense of their experience will be different and therefore divergent beliefs could be expected. Moreover, parent-child discrepancies in JIA regarding reported pain and disability correlated with low mood in the young person (Palermo, Zerbracki, Cox, Newman, & Singer, 2004). This may indicate that understanding the nature of the differences in illness beliefs may be beneficial for professionals working with families in order to further understand family functioning and well-being and how discrepancy may develop.

There were differences in the family with regards to how they spoke about their battles. The Hunter family articulated an on-going sense of battling for control with the JIA, and described these battles located in the past, the present and they anticipated battles in their future. For example, the Hunter family described an on-going battle with Annie's body 'rejecting' her medication, and that they had tried many options but were unwilling to give up trying. In contrast, the Aitkin family primarily located their battles for control in the past. The difference in accounts could be related to comparisons in functional ability and how active the JIA is. Evidence suggests that children diagnosed with JIA at an earlier age have been found to have both better physical and psychosocial functioning, as reported by parents (April, Cavillo, & Feldman, 2012). April et al. (2012) suggested that this could be because the condition was appraised as less severe in younger children and younger children have fewer on-going difficulties with social and peer relationships than adolescents. Within the present sample, Carly was diagnosed significantly earlier than Annie, at five years old, in comparison to ten years old. In addition, considering the nature of the discord (i.e. treatment related), it may have been easier for the Aitkin family to continue with their shared narrative of getting on with 'normal' family life, therefore, largely avoiding the disagreements around treatment regimes. However, within the Hunter family, the increased rates of disease activity meant that their divergent views about treatment were raised more frequently.

Furthermore, differences within the families, in relation to discussions around battling for control, could also be related to Bandura's (1977) theory of 'outcome expectancy'. This suggests that a particular outcome will be expected following the implementation of a specific strategy. For example, it could be expected that adhering to treatment and medical advice and minimising triggers to flare-ups, would result in lower disease activity and result in better functional outcomes. However, for those experiencing JIA, this is not always the case and concordance does not always predict outcome (see Ravelli & Martini 2007 for an overview). This was the case with the Hunter family whereby they described feeling disappointed and '*downbeat'* that the outcome (poor physical mobility) did not reflect the effort that they had put in to managing the JIA. This may have resulted in a sense of powerlessness over the condition and the need to battle harder to regain that control.

A potential reason for divergent views could be as a result of dominant generational discourses regarding the medial professions and treatment, which may have impacted upon the families in several ways. The medical model within health-care settings is still very much dominant (Department of Health, (DoH), 2001). The medical model implies the patient as a passive recipient to care in which the power lies within the medical professions (DoH, 2001). In recent NHS developments, the promotion of patient-centred care has been emphasised, based on the growing acknowledgement that the patient can also be an expert who can be empowered to

contribute to the management of the condition (DoH, 2001). Young people, who have been socialised to this model, and have regular contact with the healthcare professions, may be more likely to provide their own thoughts with regards to their treatment than older generations, who could have less experience of the patient-centred model. This could therefore result in divergent views with decision-making regarding an individual's health behaviours. In addition, a change in focus towards patient-centred care may also change the way health-care professionals relate to their patients, enabling a context that facilitates a sense of control and mastery over the condition and an increasing trusting relationship with professionals (Hall, Dugan, Zheng, & Mishra, 2001). The implications for this may be that the patient-centred model empowers the patient but has the potential to disempower the parents. Clinically, this could indicate that it may be important for professionals working with families to empower the family as a unit as opposed to just the individual.

One final aspect of this theme, in relation to the divergent viewpoints, that is worth identifying is contextualising 'battling' and 'fighting' within normative family life transitions. From the viewpoint of the 'family life cycle' model (Carter & McGoldrick, 1989; Vetere & Dallos, 2004) the family constantly experiences normative stressors and so they are always undergoing transitions and negotiating changes to re-establish equilibrium. A normative stressor can be a major family transition such as a young person individuating from their family during adolescence, which inevitably will cause some family disruption (Knafl & Gillis, 2002). Adolescence is a time of identity formation and often characterised by becoming autonomous, developing initiative and developing identity (Erikson, 1968; Grotevant & Cooper, 1986). Autonomy and transition for a young person may be threatened or delayed when families also experience non-normative stressors, such as a chronic condition like JIA (Power, Dahlquist, Thompson, & Warren, 2003). The difficulty negotiating non-normative stressors during adolescence may lead to tension between parents and the young person as families, such as the Hunter and Aitkin family, are also promoting autonomy alongside the additional care that is needed to manage JIA (Power et al., 2003). Furthermore, the described 'battles' can also be viewed as Annie and Carly asserting their autonomy as many adolescents would at their age, and thus, the divergent views would be observable regardless of JIA. In support of the latter point, research has found that family functioning of families experiencing a chronic condition during emerging adulthood can be comparable to that of control groups (for example, Lewandowski et al., 2010), indicating that it may be important to take into account that divergent views may be anticipated and 'normal' during this point in a family's developmental stage and should not necessarily be viewed as problematic family functioning or reflecting difficulties with cohesion.

Family communication and negotiating shared understandings

The findings provided information regarding how the participating families constructed their accounts and negotiated experiences as a unit, which relates to the second aim of this study. Meaning is constructed when people organise their experiences into stories that relate to their experiences (White, 2007). Making sense and understanding experiences occurs through storytelling of thoughts, opinions, emotions and reactions. Within the present study, the participating families had the potential to demonstrate the processes by which they negotiated JIA, in the form of accounts from their experiences and also in relation to how stories were told within the interview. It has been suggested that the processes of how families tell stories can be relevant to practitioners working with families as this gives insight into family functioning and how they create meaning (Koeing-Kellas & Trees, 2006).

According to the family systems theory, family members making sense of their experiences will not occur in isolation and understandings will be influenced by other members within the family and also outside of the family. The sharing and negotiation of experiences therefore, can lead to shared understandings and promote positive family cohesion and functioning (Fiese & Sameroff, 1999). Olsen (1993) suggested that families who demonstrate high levels of cohesiveness make decisions together and demonstrate close emotional bonds, which will lead to efficient adaptability to stressors, such as those associated with chronic health conditions. Evidence from adolescents implies that family cohesion is an integral factor in well-being and maintaining low levels of condition-related stress (Salewski, 2003). Both families spoke about working together in relation to the JIA and demonstrated shared narratives in relation to this. Many of the families' stories in relation to managing the negative impact of JIA, focused around the individual family members adopting roles to contribute to limiting the impact the JIA had upon Annie and Carly, and one another. Potentially, having a shared understanding of an experience may be an indicator of family cohesion as the family would need to develop sufficient levels of communication and a willingness to be flexible to meet one another's needs (Skettett, 2003).

Further evidence of cohesion was displayed in the sharing of accounts by both families. For example, during the interview process, the family members often completed one another's sentences and frequently used positions of 'we' and 'us' as opposed to 'I' or 'me'. Both families also took one another's perspective in trying to determine another member's experience, which may demonstrate mutual support and empathy. While the Aitkin family also spoke in 'we' and 'us' terms, there were also several examples of when difficulties recalling events and inconsistent stories stilted their joint storytelling. Stilted storytelling was less evident in the Hunter family. Potentially, this could allude to lower levels of familial communication in the Aitkin family, regarding JIA, but not necessarily lower levels of family cohesiveness. Indications of cohesiveness

may be evidenced in the way the Aitkin family negotiated their experiences within the family interview and frequently co-constructed their understandings together. In bringing together their divergent experience, the family began to renegotiate and reformulate their understandings to develop new meaning. This may demonstrate the family exhibiting flexibility and adaptability by way of renegotiating new meanings. Maintaining a degree of cohesion also appeared to be a strategy for the Aitkin family to maintain equilibrium. Simon's account suggested that he relied on the advice from the professionals in order to prevent the family from having divergent views about JIA, and to avoid the need to negotiate their individual beliefs that may lead to a breakdown in relationships. This may indicate that professionals were also an integral part in how the Aitkin family negotiated their understandings of JIA, but it could also be that Simon had a belief that as long as the family's relationship with the professionals remained intact, then family cohesion and equilibrium was inevitable.

In addition, not only did high levels of family cohesion and communication facilitate adaptation and adjustment to JIA, the JIA was also found to facilitate family cohesion and mutual support. This has been reported in the literature (Britton, 2006; Britton & Moore, 2002a, 2002b; Segrin & Flora, 2005). In the present study, Emily, for example, described that during times when Annie experienced a flare-up, she would assume the role of a care-giver which Emily noticed brought her and Annie closer together. Emily described these as 'bonding times', which she appeared to treasure.

It was also evidenced that some distress occurred at times when the families highlighted divergent viewpoints in their views about JIA. For example, Annie and Robert described tensions within their relationship as a result of their differing views about the benefits of taking medication. Both members felt that their views were non-negotiable and demonstrated a level of inflexibility, which resulted in experiences of distress. Branstetter et al. (2008) also found that a breakdown in family communications disrupted family functioning and increased levels of stress and condition-related burden. This can also relate to Olsen's (1993) model of family functioning. In addition, Emily reported experiencing uncertainty and worry as a result of being excluded in conversations about JIA. This could lead to a perceived sense of feeling confused, unimportant or isolated from the other family members. It is perhaps evident that some stories and experiences were left non-negotiated or not shared within the families. These findings are consistent with research addressing sibling adjustment, whereby siblings often experience distress as a result of feeling isolated or a burden to parents (for example, Miller, 1996; Waite-Jones & Madill, 2008a). Emily described the importance of feeling involved in caring duties which may have helped integrate her back into a significant 'shared' family experience, essentially bringing the family closer together. Indeed, Emily also described that the interview experience helped her understanding of Annie's

experiences, perhaps indicating the importance of family communication and sharing understandings in the adaptation and well-being of the family. Emily's experience is comparable to other findings that demonstrate being more informed and aware of a chronic condition increases reported connectedness between family members and improved adjustment (Lobato & Kao, 2002). It has been suggested that convergent narratives preserve the families' sense of equilibrium and demonstrate higher levels of family satisfaction (Trees & Koeing-Kellas, 2009). Therefore, it is likely that family members share a motivation to negotiate their experiences with one another. A sense of connectedness may also reinforce a sense of feeling supported (Trees & Koeing-Kellas, 2009). It is evidenced in the literature that social support is important to well-being and positive health-related outcomes (Kraemer, Stanton, Meyerowitz, Rowland, & Ganz, 2011; Woods, Yates, & Primomo, 1989; Varni et al., 1988) and mediates the psychological well-being of young people experiencing JIA (Varni et al., 1988).

A shared understanding of JIA-related experiences, may also explain how families negotiate and cope with stressors, such as a chronic condition, as a unit. Shared coping cannot always be understood by individualistic models of coping such as that offered by Folkman and Lazarus, outlined earlier (Lyons, Mickleson, Sullivan, & Coyne, 1998). Shared coping strategies have been referred to as 'communal coping' (Segrin & Flora, 2005). 'Communal coping' can be viewed in relation to the extent to which the family perceives the stressor to be 'our' problem and 'our' responsibility as opposed to 'your' or 'my' problem or responsibility (Lyons et al., 1998). Lyons et al. (1998) identified three components of communal coping, which results in the family taking joint responsibility for managing the stressor and developing strategies to combat it. Communal coping has been evidenced in couples adjusting to chronic health conditions (for example, Skerrett, 2003, Yorgason et al., 2010), but not within the paediatric literature.

Communal coping may be seen as beneficial as it may preserve personal resources and facilitate social support. Social support has frequently been found to strongly buffer against stress and promote family functioning in families experiencing a chronic condition (Kraemer et al., 2011; Rosland, Heisler, & Piette, 2012; Woods et al., 1989). Findings from the present study may provide further support for this theory. For example, both families described coming together to implement their strategy plans and routines during times of increased stress, which was often when Annie and Carly experienced an increase in disease activity. As described earlier, it was also found that the Hunter family frequently used 'we' in reference to their experiences, which may indicate the family's sense of 'togetherness'. In contrast, divergent stories regarding coping, identified by the families, could have been due to individual and personal characteristics that influenced the coping process and therefore were not shared at a family level (Segrin & Flora, 2005) or those outlined earlier in the chapter relating to impression management. Moreover, research looking into

couples adapting to a health condition demonstrated that adjustment was better if the couple had similar coping strategies as opposed to dissimilar strategies (Kraemer et al., 2011). However, this correlation was found to be weak (Kraemer et al., 2011). These findings have implications for professionals working with families. It may be important to support families as a unit to explore their coping strategies and develop a shared way of managing a chronic health condition. Skerrett (2003) proposes that those working with couples should promote: a 'we' awareness (experiences occur within a relational context), for their partner's experiences and adaptation, and healing by empowering the 'we'. This framework may also be useful for families as well as couples in promoting shared adaptation. While promoting a shared understanding is important, as discussed earlier, divergent viewpoints within families is inevitable and normal, therefore, it may also be important to support individual coping strategies, but at the same time encouraging an increased awareness of other's methods of coping.

The interactional results may be best applied to the family systems theory and symbolic interactionism theory. The underlying principles of the systems theory are that system elements are connected, systems adapt themselves based on environmental feedback, systems are not reality and interactions are an important aspect to understanding the system as a whole (White & Klein, 2008). This latter point also links with the symbolic interactionism theory that posits that meaning is created through socialisation, interaction and language within small social groups such as that of a family (Blumer, 1969). According to these theories, communication patterns and ways of interacting are central to understanding the family, and are important in self-regulation and maintaining equilibrium (White & Klein, 2002). In order to respond effectively to change, communication regarding shifts in roles, managing the demands of the condition, adjusting parameters and negotiating these additional demands are required (Branstetter et al., 2008). This was evidenced throughout the results for both families.

Spontaneous interaction and negotiation of experiences and understandings was observed during the interview processes, as indicated earlier. For example, at several points during the family interview, the Aitkin family drew together individual accounts in order to attempt to create collective meaning. Within the Hunter family, Annie's discovery that Robert had arranged a wheelchair resulted in a strong reaction from Annie. Annie's repeated reference to this disclosure was interpreted as a change in her understanding that Robert did not always share information about his JIA experience with her.

The bringing together of individual stories to develop convergent ones has also been found in a qualitative study explaining family sense-making (Koeing-Kellas & Trees, 2006). Within their study, families negotiated their understandings at three different levels: all family members contributed to the account and an understanding of the experience was accepted by all members;

understandings that were shared among some family members and not others or meaning had been made at an individual level only; and, finally, incomplete sense-making whereby individuals and families had not fully made sense of their experiences. The first two were identified in the present study. The latter level may not have been observed in the present study due to the longevity of the families' experiences, and therefore families had a significant amount of time to make sense of their experiences. It has been argued that recognising family's spontaneous sense- making process provides important information for people working alongside families about how negotiating understandings occur (Koeing-Kellas & Trees, 2006). Furthermore, it is proposed that the process of family storytelling is a better indicator of family functioning than the content of the account (Trees & Koeing-Kellas, 2009). Potentially, professionals can monitor family shared meanings and also work with families at one of these three levels in order to promote optimal adaptation (Koeing-Kellas & Trees, 2006).

The bringing together of accounts and developing understandings within families, demonstrates that making sense of experiences is an on-going dynamic process that occurs continually through the interaction with others. This can be evidenced in the present study, as meaning is spontaneously developed throughout the interview process, as discussed earlier. It can therefore be assumed that adaptation and family functioning is also an on-going dynamic process. These on-going processes are worth considering in the context of a long-term, unpredictable condition such as JIA. High levels of flexibility may be required in order for adjustment to occur. This may be important for rheumatology services to offer regular contact with families in order to monitor these processes and determine what could be hindering successful adjustment.

A final point is that the family systems theory would argue that there is no particular style of family negotiation regarding coping or adaptation that is 'normal' or 'functional'. A family who is not meeting its own needs and managing demands may be seen as struggling to adapt (Libow, 1989). The coping and adjustment a family experiences is additionally not static and is therefore a process that is constantly adapting and evolving depending on the stages at which the family believes themselves to be in, how extensive the demands of the condition are, levels of communication and cohesion at the time in which adaptability is observed. This means that it is important to continue to respond to each family as a unit in its own right and be guided by their individual experiences. In addition, assessing the functioning of families who are experiencing a chronic condition by comparing against a control group, could be argued as fundamentally unhelpful because the families are incomparable and the difference should be acknowledged (Libow, 1989). The above points may help determine the inconsistencies in the family chronic health literature outlined in Chapter One.

Critical Evaluation of the study

The following section will outline a critical evaluation of the study, highlighting the study's limitations and the study's strengths.

Methodological considerations

Sampling and recruitment procedures

There are a number of sampling related issues that are important to consider in the context of the study's findings. The study aimed to recruit a small homogenous sample size in order to complete an in-depth exploration of families' idiosyncratic experiences of JIA. The advantage of this is that utilising a case-study approach has enabled the researcher to obtain a rich amount of data, sufficient for an in-depth exploration. The families recruited in the present study had similarities, such as both the young people with JIA were female of about the same age, each with a sibling and both families were recruited from the same paediatric service. This was considered sufficient homogeneity to synthesise and compare the results.

A disadvantage of recruiting a small sample size is that it is only possible for a limited range of family experiences to be studied. Due to the present study failing to recruit a male with JIA, or families that had experienced JIA for a shorter period of time, meant that the experiential themes identified, may not resonate with other families. Additionally, the participating families reported a general sense of working well alongside JIA and did not experience any significant adverse effects; the emergent themes therefore reflected this. Recruiting a larger sample of families would have yielded a wider array of family experiential data. Moreover, a larger sample would have enabled a more detailed and richer synthesis of emergent themes that would have facilitated a greater exploration of the similarities and differences between family experiences.

In addition, the two families who agreed to participate in the present study had also participated in previous JIA related research. This could suggest that these families were initially selected by the paediatric rheumatology service because of their likelihood of taking part in the research or selected due to the positive relationships between the selected families and the rheumatology service. Additionally, it could be that families who did not choose to take part in the study were experiencing a significant burden upon their resources, as a result of JIA-related demands, and so felt that they were not able to take part. Alternatively, families may not have chosen to take part in the study because they felt that JIA had not impacted or affected them sufficiently. This was identified by two of the three families that the researcher contacted about the study. One family chose not to take part for this reason. This could therefore have biased the results and therefore the emergent themes.

It is also possible that the families who chose to participate in the present study were displaying close or stable familial relationships, and as a result, were willing to talk about their experiences with others present. It is therefore likely that this study captured families who perceived themselves to communicate well with one another. As a consequence, families who perceived themselves to not be coping well with JIA or having strained familial relationships would have been less likely to respond. Finally, due to the complexities of the group interview format and the interactional focus of data analysis, it was deemed not suitable to interview families who were unable to speak English. This would limit the usefulness of the themes in relation to families from other ethnicities or cultures experiencing JIA. It could be deemed probable that families from different cultures or ethnicities deal with emotions differently (Hedges, 2005) and employ different ways of communicating as a result (Gudykunst & Lee, 2001).

A further methodological consideration is the low response rate during the recruitment phase of this study. Of 18 information packs that were sent out, seven reply slips were returned where three families indicated that they could be contacted by the researcher. Low response rates could have been due to several reasons. Firstly, some families claimed that they had not received the information packs through the post, indicating a low response rate was as a result of suitable families not being informed about the study. Secondly, due to recruiting from a considerably busy service, accurate records had not been sufficiently kept of the families who had been identified as meeting the inclusion criteria. This meant that follow-up telephone calls had not been made to families who had not returned their response slip. Thirdly, the study requested entire family units to consider participation in the study. This may indicate that within some families, members did not want to participate, preventing other consenting family members to contact the researcher. Fourthly, the study required several hours of total participant time to complete both interviews, which may have felt too demanding for families potentially already experiencing many competing demands as a result of JIA. Research suggests that families experiencing a chronic condition report significant strains on their resources (Britton, 2006). Finally, low response rates may have been due to an impersonal recruitment procedure. Families may have been more likely to take part in the study if they had the opportunity to discuss the research with a member of the paediatric rheumatology team or researcher during a routine clinic appointment or by telephone once they had received an information pack.

Access to experiences

Due to the longevity of the families' experiences, many of the participants relied on accurate recall to describe their experiences of JIA. It was clear during the interviews that some family members could not recall early events relating to JIA due to the longevity of their experiences and/or the

participants' young age at onset. Of the young people interviewed, only Annie could vaguely recall the immediate changes that occurred as a result of JIA and Emily could recall changes within the family approximately a year after Annie was diagnosed. Within the Aitkin family, problems with recall resulted in difficulties analysing the family's accounts and establishing shared understandings. There were many occasions where recall difficulties halted storytelling. Conversely, IPA researchers may argue that this may not need to be perceived as a limitation due to the fact that IPA is interested in experiences as the participant recalls it (Smith et al., 2009).

The interviewing style of the researcher may have also limited some access to experience. Due to the difficulty engaging some participants in the family interviews, I (as the researcher) found myself asking fact-based questions and jumping between topics in order to find experiences that could be shared by all family members and which could retain engagement. Additionally, as a result of some family members being able to better recall events or appearing to be more engaged in the interview process, I also found myself directing questions to these family members. The implications of this could be that some experiences may not have been fully explored, especially as one family member did not opt to be interviewed for a second time.

Is it well documented that using IPA to elicit experiential accounts relies on examining how meanings are storied, the language that is used to convey accounts and the use of metaphors and images (Smith et al., 2009). To do this, IPA relies on participants being able to describe their experiences as articulately as possible and with as much detail as possible. A challenge of working with adolescents, for the researcher, was gaining access to their experiences as a result of their occasional disengagement from the interviews or limited storytelling. This meant that potentially, some experiences were not communicated leaving more room for misinterpretation at the analysis stage. Conversely, these observations could also be due to the participants struggling to make sense of their experiences, therefore communicating this in a non-verbal manner. Alternatively, this could have been an indication of an insufficient relationship with the researcher. It may be that other forms of collecting data, such as diaries, could have increased the amount of data collected from these participants who struggled to verbalise their experiences.

A particular strength of this study was gaining access to family experiences via a multiple-perspective design. It is well documented that an individual does not adjust and function in isolation and will be impacted by wider systems (Patterson, 2002a). Using a multiple perspective design enabled rich in-depth accounts of families' experiences of JIA and gained access into family functioning that would not otherwise have been gained in a single perspective design. A further strength was utilising a mixed methods approach to data collection. This enabled the opportunity for the researcher to access information within different contexts, for example, accessing further information from participants away from other family members. Finally, a

proposed benefit of conducting second interviews is said to enable researchers who are novel to qualitative research to collect further data that may have been missed in the initial interview (Flowers, 2008). This means that access to experience can be facilitated by the development of the researchers' skills. Finally, the present study, also gained access to father's experiences. It has been well documented in the literature that studies relating to family adaptation and functioning that father's experiences are underrepresented and often difficult to access (MacFadyen, Swallow, Santacroce, & Lambert, 2011; Timko et al., 1992).

Quality control

Due to the rapidity at which the themes were re-clustered and changed and limited time resources, an accurate audit trail of every stage of the analytic process could not recorded. This is a limitation of the study and may have implications for the quality of the research. Conversely, the present research employed several methods, where possible to ensure this study was of a good quality. Methods included: researcher reflexivity, regular supervision, peer coding, ethical consideration and transparency.

Spontaneous meaning making and a cathartic process

A particular observation of the interviewing process was the positive impact some participants verbalised during the individual interviews. Throughout the conversations, both families demonstrated shifts in how they understood their experiences as a result of their collective conversations. In addition, two participants in the Hunter family reported that they had experienced some benefit in discussing JIA together. For example, the interview process enabled Emily to access Annie's thoughts and feelings about JIA that she had not experienced before. She stated in her individual interview that it had been useful to hear, which meant she was in a better position to support and understand Annie. It is commonly cited in research that siblings feel isolated within the family because aspects of chronic conditions have not been shared (Waite-Jones & Madill, 2008a). The interviewing process in itself may have encouraged catharsis and empowerment for Emily (Hutchinson, Wilson, & Wilson, 1994), resulting in a sense that the interview had brought her closer to her sister. This perhaps, may further support the usefulness of bringing families together to share their experiences and develop communal understandings.

Clinical implications

The following section will discuss the clinical implications of the findings of the study and research implications of the methodological approach utilised for data collection. In the context of a case-study approach, it is possible to provide some consideration to clinical application, but it is important to note that a case study approach can only point to themes that may be important to some families and are not necessarily indicative of the wider JIA cohort or to other families experiencing a chronic condition.

Firstly, the findings from both families suggest that utilising coping strategies such as striving for a sense of shared normality could be an important process in restoring equilibrium and consequently promote adaptation for families. It may therefore be useful for health-care practitioners to support families in both assimilating JIA-related care and tasks into family life so that they become routine, and also support the family in focussing on aspects of family life that are not necessarily dominated by JIA. In addition, and drawing upon systemic intervention ideas, it may be important to appropriately support and reinforce families' active coping strategies and resiliencies, including those of focussing upon optimism, hope, reframing and building upon past successful adaptation processes, in order to promote positive family functioning. The importance of continued monitoring of the family's coping, levels of cohesion and adaptation processes may also be useful, as the adaptation process is continuous. This therefore means that families are constantly changing and negotiating normative transitions in parallel to those of the chronic condition which could lead to additional pressures.

Secondly, the findings from this study indicate the importance of supporting the whole family in their adjustment to a chronic condition, including that of siblings. This study has built upon existing knowledge and theory in relation to adjustment to a chronic condition, and it can be argued that the more these adaptive processes are understood, the more opportunity there is to intervene (Biesecker & Erby, 2008). Interventions that include psychoeducation for siblings demonstrate an increase in their well-being and promote a sense of connectedness with other family members (Lobato & Kao, 2002).

A further clinical implication could be an increase in support for families at the time adolescents are transitioning into adult services. Parents discussed the difficulties adjusting to changes in their parenting roles during this stage and struggling with taking a step back from contributing to the medical consultations. The families also discussed the importance of their relationships with the health-care teams, which significantly changed in adult services, and communication thereafter suffered. Rheumatology services may benefit from utilising transition models (McDonagh, 2007; McDonagh & Kaufman, 2009; McDonagh, Southwood, & Shaw, 2007; Shaw, Southwood, & McDonagh, 2004) to prepare and support families into adult services. In

addition, access to professionals in supportive or counselling roles, such as clinical psychologists or family therapists, may be beneficial for families who are struggling with both normative transitions and transitions within the healthcare services, that may result in poorer health outcomes should support not be offered.

Moreover, both families described that their most difficult experiences were, in fact, related to the difficulties around medication as opposed to the JIA itself. These findings suggest that increased support and/or early support for the families in managing the anxieties around procedural distress may be helpful. For example, cognitive behavioural therapy (CBT) for procedural distress has been shown to be beneficial for young people in reducing anxiety (for example, Uman, Chambers, McGrath, & Kisely, 2010).

The study also highlights the importance of understanding JIA within the context of the family and the family's relationships within the context of the healthcare system. A greater understanding of the family processes of adaptation may be important for professionals in encouraging positive relationships between patient (and family) and the professionals. Positive family-professional relationships have been correlated with better patient outcomes both physically and psychologically (Hall et al., 2001), therefore it can be argued that increasing awareness of families' experiences can aid professionals to have a greater understanding of the families' needs.

A final clinical implication is that families appear to demonstrate that adaptation and adjustment to a chronic condition occurs at both a family level and an individual level. This highlights a need to consider these different levels when supporting families or individuals. This may especially be pertinent when working with adolescences who often attend appointments independent of parents. Identification of shared family coping strategies and individual coping strategies, for example, may help professionals to identify and promote alternative coping strategies and facilitate communal coping.

Research Implications

Implications for future research include addressing some of the limitations of the present study, such as the sample. It would be useful to explore how the themes identified in the study are experienced by a more diverse sample of families. This could include recruiting families with different structures such as more than one sibling or families whereby the young person with JIA is male or younger than the adolescents in the present sample. Additionally, recruiting families in which the young person has been recently diagnosed may yield different themes relating to communication or adaptation processes, which would be useful to compare against the themes that were identified in the present study.

It is evident from this study that the process of negotiating understandings and the adaptation process is an on-going development for families and little is known about how shared and divergent narratives are negotiated over time in families experiencing JIA. A longitudinal study investigating the communication processes that lead to shared and divergent stories within families would be particularly interesting. JIA is usually a long-term condition characterised by an unpredictable flare-ups, intermittent periods of limited functioning and an uncertain trajectory. Families, therefore, need to be flexible to deal with a number of uncertainties and adapt quickly to the changeable circumstances. This means that investigating these processes over a number of years would yield useful information for health care professionals who often work with families for the duration of their time in the service.

Conclusion

This study aimed to explore family experiences of JIA. This was the first study within this field to investigate family experiences as a unit that included an explicit analysis of how families relate to one another in the context of adapting to a chronic condition. The findings of this study were consistent with both the chronic health literature and family communication literature. The families placed a strong emphasis on trying to maintain a sense of normality in the face of uncertainly and disruption to 'normal' family life and also made efforts to accommodate JIA but to keep the impact to a minimum. In addition, the family utilised positive reframing strategies and comparisons against those who they perceived to be 'worse off' than them in order to try and maintain perspective and 'get on' with family life. Maintaining a sense of normality appeared to work well at a family level as a shared way of coping, however, at an individual level this appeared harder for family members to maintain. The families also described that different understandings, experiences and beliefs about the JIA created some divergent narratives between family members, which, at times, developed into discord. This was especially the case with regards to the prescribed medication and who could be considered an 'expert' about the JIA. Finally, during the interviews, families spontaneously and jointly constructed meanings relating to their experiences of JIA, which suggests that adaptation and making sense of experiences is constantly evolving and changing. Further research addressing how divergent and convergent meanings develop and change over time could be important in helping rheumatology services support families to promote optimal adjustment to a chronic condition.

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APPENDICES

APPENDIX 1: Literature search.

The literature search for related articles is outlined below:

1. The databases 'PubMed', 'psyc Info', 'Medline' and 'CINHAHL' were searched to yield any literature on qualitative research using interviews with more than one family member to elicit family experiences of JIA. The search terms included the key words identified below:

    - 'Juvenile'

and

    - 'Arthritis'

and

    - 'family'

Papers excluded:

- If not multi-perspective studies (i.e. interviews with only one family member)
- Quantitative methodology.
- Children had a diagnosis of fibromyalgia as opposed to Juvenile idiopathic arthritis, juvenile rheumatoid arthritis or juvenile arthritis.
- Commentary papers or reviews of quantitate research
- Not translated into English

psycInfo:

search terms	**number of articles**	**relevant to present research**
‘juvenile’	114	7 papers from 4 studies
and		
‘arthritis’		
and		
‘family’		

2. PubMed database was searched

Search terms	**Number of articles**	**Relevant to research**
As above	49	no additional papers to psycInfo

3. Medline database was searched

Search terms	**Number of articles**	**Relevant to research**
As above	89	no additional papers to psycInfo

4. CINHAL

Search terms	**Number of articles**	**Relevant to research**
As above	92	One additional paper found

5. Four studies were of interest after the inclusion and exclusion criteria was applied. Abstracts were read to assess if the papers were suitable ornoe.
   No further studies were found in Medline or PubMed that were not found in the Psych Info database.

Three of these studies quantified interviews for statistical analysis and were therefore not included as experiential literature.

6. References of the three remaining studies were examined and no further qualitative studies utilising more than one family member to give an account of their family experiences of JIA was found.

The three studies were those investigated by Waite-Jones and Madhill* (2008a, 2008b), Britton (2006), Britton and Moore* (2002a, 2002b) and finally Rosatto, Angelo, & Silva (2007).
*Authors produced more than one paper.

APPENDIX 2: Ethics approval letter.

NHS
Health Research Authority
NRES Committee North East - Newcastle & North Tyneside 1
TEDCO Business Centre
Room 002
Rolling Mill Road
Jarrow
NE32 3DT

Telephone: 0191 428 3564
Facsimile: 0191 428 3432

07 June 2012

Miss Rachel L V Notman
Leeds Institute of Health Sciences
Room G.04 Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds
LS2 9LJ

Dear Miss Notman

Study title: **Exploring family understandings of Juvenile Idiopathic Arthritis and the processes by which these understandings are negotiated.**
REC reference: **12/NE/0236**
Protocol number: **n/a**

The Proportionate Review Sub-committee of the NRES Committee North East - Newcastle & North Tyneside 1 reviewed the above application on 07 June 2012.

Ethical opinion

The Proportionate Review Sub-Committee reviewed the above study.

The Sub-Committee confirmed that this study has no material ethical issues.

The Sub-Committee felt that, whilst this study has the potential to cause distress during focus groups/interviews, a satisfactory strategy had been provided for addressing any participant distress that may occur.

Members of the Sub-Committee noted that, whilst the University of Leeds reviewers appear happy with the proposed sample size of three families, there was some vagueness about actual numbers. Clarification was therefore requested of the minimum overall sample size for both focus group and interview.

The researcher replied that the sample size is aimed to be three families. The minimum number of family members expected to participate is three members per family (the adolescent with a chronic illness, one parent and a sibling/second parent), and therefore this would mean a minimum number of 9 family members to take part in the focus groups. It is also aimed to interview a minimum of two family members in subsequent individual follow-up interviews. Therefore a further six participants will be interviewed in this second stage

It was noted that all versions of the Participant Information Sheet needed to contain a statement reflecting how any criminal or other disclosures would be dealt with.

The researcher confirmed that she had added a sentence to the Participant Information Sheets regarding limitations to confidentiality.

The Sub-Committee also noted that the correct REC title needed to be added to the Participant Information Sheets.

The researcher replied that she was unsure what is meant by the 'correct REC title' and had assumed this meant the full title of the study – in which case the documents have been amended

The researcher provided a copy of the following documents:

- *Participant Information Sheet for Young People, Version 2, dated 04/06/2012*
- *Participant Information Sheet, Version 2, dated 04/06/2012*

Members of the Sub-Committee noted that all versions of the Consent Form needed to include an option based on the NRES standard statement regarding access to research records (this does not need to refer to medical records)

The researcher confirmed that she had added the recommended statement re: access to research data to the Consent Forms.

The researcher provided a copy of the following documents:

- *Consent Form for Family, Version 2, dated 04/06/2012*
- *Consent Form for Adults, Version 2, dated 04/06/2012*
- *Consent Form for Parents, Version 2, dated 04/06/2012*
- *Consent Form for Young Person, Version 2, dated 04/06/2012*

The Sub-Committee informed the researcher that in the Participant Information Sheets there is a section titled 'Who has reviewed this study?' which should name the committee that reviewed this application (Newcastle and North Tyneside 1 REC)

Members of the Sub-Committee requested that the reference to regulatory authorities in the access to records statement be added to the Young Person Consent Form

The researcher confirmed that she had added this information to these documents.

The researcher provided a copy of the following documents:

Participant Information Sheet for Young People, Version 3, dated 05/06/2012
Participant Information Sheet, Version 3, 05/06/2012
Consent Form for Young Person, Version 3, 05/06/2012

On behalf of the Committee, the Proportionate Review Sub-Committee gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. Confirmation should also be provided to host organisations together with relevant documentation.

Approved documents

The documents reviewed and approved were:

Document	*Version*	*Date*
Evidence of insurance or indemnity	David Galey (Henderson Corporate)	28 September 2011
Interview Schedules/Topic Guides	Version 1	28 May 2012
Investigator CV	Rachel Notman	28 May 2012
Investigator CV	Dr Catherine A Brennan	
Letter of invitation to participant	Version 1	28 May 2012
Participant Consent Form: Family	Version 2	04 June 2012
Participant Consent Form: Adult	Version 2	04 June 2012
Participant Consent Form: Parental	Version 2	04 June 2012
Participant Consent Form: Young Person	Version 3	05 June 2012
Participant Information Sheet: Young People	Version 3	05 June 2012
Participant Information Sheet: Adult/Parent	Version 3	05 June 2012
Protocol	Version 1	28 May 2012
REC application	IRAS Version 3.4 102435/329214/1/462	28 May 2012
Referees or other scientific critique report	Transfer Recommendation Form - University of Leeds	12 March 2012

Membership of the Proportionate Review Sub-Committee

The members of the Sub-Committee who took part in the review are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

12/NE/0236	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

L. Kirkbride

(pp)

Mr Chris Turnock
Chair

Email: laura.kirkbride@sotw.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

"After ethical review – guidance for researchers"

Copy to: *Faculty Research and Ethics and Governance Administrator*

Mr Derek Norfolk, Leeds Teaching Hospital NHS Trust

NRES Committee North East - Newcastle & North Tyneside 1

Attendance at PRS Sub-Committee of the REC meeting on 07 June 2012

Name	*Profession*
Dr Pamela Davies	Principal Lecturer
Mr Gary Player	Advanced Practitioner - Cytology
Mr Chris Turnock (Chair)	Learning & Teaching Advisor

APPENDIX 3: Cover letter to families.

Leeds Institute of Health Sciences
Faculty of Medicine and Health

Charles Thackrah Building
University of Leeds
101 Clarendon Road
Leeds LS2 9LJ

www.leeds.ac.uk/lihs

UNIVERSITY OF LEEDS

Cover letter (version 1. 28.05/2012)

Family understandings of juvenile idiopathic arthritis.

Dear Parents/ Guardians,

I am Rachel Notman, a Psychologist in Clinical Training at the University off Leeds. I am currently undertaking a research project investigating families' understandings of Juvenile Arthritis. The Paediatric Service at St. James's University Hospital has sent this letter to you as they have identified you as a family that may be interested in taking part.

I am looking to invite families who have an adolescent diagnosed with Juvenile Arthritis to take part in this research. The research will involve a 60-90 minute family interview to explore your understanding and experience of arthritis. I would like to hear the viewpoints of all family members, as chronic illness impacts the whole family in one way or another. I would also like to interview individual family members to further understand individual experiences.

I am the researcher who will be facilitating the interviews and I would be very grateful if you would take the time to read the enclosed information sheet to get a better understanding of the research and what it will involve.

I hope that through this research project we will gain a better understanding of how families experience Juvenile Arthritis and this may help us develop services best able to support them. Whilst the Paediatric Rheumatology service is supporting this research, they will not have knowledge of which families have decided to take part or opted out of the research.

I would appreciate it if you could please complete the return slip to let me know if you would like more information about this project or would prefer not to be contacted again. If you would like more information a researcher will contact you so you have the opportunity to ask questions before

you decide if you want to take part. Please complete and return the form in the pre stamped and addressed envelope. The reply slip can be found at the bottom of the information letter.

Many thanks

Rachel Notman

Researcher
University of Leeds
0113 3430815

Dr Cathy Brennan

Lecturer in Public Health
University of Leeds

Dr Becky Hames

Senior Clinical Psychologist
St. James's University Hospital, Leeds

APPENDIX 4: Information sheets for parents and young people aged 16 and above.

eeds Institute of Health Sciences
aculty of Medicine and Health

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UNIVERSITY OF LEEDS

Study information Sheet (version 3. 05/06/2012)

Family understandings of juvenile idiopathic arthritis

Dear family,

Introduction

With the support of the Paediatric Rheumatology Service at St. James's University Hospital, Leeds, the Clinical Psychology department at the University of Leeds is carrying out a research project, exploring family experiences and understandings of Juvenile Arthritis. We understand that it is not just the individual with a diagnosis who is affected by chronic illness, but the whole family and so it is important to hear every voice to learn as much as we can. I would like to invite you to take part in this research with the hope of understanding more about how families adapt following a diagnosis of Juvenile Arthritis. Conducting research in this area is important in order to help paediatric services offer the best possible care to families. I hope that this research will contribute towards future service development.

I (Rachel Notman) am the researcher and I am carrying out this research as part of my professional training in Clinical Psychology.

What does the research involve?

Should you choose to take part in the research I will invite you and your family to take part in a family group interview lasting for approximately 60-90 minutes. I will ask you and your family several questions about your experiences of Juvenile Arthritis where you will be able to discuss your thoughts with one another, and myself. It may be the case that two or three individuals will also be invited to a second interview if there were any discussions from the family interview that would be useful to hear more about.

You can choose to have this interview at your home for your convenience, or the researcher can arrange a confidential room at the University of Leeds if this is your preference. Travel and parking expenses will be reimbursed should the latter option suit you better.

All interviews will be audio recorded as a memory aid for what was discussed. To ensure your responses are confidential, your names and any identifying information will be changed.

Why have I been sent this information?

We are writing to a selection of families in Yorkshire area where a member above the age of 12 years has a current diagnosis of Juvenile Arthritis. We are especially interested in talking to families who received a diagnosis at least 18 months ago.

In accordance with the Data Protection Act (1998) the researcher does not have contact to your personal or medical details. The Paediatric Rheumatology Service has identified that your family may wish to take part in the research, but they will not be directly involved in the research.

There is a reply slip at the end of this information sheet. If you would like further information about this research and for myself to contact you, you will need to fill in this slip and include your contact details. I will then contact you and answer any questions you may have.

What will happen to the information collected?

The researcher will look at all your interview discussions, along with the discussions from other families who are taking part. These will be accumulated to get an overall picture of the topics discussed. The results from all the interviews will be fed back to the Paediatric Rheumatology Team at St. James's University Hospital and published in a doctoral thesis. It is also possible that the results will be published in an academic journal.

All your personal details and identifying information will be anonymised and your names will be changed. Researchers often used quotes from interviews to illustrate what themes were discussed. These will also be anonymised to protect your identity.

Confidentiality

All paper and electronic records of your discussions will be kept safe and confidential in a locked cabinet at the University of Leeds.

The researcher for this study also has a professional duty to keep yourself and others safe. If during the interviews you disclose information that suggests you or someone else is in danger, then Rachel will need to report this. Rachel will not report any disclosures without discussing this with you first.

Is participation voluntary?

Participation is entirely voluntary. It is you and your families decision whether or not you take part. You may also choose to take part in the research and withdraw at any point without giving a reason. If you decide not to participate or choose to withdraw from the study, the care you receive at St. James's University Hospital will not be affected.

You can choose to have this interview at your home for your convenience, or the researcher can arrange a confidential room at the University of Leeds if this is your preference. Travel and parking expenses will be reimbursed should the latter option suit you better.

All interviews will be audio recorded as a memory aid for what was discussed. To ensure your responses are confidential, your names and any identifying information will be changed.

Why have I been sent this information?

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Is participation voluntary?

Participation is entirely voluntary. It is you and your families decision whether or not you take part. You may also choose to take part in the research and withdraw at any point without giving a reason. If you decide not to participate or choose to withdraw from the study, the care you receive at St. James's University Hospital will not be affected.

APPENDIX 5: Information sheet for children.

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www.leeds.ac.uk/lihs

UNIVERSITY OF LEEDS

Information Sheet for young people (version 3. 05/06/2012)

Family understandings of juvenile idiopathic arthritis

What is this about?

My name is Rachel Notman and would like to talk to you and your family about Juvenile Arthritis. I would like to hear your thoughts about arthritis and how it affects you and your family because there are still some things we don't know about it yet. If those people helping families, like yours, know more about Arthritis, it may mean we can do more things to help.

What do I need to do?

If you and your family think it would be useful to talk to me about arthritis, I will meet up with you all and we can all talk about thoughts of Arthritis. I would like to know what you think about arthritis and some of the good and not-so-good bits about it.

I will meet with you and your family together and I will ask you all some questions. If you want to say more about your thoughts about Arthritis I can also talk to you on your own or with one of your parents. What you and your family discuss with me will be kept private, when I write the report you may recognise some of the things you said, but we will change names and personal details so only you will know who said this.

You may feel a bit nervous to start off with, this is ok as you don't know what to expect. You can talk to your parents about it and they can talk you through what will happen. You can also talk to me as well if you want to ask me any questions about what to do.

What will happen to the information I give?

I will record what you and your family say and will need to keep this to be able to write about all the conversations I have had with you and other families. These recordings will be kept safe so no-one else will be able to listen to them.

There may be a time when I will need to tell someone what you have talked to me about. This will only happen if you explain to me that you or someone you know is in danger and I will need to keep you, or them safe.

Do I have to take part in this?

No you don't have to talk about Arthritis if you don't want to. It is for you to decide if you want to talk to me. Some people find it easier than others to talk about their experiences.

You may feel you want to try it to see what it will be like, and if you don't like it you can tell me you don't want to carry on. You can choose not to answer a particular question or ask to stop altogether. You don't have to say why you want to stop.

Is this study safe?

This study has been checked by the Newcastle and North Tyneside 1 Proportionate Review Research Ethics Committee who has agreed that this study is ethical for families to take part in.

Thank you very much for reading this information

Rachel Notman

APPENDIX 6: Reply slip.

.....

Reply Slip

Please return this reply slip in the envelope provided, even if you do **NOT** wish your family to take part in the research.

1. Print your name .....
2. Please Tick one box below
   - **We would like you to contact us with further information** about the study to help us decide whether to take part. ☐
   - **We do not want to take part** in the research ☐
3. If you ticked either of the first two boxes please leave your preferred contact details below:

Address .....

.....

Home telephone number .....

Mobile phone number .....

Email address .....

4. Would either parent please sign your name and date the form

Name ..... Date .....

Thank you for returning this slip.

APPENDIX 7: Adult consent form.[7]

Leeds Institute of Health Sciences
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Adult consent Form (version 2. 04/06/2012)

Family understandings of juvenile idiopathic arthritis

The researcher involved in this study is Rachel Notman. Rachel is working in collaboration with the University of Leeds and the Paediatric Rheumatology Service at St. James's University Hospital, Leeds. You should have received an information sheet that explained what this research involves and also had the opportunity to ask Rachel any questions you have had.

Please fill in the form below indicating your consent to take part in this study. You may withdraw this consent at any time without giving a reason.

	Please tick box to confirm
• I have read the information sheet for the above study	☐
• I have had the opportunity to ask questions about the study and discussed it with my family	☐
• I have received satisfactory answers to my questions	☐
• I have received enough information about the study	☐
• I understand I am free to withdraw from the study at Any time, without giving a reason and without it affecting the care we receive.	☐
• I understand that I may withdraw information that I have given at any time, even after I have finished and left the interview.	☐

[7] The family consent form, parental consent form and young person consent form follow a similar format to the example present in Appendix 6.

- I agree to my conversations with Rachel to be audio recorded and understand these recordings will be kept confidential ☐

- I understand that the data collected during the study may be looked at by individuals from The University of Leeds, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records. ☐

Please tick box that applies

- I agree to take part in the above study ☐
- I do NOT agree to take part in the study ☐

Signed _____ Date _____

Researcher:

Signed _____ Date _____

APPENDIX 8: Example of family interview topic guide.

1. **Example pre-diagnosis questions**
    - Can you tell me about family life before the onset of JIA?
    - Did you know anything about arthritis before the onset?

2. **Example diagnostic experiences**
    - What happened at the onset of symptoms of JIA?
    - Can you tell me about your experience at diagnosis?
    - What were your reactions?
    - How were your reactions the same/different?

3. **Example treatment questions**
    - What were/are the treatment regimens? If any
    - Any changes to daily life? How did it impact on the family?/how do they impact on the family?
    - Experiences of treatment regimes? Treatment changes?
    - Who was involved in the treatment regimens?
    - Can you give examples of how you helped X?

4. **Example adjustment questions**
    - Can you tell me about the adjustments that were needed? If any
    - Experiences of adjustment? Who noticed?
    - How did this affect you?
    - Has the way you respond changed over time?
    - Did you notice any changes in your relationship with one another?
    - Do you feel you are different or the same in the way you have adjusted?
    - Has your experienced changed the way you view yourselves?

5. **Example coping questions**

- Can you discuss how you have all managed or dealt with the changes you have experienced?
- What helped/did not help?
- Who helped/ did not help?
- What are you doing now to cope on a daily basis?
- Do you anticipate difficulties?
- Di you think you are the same/different in the way you cope?

6. **Communication**

- Has JIA impacted on the way you communicate as a family?
- Do you discuss JIA? How do you experience conversations?
- Can you please discuss a time where you feel you have worked well as a family?
    - Why do you think you had similar views about this?
- ……had different views as a family?
    - Why do you think you had different views about this?
- Do you think different views are a problem?
- How do you make decisions as a family?

7. **Closing Questions**

- Is there anything we have missed that you would like to add?

Prompts

- Can you tell me more about that
- Then what happened next
- How did you feel about that
- Can you give me an example
- Who agrees/disagrees with that
- Are there any differences in your views
- Who has similar views
- and how did you all react to that
- Was that the same/different for you

APPENDIX 9: Sample of interview questions with Annie Hunter.

Annie Hunter

Clarification of family interview comments

- You said at the end of the interview that you learnt some things from taking part in the interview. What were those things?
- Were you surprised at anything spoken about in the family interview?

Coping based questions

- You mentioned that the arthritis, especially with regards to the medication, is unpredictable, how do you manage that?
- Can you tell me more about?
    - Wheelchair experience- how did it make you feel?
- Not all families would have the same outlook as yours, in terms of trying not to let JIA take over, why do you think you have this outlook? Do you feel this is a good or bad thing? Or both?
    - Has this always been the case?
    - Have there been times when it has been more of a struggle to do this?
    - What do you think influenced this way of viewing things?

Support-based questions

- How does family help or not help?
- How do friends help not help?
- I asked you all about a time when you worked well together, you chose the times when you come back from hospital?
    - Why do you think you work well together following your steroid injections?
    - What contributes to you all working well together?
    - Would Emily and Robert agree with that?
- How does it make you feel to have your family helping you manage things, on the days when it is more of a struggle?
- Tell me about adult services
- How was the decision made to attend appointments on your own?

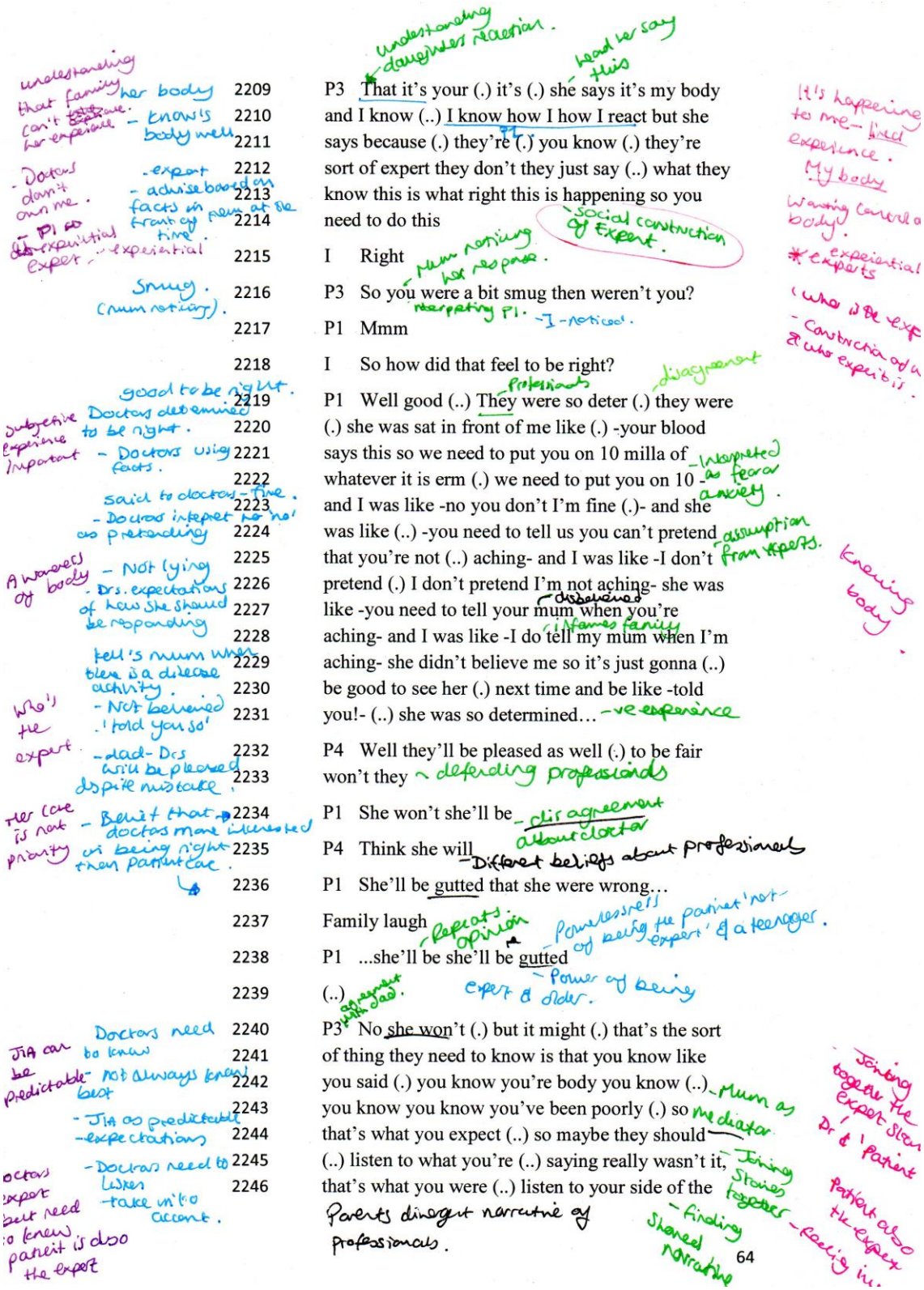
Reflecting upon experiences questions

- If you could go back in time, to when you first started experience symptoms, what advice might you give yourself?
- Do you think about your future much?
    - Do you think Emily or your Robert does?
        - If so, what do you think they are thinking about it?
- Have you and your family met any other young people with arthritis?
    - What was that like?
    - Did it change the way you thought about arthritis?

Closing questions

- Anything else you wish to add?

APPENDIX 10: Sample transcript and coding of Aitkin Family interview.



P3 That it's your (.) it's (.) she says it's my body and I know (..) I know how I how I react but she says because (.) they're (.) you know (.) they're sort of expert they don't they just say (..) what they know this is what right this is happening so you need to do this

I Right

P3 So you were a bit smug then weren't you?

P1 Mmm

I So how did that feel to be right?

P1 Well good (..) They were so deter (.) they were (.) she was sat in front of me like (.) -your blood says this so we need to put you on 10 milla of whatever it is erm (.) we need to put you on 10 - and I was like -no you don't I'm fine (.)- and she was like (..) -you need to tell us you can't pretend that you're not (..) aching- and I was like -I don't pretend (.) I don't pretend I'm not aching- she was like -you need to tell your mum when you're aching- and I was like -I do tell my mum when I'm aching- she didn't believe me so it's just gonna (..) be good to see her (.) next time and be like -told you!- (..) she was so determined…

P4 Well they'll be pleased as well (.) to be fair won't they

P1 She won't she'll be

P4 Think she will

P1 She'll be gutted that she were wrong…

Family laugh

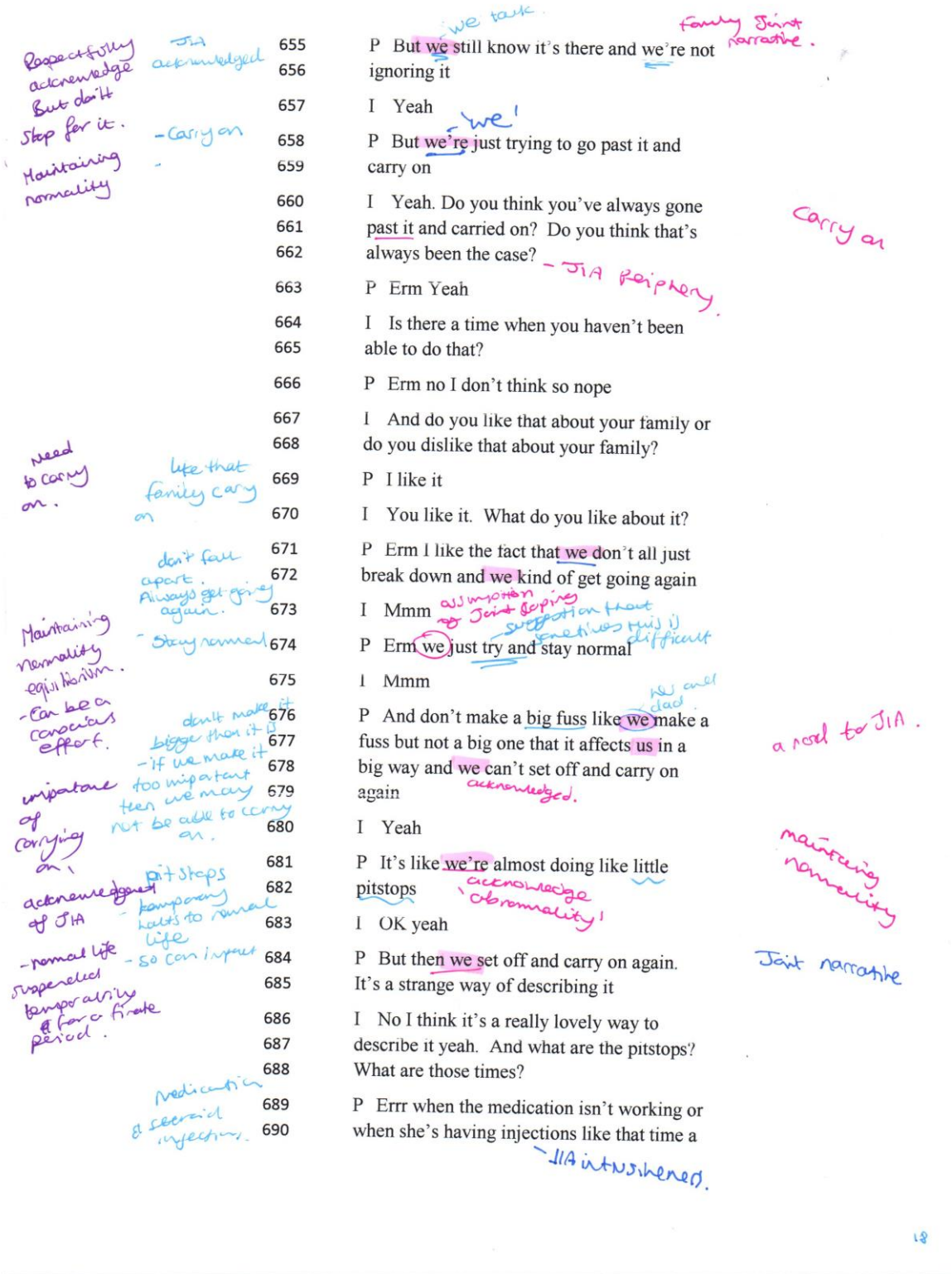
P1 ...she'll be she'll be gutted

(..)

P3 No she won't (.) but it might (.) that's the sort of thing they need to know is that you know like you said (.) you know you're body you know (..) you know you know you've been poorly (.) so that's what you expect (..) so maybe they should (..) listen to what you're (..) saying really wasn't it, that's what you were (..) listen to your side of the

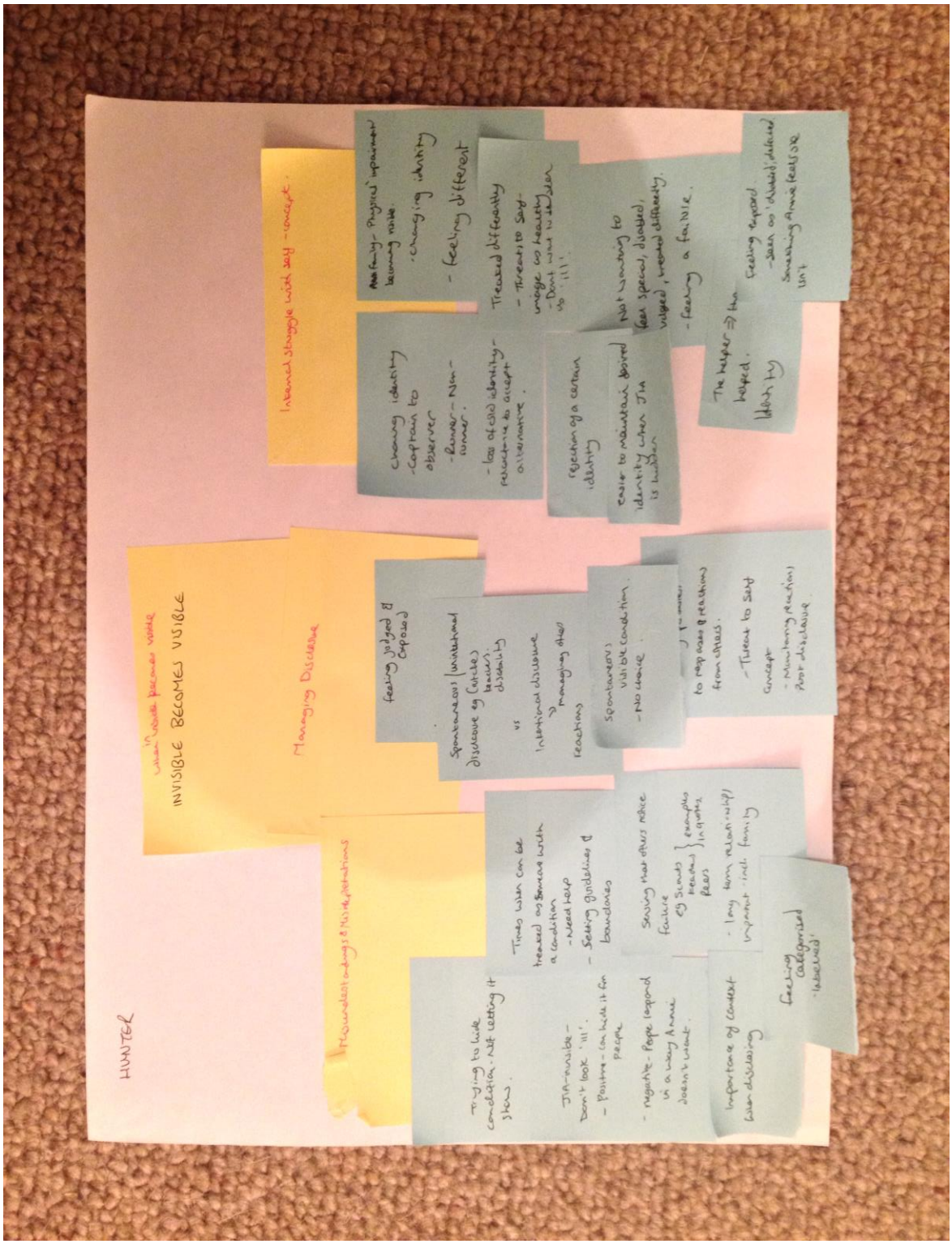
Parents divergent narrative of professionals.

APPENDIX 11: Example transcript and coding from individual interview (Emily Hunter).



655 P But we still know it's there and we're not
656 ignoring it
657 I Yeah
658 P But we're just trying to go past it and
659 carry on
660 I Yeah. Do you think you've always gone
661 past it and carried on? Do you think that's
662 always been the case?
663 P Erm Yeah
664 I Is there a time when you haven't been
665 able to do that?
666 P Erm no I don't think so nope
667 I And do you like that about your family or
668 do you dislike that about your family?
669 P I like it
670 I You like it. What do you like about it?
671 P Erm I like the fact that we don't all just
672 break down and we kind of get going again
673 I Mmm
674 P Erm we just try and stay normal
675 I Mmm
676 P And don't make a big fuss like we make a
677 fuss but not a big one that it affects us in a
678 big way and we can't set off and carry on
679 again
680 I Yeah
681 P It's like we're almost doing like little
682 pitstops
683 I OK yeah
684 P But then we set off and carry on again.
685 It's a strange way of describing it
686 I No I think it's a really lovely way to
687 describe it yeah. And what are the pitstops?
688 What are those times?
689 P Errr when the medication isn't working or
690 when she's having injections like that time a

APPENDIX 12: Example of analysis process for the Hunter family.



APPENDIX 13: Example of analysis process for Aitkin family.