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Beth Coleman BSc Hons

**EXPLORING THE EXPERIENCE OF PAIN IN ADULTS WITH
SICKLE CELL DISEASE**

Section A: Living with Chronic Illness
Word Count: 5,500 (plus an additional 1,819 words)

Section B: The Experience of Pain in Adults with Sickle Cell Disease
Word Count: 8,000 (plus an additional 66 words)

Section C: Critical Appraisal
Word Count: 1,979

Overall Word Count: 15,479 (plus an additional 1,883 words)

A thesis submitted in partial fulfilment of the requirements of
Canterbury Christ Church University for the degree of
Doctor of Clinical Psychology

SEPTEMBER 2013

SALOMONS
CANTERBURY CHRIST CHURCH UNIVERSITY

Acknowledgments

I would like to thank my participants, without them none of this would have been possible. Thank you to my supervisors who went beyond what I expected of them. To the wonderful library staff at Salomon's thank you for all your help. I cannot thank my amazing friends and family enough for all the help and on-going support right until the very end; I am a lucky girl to have you all in my life. And last but most important I thank God for in whom all things are possible.

Summary of the MRP portfolio

Section A is a literature review critically evaluating research related to the experience of living with chronic illness. Focusing on three illnesses cystic fibrosis, sickle cell disease and diabetes mellitus type one 18 qualitative papers were identified yielding 84 themes which were collated into a model of four categories devised from the material ‘the impact on everyday life’, ‘relationships’, ‘coping’, and ‘acceptance.’ The evidence suggested commonalities across the conditions but also some key differences. Gaps in the existing evidence were discussed, in particular the lack of research related to the experience of illness specific symptoms leading to suggestions for future research including studies exploring the experience of symptoms.

Section B presents an Interpretative Phenomenological Analysis of the experience of pain for seven adults with Sickle Cell Disease. The analysis revealed that participants had difficulty describing pain, favouring the use of analogy in an attempt to describe unimaginable pain. They faced a dilemma when considering medical treatment as treatment was necessary but difficult relationships with health care professionals meant this was not preferable. Participants went through a process of understanding and managing the disease in order to find a life with pain. Implications for clinical practice and future research are discussed.

Section C is a critical appraisal of the study considering research skills, what would be done differently if the study was repeated, and implications for clinical practice and future research.

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BSc Hons

Section A: Literature review

Living with Chronic Illness

Word Count: 5,500 (plus an additional 1,819 words)

JULY 2013

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Abstract

Many people in the UK live with chronic illness (CI). This paper presents information about the experiences of people with three CIs, Sickle Cell Disease (SCD), Cystic Fibrosis (CF) and diabetes mellitus type one (T1DM).

Eighteen qualitative research papers relating to the experiences of living with SCD, CF or T1DM were identified. The research findings were presented using a model of themes devised from the material encompassing four categories: 'the impact on everyday life', 'relationships', 'coping', and 'acceptance'.

Despite the different medical presentations this review indicates that there are a number of similarities across these CIs. CI impacted individuals in numerous varied ways. Individuals strived to cope with the impact of the illness, and had hopes to live a normal life. Some individuals appeared to have accepted the impact of the illness and that it did not have to be all encompassing. However, research on the experience of specific illness symptoms was minimal.

Future research should endeavour to more fully explore the experience of those living with CI and consider how such experiences can enhance knowledge of health and illness.

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Chronic illness

One in three people in the UK live with chronic illness (CI), such as heart disease, epilepsy, asthma and diabetes (Department of Health (DoH), 2012). CIs are diseases which medical interventions cannot cure only control therefore the life of the person is forever altered. Most CIs share several common features; complex causality; gradual onset; lengthy progression; functional impairment or disability; other health complications; risk of premature death (DoH, 2004). CI is the leading cause of mortality in the world, representing 63% of all deaths; 36 million people died from CI in 2008 (World Health Organisation, 2011).

Many CIs, such as cystic fibrosis (CF), sickle cell disease (SCD) and diabetes mellitus type one (T1DM), are genetic and diagnosed in childhood, although symptoms may not be visible early on. Other conditions, such as stroke and cancer, can develop at any point throughout life. The experiences of those living with CI vary in clinical presentation but all conditions affect multiple domains of life including education, social and employment (DoH, 2004).

Health beliefs and behaviours

An important task for health professionals has been to understand the factors that influence an individual's health behaviour, or their adherence to medical treatment. Laing (1971) first used the term 'medical model' to describe 'the set of procedures in which all doctors are trained.' This important development recognised the disease process; what was wrong with an organism, what was not functioning as it should, and considered how this could be put right. Thus traditionally biomedical models suggest a direct causal relationship between illness and symptoms (Walker, Jackson & Littlejohn, 2004). Consequently,

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biomedical models of illness concentrate more on the physical cause of illness, whereas it has since been recognised that physical, psychosocial and cultural factors are also determinants for health and illness (Marks, et al., 2011). As a result Engel (1977) proposed the biopsychosocial model which encompassed these factors and suggested that illness may have physical and psychological consequences (Ogden, 2007). This understanding of health and illness is the foundation of health care provisions today and has contributed to the decline of disease in the Western world (Marks, Murray, Evans & Estacio, 2011).

A further development has been the recognition of psychological causes of illness (Ogden, 2007). For example, heart failure can cause anxiety but mood is also related to either the onset or progression of the illness. Equally, not all disease-specific symptoms need to be present for someone to be ill; indeed it is not uncommon for someone to feel ill without any physical signs of disease thus it is important to understand the impact patients' psychological processes have in the appraisal of symptoms, health and illness.

The Health Belief Model (HBM) for example, (see Becker, 1974 for a comprehensive overview), highlighted the importance of patients' beliefs about health in understanding health and illness. The model aimed to predict patient health related behaviours, incorporating both preventative behaviours and behaviours in response to treatment for both acute and chronic illness. It proposed that behaviours result from a set of core beliefs which can be used to predict the likelihood that a behaviour would occur (Ogden, 2007). Although there is much empirical support of this model (Diefenbach & Leventhal, 1996), several studies reported conflicting findings (Ogden, 2007). Criticisms included; the focus on conscious cognitive processing and whether individuals think in probabilistic terms; the individual focus and therefore the absence of cultural and social factors; no consideration of

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the role of emotions such as fear and denial; the possible interaction between core beliefs has not been adequately explored.

Social cognitive models improved on some of the critics of the HBM, by attempting to place an individual within the social context of other people and society (Ogden, 2007). The theory of reasoned action (TRA; Fishbein, 1967; Ajzen & Fishbein, 1970; Fishbein & Ajzen, 1975) emphasised the key role of social cognitions as subjective norms (beliefs about the social world) and incorporated both beliefs and evaluations of these beliefs. The theory of planned behaviour (TPB; Ajzen, 1985; Ajzen & Madden, 1986; Ajzen, 1988), a development of TRA, proposed that behavioural intentions, defined as 'plans of action in pursuit of behavioural goals' (Ajzen & Madden, 1986), are the outcome of a combination of several beliefs. The TPB has been widely used to successfully assess a range of health-related behaviours and has more effectively included social and environmental elements (Ogden, 2007). However, it has been criticised as there is no order to the beliefs or any suggestion about the direction of causality nor does it not stipulate what values or beliefs are of particular relevance for the evaluation of health behaviours. Further, the model does not acknowledge that health behaviours can occur for non-health reasons and the environmental cues to attitudes and actions. Thus we are unable to access the catalogue of signs/symptoms that people use to recognise a threat or the usefulness of an action to prevent it (Diefenbach & Leventhal, 1996).

Some researchers have suggested that the patients' perceptions are central to their understanding and ability to manage illness. This is the central concept of the Common Sense Model (Leventhal, Meyer, & Nerenz, 1980). A self-regulation model it predicts that illness representations are directly related to coping behaviour and, via this, to a client's

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health outcome. This implies that the impact of a problem will be mediated by a person's ideas about such things as discomfort, disability and dependence, and that these will in turn influence their emotional responses, coping behaviours and appraisals (Hagger & Orbell, 2003). Threats to health are understood by the patient according to their own cognitive and emotional representations of that threat. These representations are influenced by various cultural and social experiences, such as the media, previous experience of illness and amount of health education received. Both intellectual and emotional representations impact how a patient accepts their illness. This idea can be seen in patients living with chronic kidney disease (Jansen, Rijken, Heijman, Kaptein & Groenewegen, 2012) who found that generally perceived autonomy and self-esteem were linked to beliefs about more personal control, less impact of the illness and its treatment and less concern.

Other researchers have highlighted the importance of psychological adjustment to illness as key to understanding health behaviour. Taylor (1983) suggested a cognitive adaptation model in response to threatening events such as serious illness. Taylor proposed that psychological adjustment was grounded in three key themes; finding meaning, a sense of mastery or control over the illness and rebuilding self-esteem. Taylor suggested that psychological adjustment played a major role in illness resistance and disease course. This may be not surprising when considering that individuals with CI face on-going symptoms of illness, possible disability, uncertainties about the future and the progression of their illness, and concerns about their ability to continue in their accustomed lifestyle. These consequences of CI can result in irreversible changes to everyday life. As a result individuals may find themselves in a position of needing to redefine meaning in order to limit stress and enhance coping. Meaning making is influenced by the individual's own experience, cognitive response to and appraise of an event and their social context. In CI this making sense can

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change and develop over time as the condition changes eventually reaching acceptance of the condition (Walker et al., 2004).

Similarly, Karnilowicz (2011) presented the notion of psychological ownership in relation to CI. He proposed that identity and psychological ownership are inseparably linked so when presented with a life threatening illness the individual faces a sudden and often unexpected change to life and the future that may result in changes to the individuals concept of self, ideology which can lead to new insight. The illness process itself often requires the individual to gain control over the impact of the illness by assuming some level of ownership. This occurs within a culture and social context and can ultimately result in an individual's personal knowledge and experience enhancing their sense of ownership of the illness. Karnilowicz experienced this himself when diagnosed with prostate cancer, describing a process of developing his own psychological ownership and thus a "more balanced interpersonal relationship" (p.279) with his oncologist. This acknowledged the doctors expertise but also established a less authoritative and more collaborative approach to discussing his illness and treatment.

The models presented represent the prominent models used to explain how individuals adapt to living with CI. Walker, et al. (2004) suggested that the key components of health and illness are organised into three major paradigms; the biomedical model which emphasizes disease; psychological models of adaption to illness and biopsychological frameworks which both focus on health, functioning and well-being. The authors presented a comprehensive critique of these three approaches in relation to rheumatoid arthritis, suggesting that each approach had merits in its attempt to understand how individuals with rheumatoid arthritis adapt to the illness but that no single approach fully explained the experience. They concluded that a biopsychosocial model of adjustment to CI that developed

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the stress and coping paradigm to improve psychofunctioning and psychological adjustment might provide a more comprehensive approach to investigating and treating chronic conditions.

Individual experience of CI

Models of health and illness recognise the impact of numerous factor on the experience of illness, such as individual experiences, personal health beliefs, appraisals and cognitions as well as the influences of others and society (Diefenbach & Leventhal, 1996). Psychological factors such as emotions, psychological adaption and ownership are also considered. In order to acknowledge such variable factors it is important to consider the patient's individual lived experience. Qualitative research primarily focuses on human experience (Van Manen, 1990) but the concept of "lived experience," from the German *erlebnis*,¹ expresses more than just experiencing the events of life but the accumulation of experiences and the understandings, reflections and self-awareness we make of those experiences (Van Manen, 1990). It is therefore arguable that a patient's lived experience of CI and the accompanying processes contribute to the individual experience of and response to CI.

Rationale

Given the prominence of CI in society and the importance of understanding health and illness behaviours it the important to gain a wide, holistic perspective on illness, it is therefore important to consider what research is available looking at the experience of those living with CI.

¹ The etymology of the English term 'experience' derives from the Latin *experientia*, meaning "trial, proof, experiment, experience." It does not include the meaning of lived whereas the German word for experience, *erlebnis*, already contains the word *Leben*, "life" or "to live." The verb *erleben* literally means "living through something." (Lewis-Beck, Bryman, & Liao, 2004).

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Research which focuses on the individual's experience is of particular importance to understand how individuals experience their own CI and what factors may influence their health behaviours, coping and treatment adherence. The majority of research which looks at the lived experience of CI, for example, quality of life (QOL) research is questionnaire based. Questionnaire based studies often impose responses on participants rather than give individuals the opportunity to respond using their own words (Barker, Pistrang, & Elliott, 2002). Qualitative designs allow a search for meaning through in-depth and detailed descriptive interpretations of individual discourses, which is not possible from quantitative analysis (Smith, Harré, & Van Langenhove, 1995). This is important when considering an individual's lived experience of CI. Radley (1999) argued that illness can only be studied qualitatively because illness always appears in relation to how individuals represent their world as a result of the occurrence of disease and this is embedded in experience and the social context.

This review seeks to critically evaluate the qualitative literature concerning living with CI. The focus on qualitative work enables the review to access the richness of individuals experience through their own words (or the interpretations of their words). The exclusion of quantitative research means this focus on individuals own accounts will be maintained. It would however be interesting to compare measurable factors of illness, such as lung capacity in CF, or established measures of mood with patients reported experience however as this review is primarily focused on the lived experience of CI it is the reported narratives of adults with CI that is of interest.

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The review will focus on three genetically inherited CIs SCD, CF and T1DM. SCD is a haematological disease where red blood cells become sickle shaped and block blood vessels resulting in episodes of severe pain known as crisis. CF is a pulmonary condition where the lungs and digestive system can become clogged with sticky mucus most commonly resulting in a persistent cough and reoccurring chest and lung infections. T1DM is a blood disorder where blood sugar levels become too high due to the pancreas not producing insulin. (Kumar & Clark, 2009). Despite these differences in presentation these conditions share many key features such as early onset, risk of other health complications and early death.

Method

Search procedures

An electronic literature search was conducted for relevant papers using PsycINFO, Medline, Web of Science, ASSIA and the Cochrane Database of Systematic Reviews. A range of keywords were used including: sickle cell, cystic fibrosis, type 1 diabetes, chronic illness, lived experience and psychosocial experience. Such search terms were used in multiple combinations. A manual search was conducted on significant journals and reference lists.

Inclusion/exclusion criteria

The search yielded 49 results, which were then screened manually for suitability. Articles were included if they utilised a methodology which identified themes related to an aspect of the experience of adults (over 18 years old) with either SCD, CF or T1DM. Articles were excluded if they included multiple diagnoses when it was not possible to separate the findings for the illness of interest, or, in the case of diabetes, if the population was not clearly

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defined as T1DM. Further, articles that focused on caregiver perspectives, unpublished dissertations, or articles not available in English were excluded. Based on this criteria 18 articles were identified.

Review of studies

The studies selected included five CF, eight SCD and five T1DM papers; ten utilised individual interviews, four focus groups, three a combination of interviews and focus groups and one used interviews and diaries. A variety of qualitative techniques were represented including interpretative phenomenological analysis (IPA; one), grounded theory (six), thematic analysis (three), narrative analysis (one) and framework analysis (one). The remaining six studies did not specify a specific model or used an approach from an earlier paper (see Appendix 2 for information on study designs).

In total the articles presented 84 themes relating to different aspects of an individual's experience of living with CI. The review below will present this information using a model devised from the material. The published themes were compiled into four categories and 11 subthemes based on patterns within the topics using thematic analysis. The model was audited by a psychologist with experience in thematic analysis as a form of credibility check (Elliott, Fischer & Rennie, 1999). The categories generated were 'the impact on everyday life', 'relationships', 'coping', and 'acceptance'. It should be noted, that within these domains there was much overlap and interrelation, and some of the themes gleaned from the research address more than one domain (see Appendix 3). Due to the volume of data each section will present a summary of the literature linked to the theme in relation to the experience of individuals living with SCD, CF and/or T1DM.

Impact on Everyday Life

The physical manifestations of SCD, CF and T1DM differs greatly. CF patients often suffer from a persistent cough and chest infections due to sputum, a thick sticky mucus that obstructs the lungs and digestive system, requiring frequent physiotherapy. SCD patients suffer from acute painful episodes called crisis, everyday chronic pain and numerous complications due to blood clots throughout the body. T1DM patients are required to take insulin daily in order to regulate blood glucose levels (BGLs) in order to avoid hypoglycaemic or hyperglycaemic episodes (Kumar & Clark, 2009). 15 papers considered how these unique presentations impact daily life (CF four, SCD eight, T1DM three).

Unpredictability and everyday life. Twelve studies (CF two, SCD eight, T1DM two) considered the unpredictable nature of CI a major life limiting factor across many domains. Rasmussen, O'Connell, Dunning, and Cox, (2007) described the changeable facet of T1DM in young women as 'being in the grip of BGLs.' The uncontrollable aspect of BGLs fluctuation with no or minimal warning caused the women difficulties in relation to starting work, becoming pregnant and entering motherhood. Likewise, university students in the UK were worried that diabetes would prevent them from experiencing 'normal' student life (Balfe, 2009). Similarly, 15 young adults with CF feared the unpredictable nature of CF, the resulting impact on life and the future (Higham, Ahmed, & Ahmed, 2013). Meanwhile adults with SCD reported finding the unpredictable nature of SCD pain and crisis the most difficult aspect to deal with, due to "unplanned interruptions" to life (Booker, Blethyn, Wright, & Greenfield, 2006, p.45). Other patients felt the unremitting unpredictable nature of SCD crisis was "taking over" to the extent that they were not able to enjoy life (Thomas & Taylor, 2002, p.353).

SCD studies show that multiple hospitalisations and time consuming treatment regimens resulted in absences from work or education from an early age (Weisberg, Balf-Soran, Becker, Brown, & Sledge, 2013). Older SCD patients reported being concerned about finding and maintaining employment (Booker, et al., 2006; Anderson & Asnani, 2013).

Loss. Three papers looking at SCD considered the theme of loss; research in CF and T1DM alluded to this notion but only the SCD papers directly considered loss. Booker, et al. (2006) noted that male participants experienced a loss of virility. More broadly, Caird, Camic and Thomas (2011) commented that both psychological and social functioning were undermined in SCD patients by a loss of control alongside emotional and physical suffering. Anderson and Asnani's (2013) thematic analysis encapsulated this by suggesting that individuals were faced with numerous important losses causing a negative impact on life; loss of social contacts, their own autonomy or ability to fully assume roles, loss of employment/education and independence and ability to perform typical roles. The ultimate loss was death, both the loss of their own life and the death of others with SCD. The authors remarked that in response to these losses some interviewees suffered depression/suicidal ideation.

Emotional impact. Twelve papers reported a range of emotions in response to CI (CF four, SCD six, T1DM two). Negative emotions were not uncommon. Participants across the research reported feeling depressed (Strickland, Jackson, Gilead, McGuire, & Quarles, 2001) suicidal (Weisberg et al., 2013), angry (Booker, et al., 2009), hostile (Strickland et al., 1999), embarrassed (Addis, Spector, Shaw, Musumadi, & Dhanda, 2007), shamed (Lowton, 2004), and disgusted (Tierney, Riley, Jones, Webb, & Horne, 2010).

Fear, worry and anxiety were frequently expressed (e.g. Ingadottir & Halldorsdottir, 2008). Death anxiety or fear of complications and the future were particularly prominent across all populations. Such feelings had been experienced by participants from an early age and continued throughout life. Young CF patients reported a fear of death and dying (Higham, et al., 2013) and young women with T1DM feared complications such as hypoglycaemia (Rasmussen, et al., 2007), as did older participants (Ingadottir & Halldorsdottir, 2008). Booker et al. (2006) found SCD patients of various ages (22-53) were worried about death and the future, fearing there was little they could do to prevent the course of the disease. There was also a tendency for patients to 'fear the truth' or be in denial about the reality of the illness (Ingadottir & Halldorsdottir, 2008, Caird et al., 2011).

Despite experiencing these difficult emotions, many individuals also exhibited an appreciation of life and attempted to view the illness in a positive light. Participants took pleasure in the details of life, were pleased to be alive and felt fortunate for various aspects of their life, such as children (Chapman, 2002; Rasmussen et al., 2007; Caird, et al., 2011). Several studies also recognised strength and resilience despite the suffering (Caird, et al., 2011; Weisberg et al., 2013) and a hope for the future (Higham, et al., 2013).

Experience of symptoms. Surprisingly few papers (five) included information about the experience of illness symptoms. Two CF papers considered the impact of coughing and sputum production, two SCD studies looked at the experience of pain and one the experience of priapism. There were no articles that investigated the impact of hypoglycaemic or hyperglycaemic episodes for individuals with T1DM, although one considered the impact of fluctuating BGLs (Rasmussen et al., 2013).

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In CF, coughing was seen as intrusive and an outward sign of illness that would inform others that the individual was unwell (Lowton, 2004; Tierney, et al., 2010). Sputum production in particular was considered dirty and participants did not want this symptom to define them (Tierney, et al., 2010.) Some participants spoke about the inconvenience of the cough, the impact on daily life, the pain involved in coughing and how important it was to “take advantage of when you’re well” (Tierney et al., 2010, p197). The paper gave the reader an idea of what it is like to live with a persistent cough rather than just the disadvantages such symptom produces, however none of the papers considered the impact of not being able to breathe properly.

For SCD patients, pain was presented in the context of the handicap it produced, the impact on relationships, the emotional impact and the need for medication/hospital treatment (Booker et al., 2006; Weisberg, et al., 2013). Booker et al. (2006) noted that although their study focused on pain, participants directed the discussion towards the wider impact of pain; although this was clearly important to participants it does not leave the reader with a sense of what it is like to experience such disabling pain.

One SCD study focused on a common but not often discussed problem in SCD, priapism, the prolonged and extremely painful penile erection that does not diminish on orgasm (Melamn & Serals, 2000). Addis, et al. (2007) interviewed six men (age not recorded) who had all experienced priapism, mainly beginning in early adolescence. This paper reported in detail what it was like to experience priapism and the impact this had on life for example one participant said “You just don’t know how to position yourself, how to sit down or stand up or whatever” (p.149).

Relationships

Fourteen papers suggested that living with CI had an impact on different types of relationships (CF four, SCD seven, T1DM two).

Family and friends. Nine studies considered relationships with friends and family (CF two, SCD five, T1DM two). Some studies found that individuals avoided or delayed disclosing their diagnosis because of concern for how this would affect their relationships. Fear of being misunderstood or stigmatised for being medication dependent was a concern for individuals with SCD (Booker, et al., 2006). Some adults with CF expressed a fear of “being seen as dirty” (Tierney, et al., 2010, p.195) due to the need to dispose of sputum so would try and disguise their illness. Young adults with T1DM starting university reported feeling worried that their new peers would view them as disabled, damaged or different (Balfe, 2009).

Adults with CF reported having mixed experiences of disclosing their diagnosis, some had found it positive, helpful and supportive whereas others felt that their friends were ‘showing them off’ or treating them as patients (Lowton, 2004). A lack of understanding made it difficult for SCD participants to maintain friendships (Caird, et al., 2011) particularly in relation to the unpredictable nature of SCD making it impossible to commit to social events. Fear of social isolation (Booker, et al, 2006) or feeling marginalised (Strickland, et al., 1999) was common. The unpredictability of SCD sometimes made patients want to hide away during crisis, but this isolation could be experienced negatively by significant others affecting intimacy and trust (Thomas & Taylor, 2002).

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Many adults with SCD were acutely aware of the disease's impact on their relationships, especially that significant others found their constant suffering difficult to cope with (Thomas & Taylor, 2002). Despite this, SCD patients described requiring a lot of support and therefore relied heavily on a select number of close and trusted family and friends (Booker, et al., 2006). Having this supportive network was crucial to patients in managing their condition (Caird et al., 2011). Likewise young women with T1DM described requiring a lot of help but felt this was a burden on their social support network and wanted more support for their partners (Rasmussen et al., 2013). SCD participants expressed concern that the heavy burden of support required would impact the lives of others resulting in some patients hiding their symptoms from loved ones (Booker, et al., 2006).

Intimate relationships. Six studies mentioned intimate or sexual relationships (CF three, SCD two, T1DM one). Disclosure of SCD to potential partners was problematic for patients. They expressed concerns that partners would not form a long-term relationship with a CI patient. For CF patients the implications on fertility affected their decision to disclose their diagnosis (Lowton, 2004); men are usually sterile and women can have reduced fertility (Sawyer, 1996). Consequently, the seriousness of the relationship, level of intimacy and the perceived reaction were important factors in deciding to disclose (Lowton, 2004).

Young adults with CF understood that they might need In Vitro Fertilization (IVF) in order to have children, however they expressed a desire to conceive naturally in an attempt to be normal (Higham, et al., 2013). Young women with diabetes experienced increased anxiety around having children due to the change of hormones associated with pregnancy and lactation. The women who had children had found it a struggle to balance diabetes management, their own needs and the needs of their children (Rasmussen et al., 2007).

The majority of the SCD men who suffered from priapism (Addis, et al., 2007) had told their sexual partners about the condition but had found disclosure difficult. Half reported a detrimental effect on sexual relations and all reported feeling anxious about sex and relationships. In addition to priapism three participants experienced erectile dysfunction as a result of SCD damaging the peripheral nerves involved in penile erection.

Professional relationships. Professional relationships were considered by four papers (CF two, SCD one, T1DM one). There appeared an increased reluctance to disclose some diagnoses in a work situation. Lowton (2004) explained that CF participants considered themselves less “suitable” (p.178) for employment compared to those without a CI; consequently patients thought disclosure would hinder their chances of employment. Young adults with CF expressed the importance of having a career as part of living a normal life (Higham, et al., 2013) but some participants reported their illness having been ‘uncovered’ resulting in dismissal from employment (Lowton, 2004). Likewise young women with T1DM found variable BGLs affected their ability to work and also had difficulty deciding whether to disclose their illness (Rasmussen et al., 2013).

Relationships with medical professionals. Six papers discussed relationships with medical staff, the majority were SCD studies (five) but a single paper for CF mentioned that participants did not like to be seen as sputum producers by practitioners (Tierney, et al., 2010).

Booker et al. (2006) found that some SCD patients described interactions with medical staff as a ‘battle’ where they had to work hard to convince staff that they genuinely needed help. Some actively avoided medical professionals whilst in crisis, as a result of

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repeated experiences of not feeling believed. Patients reported hospital staff lacked empathy for the excruciating pain experienced (Thomas & Taylor, 2002) and that this resulted in distrust regarding pain management medication, with medical staff believing they were merely drug seeking (Maxwell, Streetly, & Bevan, 1999; Strickland et al., 2001; Weisberg et al., 2013). Patients believed that due to this stigmatisation they were treated differently (Maxwell et al., 1999). Many SCD participants felt some clinicians were racist (Weisberg, et al. 2013), did not have enough knowledge to make appropriate treatment decisions (Booker et al., 2006), that basic physical and personal needs were not fully attended to (Maxwell et al., 1999) and that receiving hospital treatment increased overall stress levels (Thomas & Taylor, 2002).

Despite these difficulties SCD patients required regular medical treatment and, thankfully, not all the experiences reported were negative. Some patients reported appreciation to staff for their efforts in promptly dealing with pain, especially in accident and emergency, and that it was comforting to know there were people that could care for them (Thomas & Taylor, 2002). Others commented on the affection and care from staff members and described close relationships with staff when in children's hospitals (Weisberg, et al., 2013). Those who required more hospital treatment advocated the benefits of building positive relationships with staff (Maxwell et al., 1999).

Coping

Fourteen articles presented a number of ways in which individuals with CI cope (CF two, SCD seven, T1DM five).

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Regaining control. Six papers across the SCD (two) and T1DM (four) literature presented regaining control as a key concept of coping. University students with T1DM expressed anxiety that diabetes would control their lives and thus form their identity (Balfe, 2009). Ingadottir and Halldorsdottir (2008) described the constant attempt to master diabetes in order to live a normal life balancing wellbeing with medical regimen as “disciplining the dog” (p.610). Paterson, Thorne, Crawford, and Tarko (1999) described a gradual process in which T1DM patients discovered that diabetes did not have to control them but rather they could control their diabetes. This is important as being in control of diabetes was strongly associated with increased QoL (Rasmussen et al., 2007).

SCD patients also expressed frustration at their lack of control. Coping involved regaining control of the physical aspects of the illness and managing their own and others responses to SCD. One strategy was to engage support from friends and family who could check that the individual was looking after themselves (Anderson & Asnani, 2013). Others had a strong sense of self-responsibility for managing their own illness (Maxwell et al., 1999).

Self-care. Eleven papers considered the role of self-care (CF two, SCD five, T1DM four). In order to effectively utilise self-care strategies it was important for individuals to understand how their body worked including the unique impact of their condition. In CF understanding that the lungs and digestive system can become clogged with sputum was an important part of how individuals established what activities they could and could not manage (Tierney, et al., 2010). Huyard (2008) called the process of learning to notice and interpret the manifestations of CF “critical internalization” (p.540). Similarly, T1DM participants grew to understand the interaction between diabetes, insulin and the body in

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order to maintain healthy BGLs (Ingadottir & Halldorsdottir, 2008). In SCD, understanding the difference between everyday pain and crisis pain, as well as analgesic medications was crucial to maintaining a sense of normality (Weisberg, et al., 2013).

The importance of being self-aware and knowing their bodies in order to notice any changes was emphasised by T1DM patients; consequently patients would be hyper-vigilant for any fluctuations (Hernandez, Bradish, Rodger, & Rybansky, 1999). Some reported patterns within their bodies signalling hypoglycaemia (Paterson, et al., 1999). Similarly, SCD patients noticed symptoms such as headache or fatigue as indicative of an impending crisis (Strickland et al., 2001). Many SCD patients reported engaging in self-care techniques in an attempt to control the physical manifestations of the illness, believing they would prevent a crisis by, for example, taking a warm bath (Strickland et al., 2001; Anderson & Asnani, 2013). Others, however, were less convinced such actions were consistently helpful, some believed that nothing could be done to ease the pain (Addis et al., 2007).

Religious beliefs and meaning making. The importance of faith in coping, particularly during crisis, was reported in four SCD studies. Patients reported prayer and going to church as beneficial and comforting (Strickland, et al., 2001). Moreover, a divine reason for being ill presented an explanation for the suffering which helped make sense of the illness. This process appeared to reduce the emotional impact of SCD, such as depression and stress. It also helped individuals turn their attention to other matters (Anderson & Asnani, 2013), and sustain hope (Caird, et al., 2011).

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Meaning making also existed outside a religious context, in particular increasing awareness about the diseases and showing others that CI could be something constructive, appeared to create a focus and bring meaning to the experience for some (Caird, et al., 2011).

Identity. The strength and depth of an individual's personal identity was considered an important aspect in the ability to cope in four papers (SCD one, T1DM three). For some putting their experiences into perspective based on other aspects or roles in life, such as being a mother or a professional was key (Rasmussen et al., 2007; Caird, et al., 2011). Others attempted to develop different interacting identities which balanced together in order to live unaffected by illness (Balfe, 2009). Others still found a more fundamental transformation took place that allowed them to differentiate the self from the body, in doing so a positive identity emerged "perceiving the self as the subject and no longer the object of the disease" (Paterson et al., 1999, p.795).

Acceptance

Despite the numerous and varied ways of coping with CI the research suggested that the most common way to cope was through acceptance. Four papers presented the idea of acceptance (CF one, SCD two, T1DM one).

SCD patients accepted that there was no better option than to cope with the challenges faced. As there was no cure, they accepted that they 'just have to live with it' (Anderson & Asnani, 2013, p.658). Some believed that mental strength was the key to maintaining physical health thus acceptance was perceived as an important aspect that developed over time. Others spoke about a changing relationship to SCD, moving from denial to active disease management, onto acceptance. Many had been shocked out of denial as a result of

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external life events such as becoming a parent. Acceptance appeared to develop with age and recognition of the importance of coming to terms with the diagnosis in order to live with the impact of the disease (Caird, et al., 2011).

CF patients also reported a journey from not considering themselves ill to accepting the illness. Huyard (2008) termed this an “illness career” (p.538) with diagnosis as the first stage, followed by becoming aware of the relationship between the disease and the symptoms. The critical internalization process mentioned above involves putting what has been learnt to the test, including what happens if treatment schedules are not followed. Through this process clients develop a detailed knowledge of how their body functions with CF and an acceptance of limitations accompanied by a pragmatic approach to life with the condition.

Diabetes patients described living with the illness as a transformation process of on-going preserving and renegotiating; “light dawning that things could be different” (Paterson, Thorne, Crawford, & Tarko, 1999, p.791). This process started when the individual discovered that they knew what was best for them and that they had the power to make positive decisions in their lives. There were many barriers in finding acceptance; however several individuals choose to face these as challenges rather than problems as this enabled growth. Again knowing bodily signs, symptoms and individual limitations was an important aspect of this. Finally, finding an identity separate to the illness was central to participants’ ability to live life and focus on life goals.

Discussion

Summary

The research presented provides information on the lived experiences of people with SCD, CF and T1DM. Despite the different medical presentations this review indicates that there are a number of similarities across these CIs.

Living with CI impacted on many domains of life independent of diagnosis, including schooling, employment, relationships, mood as well as causing physical limitations. There was a sense of loss evident throughout the research, and an on-going struggle to overcome the limitations of the diseases - to find a way to reconcile the impact of the illness, the necessity of a medical treatment without jeopardizing independence and a 'normal life.'

This struggle appeared to be a process or journey that developed over time. Learning about the illness, how it affected the individual and how best to look after themselves through various self-care strategies were important aspects of successful functioning. Finding a positive identity or identifying a meaning for the CI, whether religious or otherwise, was reported as another important aspect of coping and a positive step towards acceptance.

Growing to accept the illness and its impact on life appeared to empower individuals to take control of their lives and live less impinged by the illness. True acceptance is perhaps the realisation that life will not be 'normal' due to the effects of the illness but that life can still be enjoyable and productive.

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There were also differences between the conditions. The idea of managing the illness and transitioning across different stages of life such as starting university and becoming a mother appeared popular in diabetes research. CF research appeared to focus more on achieving a normal life through hiding symptoms, not disclosing or acceptance. In SCD the relationship with medical staff was prominent; this is perhaps because these relationships are typically problematic. Also the role of faith was reported by a number of SCD papers but not mentioned in any of the others. Although lessons can be learnt by comparing CIs these key differences highlight the importance of considering the unique factors of each disease.

Critique of the studies

Most of the research related to the psychosocial impact of living with CI rather than the lived experience of specific disease related symptoms, for example living with breathing difficulties in CF, disabling pain in SCD or hypoglycaemia in T1DM. Although there was some information available about the experience of symptoms in CF and SCD it was limited and did not provide a detailed picture about what it is like to experience such symptoms.

Although important information can be gleaned about the impact of CI the conclusions drawn from the research presented must be tentative due to methodological limitations. Yardley's (2008) quality assurance criteria and Elliott et al. (1999) guidelines for evaluating qualitative studies (see Appendix 4) were used to inform the critique including methodological details, the ability to situation the sample in order to demonstrate how breadth of the finding and considering the presence of creditability checks in relation to theme development.

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The studies reviewed utilised a number of qualitative methodologies. Approaches such as IPA, Grounded Theory and Narrative Analysis have been developed, tested and demonstrated in a variety of studies as useful methods to gain insight into health psychology topics (Murray & Chamberlain, 1999). The process of coding and developing themes is dependent on subjective interpretations; consequently these established approaches encompass methodological procedures that promote rigor and credibility (Silverman, 2001). Some of the papers do not clearly state the type of analysis, instead using terms such as ‘qualitative’ (Maxwell, et al., 1999) ‘content themes’ (Strickland, et al., 1999) or ‘thematically analysed’ (Balfe, 2009). Others cited less well known methods such as the Vancouver School (developed by Halldorsdottir, 2000). Not clearly defining the methodology means it is difficult to establish the epistemological background of the research and thus reduces the transferability and credibility of the studies (Saldaña, 2013).

Qualitative researchers do not normally set out to generalise findings (Ryan, Coughlan, & Cronin, 2007) however situating the sample helps the reader judge how widely the finding might apply (Barker, et al., 2002). The amount of situational information varied greatly across studies; some providing a lot, for example Caird, et al. (2011) who included numerous demographic details as well as providing a context for the research. Meanwhile others reported much less, such as Addis, et al. (2007) who did not include participant ages.

It is important to ensure credibility and validity across all types of research (Willig, 2001). There are a number of options available to qualitative researchers; however the number of studies that reported such processes varied, and worryingly three studies did not report any validation activities (Elliott et al., 1999; see Appendix 4 for details).

Future Research

In recent years there has been an increase of qualitative research in the area of CI in adulthood, however further research is needed to explore the lived experiences of those with CI. Future research could:

1. Explore the lived experience of specific disease related symptoms for example breathing difficulties in CF, living with disabling pain in SCD or hypoglycaemia in T1DM.
2. Investigate whether the research on lived experience can be accommodated by current models of health and illness.
3. Further investigate the psychosocial impact of CI, including complications, multiple conditions and factors affecting acceptance, meaning making and identity.

Conclusion

Living with CI affects individuals in numerous varied ways. Despite differences in clinical presentation SCD, CF and T1DM patients all experience restrictions on life, problems with schooling or employment and complications with relationships. Individuals strive to cope with the impact of the illness, and have hopes to live a normal life. Some individuals appear to have reached an acceptance about life with CI; accepting the impact of the illness and that it does not have to be all encompassing.

Although there is a growing body of literature considering the lived experiences of individuals with CI there are many areas unexplored. It is important to continue to investigate the experiences of those living with CI.

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BSc Hons

Section B: Empirical Paper Literature Review

The Experience of Pain in Adults with Sickle Cell Disease²

Word Count: 8,000 (plus an additional 66 words)

All names and other identifying features have been anonymised to protect participant's identity

JULY 2013

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² This manuscript is intended for submission for publication to the British Journal of Health Psychology. Wherever possible, authors' guidelines (Appendix 1) have been followed.

Abstract

Sickle Cell Disease (SCD) is England's most common blood disorder whereby sickled shaped red blood cells block small blood vessels causing both acute and chronic pain. Currently there is poor understanding about the experience of SCD pain. Furthermore, there is a recognised difficult relationship between SCD patients and health care professionals (HCP) which has been found to prevent patients seeking medical assistance.

Seven adults with SCD participated in semi-structured interviews regarding their experience of pain and receiving medical treatment. Interviews were transcribed and analysed using Interpretative Phenomenological Analysis (Smith, Flowers & Larkin, 2009). The analysis revealed three overarching themes: experiencing unimaginable pain, the dilemma of treatment and finding a life with pain.

Findings suggest describing SCD pain is extremely difficult, participants favoured using analogies to attempt to communicate the constant, agonising, limitless, inescapable pain. Participants described that normal rules do not apply in that pain medication does not always relieve pain. These factors have caused misunderstanding with staff thinking SCD patients are drug seeking. Participants wanted HCPs to listen to their own expertise when considering treatment options.

Study limitations, implications for clinical practice and current models of health and illness are considered, as well as possible for future research.

Key words: Sickle Cell Disease, experience, pain, qualitative

PAIN IN SICKLE CELL DISEASE

Sickle cell disease (SCD) comprises a group of genetic blood disorders with an estimated prevalence of 12,500-15,000 in England (NICE, 2012) making it the country's most common blood disorder, affecting predominately people of black African and Caribbean origins. In SCD red blood cells distort into a sickle shape causing vascular occlusion, leading to a series of both acute and chronic complications (Kumar & Clark, 2009). SCD is a chronic illness, an incurable, controllable disease resulting in the life of the person being forever altered. Severity of SCD varies, and treatment is usually symptom based. The pain experienced in SCD is unlike most other conditions, with patients experiencing both chronic and acute pain. Although recurrent and unpredictable acute pain episodes (known as crisis) is the most common reason for hospitalisation, the pain experience in SCD is poorly understood.

Research attempting to understand the experience of pain has concentrated on chronic pain, the predominate being the gate-control theory (Melzack & Wall, 1965). This suggests that pain can be understood through a complex stimulus-response pathway, mediated by a network of interacting processes (Ogden, 2007). In crisis pain the 'sickling' of red blood cells block small blood vessels which stops the flow of blood. This sickling can be severe and enduring (Ballas, 2007) resulting in both acute and chronic pain, sometimes simultaneously. It is unclear whether chronic pain in SCD is caused by a similar process as suggested by the gate-control theory or through on-going red blood cell sickling, or other complications (Jacob, 2001).

Research has moved towards a bio-psychosocial model of pain, where psychological, social, cognitive, physiological, and behavioural factors are hypothesized to interact and result in individual pain experience. For example the common sense model of

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illness representations (CSM; Leventhal, Meyer, & Nerenz, 1980) highlights the importance of individual beliefs, specifically about illness. The model encompasses the idea that illness representations provide patients a framework for coping with and understanding illness including symptoms like pain. However, this model considers single episodes of illness rather than a chronic condition where illness and 'normality' become one. Thus, theories focus on the acute pain experience; chronic pain and the interaction between the acute and chronic pain are not considered. There is no over-arching model that attempts to account for this pain experience (Smith, et al., 2005).

Investigating the individual's experience is of particular importance to understand how individuals with SCD understand their pain, as well as what factors influence their health behaviours, coping and treatment adherence. McClish et al. (2009) found an average of three out of 16 possible pain locations which varied with age, depression, frequent pain days, crisis and unplanned hospital visits. When considering quality of life McClish et al. (2005) reported that SCD participants scored significantly worse than national norms on all subscales except mental health, and that increased levels of pain were represented by a significant reduction in scores.

Such studies provide initial information about the experience of SCD pain, however questionnaire based studies often impose responses on participants rather than give individuals the opportunity to respond using their own words (Barker, Pistrang, & Elliott, 2002). Qualitative designs allow a search for meaning through in-depth and detailed descriptive interpretations of individual discourses (Smith, Harré, & Van Langenhove, 1995). Radley (1999) argued that illness can only be studied qualitatively because illness always

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appears in relation to how individuals represent their world as a result of the occurrence of disease and this is embedded in experience and the social context.

Some studies have attempted to understand the experience of those with SCD using qualitative methods. Caird, Camic and Thomas (2011) found that SCD had an adverse effect on the physical, psychological and social functioning of individuals. Thomas and Taylor (2002) highlighted that SCD affected multiple domains of life including education, employment and relationships. A few studies have specifically focused on the experience of pain, for example Booker, Blethyn, Wright and Greenfield (2006) identified that feeling isolated, not listened to and limitations in social support affected patients' ability to manage pain. Such research however, does not explicitly focus on the experience of pain. Booker et al. (2006) stated that despite the focus of the study being pain, participants drew the investigation away from the experience of pain to the wider impact that pain imposes on everyday life. Although this was clearly important to the participants, and has been noted by previous authors to be common within CI (Miles, Curran, Pearce, & Allan, 2005), it does not leave the reader with a sense of what it is like to experience debilitating pain.

Understanding such pain is vital as crisis pain is the most common reason for hospitalisation. Especially as research has identified that some participants actively avoid health care professionals (HCPs) whilst in crisis as a result of repeated experiences of not feeling believed or understood (Booker, et al., 2006). Hayward et al. (2011) found that negative HCP attitudes towards SCD patients was an important barrier to receipt of appropriate pain management. Patients reported hospital staff lacked empathy for the excruciating pain experienced (Thomas & Taylor, 2002) and that this resulted in distrust regarding analgesia, with HCPs believing they were merely drug seeking (Weisberg, Balf-

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Soran , Becker, Brown, & Sledge, 2013). Patients believed that due to this stigmatisation they were treated differently (Maxwell, Streetly, & Bevan, 1999)

An in-depth exploration of the experience of SCD pain will provide additional understanding about the subjective and physical experience of pain specific to SCD. Such an account will give a valuable foundation on which a model of pain in SCD can be developed. This research will also support HCPs ability to understand patient experience, with the potential to improve relationships and health outcomes. This includes the ability of psychologists to understand the specifics of patient experience and provide support and therapy in a way which feels meaningful to patients.

Aim

This study aimed to explore the following four questions:

- How do adults with SCD experience acute and chronic pain?
- How do people with SCD experience HCPs?
- How do adults make sense of their pain?
- How does the experience of SCD pain fit with current theories and models from the illness representation and chronic pain literature?

Method

This study qualitatively explored the experience of SCD pain using Interpretative Phenomenological Analysis (IPA) to analysis individual interviews.

Participants

Participants were adults with a diagnosis of SCD who experienced SCD pain daily or on most days. All had attended or were in contact with a community based SCD support group in north London. A total of 7 participants were interviewed, 4 women, 3 men, aged 24-57 (mean=40) (see Table 1). This sample size is consistent with that recommended by Smith, Flowers and Larkin (2009) for IPA studies. The recommended purposive homogeneity of sample (Smith, et al., 2009) was established by recruiting through a single SCD support group. The sample was not assumed to be representative of the broader population of adults with SCD.

Table 1. Participant demographics NB. Names used are pseudonyms.

Pseudonym	Age	Gender	Diagnosis	Nationality
Adebola	49	M	SS	Black British
Benjy	32	M	SS	Black British African
Charlotte	24	F	SC	Black British African
Deborah	43	F	SS	Black British African
Edith	57	F	SC	British Caribbean
Femi	36	F	SS	Black East African
Glen	41	M	SS	Black British African

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Procedure

The facilitator of the SCD support group was contacted by the researcher to discuss the project. Following this the researcher attended a SCD support group to present the project to those in attendance, including information regarding informed consent, confidentiality and right to withdraw. A time for questions was given and patient information sheets provided (Appendix 5). At the end of the evening seven individuals agreed to be interviewed. Interviews were arranged at times and locations convenient to participants, mainly at the location of the SCD support group but also at individual's homes and workplace.

Ethical information was repeated and consent forms (Appendix 6) signed immediately before interview. The length of interviews ranged from 30 to 120 minutes. Interviews were recorded and transcribed. Post-interview participants were given the opportunity to discuss their interview experience and responses to the topic.

Interview

A semi-structured interview schedule (Appendix 7) was developed to flexibly explore participants' views. The schedule was based on the research questions and reflected IPA best practice by ensuring open ended questions (Smith, et al., 2009). Interviews aimed to capture participants' pain experiences, how they made sense of this and communicated it with others, in particular HCPs. The interview was piloted with a patient-consultant which encouraged helpful reflection and led to an additional question related to changing experiences.

Analysis

Interviews were transcribed and analysed using IPA (Smith et al., 2009). IPA allows the focus on subjective meaning and how people make sense of their experiences (Smith et

al., 2009). IPA is interpretative because it explores the “double-hermeneutic” of the researcher trying to make sense of participants’ attempts to make sense. It is phenomenological as it examines participants’ experiences in detail (Smith & Osborn, 2003).

Each transcript was read several times. Notes were made about questions, meanings, understanding and the sense made by the participant and researcher. Relevant text was coded, and re-examined using the researcher’s interpretative lens (Appendix 8). Codes were gathered together from all seven transcripts to represent emerging themes. Master themes were created, adjusted and rearranged until they reflected the researcher’s interpretation of participant experience. Master themes were grouped into overarching themes, which reflected the experiences of adults with SCD pain.

The researcher is a trainee clinical psychologist with some experience in qualitative studies and an interest in chronic pain.

Quality Assurance

Prior to conducting the project, guidelines for conducting IPA were consulted (e.g. Saldaña, 2013; Smith et al., 2009) and established guidelines for carrying out quality assurance (e.g. Elliott, Fischer, & Rennie, 1999; Yardley, 2000) were followed in order to assure ‘commitment and rigour’. To demonstrate ‘coherence and transparency’, an independent audit of codes was carried out by a psychologist and an academic with experience in IPA. This aimed to ensure that themes were grounded in the data (Yardley, 2008). The model of themes generated was sent to the first participant (the SCD support group facilitator) to ensure it reflected participant experience. In addition a clinical psychologist working in an SCD service (supervisor) was consulted regarding model

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development. A reflective journal was kept (Appendix 9) and regular reflective discussions with supervisors took place. A ‘paper trail’ was kept showing all stages of the analysis.

Ethics

Ethical approval was received from the Salomons Ethics Committee (Appendix 10) at Canterbury Christ Church University. The BPS code of Ethics and Conduct (2006) was adhered to throughout the study.

Results

Three overarching themes and 11 master themes were identified, each broken down into numerous sub-themes. Master themes are identifiable both within and across participants; narratives around an aspect of their shared views or experience are represented by master themes. Themes do not always represent independent concepts.

Overarching themes	Master themes	Themes
Experiencing unimagined pain	Indescribable pain	Worse than the worst pain imaginable
		Use of analogy
		Varied within the same person and between different people
		Unpredictable
	Affecting every aspect of life	Every-day functioning
		Planning and the future
		Parenthood
		Emotional impact
		Complications

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	Normal rules don't apply	Crisis pain vs every-day pain Being misunderstood
The dilemma of treatment	Treatment limitations	Medication is not enough - Pain breaks through
		Tolerance to medication
		Own expertise ignored – misunderstood/not believed
		Understanding HCPs position
		Passports didn't work
	The impact of Health Care Professionals	Level of experience and knowledge
		Level of interest
	Building relationships	Inconsistent staffing
Positive relationships make a difference		
Finding a life with pain	Struggling to understand	Varied explanations for pain
		Unknown triggers
		Role of self – blame
		Inseparable part of life
	Pain as a thing	Relationship with the pain
		Fighting vs appeasement
	Living with pain	No choice but to cope
		Knowing self
		Self-care
		Support networks
	Critical sense of time	Knowledge

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		Age
		Experience
	Acceptance	It's not going away
		Appreciating the small things/life
		Positives lessons about self

Table 3: Overview of themes

Overarching themes will be discussed sequentially. Master themes will be illustrated using verbatim quotations³. Due to space limitations not all themes and sub-themes can be discussed in depth. (See Appendix 11 for additional quotes.)

Overarching theme 1: Experiencing unimaginable pain

All participants spoke about living with extreme pain as a result of SCD.

Indescribable pain. All participants' had difficulty describing SCD pain; Edith said "I don't know how to describe pain." Participants declared that it was "the worst pain." Adebola described it as having "no cut-off point." Deborah reported that "on a scale of one to ten ... it was twenty." Those who had given birth rated SCD pain as much worse, Edith said during crisis pain she thinks "just let me have another baby... labour is piece of cake (compared) to this."

Consequently participants tended to use analogies to try and describe the pain. For example Charlotte had this "visualisation" ready as she was often asked to describe the pain:

³ . Short pause
 ... Brief section removed for clarity
 (to) word added for clarity
 [doctor] Altered for confidentiality

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It's just like somebody, is like hammering inside your bone and drilling, and it's like all the way (inside), it's like you can't even point to it, it's like so far in that it's yeah, wherever it is its bad, it's horrible.

Similar to Charlotte, Deborah said "it's like knocking, it's like a wood pecker that's go ddddddddddddddd in the blood stream." However, analogies varied greatly: Adebola described it as a "being set on fire continuously;" Benjy pictured it as a "lightning and thunderous storm, cause when the pain first starts, it strikes at different positions around my whole body;" Femi described it "like a scorpion" that stings with a poison that spreads throughout the whole body; Glen explain that it is like being in a leaky boat in the dark surrounded by sharks, "you've got this boat, which is better than being in the water with the sharks but you know that gradually this boat is going to start sinking."

Even when using such descriptive techniques participants' remained unsure if this accurately explained the pain. Benjy said "I don't know if my description would fully explain." For many the pain could not fully be understood by someone who had not experienced it

The difficulty describing pain was made more complex as everyone's experience is different. Edith said "there's no two sicklers the same." Furthermore, an individual can experience a variety of different pain experiences. When talking about his pain Adebola said "it isn't just the one type of pain...it depends on the intensity of the pain and where that pain is."

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The unpredictable, changeable nature of SCD crisis was also problematic. Femi described that her pain “just comes suddenly” and Glen commented that it is “without warning.” A few participants said that it is this unpredictable element that makes SCD so difficult to live with. Benjy put it this way “it’s almost like it would be easier if it was a constant thing, then everyone would know what parameters you would work to and within.”

Affecting every aspect of life. Participants reported that SCD impacted every aspect of life: Every-day functioning, Adebola said “if I were to get up I’d be in more pain but I need to go to the bathroom;” employment and social life Glen said “you can make commitments to other people and then have to just drop that commitment;” planning for the future, Glen reported that he “couldn’t get a mortgage ... because for the mortgage you have to have life insurance and they would not give (him) life insurance because (he) had this pre-existing condition.”

Difficulties emerged when considering marriage and parenthood. Femi was angry that her parents did not consider their haemoglobin before they had children; “my parents are stupid enough not to check themselves.” Glen spoke about the reality that SCD ultimately reduces life expectancy; “I can find myself wondering will I see my daughter grow up.”

The emotional impact of living with SCD was another consideration; all participants mentioned negative feeling such as feeling sad or lonely. Adebola recognised that “with pain all the time your mood does change.” Deborah claimed every SCD patient would experience such emotions “the first thing that we as a sickle cell suffer, that you go through, you get depressed.”

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Other health complications such as stroke were also frequently recognised; which increased participant's suffering and further limited life. Adebola explained that "in some ways that (SCD pain) is the easy bit:"

Normal rules don't apply. Participants explained that they experienced both chronic and acute pain simultaneously, that is an every-day pain and crisis pain. Adebola differentiated between the two "every day is just the ache. The sickle cell crisis is the sharp intensity, and the ache." There was a sense that SCD is unique and that the normal rules do not apply for example, Deborah explained that when considering blood transfusions if "it goes below seven... that's transfusion time but my blood level has been six for the last seven years, eight, nine and ten years and I am OK."

Some participants suggested that SCD pain was different from the accepted concept of pain, which is "generally a signal that some sort of trauma had occurred or that it was a way for you to avoid particular stimulus" (Glen). Others however saw SCD pain as a warning sign that something was wrong, like traditional models of pain suggest, Deborah said it "means something's wrong within the system."

Regularly experiencing such extreme levels of pain required strong medication however participants reported that this did not fully control the pain; Femi declared "the pain keeps shooting though the medication." Adebola said he had "been in an induced coma and still been in pain." Deborah had experienced the same.

Overarching theme 2: The dilemma of treatment

All participants needed some kind of regular medical regimes, including daily medication, analgesics and blood transfusions. In addition to this, medical support is often sought when experiencing crisis. However, treatment interrupted life, had limited effect and often increased stress due to problematic relationships with HCPs.

Treatment limitations. Participants suggested that regularly experiencing extreme levels of pain had resulted in a higher threshold for pain. Deborah said “I can handle pain much more because I’ve been through sickle pain.” This resulted in participants presenting for treatment when pain levels are at unbearable levels. Edith said “if you can manage the little ones at home, you don’t need to go to hospital you can still get on with life somehow.” Consequently when participants sought help they often required more analgesia than HCP could prescribe.

They give you more and more opiates and it takes away a certain bit and then you build up a tolerance for that ... and it gets to the point where nothing happens and you just, . you’ve just got to put up with it, and it just, ... it’s like a huge silent scream. (Adebola)

Unfortunately, as “you can’t see any physical proof” (Charlotte) of pain participants often felt misunderstood or not believed especially in relation to the need for extra medication, at times causing mistrust and misunderstanding with HCPs. Participants described how they or someone they knew had not been believed about their level of pain and need for medication. Adebola had created a saying to explain this; “pain is what the patient says it is unless... you’ve got sickle (cell) and then it’s what the doctor says it is.”

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Frustration was expressed at having a lifelong condition yet personal experience being ignored, Glen explained that:

They want to try this thing ... that you know is really going to be a very tortuous experience for you but you have to do it because otherwise it's like, well why is this guy being so resistant to this thing or maybe he does want drugs.

Participants were aware that not complying with the HCPs recommendations could leave them considered as drug addicts, Adebola said "the one thing we get labelled as is junkies. They just think we're there for opiates."

Yet most participants expressed understanding for HCPs due to the restrictions imposed by the profession. Femi described the process:

It's also very difficult ... for them 'cause ... they kind of just can't believe that you've just had this dose and within 30 minutes or so you're still feeling the pain and you're wanting more medication ... and probably ... their profession doesn't allow them to give you any more.

Patients also talked of resenting spending time repeating standard and possibly irrelevant information, such as when they were diagnosed. Femi said "...all (they) really need to know is what medication are you taking, what have you taken, or y'know who is your consultant." Femi mentioned that at one point she had a passport to give to HCPs which contained most of this information in an attempt to overcome this difficulty and

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provide a fast-track system however she said “it was useless” because staff did not look at it.

The impact of HCPs. A major influence on experience that was recognised by most participants was the importance of the particular HCP managing their care. HCPs with less knowledge and experience seemed to lead to misunderstandings and at times incorrect treatment. Glen explained the difficulties with HCPs, “they’ll always use their past experience (which) will tend to be at odds with the way erm the experiences that someone with sickle cell has” whereas he said “when you’re dealing with more experienced doctors they understand.” Many participants had been left waiting hours whilst in agonising pain; Charlotte explained “I’ve been to A&E and been given my morphine and just been left there. Like literally for hours.”

For others lack of knowledge had a more harmful outcome, both Deborah and Benjy spoke about medical mistakes in the past that had detrimental effects on their long term health; Benjy had suffered a stroke when 12 years old as a result of HCPs not bringing him to the attention of the doctor because they “weren’t aware how bad it (SCD) was.” Glen spoke about a friend who had died in agony at the hands of an inexperienced doctor who would not prescribe analgesics; he said “it was the fact that she ... was so uncomfortable before she died.” Glen reported that this incident left him feeling “paranoid” about receiving treatment from inexperienced HCPs. Deborah too had reservations about receiving care from those with poor knowledge she said “I have (had) doctors that tell me they don’t know how to treat me... literally never heard of sickle cell disease.”

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Other participants described varying levels of interest from HCPs, for example Glen spoke about doctors on rotation who “could not care less.” Charlotte described HCPs attitudes as uncaring, that they would “just give her morphine and shut her up.” Adebola reported that it was “quite common” for HCPs to hold the opinion that patients just “wanted drugs.” He reflected how this judgment affected how HCPs treated patients, reporting a conversation following a comment about him “liking drugs;” “You’re supposed to be looking after me and people like me and your attitude, and your opinions affect the treatment you give me.”

Another problem highlighted was being treated by numerous different HCPs. This increased the amount of repeated information, the quantity of unsuccessful treatment trails and/or wait times and increased the likelihood of meeting someone with an unhelpful attitude. Where possible participants would go to the same hospital in order to avoid this but it was not always possible.

There was general agreement that haematologists and specialist teams showed genuine interest and provided superior treatment. Charlotte considered that “anybody that’s not a haematologist or specialist has some sort of attitude.” Glen reported that “the house that we’re moving into now, (is) simply so that I would be in the catchment area for [hospital].” Developing a personal relationship was identified as an important aspect of good quality care.

Building relationships.

For Femi building relationships meant “(knowing) which doctor (you’re) dealing with or which profession you’re dealing with, whether they really understand the illness

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and how it affects you.” Many participants reported improved quality of life as a result of receiving treatment from consultants and specialist nurses they knew well, for example Edith stayed in work longer, Benjy was able to reduce his medication and Glen reported:

the actual basic way they treat me, on observing me, on my experiences, and they treat, they also tend to treat us as a whole, more like adults...the fact that they are willing to make that effort for you ... they improve my quality of life no end! ... it really makes a huge huge difference.

Overarching theme 3: finding a life with pain

Participants talked about the difficulty in maintaining a life living with unimaginable pain while complying with the difficulties of a medical regimen.

Struggling to understand. Throughout their lives participants had attributed different explanations for the pain. Growing up in Africa or away from metropolitan London appeared to increase the number of non-medical explanations. Benjy grew up in Nigeria where he reported that “there was absolutely nothing that I did differently from the other kids.” Femi did not receive a diagnosis until she moved to England aged 20. She reported:

“in Africa there are associated with so many other things... maybe you have an infection, or its malaria or it’s, it’s got to do with your genes, or it’s y’know, things like suspicions,... sometimes they think oh maybe they have ah sent you bad omens...”

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All participants demonstrated some level of medical knowledge for the presence of SCD and crisis however there remained uncertainty regarding individual differences such as crisis triggers. Causation was questioned and phrases such as “for some reason” were often used for example Adebola said “the pain in my legs, which for some reason I tend to get a lot of” and Edith “I have had pain and I’ve thought where’s this coming from, why am I getting pain there?”

Participants often looked to themselves and their actions in relation to unknown triggers. Edith said “I make it worse because if I do move ... then I regret it and it makes me feel worse.” This highlights the role of self in SCD pain. Glen said “if you start to do something and the pain starts flaring up, stop doing that!” Adebola noted the role of emotions on pain, “if things are troubling you, how you are dealing with it... so you’re not as effective at dealing with it or the levels of the pain that day.” Edith also noted the role of emotions “I think what have I done? Am I stressed? Do I have a problem? I always I sort of analyse myself.”

For all participants pain had become an inseparable part of life, Benjy said “it’s part of who I am.” Glen suggested that he would “have no idea what life would be like without it.” Adebola suggested that pain was so much a part of his identity that removing it would be “like trying to separate me from my colour.” He also suggested that if the pain was taken away “... you’d probably feel some kind of bereavement.”

Pain as a thing. Participants spoke about the pain as if it was a tangible thing, a living entity almost. Participants established a relationship with their pain; the majority negative. Deborah said “you wanna kill the pain” and Glen expressed a “difficult relationship,” “utter

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hatred” and that the pain was “kind of spiteful.” Charlotte said the pain is “my enemy like I just hate it.” Benjy however chose not to personalise the pain, he saw it “just as a condition that has afflicted me from birth and I deal with it daily... I'm used to it.”

Edith reported “I hate it obviously but I try to be friends with it I try and be friendly with it, you know because I don't want it to get worse.” Similarly Femi describes building a relationship with different aspects of the pain:

you just hate it ... then there's the one you know which is ... the day-to-day ... you ... accept it ... you build a relationship with it, its going to be there you've got to work around it and deal with it every day.

There was a sense of a fight or an on-going struggle to with this thing called pain. Benjy recognised that this is not always easy “I do find it extremely hard to cope, but I am very strong willed and love being alive even if not always pain free.”

Participants approached this in different ways; some participants declared war on their pain. Adebola described entering “battle mode” and that although he was aware of the pain it “wouldn't sort of stop (him) from doing stuff.” Femi described the pain as “it's either there and you fight it or not there at all.”

Others attempted to appease it, Glen said “I had this erm pain sort of flaring up and so sort of have to stop and take it easy for maybe a couple of hours cos you need it to to sort of subside.” Whereas Edith went a step further and tried to befriend the pain she said “there's no

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good sort of getting angry and fighting” for example if she woke up in pain she would think “I know you there ... so I think ah well take it easy today, don’t upset it.”

Living with pain. Participants vocalised their lack of choice in the situation. Deborah said “its constant and I never can chose whether it’s there or not it’s just always there.” Likewise Adebola reported “it’s just always there, so you, y’know you both cope but it’s very different. There isn’t a sense of ‘I’ll cope here’ I cope because . Errrr . there’s no choice.”

A tactic in this struggle was for participants to know themselves and their bodies. Charlotte said that she is “more conscious of (her) body” and that she has to “make sure I’m wrapped up warm” because “if I like let it slip or let my guard down I would have ... a crisis.” Similarly Deborah said “I know my system well enough” she had a series of checks she performed when feeling unwell and advocated that all SCD patients should “know (their) body because your body speaks to you.”

Knowing one’s body allowed participants to partake in self-care activities. Femi noted “when pain sets in, I go ... what do I need to do, ... (I) do the things you are supposed to be doing properly and you recover from it.” Charlotte recognised the importance of self-care, “I used to sickle so much more, which is why I believe that doing little things like making myself warm and stuff makes a big difference.” Benjy spoke about the important role of managing medication “I always need to have medication on me you know because (crisis is) that quick.”

A number of participants reported the important role support networks had in achieving a life with pain. Glen reported that at his worst his wife “can step in and help.”

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Edith recognised that a reason she is able to continue working is because of the supportive environment of her workplace; “it is very good ... because if I didn’t have them round me it (SCD) would have more of an effect on me.”

Critical sense of time. Initially having limited knowledge about SCD was common; Femi reported that “I didn’t know much about it.” Edith’s consultant provided her with a lot of knowledge “he told me about, all about, what, how, how my, my diagnosis will effect me.”

Learning about SCD and how to manage it appeared to develop over time with age. Charlotte reported “when you’re younger you don’t really realise that the cold makes you sickle and stuff.” For Glen age was critical, he said “most of the time young people are not understanding about illness at all, y’know? When you’re 16, as far as they’re concerned they’re indestructible.”

Experience also played a big part in managing the pain. Benjy said “when I’m in pain now I know what I need to take.” Edith noted that in the past she used to “be running around too much” when feeling well which made herself ill and later “regret it” however repeated experience taught her to slow down. Adebola summed it up: “hopefully the, the older I am, the longer I live, the better I get at dealing with (pain).”

Acceptance. Similar to learning to manage pain participants felt there was no choice but to accept that pain was part of their lives and it was not going away. Charlotte said there is “No choice but to accept, we won’t be cured, it’s either you live with it or you die.” Adebola said:

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I kind of figured out a long time ago that if I sat in bed and did absolutely nothing, didn't go anywhere, didn't do anything, didn't take any risks, didn't play a sport, I'd still be in pain, I would hurt no matter what I did... So if that's the case I might as well enjoy myself.

Edith described a process:

Every time I had a crisis I think I started cussing less cause I knew I just had to get used to it, it wasn't going anywhere it was here there's no cure ... but its taken probably about 30 40 years to get to this.

Part of this process appeared to be developing an appreciation for the minutiae of life or life itself. Benjy reported he 'loves being alive' while Adebola when talking about material things said "I don't get caught up in all that stuff, it's not really important;" he had learnt to appreciate "the simple things."

Participants also acknowledged what the experience of living with SCD had taught them about themselves. Femi reported that the experience teaches you about "what kind of a person you are... either you're somebody impatient or somebody very patient or somebody strong." Similarly Charlotte had learnt that "I've overcome it so many times... it makes you stronger."

Summary of Overarching themes

For the seven individuals with SCD pain the experience was indescribable, the use of analogy suggested unbearable pain that was agonising, constant, inescapable and without

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limit. Some participants reported feeling pain despite strong analgesics, while others were placed in an induced coma because the pain was so severe yet still felt pain. It is perhaps impossible to understand such unimaginable pain without experiencing it. Accordingly individuals experienced being misunderstood or not believed about the severity of the illness. This was compounded by the vast variety of experiences both within the individual and between different people. The disease impacted every area of life from day-to-day activities, planning, hopes for the future and emotional distress.

Participants had no choice but to engage in medical treatment however this posed a significant dilemma; although medication helped at times it often was not enough to completely relieve the pain; consequently patients were often mistaken for drug seeking. This unhelpful HCP attitude was compounded by a lack of knowledge, experience or interest in SCD. However, most participants had experienced good health care provision, to the extent that individuals would do their utmost to ensure their treatment continued at a location deemed suitable.

Alongside this dilemma participants attempted to understand the disease and the impact on life in order to live a life despite the pain. Participants described the pain as a thing they chose to fight, not giving into it whereas others befriended the illness so as to appease it and not make symptoms worse. Knowing one's self, including bodily symptoms and the implications of individual choices was key to implementing self-care strategies. Participants noted that such abilities developed as their knowledge of SCD matured with age and experience. Ultimately participants felt obliged to accept their illness as it would never be cured but were able to appreciate life and recognise positive life lessons as a result of living with SCD.

Discussion

This study aimed to gain a clearer understanding about the experience of pain in SCD and the relationship with HCPs. The research questions will be addressed below.

How do adults with SCD experience acute and chronic pain?

The variety of pain experienced both within an individual and between different people has been identified in literature (Taylor, Stotts, Humphreys, Treadwell, & Miaskowski, 2010). The unpredictable nature of crisis pain is also well documented as causing difficulty for patients to actively function in society (Caird, et al., 2011). Further the notion that pain is often misunderstood is documented (e.g. Thomas & Taylor, 2002). Participants found it difficult to describe this pain, referring to it as the worst pain ever. The use of analogy was vital to help others understand this experience however it was felt that such strategies were not sufficient to explain unimaginable pain.

The use of analogy to describe SCD has not been widely documented, if at all. Some participants had analogies prepared in order to communicate what they otherwise found impossible. Analogy content varied and some participants' descriptions were complex and detailed allowing for the complexity and variety of life limiting SCD pain. The use of varied analogy suggests that traditional measures of pain, such as the McGill Pain Questionnaire (Melzack, 1987) with standard rating would not fully encompass the pain experience for those experiencing SCD.

Participants experienced 'pain as a thing,' by attributing human characteristics and emotions to the pain, such as it being spiteful or an enemy. This personification of the pain

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appeared a key element in managing the pain experience and perhaps successfully establishing a life with pain.

A number of participants expressed the idea that SCD pain is different from other experiences of pain; that the normal rules do not apply. Hence it may be pertinent to suggest that SCD requires its own pain classification and acknowledgment that current models of health and illness do not fully explain the experience. Additionally it may be appropriate for patients presenting for medical interventions, particularly in crisis, not to be asked to rate their pain based on the current understanding of pain but a new enhanced model incorporating the unique features of SCD pain. However, other conditions share simultaneous occurrence of acute and chronic pain (e.g. fibromyalgia ‘flare ups’, Sturge-Jacobs, 2002) signifying that SCD might not be fully unique in all aspects identified by this research.

How do people with SCD experience HCPs?

SCD treatment is varied and essential, but medical intervention is limited, both in terms of pain relief and in the safety restrictions on medications (Taylor et al., 2010). Participants wanted HCPs to understand SCD and appreciate their own expertise however the problem of opioid addiction is well documented (Ballantyne & LaForge, 2007). Research has shown that when using DSM-IV criteria for substance dependence 31% of SCD patients met the criteria when pain-related symptoms were included whereas only 2% met the criteria when focused on non-pain symptoms (Elander, Lusher, Bevan, & Telfer, 2003). This is a complex issue, patients are experiencing extreme levels of pain that require high dosage analgesics but, HCPs are bound by professional guidelines and procedures. Some participant’s recognised the difficult situation HCPs faced but felt their personal experience and expertise from living with the disease most of their life was ignored. It is arguable that

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during crisis patients are seeking drugs as a means to relieve crippling pain. However, participants in this study expressed that this did not indicate drug dependency rather it was necessary to achieve some pain relief.

How do adults make sense of their pain?

Participants attempted to understand and manage the pain in order to find a way to live despite the pain. The search for meaning varied across participants, with various explanations for pain and the experience of crisis. The variety of explanations suggested a lack of knowledge amongst participants, with some demonstrating more accurate knowledge than others. This could be crucial in terms of acceptance but also in the argument about patient expertise not being valued by HCPs. Positive relationships with HCPs appeared to help develop mutual understanding and knowledge.

Interestingly all participants recognised that SCD was an integral part of the self but few mentioned other identities, such as motherhood. This is in contrast to other research that identified the importance of participants being able to define themselves as separate from SCD (Caird et al., 2011). It appears that for this group of SCD patients recognising the fundamental part SCD plays in their lives was important to acceptance and coping. Also this studied differed as spirituality did not seem to come up as a particular ‘meaning’ or way of making sense of the pain.

Various self-care strategies have been documented throughout the literature (e.g. Anderson & Asnani, 2013). This population also noted the role they had in the pain experience, such as knowing their body and its limits. There was a critical sense of time in developing this understanding; Edith described a process that took her 30 to 40 years to

complete. This supports the idea that coping is a process that proceeds through stages with the inclusion of learning to appreciate the minutiae of life or positive lessons learnt (Caird, et al., 2011).

How does the experience of SCD pain fit with current theories and models from the illness representation and chronic pain literature?

The CSM incorporates the importance of individual beliefs or illness representations (Leventhal et al., 1980) based on five belief domains; identity; cause; time line; consequences; curability and controllability (Leventhal, et al., 1997). The themes identified in this research map onto these domains (see Appendix 12). Thus the model provides some insight into how the participants have arrived at their particular place of understanding; for example it identifies the influences of self, interpretations, appraisals and coping on the formation of illness representations, and the impact of these representations on these areas. The idea of learning through time and experience can be represented by the problem solving element of the model, where patients make appraisals and implement change based on the outcome.

The CSM, however, is less equipped to capture the continual nature of this life long, chronic illness. The model is circular, in that the process is continually repeated in a cycle whereas in CI illness representations must be constantly updated, with participants moving towards acceptance over time. The model is not able to explain this progression and moving forward. Therefore a model is required that would be able to incorporate this process of moving forward.

Limitations of the study

A potential limitation of IPA is the small, homogenous, purposive sampling. Participants were recruited through a single SCD support group and although a gender mix was present and age varied, ethnicity was similar across the sample. Small study sizes are crucial to allow in-depth analysis which would not be possible with large sample sizes. Further, it has been argued that the ideographic nature of IPA is suited to sample specificity which allows for better pattern analysis (Smith, et al., 2009). Qualitative studies do not normally set out to generalise findings (Ryan, Coughlan , & Cronin, 2007) but a process of theoretical transferability has been suggested, whereby linking findings to existing literature enhances current research (Smith, et al., 2009).

The sample population was limited; participants were recruited from a single SCD support group. Membership in such groups potentially demonstrates certain personal characteristics, for example interest in engaging with other individuals with SCD or a desire to increase awareness (a focus of the group) therefore such individuals might be more interested in partaking in research and may have a different narrative about their experience of SCD than others who do not engage in such groups. An inclusion criteria was to experience pain daily, some people may have SCD and experience pain much less regularly. In contrast, those with more severe pain may not be able to attend the group meetings and so were not included in the sample. Consequently, the sample's experience of pain might not have represent that of others with SCD.

This research considered the relationship with HCPs however the opinion gathered was localised to this participant group. The research could have been enhanced had it considered the relationship from the perspective of HCPs and triangulated the findings.

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Triangulation can increase rigour by enhancing the completeness of the findings (Yardley, 2008), thus doing so could have identified different elements and subsequently improved understanding.

Future Research

SCD is predominately in black African and Caribbean populations however it is also present among other populations, such as Asian and Mediterranean. The current research was undertaken with a mainly black British African population, further research is required to investigate if this model is representative of adults with SCD across different ethnic backgrounds. It would be of particular interest to investigate the relationship with HCPs in Caucasian individuals as some studies have suggested that HCPs attitudes and behaviours may have a racist element (Weisberg et al., 2013).

The idea of a critical sense of time affecting acceptance of SCD pain was raised in this research. Participants suggested it was related to age, knowledge and experience, however if critical components can be identified through further research it may be possible to help individuals with SCD reach acceptance earlier.

The present study highlights that although the CSM does provide a way to understand the data, it does not fully explain the process involved for SCD patients. More research is required to explore this in order to incorporate the process of acceptance and moving forward that appears to exist in SCD pain.

This research suggested that existing measures of pain do not fully represent the experience of SCD pain. Future research could consider more appropriate ways to measure

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such pain, or indeed if measuring pain in a clinical context is necessary in SCD. Consideration should also be given to models of health and illness to assess their suitability to represent chronic conditions such as SCD.

Clinical Implications

The findings of this study support previous work regarding the variety of SCD pain experienced and the vast array of implications the condition creates. Clinicians working with SCD should therefore not only focus on the physical pain but the psychological impact. This indicates that MTD working is essential to ensure that all psychosocial needs are addressed. Psychologists should play an important role in such teams in order to keep psychological aspects central in all facets of treatment.

Participants found it difficult to communicate their experience of pain and often felt misunderstood by both personal and medical contacts. A potential therapeutic role for psychologists could be developing existing communication skills to help individuals articulate their experiences, perhaps using analogies as the participants in this study favoured. Additionally, this study supports findings that individuals pass through stages in the process of acceptance which includes learning to appreciate life. It is therefore important that psychological interventions do not focus exclusively on deficits but incorporates the strengths and expertise of those with SCD.

According to this research misunderstanding and lack of knowledge about the experience of SCD pain in HCPs has a detrimental impact on SCD patients. It would therefore be important for findings such as these to be made available to a wider audience, for example through a leaflet for HCPs not based within specialist teams. Psychologists could

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also be involved in developing and delivering training packages for HCPs in order to increase knowledge and understanding and ensure a bio-psychosocial approach is presented.

Conclusion

The findings indicate that SCD pain is unbearable, agonising, continuous, inescapable and limitless. Analogy can be used in an attempt to describe pain but patients repeatedly experience being misunderstood and not believed about the severity suggesting that it remains indescribable. Participants develop understanding and coping skills over time but medical treatment raises complex difficulties; pain does not always diminish with medication and relationships with HCPs can be difficult at times making the treatment experience worse, especially in relation to the need for high levels of analgesia and the unimaginable extremes of SCD pain.

These initial findings suggest that psychosocial training may be required for HCPs not based within specialist teams. Current models of health and illness are useful to understand some aspects of SCD pain but are limited in describing the process of acceptance and moving forward. However further research is needed across a wider SCD population and incorporating the perspectives of HCPs in order to corroborate the findings.

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Section C: Critical Appraisal

Word Count: 1979

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Running heading: CRITICAL APPRAISAL

This is a critical appraisal of an interpretative phenomenological analysis (IPA) study exploring the experience of pain in adults with sickle cell disease (SCD) including consideration of the relationships with health care professionals (HCPs). Four questions will be answered and wider clinical and research issues will be considered.

Skills Learnt

What research skills have you learned and what research abilities have you developed from undertaking this project and what do you think you need to learn further?

Prior to the clinical training course I had some minor involvement with a few research projects but little formal research experience. This experience had mainly been quantitative but had provided me with some exposure to qualitative methodologies. I had often found quantitative practices frustrating as I wanted to know what participants were thinking or feeling about the topic, or about their experiences and understanding of the situation. Naturally this led me to developing a research project incorporating these areas using a qualitative approach.

Due to my lack of experience with qualitative methods, an initial challenge was identifying research aims and choosing an appropriate methodology. This time consuming process involved reading about different approaches, thinking about exactly what it was I wanted to explore and considering existing literature. Through this process, and specifically finalising research aims, it became clear that a qualitative method was indicated, which led me to an increased appreciation for the value of this methodology (Elliott, Fischer, & Rennie,

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1999). Furthermore, the process enhanced my knowledge about qualitative methods, particularly my chosen method, IPA (Smith, Flowers, & Larkin, 2009) and complementary guidelines on assuring quality (Elliott, Fischer, & Rennie, 1999; Yardley (2000).

In qualitative research it is important for the researcher to be aware of their own opinions about the topic and the potential influence this could have on the research process. I had first encountered SCD as a teenager when I babysat for a child with the illness. I remember back then being shocked that I had never heard of a disease that afflicts so many people and not being able to fully comprehend what it was like for a little girl to experience such pain. Later, during my CAHMS placement, I worked with a teenager with a severe form of the disease. This experience reminded me of my initial feelings about the disease and that in the ensuing years I had not heard much about it. Then through starting this project I discovered that some of my closest friends had SCD or the trait, yet we had never talked about it. I felt this was indicative of the hidden nature of the disease and the stigma that can be attached. Reflecting on aspects such as these in my clinical work has become normal. However, despite trying to 'bracket' my own ideas, I began to realise that it is impossible to completely separate interpreting data from your own pre-conceptions. Through the use of a reflective diary and supervision I was able to consider my own experience, the development of ideas and how this could impact my research (Smith et al. 1999).

It was interesting for me to note that initially I was unaware of how similar my own chronic pain issues were to some of the participants' experiences. Initially I was not aware of this connection with myself, perhaps because I was in the process of receiving a diagnosis of chronic pain and was yet to accept my own diagnosis. It was particularly difficult when a couple of participants assumed that I did not experience pain, let alone live with chronic pain.

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One patient told me I did not live with pain which was untrue. I appreciate my pain level is to a lesser extent but I have experienced acute and chronic pain simultaneously during flare-ups, and at times have been unable to walk due to the pain. It was therefore, particularly important to reflect on this during supervision and for me to withhold my own opinions and personal experiences from participants in case my own experiences altered participant dialogue in any way.

Working as a researcher can be a difficult transition for a clinician. When listening back to some interviews, I noticed that at times I was drawn into a therapeutic role, particularly when participants were talking about the future and the progressive nature of SCD. Becoming aware of this taught me that even in a research situation where therapy is not the aim, I am drawn to help people, perhaps entering into a rescuer role. Noticing helped me focus on my position as researcher in subsequent interviews.

Conducting research interviews uses different skills to therapeutic assessment. Interviewing is a critical skill which develops over considerable time (Smith, 2011). Reading through the transcripts highlighted how critical experience was in developing my interview skills. Initially utilising a pilot interview and receiving feedback from a participant helped me experiment with techniques and develop confidence. It was clear as I conducted more interviews that my confidence and ability continued to grow; I was able to ask about more difficult aspects of SCD and was more comfortable to ask questions that diverged from the interview schedule. This enabled a richer level of dialogue to emerge that was more linked to the participant's narratives than my own interests (Yardley, 2008). Additionally, interviewing people in their homes or workplace advanced my ability to manage some challenging issues associated with this, e.g. interruptions from colleagues and children.

Study Improvements

If you were able to do this project again, what would you do differently and why?

Purposive sample of SCD patients was utilised to attempt to select a sample representative of the SCD population (Yardley, 2008) however, there are likely to be important differences between sub-groups of SCD patients which this research cannot comment upon. For example those who grew up in Africa appeared to learn about their diagnosis later, their symptoms being initially regarded as part of 'normal' life experience. It might therefore be necessary to limit a sample by ethnicity in order to identify if such experiences affect the experience of pain, understanding or acceptance.

I would seek to include the experiences and opinions of others. Although the experience of pain is within the individual, partners and family members may be able to provide important insight to how they perceive the individual is managing pain. They may also be able to contribute ideas about understanding and acceptance. Regarding relationships with HCPs, family members might be able to collaborate the experiences of those with SCD and give account of their own experiences. It would also be important to consider the relationships from the perspective of the HCPs in order to develop a clearer picture of the dynamics involved.

A minimal amount of testament validity took place (Elliott et al, 1999) however I would carry out further validity checks to ensure the results with the participants, or ask similar others to check if the model developed represented their experience.

Clinical Implications

As a consequence of doing this study, would you do anything differently in regard to making clinical recommendations or changing clinical practice, and why?

Due to the diverse impact of SCD I would recommend that any psychologist working with an individual with SCD should do so within an MDT. Or if the client is seen in a non-health setting, as for example when I saw the teenager in CAMHS, I would strongly recommend linking with all other professionals involved in the clients care. I did not do this to a great extent and in light of this research regret not doing so. It is important to consider all clients holistically. Because of the complex and varied nature of living with SCD, it is vital that all professionals involved have a good understanding of the experience of living with SCD and that they work together to help improve the individual's quality of life.

This research highlights how difficult it is to understand and appreciate the extreme nature of SCD pain. Consequently this pain is often misunderstood by HCPs. It is important to find a way to decrease these misunderstandings which often lead to tension in relationships and can dissuade patients from seeking treatment (Thomas & Taylor, 2002). This could start with the production of an information leaflet for staff about the experience of SCD pain. Specialist training could be developed, either as individuals or as teams but in order to elicit sustainable change more fundamental action would be required. A shift in the culture of non-specialist SCD teams would be necessary so that all aspects of clinical practice of healthcare teams are affected, for example increased emphasis in professional training programmes.

When talking about my project with various colleagues on placement, most people were not aware of many of the aspects SCD can have. This could be because the population

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of the areas where I worked are predominantly Caucasian;; no one was aware that SCD is present in non-Black populations. These responses replicated my own when I first heard about SCD approximately 15 years ago, suggesting that there has not been much change in awareness over this time. It might not be practical for all staff to receive face-to-face training about a rare genetic condition such as SCD, but I would advocate information being readily available to all professionals about SCD independent of the service or area they work in.

Future Research

If you were to undertake further research in this area what would that research project seek to answer and how would you go about doing it?

Having now completed the research I think a major drawback was not consulting HCPs about their relationship with SCD patients. In future, I would ask what factors contribute to the difficult interactions between SCD patients and HCPs. It would be important to consider both positive and negative relational experiences. It would be interesting to ask SCD participants to identify an HCP with whom they had good experiences, and one with whom they did not. However, this could be difficult to manage in terms of the study being transparent; I would not like to approach a professional for help and subsequently have to explain that a patient thinks they are not good at their job. It could be possible to not tell the HCP how they have been selected but ethical considerations would have to be given to this. Another option would be to conduct the research at two hospitals. One would have a specialist SCD service as well as an Accident and Emergency department, and the other would not have a specialist service. Patients and staff would be selected indiscriminately.

Another possible area for future research is the experience of non-black SCD patients. Although SCD is predominately in black African and Caribbean populations it is also present among other people groups, such as Asian and Mediterranean. It would be of particular

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interest to investigate the relationship with HCPs in Caucasian individuals as some studies have suggested that HCPs attitudes and behaviours may have a racist element (Weisberg, Balf-Soran, Becker, Brown, & Sledge, 2013). In order to investigate this claim a similar research project could be developed that involves interviewing both patients and HCPs about their views of this specific population. The findings could then be compared with the experiences and opinions of black SCD patients and staff working with these patients to identify if there is any difference in perception or treatment.

I would also be interested to investigate the outcome of the introduction of SCD training or an SCD information leaflet in a specific setting, such as A and E or a GP surgery. Baseline measures regarding level of knowledge, reported understanding, frequency of holistic consideration for SCD in patient notes, patient experience of a service and frequency of use could be gathered. These could be repeated after a set period of time to assess for any change in attitude or patterns of use.

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Section D: Appendices of supporting material

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Appendix 1: Authors guides for submission to Journal of Health Psychology

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research relating to health and illness. The scope of the journal includes all areas of health psychology across the life span, ranging from experimental and clinical research on aetiology and the management of acute and chronic illness, responses to ill-health, screening and medical procedures, to research on health behaviour and psychological aspects of prevention. Research carried out at the individual, group and community levels is welcome, and submissions concerning clinical applications and interventions are particularly encouraged.

The types of paper invited are:

- papers reporting original empirical investigations;
- theoretical papers which may be analyses or commentaries on established theories in health psychology, or presentations of theoretical innovations;
- review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology; and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

1. Circulation

The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length

Papers should normally be no more than 5000 words (excluding the abstract, reference list, tables and figures), although the Editor retains discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

- the content of the paper falls within the scope of the Journal
- the methods and/or sample size are appropriate for the questions being addressed
- research with student populations is appropriately justified
- the word count is within the stated limit for the Journal (i.e. 5000 words)

4. Submission and reviewing

All manuscripts must be submitted via [Editorial Manager](#). You may like to use the [Submission Checklist](#) to help you prepare your manuscript. The Journal operates a policy of anonymous peer review. Authors must suggest three reviewers when submitting their manuscript, who may or may not be approached by the Associate Editor dealing with the paper. Before submitting, please read the [terms and conditions of submission](#) and the [declaration of competing interests](#).

APPENDIX

5. Manuscript requirements

- Contributions must be typed in double spacing with wide margins. All sheets must be numbered.
- Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. A template can be downloaded from [here](#).
- Statement of Contribution: All authors are required to provide a clear summary of ‘what is already known on this subject?’ and ‘what does this study add?’. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.
- Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript with their approximate locations indicated in the text.
- Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi.
- For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions.
- For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles.
- SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.
- In normal circumstances, effect size should be incorporated.
- Authors are requested to avoid the use of sexist language.
- Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the [APA Publication Manual](#) published by the American Psychological Association.
- Manuscripts describing clinical trials are encouraged to submit in accordance with the [CONSORT statement](#) on reporting randomised controlled trials.

6. Supporting information

Supporting Information can be a useful way for an author to include important but ancillary information with the online version of an article. Examples of Supporting Information include appendices, additional tables, data sets, figures, movie files, audio clips, and other related nonessential multimedia files. Supporting Information should be cited within the article text, and a descriptive legend should be included. Please indicate clearly on submission which material is for online only publication. It is published as supplied by the author, and a proof is not made available prior to publication; for these reasons, authors should provide any Supporting Information in the desired final format.

For further information on recommended file types and requirements for submission, please visit the [Supporting Information page](#) on Author Services.

7. OnlineOpen

APPENDIX

OnlineOpen is available to authors of primary research articles who wish to make their article available to non-subscribers on publication, or whose funding agency requires grantees to archive the final version of their article. With OnlineOpen, the author, the author's funding agency, or the author's institution pays a fee to ensure that the article is made available to non-subscribers upon publication via Wiley Online Library, as well as deposited in the funding agency's preferred archive. A full list of [terms and conditions](#) is available on Wiley Online Library.

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8. Author Services

Author Services enables authors to track their article – once it has been accepted – through the production process to publication online and in print. Authors can check the status of their articles online and choose to receive automated e-mails at key stages of production. The author will receive an e-mail with a unique link that enables them to register and have their article automatically added to the system. Please ensure that a complete e-mail address is provided when submitting the manuscript. Visit [Author Services](#) for more details on online production tracking and for a wealth of resources including FAQs and tips on article preparation, submission and more.

9. Copyright and licences

If your paper is accepted, the author identified as the formal corresponding author for the paper will receive an email prompting them to login into Author Services, where via the Wiley Author Licensing Service (WALS) they will be able to complete the licence agreement on behalf of all authors on the paper.

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10. Colour illustrations

Colour illustrations can be accepted for publication online. These would be reproduced in greyscale in the print version. If authors would like these figures to be reproduced in colour in print at their expense they should request this by completing a [Colour Work Agreement](#) form upon acceptance of the paper.

11. Pre-submission English-language editing

Authors for whom English is a second language may choose to have their manuscript professionally edited before submission to improve the English. A list of independent suppliers of editing services can be found in [Author Services](#). All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

12. The Later Stages

APPENDIX

The corresponding author will receive an email alert containing a link to a web site. A working e-mail address must therefore be provided for the corresponding author. The proof can be downloaded as a PDF (portable document format) file from this site. Acrobat Reader will be required in order to read this file. This software can be downloaded (free of charge) from [Adobe's web site](#). This will enable the file to be opened, read on screen and annotated direct in the PDF. Corrections can also be supplied by hard copy if preferred. Further instructions will be sent with the proof. Hard copy proofs will be posted if no e-mail address is available. Excessive changes made by the author in the proofs, excluding typesetting errors, will be charged separately.

13. Early View

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Appendix 2: Article summary table

paper	Diagnosis	Purpose		Sample Characteristics	Themes	Conclusion
Anderson & Asnani (2013).	SCD	To highlight Jamaican SCD patient voices regarding coping strategies	thematic analysis - individual	N=30 Age: 32 (18-46) Race/ethnicity: 100% Jamaican Gender: M - 15, F - 15 illness severity: SC - 26, SS - 3, thalassemia - 1	Loss; Control (controlling own responses to SCD; Controlling other's responses to SCD; Controlling the physical manifestations of SCD)	Faced with important losses, respondents used cognitive and behavioural coping strategies to re-establish control over their response to SCD, others' responses to SCD, and SCD's physical manifestations. Although the adaptive nature of some of these strategies is debatable, many facilitate management of the illness. Health care practitioners should encourage positive coping strategies and have non-judgmental discussions with patients about (potentially) negative ones. They should also share information with no specialist doctors and nurses to reduce stigmatization around the illness. More exploration of this under researched topic is needed.
Chapman (2002)	CF	Present empirical data on subjective levels of health and quality of life for individuals with early- or late- onset genetic conditions (Cystic fibrosis & Huntington Disease)	IPA - individual	N= 24 (12 CF, 12 HD) Age: CF 26;19-48; HD 48, 26-62 Race/ethnicity: NR Gender: CF 6 F, 6 M; HD 7 F, 5 M Illness severity: not given. CF, 1/12 was a parent; HD 11/12 were	"What is a life worth living?" (How do respondents view their quality of life?; Prenatal testing and choices; future implications)	Different emphases in judgements and definitions of QoL between HD and CF. Also emphasizes a convergence between CF & HD in terms of overall views of QoL confirming that people living with genetic conditions should form part of wider bioethical debates arising from such advances.

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				parents		
Caird et al (2011)	SCD	To investigate the lived experience of adults over 30 living with SCD and sought to develop a model understanding participants ability to function.	Grounded Theory - individual & focus group	<p>N= 15 Age: 17-68 Race/ethnicity: Black African-8; Black-Caribbean-3; British-Asian-1; Black-South American-1; NS-2 Gender: M-4; F-11 Illness severity: SS-9; SC-3, Beta-thal-1, NR-24.</p>	On-going suffering (reduction control; physical suffering; emotional suffering); Social capital; Changing relationship to SCD (Denial; Motivation for change; Moving to acceptance); Building resilience (creating meaning out of SCD; identity development; positive coping); Appreciating life	The study demonstrates the considerable resilience of people over 30 with SCD, providing a positive and hopeful model
Strickland et al (2001)	SCD	assessed the effectiveness of using focus groups to obtain info about the characteristics of pain and QoL in adults with SCD and their families	content themes - focus group (patient & family)	<p>N= 21 Age: 25-46 Race/ethnicity: NR Gender: M-3; F-7; family-11 who lived with SCD pt. Illness severity: low to moderate-high rates of pain as determined by mean painful</p>	Experiences of persons with SCD (self-care techniques and coping with disabling pain; Emotional responses of persons with SCD (Anger and Hostility; Depression; Disenfranchisement; Death Anxiety and Fatalism); Beliefs by others of drug dependency in persons with SCD; Use of Religion for Coping) Family Members' Perceptions (emotional responses of family; the impact of SCD on Family	SCD has profound consequences for QoL for individual and family. Health care providers could use info gained through this study (emotional, physiological and social effects) to positively influence the care of adults with sickle cell

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				episodes in previous 2 year: low 2 or less; mod to high 3+.	Relationships; Family's Concern about others' perception of the person with SCD)	
Thomas & Taylor (2002).	SCD	To gain understanding of the psychosocial impact of SCD on life. Determine how these experiences be conceptualized in terms of QoL	Thematic Analysis - focus group	N= 17 Age: 25.7 (15-35) Race/ethnicity: 49% West African; 51% African-Caribbean Gender: M-7; F-10 Illness severity: SS and 3 or more hospital admissions in previous yr.-mean 6 (3-18).	Growing up with SCD; Education; Impact of the unremitting nature of the disease; Employment; Effects on relationships; Hospitalization.	SCD carries a huge psychosocial burden impacting on physical, psychological, social and occupational well-being as well as levels of independence and environment. These aspects of life are equivalent to the core domains of the multi-dimensional WHOQOL and consequently we have argued that SCD undermines quality of life in important ways.

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<p>Huyard (2008)</p>	<p>CF</p>	<p>To describe illness-related learning in persons affected by a genetic disorder with early onset (not clearly defined)</p>	<p>Grounded Theory - individual</p>	<p>N= 11 Age: 5 less than 30, 4 in 30s, 2 older than 45 Race/ethnicity: French Gender: W-6, M-5 Illness severity: Diagnosis range from 0-30 yrs. 3 pts. Waiting lung transplant; 2 pts. recently started intravenous antibiotics. 5 pts. have siblings with CF, 2 had lost a sibling to CF.</p>	<p>Becoming aware of one's condition: a specific illness career; learning, putting to the test, judging: the critical internalization process; being a clinically minded but not health-conscious patient: the cases of clinical trials and alternative medicine.</p>	<p>The diagnosis of genetic disease does not imply that the pt. considers themselves to be ill. Patients gain knowledge about their illness that helps them undertake or avoid certain actions to achieve goals they consider important.'</p>
<p>Lowton (2004)</p>	<p>CF</p>	<p>explore how decisions of disclosure of CF are made in adulthood</p>	<p>qualitative - individual</p>	<p>N= 31 Age: 18-40 F mean 29, M mean 32 Race/ethnicity: NR Gender: W-17, M-32 Illness severity: Diagnosis age: 1-22 (only 2 older than 7</p>	<p>Low-risk situations: The casual encounter; medium-risk situations: friendships; high-risk situations: potential partners and prospective employers</p>	<p>A multiplicity of factors including perceived social support and disease progression, are seen to influence adults' decisions to disclose their disease.</p>

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				(14&22)). 9 had children or had attempted to have children. 19 working or studying.		
Addis et al (2007)	SCD	To investigate the physical, social and psychological impact of priapism on adult males with sickle cell disorder	Grounded Theory - individual	N= 6 Age: NR Race/ethnicity: NR Gender: all men illness severity: NR	First occurrence of Priapism; Pain associated with priapism; Precipitants; emotional consequences of priapism; self-management; experience of hospital; impact on work and social life; impact on sexual relations; erectile dysfunction; disclosing priapism	The study powerfully highlights the social and psychological significance of priapism.
Booker et al (2006)	SCD	To investigate the nature of the recurrent 'crisis' pain.	Thematic Analysis - focus group	N= 10 Age: 32 (22-53) Race/ethnicity: 8 - afro Caribbean; 1 - African; 1 - Portuguese Gender: F- 4, M-6 illness severity: at least 1 self-reported crisis in preceding 12 months	Fear (Death; uselessness; helplessness; loss of virility; isolation; worsening ill-health); Coping (nature of the pain and strategies; faith; medications; anger); Interacting with others (telling others; support from others; social isolation; communication with healthcare professionals	Feelings of isolation may drive maladaptive coping strategies and manifest in anger, aggression and active avoidance of service use. Suggested service improvements include the active targeting of isolated
Paterson et al (1999)	D	An attempt to explicate the structures and processes of	Grounded Theory - interview, think aloud	N= 22 Age: 43.3 (24-81) Race/ethnicity:	The experience of transformation; Challenges leading to transformation; Differentiation of self and body.	Transformation was the results of a conscious decision to identify and interpret a challenge and, in doing so, create a new relationship with the illness

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		transformation for individuals with Type I diabetes.	recordings & focus group	Living in British Columbia, Candia. Caucasian Gender: F-11, M-8 Illness severity: diagnosed for 15-41 yrs.; all but 4 had high school or postsecondary education; 8 participants had diabetes related complications.		and with those who provide healthcare.
Tierney et al (2010)	CF	to explore the struggle of cystic fibrosis patients to produce sputum samples	framework analysis - individual	N= 18 Age: 25.9 Race/ethnicity: NR Gender: M - 44% illness severity: median age diagnosis - 11 (0-19)	Sputum coming to define interviewees (being seen as dirty; exposing difference; a reminder of having CF; sputum providers	Patients' difficulties with expectorating sputum were multifaceted and included psychosocial and physical factors, for some people, a request for sputum can cause significant discomfort.

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Balfe (2009)	D	To examine tension regarding engaging (or not) in diabetes self-care practices in university students and what being normal means for these students.	thematically analysed - individual & diary	N= 17 Age: 18-25 Race/ethnicity: NR. All studying at university in the Midlands, UK. Gender: illness 6 - M, 11- F severity: NR	The student body; the toned body; the healthy body; Difficulties in Actualizing Body Projects; Balancing projects.	Normalcy for these students is constructed and maintained through the body by developing “body projects.” Each body projects is intended to produce a particular kind of normal embodied identity that is unaffected by diabetes. Unfortunately, body projects often undermine each other which risky for students’ diabetes control. Students have to reach a balance between their different body projects.
Hernandez et al (1999).	D	To identify the cues, sensations and circumstances that people with Diabetes and their families associate with symptoms the types of strategies that people with diabetes use to tune in to body cues and sensations	identify symptoms, cues, circumstances & strategies - focus group	N= 4 (& 4 family members) Age: 2 in 30s, 1 in 40s, 1 in 60s Race/ethnicity: NR Gender: all Female- 4 illness severity: type 1 diabetes at least 15 years	Body cues; contextual circumstances; self-awareness strategies (constant self-awareness/tuning in; body checks; knowing one's body, personal responses, body patterns and norms; mental tests to detect a low; doing a blood glucose test; collaborative alliance)	Personal cues of different levels of glycaemia may be different than classic textbook symptoms therefore people should be encouraged to identify their own body cues. Even people with hyperglycaemia unawareness may recognise cues that replace the automatic ones they have lost.

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<p>Weisberg et al (2013)</p>	<p>SCD</p>	<p>To characterize the subjective experience of extremely high hospital use in patients with sickle cell disease, and generate hypotheses about the antecedents and consequences of this phenomenon.</p>	<p>narrative analysis - individual</p>	<p>N= 8 Age: 29 (22-37) Race/ethnicity: NR Gender: M-3, F-5 Illness severity: SS-5; SC-1; beta thal - 2. Ave no of hospital days per year - 152 (94-263)</p>	<p>pain and opioid medication use; interpersonal relationships; person development</p>	<p>Results suggested a systematic, self-reinforcing process of isolation from mainstream society, support structures, and caregivers, based on increasing hospitalization, growing dependency on opioid medications, as well as missed developmental milestones.</p>
<p>Rasmussen et al (2007)</p>	<p>D</p>	<p>Explore and develop a substantive theory to explain how young women with type 1 diabetes managed their lives when facing turning points and undergoing transitions.</p>	<p>grounded theory - individual</p>	<p>N= 20 Age: 28 (20-30) Race/ethnicity: NR Victoria, Australia Gender: all F - 20 Illness severity: diabetes since 5-11 yrs.</p>	<p>Being in the grip of blood glucose levels (BGLs) (the impact of being susceptible to fluctuating BGLs; the responses of other people to the individual woman's diabetes; the impact of the individual women's diabetes on other people's lives; Creating stability; Consequences of being in the grip of Blood Glucose Levels</p>	<p>The women used a basic social process to overcome the basic social problem by creating stability, which involved using three interconnected sub processes: forming meaningful relationships, enhancing attentiveness to blood glucose levels, and putting things in perspective.</p>

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<p>Ingadottir, & Halldorsdottir, (2008).</p>	<p>D</p>	<p>To explore the essential structure of mastering diabetes from the patient's viewpoint, emphasising lived experience of adhering to and not adhering to a complex treatment regimen.</p>	<p>Vancouver school - individual</p>	<p>N=11 Age: 48 (28-84) Race/ethnicity: NR Gender: F - 6, M-5 Illness severity: living with diabetes 5-60 years (29) 10 type 1, 1 type 2 - 2 diagnosed as adult. 5 had various diabetes complications</p>	<p>Mastering diabetes: disciplining the "dog" (knowledge, understanding and experience; Fighting fear in the search for safety; dealing with distress; freedom or constraint? The question of autonomy)</p>	<p>It revealed how people with diabetes exist in the world and how they strive to gain meaning from the experience of being diagnosed with a disease, of being ill, and of being embodied in an ever-unpredictable and changing body.</p>
<p>Maxwell et al (1995)</p>	<p>SCD</p>	<p>To investigate how sociocultural factors influence management of pain from sickle cell disease by comparing the experiences of those who usually manage their pain at home with those who are more</p>	<p>Qualitative - focus groups (40) and interviews (9) both (6)</p>	<p>N=57 Age: 34 (20-60) 2 missing Race/ethnicity: west African - 29 (51%), afro-caribbean-26 (46%), other African - 2 (4%) Gender: F - 32 (56%), M - 25 (44%) illness severity: 44 - SS or Beta thal, 9- SC, 4-</p>	<p>Experience of hospital care (mistrust; stigmatisation; control; neglect); Strategies for management of pain and treatment seeking ; Strategies of patients managing pain at home (assertiveness; self-education; resistance); Strategies of patients frequently admitted to hospital (developing relationships; aggression; passivity; use of multiple hospitals).</p>	<p>The current organisation and delivery of management of pain for sickle cell crisis discourage self-reliance and encourage hospital dependence. Models of care should recognise the chronic nature of sickle cell disorders and prioritise patients' involvement in their care.</p>

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		frequently admitted to hospital for management of their pain.		unknown		
Higham et al (2013)	CF	To explore the hopes and fears of young adults with cystic fibrosis	Grounded Theory - individual	<p>N=15 Age: 24 (18-29) Race/ethnicity: NR Gender: f=8, m=7 illness severity: diagnosed in childhood, had Delta F508 mutation and FEV1<40%</p>	Living with unpredictable health and the fear of death and dying; Hopes for normality; Hopes for a normal relationship and/or marriage; Hopes to become a parent; Hopes for a normal work life.	Participants feared the unpredictable nature of CF and the suffering they believed they would endure before premature death. Despite their fears participants hoped for key aspects of a 'normal' life.

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Appendix 3: Model of theme development

Review theme	sub theme	No.	study	diagnosis	theme label in text						
The impact on everyday life	Unpredictability and day-to-day life	12	Addis et al. (2007)	SCD	Impact on work and social life						
		2 CF	Anderson & Asnani (2013).	SCD	Loss	Pain associated with Priapism	first experience of priapism				
		8 SCD	Balfe (2009)	TI DM	The student body	Difficulties in actualizing body projects	Balancing projects				
		2 TI DM	Booker et al. (2006)	SCD	Interacting with others	Coping					
			Caird et al. (2011)	SCD	On-going suffering						
			Chapman (2002).	CF	How do respondents view their quality of life	Prenatal testing and choices	Future implications.				
			Higham (2013)	CF	Living with unpredictable health and the fear of death and dying						
			Maxwell et al. (1995)	SCD	Experience of hospital care						
			Strickland et al. (2001)	SCD	self-care techniques and coping with disabling pain						
			Rasmussen et al. (2007)	TI DM	Impact of being susceptible to fluctuating BGLs	Creating stability: A basic social process					

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			Thomas & Taylor (2002).	SCD	Growing up with SCD	Education	Impact of the unremitting nature of the disease			
			Weisberg et al. (2013)	SCD	Personal development					
The impact on everyday life	Loss	3 0 CF	Anderson & Asnani (2013).	SCD	Loss					
		3 SCD	Booker et al. (2006)	SCD	Fear (loss of virility)					
		0 T1D M	Caird et al. (2011)	SCD	On-going suffering					
The impact on everyday life	Emotional impact	12 4 CF	Addis et al. (2007)	SCD	Emotional consequences of priapism					
		6 SCD	Booker et al. (2006)	SCD	Fear	Coping				
		2 T1D M	Caird et al. (2011)	SCD	On-going suffering	building resilience	appreciating life			
			Chapman (2002).	CF	How do respondents view their quality of life	Future implications				
			Higham (2013)	CF	Living with unpredictable health and the fear of death and dying	Hopes for normality	Hopes for a normal relationship and/or marriage	Hopes to become a parent	Hopes for a normal work life	

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			Ingadottir, & Halldorsdottir, (2008).	TIDM	Fighting fear in the search for safety					
			Lowton (2004)	CF	Low-risk situations: The casual encounter	medium-risk situations: friendships	high-risk situations: potential partners and prospective employers			
			Maxwell et al. (1995)	SCD	Experience of hospital care	Strategies for management of pain and treatment seeking				
			Rasmussen et al. (2007)	TIDM	Impact of being susceptible to fluctuating BGLs					
			Strickland et al. (2001)	SCD	Emotional responses of persons with SCD	Anger and hostility	Depression	Death anxiety and fatalism		
			Tierney et al. (2010)	CF	being seen as dirty	exposing difference	A reminder of having CF			
			Weisberg et al. (2013)	SCD	Personal development					
The impact on everyday	Experience of symptoms	5 2 CF	Addie at al. (2007)	SCD	First occurrence of Priapism	Pain associated with	emotional consequences of priapism	impact on work and social life	impact on sexual	erectile dysfunction

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life		3 SCD	Booker et al. (2006)	SCD	Fear	priapism	Coping	Interacting with others		relations	
		0 T1D M	Lowton (2004)	CF	Low-risk situations: The casual encounter	medium-risk situations: friendships	high-risk situations: potential partners and prospective employers				
			Tierney et al. (2010)	CF	being seen as dirty	exposing difference	a reminder of having CF	sputum providers			
			Weisberg et al. (2013)	SCD	pain and opioid medication use	interpersonal relationships	Personal development				
Relationships	Family and friends	9	Addis et al. (2007)	SCD	disclosing priapism	first occurrence of Priapism					
		2 CF	Balfe (2009)	T1DM	The student body	The tones body	The healthy body				
		5 SCD	Booker et al. (2006)	SCD	Interacting with others						
		2 T1D M	Caird et al. (2011)	SCD	Social capital						
			Lowton (2004)	CF	medium-risk situations: friendships						
			Rasmussen et al. (2007)	T1DM	Responses of other people to the individual woman's diabetes						
			Strickland et al. (2001)	SCD	Disenfranchisement	Family members'					

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						perceptions					
			Thomas & Taylor (2002).	SCD	Relationships						
			Tierney et al. (2010)	CF	exposing difference						
Relationships	Intimate relationships	6	Addis et al. (2007)	SCD	impact on sexual relationships	erectile dysfunction	disclosing priapism				
		3 CF									
		2	Chapman (2002).	CF	Prenatal testing and choices	Future implications					
		SCD									
		1	Higham (2013)	CF	Hopes for a normal relationship and/or marriage	Hopes to become a parent					
		T1D									
		M									
			Lowton (2004)	CF	high-risk situations: potential partners and prospective employers						
			Rasmussen et al. (2007)	T1DM	Responses of other people to the individual woman's diabetes	impact of the individual woman's diabetes on other people's lives	Creating stability: A basic social process				
			Thomas & Taylor	SCD	Relationships						

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			(2002).							
			Higham (2013)	CF	Hopes for a normal work life					
			Lowton (2004)	CF	high-risk situations: potential partners and prospective employers					
			Rasmussen et al. (2007)	T1DM	Responses of other people to the individual woman's diabetes					
			Thomas & Taylor (2002).	SCD	Employment					
Relationships	Relationships with medical staff	6	Booker et al. (2006)	SCD	Interacting with others	Coping				
		1 CF								
		5 SCD	Strickland et al. (2001)	SCD	Beliefs by others of drug dependency in persons with sickle cell disease	Family members' perceptions				
		0 T1DM	Thomas & Taylor (2002).	SCD	Hospitalization					
			Tierney et al. (2010)	CF	sputum providers					

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			Weisberg et al. (2013)	SCD	Interpersonal relationships					
			Maxwell et al. (1995)	SCD	Experience of hospital care	Strategies of patients managing pain at home	Strategies of patients frequently admitted to hospital			
Coping	Regaining control	6	Anderson & Asnani (2013).	SCD	Controlling own response to SCD	Controlling others' responses to SCD	Controlling the physical manifestations of SCD			
		0 CF								
		2	Balfe (2009)	T1DM	The student body	Difficulties in actualizing body projects	Balancing projects			
		4								
		T1D	Ingadottir, & Halldorsdottir, (2008).	T1DM	Mastering diabetes: Disciplining the "dog"					
		M	Maxwell et al. (1995)	SCD	Experience of hospital care	Strategies of patients managing pain at home	Strategies of patients frequently admitted to hospital			
			Paterson et al. (1999)	T1DM	The experience of transformation					
	Rasmussen et al. (2007)	T1DM	Creating stability: A basic social process							

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Coping	Self-care	11	Addis et al. (2007)	SCD	self-management	Precipitants				
		2 CF	Anderson & Asnani (2013).	SCD	Controlling the physical manifestations of SCD					
		5 SCD								
		4 T1DM	Hernandez et al. (1999).	T1DM	Body cues	contextual circumstances	self-awareness strategies			
			Huyard (2008)	CF	Becoming aware of one's condition: a specific illness career	learning, putting to the test, judging: the critical internalization process	being a clinically minded but not health-conscious patient: the cases of clinical trials and alternative medicine			
			Ingadottir, & Halldorsdottir, (2008).	T1DM	Mastering diabetes: Disciplining the "dog"	Knowledge, understanding and experience	Dealing with conflicting desires	Freedom or constraint ? The question of autonomy		
			Maxwell et al. (1995)	SCD	Strategies of patients managing pain at home					

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			Paterson et al. (1999)	T1DM	Challenges leading to transformation					
			Strickland et al. (2001)	SCD	self-care techniques and coping with disabling Pain					
			Rasmussen et al. (2007)	T1DM	Creating stability: A basic social process					
			Tierney et al. (2010)	CF	sputum providers					
			Weisberg et al. (2013)	SCD	Pain and opioid medication use					
Coping	Religious beliefs and meaning making	4	Anderson & Asnani (2013).	SCD	Controlling own response to SCD					
		0 CF								
		4 SCD	Booker et al. (2006)	SCD	Coping					
		0 T1DM	Caird et al. (2011)	SCD	Building resilience					
			Strickland et al. (2001)	SCD	Use of religion for coping					
coping	Identity		Balfe (2009)	T1DM	The student body	Difficulties in actualizing body projects	Balancing projects			
		4	Caird et al. (2011)	SCD	Building resilience					

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		0 CF	Paterson et al. (1999)	T1DM	Differentiation of self and body					
		1 SCD	Rasmussen et al. (2007)	T1DM	Creating stability: A basic social process					
		3 T1DM								
Acceptance		4	Anderson & Asnani (2013).	SCD	Controlling own response to SCD					
		1 CF								
		2 SCD	Caird et al (2011)	SCD	Ongoing suffering	Changing relationship to SCD	appreciating life	Building resilience		
		1 T1DM	Huyard (2008)	CF	Becoming aware of one's condition: a specific illness career	learning, putting to the test, judging: the critical internalization process	being a clinically minded but not health-conscious patient: the cases of clinical trials and alternative medicine			
			Ingadottir, & Halldorsdottir, (2008).	T1DM	Mastering diabetes: Disciplining the "dog"	Dealing with conflicting desires	Freedom or constraint? The question of autonomy			

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Appendix 4: Critiquing and quality assurance information (Elliott, Fischer & Rennie, 1999).

Paper	Diagnosis	Owning one's perspective	Situating the sample	Analytic auditing	Triangulation	Testimonial validity	Non-participants check
Addis et al (2007)	SCD	Yes	No	Yes	No	No	No
Anderson & Asnani (2013).	SCD	No	No	Yes	No	No	No
Balfe (2009)	D	No	No	No	Yes	No	No
Booker et al (2006)	SCD	No	No	Yes	focus group	Yes	No
Caird et al (2011)	SCD	No	No	Yes	Yes	No	No
Chapman (2002)	CF	No	No	No	No	No	No
Hernandez et al (1999).	D	No	No	Yes	focus group	Yes	Yes
Higham et al (2013)	CF	No	No	Yes	No	No	
Huyard (2008)	CF	No	No	No	No	No	No
Ingadottirr, & Halldorsdottir, (2008).	D	No	No	No	No	Yes	Yes
Lowton (2004)	CF	No	No	No	No	No	No
Maxwell et al (1995)	SCD	No	No	No	Yes	No	No
Paterson et al (1999)	D	No	No	No	Yes	Yes	No
Rasmussen et al (2007)	D	No	No	No	No	Yes	No
Strickland et al (2001)	SCD	No	No	No	Yes	No	No
Thomas & Taylor (2002).	SCD	No	No	Yes	focus group	Yes	No
Tierney et al (2010)	CF	No	No	Yes	No	No	No
Weisberg et al (2013)	SCD	No	No	Yes	No	Yes	No

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Appendix 5: Patient information sheet

Experiences of Sickle Cell Disorder Related Pain

Hello. My name is Beth Coleman and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study looking at the experiences of sickle cell disorder related pain. Before you decide if you want to take part it is important that you understand why the research is being done and what it would involve for you. You may also wish to talk to others about the study.

What is the purpose of the study?

This study is interested in how people with SCD experience and describe pain, both acute crisis pain and chronic day-to-day pain. It aims to talk to a number of individuals with SCD in order to collate their experiences to try and develop a better understanding about the nature of SCD related pain.

Why have I been invited?

You have been invited to join in a research project as you have SCD and attend a SCD support group who has expressed an interest in supporting the project.

Do I have to take part?

No, it is up to you if you join the study; involvement in the project is entirely voluntary. If you choose to take part, I will then ask you to sign a consent form. You are free to withdraw from the project at any time, without giving a reason. This would in no way affect the service you receive.

What will I have to do?

You will be asked to attend an informal discussion to talk about your experience of pain. This meeting should last no more than 90 minutes and can take place in your home, at the centre where your support group meets or, if possible, at another venue of your choosing. The interview could take place in one go, perhaps with a break in the middle, or in two 45 minute settings a week or two apart. Potentially the second interview could be via telephone if more convenient for you.

The discussion will involve a number of questions about your experience of SCD related pain, for example how do you talk about your pain? How do you understand your pain? What is your relationship with your pain, are you enemies, friends, family? Would you describe the pain differently to someone who does not have SCD? I would like to record this conversation so I can accurately represent what you say, I will ask for your consent to do this.

Will my taking part in the study be kept confidential?

Yes. We follow ethical and legal practice and all information will be handled in confidence. The information you provide will be kept strictly confidential. Any information about you which leaves the interview venue will have your name removed so that you cannot be recognised. The interviews will be stored anonymously on encrypted data drives during the period of the project and transcribed by the researcher. This will not be identifiable to you in any way other than to the researcher who will conduct the interviews. It will then be stored on a password protected CD in lockable cupboard for 10 years following the completion of the project after which it will be destroyed.

What are the possible disadvantages and risks of taking part?

The interviews are focused on your experiences of SCD related pain. You are free to talk about this as much or as little as you like however such discussions may include topics of a difficult or personal nature. The interviewer will be able to offer you support if this is the case. Other members of your support group or your NHS team may also be able to support you. Furthermore you could contact the Samaritans on 08457909090.

What are the possible benefits of taking part?

The information you provide will be collated with others who experience SCD related pain and used to try and develop a greater understanding about the nature SCD related pain. We then hope to develop a means of communicating this information to other health professionals to help them have a better understanding of SCD related pain.

Expenses and payments

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Travel expenses to and from the discussion will be covered and a £5 thank you voucher will be given to those who participate.

What will happen if I don't want to carry on with the study?

You have the right to withdraw from the project at any time. Your views, comments or withdrawal from the project will in no way affect the service you receive. If you withdraw from the study, we would like to use any information you have given us up to your withdrawal, but that is your decision, you can withdraw all of your information with no consequences.

What if there is a problem?

If you have a concern about any aspect of this study you could ask to speak to me and I will do my best to answer your questions. Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed.

Complaints

If you remain unhappy and wish to complain formally, you can do this by contacting Paul Camic, Professor of Psychology & Research Director at:

Department of Applied Psychology
Canterbury Christ Church University
Broomhill Road
Tunbridge Wells, Kent TN3 0TG.
Tel: 01892 507 773
Email: paul.camic@canterbury.ac.uk

Involvement of the General Practitioner/Family doctor (GP) and other health care professionals

Your GP does not need to be notified of your involvement with this project. It may however be useful for any health professionals linked to your SCD to know you are taking part so that they can support you if the need arises.

What will happen to the results of the research study?

The information you provide will form part of a wider project that is part of the Clinical Psychology Training Programme at Canterbury Christ Church University and will be written up as a piece of assessed work. Furthermore it is possible that the findings will be published in an academic journal. Anonymous quotes from your material may be used in published reports of the study.

Following the project the interviewer can provide you with information regarding the outcome in a format convenient to you such as a summary letter or a feedback meeting.

Who is organising the research?

This study is part of the clinical psychology doctoral course at Canterbury Christ Church University. The university is funding the research project.

Who has reviewed the study?

This research has been reviewed by the Salomons' Ethics panel at Canterbury Christ Church University to protect your interests. The study has been given a favourable opinion.

Further details and contact details

If you would like to speak to me and find out more about the study or have questions about it, you can leave me a message on a 24-hour voicemail phone line at 01892 507673. Please say that the message is for me, Beth Coleman, and leave a contact number so that I can get back to you. Alternatively you could speak to your SCD support group facilitator or a member of your NHS team who can contact me on your behalf.

Thank you

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Appendix 6: Participant consent form

Centre Number:

Study Number:

Participant Identification Number for this study:

Consent form

The Experience Sickle Cell Disorder Related Pain

Thank you for agreeing to take part in this research project. The project is looking at how people with SCD experience and describe pain, both acute crisis pain and chronic day-to-day pain. The purpose of this meeting will be to find out about your experience of SCD related pain.

The meeting is an informal discussion that should last no more than 90 minutes, either in an individual session or split across two 45 minute sittings. For the purpose of the research project I would like to record the interview. The information you give in this recording will be confidential and not identifiable to you. It will be stored confidentially on a password protected CD in lockable cupboard until the completion of the project.

The information you provide will be collated with others who have SCD and used for this research project to try and develop a better understanding about the nature of SCD related pain. The project will be written up as a piece of assessed work for the Clinical Psychology Training Programme at Canterbury Christ Church University. Anonymous quotes from your material may be used in published reports of the study. The data will not be used for any other purpose than those listed above.

You have the right to withdraw from the project at any time. Your views, comments or withdrawal from the project will in no way affect the service you receive.

I have read the above information and give my permission for the interview to be recorded and for the information I provide to be used for the purposes of the research project, an assessed piece of work for Canterbury Christ Church University including the use of anonymous quotes.

Please initial boxes below:

1. I confirm that I have read and understood the information sheet dated Nov 2012 (version 1) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being effected.
3. I give permission for the interview to be audio recorded and transcribed.
4. I agree to anonymous quotes from my interview may be used in published reports of the study finding.
5. I agree to take part in the above study.

Name of Participant.....
Signature..... Date...../...../.....

Name of Person taking consent.....
Signature..... Date...../...../.....

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Appendix 7 Interview schedule

1. Please can you tell me about the pain you are experiencing today?

Possible prompts: Is this a normal day – what different kinds of days do you have?

2. Do you have different types of pain? Describe these to me.

Possible prompts: What is the worse pain for you? What is the easiest pain for you?

3. What are the main differences between a good and bad pain day?

Possible prompts: how do you cope? Has this changed over time?

4. Do you have a name for your pain?

5. What does your pain look like, sound like?

6. How do you imagine the pain in your body? / What images come to mind when you talk about the pain and when you experience the pain?

Possible prompts: creative expressions or ideas to explain your pain? If your pain was like something in a natural world what would it be?

7. What is your relationship to the pain?

Possible prompts: Are you enemies, friends, family?

8. Can you describe how you talk about your pain with other people?

Possible prompts: friends, family, people who don't have SCD (would this be different?), in hospital?

9. What do you think your friends/family think about your pain experience?

10. What do you think health care professionals think about your pain experience?

Possible prompts: What is it like trying to explain pain in hospital?

11. How do you understand the pain? What does the pain mean to you?

12. Has your experience taught you anything?

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Appendix 8: Example transcript

This has been removed from the electronic copy.

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Appendix 9: Excerpts from reflective journal

Fri 26th October 2012 – submitted second MRP proposal

Bit of a stress getting it all together but as I have had to start all over again timing is so crucial and I am so behind.

Fri 2nd November – MRP Review

Really helpful, it was unclear if the panel could meet my deadline but they did all they could to help me. Got to make sure I have theory at the heart of the project, not my strength I know.

Friday 9th Nov – Ethics deadline

That was an even quicker turn around but hopefully there won't be any issues! Had to get it in today otherwise it wouldn't be reviewed by the panel until Jan!

28th November - Got ethics approval

13th December – pilot interview

Useful practice, strange to be a researcher instead of a clinician, felt being pulled into the clinician role a bit. Will have to keep a check on that.

Friday 4th Jan – Booked to attend sickle cell support group

Group got cancelled because of the snow so couldn't recruit. The cold is not good for people with SCD as it increasing sickling. Still I have one interview booked in with the facilitator and I can go next month.

Monday 14th Jan 2013 – 1st Interview Adebola

First interview today. Really struck by the amount of pain. Adebola was very articulate at describing it both verbally and with images. What a lot of pain! Was slightly annoyed to be told that I don't live in constant pain. One of the things he said was that people don't see the pain so don't know and make assumptions but that's exactly what he did. Was slightly uncomfortable about comparisons to other conditions like cancer, saying that when people suffer with cancer they know that they will go get treatment and won't be back whereas with SCD they will always be back. If only cancer was that straight forward to treat!

Fri Jan 25th – IPA workshop

Lecturer is amazing! IPA feels so manageable. Really useful 2-ish hours. Changing interview script based on her advice, making it more 'free' to let them do the talking. Slightly scary but based on first interview confident good stuff will come up.

Friday 1st Feb 2013 – Attended sickle cell support group

Wow that was a long group. There seems a lot of frustration about treatment from medical staff; people liked to tell their story in detail. Lots of ideas about doing something but not much action. Wonder why? Is there something about a victim role? Got 6 interviews booked over the next 3 weeks though!

Feb 2013 – contacting other support groups

Emailed lots of support groups across London, had some interest and some asked be to attend but was not able to because of personal commitments.

Monday 4th Feb – 2 interviews Benjy and Charlotte

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The contrast been Charlotte who is 24 and angry at her parents for having a child without knowing they could pass on SCD was and Benjy (32) who has come to accept and manage life as best he can is stark. Is age a factor? Does perception change with age? Or does acceptance come with age? Or is life circumstances? Benjy seems relatively well off? Son of a brief love affair (Nigerian culture second wife) while Charlotte grew up in London – they seem to be worlds apart.

Tue 5th Feb – interview Deborah at her house

I found this interview difficult because of this lady's presumptions; she assumed I do not have live in pain too. I agree it's not to the same degree but I live everyday in pain. How did I end up doing a project on chronic pain? Partly because of my first project fell through and this being almost ready to go but also my pain wasn't as flared up when I took up the project. It's flared up much worse since then. Must talk to my supervisor about this.

Tue 5th Feb – emailed to book a supervision session given my link with chronic pain. Booked for 1st March, unfortunally after all my interviews are scheduled but timing did not allow for an earlier meeting.

Thurs 7th Feb – interview booked with H

I would have had to cancel interview with J due to my hip scan but she cancelled first. My own pain needs were in the forefront of my mind.

Monday 11th Feb – 2 interviews Glen and Edith

Glen cancelled but met with Edith, she has twin boys! (SCD trait) Also although SS her symptoms do not seem as server as Adebola or Benjy. Maybe that is just in the reporting but it highlights the massive range of experiences in SCD even with the same diagnosis. Felt myself being pulled into therapist roll again with Edith. It's not happened since the pilot interview, wonder why that was?

Thurs 14th Feb – Interview Femi

Femi was quite reserved, and it was difficult to get info from her. Her English was not always great so at times it was difficult to understand her. She has a daughter with SCD so hearing about the perspective of being a mother was interesting too.

Monday 18th Feb – 2 interviews Glen and H

Glen came this time but H cancelled again.

Glen made me really think about psychological pain – e.g. thinking about who to have children with due to the risk of passing on SCD, not knowing if you will be around to see your child grow up, not being able to book holidays or fun things as you just don't know if you'll be well enough. Tough. Think he helped me appreciate how many areas of life it affects.

1st March – supervision

Talked about my experience of chronic pain, how at times I have had to put my own pain needs first and how participant assumptions that I am not in pain have been difficult.

Feb/March 2013 – transcribing

Transcribing takes a long time, but I feel a lot more familiar with the data and am already thinking about similarities, differences and themes.

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Feb/March

Not heard back from other support groups. Also two possible participants, H and I kept cancelling and now I cannot get in touch with them. I'm taking that as them deciding not to be a part of the project. I have 7 interviews that is one more than Smith recommends so I am happy with that. And there is a lot of good stuff in the interviews I'm transcribing.

Monday 22nd April – started pain placement

I have 1 day a week with a community pain team. Interesting how this came about as my OA supervision arranged it 'in-house' without Salomon's knowledge. Think I'm going to learn a lot about my own pain here.

Thursday 25th April – physio

I have now officially moved into the realm of chronic pain. I sort of knew this already it had't be classified so by medical staff. Physio was somewhat reluctant to talk about it, like it's a dirty condition somehow.

Also been thinking about SCD and the chronic and acute pain within one disorder and well I get both, I have an everyday pain that is always there, sometimes worse than other times, and then I get an acute flare up that is much more painful and debilitating. Again not to the same extremes but is it that different?

May 2013 – Section A

I'm currently focusing less on my analysis and taking some time to work on Section A. as taking a while to focus on this while my friends and family transcribe for me. But reading about the different chronic conditions (CF and T1DM) has raised more questions about illness in SCD and how different it really is. Yes so SCD main factor is pain but CF is lung related, breathing, coughing etc which also has a chronic component. Also the SCD literature is the only one that mentions the relationship with medical staff being difficult. Perhaps it is because pain is not visible that they get branded 'druggies' or maybe there is/has been a culture clash/racism? Black people do tend to be more direct and I know from my friends that sometimes this can come across as cold, harsh, confrontational, aggressive almost so maybe that has been a historical issue. Or is it something to do with who gets affected and how pushy their parents are in relation to research? Traditionally it has been white professionals that push for research; the SCD population would not be included in this.

June 2013

On-going analysis. Starting with Adebola

Feeling rather frustrated about the assumptions about other conditions experiencing pain! Maybe because I have chronic pain and nothing takes that away and assumptions were made about my pain experience. But also from my day at the Pain service. Other people have experiences of not being believed about pain too, SCD is not unique in that yet I get a sense that they want special treatment, to be privileged and although the pain experience sounds hideous I'm not sure why they should by-pass all the questions in hospital. Do other conditions who frequent hospital regularly get this too? Why is SCD so special?

18th June - My landlord is a GP and when I mentioned my project to him he mentioned the medication problem and how he as a doctor has to be careful because it is such strong stuff they are asking for.

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21st June - Is pain quantifiable? Is it measurable? The same injury can cause pain, but the experience is different. Nerve pain, the scientific chemical stuff etc. is quantitative but how you experience it is qualitative E.g. stubbing your toe. Two people would feel pain but the experience would be how you deal with it different. Same with SCD; even those with SS don't have the same amount of crisis in the same way with the same complications. But for those that do if you, they know what SCD pain is, how you deal with it is the difference. So it can be quantified in the sense that if you understand what that pain means, you know what that is to them but how they deal with that is on top of that – but the basic thing of what the pain feels like is the same in some respects.

22 June model of themes is coming together slowly. Attempted to use NVivo to analysis but it does not allow a way to include the notes stage of the analysis. I have tried a few ways but it does not seem possible. This is very frustrating as doing it my hand will be extra time consuming, especially when I come to gather quotes together at the end. However this is not avoidable so I am not going to spend any more time seeing if there is a way. Possible overarching themes – experience of pain

1st July

Yes the pain is really that bad! Getting a real sense about SCD as a hidden illness- not wanting to be seen as different yet feeling misunderstood and wanting others to appreciate just how bad it is!

10th July

Finding quotes to write up would be so much easier with Nvivo! Wish it had worked with IPA. I think there are several more themes that could be drawn out but there is not enough time or space to incorporate everything. I am having to use my interpretation to decide which are most valid, I hope I do the participants experiences justice.

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Appendix 10: Letter of full ethical approval from the Chair of the Salomons Ethics Panel

This has been removed from the electric copy.

Appendix 11: Additional illustrative quotes

Overarching theme 1: Experiencing unimaginable pain

Indescribable pain	
Worse than the worse pain imaginable	<p>Adebola: Worse pain is ouu it would be off the scale. Adebola: I've been in an induced coma and still been in pain. Adebola: it doesn't have a limit and y'know it doesn't have a limit so however bad you feel it gets worse.</p> <p>Benjy: Oh-h God the worst pain I've experienced.. . Would I mean in hospital they would say one to ten where would you be on that level mine would be eleven twelve but they wouldn't I mean telling someone a number actually try and explain pain to them is so deadly different I mean I used to scream my head out</p> <p>Charlotte: I think pain, sickle cell pain, is the worse pain ever imaginable. It's like just someone please come and kill me, yeah, that bad, really bad yeah.</p> <p>Deborah: I was in a lot of pain even with my caesarean section because they did not. And I will repeat that they did not medicate me enough. Erm.. the epidural I got did not work, and so it probably numbed the section where. They opened me up just probably that bit but the rest of the child bearing pain I felt every bit of it. I knew when they moved my son's arm I knew when they moved his leg, I knew when they moved him out of position, I knew it was like going through child birth normal child birth and I can tell you sickle cell pain on a scale of one to ten is worse..</p> <p>Edith it's like being punished</p>
Use of analogy	<p>Adebola: It would be fracture, broken bones, erm severe infection all rolled into one Adebola: I mean at its best where it's the perfect day would be like background noise. So it would be mmm an irritating noise you would hear in the background. Adebola: on a graph it would look like spikes and every time you got a spike you got a spike of pain or intensity. Adebold: it's like being in a fire and smoke, having smoke inhalation damage where ye your natural instinct is to breath but then every time you breath you get more smoke so you hold your breath but you can only do that for so long as you have to keep breathing and it's a vicious circle</p> <p>Benjy I picture my crisis like a lightning & thunderous storm, cause when the pain first starts, it strikes at different positions around my whole body. Banjy: a monster... a painful one. A big painful one</p> <p>Charlotte: It's just like somebody, is like hammering inside your bone and drilling, and it's like all the way (inside), it's like you can't even point to it, it's like so far in that it's yeah, wherever it is its bad, it's horrible.</p> <p>Deborah Its, that blood vessel that's pulsating its just going D D D D D D D so its like knocking its like a wood pecker that's go D D D D D D D D D</p>

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	<p>D D D D D in the blood stream</p> <p>Deborah: We're talking about pain you cannot quantify. We're talking about pain you can't even express. You know we're talking about things that you probably don't know how to say to another person, Deborah</p> <p>Funmi: like a scorpion... Or that, burning sensation of y'know of which it just spreads .Y'know it's like a sting but then y'know it's like poison, y'know you've been stunk and then for some reason y'know it's getting higher and y'know it's like poison spreading in the whole body</p> <p>Glen And the boat has a leak (Okay) so there's the ocean. It's dark maybe and the boat has a leak (Okay) so there's the ocean it's dark maybe there are sharks in it something like that. And you've got this boat, which is better than being in the water with the sharks but you know that gradually this boat is going to start sinking</p>
<p>Varied within the same person and between different people</p>	<p>Adebola: I get pain ... coz not all sicklers are the same. Some get pain intermittently. Some get it every day. I get pain all the time. So I woke up in pain, I went to bed in pain. Adebola Adebola: in different parts of your body it feels different. Adebola: not everyone who has sickle cell the same. Not everyone who's SS is the same.</p> <p>Benjy: I don't think there's any two sicklers that are the same or with the pain they have</p> <p>Charlotte: See this is like I think everybody's had different experiences Charlotte: It's the same pain, I don't doubt that it's the same pain coz that pain I feel is like, I, from those conversations I've had it's the same reaction that other people have.</p> <p>Deborah: "we all suffer the same sickle cell, it's the same crisis but pain levels vary."</p> <p>Edith: everybody's pain is different isn't it? Its describe someone's describing to me their pain mines not like that mines not like that cause that bad? But you know everyone's is different yeah . everybody's is different</p> <p>Femi: So really y'know it depends where it attacks it kind of feels differently there are those who get it in their chest, know you, that is also different kind of sensation or pain Femi: I mean we all have difference, sometimes y'know we, depends to each individual</p> <p>Glen: "The unpredictability. And the variety. Different pains people can have. Crises in different places. And all the complications that can have a variety of effects on life."</p>
<p>Unpredictable</p>	<p>Adebola: it's beyond ridiculous where literally one second you are laughing, we could be sitting here laughing and joking and I said to you do you want a cup of coffee and I go and make a cup of coffee and err it's just that quick and it starts anywhere, from your finger to your shoulder joint wherever.. and it just.. it can build or it can just be intense. It can go from 0</p>

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	<p>to full intensity really quickly... erm and it's the er sort of surprise nature of it that's a problem. So there's no way of predicting</p> <p>Charlotte: Sometimes you can't control it, sometimes it lit, you can just wake up in the middle of the night and it could just be there</p> <p>Edith: but if you just sitting there and its just suddenly you get this pain its [chuckles] quite kinda quite strange</p> <p>Glen: it it can often, put maybe a distance between you and other people because it's so unpredictable. So, you can make commitments to other people and then have to just drop that commitment.</p> <p>Glen: So it's that changeable, unpredictable aspect, effectively. It's almost like it would be easier if it was a constant thing, then everyone would know what parameters you would work to and within, and, and it would be.. rather than somewhat changeable.</p>
<p>Affecting every aspect of life</p>	
<p>Every-day functioning</p>	<p>Adebola: it's got your attention constantly and then it it stops you from doing or just functioning normally. So how long you sit, to how you sit, whether you lie down, or just doing stuff. And what you do ermm whether you choose to go out or not go out.</p> <p>Adebola: because of the nature of it makes it hard to sit still. I tend to fidget a lot</p> <p>Adebola: the pain in your chest tends to make you hold your breath, it catches your breath. So breathing becomes painful</p> <p>Adebola "if I were to get up I'd be in more pain but I need to go to the bathroom."</p> <p>Deborah, "I want to go to work but (I can't) because I'm in so much pain."</p> <p>Glen "it's affected my ambitions."</p>
<p>Planning and the future</p>	<p>Benjy : Umm well now I live day to day really.</p> <p>Benjy: I try not to look to far into the future cause of the constant pain I suffer from, I try to live a day at a time,</p> <p>Charlotte: I question my future a lot because again like I said I've got two metal hips 18 and 21 so, they don't last forever you can only have a certain amount so I do wonder like what am I gonna be like when I'm forty am I gonna be in a wheelchair, and then also every single crisis you have you wonder, you actually gonna survive this one, you know like I could here and sickle down the road I could end up having a stroke God forbid. There's just so many things.. that can happen..</p> <p>Deborah: why I feel sad all my friends have boyfriends I don't have any. That's the other thing you have to deal with. You don't get many men coming forward to say I want to marry you. Or I want to date you. Because first of all they look at you and they can see the distortions in your body frame. Because if you look at ninety percent of sickle cell patients people that don't look after themselves you can see distortions within the frame. They either too skinny or they look like something's wrong.</p> <p>Glen: okay, so, so for example when we were going on honeymoon (Yeh) er [name] and myself, we, obviously there were all these things about insurance and so, insurance ordinarily for two people that age, where we</p>

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	<p>went, it would have cost us not even £40. But because of my medical condition and so it ended up coming to well so the best quote was about £250 I found and obviously we were going for slightly less than 2 weeks. So these, there are all those things as well. And then I couldn't get a mortgage oh no no no because for the mortgage you have to have life insurance (Okay) and they would not give life insurance because I had this pre-existing condition</p>
<p>Parenthood</p>	<p>Charlotte: And my parents claim that they didn't know that they were carriers until I had my first crisis</p> <p>Edith: I think from 7 months every fortnight yeah just to keep to make sure the babies were gonna come out come out well .</p> <p>Debroah: why I feel sad all my friends have boyfriends I don't have any. That's the other thing you have to deal with. You don't get many men coming forward to say I want to marry you. Or I want to date you.</p> <p>Glen It was (important). It was because ...knowing its impact on me, I could not bear... Having my daughter now, the idea of watching her going through that kind of (pain), er, y'know, I couldn't. I would hate that so much.</p> <p>Glen; sickle cell does affect life expectancy and you do y'know I can find myself wondering well, will I see my daughter grow up</p>
<p>Emotional impact</p>	<p>Adebola: coz problem with pain all the time your mood does change ermm and I'm no different. So but I'm just more aware of it so I have to work harder to control it</p> <p>Adebola: generally I try to sort of work really hard to not let it.. I don't I don't put THIS on other people.</p> <p>Adebola: "I have to work hard to say right ok, I'm staying on an even keel."</p> <p>Deborah: It takes a toll on the body because you become excessively tired..... and that's how it is. You become, sometimes you become mentally, you get mentally tired. You get emotionally tired you get drained. And you just don't even want to speak about it.. and that's exactly what it is.</p> <p>Deborah: Yes so when you are talking about things like pain you also have the emotional, I'm not just talking about the physical pain now if you see where I am coming from. You have the pain the emotional bits, you have the, you know you have the emotional bits, you have the mental bits, why do I have to go through this. Why me. Ah am am why I feel sad all my friends have boyfriends I don't have any.</p> <p>Edith: I try not to be angry at the world you know it's not their fault you know it's just something that happened.</p> <p>Edith: I don't I don't put it on you guys I don't start going round being moody and miserable you know I don't try to give them I don't put it on them at all</p>
<p>Complications</p>	<p>Adebola: The only difference as time goes on is some of the damage it does to other parts, so joints shoulder, hips, those those sorts of things change</p> <p>Adebola: So you you develop a problem with your hips for example, because your having a crisis because of how the sickle affects your bones and in your born marrow.</p>

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	<p>Adebola: So the pain is the constant but it's all the other bits that come along from strokes, er to sickling in the brain, to priapism, to erm leg ulcers, to necrosis, to mellitus, endless, problems with eyes, problems with iron overload, lots and lots, heart...</p> <p>Benjy: arhh the age of twelve when I had a stroke yeah hah so erm yeah had a stroke and totally my world fell apart</p> <p>Benjy: its because of sickle cell I've got my veins thrombosed because of the amount of usage that they you know erm when I was younger for instance every single time they needed blood they'd have to take it from one of my veins with a huge syringe. I mean now I've got the port which is a lifesaver really ermm, so now whenever they want the transfusion they can put it in there.</p> <p>Danielle: this is why we say you can't die from sickle cell you can die from the infection and complications of not being treated.</p> <p>Glen: The unpredictability. And the variety. Different pains people can have. Crises in different places. And all the complications that can have a variety of effects on life.</p>
<p>Normal rules don't apply</p>	<p>Adebola: I've been in an induced coma and still been in pain. Adebola: even if you're sleeping you can feel it in your sleep and that's a weird sensation.</p> <p>Benjy: to me pain is very different to what other people I think might umm because I live with it every day of my life unfortunately I, sometimes wake up in pain most times go to sleep in pain you know so constantly on the morphine (sneezes) constantly on morphine tablets</p> <p>Deborah: Because you think or your hospital policy is if it goes below seven its. That's transfusion time. But my blood level has been six for the last seven years eight nine and ten years and I am OK</p> <p>Glen: the thing we got taught at school was that pain was generally a signal that some sort of trauma had occurred or that it was a way for you to avoid particular stimulus so on and so forth. But for me in a way it's sort of been a.. control and by that I just mean that it it stops me doing particular things sometimes things I might have wanted to do are the times I should not kind of kicks in and tells you well yet this is where you may need to stop</p>
<p>Crisis pain vs every-day pain</p>	<p>Adebola: And as far as I am aware most other pain have a limit and a cut-off point sickle cell doesn't.</p> <p>Adebola: Yep coz I work on a scale form 1 – 10 so anything below 5 I am home. Anything above 5 I'm considering going to hospital but that depends how far above 5 that it. So a crisis for me would be where being in hospital needing ermm a lot stronger analgesia then I would take at home or could take at home safely to control the pain</p> <p>Adebola: If you shut your hand in the door that goes away eventually and you just get the ache. With sickle cell pain that doesn't go, it stays and you get the ache and the sharpness. That's the difference.</p> <p>Adebola: On a day to day basis if I were to describe pain in terms of colour, sound, and texture it would be blue/purple. Blue would be a calmer.. more colder sensation. So you're aware of the blue but it has no heat, it has no fire, it has no punch to it no intensity to it. The purple, there is a bit more intensity to purple and the strength of it. So every day it goes from the blue</p>

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	<p>to where you aware of it but it hasn't got the intensity of the purple. The purple has a bit more intensity so it grabs your attention. A crisis goes from purple to red blood red, fiery red. So you've got the yellows, you've got the red you've got all that mixed in and then if you imagine a sound wave where you're looking at a graph and I played you a sound and it and it spiked. That kind of chaos is sickle cell pain at it's worse.</p> <p>Benjy: but yeah from daily pain to crisis pain I'd say every day is a crisis pain, Benjy: I'd say there isn't a difference because everyday constant pain could ever turn into crisis pain if I didn't take.. medication for it yeah erm doesn't have to be morphine</p> <p>Charlotte: Well I get like the little aches and niggles and stuff like cold weather and stuff I don't really call them, I personally don't call them crisis cos I can deal with them like I can still kinda do my day to day. For me sickle cell pain is really debilitating like when you're in serious crisis you can't do anything like you can just about think of what your name is, you know so. I do like get the little pains like niggling pains or maybe crisis that will last like a day or two, three days but I, they're, they're alright like, it's annoying, but I can deal with them.</p> <p>Edith: because even between the even between the acute crises you can still have you can still have little crises in between yeah so end up getting all the milder ones without the biggie at the end that's how I look at it</p>
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Overarching theme 2: The dilemma of treatment

Limits of treatment	
Medication is not enough - Pain breaks through	<p>Adebola: even that's a weird thing as it doesn't take away the pain, it just takes the edge away. Adebola: you take ermm analgesia like morphine or something, the relief you feel from that is like to take away the extreme intensity and it it you see the misconception is that it takes away the pain, nothing does that, it takes away the extreme intensity. And it sort of takes the brakes off it for a bit but you still feel that pain</p> <p>Femi: So y'know after again, awhile, I think also as time goes on y'know it wears down obviously then the pain keeps shooting though the medication and then that's when you you want to ask for more because y'know the worst is coming, once it wears down</p>
Tolerance to medication	<p>Emily: I took my pain killers and then that erm that that does help but you always know it's there and you think when the pains gonna wear off is it still gonna be there or have I got to take more pain killers</p> <p>Deborah: .Definitely most definitely my base line tolerance pain may probably be seven. Your base line and because you don't go through pain your base line pain's probably two.</p> <p>Glen: I'll give you an example... my brother is a big man... At a point he</p>

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	<p>broke his leg... it's very painful. They give him I don't know, ten milligrams diamorphine okay. That was the end of that for him ... so then you've got this other guy who's what.. I'm only eighty kilo... and so yet he's saying oh we need to give him like thirty milligrams? Or whatever? It doesn't compute in our minds. So what we'll do, we'll try and we'll take ten and well see how he goes.</p>
<p>Own expertise ignored – misunderstood/not believed</p>	<p>Adebola: because within the medical profession there's a saying "pain is what the patient says it is" but with sicklers, actually I came up with this a few years back, ermm "pain is what the patient says it is unless your sickle, you've got sickle and then it's what the doctor says it is."</p> <p>Adebola: It's not the same thing. The doctor is not going to go through the whole process with you cus he understands that if you've broken something, or you've had a serious fall you're in pain and there is a reason why you're in pain, so the best thing to do is to treat that is for you to get the pain under control so he can look at whatever your injury is. With me and people like me they are not looking at it that way, unfortunately, they are not looking at what, something, we know what's causing you pain, causing discomfort, let's get it under control so we can make you as comfortable as possible and then we can deal with some of the other problems that that are related to that. That's not the general attitude.</p> <p>Adebola: there's a handful of people who know me really well that, that can tell ermmm some other people who think they can tell because they think my mood changes</p> <p>Adebola: if you're, y'know if you went into hospital, into A&E as [researcher] and the doctor said what's wrong and you said I fractured my leg, I fell down the stairs and hurt myself; I'm in tremendous amount pain and he could see you're in pain the doctor would be sympathetic.</p> <p>Benjy No No erm obviously, those ones where I wasn't believed it did affect my opinion really hardly I always felt I took it personal why would anyone want to lie about pain and why would think that I would be the sort of person that would be lying about how I'm feeling you</p> <p>Deborah: So it went on from zero because at the time I felt there was no medical practitioner that understood what I was going through. When I was in hospital the nurses there were horrendous.. They were horrible they didn't understand the skill of the pain that one was going through</p> <p>Deborah: So it went on from zero because at the time I felt there was no medical practitioner that understood what I was going through. When I was in hospital the nurses there were horrendous.. They were horrible they didn't understand the skill of the pain that one was going through</p> <p>Edith: I have a prob a lot of people don't believe they say are you disabled Jo and I say yeah you don't look it but you don't have to look it it's a hidden disability</p> <p>Femi: they question the pain</p> <p>Glen: the idea that saying well that I did do this back in 1998 or 2007 or whatever and it's not enough... It's virtually ... sort of experimenting on you</p> <p>Glen: There are others who will say right well you've had some opiates so, so you can't get out of bed. Well I want a newspaper because obviously you can't lie in bed staring at the ceiling. No, no, no. If you're well enough to go</p>

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	to shop downstairs to get a newspaper then y’know, you don’t need any painkillers you’re ready to go home.
Understanding HCPs position	<p>Deborah: So you have to wait it out or it might take it away for an hour and bring it back and the pain comes back within. Three four you know two hours so an hour you’re not in pain your waiting another two hours because they won’t give you the medication until it’s four hours so that they don’t overdose you</p> <p>Femi: you’re hands are tide, basically, you can’t do anything, you’re in pain, the person you think y’know you can share you’re pain with can’t do anything so you’re left on your own to y’know deal with it, so it’s really frustration, annoying, y’know you just want to scream on top of your voice. Much as maybe the pain may not be the one making you to scream but its just the being aggravated, y’know, nothing can be done, but what you’re feeling at the moment y’know until they feel ok this is maybe the time can do something about feel very frustrated.</p> <p>Femi: its also very difficult y’know for them cause y’know they kind of just can’t believe that you’ve just had this dose of and within 30 minutes or so you’re still feeling the pain and you’re wanting more medication y’know pain killers and and properly y’know they’re profession doesn’t allow you then to give you any more so that’s where the problem</p> <p>Glen: I think, I, because I think it’s a fundamental. . It’s a, it’s a problem with human beings really y’know. A lot of the time they’re always going to want to see this cos when they look at it they think well, ahm. Okay</p>
Passports didn’t work	Femi: There was a time they said they are, um, what are they called, passports, they give you ... You know I don’t even know, I don’t know where I put it cause it was useless, I say what is the point of when you are in pain trying to scratch it where it is and go with it so that it helps you, you know.
The impact of Health Care Professionals	
Level of experience and knowledge	<p>Adebola: the one thing we get labelled as is junkies. They just think we’re there for opiates. They just look at it as, as it’s, it’s, that’s just how they see us.</p> <p>Adebola: Y’know and you have doctors that have ridiculous opinion, y’know and make silly comments or pft erm prescribe things that you’re thinking.. to be honest, either you’re deliberately trying to antagonise me... ok or you shouldn’t have made it through medical school to be honest you’re an idiot because if I come in and I’m sicking in crisis why would you prescribe me something that if I stubbed my toe you would probably give me.</p> <p>Adebola: But there are too many like that. Whether it be doctors or nurses. Er some it’s through ignorance through not knowing.</p> <p>Benjy: the one that was supposed to be looking after us was still on her holiday so people that were there looking after us were people that weren’t were aware of my illness and condition but weren’t aware how bad it was ... So the doctor came in and was like erm ohh how come I didn’t see him this morning coz I was in earlier on and she was like oh he wasn’t bad then we just didn’t think he needed seeing to you know umm and she was like well I wish you hadn’t done that because he’s quite very much more ill than you think he is he’s actually had a stroke right now and we need to get him into hospital as soon as possible..</p>

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	<p>Femi: Yeah could be in A&E or could be your GP or y’know, could be whoever, not your consultant. Your consultant gets to know if they remember even to tell the consultant cause they’re supposed to notify the consultant that you’ve come in then consultant sees the other side of the SC, of things, then raises y’know issue with them, so ok, person with SC this will happen if this doesn’t happen if y’know do this then they combine their own understanding of it then general well, well-being of a normal, y’know, a normal person without SC who is ill into what the haemoglobin doctor is saying. So in, y’know, y’know in in normal circumstances you are dealing with those who don’t have the knowledge of what you’re illness is they’re just dealing with, I don’t know how to, if you understand, they’re just dealing with the normal situation of someone being ill, y’know someone being in pain.</p> <p>Glen: So the ones that have more knowledge, experience maybe understanding the situation (yeh)I value them very highly</p>
<p>Level of interest</p>	<p>Adebola: occasionally you’ll get a doctor or nurse or someone medical who actually asks you a question, or what’s like, not because but just because they want to know and they’re interested and they they ask you and y’know they don’t want you to give them some sort of standard answer so they can tick their boxes they are actually asking you as you are asking me know, which is different so I’ll take the time to explain. Erm (cough) and then you get others who couldn’t care less, and it’s quite obvious.</p> <p>Adebola: if you have an ill-informed opinion of me or people like me that’s going to affect how you do your job, how you approach the job, how you come into work in the morning. Everything you do</p> <p>Charlotte: they literally do have that attitude as in like just give them what they want and shut up let them shut up and just leave them there</p> <p>Glen: you see it with um doctors through doing it with their rotations and your talking to them, I mean (exhalation of breath) they could not care less. They have to do it. They don’t want, they definitely don’t want to specialise in it and they want to waste as little mental energy on it as possible. You can see it very plainly.</p>
<p>Building relationships</p>	
<p>Inconsistent staffing</p>	<p>Adebola: I’m there every week. So if I have a bad experience last week, or a good experience last week, which I did, I know the fact that I am going to go there this week may be different.</p> <p>Femi: That is also varies cause most of the time you don’t see the same person</p> <p>Glen: And then you have other doctors who always come in and you they always have something that they want to try that you’ve been through before.</p>
<p>Positive relationships make a difference</p>	<p>Charlotte: I come really close with doctors and nurses and stuff so I can’t really complain too much</p> <p>Edith: I got pregnant and I went I had to go to [hospital] and they said oh you know it was quite serious you know erm being a woman having your this diagnosis your gonna have children and your gonna have twins I</p>

	<p>thought oh my god so they really closely monitored me for that so erm they they really the [hospital] the medical staff were really good they monitored me all the time</p> <p>Edith: I prefer to work than erm not work I prefer to be active than not active cause then [consultant] said to me oh EDITH you need to work and he was I mean cause maybe 2, 4 or 5 years ago I might of you know taken early retirement or just stop work but [consultant] was always saying erm ... oh “you know keep working it’s good for your back its good for this its good for that” so I I just kept working</p> <p>Femi: y’know, who, which doctor dealing with or which profession you’re dealing with, whether they really understand the illness and how it affects you.</p> <p>Glen: I value them very highly y’know for example, that’s to give you an example of how extreme that can be, that’s what changed where I live now, well the house that we’re moving into now, simply so that I would be in the catchment area for [Hospital] because of the culture there in the haematology department because the experiences my wife and I have had for example, um, obviously we weren’t married then but when we went to the same university. So there, at that [hospital], erm, [hospital] un, erm, I mean horrible beyond belief and so the stress involved.</p> <p>Glen: Name] is not a doctor but also I regard [name] as a friend and even the um successor, er, in the job? [name2] as well because the fact that they are willing to make that effort for you, it means that really ... make. I mean. They improve my quality of life no end! So, there’s nothing, I don’t know it, really makes a huge huge difference</p> <p>Glen: the fact that I had a team of doctors and nurses who were competent and who did care, it made a huge difference it’s made a difference to my family.</p> <p>Glen: For example one of my doctors came to the naming ceremony for my daughter.</p>
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Overarching theme 3: finding a life with pain

Struggling to understand	
<p>Varied explanations for pain</p>	<p>Deborah: it something with sickle pain and oxygen as well.. because I notice that when I have sickle pain when I hold my breath. I don’t have sickle pain anymore. ..When I let the air out. and the blood’s flowing very freely I get sickle pain so theres something with sickle pain and oxygen levels. Yeah. Id Id It lot of sicklers that’s exactly how they hold onto the pain..breath in you don’t get the pain. you breath out so theres something with the oxygen levels and the oxygen levels in the bloodstream must be very very low, extremely low, for you to have that pulsating pain. where theres less oxygen getting into that blood stream because if the blood was flowing freely you shouldn’t get that pa, if the oxygen levels were flowing in the blood stream you shouldn’t get that pain.</p> <p>Deborah: When was the last time I juiced. Vegetables. Because I find that water and vegetable juicing do exactly the same thing or exactly the same. Give exactly the same results but vegetable juicing is much better because it gives you the extra oxygen from the plants and then also gives you the nutrients that come with the vegetables that your body needs.</p> <p>Femi: well you sometimes associate it... y’know being in Africa there are</p>

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	<p>associated with so many over things, y’know, either, y’know, being, y’know good thing maybe you have an infection, or its malaria or it’s, it’s got to do with your genes, or it’s y’know, things like suspicions, they would associate it y’know, with different things, y’know, sometimes they think o maybe they have ah sent you bad omens or something like that, they call it.</p>
Unknown triggers	<p>Adebola: “the pain in my legs, which for some reason I tend to get a lot of and always have done”</p> <p>Edith: I’ve thought where’s this coming from why am I getting pain there</p> <p>Glen: But the thing is I don’t know if it’s the physical injuries are then triggering the sickle pain if that makes sense.</p> <p>Glen: And then i will have crises pain and I often find myself wondering, is the one triggering the other, sort of thing.</p>
Role of self	<p>Adebola: depending what I’m doing, how long I’m sitting still. Whether I suddenly do anything.</p> <p>Adebola: and then if you’re having a bad day then it’s all over and it’s not good. It’s not good at all.</p> <p>Adebola: y’know there are certain things that if you do or if you are feeling unwell that are more likely to bring on a crisis</p> <p>Benjy: there’s many times that I’ve caught it late and gone through a crisis phase</p> <p>Charotte: I think that if I get those niggling pains and stuff I think that if I treated it as if it were a sickle cell crisis it would actually turn into a big sickle cell crisis</p> <p>Danielle: I drink loads of water when its cold I find myself drinking loads of tea. And less of water but I do drink. I could have like four cups of tea in a day and that’s why I say I’m bad because I take in more caffeine than I will take in water if its cold</p> <p>Femi: so but you know sometimes then sometimes gets ignored were you are looking and like um this is not, cause the more it gets unwell doesn’t drink much enough, not eating and not very, then the body gets more and more and then crisis you know sets in.</p>
Inseparable part of life	<p>Adebola: it would be like trying to separate me from my colour.</p> <p>Adebola: this kicked in at 6 months old so before I had awareness of anything, self, whatever I had awareness of pain so I couldn’t understand or even put I name to it but I could feel pain.</p> <p>Adebola: if someone could wave, sort of wave a magic wand and take that pain away .. you’d probably feel some kind of bereavement because you’d think “errmmm you know you can deal with that”</p> <p>Benjy: Overall its part of my life. I don’t know if I can say it means something separate because it’s part of who I am</p> <p>Charlotte: Yeah it’s part of me yeah.</p> <p>Emily: it’s with me all the time so it’s a part of me now</p>
Relationship with the pain	<p>Adebola: I suppose... how... at it’s worse.. hmpf you’d treat it almost like erm almost combatant... you become combatant, and you become sort of</p>

	<p>emmm battle mode because to be able to to fight the pain affectively and to to be able to deal with that level of pain you have to have ... you have to almost get angry with the pain, you have to almost show aggression towards the pain</p> <p>Benjy: I find and see my pain with indifference, I have never or will ever personalize my pain with any of the descriptions that you have given above. I see It just as a condition that has afflicted me from birth & I deal with it daily so not meaning to demean it in any way but I'm used to it.</p> <p>Charlotte: It's your enemy and you hate it but it's part of you</p> <p>Deborah: I want to strangle pain every night.. I wish I could take pain and put it on a noose and put it to hang and that's how I feel but to have a relationship with it, no I don't want you as a relationship, I don't need that relationship.</p> <p>Edith: Well I'd advice anybody who has pain to befriend it . yeah I think I think that would that helps a lot well I cant really talk for everyone ... Yeah I find I know it might be a safe side, a safe thing to do but [sigh] . I just I just don't want to have pain and if I can . squash it a bit or keep it down . but I do that in anything I have if I have a problem in life sort of try and that's me I don't know if its the right thing to do try and squash it stop it you know keep it down but I do it on a friendly I'm not I'm a non I'm not very aggressive.</p> <p>Femi: Day to day one you know you would call it companion really. Day to day one you know is your companion you know its there, it will always be there, you know its accompanies you everywhere you go but the other you know you can't call your companion cause you can't live with it, you can't tolerate it you can't, so really you can't have a relationship with it, it's either there and you fight it or not there at all, so yeah</p> <p>Glen: I, er have a troublesome relationship with, because, and even though it doesn't make sense? I find sometimes I'm personifying it. It's er. It's like, it cos it's like. As I said. Kind of spiteful, something you could hate.</p>
<p>Living with pain</p>	
<p>No choice but to cope</p>	<p>Adebola: And... it... it's just always there... so you, y'know you both cope but it's very different. There isn't a sense of 'I'll cope here' I cope because.... Errrr.</p> <p>R: there is no choice?</p> <p>Adebola: yeh!</p> <p>Edith: it's just it just can't there's no control you can't control it you can't control it at all yeah you can't control it at all yeah pressure and you can't do anything physical you can't you just don't wanna do anything you just wanna ooh yeah hmm</p> <p>Edith: I think I have I think I have I mean I've had too there's no one else who can help me and people you know when you talk to friends oh EDITH you know they don't wanna hear it you don't want to bog bog down people with your pain all the time you know they get fed up with it as well</p> <p>Glen: I'm walking around, and these are my companions! [shows</p>

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	<p>painkillers] Do you see what I mean? And you have to carry them everywhere with you. And (Yeah) I might not need them today, but, so. It's that, that ridiculous sort of situation, like, carrying painkillers around. So I've got my little bottle of water and my painkillers and er, yeah. So. But anyway that's the end of my ramble.</p>
<p>Fighting vs appeasement</p>	<p>Adebola: it wouldn't sort of stop me from doing stuff. Adebola: at it's worse.. hmpf you'd treat it almost like erm almost combatant. Adebola: you become combatant, and you become sort of emmm battle mode because to be able to to fight the pain affectively and to to be able to deal with that level of pain you have to have ... you have to almost get angry with the pain, you have to almost show aggression towards the pain Adebola: Yeh so I can be angry at the pain, combatant with the pain and and think right ok lets go to war. Stand toe to toe, Marquis de Queensbury rules... last one standing wins. Adebola: yeh so I keep ... I get up and I relish the fight. Because it's gonna, the fight is going to happen whether I want it or not so I relish the fight so rather than think 'right ok' I think 'let's go to war.' Edith: it's like a lot of things you'd do with a friend you've worked into your relationship with pain to get on with it Glen: I had this erm pain sort of flaring up and so sort of have to stop and take it easy for maybe a couple of hours cos you need it to to sort of subside</p>
<p>Knowing self</p>	<p>Benjy: I know that it is just a few days before I'm gonna be, in for my transfusion I just, take the trans the morphine Charlotte: I honestly think its just people learn their body and stuff. Deborah: if I get ill I know I have to check three things... the first thing I do is check my diet and check what have I been doing. Have I had water.. have I been eating vegetables. Femi: I mean the body tells you you know it that you know the usual and also when you get something else still the body reacts differently from the usual to the un-usual. cause when get something you feel say you know this is not going you know to end up you know well Glen: By that time you, like by 30 even, you probably know the way your body works, you know your limits, you also know the ways it can help you cope with the, the, um, condition.</p>
<p>Self-care</p>	<p>Benjy: So yeah um so um and those are just the normal medications that I.. I've always taken medication Charlotte: for me like when I get the little niggling pains I will extra wrap up warm ensure I've got layers on, have really really hot baths, to make sure you know like I'm hot through and through, through the bones and everything or hot water bottles or stuff like that. Danielle: cold I'm sitting here because I'm right by the heater. I won't sit close to you because its cold it's I'm in a cold room. I need this to stop me from getting cold or from getting for my body getting.. or for me changing</p>

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	<p>the temperature in my system</p> <p>Femi: Properly like, um drinking a lot, obviously um, medication, y’know you have regular medications and um, and also y’know eating healthy as well and also having a lot of rice because sometime y’know people sometimes they forget to, y’know you forget and over y’know over work y’know yourself and then before y’know it your breaking down and then of course that’s how the whole system kind of gets into shock and then that could send you into a crises.</p>
<p>Critical sense of time</p>	
<p>Age</p>	<p>Adebola: I think the younger me would have reacted to it. Adebola: “hopefully the, the older I am, the longer I live, the better I get at dealing with (pain).”</p> <p>Glen: I was far more ignorant of my condition when I was younger, Glen: By that time you, like by 30 even, you probably know the way your body works, you know your limits, you also know the ways it can help you cope with the, the, um, condition</p>
<p>Experience</p>	<p>Adebola: if you have to develop it or learn how to deal with it and err your coping mechanism is different, so how you view the pain and how you cope with it and.. it is very different from something that you have always had. Adebola: I’m someone who’s been in and out more times then he has actually be there. I’ve spent more time in hospital than he does probably.</p> <p>Charlotte: I’ve tried to explain to my mum how it feels and you just really can’t. Everybody said any sickler will say I don’t think, I really wish they could simulate a machine that could make people feel it, but its just something you just you can’t explain that pain.</p> <p>Deborah: I can handle pain much more because I’ve been through sickle pain</p>
<p>Knowledge</p>	<p>Adebola: I’d say the best comparison would be trying to ask someone who’s who’s born blind and someone who went blind ermmm someone who’s born blind is aware of the fact they can’t see but you try and describe colour. Try and describe the colour blue or the colour red or soft or .. and even if you said can you feel that it’s rough and can you feel that it’s smooth how are they going to get a picture of what rough and smooth is, there is no picture just texture. Because there is nothing to compare that too so their experience is... and someone and .. they both have loss and a sense of not having something but it’s different, the one who’s lost their sight knows what they’re losing. You you had something to loose, you’ve had something taken away whereas if you were born blind you’ve had your sight taken away but all the things that come with sight you didn’t have them to begin with. So you’ve lost them but you haven’t experienced it. But whereas if you lose your sight you’re losing more because you’ve experienced it, you’ve gained knowledge from it.</p> <p>Femi: To be honest I didn’t know, y’know, what it, exactly what it was. Y’know, until I y’know probable, y’know, got the counselling here when I got ah pregnant and all that so I found out then, y’know, how it would y’know affect y’know everything Femi: well how you come to accept it is first accepting that you have a long</p>

APPENDIX

	term illness, then also learning more about your long term illness, how it's going to affect you in the future and what you can do about it,
Acceptance	
It's not going away	<p>Adebola: And the difference is where someone that has an illness, even say cancer or whatever, and goes into hospital and has a good or bad experience it's a once in a lifetime thing. They're not going to come back again. And if they do it may be in the distance, but for me I know I'm going to be there a lot</p> <p>Adebola: I'm going to have to keep going back. Whether I want to or not</p> <p>Charlotte: we won't be cured, it's either you live with it or you die.</p>
Appreciating the small things/life	<p>Adebola: I don't get caught up in all that stuff, it's not really important. Something's are nice to have, but it's not like the be all and end all.</p> <p>Edith: that's another reason why I go to work because they're there for me [chuckle] they're not there for me but they they help me a lot</p>
Positives lessons about self	<p>Adebola: And I use I use the fact that the pain is there as a way to motivate me and to push me.</p> <p>Adebola: you spend an awful lot of time on your own. Y'know I like my own company.</p> <p>Edith: To be tolerant I think I just to be tolerant erm to be mature about it er And then well yeah well not to laugh at anyone who's in pain or who's ill or is unwell you know never take advantage if anyone's vulnerable I never take advantage of that person</p> <p>Femi: as you go along the way you know, you kind of discover what kind of person you are and what kind of things actually you can cope with, and things you can't cope with</p> <p>Glen: Maybe a more realistic view of life. Maybe a better understanding of the struggles that people can go through and as a consequence I'm probably more, I dunno, maybe more compassionate than they would tend to be.</p>

Appendix 12: Common Sense Model's illness representations and research themes

Illness representation/ cognition	Definition	Example	Themes
Identity	Beliefs about the illness label (medical diagnosis) and the symptoms experienced.	The diagnosis –a cold; The symptoms - runny nose.	Normal rules don't apply
Cause	Perceived cause of the illness. Causes may be biological, (a virus or a lesion), psychosocial, (stress or health-related behaviour), or environmental (pollution or chemicals). Patients may also hold illness representations that include multiple causal factors.	'My cold was caused by a virus.' 'My cold was caused by being run down'.	Struggling to understand
Time line	The patients' beliefs about the course of the illness and the time scale of symptoms; short-term (acute) or long-term (chronic).	'My cold will be over in a few days.'	Critical sense of time
Consequences	Beliefs about the possible impact of illness on overall quality of life and functionality. Such consequences may be physical (e.g. pain, lack of mobility), emotional (e.g. loss of social contact, loneliness) or a combination of factors.	'My cold will prevent me from playing football, which will prevent me from seeing my friends.'	Indescribable pain, Affecting every aspect of life
Curability and controllability	Beliefs regarding treatability and cure, degree in which the outcome of illness is controllable either by themselves or by powerful others; feeling of empowerment regarding performance of coping behaviours.	'If I rest, my cold will go away.' 'If I get medicine from my doctor my cold will go away.'	Acceptance, Limits of treatment, Living with pain, Critical sense of time, The impact of Health Care Professionals, Building relationships