

**Trait Anxiety in Sickle Cell Disease: investigating
and exploring links to the management of pain**

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DPsych, University of East London, 2014

A thesis submitted in partial fulfilment of the
requirements of the University of East London for the
Professional Doctorate in Counselling Psychology

University of East London

July 2014

ABSTRACT

The current sequential explanatory mixed-methods study investigated the relation of trait anxiety and health outcome variables (pain intensity, depression, quality of life, hospitalisation frequency) in Sickle Cell Disease (SCD). SCD is a hereditary blood disease that causes frequent pain in people with the disease. Pain management is a goal of disease management in SCD. Trait anxiety was used as a hyper-vigilance construct to investigate the relationship between hyper-vigilance and health outcomes in SCD and across an illness comparison group (Blood Cancer, BC) and an illness exposure control group (Carers). Participants ($N = 51$) completed online self-report measures of anxiety, depression, pain intensity and quality of life. Hierarchical regression results showed that the variance in trait anxiety was significantly predicted by: depression scores (48.1%, $p = .001$) and quality of life scores (27.1%, $p = .001$) in the complete sample (SCD, BC, Carers); and by depression scores (67.2%, $p = .001$) and quality of life scores (17.7%, $p = .021$) in the complete SCD sample ($n = 28$). Multivariate results with equal group sizes ($n = 24$) revealed the SCD group experienced significantly greater sensory pain ($p = 0.018$) and lower general health ($p = 0.019$) relative to Carers. The SCD group also experienced significantly lower depression ($p = 0.044$) relative to the BC group despite having similar levels of trait anxiety as the BC group. Thematic analysis of qualitative semi-structured interview data revealed six themes: pain appraisal, purpose and change in identity, coping strategies, anger and frustration, social construction of illness and personal control. Data integration showed hyper-vigilance behaviour was more prominent in SCD, relative to BC and more likely to be used as an adaptive coping strategy in pain monitoring and prevention, or in pain adjustment. Counselling psychologists and healthcare practitioners may need to consider that reducing hyper-vigilant behaviour in SCD may increase pain experience and affect pain management negatively.

Dedications

This dissertation is dedicated to all of the courageous participants in this study who have contributed to the knowledge of pain management in Sickle Cell Disease, in Blood Cancer and also in Carers.

A special dedication is given to my beloved family who have shared in the trials and tribulations of my progression through the course and in the production of this thesis. They always believed in me and I would not have been able to reach this goal without them. I have to specifically thank both of my parents who have always instilled the love of truth and learning in me and whose unwavering value of education has led me to pursue this degree. Their emotional, financial and physical support has helped me complete this course and this accomplishment is as much theirs as it is mine.

A special mention must go to my mother who passed away during the course of this study and to whom I owe my integrity, my passion and my resilience.

For my Mother

Acknowledgements

This thesis came together as a result of the guidance, support and assistance of many different individuals and groups of people. I am deeply grateful for the patience, support, knowledge, love and kindness that I have received from everyone who has helped me on my four year journey towards this counselling psychology doctorate.

I would like to thank all the men and women who participated in this study. Thank you for sharing your experiences and personal information with me – this study would not have been possible without you.

I would specifically like to thank Sam and Cecilia Shoetan from the Barking, Dagenham and Havering Sickle Cell and Thalassaemia Support group for including me in their support group. They allowed me to participate as a member of the support group and gave me assistance in recruiting participants and in understanding the psychological effects of the disease. I would also like to thank the members of the support group who welcomed me as a member of the group and who offered support and advice along the way. Thank you to the Sickle Cell Society for providing recruitment advice and allowing me to recruit at their conferences. Thank you to the Ealing, Hammersmith and Fulham Sickle Cell and Thalassaemia Support Group and to the numerous Facebook sites supporting Sickle Cell Disease and Thalassaemia and the carers of people with these diseases.

Thank you to Leukaemia Care and the Blood Genetic Diseases support groups for their platforms that helped me to recruit participants and to the numerous myeloma, leukaemia and lymphoma Facebook support groups that supported the study and assisted in participant recruitment.

A special thank you goes to my supervisory committee: Dr Meredith Terlecki, the director of studies, Professor Rachel Tribe and Dr Kendra Gilbert. The ideas of this thesis and its execution are the result of a collaborative effort and would not have been completed without their expertise and wisdom and also their patience and latitude! Thank you so much.

I would like to thank the Counselling Psychology teaching team: Dr Kendra Gilbert, Dr Jane Lawrence, Dr Melanie Spragg, Professor Rachel Tribe and Dr David

Kaposi. Thank you for all your support, guidance, kindness and compassion over the last four years; especially when it was really difficult for me to continue. I really appreciate your support.

I would like to thank Dr E.M Camm, Nadia Al-Khudairy, Dragana Savcic-Sanders and Dr Mark Kingsley for helping me to complete this course in different ways. I appreciate the time and effort that it took for them to assist me and I am deeply grateful.

My family and close friends have unlimited levels of patience. Thank you for going on this journey with me and giving me your unwavering support. Thank you for being there and for riding the highs and lows with me. Thank you very much.

And finally, I would like to thank the Professional Doctorate in Counselling Psychology class of 2014. You are all amazing and wise people. We embarked on this four-year journey together and together we weathered many storms. Thank you for your kindness, support and honesty. I have learned much about myself from all of you. Thank you.

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List of Abbreviations

| | |
|------------|---|
| ANOVA | Analysis of Variance |
| BC | Blood Cancer |
| BDI - II | Beck Depression Inventory (1996 revision of the original BDI) |
| CBT | Cognitive Behavioural Therapy |
| CSQ | Coping Strategies Questionnaire |
| DV | Dependent Variable |
| GCT | Gate Control Theory (of pain) |
| HADS | Hospital Anxiety and Depression Scale |
| IV | Independent Variable |
| IP address | Internet Protocol address |
| M | Mean |
| MANOVA | Multivariate Analysis of Variance |
| NHS | National Health Service |
| PHQ | Patient Health Questionnaire |
| SCD | Sickle Cell Disease |
| SD | Standard Deviation |
| SPSS | Statistical Package for the Social Sciences |
| STAI-S | State -Trait Anxiety Inventory – State version |
| STAI-T | State - Trait Anxiety Inventory – Trait version |
| SF - 36 | Short Form 36 (Medical Outcome Survey) |
| SF - MPQ | Short Form McGill Pain Questionnaire |
| UK | United Kingdom |
| US | United States of America |

Chapter 1: Introduction

Sickle Cell Disease (SCD) is one of the most common single gene disorders in the world (Rees, William & Gladwin, 2010). There are 250 000 people in the United Kingdom (UK) who are carriers of the sickle cell gene and 13 500 people who have SCD (National Health Service; NHS, 2006). In addition, Brousseau, Panepinto, Nimmer & Hoffman (2010) estimated that 89 000 people have the disease in the United States (US). Consequently, it is the most common inherited blood disorder in the UK (NHS, 2006; Rees et al., 2010) and in the US (Brousseau et al., 2010; Hassell, 2010). These figures indicate the presence of SCD in countries outside the geographical areas where there is a selective advantage for people who are carriers of the gene, but do not have the disease itself; resistance to malarial parasites in the red blood cells is increased in people who are carriers of the SCD gene and also, to a lesser extent, in people who have the disease (Rees et al., 2010).

Rationale for the Current Study

SCD has traditionally been conceptualised and managed using the medical model (Anie, 2005; Anie & Green, 2002; Rees et al., 2003), which focuses on the biological and physical symptoms of the disease and on medication adherence. Psychological interventions are used to assist disease management in SCD (Anie, 2005; Midence & Elander, 1994; Rees et al., 2010); managing pain symptoms in particular. Reporting the development or efficacy of psychological interventions for ongoing management of SCD is in its infancy in academic research (Anie & Green, 2002; 2012). Only 11 research papers (randomised or quasi-randomised methodologies) reporting the efficacy of psychological interventions for the ongoing treatment of SCD emerged in an international review in 2012, six of which were suitable for the review: Borroffice (1991), Broome, Maikler, Kelber, Bailey & Lea (2001), Gil et al. (1996), Gil et al. (1997), Kaslow et al. (2000) & Thomas, Dixon & Milligan, 1999 (Anie & Green, 2012). Anie & Green (2012) reported that cognitive-behavioural therapy (CBT), psycho-education and psychodynamic psychotherapy were the main psychological interventions used in SCD to support ongoing disease management. Anie & Green (2012) only reviewed randomised or quasi-randomised studies in their report; however, other non-randomised studies have suggested the

usefulness of including psychological interventions in the ongoing management of SCD (Cummins & Anie, 2003; O'Connell-Edwards et al., 2009; Thomas, Wilson-Barrett & Goodhart, 1998). These papers were not included in Anie & Green's (2012) review because they were not thought to be methodologically rigorous; however, the results from these studies do indicate that there are different methodological ways of demonstrating psychological relevance in SCD management, including mixed methods studies and cross-sectional studies. It is apparent that whilst psychological research in SCD exists, there is room to add to this literature through mixed-methods research. It is important to hear the participants' voices and their perspectives of their problems, in order to make subjective sense of measurable objects i.e. pain experience (Bhaskar, 1998).

An important psychological variable to consider in pain management is trait anxiety. Trait anxiety (as opposed to state anxiety) is considered to be an enduring personality trait or an enduring characteristic (Block, 2002; Spielberger, O'Neil & Hansen, 1972; Spielberger & Smith, 1966; Spielberger & Vagg, 1984). Individuals with high trait anxiety show a constant awareness of potentially impending primary anxiety, rather than anxiety in response to the presentation of an actual anxiety-causing trigger (Block, 2002; Spielberger et al., 1972). According to Spielberger's state-trait model (Spielberger et al., 1972; Spielberger & Vagg, 1984), external stimuli (environmental, social or situational stressors), as well as internal stimuli (thoughts, feelings and biological needs), are appraised in a way that either induces anxiety in a person or not. The appraisal of the stimuli is thought to be a function of a person's level of trait anxiety (Spielberger et al., 1972; Spielberger & Vagg, 1984). If the stimuli are cognitively appraised as being threatening, defence mechanisms are engaged that adjust for avoiding or reducing the threat to the person, which in turn causes behavioural change. Trait anxiety, as measured on the *State-Trait Anxiety Inventory (STAI-T)* (Spielberger, Gorsuch, Lushene, Vagg & Jacobs, 1983), is known to be stable over time and is, therefore, considered to be a measure of dispositional tendency towards anxiety. Research has shown that the trait anxiety personality type does bias the memory for items that cause worry and elicits the fear response in the presence of these items (Reidy, 2004) and creates a bias towards negative events associated with these items (Eysenck & Derakshan, 1998). In addition, Eysenck (1992) reported a hyper-vigilance theory that proposed that hyper-vigilance caused

by high levels of trait anxiety influences the cognitive vulnerability factor for clinical anxiety. Hyper-vigilance has been defined as the behaviour of constantly scanning the body for somatic sensations; pain in particular (Chapman, 1978). High trait anxious participants have been found to perceive and detect threats faster than low trait anxious participants in a study that aimed to examine the relationship between threat estimation and trait anxiety (Longin, Rautureau, Perez-Diaz, Jouvent & Dubal, 2013). From these findings it was suggested that the hyper-vigilance to a threat may be adaptive in a person when it increases the ability and the speed of detecting risks and increases the speed with which these risks can be avoided. High levels of trait anxiety have been associated with chronic pain diseases, such as fibromyalgia (Crombez, Eccleston, Van den Broeck, Goubert & Houdenhove, 2004), rectal cancer (Ristvedt & Trinkaus, 2009) and SCD (Burlew, Telfair, Colangelo & Wright, 2000; Thomas et al., 1998; Thomas et al., 1999). Thus, the role of trait anxiety across these illnesses may differ due to the different presentations and aetiologies of these diseases. Despite this observation, very little research has focused on the relationship between trait anxiety and health outcomes, especially in relation to SCD.

The current study aims to focus on how psychological variables interact with health outcomes in SCD by focussing specifically on trait anxiety and pain management. This study aims to gain an appreciation and an understanding of the role of trait anxiety in pain management in SCD. It is hoped that attempting to understand the underlying role of trait anxiety will improve the efficacy of psychological interventions in future studies and will demonstrate how a critical realist approach to SCD can help to reveal that there are different levels of anxiety in the disease. The next section will describe the presentation, aetiology and management options for the disease to demonstrate the practical and emotional difficulties of living with the disease. Demonstrating these difficulties will highlight the relevance of including psychological perspectives and interventions in pain management for SCD.

Sickle Cell Disease

SCD is the term used to define a group of blood disorders that are caused by an inherited, mutated single recessive gene that causes the protein bonds that hold a blood cell's shape to change from creating a healthy round shape to creating an

incorrect sickle shape (Elander, 2007). Sickle cells are elongated red blood cells that give the blood cell a crescent shape rather than the normal round disc shape when oxygen concentrations in the red blood cells are lowered (Elander, 2007; Idemudia, Helen, Kolawole & Morenike, 2001). This results in a lack of oxygen in cells and a lack in capacity to transport oxygen to other red blood cells. In addition, the irregular shape of these crescent-shaped cells causes blockages in blood vessels.

Sickled-blood cells obstruct the blood vessels due to the incorrect shape of the red blood cell, which leads to pain in bones, organs and muscles (vaso-occlusive crises; Rees et al., 2010; Shapiro, 1997). Vaso-occlusive crises can also lead to subsequent organ damage, an increased likelihood of blood clots and strokes and an increased vulnerability to infections, especially if the spleen has had to be removed due to the disease (Sogutlu, Levenson, McClish, Rosef & Smith, 2011). Consequently, SCD is a multi-systemic disease (it affects all the blood vessels in the body, and therefore all the systems e.g. cardiovascular, immune, respiratory systems) with a range of acute and chronic symptoms and effects which also includes experiencing varying degrees of anaemia which affects physical mobility and functionality (Anie, Egunjobi & Akinyanju, 2010; Anie & Green, 2012). A vaso-occlusive crisis can last from a few hours to a few weeks depending on the severity of the form of disease (NHS, 2006) and can be frequent and unpredictable. Consequently, frequent hospitalisations, increased risk of bacterial infection, chronic organ damage and reduced life expectancy all become part of life for the sufferer (Idemudia et al., 2010). The physical symptoms of the disease can have acute or chronic effects on the body. This and the unpredictable nature of the disease can affect psychological processes associated with disease management.

The most common form of SCD is sickle cell anaemia HbS/S which accounts for approximately 70% of SCD presentations (Rees et al., 2010). Other forms of SCD include different strains of sickle cell anaemia and numerous thalassaemia strains (Rees et al., 2010). Thalassaemia is a form of sickle cell disease where the red blood cells do not contain sufficient haemoglobin to transport oxygen through the body (Anie & Massaglia, 2011). There can be an overload of iron compounds in the body's systems (Anie & Massaglia, 2011), however, thalassaemia is not predominantly identified by pain experience (Rees et al., 2003), whereas acute, chronic and/or recurring pain (vaso-occlusive crises) is a significant symptom of

sickle cell anaemia (Rees et al., 2003; 2010). It is reasoned that the longevity and the life-threatening nature of SCD can affect the perception and management of the disease.

Aetiology

The disease manifests from as early as six months after birth and is caused by the inheritance of a single sickle haemoglobin recessive gene from both parents (Anie, 2005). The sickle cell gene is thought to have evolved as an evolutionary adaptation against malaria and is subsequently prevalent on the African and Asian continents where malaria is prevalent and in the African and Asian Diaspora that originated from these continents (Anie & Green, 2012; Idemudia et al., 2001; Rees et al., 2010). It is believed that the advantage of being a carrier of the gene (having one copy of the gene, compared to two copies of the gene as in those with SCD) is that the presence of sickle cells in a smaller proportion of the blood in gene carriers (and in SCD to a lesser extent) prevents malarial parasites from being able to populate these cells (Rees et al., 2010). Therefore, the malarial parasites are unable to spread and populate adjacent blood cells subsequently killing the host by causing strokes, by preventing the transport of oxygen through the body or by killing red blood cells (Idemudia et al., 2001). Nigeria is thought to have the largest population with SCD in the World (Anie et al., 2010; Taylor, Stotts, Humphreys, Treadwell & Miaskowski, 2010), but the disease is also prevalent in other African countries, in Saudi Arabia, the Mediterranean, the Caribbean, India and the U.S. (Idemudia et al., 2001) and exists worldwide due to migration.

Disease Management

Disease management primarily involves medication and regular medical monitoring to reduce the likelihood of blood clot formation and anaemia (Rees et al., 2003; 2010). Folic acid, antibiotics, analgesics, hydroxyurea (a form of chemotherapy), blood transfusions and anti-coagulant medication for blood clots are the usual methods of managing the disease (Cummins & Anie, 2003; Rees et al., 2003; 2010). Pain medication and antibiotics are administered frequently for managing pain and reducing the risk of infection, respectively (Adam, Telen, Jonassaint, De Castro & Jonassaint, 2010). Bone marrow transplants in children are the only cure for SCD and the procedure is rarely used due to a lack of compatible

bone marrow donors and the high risk surgery complications associated with the procedure (Rees et al., 2010). Psychological therapy may also be provided to assist with managing the illness (Anie & Green, 2012; Cummins & Anie, 2003; Thomas et al., 1999).

Treatment for SCD is a combination of medical and psychological interventions. Of the limited studies of the different types of psychological interventions for pain in SCD, the cognitive-behavioural strategies are the most thoroughly researched (Anie & Green, 2012). SCD patients have been found to have high levels of anxiety and depression (Levenson et al., 2008; Thomas et al., 1998) and low levels of self-efficacy and illness identity (Idemudia et al., 2001; Thomas et al., 1999). In SCD, active pain management is very important outside of hospitals as approximately 90% of emergency hospital admissions in SCD are for managing pain crises (Thomas, Hambledon & Serjeant, 2001). Therefore, developing pain management strategies may help to prevent hospitalisation for pain. In addition, management strategies that avoid negative thinking about pain and avoid passive coping have been shown to predict better pain outcomes in the disease (Anie et al., 2012; Midence & Elander, 1994). Negative emotional responses to pain have been found to be associated with impaired quality of life, higher levels of distress and anxiety and more frequent hospitalisations (Anie, Steptoe & Bevan, 2002; Anie et al., 2012). Highlighting the psychological indications of the disease helps to clarify how psychological factors can affect the physical factors of the disease. There may be psychological differences between children, adolescents and with adults with SCD as these different populations may be affected by differences in age and maturation-related processes. Therefore, it is important to focus potential research on a specific age group: across age-groups, or over the lifespan cohorts, rather than as a combined group.

Investigating Psychological Differences in Adults

The decision to focus solely on adults in the current study was informed by the participant bias of past academic studies. There are comparatively fewer academic studies examining psychological variables or interventions in adults with SCD; the majority focus on children and adolescents (Anie, 2005; Burlew et al., 2000). One of the reasons suggested for this literature imbalance was that

previously, patients with SCD were likely to die before the age of 30 (Platt, Thorington, Brambilla, Milner & Ross, 1991), therefore research was conducted with the age groups that would benefit from subsequent psychological interventions i.e. children and adolescents. Recent reviews of life expectancy in SCD (Claster & Vichinsky, 2003; Taylor et al., 2010) have reported that the average life expectancy for a person with SCD has increased to between approximately 42 to 50 years of age for both men and women. The life expectancy has increased partly because treatment and disease management strategies have improved (Rees et al., 2010) and are more readily available (Rees et al., 2003; 2010). The rationale for only selecting adult participants for this study emerged from the need to contribute to the psychological literature on adults with SCD.

There are psychological consequences of having relatively low life expectancies in SCD. Taylor et al. (2010) reported that the average life expectancy for a person with SCD was 42 years for women and 48 years for men in the US. Claster & Vichinsky (2003) identified the life expectancy as being 50 years for both men and women. However, the average life expectancy in the UK is 77.7 and 81.9 years of age for men and women respectively (Office of National Statistics, 2010); and 75.6 and 80.8 years of age for men and women in the U.S respectively (United Nations, 2006). These figures show the contrast in life expectancy for the person with SCD. This and other factors e.g. frequent hospitalisation for pain crises (Anie et al., 2012) and psychological distress caused by the disease (Citero et al., 2007; Levenson et al., 2008; Thomas, et al., 2001) can affect pain experience and management in adults (Elander & Midence, 1996; McClish et al., 2005; Smith et al., 2005) and affect receptiveness to medical and psychological treatment (Caird, Camic & Thomas, 2011; Jenerette, Brewer & Ataga, 2014). The presence of these factors indicates a specific need to develop and improve the psychological understanding of disease management in SCD and this can be achieved by focussing on the psychological aspects of SCD, which was the intention of the current study.

Current Study

The current study employed a two-phased sequential explanatory mixed methods design (Creswell & Plano Clark, 2006) to explore the nature of the

relationship between psychological factors (e.g. trait anxiety) and how they affect pain management in SCD in adults. The study aim was to contribute to the psychological literature on adults with SCD in order to develop a greater understanding of psychological variables relevant to pain management in SCD. This was achieved by focussing on the role of trait anxiety in pain management. In addition, as the study examined the effect of trait anxiety in SCD, it was important to study a demographic that had more stable and identifiable traits i.e. in adults, rather than in children and adolescents who were still developing cognitively and emotionally.

This thesis continues by discussing the contribution of this study to counselling psychology and discussing the epistemological framework of the study. The literature review discusses theories of pain management in SCD, pain management and trait anxiety studies relevant to this study and how these past studies have informed the development of the research questions in this study. The methodology chapter reports the mixed-methodology used to investigate and explore the effect of trait anxiety in SCD. The two subsequent chapters report both the quantitative and qualitative analyses separately. The findings reported in the quantitative and qualitative chapters are triangulated and critically discussed in the discussion chapter, where limitations of the study and implications for future research are also discussed.

Contribution to Counselling Psychology

The current study hypothesised that high levels of trait anxiety manifested differently in SCD compared to another ill group. Rather than trait anxiety being perceived as maladaptive, as previous research has suggested in SCD (Thomas et al., 1998; 1999) and in other general chronic pain conditions (Tang et al., 2009; Van Esch, Roukema, Van der Steeg & De Vries, 2011), I suggest that high levels of trait anxiety are adaptive and are possibly advantageous in SCD given that elevated trait anxiety may facilitate disease monitoring and pain management. It is also suggested that high trait anxiety in SCD may affect the efficacy of psychotherapeutic interventions, CBT in particular and such a suggestion warrants further research in future studies. Investigating and exploring the former suggestion could add to the knowledge about the role of trait anxiety in SCD and help to understand how to

incorporate this role in psychological interventions for pain management. It is tentatively suggested that using CBT to manage anxiety and depression in pain management for SCD may be counter-productive if the role of trait anxiety is not understood properly. To explain further, it may not be helpful to reduce hyper-vigilant behaviour (measured by trait anxiety), through CBT interventions for anxiety, in people who use hyper-vigilance to monitor and manage their pain. Using hyper-vigilance to monitor and manage pain could be considered to be advantageous in a person experiencing chronic or recurring pain. Understanding how trait anxiety and its relationship with health outcomes (hospital admission frequency, depression, quality of life and pain intensity) impacts on pain management in SCD may help healthcare providers treat SCD more effectively and may inform the debate about the importance of pain monitoring and hyper-vigilance behaviours in SCD. This may be of particular interest to counselling psychologists and other practitioner psychologists who practice in pain management. It is important to clarify the perspective of this current study, as this will help the reader to understand its premise; this is discussed in the following section.

Epistemology

Counselling psychologists are trained as scientist-practitioners (Blair, 2010); the implications of which means that counselling psychologists, like other practitioner psychologists, are often negotiating conflicting ideologies, frameworks and paradigms due to the multi-disciplinary nature of their work and the union with the different counselling frameworks that emphasise subjective meaning and interpretation (Gil-Rodriguez & Hanley, 2011). Kasket (2012) argues that counselling psychologists are unique in the applied psychological sciences for having a pluralistic approach to research and its applications; that the counselling psychologist researcher has to be open to exploring all the paradoxes and research paradigms available in order to explore research questions in a manner that is akin to the profession's philosophy. Gil-Rodriguez & Hanley (2011) reflected that contrary to Kasket's (2012) aforementioned opinion, the majority of the contributions to the *Counselling Psychology Review*, a British Counselling Psychology journal, were qualitative paradigms. This suggests that there is a subconscious drive to encourage qualitative worldviews in counselling psychology. Despite this subconscious methodology drive, counselling psychologists are able to use quantitative, qualitative

and mixed-method methodologies to answer research questions. These research methodologies are determined by philosophical perspectives (Plowright, 2011) i.e. post-positivist, social constructionist, advocacy/participatory or pragmatic perspectives (Creswell, 2009). According to Blair (2010) and Kasket (2012), research methodology decisions should not be limited to personal philosophical perspectives, but rather should be determined by identifying the best approach to explore and/or examine the research question.

The traditional bipolar paradigms of positivism and constructivism (Plowright, 2011) lend themselves to quantitative and qualitative approaches respectively, and the remaining paradigms sit at varying points between the two. Positivism is the absolute realist perspective of the world (Plowright, 2011) which has evolved to post-positivism, which assumes that outcomes can be influenced by changing variables and acknowledging that there is no absolute truth (Creswell, 2009). The post-positivist view is, however, reductionist and includes empirical observation and measurement and theory verification (Robson, 2002). In contrast, the social constructivist view claims that reality is dependent on the individual and is subsequently constructed through social experience (Plowright, 2011). Robson (2002) adds that social constructivists assume that individuals seek understanding of the world that they live in by developing subjective meanings of their experiences. This view is about understanding multiple participant meanings, social and historical construction and generating theory (Creswell, 2009). Kasket's (2012) vision of the counselling psychologist researcher was that rather than focussing on research methods, the researcher should prioritise finding the best method of answering a research question by using both approaches in one study. Whilst this idea is noble, practicalities of agendas, time constraints and transferability can make combining the approaches difficult to achieve in reality (Howe, 1988; Plowright, 2011; Tashakorri & Teddlie, 1998). Merging the two paradigms can be achieved by critically evaluating a realist perspective (Bhaskar, 1998; Howe, 1988).

Traditional perspectives of realism are grounded in positivism, but modern perspectives of realism are positioned between post-positivism and social constructionism. Realism shares features with both of these paradigms, of which there are two forms: empirical realism and critical realism (Bryman, 2008). Empirical realism proposes that if researchers use the appropriate methods to study the social

world, they can really understand what reality is. In contrast, critical realism argues that there are differences between objects and the terms used to describe and understand them (Bhaskar, 1975). Considering both these stances, it could be suggested that critical realism acknowledges that there are real objects in the world that are perceived and utilised individually, whereas empirical realism suggests that all objects, even those perceived subjectively can be measured and observed if the correct methodologies are used, thus lending itself to reductionism. It can thus be deduced that studies embracing the realist philosophy employ qualitative and/or quantitative methods.

A critical realist philosophy guided the formulation of this study's objective and methodology. Bhaskar (1975; 1998) suggested that critical realist research organically identifies a research question that can be observed and measured in reality, but requires an exploration of subjective phenomena to hypothesise mechanisms that can be used to explain certain outcomes. This current study follows a perspective that rejects the traditional dualisms; constructivism did not allow for variables to be observed, which was necessary to suggest a relationship between variables i.e. to measure trait anxiety as a specific type of anxiety and to show that a relationship existed between trait anxiety and health outcomes in SCD. However, empiricism did not allow for the interpretation of subjective meaning, which is an important social construct and part of human experience and of relating to the world (Stone & Elliott, 2011). Being able to explore human experience was particularly relevant to this study – it was important to hear the participants' voices and how they experienced their illnesses; the experience of chronic pain and chronic illness is an extremely personal one and different phenomena can emerge from this type of research question. Insight into subjective phenomena is invaluable and cannot be explored without prior indication or insight. Robson (2011) confirmed this idea by positing that the world cannot be reduced to absolutes because it can also be constructed individually, for which there are an infinite number of experiences or responses. The critical realist position understands the philosophical integration of objective and subjective meanings in a method that lends itself well to the philosophical underpinnings of counselling psychology (BPS, 2005) and the generation of this current study was based on this ontology.

In summary, this chapter reported the study's objective, the epistemological framework of this study and the importance of investigating psychological variables that may influence or affect pain management in SCD. It was important to establish the framework of the study and show the perspective and intention with which the researcher followed to identify and attempt to answer the research questions. The next chapter will review theories and research on pain management in SCD. The research objectives, hypotheses and questions can be found at the end of the literature review.

Chapter 2: Review of the Literature

This chapter presents the relevant literature on SCD and pain management. The chapter reviews psychological aspects shown to be related to pain management in SCD and identifies trait anxiety and its relationship to certain health outcomes as a psychological construct that requires further investigation in SCD. There are no other identified studies specifically investigating or exploring the relationship of trait anxiety with health outcomes in the management of pain in SCD. Subsequently, this study proposed that investigating this set of relationships is important because high levels of trait anxiety may represent an adaptation to monitoring physical symptoms of the disease (through hyper-vigilance), thus enabling SCD patients to prevent or prepare for painful crises, subsequently managing their pain. This chapter is organised to show the relevance of investigating trait anxiety and its relationship with pain management in SCD and concludes with the objective of the study and the research questions that were investigated and explored in this study.

This review is based on published psychological quantitative and qualitative literature. Electronic searches of medical and psychological databases (Academic Premier, Cinahl+, Embase, Psychinfo, Psychlit, PubMed and The Cochrane Library) were conducted mainly focussing on SCD-specific papers and these key search terms: trait anxiety, mood, pain, pain management and quality of life. The inclusion criteria included English language publications and publications from and including 1992 to the present (2014). Papers were excluded if they were not reports or reviews. The focus of this study was adults with SCD; therefore, whilst this review does not specifically discuss studies concerning children and adolescents, a few studies concerning this age group have been referenced where they support or

disagree with a critique in the review. There are variations in the design, consistency and rigour of the findings presented in this review, therefore the findings are collated, criticised and presented tentatively. The content of this review includes sub-sections on pain theories, pain management research and psychological variables specific to SCD. This chapter concludes with a summary of the literature review addressing the knowledge gap highlighted by the literature review and with the research questions of this thesis.

Theories of Pain used in SCD Management

The experience of acute and chronic pain in SCD is a complex subjective and multidimensional experience influenced by biological, psychological, environmental and sociological factors (Anie et al., 2002; Elander & Midence, 1996; Gil et al., 2004; Taylor et al., 2013). Acute pain is defined as pain that lasts six months or less (Ogden, 2012) and can be managed through hospital treatments or medical therapy (British Pain Society, 2010; Rees et al., 2010). Chronic pain, in contrast, lasts longer than six months and can vary in severity, or become increasingly worse due to worsening physical conditions (Jenerette & Lauderdale, 2008; Rees et al., 2010).

The function of pain is to provide constant bio-feedback about the body's condition (Ogden, 2012) enabling adjustments to be made to either correct or maintain the body's condition. Pain has been defined as an unpleasant reaction in the body caused by illness and/or injury (Melzack & Wall, 1965). The definition of pain also incorporates emotional suffering or distress (British Pain Society, 2010). The experience of pain triggers help-seeking behaviour (Anie et al., 2012; Gil et al., 2004; O'Connell – Edwards et al., 2009) for the biological causes of pain e.g. by attending hospital for medical treatment; the experience may also generate anxiety (Forand & DeRubeis, 2013; Thomas et al., 1999) and distress (Citero et al., 2007; Howard, Anie, Holdcroft, Korn & Davies, 2005; Thomas et al., 2001; Wellington et al., 2010). Pain can be caused by anxiety, or anxiety can cause pain (Linton & Shaw, 2011). This complex relationship between anxiety and pain can influence the perception of pain and can increase pain sensitivity because of increased hyper-vigilance of pain (Crombez et al., 2004) or because of a lower tolerance of pain (James & Hardardottir, 2002).

Relevant literature on chronic pain has incorporated psychological theory in pain models (De Vries, Van der Steeg & Roukema, 2009; Min et al., 2013; Salthouse, 2012; Tang et al., 2009). Melzack & Wall (1965) developed the gate control theory of pain (GCT) to demonstrate how using psychology improved the concept and understanding of pain experience. The model hypothesises that a gate exists at the level of the spinal cord which receives sensory input from peripheral nerve fibres, from descending central influences from the brain e.g. emotions, attention or self-efficacy; and from other large and small nerve fibres in the body (Brannon, Feist & Ipdegraaf, 2013). Melzack & Wall (1965) also suggested that the gate integrates the received information and sends information to the action system in the body, which results in pain perception (Ogden, 2012). The model suggests that whilst pain may be organic in origin, the management of pain requires a combination of physical and psychological assessment (Brannon et al., 2013); the model also suggests that the pain patient is active in their experience of pain as they are actively interpreting and appraising painful stimuli (Ogden, 2012) and this process may affect pain perception.

The gate is thought to be opened by physical factors (e.g. injury), emotional factors (e.g. anxiety, depression) or behavioural factors (e.g. hyper-vigilance) (Melzack & Wall, 1965). Factors thought to close the gate and decrease pain are physical factors (e.g. medication), emotional factors (e.g. happiness, increased self-efficacy) or behavioural factors (e.g. relaxation, distraction) (Melzack & Wall, 1965). This model has been criticised for assuming that pain is purely organic in origin (Ogden, 2012), however, this model may be more useful in certain communities where beliefs about origins of disease and pain conflict with the uptake of appropriate pain management for psychological factors (Anie, Dasgupta, Ezenduka, Anarado & Emodi, 2007; Asnani, Fraser, Lewis & Reid, 2010; Dennis-Antwi, Culley, Hiles & Dyson, 2011; Idemudia et al., 2001). In addition, the GCT model (Melzack & Wall, 1965) posits that hyper-vigilance behaviour (i.e. excessive monitoring and awareness of areas of pain) increases the perception of pain, but does not allow for a possible relationship between hyper-vigilance and self-efficacy in the management of pain. Alternatively, it could be suggested that being able to detect and subsequently change the course of pain precipitators in certain pain conditions could improve a sense of personal control over pain, thus leading to increased self-

efficacy. Melzack & Wall (1965) posited that increasing self-efficacy would reduce pain perception. This paradox highlights an area in pain management that requires further investigation.

Variants of this bio-psychosocial model of pain (Melzack & Wall, 1965) have evolved over the years (Linton & Shaw, 2011) and have been adapted for disease-specific pain management models. There are four disease-specific models of pain in SCD which evolved from Melzack and Wall's (1965) gate control theory of pain. These are the health beliefs in SCD model (Leavell & Ford, 1983), the biomedical model (Maxwell, Streetly & Bevan, 1999), the conceptual explanatory model of pain and utilisation over time in SCD (Smith et al., 2005), and the bio-psychosocial-spiritual model of chronic pain in SCD (Taylor et al., 2013). All these models share the idea that psychological factors can influence pain perception. Where they differ is in the extent and depth at which the models include psychological factors.

Smith et al.'s (2005) and Taylor et al.'s model (2013) both emphasise biological, psychological and sociological factors equally and in more depth compared to the other two models. Smith et al.'s model (2005) suggests that disease-related aspects, psychosocial factors and readiness variables combine to explain pain perception and experience over time in SCD. Smith et al.'s model (2005) focuses on how these variables influence healthcare utilisation, rather than the experience of pain, but the model does demonstrate clear directional relationships between the variables. Taylor et al.'s (2013) model specifically highlights spirituality in their model, however, they also identified that spirituality could be included in the social aspect of the model as a coping mechanism or factor, rather than as a separate variable. Quantitative and qualitative studies (Jenerette & Lauderdale, 2008; O'Connell-Edwards et al., 2009) both support the addition of spirituality to Taylor et al.'s (2013) model. Smith et al.'s (2005) model does not specifically focus on spirituality compared to Taylor et al.'s (2013) model, but it does clearly demonstrate and provide evidence for how the different elements of the model are linked to each other compared to the latter model. Several published academic papers have used and provided evidence for the conceptual explanatory model of pain and utilisation over time in SCD (Citero et al., 2007; Levenson et al., 2007; 2008; McClish et al., 2005; 2006; 2009; Smith & Scherer, 2010; Sogultu et al.,

2011). The next section will look at pain management research in SCD in relation to Smith et al.'s (2005) bio-psychosocial model of pain in SCD (see figure 1).

Pain Management in SCD

The experience of acute and/or chronic pain has been described as the hallmark feature of SCD (Benjamin, 2008; Rees, 2003; 2010). Disease-related variables (e.g. disease-type, co-morbidity, pain location) are known to be the primary causes of pain perception in SCD (McClish et al., 2009; Rees et al., 2003; 2010; Smith et al., 2005; van Tuijin, van Beers, Schnog & Biemond, 2010).

Pain in SCD is known to be affected by: anxiety (Anie et al., 2012; Thomas et al., 1999), depression (Asnani et al., 2010; Grant, Gil, Floyd & Abrams, 2000; Hasan, Hashmi, Alhassen, Lawson & Castro, 2003; Laurence, George & Woods, 2006), distress (Citero et al., 2007; Howard et al., 2005; Wellington et al., 2010), quality of life (Adam et al., 2010; Anie, 2005; Asnani, Lipps & Reid, 2009; Gibson et al., 2013; Thomas et al., 2001), fatigue (Ameringer & Smith, 2011), coping (Caird, Camic & Thomas, 2011; Edwards et al., 2006; Jonassaint, Jonassaint, Stanton, De Castro & Royal, 2010), social life and relationships (Jenerette, Leak & Sandelowski, 2011; Thomas & Taylor, 2002; van Tuijin et al., 2010) and somatisation (Grant et al., 2000; McCrae & Lumley, 1998; Sogultu et al., 2011; Wellington et al., 2010). The following sections review the literature on these psychological variables and demonstrate specific gaps in the literature; some of which are included in the focus of this current study.

Mood in SCD

Pain Catastrophising

One of the well studied aspects of mood in SCD is pain catastrophising. Pain catastrophising is a negative affective reaction in response to anticipated pain based on previously experienced pain (Citero et al., 2007). Increased pain catastrophising has been associated with increased anxiety and depression (Citero et al., 2007; McCrae & Lumley, 1998). Pain catastrophising has also been associated with greater perceived pain intensity and more frequent use of medical services in SCD due to increased somatic awareness (McCrae & Lumley, 1998). McCrae & Lumley (1998) examined the relationships between somatic awareness and illness worry

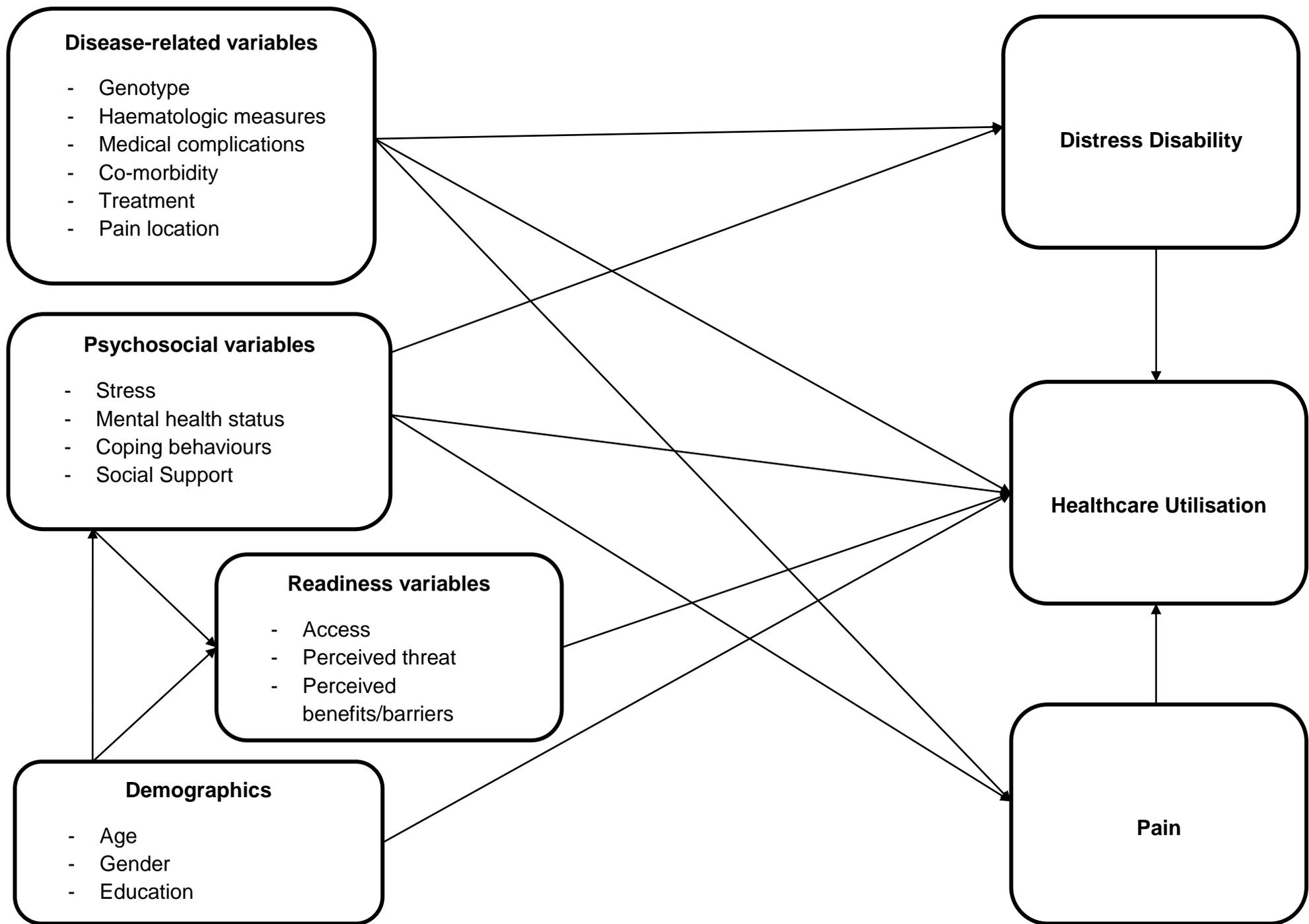


Figure 1: Draft Conceptual Explanatory Model of pain and utilisation over time in SCD

with pain severity and hospitalisation frequency in SCD and reported that 61.3% of the total variance in pain intensity in their sample was attributed to negative thinking and passive adherence in SCD. This finding indicated that catastrophising and passive coping (e.g. relying on others for support) exacerbated pain perception. In contrast, Citero et al. (2007) investigated the role of catastrophising in pain and found there were no significant differences between high-scoring catastrophisers and low-scoring catastrophisers in terms of pain intensity, medical service use or pain frequency. This finding implies that catastrophising does not affect the physical perception of pain in SCD. Citero et al. (2007) were not able to generalise the findings to other pain conditions; however the median catastrophising score in SCD was reported to be significantly higher than in other chronic pain conditions (Citero et al., 2007). Citero et al.'s (2007) finding differs from the relationship between catastrophising and pain intensity in other chronic pain illnesses e.g. in fibromyalgia (McDermid, Rollman & McCain, 1996) where higher catastrophising scores were related to increased perceived pain. Citero et al.'s (2007) finding suggests that catastrophising may have a different relationship with pain in SCD compared to other chronic pain illnesses. One explanation for this could be attributed to the nature of the longevity of SCD (from birth) and the life-threatening nature of the disease. This finding is an example of how a psychological construct can have different relationships with other psychological variables in SCD compared to with other chronic pain illnesses. Pain catastrophising is known to be related to negative thinking and depression (Barbarin & Christian, 1999) and considering how depression affects pain and distress in SCD may provide insight into pain management.

Depression

There is co-morbidity of depression in SCD. Levenson et al. (2008) reported that 27.6% of their SCD sample ($N = 308$) suffered from depression and 6.5% had an anxiety disorder, as measured by the *Patient Health Questionnaire*, a screening and diagnostic tool for depression, anxiety and somatisation (*PHQ*; Spitzer, Kroenke & Williams, 1999). Using a widely used psychometric tool allowed for depression incidence comparisons to be made across SCD studies. The aim of Levenson et al.'s (2008) study was to investigate the impact of anxiety and depression on healthcare utilisation, quality of life and medication use in SCD. The study reported that the

depressed participants experienced pain on significantly more days (71.1%) compared to the non-depressed participants (49.6%), however, pain was measured through pain diaries rather than through a standardised pain measure e.g. the *Short Form Health Questionnaire (SF – MPQ; Melzack, 1987)*. Using pain diaries instead of standardised pain measures decreased the reliability of the pain reports in Levenson et al.'s (2008) study as these pain diaries had not been validated and thus, it would be difficult to replicate their findings.

Asnani et al. (2010) investigated the co-existence of depression with loneliness in SCD in age and sex-matched non-SCD controls. Depression was found in 21.6% of the SCD sample ($n = 277$) and in 9.4% of the controls ($n = 65$). These participants were screened using the *Beck Depression Inventory II (BDI-II; Beck, Steer & Brown, 1996)*, however, the *BDI-II* is a depression screening tool and not a diagnostic tool, so conclusions about depression should have been suggested tentatively, rather than assertively. Asnani et al. (2010) also reported that depression in SCD was significantly associated with frequent hospital visits (based on the mean hospital visits of the SCD sample) and with frequent pain crises. Although, we would expect loneliness to be significantly associated with depression, Asnani et al.'s (2010) study reported that loneliness was significantly associated with unemployment and higher education attainment, rather than with depression in the SCD participants. It would have been interesting to subjectively explore the experience of loneliness in depressed SCD participants and in depressed non-SCD controls to allow similarities and differences in loneliness to emerge from the data to provide better insight into what causes depression in SCD.

The *BDI-II* (Beck et al., 1996) was used to screen for depressive symptoms in SCD during routine visits to an SCD clinic in Hasan et al.'s (2003) study. Hasan et al. (2003) reported a depression incidence of 44% ($N = 50$) in their sample, which was relatively high compared to the incidence rates reported previously by Asnani et al. (2010) and Levenson et al. (2008). The difference between the three SCD samples was the sample size; Hasan et al.'s (2003) sample was comparatively smaller ($N = 50$) compared to the other two studies – ($N = 342$) and ($N = 308$) respectively. Hasan et al.'s (2003) small sample size may not have been statistically powerful enough to reject their hypothesis had it been false. Hasan et al. (2003), however, also found significant associations between higher depression scores and poor pain control,

inadequate social support, low socioeconomic status and in being female. Whilst these depression studies report an association between hospital frequency, perceived pain and depression in SCD, only Levenson et al. (2008) measured how anxiety was related to depression and pain management in SCD. SCD studies that have measured anxiety and depression together have generally reported these variables together and termed them as the variable 'distress'.

Distress

Distress is a psychological variable that combines extreme depression, anxiety and emotional pain (Thomas et al., 2001). Distress was not measured by a specific standardised measure in Howard et al.'s (2005) study which investigated the extent of cannabis use for pain relief and the relief of emotional distress in SCD. However, Howard et al. (2005) reported that 39% ($N = 86$) of their sample used cannabis specifically to relieve their distress (self reported anxiety and depression), compared to 52% ($N = 86$) who used cannabis for pain relief. These findings indicated that anxiety and depression were secondary factors to managing perceived pain in SCD. This is an important implication, because it suggests that anxiety and depression may not be problematic factors in SCD if pain is controlled i.e. anxiety and depression may be directly influenced by pain experience. However, Howard et al.'s (2005) study was cross-sectional; therefore the results of the study could not imply causation and could only indicate avenues for further research.

Culture has also been found to be influential in the expression of distress in SCD. Thomas et al. (2001) investigated psychological distress and coping across a UK and a Jamaican SCD cohort using the *STAI-T* (Spielberger et al., 1983) to measure distress – the *STAI-T* traditionally measures trait anxiety and does not depression. Thomas et al. (2001) reported that the UK cohort had higher trait anxiety scores ($M = 52.0$, $SD = 12.4$) than the Jamaican cohort ($M = 41.5$, $SD = 9.9$). The Jamaican cohort reported less distress, lower perceived levels of pain and less frequent medical service use. Thomas et al. (2001) suggested that the Jamaican community had a more integrated sense of belonging that may have contributed to the Jamaican cohort experiencing less distress related to their perceived pain than the UK cohort and suggested that experiencing more control over pain would lead to lower trait anxiety in their sample. The focus on anxiety and distress in SCD is not as

prominent as the focus on depression and catastrophising in SCD. There appears to be a gap in the literature that examines the effect of anxiety in SCD and its role in perceived pain.

Anxiety

Whilst SCD literature does suggest an association between anxiety and perceived pain in SCD, the literature is limited in the diversity of variables assessed. As mentioned previously, Levenson et al. (2008) reported that 6.5% ($N = 308$) of their SCD sample met the threshold for an anxiety disorder (as measured by the *PHQ*, Spitzer et al., 1999), but the specific diagnoses of anxiety were not reported. Levenson et al. (2008) reported that the anxious SCD participants experienced more perceived pain, more distress and lower quality of life as measured by the *Short Form Health Outcome Survey* (SF – 36, Ware & Sherbourne, 1992) compared to the non-anxious SCD participants. Only 6.5% of the participants in Levenson et al.'s (2008) study registered as having anxiety problems compared to the 27.6% who met the threshold for depression. We would expect a similar percentage of incidences for both anxiety and depression given the large sample size and the similar occurrences of both anxiety and depression in general pain literature (Eccleston, Crombez, Aldrich & Stannard, 2001; Linton & Shaw, 2011), but Levenson et al.'s study (2008) did not reflect this. Both depression and anxiety were also measured using the *PHQ*, which may not have been sensitive enough towards anxiety (Spitzer et al., 1999), therefore not providing a true measure of the incidence of anxiety in their sample.

Anxiety and depression in SCD have often been studied together. Anie et al. (2012) investigated the association between perceived pain, mood (anxiety and depression) and quality of life during hospitalisation for vaso-occlusive crises. Their study reported a cross-sectional directional relationship between high levels of perceived pain, reduced mood and low quality of life which improved during the hospital stay of SCD participants. One criticism of this study is that it examined participants in hospital for acute pain treatment. This alone biased the study, because the participants were experiencing high levels of perceived pain at the time of assessment, which may have influenced how they perceived their mood and the perception of their quality of life whilst in the hospital environment. Future studies could examine participants who are not hospitalised at the time of assessment and

who will be, therefore, better able to provide more accurate accounts of their general perceived pain.

Separating the different types of anxiety into state and trait anxiety may also be beneficial to future studies. Leavell & Ford (1983) were one of the first SCD researchers to investigate the role of anxiety in SCD. Leavell & Ford (1983) investigated if anxiety mediated vaso-occlusive crises in SCD using the *STAI* (Spielberger et al., 1983). Their findings ($N = 16$) did not support their hypothesis that higher levels of anxiety mediated vaso-occlusive crises. This may have been due to their low sample size, which would have reduced the ability of the study to reject the null hypothesis (Coolican, 2001; Field, 2013). Leavell & Ford (1983) reported a trait anxiety score mean ($M = 40.3$) and a state anxiety score mean of ($M = 38.4$), which were scores that were below the clinical cut-off score of 44 (Spielberger et al., 1983) and indicated that their SCD participants did not have clinical levels of trait anxiety. Leavell & Ford (1983) did not report standard deviations and doing so would have shown the range of trait and state anxiety scores across their sample, which may have also shown that some participants may have experienced clinical levels of trait anxiety.

A more recent study by Thomas et al. (2001) reported trait anxiety scores of ($M = 52.0$, $SD = 12.4$) in their London cohort and ($M = 41.5$, $SD = 9.9$) in their Jamaican cohort. The state anxiety scores they reported were ($M = 50.3$, $SD = 11.8$) in the London cohort and ($M = 42.7$, $SD = 15.2$) in the Jamaican cohort. These results indicated that the London cohort experienced higher levels of both trait and state anxiety than the Jamaican cohort. It is possible that the difference in anxiety between the two groups was related to the sense of control of pain management, where the Jamaican cohort felt more in control of their perceived pain. As shown, there are differences in types of anxiety and it is not always beneficial to a study to combine anxiety and depression as one variable as both depression and anxiety may have different associations with perceived pain in SCD. Future research in anxiety in SCD could focus on separating trait anxiety from state anxiety to reveal the associations between these forms of anxiety and pain management.

Future research could also unpack the relationship between trait anxiety and depression as the relationship is unclear in both general mood and in SCD research.

Research indicates that high levels of trait anxiety co-exist with high levels of depression in breast cancer and in SCD (De Vries et al., 2009; Thomas et al. 1998), which is an expected finding. Interestingly, Min, Lee, Lee, Lee & Chan (2012) showed that depressed participants (non-pain, nor SCD) simultaneously experienced high levels of resilience with low levels of trait anxiety and that these participants responded better to anti-depressants than depressed participants with low resilience and high trait anxiety. This was an unexpected finding, as depressed participants would be expected to have low resilience and high trait anxiety as previous research between depression and resilience in SCD has shown (Burlew et al., 2000; Caird et al., 2011; Citero et al., 2007; Thomas et al., 1998). The methodological difference between the latter findings and Min et al.'s (2012) findings is that Min et al. (2012) sampled a non-clinical and non-SCD sample rather than an SCD sample, thus again demonstrating that psychological variables may have different relationships with other psychological variables in SCD. For example, Thibodeau, Welch, Katz & Asmundson (2013) showed, using a non-clinical sample, that trait anxiety and depression were not associated with pain perception, but that anxiety sensitivity was associated with increased pain tolerance in their sample. This finding implied that mood did not affect pain perception in non-clinical samples; however, previously cited research indicates that mood does affect pain perception in clinical samples.

There is specific interest in how trait anxiety is associated to health outcome variables in other pain conditions. Ristvedt & Trinkaus (2009) found higher trait anxiety was associated with lower quality of life among rectal cancer participants in remission. Employing Watson & Pennebaker's (1989) symptom perception hypothesis, Ristvedt & Trinkaus (2009) suggested that the significant relationship between trait anxiety and self-reported physical functioning was a consequence of an underlying disposition of psychosomatic distress among the participants. Thus, participants with high levels of trait anxiety were more likely to notice negative physical functioning changes in themselves than the participants with lower levels of trait anxiety. This conclusion would suggest that trait anxiety is part of a decision-making process that involves consciously paying attention to and acting on physical changes within the body.

Trait anxiety has been implicated in decision-making processes. Peng, Xiao, Yang, Wu & Miao (2014) evaluated low versus high trait anxiety groups on self-

framing and decision-making in a sample of non-SCD university students ($N = 328$). Researchers found that the high trait anxiety group tended to make more conservative and less risky decisions relative to the low trait anxiety group (Peng et al., 2014). These authors concluded that higher levels of trait anxiety were related to risk avoidance and these levels caused an over-estimation of potential risks in participants by inducing an attention bias in the participants in the high trait anxiety group. This attention bias may have caused the participants in this group to prioritise the processing of information seen as a perceived risk to the participants (Peng et al., 2014). The researchers were able to suggest that these were causal relationships because of the experimental nature of their study; they had recalled previously *STAI-T* (Spielberger et al., 1983) assessed participants to engage in a controlled and manipulated decision-making task. Peng et al.'s (2014) findings were supported by Butler & Matthews' (1987) theory that the purpose of trait anxiety in people was to induce risk avoidance of perceived risks by over-estimating potential risks. Consequently, people with high levels of trait anxiety may demonstrate an attentional bias to risk and may also present with an increased avoidance to risk, which may be pertinent to people who experience relatively higher levels of risk consistently. In chronic pain conditions, these attentional biases to risk may mean that trait anxiety may be related to perceived pain, if pain perception is perceived as a risk.

Pain tolerance has been reported to be greater in general population participants with lower, rather than higher trait anxiety (James & Hardattordir, 2002). It was suggested that high trait anxiety fostered attentiveness to possible environmental threats that would increase the experience of pain (James & Hardattordir, 2002). This finding supports Melzack & Wall's (1965) GCT model that suggests that increased attentiveness (hyper-vigilance) increases pain perception. Although James & Hardattordir (2002) employed the *SF-MPQ* (Melzack, 1987) to study pain, their study did not separate or report group differences in sensory or affective pain. It would have been useful to know if the difference in pain perception in this study was related to sensory or affective pain as this would have informed the debate on hyper-vigilance behaviour and its interaction with pain perception i.e. if hyper-vigilance behaviour was emotionally-driven or sensory-driven.

This section clarifies that there are different forms and interpretations of anxiety and whilst there are SCD studies that quantitatively measure anxiety, it can be seen that there is a need to explore subjective anxiety in order to identify and increase awareness of the different meanings of anxiety and the relationships that anxiety has with other health outcomes in SCD.

Somatisation

Somatisation is a concept that may be related to trait anxiety due to the use of attention for observing the body. Somatisation (pathological attention to a range of bodily symptoms and conditions regardless of their cause; Sogultu et al., 2011) has been found to predict negative psychological experiences in adults with SCD e.g. depression, anxiety, resistance to treatment, (Wellington et al., 2010). Findings from Wellington et al.'s (2010) study suggested that their SCD participants had a self-monitoring bias that led them to be more sensitive to small changes in their health, especially regarding their pain intensity levels. It would have been useful if Wellington et al. (2010) had utilised a self-monitoring or a hyper-vigilance tool e.g. the *STAI-T* (Spielberger et al., 1983), to support their self-monitoring bias theory. Future research could investigate how a self-monitoring bias may affect disease management in SCD. In addition, Jenerette et al.'s (2014) mixed-method study identified a theme of 'phases and cues' from interviews with SCD participants ($N = 69$) who indicated that they (84%) received signs and cues from their body before a vaso-occlusive crisis began. It could be argued that these participants may have used hyper-vigilance behaviour to monitor and assess these cues and signs in order to prepare them to either avoid or manage the impending crisis. This is not unlike high trait anxious individuals who also experience greater expectations of anxiety than low trait anxious individuals when experiencing physiological symptoms such as pain (Eysenck, 1992).

Quality of Life

Pain catastrophising, depression, distress, anxiety and somatisation are all variables that affect quality of life in SCD. Quality of life is the general wellbeing of a person and encompasses emotional, cognitive, physical and social wellbeing (Anie, 2005; Ware & Sherbourne, 1992). Pain management plays an important role in the quality of life of a person with SCD and in other chronic illnesses (Anie, 2005;

Bennett & Nelson, 2006; Malouff, Thorsteinsson, Rooke, Bhullar & Schutte, 2008). Frequent opioid analgesic use is related to experiencing lower levels of quality of life in SCD (Adam et al., 2010), however, this finding may not be surprising if the relationship between opioid analgesic need and experience of pain are considered; general wellbeing is thought to improve when perceived pain is reduced (McClish et al., 2005). In addition, mood, general health and quality of life improve when levels of pain decrease (Anie et al., 2012). However, despite a decrease in pain levels, some patients (29%; $N = 510$) often still have residual daily pain (Anie et al.'s 2012) after hospital discharge. Chronic pain is often a way of life for people with SCD and experiencing higher levels of quality of life for these people is not synonymous with not experiencing any pain. Having a greater internal locus of control is related to higher quality of life scores and lower depression scores in SCD (Gibson et al., 2013). An internal locus of control may be more influential to quality of life in people with SCD in terms of increasing the self-efficacy in managing pain, rather than trying to reduce perceived pain.

Research has shown that affective coping (catastrophising, anger and fear self-statements, praying and hoping & isolation) is related to lower levels of quality of life in SCD (Anie et al., 2002). Anie (2005) surmised that quality of life in SCD was affected by activity and functioning and by anxiety and depression. Other research studies have supported this directional relationship (Levenson et al., 2008; Taylor et al., 2010). However, McClish et al. (2005) compared quality of life subscale scores from the *SF-36* (Ware & Sherbourne, 1992) to that of the general population and to those of other chronic disease cohorts (cystic fibrosis, haemodialysis and asthma participants) and found that quality of life did not affect emotional wellbeing in SCD; SCD emotional wellbeing scores were similar to those of the general population, but were better than the other chronic disease cohorts in that study. McClish et al. (2005) suggested that this finding could mean that the SCD participants had greater social support and used different coping mechanisms to reframe their illness compared to the other chronic disease cohorts. However, it could also be suggested that anxiety and depression may have different roles and/or relationships with quality of life in SCD and these different relationships may have mediated the more positive emotional wellbeing outcome.

The previous sections have shown that there are gaps in the literature that warrant further investigation, but also indicate that there appear to be different relationships between psychological variables in SCD compared to other chronic illnesses and to non-clinical samples. It is also important to consider how to improve the relationships between these variables so that pain management is improved and quality of life is more favourable for SCD sufferers. The next sections attempt to address this.

Psychological Strategies for Improving Pain Management

This section is of particular relevance to counselling psychologists because it relates to clinical practice and discusses some of the strategies that can be focussed on in clinical work with SCD.

Social support has been found to be an influential predictor and moderator of pain intensity, pain frequency, anxiety and depression. A qualitative study reported that access to friends and family formed the context with which participants were able to manage their disease and their subsequent pain (Caird et al., 2011). Such social relationships enable the SCD sufferer to form their own identity and establish meaning and purpose in their life, thereby improving their quality of life (Caird et al., 2011). However, social support received in SCD by significant others may not always be helpful. Edwards et al. (2006) reported that parental support offered in the context of substance abuse could lead to the adoption of less adaptive coping strategies e.g. catastrophising and to an increase in intense sensory pain experiences. Such support was modelled by the adult substance-abusers to their adult children whilst as children through social learning (Edwards et al., 2006) and was not found to be helpful in adjusting to the effects of the disease. Contrastingly, van Tuijn et al (2010) did not find that social support predicted pain experiences; they found that reduced quality of life was more likely to predict or be predicted by pain experiences than social support. This finding puts the experience of pain in SCD into perspective; the perception of pain is authentic, but other psychological variables can affect the pain experience by improving or worsening it.

The unpredictability and variability of SCD has made it difficult for sufferers to relate to others and form meaningful relationships outside of their immediate families (Thomas & Taylor, 2002). In addition, Jenerette et al. (2014) surmised that young

adults with SCD found it difficult to manage their pain and care alone and required additional assistance from family and healthcare workers, but found it difficult to ask for additional help due to the stigma associated with help-seeking in SCD. Therefore, interventions that focus on improving and developing reliable social interactions and networks may be of benefit in SCD. Qualitative studies on the meaning of quality of life and adequate social support could help to unpack the subjective perceptions of these constructs and provide more meaning of the value of these constructs in quantitative research.

Using adequate coping strategies can improve pain management. Midence & Elander (1996) suggested that the use of coping strategies was an important part of managing pain in SCD and that the use of passive coping strategies (e.g. isolating, surrendering to pain) was related to less favourable health outcomes (e.g. depression, loneliness). In addition, Anie and colleagues (2002) found that active coping (intentional efforts to minimise negative effects of pain, or trying to continue despite pain) was positively correlated to the number of pain episodes requiring hospitalisation (increased number of necessary hospital visits), whilst passive adherence coping was related to increased pain intensity. Resilience is thought to improve active coping strategies in SCD. The qualitative theme of resilience emerged as a factor that facilitated the use of more adaptive coping strategies in SCD (Caird et al., 2011). Resilience was defined as an active attempt to manage the disease by: creating meaning in pain and suffering by creating a positive purpose of having the disease; developing a personal identity independent of SCD; and by developing a sense of control over the disease by using positive coping strategies (Caird et al. 2011). This definition of resilience emerged by exploring the subjective experiences of resilience in SCD and substantiates the idea that it is important to listen to participant perspectives as well as measuring quantifiable data, because new themes specific to SCD can emerge that can inform the literature and clinical interventions.

Being hyper-vigilant and maintaining healthy behaviours have also been attributed to adaptive coping. Paying attention to bodily symptoms has been attributed to longevity (Jenerette et al., 2011) and to self-care (Benjamin, 2008; Jenerette & Lauderdale, 2008). Living beyond expectations was also found to be rewarding in SCD and increased positive quality of life perceptions (Jenerette &

Lauderdale, 2008), especially when health professionals showed surprise at the longevity of their SCD patients. Benjamin (2008) suggested that a palliative care model should be introduced in the early years of disease management in SCD and used as a model of care throughout life in SCD. It is a recognised, regular framework of intervention that has been developed to help sufferers gain more control of their lives and their pain, thus improving their overall quality of life over the lifespan (Benjamin, 2008). Marlow & Chichella (2002) reported that pain management can also be affected by adherence to medication and healthy-living, which supported the ideology of healthy-living and personal control affecting pain management.

The consistency with which patients live their lives can also improve their sense of control of their condition and improve their self-efficacy and their ability to manage their pain. Such consistency can improve mood and quality of life, both of which are reported to be predictors of pain frequency and pain intensity. Abedian, Howard, Rawle & Thomas (2010) explored what would influence adherence when taking preventative medication in SCD. Four main themes thought to influence non-compliance emerged from their study: the duration, schedule and dosage of the medication; the lack of immediate consequences of not taking the medication; difficulties in patient lifestyle and social support and physical side effects from taking medication over a long period of time. Abedian et al.'s (2010) study was interesting because it showed that despite participants knowing the benefits of taking long-term medication to prevent infections that they were more susceptible to, many preferred to take the risk of contracting an infection than to continually take antibiotics over a long period of time. Perceived risk and/or benefits of taking medication would fall into the 'readiness variables' part of Smith et al.'s (2005) conceptual model. According to the literature, perceived risk is influenced by medical complications and by types of treatment, but Smith et al.'s model (2005) does not reflect this direct link between disease-related variables and readiness variables.

Psychological support and interventions have been used to improve pain management strategies. Thomas et al.'s (1999) study examined the effect of a CBT intervention on pain management in SCD. The general results supported a CBT intervention for pain management; although the effects of this CBT intervention were short-term (baseline levels of anxiety and depression returned six months post-intervention). One suggestion for this post-intervention finding could be that CBT

reduces hyper-vigilant behaviours in SCD, which would increase pain perception in the long-term, thus causing a return to hyper-vigilant behaviours over time and a return to baseline levels of anxiety and depression. Contrastingly, Cummins & Anie (2003) used a cross-sectional exploratory study to investigate how quality of life, anxiety and depression and pain differed in SCD participants ($N = 36$) who attended a CBT group ($n = 21$) compared to those in a hydroxyurea pharmacological intervention group ($n = 15$). Whilst the participants in the CBT intervention group reported a higher pain frequency than the pharmacological intervention group participants, quality of life scores, hospital admission duration and psychological coping improved favourably in the CBT group compared to the pharmacological group (Cummins & Anie, 2003). Mood, however, was not improved in either intervention, which might indicate, in this study, that mood had a different causal relationship in pain management, or that the instrument used to measure mood (*Hospital Anxiety and Depression Scale, HADS*; Zigmond & Snaith, 1983) was not sensitive enough to pick up changes in anxiety and depression in the study. Alternative and more sensitive mood-specific measures e.g. *STAI-T* (Spielberger et al., 1983) or *BDI-II* (Beck et al., 1996) could be used instead of a unitary measure. The study may also have been more sensitive to mood changes if the numbers of participants had been more equal.

It is important to briefly discuss the stigma of having the disease as perceived and actual stigma about the disease impacts on pain perception, pain management and the utilisation of healthcare services (Anie et al., 2010; Elander, Lusher, Bevan, Telfer & Burton, 2004; Jenerette & Brewer, 2010; Maxwell et al., 1999; Thomas & Cohn, 2006). The stigma around having the disease partly concerns being labelled or thought of as a 'drug addict' by hospital staff, which can lead to mistrust in hospital staff and neglect in hospital care (Maxwell et al., 1999). Mistrust and neglect can lead to low mood and anxiety about hospitalisations (Jenerette, Funk & Murdaugh, 2005; Jenerette et al., 2014) and can be a barrier to seeking help for pain (Elander, Marczevska, Amos, Thomas & Tangayi, 2006; Elander, Beach & Haywood, 2011; Thomas & Cohn, 2006). Young adults with SCD avoided attending Accident and Emergency departments in hospitals when they were in pain because of the non-effective treatment they received in the past, either through hospital staffs' lack of knowledge about how to manage their illness, or through the perception that SCD

patients were 'drug addicts' (Jenerette et al., 2014). Avoiding medical attention in hospitals can be detrimental in SCD as these patients do not access the pain relief that they need during vaso-occlusive crises and may exacerbate their condition by not receiving adequate medical attention.

Other Pain-Related Variables in SCD

So far, this literature review has focussed on mood, quality of life, somatisation, psychological strategies and stigma in SCD. Other pain-related variables in SCD concern differences and personal experiences in: gender (Leavell & Ford, 1983; McClish et al. 2006; Solomon, 2010), religion (Bediako et al., 2011; Jenerette et al., 2011; Sanders, Labott, Molokie, Shelby & Desimone, 2010), culture and identity (Rouse, 2011; Royal, Jonassaint, Jonassaint & Castro, 2011), medical attention, use and adherence (Abedian et al., 2010; Jegede & Rawle, 2008; Marlowe & Chichella, 2002), addiction (Elander, Lucher, Bevan, Telfer & Burton, 2003; Elander et al., 2006; Howard et al., 2005; Shapiro, Benjamin, Payne & Heidrich, 1997), physical functionality (Jenerette & Maurdaugh, 2008; Pells et al., 2005; Swanson, Grosse, Kulkarni, 2011), professional support (Elander et al., 2011; Maxwell et al., 1999; Thomas & Cohn, 2006), barriers to help-seeking (Marlow & Chichella, 2002; O'Connell-Edwards et al., 2009), stigma (Jenerette & Brewer, 2010; Marsh, Kamuya & Molyneux, 2011) work and employment (Barbarin, Whitten, Bond & Conner-Warren, 1999; Gil et al., 2004., Laurence et al., 2006) and lastly, education (Barbarin & Christian, 1999).

As pain management is a vital part of disease management in SCD, it is important to examine it from a psychological perspective as psychological factors have been shown to influence pain management in other chronic illnesses e.g. in chronic back pain, fibromyalgia, breast and rectal cancer (Bennett & Nelson, 2006; De Vries et al., 2009; Eccleston, 2011; Linton & Shaw, 2011; Malouff et al., 2008; Ristvedt & Trinkaus, 2009). The difference between pain management and experience in SCD compared to these other chronic illnesses is that SCD is a chronic pain disease that is present from birth and the pain can be recurring and unpredictable. This chronic and frequent pain experience, added to the life-threatening nature of the disease, creates a difference in pain management

strategies required to manage the disease compared to other chronic pain illnesses that may not be as pervasive, or as life-threatening.

Summary and Conclusions of the Review

The conceptual explanatory model of pain and utilisation (Smith et al., 2005) is a model that best describes how health outcomes affect pain and disease management in SCD. There are high incidences of depression and anxiety in SCD and pain is experienced more in depressed SCD participants compared to non-depressed SCD participants. The experience of pain, depression and anxiety is not homogenous within SCD; experiences can be grouped according to culture and disease-severity within SCD, but experiences can also differ within cultures and between different disease-types. Trait anxiety is thought to be an indicator of hyper-vigilance of risk and high levels of trait anxiety are related to risk avoidance in the general population. High trait anxiety is also related to low quality of life and higher depression levels in the general population, in other chronic pain illnesses and in SCD. Social support, religion, active coping, psychological support and interventions and paying attention to body symptoms have all been reported to improve pain management in SCD. This leads us to the current study.

Current Study

On the basis of the review, it could be reasoned that there is evidence for examining psychological differences in SCD, such as trait anxiety, pain intensity, quality of life and depression. It could be argued that elevated levels of trait anxiety may have more of an adaptive function in SCD than in other chronic pain conditions due to the adaptive role of hyper-vigilance for this pain sub-group. Monitoring and being hyper-vigilant to physical body changes and to the environment can be thought of as advantageous in SCD e.g. SCD patients are negatively affected by cold and wet weather, or by air pressure changes, therefore, noticing adverse environmental and/or physical body changes as soon as they occur can help SCD patients to counteract these changes or take preventative measures to prevent experiencing vaso-occlusive crises. Being able to prevent a vaso-occlusive crisis can empower the SCD patient and increase their sense of control over their illness, which could lead to increasingly positive health outcomes (lower depression, higher quality of life levels, lower pain intensity). Yet to date, no extant research has evaluated the

possibility of elevated trait anxiety as serving an adaptive function in SCD management.

Consequently, the objective of this thesis was to investigate if trait anxiety, as a measure of hyper-vigilance, was present in SCD and if it was, the aim was to investigate how trait anxiety was related to relevant health outcomes, as identified in the literature review. A clinical comparison group and a control group were used in the quantitative phase to investigate if any relationships between trait anxiety and the health outcomes were specific to SCD. Blood cancer (BC) was chosen as a comparison clinical pain group as patients with BC undergo similar rigorous treatments for pain and also experience acute and chronic pain until they are in remission. Carers were chosen as the non-clinical control group as they are a group known for having varying trait anxiety levels (Kuscu et al., 2009; Vignola et al., 2008; Yarkin, Tamer, Gamze, Micozkadioglu & Huseyin, 2009), for which to compare trait anxiety. It was also an intention to explore subjective perceptions of pain, anxiety and low mood in order to observe if hyper-vigilance emerged as a factor in SCD and if the role of hyper-vigilance in SCD was unique to SCD. The research questions and hypotheses follow.

Research Aims and Hypotheses

Research Aim I

To determine if there is a significant relationship between health outcome variables (depression scores, pain intensity scores, quality of life scores) and trait anxiety scores across the complete sample (SCD, BC, Carers) to determine what health outcome variables will be used to examine between-group differences [SCD, non-SCD comparison group (BC) and control group (Carers)] in the subsequent analyses (see Research Aim II).

H1: Depression scores and pain intensity scores will be positively related to higher trait anxiety scores and quality of life scores will be negatively related to higher trait anxiety scores in the complete sample.

Research Aim II

To evaluate between-group differences (SCD, BC, Carers) in self-reported health outcome variable scores (trait anxiety, depression, pain intensity, quality of life) across illness group (SCD, BC, Carers) to determine whether SCD group health outcomes are significantly different from an illness control group (BC) and a non-ill control group (Carers).

H2: There will be a significant multivariate main effect for illness group (SCD, BC, Carers) on trait anxiety and depression scores. Such that the SCD group will report lower trait anxiety and depression scores than the BC group, but higher trait anxiety and depression scores than the Carers.

H3: There will be a significant multivariate main effect for illness group (SCD, BC, Carers) on sensory and affective pain scores (pain intensity). Such that the SCD group will report higher sensory pain and lower affective pain scores than the BC group, but higher sensory pain and higher affective pain scores than the Carers.

H4: There will be a significant multivariate main effect for illness group (SCD, BC, Carers) on quality of life subscale scores. Such that the SCD group will report higher quality of life subscale scores than the BC group, but lower quality of life subscale scores than the Carers.

Research Aim III

To explore how a BC participant and an SCD participant with clinical trait anxiety experience pain, anxiety and low mood in order to expand on the quantitative findings from the first two research aims by contributing to the understanding of the suggested role of trait anxiety in SCD.

Chapter 3: Methodology

The current study investigated the significance of the relationships between health outcome variables (trait anxiety, hospitalisation frequency, depression, pain intensity and quality of life scores) in SCD. The following sections will report the research design of the study and the role of the researcher. This chapter will also present information regarding study participants, materials and procedures used and will consider the ethical considerations that were necessary for this study to occur.

Research Design

Mixed-methods research has become increasingly visible in counselling psychology research as an approach that can be used to examine and explore data holistically within the same study (Kasket, 2012). The literature speaks of the approach of combining quantitative and qualitative data in a specific research design as relatively new. However, the concept and the practice of collecting quantitative and qualitative data within the same study are not new (Creswell & Plano Clark, 2006; Symonds & Gorard, 2009). Mixed-methods research has been implemented in counselling and psychosocial studies. A systematic review of mixed-methodology studies in health-related papers established that of a total of 59 papers, 24% of the papers were in the field of nursing, 19% were in psychosocial and behavioural research studies, 14% were in public health and health policy, 8% were epidemiology studies and a further 8% concerned ageing research (Pluye, Gagnon, Griffiths & Johnson-LaFleur, 2009). Hanson, Creswell, Plano Clark, Petska & Creswell (2005) showed that mixed-methodology research does have a place in counselling psychology, demonstrating that mixed-methodology has been used for: clinical assessments, determining constraints of group and individual counselling, and has been used in the counselling process and in diversity. This current study implemented this agenda to investigate and explore the relationships of certain health outcomes with the management of pain in SCD.

Sequential Mixed-Methods Framework

The intent of this cross-sectional two-phased sequential explanatory mixed methods study was to investigate the relationship between health outcomes in SCD. In the first phase, quantitative approaches were used to examine if trait anxiety could be predicted by health outcome variables. The first phase also investigated between-group differences between the health outcome variables across three illness groups (SCD, BC, Carers). The qualitative phase of the study was designed as a multiple case study that explored subjective perceptions of the health outcomes in an SCD and a BC case; both with clinical trait anxiety.

The rationale for using a mixed methods approach for this study was defined by the outcome of the literature review. The literature indicated that trait anxiety affected health outcomes, but it was not apparent if trait anxiety could be predicted

by health outcomes in the sample utilised in this study, which was a reason for its examination. It was also apparent from the literature that different relationships may exist between the health outcomes and the different illness groups. However, reasons for these differences were not thought to be fully addressed by using quantitative methods alone and consequently qualitative methods were used to explore subjective perceptions specific to a SCD participant and a BC participant. The current study aimed to investigate and explore any relationships that existed between trait anxiety and health outcomes in SCD and used a clinical health comparison group (BC) and a non-clinical control group (Carers) to help to understand the implication of the findings regarding SCD.

Three issues were considered when designing this study: method priority, implementation and integration of methodology (Creswell & Plano Clark, 2006). The quantitative phase was given priority in this study because the study was focussed on examining the relationships between trait anxiety and the health outcomes and the relationship between the health outcomes across the illness groups. The quantitative phase had more focus and proportionally more time dedicated to it than the qualitative phase did, hence the priority of the study was quantitative (Brannen, 2005). However, the two case studies used in the qualitative phase allowed the study to explore subjective differences within the study, which enriched the data from the statistical analysis. The study was implemented using a two-phased sequential explanatory design (Creswell, 2009; Tashakkori & Teddlie, 1998). The first phase consisted of using an online self-report questionnaire to collect data and needed to be collected prior to qualitative data collection in order to inform and direct the qualitative phase and allow for the self-selection of clinically trait anxious participants for the second phase of the study. The explanatory strategy of the design was used to tentatively suggest alternative reasons and interpretations for the differences in health outcomes (Tashakorri & Teddlie, 1998) between an SCD participant and a BC participant with clinical levels of trait anxiety. Integration of the quantitative and qualitative phases occurred twice in this study. The first stage of integration occurred during the process of selecting participants for the case study phase. The second stage of integration occurred during the process of interpreting the findings for the whole study. A visual representation of the sequential explanatory nature of this design is presented (see Figure 2).

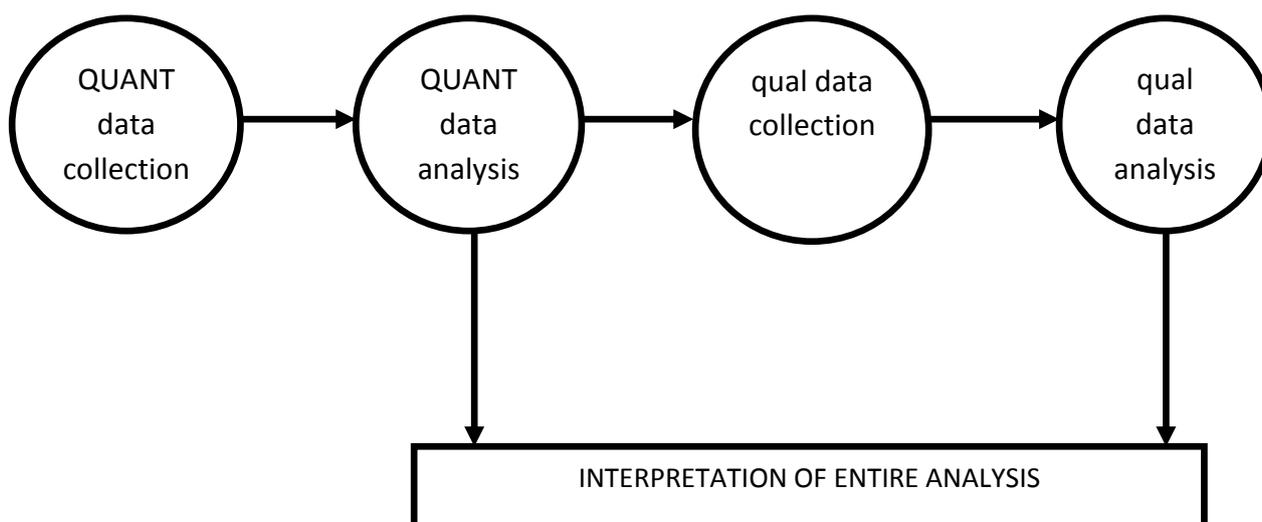


Figure 2. Visual model of the sequential explanatory framework of the present study.

Quant data collection phase = administration of questionnaires, including obtaining participant demographics; quant data analysis phase = preliminary analyses and statistical analyses leading to case selection for the qual data collection phase and to interpretation of the entire analysis; qual data collection phase = individual in-depth interviews with two participants; qual data analysis = coding and thematic analysis of transcripts; interpretation of the entire analysis = integration and triangulation of both phases of the study.

The Role of the Researcher

The principle researcher's role during the process of this study involved: applying for ethical approval to conduct the study; establishing relationships with the organisations and support groups from whom data was collected; collating research materials and collecting data; analysing the data; and discussing and disseminating the data. The researcher, with guidance from the Director of Studies, ensured that the study was conducted within ethical parameters and that the relevant skills were learned and applied in the process of data recruitment and collection and in data analysis. The researcher had the role of explaining the rationale and logistics of the study to different audiences (potential participants, support groups, conferences and workshops, and to peers and an ethics committee). Throughout the researcher's role, it was important for the researcher to remain reflexive by facilitating a critical attitude in evaluating the impact of the researcher and researcher subjectivity on the

project design, in data collection and analysis, on the research participants and in the dissemination of the findings (Finlay & Gough, 2003). Whilst researcher reflexivity is variable and indefinable to some extent, critically analysing reflexivity can be insightful in critical research and can enrich the research process (Flick, 2009).

Ethical Considerations

Ethical issues were considered during every stage of this study (BPS, 2010; BPS, 2013). Ethical approval for this study was granted by the University Of East London School Of Psychology Research Ethics Committee (see Appendix A). Subsequent approval of ethical approval amendments has also been included in Appendix A. Two separate informed consent forms were developed for the first and second phases of the study (see Appendix B). Consent was provided on the online survey by ticking 'yes' to continue, or 'no' to go to the debriefing page and exit the survey. An informed consent form was emailed back to the researcher by the participants interviewed over the telephone.

There were limitations to confidentiality. Internet Protocol (IP) addresses of the participants who participated in the quantitative phase were retained in the event that participants would decide to withdraw their data from the study and their data would need to be identified in order to do so (BPS, 2013). The IP addresses were not kept in the same dataset with other identifying information, or with the remaining data. Only the primary researcher had access to identifying information. In addition, identifying information provided by participants from the quantitative phase of the study who wished to participate in the qualitative phase was separated from the data files used for statistical analysis. The transcripts from Phase II were anonymised and Phase II consent forms were kept separately from the transcripts and the audio data (BPS, 2010).

During Phase II, it was necessary to include telephone interviews (after approval had been gained from the ethics committee) as an interview medium. Special consideration was given by being aware of emotional or physical distress in the participant during the interview and the researcher 'checked in' verbally (BPS, 2010) with the participant more frequently than would be required for a face-to-face participant.

The Phase II participants were also reminded that general and non-personified data would be kept for an additional four years, in accordance with the BPS guidelines for ethical practice (BPS, 2010).

Data Collection

Design of Phase I: Quantitative

The participants in the cross-sectional phase were asked to complete a 15-minute secure online survey (www.surveymonkey.com). The survey began by informing the participants about the nature and procedure of the study (see participant invitation letter, Appendix B). Participants were asked to provide informed consent to participate (see participant consent form, Appendix B). The following survey pages included the following standardised questionnaires (see Appendix C): the *State-Trait Anxiety Inventory (STAI-T)*; Spielberger et al., 1983), the *Beck Depression Inventory II (BDI-II)*; Beck et al., 1996), the *Short Form McGill Pain Questionnaire (SF-MPQ)*; Melzack, 1987) and the *Short Form Health Survey (SF-36)*; Ware & Sherbourne, 1992). Participants were asked to provide demographic data (e.g. age, ethnicity) and the survey concluded with a debriefing page that included helpline information for participants who required emotional and/or practical assistance (see Appendix C).

Design of Phase II: Qualitative

The participants in this quasi cross-sectional study self-referred for this phase at the end of the survey in the quantitative phase by leaving their contact details if they were interested in participating in the follow-up phase. The selection of the cases for this phase of the study was the first point of connection between the quantitative and qualitative phases in this sequential explanatory design.

Participants

Data was collected between January 2013 and January 2014. Participants were identified and sourced through purposive and snowballing sampling (Robson, 2011) from SCD, BC and Carer support groups in the UK and through the online forums and social media sites for these groups (e.g. EHF Sickle Cell and Thalassaemia Support Group, Leukaemia Care, The Sickle Cell Society and Carers

UK). Snowballing sampling is the non-probability sampling conducted by current participants who recruit future participants from amongst their acquaintances (Robson, 2002). This type of sampling assisted participant recruitment. The types of blood cancer in the BC group were (leukaemia, hairy cell leukaemia, chronic lymphocytic leukaemia, myeloma and lymphoma). The Carers were recruited through a Carer's online support website and through snowballing sampling and included Carers for SCD, BC, dementia and quadriplegia. Permission to post on forums and social media sites was provided by the administrators of the websites, who were provided with participant information sheets and consent forms prior to posting. Inclusion criteria included: adults aged 18 – 70 years; and being able to be allocated to one of the illness groups (SCD, BC, Carers). Exclusion criteria included: not being able to speak or read English at age 12 proficiency, not being able to provide informed consent, not falling within the specified included age range and not having SCD, BC or being a Carer.

Quantitative data collection

Sixty-three participants answered at least one quantitative survey question. Of those participants, twelve participants (19.7%) did not complete all the survey items and were excluded from analyses. The remaining participants ($N = 51$) answered all study questions and were included in the analyses. Sample characteristics (participant age, ethnicity and employment status are presented, see Table 1). The minimum age of the sample was 23 and the maximum age was 67. Twenty men (39.2%) and 31 women (60.8%) participated in the study. Most of the participants did not have carers ($n = 39$, 76.5%), however, six participants (11.8%) had part-time carers and five participants (9.8%) had full time carers. In addition, most of the sample was not in a romantic relationship (60.8%).

Qualitative data collection

Qualitative data was collected by purposively sampling self-selected SCD and BC participants who completed the quantitative survey and reported elevated levels of trait anxiety. Elevated trait anxiety was determined by an empirically derived cut-off score on the *STAI-T*. Scores above 44 were used to identify participants with elevated 'clinical' trait anxiety (Spielberger et al., 1983). Participants scoring below the *STAI-T* cut off were not selected for the qualitative interviews. The intention of

Table 1

Participant Characteristics

| Variable | <i>M</i> | <i>SD</i> | <i>n</i> | % |
|------------------------------|----------|-----------|----------|-------|
| Age (years) | 37.13 | 11.16 | 51 | 100 |
| Age SCD | 31.88 | 8.39 | 8 | 15.69 |
| Age BC | 50.79 | 10.68 | 8 | 15.69 |
| Age Carers | 34.42 | 7.52 | 8 | 15.69 |
| Ethnicity | | | | |
| Black African | | | 21 | 41.2 |
| Black | | | 9 | 17.6 |
| Caribbean | | | | |
| Other Black | | | 2 | 3.9 |
| White & Black | | | 1 | 2.0 |
| African | | | | |
| Other Mixed | | | 1 | 2.0 |
| White British | | | 1 | 2.0 |
| White Irish | | | 8 | 15.7 |
| White | | | 1 | 2.0 |
| European | | | | |
| Other White | | | 1 | 2.0 |
| Asian Indian | | | 2 | 3.9 |
| Asian | | | 1 | 2.0 |
| Pakistani | | | | |
| Chinese | | | 1 | 2.0 |
| Other Asian | | | 2 | 3.9 |
| Employment | | | | |
| Part time | | | 11 | 21.6 |
| Full time | | | 10 | 19.6 |
| None, unable to work | | | 12 | 23.5 |
| None, retired | | | 2 | 3.9 |
| None, student | | | 10 | 19.6 |
| None, unable to find work | | | 3 | 5.9 |

Note. (*N* = 51). SCD = Sickle Cell Disease, BC = Blood Cancer

the qualitative phase was to explore the perceptions of participants who had elevated levels of trait anxiety in order to explore differences in pain management whilst experiencing elevated trait anxiety. Subsequently, the participants who did not meet this requirement were not suitable for the interviews. Only two participants agreed to be interviewed (1 male SCD, 1 female BC). Their characteristics have been reported in detail in Chapter 5. Cross-study contamination was not an issue for this study as it was necessary to identify the participants for the qualitative phase by them completing the quantitative phase first.

Participants provided informed consent before participating in the 50 – minute semi-structured interviews (see Appendix D for the interview outline) that were audio-recorded. The participants were purposively selected (Robson, 2011). Interviews were scheduled to suit participant availability and took place individually over the telephone under conditions that maintained participant confidentiality. Topics included the self-appraisal of low mood, anxiety, pain and their perceptions of illness. The research objective was to explore these experiences in depth to elaborate on the analytical findings from phase I. No adjustments to the interview protocol were required during the interviews.

Measures

The *State-Trait Anxiety Inventory (STAI-T)* (Spielberger et al., 1983) was used to differentiate between participants with personality trait anxiety. The *STAI-T* was developed to measure trait anxiety among adults with a reading age of 12 (Spielberger et al., 1983). The 20-item *STAI-T* self-report scale is scored on a 4-point continuum that ranges from 1 (*almost never*) to 4 (*almost always*). Sample items include, '*I am a steady person*' and '*I lack self-confidence*'. Scores range from 20 - 80, where higher scores indicate greater trait anxiety (Spielberger et al., 1983). An empirically derived cut-off score of 44 and higher was used to identify participants with clinical trait anxiety (Spielberger et al., 1983). The *STAI-T* has been found to have high concurrent validity as a self-report measure ($\alpha = .85$; Tilton, 2008) and test-retest reliability ($\alpha = .86$; Spielberger et al., 1983). The *STAI-T* demonstrated good internal consistency in the current sample ($\alpha = .93$) (Mayers, 2013).

The *Beck Depression Inventory II (BDI-II)* (Beck et al., 1996) was used to assess the affective, behavioural, cognitive, motivational and vegetative aspects of

depression. It is a 21-item self-report questionnaire with questions ranging from 0 (e.g. *'I do not feel particularly guilty'*), to 3 (e.g. *'I feel guilty all of the time'*) scored on a Likert – type scale. Individual response scores are summed to create a total score (range: 0 – 63). Empirically derived cut-off scores are 0 – 13 (minimal depression), 14 – 19 (mild depression), 20 – 28 (moderate depression) and 29 – 63 (severe depression) (Beck et al., 1996). In the current sample, the internal consistency of the *BDI-II* was excellent ($\alpha = .94$) (Mayers, 2013).

The *Short Form McGill Pain Questionnaire (SF-MPQ; Melzack, 1987)*, was used to assess the severity of pain experienced (pain intensity) and has been used to assess pain intensity in studies comparing different groups' pain (Tang et al., 2009). This self-report questionnaire assessed several dimensions of pain experience using 15 (sensory and affective) verbal pain descriptors, a current pain index, and a visual analogue scale to assess pain intensity in the previous week ranging from 1 (e.g. *'no pain'*), to 10 (e.g. *'extreme pain'*). Questions 1 – 11 are sensory pain descriptors (e.g. *'throbbing pain'*), rated on a Likert – type scale from 0 (*none*), to 3 (*severe*). Questions 12 – 15 are affective pain descriptors (e.g. *'punishing – cruel pain'*) rated on a Likert – type scale from 0 (*none*), to 3 (*severe*) (Melzack, 1987). From the sum of the rank value of the pain descriptors, a sensory (0 – 3) ($\alpha = .95$; Adelmanesh et al., 2011), an affective (0 – 12) ($\alpha = .83$; Adelmanesh et al., 2011) and a total pain score (0 – 45) ($\alpha = .84$; Adelmanesh et al., 2011) have shown good internal consistency (Mayers, 2013). In the current sample, the internal consistencies of the sensory pain subscale ($\alpha = .92$), the affective pain subscale ($\alpha = .83$) and the combined total of the subscales ($\alpha = .93$) were good (Mayers, 2013).

The *Short Form Health Survey 36 (SF-36; Ware & Sherbourne, 1992)* is a non-disease-specific generic self-report measure of health that is related to functional status and wellbeing. This self-report measure has been previously been used to evaluate quality of life in SCD populations (Asnani et al., 2009; Citero et al., 2007; McClish et al., 2005). It was used to assess quality of life across eight subscales (physical functioning, physical health limitations, emotional health limitations, fatigue, emotional wellbeing, social functioning, pain and general health). Subscales are measured on a scale from 0 – 100, where higher scores on the subscales of the *SF – 36* indicated better levels of health (e.g. *'in general, would you say your health is: 100 = excellent, 75 = very good, 50 = good, 25 = fair and 0 =*

poor'). It was reported that the SF – 36 achieves high levels of criterion validity and high levels of reliability (Ware & Sherbourne, 1992). Asnani et al. (2009) reported that their subscales achieved good internal consistency ranging from ($\alpha = .70$) to ($\alpha = .93$). The subscales of the SF – 36 demonstrated good levels of internal consistency (Mayers, 2013) in this study: physical functioning ($\alpha = .94$), physical health limitations ($\alpha = .93$), emotional health limitations ($\alpha = .94$), fatigue ($\alpha = .85$), emotional wellbeing ($\alpha = .80$), social functioning ($\alpha = .82$), pain ($\alpha = .92$) and general health ($\alpha = .87$).

Data Analytic Plan (Phase I)

All quantitative analyses were conducted using the statistical package, SPSS (Version 20; IBM, 2011). Preliminary analyses were conducted to establish suitability of the data for parametric analysis and to assess applicability of potential covariates i.e. age, gender.

Suitability of Data for Parametric Analysis

The data was checked to ensure that the assumptions for multiple regression and multivariate analyses were met. There were more cases than independent variables for the regression analyses (Field, 2013); only participants who had completed the survey were included in the analyses; and univariate normality was checked by checking the normality of residuals for each dependent variable to ensure multivariate normality. However, using the latter method does not guarantee multivariate normality according to Field (2013). Distributions of study variables were not problematically skewed (Field, 2013), trait anxiety scores were normally distributed with skew of -0.25 ($SE = 0.33$) and kurtosis -0.69 ($SE = 0.66$). The regression of the standardised residual of the variables followed a normal distribution and the normal plot of the regression standardised residuals against the standardised predicted value did not show a specific pattern and thus indicated that it could be assumed that the data was linear (Coolican, 2001; Tabachnick & Fidell, 2013). This data demonstrated that assumptions of homogeneity of variance had not been violated with the data. There were no within-cell outliers using a criterion $z = \pm 3.3$ at $p < .01$ (Mayers, 2013). Results of evaluation of assumptions of normality, homogeneity of variance-covariance matrices, linearity and multi-collinearity were satisfactory.

Assessment of Covariates and Preliminary Correlations

The associations between variables were assessed with bivariate correlations amongst the following variables: trait anxiety score (*STAI-T*), depression score (*BDI-II*), age, number of annual hospitalisations, sensory and affective pain scores (*SF-MPQ*) and the eight subscale scores from the *SF-36* (physical and emotional health limitation scores, emotional wellbeing scores, fatigue scores, social functioning scores, general health scores and pain scores). The following five variables were eliminated from the analyses due to their lack of relationship with the variable trait anxiety in preliminary correlations, where $r < \pm 0.3$ (Tabachnick & Fidell, 2007) and where the significance of the relationships was greater than $p = .05$ (Mayers, 2013): hospital admission frequency over 12 months, $r = .174$, $p = .221$; physical functioning subscale from the *SF-36*, $r = -.252$, $p = 0.075$; pain subscale from the *SF-36*, $r = -.165$, $p = .249$; gender, $r = -.004$, $p = .978$; and age, $r = .181$, $p = .205$, where $N = 51$.

Hierarchical regression (Research Aim 1)

A hierarchical regression was conducted with the full SCD sample ($n = 28$) to document the relationship of trait anxiety with the health outcome variables with the main participant group, SCD. Following this, a hierarchical regression was conducted using the full sample ($N = 51$) to determine whether trait anxiety scores (*STAI-T*) (dependent variable, DV) were significantly predicted by relevant health outcome variables [independent predictor variables were mean scores of depression (*BDI-II*), pain intensity (*SF-MPQ*) and quality of life (*SF-36*)]. The independent variables (IVs) were added to the regression model in three steps. The selection and the order in which the variables were added to the regression model were determined by the strength of their individual correlations with trait anxiety in the preliminary analyses. Depression scores were added in the first step as this was the strongest correlation, sensory and affective pain scores were added in the second step and quality of life subscale scores (physical and emotional limitations, fatigue, emotional wellbeing, social functioning and general health) were added in the third step.

MANOVA analyses (Research Aim II)

Between-group differences (illness groups: SCD, BC and Carers) on mean scores of trait anxiety (*STAI-T*), depression (*BDI-II*), pain intensity (*SF-MPQ*) and quality of life (*SF-36*) were tested using three separate Multivariate Analysis of Variance Analyses (MANOVAs). Analysis of the DVs was separated across three MANOVAs because of measure scoring so that subscales of the same measure were analysed together. In addition, the separation of the DVs ensured that there were more cases than DVs in every cell of the analysis. If a cell had more DVs than cases, then every cell in the analysis would become singular and the assumption of homogeneity of variance-covariance matrices would have become un-testable (Tabachnick & Fidell, 2013). The current study had eight participants per cell and employed 10 DVs across three cells, therefore the analysis needed to have less than eight DVs per cell, per analysis. It was decided to separate the variables into three groups of a reduced number of variables: mood (measured by trait anxiety and depression scores), pain (measured by the *SF-MPQ* sensory and affective pain subscales scores) and quality of life (measured by the six selected subscales scores of the *SF-36*; physical and emotional limitations, fatigue, emotional wellbeing, social functioning, general health).

Significant MANOVAs were followed up by univariate Analysis of Variance Analyses (ANOVAs) to establish whether significant group differences existed at each individual variable level. Significant univariate analyses were conducted using pair-wise post hoc comparison tests to identify which groups were statistically different within variables and the direction of the differences (Tabachnick & Fidell, 2007). MANOVAs were chosen to investigate group differences in the three sets of variables instead of conducting separate ANOVAs and t-tests with 10 separate variables to avoid making Type I errors (Coolican, 2001) and to investigate the overall effects of the variable sets (mood, pain and quality of life), rather than simply investigating individual effects (Mayers, 2013).

Qualitative Data Analysis (Phase II, Research Aim III)

A thematic coding analysis (Braun & Clarke, 2006; Robson, 2011) was used to identify themes and thematic networks after the transcription (verbatim) of the interviews. Accuracy of the transcriptions was validated by comparing the audio data

with the transcribed data and the researcher reflected on the interview process immediately after each interview. Data collection and analysis were conducted simultaneously (Creswell, 2009; Flick, 2009). Five steps were followed for each analysis (adopted from Braun & Clark, 2006). Familiarisation with the data was the first step, by reading through each case's transcript and generating initial codes (Flick, 2009). The next step was to organise the initial codes into bigger groups and bring meaning to those groups (Robson, 2011). The third step was to identify themes by grouping similar codes together and supporting each theme with quotes that supported each theme (Braun & Clark, 2006). The fourth step involved constructing thematic networks by linking related themes together from the same case (Robson, 2011). The last step of the analysis was to construct a narrative that combined thematic descriptions for each case.

After these steps were completed, it was necessary to examine the themes across the two cases to identify themes that occurred for both cases and themes that separated both cases. After this process the qualitative data was interpreted in relation to the quantitative data through integration (Braun & Clarke, 2006; Ostlund, Kidd, Wengstrom & Rowa-Dewar, 2011). Robson (2011) identified that the quality and validity of the data and the interpretations of the data needs to be evaluated to ensure data integrity. In this study, data quality was assessed by evaluating if the data was representative of the data collection process and using convergent and divergent triangulation (Ostlund et al., 2011) to compare and integrate the data. Researcher effects were tested using the process of researcher reflection and reflection using research supervision (Finlay & Gough, 2003). It was important to weigh up the evidence and evaluate the strength of the conclusions that could be made (Robson, 2002). As the data was so specific and the sample size was relatively small, it was not possible to generalise the implications of the study to the specific groups (Flick, 2009). The findings were generalised to the sample and suggested implications were indicative of future avenues of research, rather than as conclusive evidence. This study also focussed on looking for negative evidence as a method of validating conclusions made and testing the integrity of the process and the findings.

Chapter 4: Quantitative Results

This chapter reports the findings from phase I.

Sample Characteristics

Means and standard deviations of continuous variables of the total sample ($N = 51$) are presented (see Table 2). Eight BC participants (15.7%) completed the survey, compared to 28 SCD participants (54.9%) and 15 Carers (29.4%). Due to the potential of introducing experimental error due to sample size differences across groups and the likelihood of violating the MANOVA requirement of equal sample sizes in individual groups (Field, 2013; Tabachnick & Fidell, 2007), the total sample was reduced from 51 to 24 ($n = 8$ per group) for the reported MANOVA analyses. Using unequal sample sizes in the MANOVA, as would have been the case using the full sample ($N = 51$) would have reduced the robustness of the analyses and distorted the probability values (Tabachnick & Fidell, 2013). The SCD and Carer group participant numbers were reduced through random selection to match the number of participants in the BC group, ($n = 8$), thereby leaving three groups with eight participants in each group (total $n = 24$). Means and standard deviations of the selected continuous variables across the three equal sized illness groups are presented (see Table 2, $n = 24$). The MANOVA analyses were also run with the unequal group sizes (SCD, $n = 28$; BC, $n = 8$; Carers, $n = 15$) to show that there was no significant negative impact of reducing the group sizes to $n = 8$ in each group in order to maintain equal group sizes (see Appendix E).

Hierarchical Regression (Research Aim I)

Table 3 displays bivariate correlations and significance levels of relevant variables across the complete sample ($N = 51$). Significant positive correlations existed for trait anxiety scores and scores of depression, sensory and affective pain, physical and emotional health limitations, fatigue, emotional wellbeing, social functioning and general health.

Results from the hierarchical regression demonstrated that trait anxiety was predicted by depression scores (48.1%) and by the quality of life subscale scores (physical and emotional limitations, fatigue, emotional wellbeing, social functioning and general health) (27.1%) in the complete sample. Addition of pain intensity

(sensory and affective pain) to the model did not create a significant F change. See Table 4 for regression results.

Table 3 displays bivariate correlations and significance levels of relevant variables across the complete SCD sample only ($n = 28$). Significant positive correlations existed for trait anxiety scores and scores of depression, sensory and affective pain, physical and emotional health limitations, fatigue, emotional wellbeing, social functioning and general health. These significant relationships generally mirrored the significant relationships indicated in the regression analyses with the complete sample ($N = 51$).

Results from the hierarchical regression demonstrated that trait anxiety was predicted by depression scores (67.2%) and by the quality of life subscale scores (physical and emotional limitations, fatigue, emotional wellbeing, social functioning and general health) (17.7%). As with the complete sample, the addition of pain intensity (sensory and affective pain) to the model did not create a significant F change. See Table 4 for regression results.

MANOVA Analyses (Research Aim II)

Three separate between – groups multivariate analyses of variance (MANOVA) were conducted to evaluate group differences across nine dependent variables: MANOVA 1 - trait anxiety scores (*STAI-T*) and depression scores (*BDI-II*), MANOVA 2 - sensory and affective pain intensity scores (*SF-MPQ*) and MANOVA 3 - physical limitations, emotional limitations, fatigue, emotional wellbeing, social functioning and general health scores (*SF-36*). The IV was illness group (SCD, BC, Carers). As discussed earlier, a smaller and equal group subsample was used in these analyses to match the number of complete responses in the smallest illness group, where ($n = 8$) in each illness group to prevent the violation of MANOVA. A bootstrap (a statistical technique that re-samples the data to improve accuracy and reduce bias; Field, 2013) was conducted to control for the small sample size; however MANOVA did not bootstrap the main tests. The potential covariates of gender and age were not added to the MANOVAs as there was no significant relationship between gender or any of the dependent variables (Tabachnick and Fidell, 2007). Age was not included as a covariate because when the independence of the illness group was tested against age, it was significant indicating that including

Table 2

Means and Standard Deviations of the variables in total sample (n = 51), in illness group sub-sample (n = 24) and in complete SCD sub-sample (n = 28)

| Variable | Total sample (n = 51) | Illness Groups (n = 24) | | | SCD sub- sample |
|-------------------------------|--------------------------|-------------------------|--------------------|--------------------|--------------------|
| | Mean (SD) | SCD (n=8) | BC (n=8) | C (n=8) | SCD (n=28) |
| | | M (SD) | M (SD) | M (SD) | M (SD) |
| Age (years) | 37.33 (11.77) | 31.88 (8.39) | 52.04 (13.15) | 34.42 (7.52) | 33.32 (8.83) |
| No of annual hospitalisations | 1.35 (2.95) | 2.37 (4.27) | 1.75 (3.15) | 0 (0) | 1.96 (3.48) |
| Trait anxiety | 49.49 (12.33) | 49.75 (5.15) | 56.63 (11.58) | 43.13 (9.22) | 50.07 (11.58) |
| Depression | 18.92 (13.95) | 17.63 (6.74) | 33.63 (17.44) | 11.25 (9.21) | 18.54 (11.63) |
| Sensory pain | 12.31 (9.18) | 15.88 (5.39) | 11.63 (11.07) | 3.75 (6.18) | 16.29 (6.98) |
| Affective pain | 4.04 (3.78) | 4.38 (3.07) | 3.75 (4.98) | 1.50 (2.51) | 5.18 (3.27) |
| Physical health limits | 156.86 (178.05) | 62.50 (118.77) | 125.00 (175.26) | 262.50 (192.26) | 103.57 (147.78) |
| Emotional health limits | 135.29 (142.58) | 175.00 (148.81) | 75.00 (138.87) | 200.00 (141.42) | 125.00 (137.77) |
| Fatigue | 134.12 (99.54) | 162.50 (70.46) | 65.00 (70.71) | 197.50 (89.08) | 124.29 (93.07) |
| Emotional wellbeing | 270.59 (108.32) | 272.50 (48.92) | 222.56 (116.83) | 327.50 (95.58) | 264.29 (103.76) |
| Social functioning | 94.61 (60.89) | 103.13 (52.50) | 62.50 (42.56) | 137.50 (62.68) | 80.36 (55.43) |
| General health | 177.94 (137.99) | 162.50 (60.18) | 103.13 (86.02) | 318.75 (133.46) | 131.25 (106.85) |

Note. SF – MPQ = Short Form McGill Pain Questionnaire, SF – 36 = Short Form Health Survey 36, SCD = Sickle Cell Disease, BC = Blood Cancer, C = Carer

Table 3

Bivariate Correlations of Trait Anxiety and the health outcomes in total sample (N = 51) above the diagonal line and complete SCD-only sample (n = 28) below the diagonal line

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|--------|
| 1.Trait anxiety (STAI-T) | — | .70** | .37** | .47** | -.33** | -.59** | -.70** | -.86** | -.60** | -.59** |
| 2.Depression (BDI-II) | .82** | — | .37** | .42** | -.30* | -.45** | -.57** | -.74** | -.56** | -.58** |
| 3.Sensory pain (SF-MPQ) | .37** | .48** | — | .83** | -.61** | -.34** | -.48** | -.35** | -.54** | -.60** |
| 4.Affective pain (SF-MPQ) | .56** | .65** | .73** | — | -.52** | -.45** | -.48** | -.38** | -.45** | -.53** |
| <i>SF-36 subscales</i> | | | | | | | | | | |
| 5.Physical health limits | -.34* | -.41* | -.26* | -.22 | — | .52** | .50** | .28* | .56** | .63** |
| 6.Emotional health limits | -.51** | -.41* | -.07 | -.39* | .31 | — | .70** | .49** | .58** | .52** |
| 7.Fatigue | -.63** | -.51** | -.28 | -.42* | .21 | .49** | — | .63** | .83 | .72** |
| 8.Emotional wellbeing | -.89** | -.81** | -.45** | -.60** | .38* | .41** | .60** | — | .60** | .53** |
| 9.Social functioning | -.50** | -.41* | -.32* | -.25 | .41* | .36* | .78** | .53** | — | .69** |
| 10.General health | -.53** | -.57** | -.35* | -.40* | .44* | .27 | .55** | .53** | .43** | — |

Note. * $p < .05$ ** $p < .01$; *STAI-T* = *State-Trait Anxiety Inventory* (Spielberger et al., 1983), *BDI-II* = *Beck Depression Inventory-II* (Beck et al., 1996), *SF-MPQ* = *Short Form McGill Pain Questionnaire* (Melzack, 1987), *SF-36* = *Short Form Health Survey 36* (Ware & Sherbourne, 1992)

Table 4

Summary of Sequential Regression Results and Individual Predictors of Trait Anxiety in the total sample (N = 51) and in the complete SCD sub-sample (n = 28)

| Variable (N = 51) | R ² | t | β | f ² | p |
|---|----------------|-------|------|----------------|------|
| Dependent variable: trait anxiety scores | | | | | |
| <u>Step 1:</u> | | | | | |
| Depression scores | .49 | 6.89 | .70 | .96 | 0.00 |
| <u>Step 2:</u> (F _{change} = 141.691) | | | | | |
| Sensory pain scores | | -.58 | -.11 | .04 | 0.56 |
| Affective pain scores | | 1.58 | .29 | | 0.12 |
| <u>Step 3:</u> (F _{change} = 8.431) | | | | | |
| Physical health limitations scores | .77 | .36 | .04 | .39 | 0.72 |
| Emotional health limitations scores | | -.92 | -.10 | | 0.37 |
| Fatigue scores | | -1.17 | -.18 | | 0.25 |
| Emotional wellbeing scores | | -5.84 | -.64 | | 0.00 |
| Social functioning scores | | .45 | .06 | | 0.66 |
| General health scores | | -.81 | -.10 | | 0.42 |
| Variable (n = 28) | R ² | t | β | f ² | p |
| Dependent variable: trait anxiety scores | | | | | |
| <u>Step 1:</u> | | | | | |
| Depression scores | .67 | 7.30 | .82 | 2.03 | 0.00 |
| <u>Step 2:</u> (F _{change} = 53.228) | | | | | |
| Sensory pain scores | .68 | -.65 | -.11 | .01 | 0.52 |
| Affective pain scores | | .68 | .13 | | 0.50 |
| <u>Step 3:</u> (F _{change} = 3.362) | | | | | |
| Physical health limitations scores | .85 | .22 | .03 | .22 | 0.83 |
| Emotional health limitations scores | | -1.01 | -.12 | | 0.33 |
| Fatigue scores | | -.86 | -.18 | | 0.40 |
| Emotional wellbeing scores | | -3.54 | -.7 | | 0.00 |
| Social functioning scores | | .50 | .62 | | 0.62 |
| General health scores | | -.022 | -.00 | | 0.98 |

Note. β = standardised beta weight from multiple regression

it would increase the error variance of the MANOVA (Mayers, 2013). In addition, running the MANOVA with these variables reduced the overall power of the analyses.

MANOVA 1: Group differences in mood (trait anxiety and depression)

MANOVA 1 was conducted to evaluate between-group (SCD, BC, Carers) differences in trait anxiety and depression scores. The result of the overall MANOVA was significant, *Wilks' Λ* = .532, $F(4, 40) = 3.71$, $p = .012$, Cohen's $d = .371$ (see Table 5). Follow-up univariate analyses revealed significant group differences in trait anxiety scores, $F(2, 21) = 4.46$, $p = .024$, Cohen's $d = .631$, and in depression scores, $F(2, 21) = 7.34$, $p = .004$, Cohen's $d = .835$.

Table 5

Multivariate Analysis of Variance results for Illness Group and DVs (mood, pain intensity, quality of life)

| | Wilks Lambda | df 1 | df2 | Multivariate <i>F</i> | <i>d</i> | <i>p</i> |
|---|--------------------------------|------|-----|--------------------------|----------|----------|
| <i>Mood (STAI-T) & (BDI-II)</i> | .532 | 4 | 40 | 3.71 | .371 | .012* |
| <i>Pain Intensity (SF-MPQ)</i> | .599 | 4 | 40 | 2.92 | .292 | .033* |
| | Pillai's Trace Statistic | | | | | |
| <i>Quality of Life (SF-36)</i> | .804 | 12 | 34 | 1.91 | .672 | .07* |

Note. * $p < .05$; STAI = State-Trait Anxiety Inventory (Spielberger et al., 1983); BDI-II = Beck Depression Inventory II (Beck et al., 1996); SF- MPQ = Short Form McGill Pain Questionnaire (Melzack, 1987); SF – 36 = Short Form Health Survey 36 (Ware & Sherbourne, 1992).

Bonferroni post hoc pairwise comparisons showed that the BC group reported significantly higher trait anxiety scores ($M = 56.63$, $SD = 11.58$) relative to the Carers ($M = 43.13$, $SD = 9.22$; $p = .021$). There was no significant difference in the trait anxiety scores between the SCD group ($M = 49.75$, $SD = 5.15$) and the BC group ($M = 56.63$, $SD = 11.58$; $p = .430$) or between the SCD group ($M = 49.75$, $SD = 5.15$) and the Carers ($M = 43.13$, $SD = 9.22$; $p = .473$). With regard to depression scores, the BC group also reported higher depression scores ($M = 33.63$, $SD = 17.44$) than both the SCD group ($M = 17.63$, $SD = 6.74$; $p = .044$) and the Carers group ($M = 11.25$, $SD = 9.21$; $p = .004$). There was no significant difference in depression scores between the SCD group ($M = 17.63$, $SD = 6.74$) and the Carers group ($M = 11.25$, $SD = 9.21$; $p = .904$). In summary, there was no difference in trait anxiety scores between SCD and BC, but the BC group did report significantly higher depression scores compared to the SCD group.

MANOVA 2: Group differences in pain intensity (sensory and affective)

MANOVA 2 was conducted to evaluate between-group (SCD, BC, Carers) differences in sensory and affective pain scores. The result of the overall MANOVA was significant, $Wilks' \Lambda = .599$, $F(4, 40) = 2.92$, $p = .033$, Cohen's $d = .292$ (see Table 5). Follow-up univariate analyses revealed significant group differences in sensory pain scores only, $F(2, 21) = 4.79$, $p = .019$, Cohen's $d = .675$.

Bonferroni post hoc pairwise comparisons showed that the SCD group reported significantly higher sensory pain scores ($M = 15.88$, $SD = 5.39$) relative to the Carers ($M = 3.75$, $SD = 6.18$; $p = .018$). There were no significant differences in sensory pain between the SCD group ($M = 15.88$, $SD = 5.39$) and the BC group ($M = 11.63$, $SD = 11.07$; $p = .892$), or between the BC group ($M = 11.63$, $SD = 11.07$) and the Carers ($M = 3.75$, $SD = 6.18$; $p = .183$). In summary, only sensory pain was different across the illness groups; the SCD group experienced more sensory pain than the Carers, but similar sensory pain to the BC group.

MANOVA 3: Group differences in quality of life (physical and emotional health limitations, emotional wellbeing, fatigue, social functioning & general health)

MANOVA 3 was conducted to evaluate between-group (SCD, BC, Carers) differences in quality of life subscale scores (physical health limitations, emotional

health limitations, fatigue, emotional wellbeing, social functioning and general health). Pillai's trace statistic was used to observe significance in this MANOVA because Box's test of equality of covariance matrices was significant at, $p = .001$, despite there being equal group numbers ($n = 8 \times 3$ groups) (Tabachnick & Fidell, 2007). Pillai's trace results indicated that there was a marginally significant difference in quality of life subscale scores across the illness groups, Pillai's trace $V = .804$, $F(12, 34) = 1.91$, $p = .07$, Cohen's $d = .672$ (see Table 5). Follow-up univariate analyses revealed significant group differences in fatigue scores, $F(2, 21) = 6.32$, $p = .007$, Cohen's $d = .776$; in social functioning scores, $F(2, 21) = 3.99$, $p = .034$, Cohen's $d = .617$; and in general health scores, $F(2, 21) = 9.41$, $p = .001$, $d = .947$.

Bonferroni post hoc pairwise comparisons showed that the BC group was marginally more fatigued ($M = 65.00$, $SD = 70.71$) than the SCD group ($M = 162.50$, $SD = 70.46$; $p = .059$), and was significantly more fatigued than the Carers ($M = 197.50$, $SD = 89.08$; $p = .008$). The BC group had lower social functioning scores ($M = 62.50$, $SD = 42.56$) than the Carers ($M = 137.50$, $SD = 62.68$; $p = .031$), but there was no significant difference in social functioning between the SCD group ($M = 103.13$, $SD = 52.50$) and the BC group ($M = 62.50$, $SD = 42.56$; $p = .424$), or between the SCD group ($M = 103.13$, $SD = 52.50$) and the Carers ($M = 137.50$, $SD = 62.68$; $p = .629$). The SCD group had lower general health scores ($M = 162.50$, $SD = 60.18$) than the Carers ($M = 318.75$, $SD = 133.46$; $p = .019$) and the BC group had lower general health scores ($M = 103.13$, $SD = 86.02$) than the Carers ($M = 318.75$, $SD = 133.46$; $p = .001$). However, there was no significant difference in general health scores between the SCD group ($M = 162.50$, $SD = 60.18$) and the BC group ($M = 103.13$, $SD = 86.02$; $p = .781$). In summary, there were group differences in the fatigue, social functioning and general health subscales of the *SF-36* (Ware & Sherbourne, 1992). The SCD group were marginally less fatigued than the BC group and had lower general health scores than the Carers.

Summary

Hierarchical regression showed that trait anxiety, in both the complete sample ($N = 51$) and in the complete SCD sample ($n = 28$), was predicted by depression scores and quality of life subscale scores (physical and emotional health limitations, fatigue, emotional wellbeing, social functioning and general health). Three separate

MANOVAs were used to show that there was a difference in trait anxiety, depression and sensory pain scores across illness groups and there was a difference in fatigue, social functioning and general health across illness groups.

Both the SCD group ($M = 49.75$, $SD = 5.15$) and the BC group ($M = 56.63$, $SD = 11.58$) had similar trait anxiety scores as measured by the *STAI -T* ($M \geq 44$ = clinical trait anxiety; Spielberger et al., 1983). However, the BC group had a mean severe depression score ($M = 33.63$, $SD = 17.44$) as measured by the *BDI-II* (Beck et al., 1996) that was significantly higher than that of the SCD group ($M = 17.63$, $SD = 6.74$). The SCD group ($M = 15.88$, $SD = 5.39$) had similar levels of mean sensory pain scores to the BC group ($M = 11.63$, $SD = 11.07$), but only the SCD group had significantly higher levels of sensory pain compared to the control group of Carers ($M = 3.75$, $SD = 6.18$). Despite the similar levels of sensory pain in both the SCD and the BC groups, and similar levels of trait anxiety, the BC group mean depression score was significantly higher than the SCD group mean depression score. Subsequently, Chapter 5 explores and discusses how the SCD and BC participants with clinical trait anxiety experienced their pain, anxiety and low mood to explore what could account for the difference in depression scores.

Chapter 5: Qualitative Results

The quantitative results demonstrated that trait anxiety was predicted by depression scores, physical and emotional health limitations, fatigue, emotional wellbeing, social functioning and general health. The quantitative results also showed that a difference in trait anxiety, depression scores, sensory pain scores and quality of life scores across the illness groups existed. However, despite the similar levels of sensory pain in the SCD and the BC groups and the relatively similar levels of trait anxiety, there was a significant difference in depression scores between the two groups; the BC group had higher depression scores than the SCD group. The qualitative phase was used to explore this difference (see appendix D for the interview outline) in one participant from both the SCD and the BC group.

Data collection and a thematic content analysis (Braun & Clarke, 2006; Robson, 2011) were carried out simultaneously on two cases: an SCD and a BC participant who were selected for Phase II because of their clinical trait anxiety levels

which were above the clinical cut off score of 44 (Spielberger et al., 1983) and because of the participants' above average depression scores (Beck et al., 1996).

Description of Cases

Case 1: Dee

...I know my body will respond to my emotions and um, the connection between one's mood and one's physical self, um, if I am depressed I will feel more sick. If I am laughing....when I am laughing I cannot feel ill (from the interview with Dee).

Dee was a 56 year old Asian Indian South African woman who partly lived in the UK and in South Africa. She was married and she and her husband had two adult children. She was recruited through a UK-based online support group for Hairy Cell Leukaemia, a form of Blood Cancer. She had lived with this form of leukaemia for three years and was in partial remission. She used to work as a political activist, as a writer and as a humanitarian and foster home manager, but was no longer able to work due to her illness.

She had been admitted to hospital nine times in the previous 12 months and despite the physical difficulties with her illness, she did not have a carer. Her *STAI - T* score was 54, which indicated that she had clinical trait anxiety (above the clinical cut-off score of 44; Spielberger et al., 1983) and her *BDI - II* score was 31, which indicated that she had severe depression (Beck et al., 1996). Her sensory pain score was 29 out of a possible 33 (*SF - MPQ*; Melzack, 1987), which indicated high levels of sensory pain and her general health score (*SF - 36*; Ware & Sherbourne, 1992) was 25 out of a possible 500, indicating extremely low levels of general health and, subsequently a lower quality of life level.

Dee was psychologically-minded and engaged well with the research questions. She was able to elaborate on her thoughts and feelings easily. She was interviewed via telephone because she was in South Africa at the time of the interview. Due to the emotional content, the mode of the interview and the scores indicating high levels of anxiety and high levels of low mood, extra care was taken to debrief Dee post-interview and ensure that she knew who she could contact if she felt she needed to address some of the issues raised by the interview.

Case 2: Samson

I have to be aware of my body all of the time; when I'm working, when I'm thinking, I must think 'at what level can I do this'? At what level can my body tolerate this? And I must be aware of where is the pain now? (from the interview with Samson).

Samson was a 37 year old Black African man who lived in Nigeria. He was unmarried and lived with his parents and siblings. He was recruited through an SCD conference held by the Sickle Cell Society in London. Although he had lived with SCD his whole life, he was only diagnosed with disease around the age of seven. He worked part-time as an IT manager and had a part-time carer to enable him to work part-time and manage his illness and medical treatment.

Samson had twice been admitted to hospital in the previous 12 months. His *STAI – T* score was 59, which indicated that he had clinical trait anxiety (above the clinical cut-off score of 44; Spielberger et al., 1983) and his *BDI-II* score was 18, which indicated that he had mild depression (Beck et al., 1996). His sensory pain score was 21 out of a possible 33 (*SF – MPQ*; Melzack, 1987), indicated average levels of sensory pain and his general health score (*SF – 36*; Ware & Sherbourne, 1992) was 150 out of a possible 500, indicating low levels of general health and, subsequently lower quality of life levels.

Samson engaged well with the research questions. He was proficient in English, which was his second language. He was able to express his feelings clearly, but because he was interviewed via telephone (he was in Nigeria at the time of the interview), it was sometimes difficult to hear him clearly. Care was taken to debrief the participant post-interview and ensure Samson knew who he could contact if he felt he needed to address some of the issues raised by the interview.

Findings

The qualitative findings addressed three main issues: 1) general pain experience, 2) general anxiety and 3) general low mood. The findings also addressed some general questions regarding attention to pain, changes in pain and mood, and participant identity. Six themes emerged from the data: pain appraisal, purpose and change in identity, coping strategies, anger and frustration, social

construction of illness and, personal control. Common and contrasting codes emerged from the interviews. A table of themes and sub-themes is presented (see Table 6).

Pain Appraisal

Both Dee and Samson reported that they experienced pain all the time.

Dee: 'I've always got pain....if I had one day a month pain free...'

Samson: 'The pain is always with me...'

Dee described her pain as 'completely indescribable' and reported that she found no meaning in pain, whilst Samson indicated that whilst he was always in pain, there were times where the pain was worse and these times occurred when he faced additional stressors, 'I don't really have extra pain unless I stress myself'. Both cases identified that stress made their pain worse, however, Samson regarded stressful situations as within his locus of control, '....unless I stress myself', whereas Dee did not report her stress with this same internal locus of control, 'my pain is always made worse if I'm stressed about something'.

Both cases reported different types of pain,

Dee: 'We've got neuropathy, we cannot walk, the pain in your feet and legs is too terrible...'

Samson: 'My joints, my stomach, my legs...and my back',

which was a constant battle for them. Dee described the battle as,

'A steady slide down, like snakes and ladders. You think that you are getting better and you go up the ladder and then you slide back even further than before'.

She positioned this battle in relation to the fact that she had not always been ill and that being ill had affected her life and her identity. Her statement also suggested that she subconsciously had hope that she would fully recover, but that having this hope meant that when the pain intensity increased her pain would feel

Table 6

Main Themes and Sub-themes used in the thematic analysis for exploring pain experience and general anxiety and low mood

| Themes | Sub-themes |
|--------------------------------|--|
| Pain Appraisal | Causation |
| | Constancy |
| | Description |
| Purpose and Change in Identity | Former purpose and identity |
| | Current purpose and identity |
| | Change in purpose and identity |
| | Conflict between former and current purpose and identity |
| Coping Strategies | Avoidance |
| | Distraction |
| | Emotion Release |
| | Humour |
| | Internet |
| | Medicine |
| | Practical |
| | Rationalising |
| | Social Support |
| Anger and Frustration | Conflict in accepting current identity |
| | Link to other expressed emotions |
| | Other people's perceptions of illness |
| | Reduced personal control |
| Social Construction of Illness | Available social support |
| | Isolation |
| | Lack of professionals' understanding |
| | Others' lack of understanding |
| Personal Control | Acceptance/resignation |
| | Hyper-vigilance |
| | Locus of control |
| | Mind/Body connection |
| | Powerlessness |
| | Suicidal Ideation |

worse than it did before. In contrast, as Samson had suffered from SCD from birth, it appeared that he was more able to position his pain on a time continuum,

‘I can say that it’s gone better...’

and he had developed strategies to avoid pain,

‘I usually do a lot to be able to try and stay away from pain...I’m watching everything around me. I watch everything to see what will affect my body’.

Purpose and Change in Identity

Both cases spoke about how their diseases had affected their identities and gave them different purposes in life than what they might have otherwise had. Dee spoke about having time to,

‘Reflect on your life and try to understand the meaning of life...I felt I was living a worthwhile life and now it feels like I’m not living a worthwhile life’.

Her purpose in life had changed,

‘I’ve always said life will always be worthwhile if you can make one person per day smile and forget their troubles...so far I’ve managed to make people laugh, then I feel it was not a wasted day’,

however, the change was not easy for her,

‘It’s hard to find a life that has meaning and is worthwhile. You’ve got to work at it’.

Where Dee spoke more of finding her purpose in helping others, Samson’s sense of purpose was grounded in fighting to survive and using others’ negativity towards him as a source of inner strength and resilience,

‘You only try to live and try to survive...I want to survive. I want to survive. If I don’t continue living then I won’t survive. That’s the main thing. That’s what keeps me going, I think. I just want to survive. I want to try and make it and it will even show people, at least, that they think

that 'they will die', 'they are useless', you know to prove them wrong. To prove them wrong. That's what keeps me going'.

When Samson and Dee did experience conflict in accepting their identities, the difference in experiences was that Dee reported that she did have a different identity before she developed cancer,

'I'm a human rights activist. I was in and out of court trying to help other people with human rights abuses...I founded a children's home for abandoned, abused and neglected children, children with HIV and children left and been dying on the pavement...I also got on the wrong side of the government and ...had to pull me out and take me to the UK...I've led a very, very, very busy and dangerous life...now my life is restricted to my bedroom and the hospital and my bedroom and the hospital. And my internet...I felt I was living a worthwhile life and now it feels like I'm not living a worthwhile life'.

Dee spoke about how difficult it was for her to experience and accept her new identity because of who she had been before and that this shift from an old identity and schema from before she was ill to her new identity with leukaemia often left her with internal conflict,

'I find it extremely difficult that my life has changed so drastically that I can't do what I used to be able to do. There's always fighting going on in my head. I want my old life back. I want it back and I can't get it back and I never will get it back...life has become much smaller...I have to come to terms with the fact that I'm not going to get my old life back...that's the fight and frustration that I have with myself all the time...I have a new identity. First of all I didn't want to accept it – I was in denial and chasing...I've had to accept this is the way it is right now'.

Dee also struggled with wanting to take morphine to manage her pain, but not liking the personality changes she experienced as a side effect,

'On morphine the pain is controlled, but I don't like who I am. I've become extremely aggressive and nasty. And impatient'.

Where Dee yearned for her 'old' life, it would appear that Samson yearned for a different life to the one that he had always had and did not experience the same internal conflicts that Dee experienced,

'I feel like I'm in inertia. I don't like it at all...sometimes I compare myself to my friends and what they are doing with their lives. Some of them are driving their own cars, live in their own houses, having families of their own, but I can't do that. Because really, I can't. I can see that it (SCD) has really changed everything. So, it's not something that we like, it's just something that we try to manage and live on. I don't like being sick. I don't like having Sickle Cell. I don't like it'.

Coping Strategies

A variety of coping strategies were used by both Dee and Samson; some strategies were common to both cases and some were different. Both Dee and Samson used practical coping strategies to cope with their pain,

Dee: 'I put a pillow over my face and scream...it gives me an outlet...I give myself medicine...'

Samson: 'I will take a pillow underneath my back...I will use hot water because of the joints...when I use hot water the pain will go down much more than when I don't do anything...I usually use Panadol...'

Both Dee and Samson also used distraction techniques, social interactions and the internet as effective coping strategies to help them manage their pain,

Samson: 'I know I'm in pain, but when I see them (my friends) around talking the anger goes down...I usually go online, chat with my friends or watch movies...'

Dee: 'I've also joined a group that've got the same kind of disease I have...I can also help other people to feel better...I can only help others now by the internet...you cannot help somebody else without helping yourself...I write my emotions down...'

In addition, Samson used avoidance strategies to help him manage his pain and his difficult emotions,

'I avoid using drugs that I don't know...I can't take just any drugs, or use any balm, or use any water, or use any type of soap...Me with Sickle Cell, I don't do that. I don't do things that way. I don't take things (my health) for granted. I avoid things that cause me to struggle because I know that it will cause me anxiety'.

In comparison, Dee predominantly used humour and rationalising to help her to manage her pain and her difficult emotions,

'I start thinking about all those that are much worse than me...I just have to get through for the next minute. I just have to do the next minute. For tomorrow, I will be able to stretch it to an hour...tomorrow will be better and today is better than yesterday...If I am laughing, I cannot feel ill. It's not possible to laugh and feel ill at the same time...for those few seconds, those few minutes, you get a way past your problems...I try to find the funny side in everything...'

Avoidance strategies, using humour and rationalising difficult thoughts and events are common coping strategies used to manage chronic pain (Caird et al., 2011; Edwards et al., 2006; Jonassaint et al., 2010).

Anger and Frustration

Anger and frustration were emotions that featured prominently in both of the interviews. When asked about their anxiety, both cases described having frustration and anger,

Dee: 'I don't get anxiety. I have frustration.'

Samson: 'I can't really describe it. I get angry, especially when I am having pain...so, I'm usually angry, so that's it. That's when I can tell that I'm anxious'.

Dee reported that the origin of her anger was the conflict she experienced with wanting her old life back and knowing what she once was and learning to accept this,

'I have to come to terms with the fact that I'm not going to get my old life back. And that's the fight and frustration that I have with myself all the time'.

The origin of Samson's anger was often linked to other peoples' perceptions or treatment of him,

'People treat us like some type of taboo, or we are going to die...once I hear it my blood will start boiling. Those are the things that make me angry...people do some silly things and look down on us, but it makes me angry and instead of getting better, the pain gets worse. The things that make me angry are like 'they are going to die', 'they are useless', 'they can't do this, they can't do that'.

Samson also reported getting angry when his relatives got upset when they saw him in pain and also when he would have to relinquish his personal control when in pain,

'They'll start getting upset...so...I don't like it, I even get angry. First, I can't do anything on my own. Second, I will have to rely at least 70% on my family...babysitting is the thing they usually do...when I am in pain I can get so angry. I get angry easily'.

Dee linked her frustration to her low mood,

'Sometimes the frustration gets to a certain level and then it's like you let go of the frustration and then uh, you become depressed – you can't get yourself out of this hole...'

Dee described the relationship between her anger and low mood as a dark hole which was linked to her lack of personal control,

'The feeling of helplessness and hopelessness and a certain amount of anger...it makes me very frustrated when, um, something needs to be

done and the people around me can't see the need that it must be done. And then, I'm struggling to find that because I've always been helping others, I find it difficult to ask for help for myself'.

Samson also linked his anger and frustration to his personal control, but rather than feeling a lack of personal control, it seemed that his anger and frustration were something that he had to monitor so that he could control them,

'I can't say that it (his anger) has changed and I can't say that it has not changed, I just try to avoid getting angry, that's what I do. I monitor my mood and it always starts off with me getting annoyed, then I will stop what I'm doing...'

Social Construction of Illness

Three main sub-themes emerged within this overall theme: medical professionals' lack of understanding, others' lack of understanding and isolation. Both participants felt that medical professionals did not understand how they experienced their respective diseases and did not have adequate knowledge,

Dee: 'The knowledge they do have is – mostly comes from post mortems...You can't shave your legs if you're a female because the hair follicles start bleeding. You can't shave your face if you're a man...it's all these things that the medical profession don't even know about...doctors will only see what they recognise or what they see and what they've been taught and will only recognise what they see, so if anything falls outside of that they will not actually see it'.

Samson: 'Sometimes, the doctor will give me some hard drugs and expect me to be coping, but it doesn't always work'.

In addition, Dee spoke of how doctors looked at patients with rare illnesses like hers, and detached themselves emotionally from them because they could not fully help them,

'The doctors eventually look at you and because they can't help you anymore, I've been told, 'just hang in there', uh, you know 'it's a medical science, it's getting breakthroughs every day'...and doctors

also start losing their compassion and empathy for you, because there is nothing they can do for you...you just become a number. You actually can feel that the doctor is pulling emotional attachment away from you because they've gotta cope with all their patients. If they were emotionally connected to all their patients they would constantly be grieving'.

Others' lack of understanding was associated with anger in both participants.

Dee: 'They (my friends) don't really know what's going on. They look at me and they say 'but you look so well, you wouldn't say you were ill!' I'd like to smash her teeth in...there's no empathy, there's no compassion'.

Samson: 'People treat us like some type of taboo, or we are going to die...once I hear it, my blood will start boiling...they (his work colleagues) don't know really what I go through and they don't care...I don't like it at all...I told her (a woman he had been courting) I had Sickle Cell, she said that she can't be with a man that's going to die – 'I like him, but I can't like him'.

However, both also had people in their lives that were able to provide social support in coping with their illnesses,

Dee: 'I said to my husband, I just have to get through for the next minute. I just have to do the next minute...If I go out, I must go with the wheelchair. My husband must push me...Only my children speak to me and my mother's always believed, has always stood by me'.

Samson: 'They (my parents) are used to it...so they are coping well...they are very supportive. When I don't usually feel like walking, usually they help me. They are coping well...If I have any friends I can chat with I will ask them to come over. So that time at least I will feel much happier – the mood will change. So, that's much better than just sitting there alone'.

And there was also a theme of isolation regarding managing the illness,

Dee: 'You fall outside the protocol of the norm...I'm the only one in Africa...There's a few of us where nobody knows what it's like, how horrible it is...I don't see many people because I don't have an immune system...it's completely destroyed my relationship with my family, uh only my children speak to me and my mother's always believed...it gets to this point where the doctors are now actually pulling away from me. Emotionally.'

Samson: 'I wouldn't want to leave my room because maybe my niece will come and they'll start getting upset...I'm the only Sickle Cell in my family, so it's not good...'

Personal Control

The theme of personal control appeared to be a vital theme running through both interviews. Several sub-themes emerged from this theme that helped to cluster different aspects of personal control that were highlighted in both cases. The ability to accept or be resigned to the illness demonstrated the participants' sense of their locus of control regarding their illnesses. There were times when they both felt they had no control, or were powerless and for both participants, that was a time when they would actively think of ways of regaining control of their illnesses, either through suicidal ideation or through hyper-vigilance of their symptoms. This sense of control was closely linked to their ideas of how their mind influenced their pain and vice versa. This sense of control was also linked to their ability and their identity, or to their beliefs about having a choice about what was happening to them and how they could manage what was happening to them.

Feeling powerless was closely linked to suicidal ideation in both participants. Having no control was linked to frustration, anger and depression and was also linked to having to rely on others to carry out tasks for them,

Dee: 'Sometimes the frustration gets to a certain level and then it's like you let go of the frustration and then, uh, you become depressed – you can't get yourself out of this hole. You, you know it's a struggle to get out of that dark hole...I'm so tired that I can go down the stairs, but I know that I will not be able to come up the stairs without help.'

That makes me very frustrated....I get scared that I'm not going to die. I don't want to live my life like it is now. I want to be well and I choose to live. And it's just a constant struggle to be. To be'.

Samson: '(When I'm in pain) I will have to hold someone or hold the wall if I'm trying to walk...I can't do anything, because if I'm out I will be totally stranded. I can't do anything, especially on my own when I have a crisis; totally, it spoils everything until that pain goes away...First, I can't do anything on my own. Second, I will have to rely at least 70% on my family, they do everything for me. Babysitting is the thing they usually do. I won't be able to go and relieve myself on my own. It's a struggle...Low mood will bring me down every time. I will lose focus and everything will go down... As the pain goes down, for example, the back pain, I feel so relieved because I don't want to rely on people...I have no choice. I'm trying to cope, but I am not coping well. Just trying'.

Suicidal ideation occurred when both participants felt powerless, or overwhelmed by their symptoms and was tempered by their beliefs about their ability to choose to live,

Dee: 'I am not afraid to die...I have a new treatment that just keeps me alive. My doctor says that not taking it is like suicide. Don't get me wrong, I want to live, but there have been times when I feel that it's not worth it. All of us (her support group), we have a stash of pills which we keep for when the time comes and the pain is so unbearable...There's no need to feel anxious, because when you've had enough of it, you can always take the pills and die. That's the last bit of control one has in one's life and knowing there's that option makes you feel that you still have control. You are choosing to live today'.

Samson: 'Everything just comes down. Bad images start coming up. Like, what will it be like if I go?'

The 'mind-body' connection sub-theme suggested that both Dee and Samson were aware that their emotions and thoughts affected their pain management and that their pain, in turn, affected their emotions and thoughts.

Dee: 'I know that my body will respond to my emotions and um, the connection between one's mood and one's physical self, um, if I am depressed I will feel more sick...when you are busy you are not conscious of your body at all. And when you are pain, it brings your consciousness to your own body; to your physical self...I used to be able to use my willpower to get past this tiredness'.

Samson: 'I don't really have extra pain unless I stress myself...it's like something happens in my brain. Everything just comes down. Bad images start coming up...my health, at that time will get much worse and it won't be fine you know?...Maybe it's body over mind, or mind over body, I don't know...Because the pain is always there, I usually forge ahead. I won't let it stop me'.

The 'mind-body' connection was also related to the sub-theme of hyper-vigilance. The participants used this knowledge of themselves and of their bodies to monitor mood and physical symptoms. Hyper-vigilance was more noticeable in Samson than in Dee, although Dee reported that she was conscious about what was happening in her body '95% of the time', compared to Samson's '80% of the time'.

Dee: 'When you are in pain it brings your consciousness to your own body, to your physical self...I never paid any attention to my body. Now, um, my body is pulling me back to the material me'.

Samson: 'What I usually do is watch out for signs of pain and stay there in my room 'til I know that the pain has almost gone...I will just try to sit there and the pain and the anxiety will eventually go...Where the pain is, that's where my focus will be...The pain will be quite bad and because I'm focussing on the pain it will be worse...I usually try to monitor my body so that I can be very comfortable or be prepared...at any moment I must be aware...I have to monitor my body...I have to be aware of my body all of the time; when I'm working, when I'm thinking, I

must think at what level can I do this? At what level can my body tolerate this?...I'm always aware of my body – every second. I must be aware at every moment...My body is so sensitive. It's sensitive to anything around it...I'm now watching everything around me. I will watch everything to see what will affect my body.'

Reflexivity

Data Collection

The process of interviewing the participants left a powerful impression on me. Both interviews induced feelings of empathy, concern and genuine compassion in me that did resonate for some time after the interviews. The interviews also left me feeling sad and angry on behalf of my participants after hearing some of the difficult experiences the participants shared with me. For example, both participants experienced suicidal ideation and also experienced feeling isolated, helpless, low and misunderstood. There may have been some transference and counter-transference of emotion during these occasions and it was important for me to manage these feelings effectively at that time and afterwards. I used the skills I have learned as a practitioner to stay with the participants' feelings as far as was relevant to the interview process and was safe for the participants to do so and I shared my difficult thoughts and feelings with both my second research supervisor and with my personal therapist. This helped me to explore and unpack the feelings and to think about how these feelings were related to my participants' experiences and to my own internal schemas.

There were occasions during the process of the interviews where I was conflicted about my role as a researcher and my role as a counselling psychologist in training. For example, prior to the interviews I had spoken to my research supervisor about procedures to take if the participants expressed suicidal ideation. So, when the participants spoke about suicidal ideation it was important to evaluate the urgency and context of the risk before considering acting on the risk. My assessment of both situations was that whilst it was difficult for me to hear such painful experiences and difficult thoughts, it was in my participants' best interests for me to maintain the role of the researcher and to listen to my participants' experiences. I had provided a space for them to express their difficult feelings, thoughts and experiences and it

was important for me to be reflective enough to sit with my internal conflict and provide the much needed opportunity for them to express themselves freely and without judgement. In addition, there were occasions where I felt myself wanting to slip into 'CBT mode' where I could have been more directive in my manner and challenged some of the thought distortions that the participants exhibited. Doing this would not have been helpful to the participants at that time and would have been counter-productive to data collection. Being reflective of this process helped me to maintain a researcher role, rather than a counselling role and thus maintain the integrity of the study and also the participants' best interests.

Process of Analysis

I was mindful of how my motivation and my critical realist perspective of the study could influence the decisions that I made regarding the study and also of the impact of these influences on my analysis of the qualitative data. Being aware of and considering these influences provided an awareness that helped me to 'bracket' (Finlay & Gough, 2003) these influences during data collection and analysis. I also considered the potential influence that the first phase of the study had on shaping the interviews and also on influencing the phase II participants' responses.

Thematic analysis does not follow a particular theory, but using Braun & Clarke's (2006) application of thematic analysis, it was important to consider whether I was conducting an inductive or a theoretical thematic analysis. The analysis used an inductive thematic analysis (Braun & Clarke, 2006), which meant that whilst I may have had a general idea about what themes I had been hoping to find, the themes had to emerge from the data and be clearly identifiable, rather than to emerge from pre-developed coding frames or expectations. In addition, it was also important to consider whether I was going to examine themes at the semantic or the interpretative level (Boyatzis, 1998). The analysis was carried out at the interpretative level, where there was an examination of the ideas, assumptions and ideologies that lay under the content of the data (Braun & Clarke, 2006).

Whilst using a thematic analysis was appropriate and invaluable, there were some limitations of using this analysis. Using a thematic analysis did not allow for deeper interpretations of the data other than providing detailed descriptions and groupings of codes and sub-themes. It would have been interesting to explore the

meaning of pain, anxiety and low mood with the participants to unpack any unforeseen connections and interpretations of these words. In addition, the analysis did not allow for an interpretation of language used, or an exploration of the meaning of being able to talk to the researcher about their experiences during the interviews. Pain is a subjective experience and it would have been interesting and maybe revealing to explore these terms and their meaning to the participants in more depth in both the interviews and also in the analysis.

During the analysis it was surprising to see the emergence of personal control and loci of control emerge as an important theme and sub-theme in the analysis. Whilst this finding was unexpected, the literature review indicated that perceptions of personal control can emerge as a theme in qualitative interviews (Caird et al., 2011) and as a variable in quantitative studies (Gibson et al., 2013). The emergence of these themes did increase the validity of adding a qualitative phase and analysis to the study, as the emergence of these themes added insight and richness to the quantitative data.

Summary

In summary, six themes emerged from the interviews: pain appraisal, purpose and change in identity, coping strategies, anger and frustration, social construction of illness and, personal control. Of these themes, personal control was a common thread across all the themes. There were common themes with contrasting categories present from both the interviews, which could indicate that experiencing chronic pain elicits a specific cognitive and/or emotional response that assists in maintaining physical health and survival as this was evidenced in both participants. The quantitative and qualitative results will be triangulated and discussed in the next chapter.

Chapter 6: Discussion

The aim of this sequential explanatory mixed methods study was to investigate how trait anxiety was related to specific health outcomes and pain management in SCD. The first phase of the study was quantitative and investigated relationships between trait anxiety and health outcomes (depression, pain intensity, quality of life scores) in SCD and compared these relationships to those of a

comparison group (BC) and a control group (Carers). The second phase of the study was qualitative and explored pain, anxiety and low mood in a BC and a SCD participant who both had clinical levels of trait anxiety. This chapter will critically discuss the outcome of this study and will consider the implications of the findings with specific relevance to counselling psychology practice.

Findings

The findings from both phases have initially been discussed separately and have been integrated in a separate sub-section.

Quantitative Research Aims and Hypotheses

This study investigated four hypotheses.

H1: Depression scores and pain intensity scores will be positively related to higher trait anxiety scores and quality of life scores will be negatively related to higher trait anxiety scores in the complete sample.

Trait anxiety was significantly predicted by higher depression scores and by lower quality of life scores in the complete sample and in the SCD sub-sample, thus supporting the hypothesis. This finding supports Ristvedt & Trinkaus' (2009) report that higher levels of trait anxiety were significantly related to lower levels of quality of life in rectal patients. High levels of trait anxiety have been found to co-exist with high levels of depression (Thomas et al., 1998; 2001) and whilst high trait anxiety levels have been associated with reduced pain tolerance, rather than with pain perception (James & Hardattordir, 2002; Thibodeau et al., 2013), a positive relationship was found between pain intensity and trait anxiety in this study. It was also interesting to observe that the percentage of variance attributed to depression scores was higher in the SCD sub-sample (67.2%) than in the complete sample (48.1%). An explanation for this could be that the sub-sample was a more homogenous sample compared to the complete sample and this increased the sensitivity of the analysis in observing relationships between the sets of variables (Tabachnick & Fidell, 2007).

The relationship between trait anxiety and depression was earlier reported as being unclear. The current study showed a clear directional relationship between higher levels of trait anxiety and higher levels of depression in a combined sample of

SCD, BC and Carers. In this sample, it is suggested that higher levels of trait anxiety could be related to increased worrying and rumination behaviour leading to an increase in low mood due to a perception of reduced personal control in all three sub-groups (SCD, BC, Carers). Anie et al. (2007), Caird et al. (2011) and Thomas et al. (2001) all reported that the perception of reduced control over illness and circumstances was related to increased levels of depression in SCD. Yarkin et al. (2009) also found a similar relationship between trait anxiety and depression in carers of quadriplegics and paraplegics with pressure sores. Therefore, it is suggested that the perception of personal control may have a role in mediating the relationship between trait anxiety and depression in all of the sub-groups.

A relationship was found between higher levels of pain intensity and higher levels of trait anxiety in the complete sample. Trait anxiety may have caused hyper-vigilant behaviour in all of the sub-groups, which may have caused the participants to notice their pain symptoms sooner than they would have had they not been as vigilant. Whilst the complete sample consisted of two pain groups (SCD, BC) who would be expected to show this relationship between pain intensity and trait anxiety, such a relationship may not be as expected with the Carers sub-group. Sensory pain is not considered to be a main feature of being a Carer, however, the Carers in this sample were exposed to chronic pain and illness. This, in combination with comparatively high trait anxiety levels, may have pre-disposed the Carers in this sample to be sensitive to pain. Distraction techniques are known to be effective in reducing pain perception (Anie et al., 2002; Midence & Elander, 1994; O'Connell-Edwards et al., 2009) and may be an active coping mechanism used after the perception of pain has been noticed, by engaging hyper-vigilant behaviour. Therefore, whilst higher levels of pain intensity are related to higher levels of trait anxiety, it cannot be concluded that the relationship is a causal one as this study was a cross-sectional study. It is suggested that the relationship between pain intensity and trait anxiety in the complete sample was the consequence of hyper-vigilance in the sample.

Higher trait anxiety levels have been shown to be related to lower quality of life in rectal patients (Ristvedt & Trinkaus, 2009) and in this study. It is not surprising that this finding supported the hypothesis. Lower quality of life is a variable associated with heightened sensitivity to distress (Wellington et al., 2010),

depression (Levenson et al., 2008), and increased pain experience (Pells et al., 2005). Therefore, as an increase in these variables is also related to higher trait anxiety levels, it was concluded that trait anxiety would have a negative relationship with quality of life. However, this relationship is not fully understood in this current study and could be investigated with further research.

H2: There will be a significant multivariate main effect for illness group (SCD, BC and Carers) and trait anxiety and depression scores.

The findings only partially supported the hypothesis. The BC group had higher depression scores than both the SCD and the Carer groups. Also, the BC group had significantly higher trait anxiety scores than the Carers, but not compared to the SCD group. However, the SCD group did not have significantly different trait anxiety nor depression scores than the Carers, which had not been predicted.

The BC group had higher depression scores than both the SCD group and the Carers. Whilst this was expected, the finding supports the suggestion that having high trait anxiety levels in SCD may be adaptive, rather than maladaptive. This suggestion is supported by the fact that the SCD group were not clinically depressed (Beck et al., 1996) and had relatively low depression scores despite having clinical levels of trait anxiety. Where other studies have shown a relationship exists between high trait anxiety and high depression scores in pain studies (De Vries et al., 2009; Thomas et al., 1998), this study shows that this was not the case with SCD and provides more evidence for the suggestion that there are different interpretations and expressions of depression in SCD compared to other chronic pain illnesses.

It had been predicted that the BC group would have higher trait anxiety scores than the SCD group because the role of trait anxiety in SCD, as a method of monitoring symptoms and stressors, was hypothesised to be an adaptive coping strategy in SCD, but not in BC. Consequently, since SCD is a disease that is symptomatic from the first year of life (Anie, 2005; Rees, 2003), it was rationalised that clinical levels of trait anxiety would have developed in childhood and adolescence due to the difficulties experienced with the disease (Burlew et al., 2000) and would have been maintained at relatively constant levels through adulthood as a personality trait. Since clinical levels of trait anxiety are related to poorer health outcomes, it was suggested that having excessively high trait anxiety levels would

have been detrimental to both mental and physical health and would not have been sustainable over the lifetime. Consequently, it was predicted that the BC group would have higher scores of trait anxiety because of the comparatively shorter length of time that these participants may have had their disease and because of the change in life circumstances since being diagnosed i.e. from previously not having a life-threatening disease to learning to adapt to having one, therefore experiencing increased distress. Although the BC group did have higher trait anxiety scores ($M = 56.63$; $SD = 11.58$) than the SCD group ($M = 49.75$; $SD = 5.15$), this difference was not statistically significant. In addition, whilst the SCD trait anxiety scores were not significantly different from those of the Carers, the SCD group did have higher trait anxiety scores than the Carers ($M = 43.13$; $SD = 11.25$). The lack of difference in trait anxiety score could be explained by the lack of power in the study caused by the low sample size.

It was expected that the BC group would have higher trait anxiety scores than the Carers and this hypothesis was supported by the study. High trait anxiety levels have been found in breast cancer (Van Esch et al., 2011) and in rectal cancer (Ristvedt & Trinkaus, 2009), so it was rationalised that high trait anxiety levels would also be experienced in the BC group. High levels of trait anxiety have been found in Carers (Kuscu et al., 2009; Pagnini et al., 2012; Sansoni, Vellone & Piras, 2004; Vignola et al., 2008; Yarkin et al., 2009) and have been associated with high depression scores and low quality of life. The trait anxiety scores in Carers in these studies were relatively lower than the clinical group that they were compared to e.g. in amyotrophic lateral sclerosis (Pagnini et al., 2012; Vignola et al., 2008), paraplegics and quadriplegics (Yarkin et al., 2009) and in Alzheimer's Disease (Sansoni et al., 2004). Therefore, it was expected that both the clinical groups would both have higher trait anxiety scores than the Carers, but there was no significant difference between the Carers and the SCD group. The lack of difference in trait anxiety score could be explained by the lack of power in the study caused by the low sample size. This will be discussed in the limitations section.

H3: There will be a significant multivariate main effect for illness group (SCD, BC and Carers) and sensory and affective pain scores (pain intensity).

The SCD group had significantly higher sensory pain scores than the Carers, but not the BC group. This finding shows that the SCD participants did experience more pain than the Carers, but not significantly more pain than the BC group. Whilst it was expected that the SCD group would have significantly higher sensory pain scores than the BC group, a possible explanation for the two groups having similar sensory pain scores is that people with SCD have experienced their disease since birth and so may have developed a greater tolerance for pain than the BC group. Therefore, the sensory pain scores may reflect this increased pain tolerance.

There was no significant difference in affective pain across the groups. This was an interesting finding since the Carer group was a control group, whilst both the SCD group and the BC group were diseases with acknowledged high levels of pain. The implications of this finding are that both the SCD group and the BC group were managing their affective pain effectively and this was reflected in their scores.

Differences in affective and sensory pain scores have been found within different cohorts of SCD participants i.e. in a Jamaican and a UK cohort (Thomas et al., 2001). The Jamaican cohort experienced less affective and sensory pain than the UK cohort. Thomas et al. (2001) attributed this difference to differences in perceived control, socioeconomic status and social support in the samples. This current study did not investigate or control for these variables, so whilst no comparisons can be made regarding these variables, it is possible that the differences in perceived control, socioeconomic status and social support may have biased the affective pain scores in this study.

Thomas et al. (1999) demonstrated that sensory pain may not be affected by emotional factors in SCD by finding that there was no significant difference in sensory pain across three treatment conditions (CBT intervention, attention placebo, waiting list control group). This finding implies that perceived pain is independent of psychosocial stressors. The current study supports this finding, by not finding any significant difference in sensory pain across the illness groups. Further to this, Thomas et al. (1999) also showed that there was a difference in affective pain across treatment conditions, with the participants in the two intervention groups having significantly higher affective pain compared to the control group. Thomas et al.'s (1999) finding implies that there is a social and emotional aspect of pain that exists

within SCD and contributes to the overall sense of pain. Although the current study did not examine affective pain within groups, it showed that there was no difference in affective pain across groups, which could indicate that somatisation was not present within this study. Somatisation is associated with affective pain (Sogultu et al., 2011) and where no difference in affective pain was observed across groups, it has been tentatively concluded that somatisation was not a factor in this study. Wellington et al. (2010) found that pain intensity was predicted by somatisation in SCD. However, their study did not evaluate affective and sensory pain separately, which may have shown that only affective pain was predicted by somatisation in their study.

It is important to differentiate between somatisation and hyper-vigilance in this study, because somatisation has been defined as a pathological attention to health-related factors such as pain, which was not found to be the case in this study. This study suggests that hyper-vigilance to pain in SCD may be beneficial because it can help SCD sufferers to monitor their symptoms and either prevent or prepare for a vaso-occlusive crisis in SCD.

The current study employed the *SF-MPQ* (Melzack, 1987); a well known pain intensity scale that has been utilised with SCD in previous studies (Edwards et al. 2006; Thomas et al., 1999; 2001; Wellington et al., 2010). The benefits of the scale are that it measures both affective and sensory pain, which is important in studies where somatisation may be a confounding issue; an association to affective pain can be evaluated separately from perceived pain. As such, its use in this study helped to improve the reliability, validity and comparability of the pain ratings in this current study.

H4: There will be a significant multivariate main effect for illness group (SCD, BC and Carers) and quality of life subscale scores.

The hypothesis for this research question was partially supported by the findings. The SCD group only had significantly lower general health than the Carers and not the BC group as predicted. This finding, whilst unexpected, means that the SCD group did not have lower general health than the comparison group (BC). Reflecting on the other quantitative findings in this study, it is not surprising that the SCD group had similar levels of quality of life to the BC group because the two

groups had similar levels of trait anxiety, sensory and affective pain. However, the two groups had significantly different depression scores, with the BC group being more depressed than the SCD group. Higher depression scores have been related to lower quality of life scores in previous SCD studies (Anie, 2005; Gibson et al., 2013; Levenson et al., 2008; Taylor et al., 2010). This study, however, showed that the SCD participants had relatively lower depression scores (compared to the comparison group), despite also having lower quality of life scores (compared to the control group). Whilst McClish et al. (2005) did not find that quality of life scores related to anxiety and depression were not significantly lower in SCD compared to haemodialysis and cystic fibrosis participants, the findings of this current study indicated that SCD quality of life scores were lower than those of the control group, but better than those of the clinical comparison group.

This study employed the *SF-36* (Ware & Sherbourne, 1992), which has been used in previous SCD studies (Anie et al., 2002; Asnani et al., 2009; Cummins & Anie, 2003; Edwards et al., 2006; Jenerette et al., 2012; McClish et al., 2005, 2006; Sogutlu et al., 2011) and its subscales are often separated and analysed individually, as this current study has also done. The *SF-36* (Ware & Sherbourne, 1992) has been validated for use in Jamaicans with SCD (Asnani et al., 2009), which indicates the relevance of this quality of life measure for SCD. The SCD mean subscale scores from this study indicated better quality of life compared to the SCD mean subscale scores from Anie et al.'s (2002) study, except for physical functioning which was the same mean score in both samples.

Eliminated Variables

As mentioned earlier, five variables were eliminated from the analyses, even as covariates, due to the lack of relationship with trait anxiety in the preliminary correlations. It was surprising that gender did not correlate significantly with trait anxiety because previous research has indicated that anxiety influences men and women differently (Egloff & Schmunkle, 2004; Leavell & Ford, 1983; Thibodeau et al., 2013) with women supposedly more prone to anxiety than men. However, in studies that investigate pain, it appears that men may experience greater emotional difficulties than women (Leavell & Ford, 1983) and anxiety may influence pain perception in men more than in women (Thibodeau et al., 2013) and that this may

have balanced out any gender differences in the relationship between trait anxiety in this study. In addition, neither Citero et al. (2007) nor McClish et al. (2006) found that gender differences existed in pain experiences in SCD.

It was not surprising that age did not correlate with trait anxiety, because the literature posits that trait anxiety is a construct that is stable over time and can be identified as a personality disposition towards anxiety (Eysenck & Derakshan, 1998; Spielberger & Smith, 1966). This would indicate that trait anxiety levels would remain stable after the emotional and cognitive development phases in childhood and adolescence and, therefore, a significant relationship between trait anxiety and age in this study was not expected, since only adults over the age of 18 participated in the study.

There was no relationship between hospital admission frequency over the previous 12 months and trait anxiety. This was an interesting finding because according to the premise of this thesis, higher levels of trait anxiety were expected to be related to an increasing number of hospital admissions over 12 months. An explanation for this could be that there is stigma concerning hospital attendance which may have been a deterrent to attending hospitals for treatment despite observing or experiencing negative body changes or an impending vaso-occlusive crisis. Jenerette et al. (2014) reported that hospitals were avoided because of negative past treatment, which left sufferers feeling vulnerable and ashamed due to being perceived as drug addicts (Elander et al., 2006; Shapiro et al., 1997; Thomas & Taylor, 2002), or because sufferers did not want to miss life events or every day activities (Jenerette et al., 2014; Thomas & Taylor, 2002).

No relationships were found between trait anxiety and physical functioning (from *SF-36*) or trait anxiety and bodily pain (from the *SF-36*). Since physical functioning is more dependent on pain intensity, physical disability and fatigue (Ameringer & Smith, 2011; Caird et al., 2011; Swanson et al., 2011) than on cognitive functioning, it is not surprising that no direct relationship was found between physical functioning and trait anxiety. It was surprising that no relationship was found between bodily pain and trait anxiety, because it was thought that higher levels of trait anxiety would be related to pain perception due to the symptom perception hypothesis (Watson & Pennebaker, 1989). However, the bodily pain sub-

scale of the *SF-36* only had two items which investigated pain magnitude and pain interference. These items may not have been sufficient to measure or detect a relationship with trait anxiety considering the limited sample size of this study.

Limitations of the Quantitative Phase

An obvious shortcoming of the quantitative phase was the limited sample size that was used for the across illness group comparisons. The sample size was reduced from 51 to 24 to equalise the three illness groups because there were only eight participants in the BC group and the other groups were matched to the size of this group. The reduced sample size did affect the power of the study, thus the probability that the MANOVAs correctly rejected the null hypotheses when the null hypotheses were false was weakened (Field, 2013; Tabachnick & Fidell, 2013) and has affected the generalisability of the study. All of the quantitative findings may have been more robust with a larger sample size and with less variability between the groups. Mertens (1998) recommended that there should be at least 15 participants per variable in correlational designs. This study examined 51 responses per variable to answer the first research question and only eight participants per variable to answer the remaining three quantitative research questions. The study accommodated for this by ensuring the groups used for the MANOVAs were equal in size to help to ensure that the assumptions of homogeneity of variance had not been violated (Field, 2013; Tabachnick & Fidell, 2013). There is heterogeneity within the overall sample and within the groups and future studies could keep this in mind regarding sample size; the more heterogeneity there is within a sample, the larger the sample size needs to be. Effect sizes were included in the quantitative results report to emphasise the size of the comparative differences between the groups (Field, 2013) and also for use as reference for comparisons with other studies.

In addition, the sample used for the study was recruited through the community rather than through the NHS and medical sites. This recruitment strategy was chosen because of the study's perspective; one of the aims of the study was to investigate if trait anxiety was adaptive in SCD. From this perspective and the critical realist perspective, it would not have been prudent to recruit directly from hospital sites because of the implication that patients who are hospitalised are more likely to be emotionally and physically distressed (Anie et al., 2012; Elander et al., 2006;

Harris, Parker & Barker, 1998; Jenerette et al., 2014; Thomas & Taylor, 2002) and are therefore, more likely to report negative health outcomes. This would have been counter-productive to this study and would have biased the study results by the participants being less able to objectively and accurately report how they experience their health outcomes generally. Not recruiting through the NHS did increase the difficulty in obtaining sufficient participant numbers for statistical analysis, but did maintain the integrity of the study.

Another shortcoming of the quantitative aspect of the study is that the survey did not ask the BC participants how long they had cancer. If this variable had been reported, then the age of the SCD participants (length of time having a disease) could have been compared to the length of time since BC diagnosis to remission to examine if there was a relationship between trait anxiety and length of time of illness.

It would have been interesting to investigate the relationship between coping and the health outcomes used in this study. Several SCD studies report assessing coping (Anie et al., 2002; 2007; Cummins & Anie, 2003; Thomas et al., 1999; 2001). These studies used the *Coping Strategies Questionnaire* for Sickle Cell disease, which is a disease-specific measure for use in adults (CSQ; Gil, Abrams, Phillips & Keefe, 1989), with an adapted version available for use in children and adolescents (Gil, Williams, Thompson & Kinney, 1991). This measure assesses coping attempts, negative thinking and passive adherence – factors which are all relevant to the disease. It would have been interesting to examine the relationship between trait anxiety and coping in relation to the experience of pain to examine if there was a relationship between coping and hyper-vigilance.

Several questions were left unanswered by the quantitative results. Pain intensity, quality of life and trait anxiety levels were relatively similar between the SCD and the comparison group, despite a significant difference in depression scores between the illness groups and contrary to the hypotheses. There was no clear reason for the difference in these depression scores and how these differences may have affected pain management in SCD. The following sub-section discusses the results of the qualitative analysis.

Qualitative Research Aim

To explore how a BC participant and an SCD participant with clinical trait anxiety experience pain, anxiety and low mood in order to expand on the findings from the first two research aims by adding to the understanding of the suggested role of trait anxiety in SCD.

The qualitative phase of the study was secondary to the quantitative phase. The third research aim explored the pain, anxiety and low mood experiences in two comparison cases. Only two participants self-selected for phase II, but both participants met the requirements of having elevated trait anxiety levels above the clinical cut-off score of 44 (Spielberger et al., 1983). Six themes emerged from the data: pain appraisal, purpose and change in identity, coping strategies, anger and frustration, social construction of illness and, personal control.

Both participants reported that they experienced pain all of the time and expressed that stress exacerbated their pain. Findings by Anie et al. (2012) support this finding. Anie et al. (2012) showed that mood levels improved when pain levels decreased in SCD, but that a residual more manageable pain remained regardless of this. In addition, one of the themes that emerged from Thomas & Taylor's (2002) study was that SCD was an unremitting disease. There were also differences in the issues that emerged from pain appraisal between the two participants. Sam (SCD) expressed that his pain was more within his locus of control, compared to Dee (BC) who exhibited an external locus of control. Locus of control refers to an individual's perception about the underlying causes for events in a person's life. Rotter (1966) identified the internal locus of control (where there is a belief that outcomes of our actions are self-dependent) and the external locus of control (the belief that events are outside our personal control). Rotter (1966) suggested that whilst having an internal locus of control was more advantageous because it was healthy for a person to believe that they had the ability to control things which they are capable of influencing, an external locus of control was more beneficial in chronic or terminal illness. An external locus of control was thought to remove some of the responsibility that a person would have regarding a chronic or terminal illness, where having personal control could not influence the progression of such an illness (Rotter, 1966). Regarding this study, Sam (SCD) exhibited internal locus of control, which according

to Rotter (1966) would not be as beneficial to him as having external locus of control, considering that he had a chronic and potentially terminal illness. The other difference in pain appraisal between the two participants was that they reported different types of pain: Sam (SCD) reported more specific areas of pain, which have been found to be common pain sites in SCD (McClish et al., 2009), whereas Dee (BC) experienced neuropathy (peripheral nerve damage).

Finding meaning and purpose and experiencing a change in identity were issues that arose for both the participants. Finding a purpose other than experiencing and managing pain was important to both Sam (SCD) and Dee (BC). This finding supports qualitative research by Caird et al. (2011) who found that creating more purposeful meaning independent of SCD appeared to reduce feelings of distress and was related to experiencing a certain degree of external locus of control e.g. by being religious or spiritual.

Coping emerged as an important theme when experiencing anxiety, low mood and pain. Both participants used distraction techniques, social interactions and the internet to manage their pain and distress. Social support has been found to be helpful in managing distress and the emotions related to poor pain control (Caird et al., 2011; Jenerette et al., 2014; Thomas & Taylor, 2002). Distraction techniques are well known in pain management literature (Adam et al., 1996; Eccleston et al., 2001, Linton & Shaw, 2011). Using the internet to connect to social networks and maintain social support and purpose appears to be a more recent and helpful communication tool for people suffering from chronic pain. It may help to prevent chronic pain sufferers from feeling isolated when they are physically restricted by their pain or disease-specific symptoms and can improve access to friends and family.

Anger and frustration were emotions that appeared frequently in the interviews. For Dee (BC) the anger was mostly about her conflict about her changing identity and the sense of powerlessness she experienced because of this internal conflict. For Sam (SCD), his anger was linked to other people's negative perceptions of him, because of his disease, and fuelled his sense of purpose. Personal control and finding a balance between external and internal loci of control were associated to anger in both participants. Levels of anger and frustration increased in both participants when they felt increasingly powerless. This association demonstrates

the value in examining and trying to improve pain control and the sense of purpose in chronic pain conditions; negative emotion levels and distress appear to increase when a reduced sense of self-efficacy and/or powerlessness is felt. Research by Anie et al. (2010) and Taylor & Thomas (2002) support this finding.

A lack of understanding from medical professionals was more of an issue for Dee (BC) than for Sam (SCD) as it seemed, from her perspective, that her local doctors were not cognisant of her symptoms and presentation and therefore could not help her appropriately. In SCD, a lack of understanding from medical professionals is more linked to misunderstanding the need for medication and analgesics (Edwards et al., 2006; Elander et al., 2003; 2004; 2006; Howard et al., 2005; Maxwell et al., 1999; Solomon, 2010; Thomas & Cohn, 2006) and to being passively adherent (Anie et al., 2007; 2012; Thomas & Taylor, 2002).

Personal control was an important theme that emerged from the interviews. With both participants, feelings of powerlessness and reduced self-efficacy were related to anger and frustration and to low mood. These findings are supported by previous studies (Caird et al., 2011; Thomas & Taylor, 2002). The participants attempted to regain their sense of control through suicidal ideation or through hyper-vigilance behaviours. Hyper-vigilant behaviour was expressed more by Sam (SCD) than by Dee (BC). Sam (SCD) used his hyper-vigilant behaviour to monitor signs of pain to prepare himself for or, to try to prevent a vaso-occlusive crisis from occurring. Doing this appeared to help his sense of personal control over his pain management as he was able to either prepare for, or prevent a crisis. Hyper-vigilance may have been adaptive in this circumstance compared to other studies of hyper-vigilance where health outcomes were reduced because of increased hyper-vigilance behaviours (Crombez et al., 2004; McDermid, et al., 1996).

Advantages and Disadvantages of Using Qualitative Methodology

There were several advantages and disadvantages of using qualitative methodology. Both participants were interviewed over the telephone due to geographical distances. Whilst both participants had been recruited in the UK, both had travelled abroad since they had participated in Phase I and were thus interviewed whilst they were abroad. Although it can be difficult to monitor emotional distress over the telephone, counselling psychology practice requires a high level of

skill in attending to voice nuances (Egan, 2010) and the researcher was able to use this skill to attend to emotional distress in the participants. The fact that the participants' primary residence was abroad may have added uncontrollable variance to the study. For example, the participants may have had different experiences in healthcare and stigmatization in these other countries compared to predominantly UK participants. However, this current study's findings have not been generalised to the UK population and the study has been positioned as a 'pilot' or introductory study whose findings require future research with a larger, more controlled sample size.

Semi-structured interviewing was used to interview the participants (see Appendix D). This followed the critical realist position that specific research questions needed to be answered, but that an informal approach allowed the participants to 'feel' their way through their thinking with the guidance of the researcher. This process enabled the conversation to flow more naturally, in a way similar to a real world conversation (Robson, 2011). Other advantages of using this method of interviewing were that there was freedom for the participant to explore unpredicted avenues of thought and provide data-rich responses and there was also interviewer flexibility in selecting aspects of the discourse to follow up (Coolican, 2001). The disadvantages of using this method were that there was weak reliability between the participants, according to the post-positivist view (Creswell, 2009; Creswell & Plano Clark, 2006) but this was to be expected considering this was a socially constructed method of data collection and participants experiencing two different diseases were interviewed. This form of data collection was time consuming because of the flexibility of the structure of the questioning and the time taken to transcribe the interviews.

This study used a thematic analysis guided by Braun & Clarke (2006). This method of analysis was advantageous to the study as it summarised key features of the data and was not tied to a particular level of interpretation (Robson, 2011). This method also fit the critical realist paradigm (Bhaskar, 1988). However, this form of analysis limited the study to describing themes. Further exploration in the interviews and an in-depth data analysis would be required for more in-depth subjective experiences to be explored. Another disadvantage of using thematic analysis to analyse the data was that researcher reliability was reduced as the interpretation of themes was subjective to the researcher with supervisory support. It was also

important to be aware of internal consistency in the researcher and of researcher reflexivity. This was achieved by receiving supervisor feedback, keeping a record of analysis and analytical process and by examining researcher process throughout the analysis.

Integration of Quantitative and Qualitative Findings

Depending on the perspective of the reader, the first advantage/limitation of the study was that it was a mixed method study. Despite its difficulties, the integration of quantitative and qualitative elements was beneficial to the study and provided a holistic view of trait anxiety in SCD and how it may be related to pain management. From a critical realist perspective, it was an advantage and a privilege to be able to investigate and explore the personal experiences regarding pain. Reality was tested in the quantitative phase and was subjectively explored when it was observed that testing reality failed to provide a whole truth. Trait anxiety is a quantitative construct which needed to be quantified in the participants first; subsequently, potential relationships to pain management needed to be investigated and established. Once this was completed, it was apparent that further explorations were needed to source alternative issues or constructs that would enhance the understanding of the concept of trait anxiety in SCD and its relationship to pain management.

There were two occasions where there was mixing of the quantitative and qualitative data. The first point was at the intermediate stage when participants for the second phase were selected based on the responses given in the first phase. Both participants were selected to be interviewed for the qualitative phase because they had clinical trait anxiety; trait anxiety scores greater than 44 (Spielberger et al., 1983). The second point of integration was in this section where there is both complementary and convergent triangulation (Ivankova, Creswell & Stick, 2006; Ostlund et al., 2011).

In the first phase of the study, it was shown that both the SCD and BC participants had higher levels of trait anxiety than the controls. Although the SCD group's mean trait anxiety score was not significantly different to the BC group's mean score, the results from phase II show that the SCD participant reported more hyper-vigilant behaviours than the BC participant did . This supports the argument

that the role of trait anxiety in SCD may be different to the role of trait anxiety in BC. Past research has argued that hyper-vigilance in fibromyalgia was related to increased pain intensity and lower pain thresholds (McDermid et al., 1996) and to increased pain intensity, higher levels of distress and higher levels of catastrophic thinking about pain (Crombez et al., 2004). In this current study there was no significant difference in sensory or affective pain between the SCD and the BC group, which indicated that increased hyper-vigilance did not always lead to increased pain intensity across groups and the relationship with and purpose of hyper-vigilance and pain may be different across pain groups. For example, hyper-vigilance was associated with lower health outcomes in fibromyalgia compared to chronic low back pain (Crombez et al., 2004) and between fibromyalgia and rheumatoid arthritis (McDermid et al., 1996).

Hyper-vigilance was not measured directly in this study. However, a relationship between high levels of trait anxiety and hyper-vigilance has been established in the literature (Cisler, Bacon & Williams, 2009; Crombez et al., 2004; Eysenck, 1992; Ouimet, Gawronski & Dozois, 2009; Yiend, 2010). In addition, experimental studies have evidenced a relationship between high levels of trait anxiety and vigilance for threat (Broadbent & Broadbent, 1988; Mogg, Bradley & Hallowell, 1994), thus showing a link between the concept of hyper-vigilance and trait anxiety. The *Pennebaker Inventory for Limbic Languidness* (Pennebaker, 1982) has been used to measure hyper-vigilance (McDermid et al., 1996), but does not specifically measure hyper-vigilance. This inventory was inappropriate for use in this study because participants would have to rate common symptoms and bodily sensations on a 5-item rating scale that has items that are genuine symptoms for both SCD and BC in differing degrees, for example, swollen joints are persistent symptoms in SCD. Therefore, using this measure may have biased the results. In addition, the *Pain Vigilance and Awareness Questionnaire* (McCracken, 1997) is a broad measure of attention to pain which can be applied to various pain populations. This measure was not used in this study because of its focus on attention to pain, whereas the objective of this study was to measure how hyper-vigilance could be an adaptive construct. It was decided to use a more generic hyper-vigilance measure i.e. *STAI-T*, that did not focus specifically on pain.

Another interesting finding was that the BC group had higher depression scores than the SCD group. The BC participant in phase II was shown to have a higher depression score and a more external locus of control compared to the SCD participant. Benassi, Sweeney & Dufour (1988) conducted a meta-analysis that effectively showed that external locus of control was related to depression scores; greater externality was significantly associated with higher depression scores and this relationship was consistent across the studies they investigated. The findings from this current study do appear to support this relationship. Thus, it could be suggested that exhibiting an internal locus of control could be related to lower depression scores. Applying this hypothesis to this study, it is suggested that the SCD participant simultaneously had clinical levels of trait anxiety and a non-clinical depression score because he exhibited an internal locus of control. This is an interesting finding because past studies have indicated that high trait anxiety is reported with high levels of depression as reported in literature review, but this was not the case with the SCD participant in phase II. This finding is supported by Thomas et al. (1999) who found that reducing anxiety and depression in SCD did reduce external locus of control perceptions, and by Gibson et al. (2013) who found that internal locus of control was associated with higher quality of life scores and lower depression scores.

Phase II also showed that feeling powerless was linked to distress and was linked to locus of control. A shortcoming of the current study is that locus of control was not quantitatively examined. Locus of control has been examined in SCD (Anie et al., 2007; Gibson et al., 2013; Thomas et al., 1999) and has demonstrated the importance of considering the relationship between locus of control and depression in SCD. Future studies could examine the relationship between locus of control and hyper-vigilance to examine if being hyper-vigilant in SCD is related to a greater sense of pain control, thus improving their pain management.

An unexpected finding was that both participants, despite having elevated levels of clinical anxiety, both identified experiencing anger and frustration rather than anxiety. This is an interesting finding because it directly links the emotions of anxiety and anger. One interpretation is that expressions of anger and frustration in chronic illness could be an indication of underlying high levels of anxiety and that the higher the levels of anxiety, the more likely feelings of anger and frustration will be

expressed and projected. Future research could investigate the relationships between trait anxiety, depression and loci of control to determine if relationships exist between the three variables and what direction, or influence these variables have on each other in chronic pain illnesses.

This study did show that having a life purpose that was separate to managing pain may protect against depression and distress. The internet was shown to be a useful medium and coping strategy to communicate with 'the outside' world when isolated due to physical disability and or prevention of pain.

Coping was not quantitatively assessed in this study. Coping has been extensively researched in SCD (Anie et al., 2002; 2007; Cummins & Anie, 2003; Simon et al., 2009; Thomas et al., 1999; 2001); passive adherence coping is associated with higher levels of depression and anxiety and active coping is associated with an internal locus of control. Investigating coping in this study may have provided some insight into how coping may have mediated the potential relationships between loci of control, trait anxiety and how pain is managed in SCD.

There are a number of advantages of using the mixed method approach for this study. Using both quantitative and qualitative approaches produced a more complete understanding of how trait anxiety affected health outcomes in SCD. In addition, there were two different types of research questions being asked: one type required nomothetic responses, the other required idiographic responses; hence the necessity for this mixed approach. The combination of research approaches is transferable to real world settings because of the complexity of the phenomena concerning the approach that mimics real world situations (Robson, 2011) and consequently increases the generalizability of the study's results (Creswell, 2009; Pluye et al., 2009). However, the findings from this study cannot be generalised due to the relatively small sample size.

Using this type of design was time consuming (Burke & Onwuegbuzie, 2004) and progress to phase II was hampered by the low sample size and recruitment difficulties experienced in phase I. It was also possible that the research outcomes were the result of heterogeneity which the study was unable to control for. The qualitative part of the study depended on interviewing an SCD and a BC participant, but was dependent on participants in phase I being willing to participate in phase II

and meeting the criteria of having clinical levels of trait anxiety. Another difficulty of using mixed method designs in this study was that it was difficult to judge what had been gained by utilising both approaches in the study. It was especially important during this study to constantly reflect and examine what the intention of the individual phases of the study were and what was being gained from integrating them. This process helped to maintain the integrity of both approaches in the study and maintain an integrated study. This was done by maintaining research supervision, by reviewing research process and by analysing data as it was received.

In general, it appears that integrating quantitative and qualitative data and methods was beneficial to the study and helped to answer the research questions holistically.

Implications, Relevance and Recommendations of the Study

Implications for Practice and Relevance to Counselling Psychology

This study highlights that psychological variables do affect pain management in SCD. Thomas et al. (1998; 1999) showed that a CBT intervention to improve pain management was only effective for 6 months. After 6 months, levels of anxiety and depression went back to pre-intervention levels. Thomas et al. (1999) suggested that this return to pre-intervention levels of distress, anxiety and depression was related to a gradual increase in 'less adaptive' coping strategies over the six month period after the CBT intervention had been completed, for example, noticing and acting on pain sensations and focussing attention onto the body. It may be worth considering if this occurred because elevated trait anxiety levels in SCD may have facilitated hyper-vigilant behaviour that monitored bodily symptoms to prevent or prepare for vaso-occlusive crises. According to this theory, engaging strategies to actively reduce anxiety levels could lead to more vaso-occlusive crises being experienced, which would increase levels of powerlessness and distress in SCD and thus may not be beneficial to pain management. Health professionals developing psychological interventions for pain management in SCD may need to consider supporting hyper-vigilant behaviour to a degree where it is not counter-productive and could lead to increased pain intensity as reported in previous studies (Crombez et al., 2004; McDermid et al., 1996).

It is important for psychological practitioners to be mindful that anxiety and depression can interact in different ways in different pain conditions. This study demonstrates this and supports the argument that pre-intervention anxiety can predict patterns of change in treatments for depression and that practitioners need to be mindful of the lack of ability to generalise regarding all pain conditions, but SCD in particular. Forand & De Rubeis (2013) support this argument. In addition, practitioners may need to be mindful that expressions of anger and frustration may indicate underlying and unrecognised anxiety that may need to be addressed as a primary concern before addressing emotions of anger and frustration.

The qualitative results indicated that psychological practitioners should consider developing interventions that focus on exploring anger and frustration, rather than depression. Whilst these emotions are related, it may be that the approach taken may focus on reducing depression by managing anger and increasing the perception of personal control, rather than focussing generally on reducing anxiety and depression. It was also shown that how health professionals view SCD participants can affect healthcare utilisation and passive adherence coping. Consequently, it is important that healthcare practitioners are more informed in their decision-making regarding patient care in SCD and are more sensitive to cultural beliefs about pain management.

Counselling Psychologists work within general medicine settings where SCD clients present with acute and chronic pain and distress. This study demonstrates the importance of considering the role that psychological variables play in pain management; however, it is also important to consider that SCD clients may not believe that psychology plays a role in pain management. It may be difficult to access SCD clients because of this belief and increasing awareness of the counselling psychology role in pain management may be an essential part of the role of a counselling psychologist.

Implications for Research

As mentioned earlier, future research could investigate the effectiveness of personal control/anger management interventions for pain management. A longitudinal study examining trait anxiety from childhood through to adolescence could identify if trait anxiety is a trait that develops in the early years of the disease to

manage pain in the illness, or if it is a situational trait regarding the disease. A shortcoming of the survey used in this study is that it did not measure the time between diagnoses to recruitment of the BC participants. Thus, a comparison between SCD and BC examining the effects of the longevity of the diseases was not possible in the study. A comparison can be made by examining the relationship between the ages of SCD participants and the BC participants' time from diagnosis. It would also be interesting to compare the relationship between trait anxiety and pain in SCD compared to fibromyalgia to examine the effect of hyper-vigilance on the experience of pain. An in-depth phenomenological approach to exploring the experience of SCD may give a more thorough understanding of the experience of the disease and of how locus of control and/or hyper-vigilance may affect pain management.

Reviewing Smith et al.'s (2005) draft conceptual model, it was observed that the model emphasised healthcare utilisation rather than pain experience. The current study placed emphasis on pain management, with pain experience being the most prominent and persistent feature of the disease. Considering this and integrating the current study's findings into the model (see Figure 3), it has been suggested that there is a directional link between disease – related variables and psychosocial variables that was not in the original model. This study demonstrated that treatment and pain location can affect quality of life, coping behaviours (hyper-vigilance) and social support and that these variables can also affect treatment efficacy and pain location. It is suggested that the relationship between distress disability and psychosocial behaviours is a two-way relationship because distress disability was found to be related to hyper-vigilance (coping behaviours). Finally, the findings from this study suggest that there is a two-way relationship between readiness variables and pain. It is hypothesised that a relationship exists between hyper-vigilance of the perceived threat of pain and pain experience/management and also that pain intensity and experience can affect the perceived threat. Future studies can focus on improving and refining the model and evaluating the relevance of the suggestions that this study has made.

Conclusion

In conclusion, the findings from this study suggest that trait anxiety plays a role in SCD; hyper-vigilant behaviour (as represented by elevated levels of trait anxiety in SCD) may help sufferers to monitor their physical symptoms and alert them to negative changes in order to prevent or prepare for vaso-occlusive crises. By investigating the relationship between trait anxiety and several health outcomes it was possible to refine the specific variables that needed to be explored further through subjective experience. It is hoped that this thesis has added to the psychological knowledge about SCD and has highlighted the impact of psychological processes on pain management in SCD in adults. This study could have focussed on either the quantitative or the qualitative phase, but it was only by mixing methodologies that a better understanding of the research problem was achieved. Several avenues for future research have been highlighted throughout the discussion and it is hoped that the results of this study will be productive and beneficial for the people who suffer from Sickle Cell Disease.

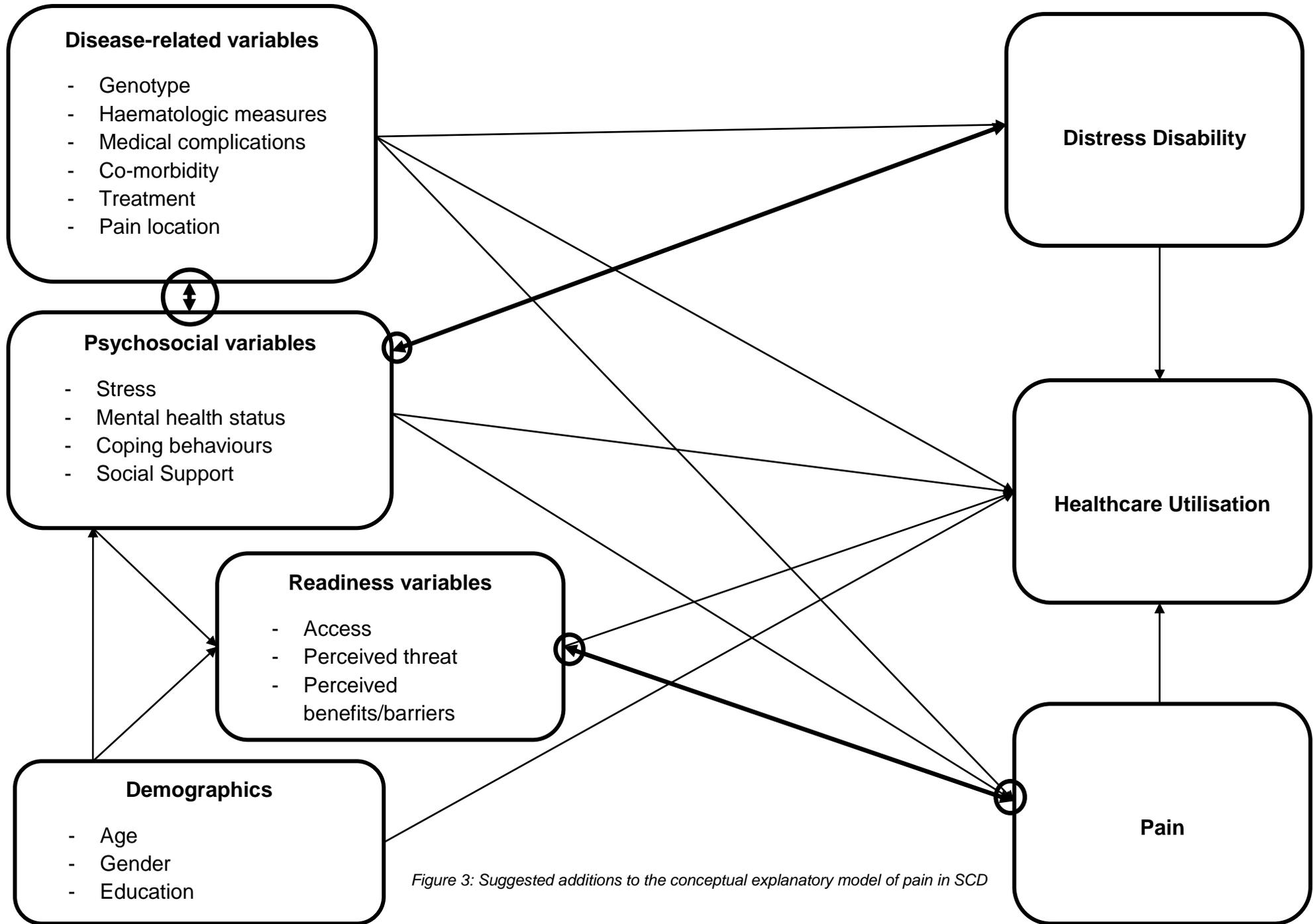


Figure 3: Suggested additions to the conceptual explanatory model of pain in SCD

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Appendix A
Ethical Approval Documents

ETHICAL PRACTICE CHECKLIST (Professional Doctorates)

SUPERVISOR: Meredith Terlecki

ASSESSOR: Virginia Lam

STUDENT: Dede-Kossi Osakonor

DATE (sent to assessor): 26/06/2012

Proposed research topic: Trait anxiety in Sickle Cell Disease: investigating and exploring links to the management of pain in Sickle Cell Disease.

Course: Prof. Doc Counselling Psychology

1. Will free and informed consent of participants be obtained? YES
2. If there is any deception is it justified? N/A
3. Will information obtained remain confidential? YES
4. Will participants be made aware of their right to withdraw at any time? YES
5. Will participants be adequately debriefed? YES—see below
6. If this study involves observation does it respect participants' privacy? NA
7. If the proposal involves participants whose free and informed consent may be in question (e.g. for reasons of age, mental or emotional incapacity), are they treated ethically? NA
8. Is procedure that might cause distress to participants ethical? NA
9. If there are inducements to take part in the project is this ethical? NA
10. If there are any other ethical issues involved, are they a problem? NA

APPROVED

| | | |
|-----|--------------------------------------|----|
| YES | YES, PENDING MINOR CONDITIONS | NO |
|-----|--------------------------------------|----|

MINOR CONDITIONS: Debriefing (in the form of a passage after participation) should also be available for online participation of phase 1.

Assessor initials: VL

Date: 3 July 2012

RESEARCHER RISK ASSESSMENT CHECKLIST (BSc/MSc/MA)

SUPERVISOR: Meredith Terlecki

ASSESSOR: Virginia Lam

STUDENT: Dede-Kossi Osakonor

DATE (sent to assessor): 26/06/2012

Proposed research topic: Trait anxiety in Sickle Cell Disease: investigating and exploring links to the management of pain in Sickle Cell Disease.

Course: Prof. Doc Counselling Psychology

Would the proposed project expose the researcher to any of the following kinds of hazard?

- 1. Emotional NO
- 2. Physical NO
- 3. Other NO

(e.g. health & safety issues)

If you've answered YES to any of the above please estimate the chance of the researcher being harmed as: HIGH / MED / LOW

APPROVED

| | | |
|-----|----------------------------------|----|
| YES | YES, PENDING MINOR CONDITIONS | NO |
|-----|----------------------------------|----|

MINOR CONDITIONS: N/A

REASONS FOR NON APPROVAL: N/A

Assessor initials: VL

Date: 3 July 2012

SCHOOL OF PSYCHOLOGY

Dean: Professor Mark N. O. Davies, PhD, CPsychol, CBiol.



**School of Psychology
Professional Doctorate Programmes**

To Whom It May Concern:

This is to confirm that the Professional Doctorate candidate named in the attached ethics approval is conducting research as part of the requirements of the Professional Doctorate programme on which he/she is enrolled.

The Research Ethics Committee of the School of Psychology, University of East London, has approved this candidate's research ethics application and he/she is therefore covered by the University's indemnity insurance policy while conducting the research. This policy should normally cover for any untoward event. The University does not offer 'no fault' cover, so in the event of an untoward occurrence leading to a claim against the institution, the claimant would be obliged to bring an action against the University and seek compensation through the courts.

As the candidate is a student of the University of East London, the University will act as the sponsor of his/her research. UEL will also fund expenses arising from the research, such as photocopying and postage.

Yours faithfully,

Dr. Mark Finn

Chair of the School of Psychology Ethics Sub-Committee

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The University of East London has campuses at London Docklands and Stratford
If you have any special access or communication requirements for your visit, please let us know. MINICOM 020 8223 2853



UNIVERSITY OF EAST LONDON

School of Psychology

REQUEST FOR AMENDMENT TO AN ETHICS APPLICATION

FOR BSc, MSc/MA & TAUGHT PROFESSIONAL DOCTORATE STUDENTS

Please complete this form if you are requesting approval for proposed amendment(s) to an ethics application that has been approved by the School of Psychology.

Name of applicant: Dede-Kossi Osakonor

Programme of study: Professional Doctorate in Counselling Psychology

Title of research: Trait anxiety in Sickle Cell Disease: investigating and exploring links to the management of pain

Name of supervisor: Dr Meredith Terlecki

Briefly outline the nature of your proposed amendment(s) and associated rationale(s) in the boxes below

| Proposed amendment | Rationale |
|--|---|
| I would like to change the comparison group from Bowel cancer to Blood Cancer as I have found it difficult to recruit Bowel Cancer participants. | My study is a mixed-method study which includes three participant groups: Sickle Cell Disease, Bowel Cancer and Carers. This change does not affect any aspect of the study, other than that the comparison group will be Blood cancer, rather than Bowel cancer. |
| To be able to interview participants over the telephone, in addition to the previously | The ethical approval I received includes me collecting qualitative data through face-to-face semi-structured interviews. I am asking to conduct recorded interviews through the |

| | |
|--|--|
| approved method of interviewing in person. | telephone for the convenience of the participants who may find it difficult to physically attend meetings. |
|--|--|

| Please tick | YES | NO |
|---|-----|----|
| Is your supervisor aware of your proposed amendment(s) and agree to them? | X | |

Student's signature (please type your name): Dede-Kossi Osakonor

Date: 13.04.2014

| TO BE COMPLETED BY REVIEWER | | |
|------------------------------|-----|--|
| Amendment(s) approved | YES | |
| Comments | | |

Reviewer: M. Finn

Date: 21/04/14

Appendix B

Participant Consent Forms and Information Sheets

UNIVERSITY OF EAST LONDON

School of Psychology
Stratford Campus
Water Lane
London E15 4LZ

The Principal Investigator(s)

Dede-Kossi Osakonor
U1019289@uel.ac.uk

I would like to invite you to participate in a research study that is being conducted as part of my Professional Doctorate in Counselling Psychology degree at the University of East London.

Project Description

The study is exploring how mood affects pain management and quality of life in Sickle Cell Disease, Blood Cancer and in Carers. Participants will be asked to go to this website <http://www.surveymonkey.com/s/UELpainmood2013> and complete as much of it as possible. The questions should not take more than 15 minutes to answer and you can save your answers and return to complete the questionnaire when you have time.

It is possible that participants may feel distressed during or after completing the questionnaire – counselling agencies and help lines have been provided at the end of the questionnaire to help with the distress.

Confidentiality of the Data and safety

Your data will be anonymous unless you wish to participate in the 2nd phase of the study as you will have to provide your contact details to be contacted to participate in the 2nd phase of the study. However the data provided in the 2nd phase of the study will be anonymised and will not be linked to any contact details or personal information. The primary researcher will be the only person who has access to the data.

All the data will be kept electronically for 3 years after the conclusion of the study on a password protected external hard-drive. After 3 years the electronic data file will be deleted and only reports and articles summarising the data will continue to exist.

You are not obliged to take part in this study. You are free to withdraw at any time. Should you choose to withdraw from the study you may do so without any consequences.

Please feel free to email me any questions that you may have at u1019289@uel.ac.uk

If you have any questions or concerns about how the study has been conducted, please contact the study's supervisor: Dr Meredith Terlecki, School of Psychology, University of East London, Water Lane, London E15 4LZ, +44 (0)20 8223 4463, m.terlecki@uel.ac.uk

Or

Chair of the School of Psychology Research Ethics Sub-committee: Dr. Mark Finn, School of Psychology, University of East London, Water Lane, London E15 4LZ, 020 8223 4493, m.finn@uel.ac.uk

Thank you.

Dede-Kossi Osakonor

UNIVERSITY OF EAST LONDON
School of Psychology
Stratford Campus
Water Lane
London E15 4LZ

The Principal Investigator
Dede-Kossi Osakonor

U1019289@uel.ac.uk

The purpose of this letter is to provide you with the information that you need to consider in deciding whether to participate a research study. The study is being conducted as part of my Professional Doctorate in Counselling Psychology degree at the University of East London.

Project Title

Trait anxiety in Sickle Cell Disease: investigating and exploring links to the management of pain in Sickle Cell Disease.

Project Description

The study is exploring how mood affects pain management and quality of life in Sickle Cell Disease and in Blood Cancer. Participants will be asked questions over a 50 minute period in a one-to-one interview about how they describe their moods, their hospital admissions, their pain and their quality of life. The interviews will be audio recorded and participants will be able to ask questions about the research and about the questions that will be asked. The interviews may also be conducted over the telephone if participants have mobility difficulties and may find it difficult to physically attend a meeting. The researcher's phone will not be on speakerphone during this interview and the researcher will be alone in a private and secure room to ensure participant confidentiality and privacy. The telephone interviews will also be recorded through a recording device application on the researcher's phone.

It is possible that participants may feel distressed during or after the interview and counselling agencies and help lines will be provided to help with the distress.

Confidentiality of the Data and safety

The interviews will be coded by providing a serial number on the interview that will be stored separately from contact information. The primary researcher will be the only person who has access to either form of data which will not be shared with anyone else. In the transcriptions all identifiable names will be changed. The audio data will be stored digitally on a password locked external hard-drive.

All the data will be kept electronically for 3 years after the conclusion of the study on a password protected external hard-drive. All paper records will be destroyed at the end of the study.

The interviews will take place at a room in your local support group in London, or over the telephone.

You are not obliged to take part in this study and should not feel coerced. You are free to withdraw at any time. Should you choose to withdraw from the study you may do so without disadvantage to yourself and without any obligation to give a reason.

Please feel free to ask me any questions. If you are happy to continue you will be asked to sign a consent form prior to your participation. Please retain this invitation letter for reference.

If you have any questions or concerns about how the study has been conducted, please contact the study's supervisor: Dr Meredith Terlecki, School of Psychology, University of East London, Water Lane, London E15 4LZ, +44 (0)20 8223 4463, m.terlecki@uel.ac.uk

or

Chair of the School of Psychology Research Ethics Sub-committee: Dr. Mark Finn, School of Psychology, University of East London, Water Lane, London E15 4LZ.

(Tel: 020 8223 4493. Email: m.finn@uel.ac.uk)

Thank you in anticipation.

Yours sincerely,

Dede-Kossi Osakonor

UNIVERSITY OF EAST LONDON

Consent to participate in a research study

Trait anxiety in Sickle Cell Disease: investigating and exploring links to the management of pain in Sickle Cell Disease.

I have read the information sheet relating to the above research study and have been given a copy to keep. The nature and purposes of the research have been explained to me, and I have had the opportunity to discuss the details and ask questions about this information. I understand what is being proposed and the procedures in which I will be involved have been explained to me.

I understand that my involvement in this study, and particular data from this research, will remain strictly confidential. Only the researcher involved in the study will have access to identifying data. It has been explained to me what will happen once the research study has been completed.

I hereby freely and fully consent to participate in the study which has been fully explained to me. Having given this consent I understand that I have the right to withdraw from the study at any time without disadvantage to myself and without being obliged to give any reason.

Participant's Name (BLOCK CAPITALS)

.....

Participant's Signature

.....

Researcher's Name (BLOCK CAPITALS)

.....

Researcher's Signature

.....

Date:

Appendix C

Survey Questions and End of Survey Debrief

Participant Information

I would like to invite you to participate in a research study that is being conducted as part of my Professional Doctorate in Counselling Psychology degree at the University of East London. Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully.

Project Description

The study is exploring how mood affects pain management and quality of life in Sickle Cell Disease, Blood Cancer and in Carers. Participants will be asked to go to this website <https://www.surveymonkey.com/s/UELpainmood2013> and complete as much of it as possible. The questions should not take more than 15 minutes to answer and you can return to complete the questionnaire when you have time.

It is possible that participants may feel distressed during or after completing the questionnaire – counselling agencies and help lines have been provided at the end of the questionnaire to help with the distress.

Confidentiality of the Data and safety

Your data will be anonymous unless you wish to participate in the 2nd phase of the study. The primary researcher will be the only person who has access to the data.

All the data will be kept electronically for 3 years after the conclusion of the study on a password protected external hard-drive. After 3 years the electronic data file will be deleted and only reports and articles summarising the data will continue to exist.

You are not obliged to take part in this study. You are free to withdraw at any time. Should you choose to withdraw from the study you may do so without any consequences.

Please feel free to email me any questions that you may have at u1019289@uel.ac.uk

If you have any questions or concerns about how the study has been conducted, please contact the study's supervisor: Dr Meredith Terlecki, School of Psychology, University of East London, Water Lane, London E15 4LZ, +44 (0)20 8223 4463, m.terlecki@uel.ac.uk

Or

Chair of the School of Psychology Research Ethics Sub-committee: Dr. Mark Finn, School of Psychology, University of East London, Water Lane, London E15 4LZ, 020 8223 4493, m.finn@uel.ac.uk

Thank you.

Dede-Kossi Osakonor

Participant Consent Form

*** 1. I would like to invite you to participate in a research study that is being conducted as part of my Professional Doctorate in Counselling Psychology degree at the University of East London. Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully.**

Project Description

The study is exploring how mood affects pain management and quality of life in Sickle Cell Disease, Blood Cancer and in Carers. Participants will be asked to go to this website

<https://www.surveymonkey.com/s/UELpainmood2013> and complete as much of it as possible. The questions should not take more than 15 minutes to answer and you can save your answers and return to complete the questionnaire when you have time.

It is possible that participants may feel distressed during or after completing the questionnaire – counselling agencies and help lines have been provided at the end of the questionnaire to help with the distress.

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You are not obliged to take part in this study. You are free to withdraw at any time. Should you choose to withdraw from the study you may do so without any consequences.

Please feel free to email me any questions that you may have at u1019289@uel.ac.uk If you have any questions or concerns about how the study has been conducted, please contact the study's supervisor: Dr Meredith Terlecki, School of Psychology, University of East London, Water Lane, London E15 4LZ, +44 (0)20 8223 4463,

m.terlecki@uel.ac.uk

Or

Chair of the School of Psychology Research Ethics Sub-committee: Dr. Mark Finn, School of Psychology, University of East London, Water Lane, London E15 4LZ, 020

8223 4493, m.finn@uel.ac.uk

Thank you.

**Dede-Kossi
Osakonor**

I agree to participate

I do not wish to participate

Participant Group

*2. Which of these applies to you?

I have sickle cell disease

I have thalassaemia

I have a blood cancer

I am a carer

(please write what type in the
'other' box)

Other (please specify)

3. What is your gender?

- Male
- Female

*4. Do you consider yourself to have a disability?

- No
- Physical impairment
- Sensory impairment
- Mental health condition
- Learning disability/difficulty
- Long-standing illness
- I do not wish to disclose
- Other

Other (please specify)

5. How old are you?

Years

***6. What is your ethnic origin?**

- Black or Black British African
- Black or Black British Caribbean
- Any other Black background
- White and Black African
- White and Black Caribbean
- White and Asian
- Any other mixed race background
- White British
- White Irish
- White European
- Other white background
- Asian or Asian British Indian
- Asian or Asian British Bangladeshi
- Asian or Asian British Pakistani
- Other asian background
- Chinese
- Other chinese background

Other (please specify)

***7. How many times have you been admitted to hospital because of your illness within the last 12 months?**

***8. How many unbearably painful episodes did you experience within the last month?**

9. Have you had any psychological therapy to help you manage your illness?

YES, please specify:

NO

10. What is your marital status?

- Single
- Married
- Civil partnership
- Divorced
- Widowed
- Separated
- A member of an unmarried couple

11. Do you currently have a carer?

- Yes, full time
- Yes, part time
- No

12. Are you employed?

- Yes, full time
- Yes, part time
- I am self-employed
- No, I am unable to work
- No, I am retired
- No, I am a homemaker/housewife/househusband
- No, I am a student
- No, I am unable to find work

Other (please specify)

A number of statements which people have used to describe themselves are given below. Read each statement and then choose the statement that applies to you and that indicates how you generally feel. There are no right or wrong answers. Do not spend too much time on any one statement and click the answer which seems to describe how you generally feel. PLEASE RESPOND TO ALL ITEMS.

*** 13. I feel pleasant**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 14. I feel nervous and restless**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 15. I feel satisfied with myself**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 16. I wish I could be as happy as others seem to be**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 17. I feel like a failure**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 18. I feel rested**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 19. I am 'cool, calm and collected'**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 20. I feel that difficulties are piling up so that I cannot overcome them**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 21. I worry too much about something that really does not matter**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 22. I am happy**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 23. I have disturbing thoughts**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 24. I lack self-confidence**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 25. I feel secure**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 26. I make decisions easily**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 27. I feel inadequate**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 28. I am content**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 29. Some unimportant thought runs through my mind and bothers me**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 30. I take disappointments so keenly that I cannot put them out of my mind**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 31. I am a steady person**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

*** 32. I get in a state of tension or turmoil as I think over my recent concerns or interests**

| | | | |
|---|---|---|---|
| Almost Never | Sometimes | Often | Almost Always |
| <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |

For this part of the questionnaire there are groups of statements. Please read each group of statements carefully, then pick out the one statement in each group which best describes the way you have been feeling in the PAST MONTH. Tick the box beside the statement you picked. If several responses seem to apply to you, choose the one that applies to you TODAY.

There are no right or wrong answers. Do not spend too much time thinking about each item.
PLEASE RESPOND TO ALL ITEMS.

*** 33. BDI1**

- I do not feel sad
- I feel sad
- I am sad all the time and cannot snap out of it
- I am so sad or unhappy that I cannot stand it

*** 34. BDI2**

- I am not particularly discouraged about the future
- I feel discouraged about the future
- I feel I have nothing to look forward to
- I feel that the future is hopeless and that things cannot improve

*** 35. BDI3**

- I do not feel like a failure
- I feel I have failed more than the average person
- As I look back on my life I can see a lot of failures
- I feel I am a complete failure as a person

*** 36. BDI4**

- I get as much satisfaction out of things as I used to
- I do not enjoy things the way I used to
- I do not get real satisfaction out of anything anymore
- I am dissatisfied or bored with everything

*** 37. BDI5**

- I do not feel particularly guilty
- I feel guilty a good part of the time
- I feel quite guilty most of the time
- I feel guilty all of the time

***38. BDI6**

- I do not feel I am being punished
- I feel I may be punished
- I expect to be punished
- I feel I am being punished

***39. BDI7**

- I do not feel disappointed in myself
- I am disappointed in myself
- I am disgusted with myself
- I hate myself

***40. BDI8**

- I do not feel I am any worse than anybody else
- I am critical of myself for my weaknesses and mistakes
- I blame myself all the time for my faults
- I blame myself for everything bad that happens

***41. BDI9**

- I do not have any thoughts of killing myself
- I have thoughts of killing myself but I would not carry them out
- I would like to kill myself
- I would like to kill myself if I had a chance

***42. BDI10**

- I do not cry more than usual
- I cry more than I used to
- I cry all the time now
- I used to be able to cry, but now I cannot cry even if I want to

***43. BDI11**

- I am no more irritated now than I ever am
- I get annoyed or irritated more easily than I used to
- I feel irritated all the time now
- I do not get irritated at all by the things that used to irritate me

***44. BDI12**

- I have not lost interest in other people
- I am less interested in other people than I used to be
- I have lost most of my interest in other people
- I have lost all of my interest in other people

***45. BDI13**

- I make decisions about as well as I ever could
- I put off making decisions more than I used to
- I have greater difficulties in making decisions than before
- I cannot make decisions at all anymore

***46. BDI14**

- I do not feel I look any worse than I used to
- I am worried that I am looking old and unattractive
- I feel that there are permanent changes in my appearance that make me look unattractive
- I believe that I look ugly

***47. BDI15**

- I can work about as well as before
- It takes an extra effort to get started at doing something
- I have to push myself very hard to do anything
- I cannot do any work at all

***48. BDI16**

- I can sleep as well as usual
- I do not sleep as well as I used to
- I wake up 1-2 hours earlier than usual and find it hard to get back to sleep
- I wake up several hours earlier than I used to and cannot get back to sleep

***49. BDI17**

- I do not get more tired than usual
- I get tired more easily than I used to
- I get tired from doing almost anything
- I am too tired to do anything

*** 50. BDI18**

- My appetite is no worse than usual
- My appetite is not as good as it used to be
- My appetite is much worse now
- I have no appetite anymore

*** 51. BDI19**

- I have not lost much weight, if any, lately
- I have lost more than 5 pounds
- I have lost more than 10 pounds
- I have lost more than 15 pounds

I am purposely trying to lose weight by eating less, yes or no?

*** 52. BDI20**

- I am no more worried about my health than usual
- I am worried about physical problems such as aches and pains; or upset stomach; or constipation
- I am very worried about physical problems and it is hard to think of much else
- I am so worried about my physical problems that I cannot think about anything else

*** 53. BDI21**

- I have not noticed any recent change in my interest in sex
- I am less interested in sex than I used to be
- I am much less interested in sex now
- I have lost interest in sex completely

***54. The purpose of this checklist is for you to give us an idea about what your physical pain feels like. Each of the words in the left column describes a quality or characteristic that pain can have. So, for each pain quality in the left column, CHOOSE the response that tells how much of that specific quality your pain has. Rate every pain quality.**

| | None | Mild | Moderate | Severe |
|-------------------|--|--|--|--|
| Throbbing | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Shooting | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Stabbing | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Sharp | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Cramping | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Gnawing | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Hot-burning | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Aching | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Heavy | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Tender | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Splitting | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Tiring-exhausting | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Sickening | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Fearful | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| Punishing-cruel | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |

***55. Please choose the number that represents how bad your pain is right now from 0 (no pain) to 10 (the worst possible pain).**

***56. Please choose the statement that best describes your present pain.**

- 0 no pain
- 1 mild
- 2 discomfoting
- 3 distressing
- 4 horrible
- 5 excruciating

***57. Is your pain...**

- Brief
- Intermittent
- Continuous

For each question, tick the answer that best applies to you.

*** 58. In general, would you say your health is:**

- Excellent
- Very Good
- Good
- Fair
- Poor

*** 59. Compared to one year ago, how would you rate your health in general now?**

- Much better now than one year ago
- Somewhat better now than one year ago
- About the same
- Somewhat worse now than one year ago
- Much worse now than one year ago

*** 60. The following items are about activities you might do during a typical day. Does your health now limit you in these activities? If so, how much?**

| | Yes, limited a lot | Yes, limited a little | No, not limited at all |
|---|---|---|---|
| Vigorous activities, such as running, lifting heavy objects, participating in strenuous sports | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Lifting or carrying groceries | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Climbing several flights of stairs | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Climbing one flight of stairs | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Bending, kneeling, or stooping | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Walking more than a mile | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Walking several blocks | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Walking one block | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Bathing or dressing yourself | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |

***61. During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of your physical health?**

| | Yes | No |
|--|--|---|
| Cut down the amount of time you spent on work or other activities | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Accomplished less than you would like | <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> |
| Were limited in the kind of work or other activities | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Had difficulty performing the work or other activities (for example, it took extra effort) | <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> |

***62. During the past 4 weeks, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?**

| | Yes | No |
|---|--|---|
| Cut down the amount of time you spent on work or other activities | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Accomplished less than you would like | <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> |
| Didn't do work or other activities as carefully as usual | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |

***63. During the past 4 weeks, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?**

- Not at all
- Slightly
- Moderately
- Quite a bit
- Extremely

***64. How much bodily pain have you had during the past 4 weeks?**

- None
- Very mild
- Mild
- Moderate
- Severe
- Very severe

***65. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?**

- Not at all
- A little bit
- Moderately
- Quite a bit
- Extremely

***66. These questions are about how you feel and how things have been with you during the past 4 weeks. For each question, please give the one answer that comes closest to the way you have been feeling.**

How much of the time during the past 4 weeks . . .

| | All of the time | Most of the time | A good bit of the time | Some of the time | A little of the time | None of the time |
|---|--|---|---|---|---|---|
| Did you feel lively? | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Have you been a very nervous person? | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Have you felt so down in the dumps that nothing could cheer you up? | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Have you felt calm and peaceful? | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Did you have a lot of energy? | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Have you felt downhearted and blue? | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Did you feel worn out? | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Have you been a happy person? | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |
| Did you feel tired? | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> |

***67. During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting with friends, relatives, etc.)?**

- All of the time
- Most of the time
- Some of the time
- A little of the time
- None of the time

*** 68. How TRUE or FALSE is each of the following statements for you.**

| | Definitely | Mostly true | Don't know | Mostly false | Definitely false |
|--|--|--|--|--|--|
| I seem to get sick a little easier than other people | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| I am as healthy as anybody I know | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| I expect my health to get worse | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> | <input type="radio"/> <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |
| My health is excellent | <input type="radio"/> <input type="radio"/> <input checked="" type="radio"/> |

Opt-in for second part of the research

OPT-INTO PHASE II of the study

The first part of the study looked at how your mood affected your hospital admissions, pain and quality of life over the last year.

The second part of the study aims to explore how your experiences of your disease/illness have affected your mood over the last year by asking you questions that the first part of the study did not ask or allow you to answer.

If you feel that you have more to say about how your illness affects you and would be prepared to meet with the researcher for 50 minutes of your time for a confidential and anonymous interview in a safe and secure place, then please leave your email address and/or your contact phone number and I will get back to you when the first phase of the study is completed within the next 3-4 months.

If you are not interested in taking part in the personal interviews then please move on to the next page.

*Please note that you may withdraw from participating in phase II at any time without having to give any reasons.

69. If you would like to participate in phase II of the study then please complete the boxes below.

***you do not have to leave your contact phone number**

Forename

Email address

Contact phone number

***70. Where did you find out about this study?**

- A support group
- A charity
- GP or other health professional
- Psychologist
- An NHS service
- Health forum
- Social media
- Word of mouth
- Through the researcher

Debriefing Page

Thank you for participating in this research.

This study is exploring how mood affects pain management and quality of life in chronic disease e.g. Sickle Cell Disease, thalassaemia, blood cancers and also in Carers.

If you feel distressed after completing the questionnaire then there are some contact numbers and help lines that you can contact if you would like help with the distress.

Pain helpline: Pain concern: 03001230789, www.painconcern.org.uk

Sickle Cell Disease helpline: Sickle Cell society: 020 8961 7795, www.sicklecellsociety.org

Cancer Help UK helpline: 0808 800 4040, www.cancerhelp.org.uk

Carers helpline: Carers UK: 0808 808 7777, www.carersuk.org

List of psychologists in your area: <http://www.bps.org.uk/psychology-public/find-psychologist/find-psychologist>

Your data will be anonymous unless you wish to participate in the second phase of the study. The primary researcher will be the only person who has access to the data.

All the data will be kept electronically for 3 years after the conclusion of the study on a password protected external hard-drive.

You are not obliged to take part in this study. You are free to withdraw at any time. Should you choose to withdraw from the study you may do so without any consequences.

If you have any queries about the study, then please contact me, Dede-Kossi Osakonor at u1019289@uel.ac.uk.

If you have any questions or concerns about how the study has been conducted, please contact the study's supervisor: Dr Meredith Terlecki, School of Psychology, University of East London, Water Lane, London E15 4LZ, +44 (0)20 8223 4463, m.terlecki@uel.ac.uk

Or

Chair of the School of Psychology Research Ethics Sub-committee: Dr. Mark Finn, School of Psychology, University of East London, Water Lane, London E15 4LZ, 020 8223 4493, m.finn@uel.ac.uk

Thank you for your time and for participating in this study.

Appendix D

Interview Outline

Interview Outline

1. *General experience of pain*

- How would you describe your pain?
- What makes your pain worse?
- What helps to alleviate your pain?
- How does it affect your day to day life?
- What do you think it means when you experience general pain?

2. *General anxiety*

- How would you describe your anxiety?
- What causes you to feel more anxious?
- Is there anything that reduces your anxiety?
- How do you think your anxiety affects your pain?
- How do you think your anxiety affects your general health?

3. *General low mood*

- How would you describe your low mood?
- What causes you to feel low?
- What helps your mood to improve?
- How does your low mood affect your pain?
- How does your low mood affect your general health?

4. *General questions*

- When you have experienced general pain, have you become more conscious of what is happening in your body? If so, what percentage of time would you say you are pre-occupied by the pain?
 - How has your experience of pain, anxiety or low mood changed over time?
 - What does having this illness mean to you about your identity?
 - Is there anything else you would like to tell me about your illness?
-

Appendix E

MANOVA analyses with unequal group sizes

(SCD = 28; BC = 8; Carers = 15)

The MANOVA conducted to evaluate between-group differences in trait anxiety and depression scores across the unequal groups showed that Box's test of equality covariance matrices was significant at $p = .020$, indicating the covariance matrices were unequal, thus violating the conditions of the MANOVA. The result of the overall MANOVA was significant and has been included in Table 7. Bonferroni post hoc pairwise comparisons showed that the BC group reported significantly higher trait anxiety scores ($M = 56.63$, $SD = 11.58$) relative to the Carers ($M = 44.60$, $SD = 12.75$; $p = .077$). There was no significant difference in the trait anxiety scores between the SCD group ($M = 50.07$, $SD = 11.58$) and the BC group ($M = 56.63$, $SD = 11.58$; $p = .531$), or between the SCD group ($M = 50.07$, $SD = 11.58$) and the Carers ($M = 44.60$, $SD = 12.75$; $p = .475$). With regard to depression scores, the BC group also reported higher depression scores ($M = 33.63$, $SD = 17.44$) than both the SCD group ($M = 18.54$; $SD = 11.63$; $p = .011$) and the Carers group ($M = 11.80$, $SD = 10.21$; $p = .001$). There was no significant difference in depression scores between the SCD group ($M = 18.54$; $SD = 11.63$) and the Carers group ($M = 11.80$, $SD = 10.21$; $p = .279$). In summary these results from the unequal MANOVA, whilst violating the conditions of the MANOVA, were still consistent with MANOVA 1 reported in the quantitative results section.

The MANOVA conducted to evaluate between-group differences in sensory and affective pain scores across the unequal groups showed that Box's test of equality of covariance matrices was not significant at $p = .714$, indicating the covariance matrices were not unequal. The result of the overall MANOVA was significant and has been included in Table 7. Bonferroni post hoc pairwise comparisons showed that the SCD group reported significantly higher sensory pain scores ($M = 16.29$, $SD = 6.98$) relative to the Carers ($M = 5.27$, $SD = 7.81$; $p = .001$). There were no significant differences in sensory pain between the SCD group ($M = 16.29$, $SD = 6.98$) and the BC group ($M = 11.63$, $SD = 11.07$; $p = .449$), or between the BC group ($M = 11.63$, $SD = 11.07$) and the Carers ($M = 5.27$, $SD = 7.81$; $p = .221$). These findings were consistent with the findings from MANOVA 2. The difference between MANOVA 2 and this current MANOVA was that there was a significant difference reported in the affective pain scores. The Bonferroni post hoc pairwise comparisons showed that the SCD group reported significantly higher affective pain scores ($M = 5.18$, $SD = 3.27$) than the Carers ($M = 2.07$, $SD = 3.35$; $p = .001$).

= .028), but not significantly different from the BC group ($M = 3.75$, $SD = 4.98$; $p = .978$). In summary, the sensory pain results from this MANOVA were consistent with the results from MANOVA 2; however the affective pain results which were insignificant in MANOVA 2 were significant for the unequal groups MANOVA.

The MANOVA conducted to evaluate between-group differences in quality of life subscale scores (physical health limitations, emotional health limitations, fatigue, emotional wellbeing, social functioning and general health) across the unequal groups showed that Box's test of equality covariance matrices was significant at $p = .012$, indicating the covariance matrices were unequal, thus violating the conditions of the MANOVA. The result of the overall MANOVA was significant and has been included in Table 7. Bonferroni post hoc pairwise comparisons showed that there was no significant difference in fatigue between the SCD group ($M = 124.29$, $SD = 93.07$) and the BC group ($M = 65.00$, $SD = 70.71$; $p = .347$). The SCD group had more physical health limitations ($M = 103.57$, $SD = 147.78$) than the Carers ($M = 273.33$, $SD = 186.96$; $p = .007$). Both the SCD group ($M = 80.36$, $SD = 55.43$; $p = .005$) and the BC group ($M = 62.50$, $SD = 42.56$; $p = .008$) had lower social functioning scores than the Carers ($M = 138.33$, $SD = 58.18$). Both the SCD group ($M = 131.25$, $SD = 106.85$; $p = .001$) and the BC group ($M = 103.13$, $SD = 86.02$; $p = .001$) had lower general health scores than the Carers ($M = 305.00$, $SD = 132.02$). In summary, there were no group differences between the SCD group and the BC group, but there were group differences between both these groups and the Carers in physical health limitations, social functioning and general health scores in this MANOVA that were slightly different to the findings from MANOVA 3. In addition, there were no group differences in fatigue scores in this MANOVA compared to MANOVA 3.

Table 7

Multivariate Analysis of Variance results for unequal Illness Groups and DVs (mood, pain intensity, quality of life) (SCD, n = 28; BC, n = 8; Carers, n = 15)

| | Pillai's Trace | df1 | df2 | Multivariate F | d | p |
|-------------------------------------|----------------|-----|-----|----------------|------|--------|
| <i>Mood (STAI-T) & (BDI-II)</i> | .270 | 4 | 96 | 3.75 | .156 | .007** |
| | Wilks' Lambda | | | | | |
| <i>Pain Intensity (SF-MPQ)</i> | .698 | 4 | 96 | 4.64 | .178 | .002** |
| | Pillai's Trace | | | | | |
| <i>Quality of Life (SF-36)</i> | .500 | 12 | 88 | 2.45 | .333 | .009** |

Note. * $p < .05$, ** $p < .01$; STAI = State-Trait Anxiety Inventory (Spielberger et al., 1983); BDI-II = Beck Depression Inventory II (Beck et al., 1996); SF-MPQ = Short Form McGill Pain Questionnaire (Melzack, 1987); SF-36 = Short Form Health Survey 36 (Ware & Sherbourne, 1992).