Portfolio Volume 1: Major Research Project

The Experience of Body Image in People with Sickle Cell Disease

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Abstract

Sickle Cell Disease (SCD) is a significant genetic blood disorder, causing a vast number of possible health complications, with a potentially huge impact on adjustment and quality of life. As with many other chronic illnesses, there has been some interest in the self-concept of people with SCD, and within that on their body image. However, research into body image for those with SCD remains limited in scope, and has frequently imported pre-existing and potentially culturally inapt concepts and measures to do so.

This study, therefore, is an exploratory qualitative project, to look at how people with SCD relate to their bodies and conceive of their body image. Data was collected from online blogs written by people with SCD, with approximately 500 posts collected. Data was analysed in the Reflexive Thematic Analysis framework, using an inductive approach and social constructionist epistemology.

Two themes, with two sub-themes each, were generated by the researcher: 'The Battle with my body, and how to win it' including 'Body, blood, cells, genes as enemy' and 'Victory through loving one's enemy', and 'My pain-filled body is unique and central to my identity' including 'Identification with the body strengthened by SCD' and 'Pain maintains the awareness of SCD'. The findings suggest that there are a range of ways of relating to the body for people with SCD: The body is made a central part of one's identity through the experience of the illness and that one's relationship to it can be one of hostility and enmity, but this can develop into one of love and nurture. Clinical implications of these findings could be the development of interventions for those with SCD where this body relating impacts on their well-being, as well as increasing awareness for physical health practitioners and joining up with other love-as-liberation movements.

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Chapter 1: Introduction

1.1 Overview

This introduction comprises a summary of how this project came to be, my place within it, the epistemology that undergirded it, and a look at some of the research in body image and Sickle Cell Disease (SCD).

1.2 Personal and epistemological position

1.2.1 The Journey To This Project

The former Psycho-haematology Research Unit at the University of Hertfordshire was developing a project on body image in people with haemophilia, and it was a logical progression to consider a project that considered the same in people with SCD. SCD is also a genetic blood-based disorder, with a lifelong impact on both physical health and psychological well-being. People with SCD are also a population who are considered underserved by research.

I therefore developed a plan to complete interviews with people with SCD regarding their body image. When the pandemic occurred, I turned to online blogs, to continue pursuing the research question of how people with SCD relate to their body.

1.2.2 Locating Myself

In all projects, who the researcher is constitutes an important question. This was especially so in this project. People with SCD are usually of ethnic groups who are subject to oppression in the white-dominated hegemony of the world. I took the position, put forward by Bahr & Song (2015), that the reason SCD does not receive more research attention is due to structural violence. They contend that people with SCD experience both the direct violence

caused by racism and classism and the structural violence of reduced political prioritisation and restriction of societal opportunities. This applies to health disparities including disparities in research.

Structural violence does not occur separately from those whom the structure benefits. It is therefore important to locate myself, the researcher, in relation to those structures and the people with SCD whom this research concerns. A conceptual framework that I found helpful to do this is the Social GRRRAAACCEEESSS (Burnham (2018)), the ideas of Schüssler Fiorenza (1992) and those of Crenshaw (1989). They suggest that aspects of identity firstly become differently salient at different times, dependent on context, and secondly that aspects of identity have interacting effects. Hence, the visible parts of my identity – I am an educationally privileged, white, middle-class cisgender man – work together to position me as a person who is privileged by the structures of society, in many of these ways different from the participants in the research. Richards & Schwartz (2002) suggest all research involves a power relationship where the researcher has power over the participants. Although being a researcher is not an aspect of identity, it is the key role I take in this research and with the research participants. Therefore it too connects with the commingled parts of myself given power by society.

Some, like Allen & Baber (1992), argue that privileged people should not conduct research with those who are not privileged. Some, like Robley (1995), suggests that one should use one's privilege to ensure less privileged voices are heard. Intersectionality means that one is rarely in a wholly privileged or unprivileged relationship with another person. Harry (1996) suggests that researchers must maintain a clear awareness of the various aspects of their identity in order to keep balance and not re-enact oppression. This is where I try to

position myself – recognising all of my identity. One part of that is acknowledging the invisible parts of my identity – being bisexual, developmentally disabled, having lived experience of mental health, being a Christian and experiencing chronic pain, that put me in different places of power relative to the participants of this research. Another part is recognising the context – whilst focusing on SCD, the more significant lenses (Schüssler Fiorenza (1992)) are those in which I have more privilege, rather than less, and voicing them does not diminish that. The final part is my aspiration to the optimistic view that privilege can be used – to put research effort and energy into an area that impacts significantly on the less privileged. This requires staying on guard against the risk of being exploitative, through various processes, such as service user consultation, supervision and reflexivity.

1.2.3 Epistemology

The epistemological position that I took in this research is a social constructionist position. Social constructionist authors have pointed out the difficulty with defining social constructionism due to its own critiques of making fixed or rigid definitions. Burr (2003) describes the various definitions as having family resemblances – enough shared between them that they are recognisably similar. One highly influential family member was the set of principles described by Gergen (1985).

Gergen (1985) described the key features of social constructionism. I summarise my construal of them in the following five points:

First, following post-modernism and post-structuralism, there is no way of following signifiers to something that does not itself refer to other things. Since all meaning can shift based on context, knowledge cannot be said to have a direct or stable relationship with its

subject. Objectivity and its implications, such as positivistic approaches to science, are not possible and pursuing them is not helpful.

Second, knowledge, not being generated from direct observation, comes about from people interacting with each other. This happens in their place in history and under the influence of existing ideas and how those ideas are interacted about by those people.

Third, in positivism, the value and endurance of ideas is how accurately they describe reality. For Gergen, ideas prosper depending on social processes, most significantly the operation of power.

Fourth, knowledge is not just a realm of knowing, but shapes doing. Therefore our understanding has consequences for our actions. People's choices are determined and constrained by their understanding, which is shaped by power.

Fifth, and last, the knowledge developed in this way is not a different way of being correct or True, but a living, continuing process, which does not offer certainty, but nor does it offer endless relativity. If there are structures by which competing ideas of knowledge may be shaped, one has to choose which of those structures are adhered to. Gergen argues therefore for the morality of social constructionism, that it brings questions of justice into all creation of knowledge. It is a hermeneutic of justice, as I understand it, that I intend to take. This is similar to the position put forward by Van Dijk (1993) in his conception of critical discourse analysis – that wherever possible one should take the perspective and prioritise the insights those treated most unequally.

In short: Knowledge is socially constructed. The social construction of knowledge is not a neutral process. It is principally influenced by power. Due to my societal position in relation to my identities, I have power over my participants. This power will impact upon the knowledge creation process of this research, even without direct interaction with the

participants. I chose to do whatever I can to prioritise the views and positions of those who are oppressed.

1.3 Sickle Cell Disease

    1.3.1 Physical consequences

    Sickle Cell Disease (SCD) is the most common blood disorder with a genetic cause (Edwards et al (2005)). The name SCD describes several illnesses where the shared feature is abnormal haemoglobin. This leads to 'sickling' of red blood cells, which obstruct blood vessels and prevent oxygen reaching some body tissues. This tends to occur in various locations in the body, leading to a range of medical problems which can affect the lungs, the brain, growth, the hips and shoulders, the eyes and the skin. One of the primary features is severe and chronic pain, a direct result of the blocking of blood vessels. Over their life, a person with SCD is likely to experience a number of 'crises' – marked increases in pain that may relate to an increase in sickled cells. Triggers for these crises are not always predictable or avoidable, but can include dehydration, over-exertion, cold weather and psychological stress.

    1.3.2 Psychological factors of SCD

    According to Edwards et al (2005), SCD is linked with a number of psychological difficulties, which are theorised to stem from the experience of pain and also influence the level of pain. These include low mood, anxiety and anger. Psychological problems may be present since childhood, given the lifelong nature of the illness. These can include poor self-concept and body image. Compas et al (2012) state that all chronic illness causes and is affected by chronic stress from childhood onward, including SCD alongside their discussion of cancer, diabetes and asthma. Edwards et al (2005) suggest that disruption in self-concept

may emerge in adolescence, which in turn could impact self-worth and relating to other people. They describe various psychosocial factors in adulthood which predict elements of both physical and mental health. For example, negative cognitions predict poor health, and positive thinking about pain correlates with adjustment. Bruton et al (2015) reported higher anxiety, depression and alcohol difficulties in people with SCD compared to the general population. Given the link between psychological factors and physical health factors in a possible cyclical relationship, it is worth considering specific psychological factors that may underlie distress in people with SCD.

1.3.3 Contextual factors for people with SCD

Anie et al (2002) state that SCD principally affects people who originate in Africa, Asia or the Caribbean, and to a lesser extent people from South Europe/the Mediterranean and the Middle East. In their sample of patients in London, almost all were from African or Caribbean backgrounds (split about evenly between these two groups). This is important to note in the UK context, as these groups face societal discrimination. Racism is a factor in the experience of SCD, argued Atkin and Ahmad (2001). People with SCD face difficulties with professionals in health and education, intersectional sexism and ableism in the labour market, in different ways to other chronic illnesses. Literature on SCD does not seem to consider the link between this prejudice and ideas of the body, with a PubMed research of "body image" AND "sickle cell" and prejudice returning zero results (NCBI (2019)). As my research does not directly address race, my position is that it is important to consider as part of the research process – in how it impacts on my privileged position in relation to the research participants, for example, and be open to possible connections with body relating.

1.4 Body Image

Although this project is going to take a more exploratory position on body image, it is important to touch upon some of the existing concepts of body image.

1.4.1 Concepts

Grogan (2016), describing the history of body image academia, puts forward that it began focused on weight and shape, because of the clinical psychology of eating disorders, but has become increasingly multifaceted. She explains the focus on body image as principally negative has moved to one that also includes positive elements – concepts such as body appreciation and acceptance. As early as 1950, she reports, body image included the perception of one's body, intrapersonal attitudes, and social factors. She takes a view that body image is the person's perceptions, thoughts and feelings, in relation to their body – and that behaviours may demonstrate some of these. She suggests that there are a number of key approaches which are very briefly summarised here:

1.4.1.1 Sociocultural View

Van Den Berg et al (2002) broke down the influences on a person's body image into the sources of the media, family and peers. They suggest that the media's role is by providing a comparative ideal image for the body. Tiggemann (2012) state that these ideals and comparisons are internalised, though not equally in everyone. She suggests other psychological factors, like self-esteem, mediate the extent of internalisation. She also acknowledges that the focus has been on weight and shape, and the model is limited by its focus on the hegemonic culture. Taleporos & McCabe (2002) describe a similar process occurring for people with physical disabilities, wherein media impacts on body image by the portrayal of people without disabilities and 'physical fitness' creating an ideal that both opposes and erases those with disabilities.

1.4.1.2 Cognitive-Behavioural View

Cash (2012) summarises the cognitive-behavioural view. He suggests that influences from a person's history affect the way a person evaluates their body, and the investment they have in it. He suggests that the socio-cultural view forms part of this. When an activating event occurs, as with other cognitive-behavioural models, the impact it has is due to the cognitions the person then has, which are shaped by these self-evaluations. This has an interacting relationship with behaviour and emotion in relation to the body image. He identifies coping approaches that are specific to body image, but parallel other cognitive-behavioural ideas – such as avoidance and over-compensation.

Within a cognitive-behavioural view, it can be suggested that people can experience emotions in relation to their body, including more complex emotions. Troop (2016), for example, elaborates on the concepts of body pride and body shame for people in the general population, by showing how they can be experienced contemporaneously and in anticipation. These anticipated emotions were found to impact upon behaviour – that anticipating one will feel body shame may lead to avoiding behaviour even if the person is not at that time feeling body shame. Troop & Redshaw (2012) found similar patterns in people with an eating disorder – that body shame, both present and anticipated, had impacts on their emotions towards their body and their weight, and predicted symptoms of anorexia.

1.4.1.3 Feminist/Objectification View

Although the sociocultural view takes some notice of the way in which can marginalise certain groups in their images of their bodies, Objectification Theory, posited by Fredrickson & Roberts (1997), views the relationship between body image and society as more pervasive. Rather than a set of information that informs one's conceptions, societal views of what a body should be (especially a female body) are key in the social constructive process of body image. The core theme of this process, they explain, is that the person is seen

purely in relation to the body and its use by others. McKinley (2011) describes this being akin to the panopticon – a Foucauldian metaphor about how when believing oneself to be continually scrutinised, one starts to do so for oneself. She suggests that although self-objectification occurs, it results from objectification by those who have power.

Although originating as a feminist approach, one can analogise to Marxism in terms of objectification as worker, and anti-racism in terms of objectification as colonial subject or subjugated race. McKinley, Fredrickson and Roberts encouraged such investigations in their papers. Buchanan et al (2008) researched whether skin lightness/darkness was a way in which Black women experience the process of objectification. They found that self-objectification occurred for these Black women, again through self-monitoring.

1.4.1.4 Positive Body Image View

Tylka & Wood-Barcalow (2015) made a case for theoretical attention to positive body image, arguing that previous theoretical approaches focused overly on the negative to the detriment of research and interventions. They draw on positive psychology, the Health at Every Size movement, cognitive-behavioural concepts and positive connected embodiment (joy in one's body). The authors give a definition of positive body image focused on acceptance, comfort and positive interpretation, not just the opposite of negative body image.

Tylka (2012) suggests that this leads to minimising perceived imperfections, focusing on the parts of one's body one appreciates, listening to and taking care of the body. There is a focus on each body being unique. She also suggests that positive body image is maintained by filtering out or otherwise managing the influence of culture, and by selecting peers who reciprocally reinforce positive body image.

1.4.2 Body Compassion

One newer area in body image is body compassion. Building upon ideas of self-compassion, body compassion concerns relating to one's body kindly and mindfully, recognising difficulties and pains as part of the human experience (Murn (2014)). This includes being sensitive to distress and pain which might be felt in/by the body, and being motivated to respond to this (Beadle, Cain, Lennox, et al (n.d.)). In a range of groups, with and without health difficulties, people have described relating to their body as unique, self-owned, having commonalities with others, and productive, when having body compassion.

This concept is being applied to research where body image has been salient such as with cosmetic surgery (Khanjani et al (2020)) or disordered eating (Oliveira et al (2018)). In one study, those who identified as disabled had different mood related to those who did not identify as disabled, dependent on levels of body compassion – those who were lower in body compassion and were disabled had lower mood, and those who were higher in body compassion and were disabled had higher mood (Beadle, Cain, & Troop(n.d.)). Body compassion could have significant importance for one's coping or adjustment in a range of chronic illnesses or disabilities. Body shame and body compassion inform an idea of a relationship everyone has with their body going beyond satisfaction with body image to reflect emotions and attitudes in a richer, wider way.

1.4.3 In relation to race and ethnicity

In Grogan (2016), the section on race focuses on body satisfaction, reporting mixed results from quantitative studies in the UK and US regarding body satisfaction in Black, white and Asian groups. The sociocultural and objectification views on this area discuss the way society frames the Black body as sensuous in contrast to white purity, and a lack of

Black body representation in Eurocentric culture. Tylka (2012) said that for People of Colour there is a relationship between racial acceptance and body image – that positive body image is dependent on acceptance by one's own racial group and the societally dominant one. Some research has suggested that marginalisation can lead to a space where the impossible demands of said culture can be transcended. Parker et al (1995) found that Black teenagers in the USA saw body image as expression of one's personality, flexibly making it work for the individual rather than a shared ideal. McHugh et al (2014) found that when Native Canadian teenage girls participated in traditions of their cultures, they could feel body pride in their Native features, alongside health and body.

1.4.4 In relation to medical conditions other than Sickle Cell Disease

Body image and mental well-being have been linked in a number of medical conditions, especially the more visible. Taylor and Pooley (2017) argued that body image was a significant issue in the mental health of people who had had limb surgery due to sarcomas. Łakuta et al (2016) looked at the mediation relationship of body image and mental health in psoriasis. They found that experiences of stigmatisation led directly, and indirectly by increasing the salience of appearance in self-evaluation, to a negative emotional attitude to the body. This then fostered depressive symptoms. They also found that the earlier one developed psoriasis, the more negative beliefs about the body developed. This is relevant when comparing psoriasis with SCD, since SCD is usually first identified in the first year of life and is a lifelong genetic condition. It may be that people with SCD could experience especially negative beliefs in relation to the body. Łakuta et al (2016) recommend considering cognitive-behavioural approaches to body image for intervention. Holzer et al (2014) measured the impact of lower-limb amputation on body image, using a scale that measured appearance, fitness, health and illness. They found significant reductions compared to

controls on body image overall, on evaluation of their appearance, fitness and health, and reduced investment in being fit, maintaining health and reacting to illness. They identified dissatisfaction with the area of the body where the amputation had taken place. This change in behaviour, which could then impact one's health, could be relevant, though this is a different medical condition to SCD.

Olsson et al (2016) longitudinally studied people with blood cancer. During treatment, their participants showed some reduction in their positive body image which predicted changes in health-related quality of life. This was only the case for men. The quality of life measure showed changes in Physical, Social and Role functioning, not in Cognitive or Emotional functioning. They noted that changes in body image were quite limited in this quantitative study and cited Olsson et al (2013), a qualitative paper where people being treated for blood cancers described their body image affected, especially around ideas of strength. They noted that the measure of body image focused on external ideas of body image, not the internal sense of the body – such as a sense of wholeness. It would be important for this study to ensure that both internal and external aspects of body image are considered. The mental health impact of body image varied in different health conditions, so SCD may have its own relationship too.

1.4.5 In relation to Sickle Cell Disease

There are a number of physical difficulties in SCD that might lead to a negative body image. Edwards et al (2005) describe how pain in SCD is cued by a wide range of triggers, lasting for a varying amount of time, occurring in irregular episodes. Common other effects that may impact body image can come in adolescence – delayed growth and delayed sexual development. Roseff (2009) describes a range of SCD complications: the blockage of small

blood vessels – which leads to pain, swelling, bone damage, ulcers on the skin, priapism and stroke. Rees et al (2010) add gallstones, high blood pressure, kidney disease, greater risk of infection, damage to the retina, inflammation, and greater tendency towards blood clots as possible complications. Alao et al (2003) describe another possible symptom – spontaneous abortion. Whilst not a symptom, Samuels-Reid & Scott (1985) describe that starting menstruation may be a cause of SCD crises, which could also impact on body image. Because of these symptoms, people with SCD can have a shortened lifespan, due to the risk of cardiovascular problems and organ failure. One of the principal treatments for SCD is transfusion. Pain management often involves intravenous opiates. Both of these can lead to scarring.

Some studies have discussed the links between body image and mental health in SCD: Alao et al (2003) report that children and young people with SCD may have a greater incidence of depression than the general population, which they suggest relates to body image and pain. They reviewed research in adults that reported greater anxiety and depression, including possible differences in body image. These differences in mental health are suggested to be due to psychological and social factors, pain, trauma and side effects of opiate medication. Brown et al (1993a) reviewed studies on risk and resilience factors for young people with SCD. They identified that self-esteem was lower in children with SCD compared to controls, and they related this to body image, especially for young women in relation to sexuality. Researching these links directly, Cepeda et al (2000) assessed young people with SCD between the ages of 8-19 on their self-esteem and body image whilst also measuring height, weight and maturation of secondary sexual characteristics, comparing them to matched controls without chronic illness. They did not find significant differences in body image and self-esteem, which they said may be because physical differences are not outside the range

found in those without chronic illnesses – although children with SCD are significantly

smaller as a group, they are not outside the usual range of possible development. Cepeda et al

(1997) had previously found no significant difference in clinical diagnosis of mental health

difficulties between those with SCD and race-matched controls. They argued that other

studies showing links did not sufficiently account for the overlap between some of the SCD

symptoms and some of the symptoms used in screening for depression. Alao et al (2003)

argue, however, that screening tools may ask about a wider range of manifestations of low

mood compared to clinical interview. As can be seen, there are differing views in the

literature on both the presence of differences in body image between those with SCD and

those without, and the connection between this and mental health difficulties.

1.4.6 Bringing It Together

There are numerous threads that overlap and intersect providing a partial view of how

body image might operate in SCD. It seems reasonable to expect that there will be some

commonalities with the features of body image in all people, both positive and negative; with

others specific to being in a more marginalised racial groups; with others still to living with

chronic health difficulties. There is, as well, the unique viewpoint that each individual has.

However, to more firmly establish the rationale for the project and what has not yet

been explored in depth by researchers, it is necessarily to take a more systematic approach, as

detailed in the next part.

Chapter 2: Systematic Review

2.1 Scope of the Review

The introduction presented a narrative wherein body image, mental health and chronic conditions can be seen as interrelated. However, it seemed that there was a paucity of research assessing this in people with Sickle Cell Disease (SCD). It is appropriate to systematically review the literature to see if this is the case, and what has been shown by the research so far.

Preliminary searches showed that using the search 'what is the relationship between sickle cell and body image?' would return too few papers for a systematic review. Attempts to widen the search to look at the body in general would be over-inclusive of papers regarding physical health. Searches based on the question 'What is the psychological impact of sickle cell disease?' similarly generated an overly large, heterogeneous collection of results.

Looking into the literature for ideas that had a scope larger than body image but smaller than psychological impact, one area that was identified was self-concept. Gana (2012) explains how the idea of self-concept has both included one's body but also included other aspects, since its first inception by William James. Oyserman et al (2014) suggest that self-concept is the sum of one's identities and one's images of oneself, both in terms of psychological views and sensory experience. They define self-concept as being the ideas one has about who one is, was and will become. Giving examples, they suggest this can be studied at various sub-levels, whether the very specific, such as how athletic a person thinks they are, or the more general but still subsidiary to overall self-concept, such as self-esteem. Hattie (2014) notes the following synonyms for his key terms of self-concept and self-esteem – self-identity, self-image, self-perception, self-regard, self-worth and self-evaluation. This

could therefore be a good fit for a wider set of research than body image, but narrower than the total psychological impact. This does not suggest that there is a single unified idea of self-concept, but that it is a network of ideas that includes (without being limited to) the ideas already mentioned. Inevitably, it will be defined differently by different researchers, who will take their own positions on whether there is one such thing as self-concept. Nevertheless, it is reasonable to expect there will be a collection of research that is relevant to the topic of this study and could give context and understanding of the psychology of SCD.

Therefore the question to be addressed by the systematic review was decided to be 'what is the nature of self-concept in people with sickle-cell disease?'

2.2 Developing the Search Strategy

2.2.1 Sources

Vassar et al (2017) argue that for best sensitivity and completeness, a range of sources should be searched, including multiple databases. There were three available psychology-focused databases. These were PubMed, Scopus and PsycArticles. However, the PsycArticles database did not produce any results with the initial versions of the searches, and so was excluded at this stage. PubMed and SCOPUS were thus the databases used.

2.2.2 Search Terms

I made use of MeSH terms (NCBI (n.d.)) and the synonyms used by Hattie (2014) to ensure comprehensive search terms for self-concept and SCD. Therefore the complete search was: ("sickle cell d*" OR "sickle cell a*" OR "sickle cell c*" OR "Sickling Disorder Due to Hemoglobin S" OR "HbS Disease OR Hemoglobin S Disease") AND ("Body Dissatisfaction" OR "Body Image Dissatisfaction" OR "Negative Body Image" OR "Self Concept" OR "Self-Perception" OR "Body Representation" OR "Body

Schema" OR "self-esteem" OR "self-identity" OR "self-image" OR "self-concept" OR "self-regard" OR "self-worth" OR "self-evaluation") This is in the SCOPUS grammar, but the PubMed grammar was identical save for "sickle cell" being the first part prior to the AND operator, which functionally led to the same search.

2.3 Refinement

In reference to the review question and informed in part by the pilot searches, the following inclusion and exclusion criteria were created and then applied to the results of the search:

Inclusion Criteria	Exclusion Criteria
Focuses on SCD	Do not focus on SCD
Focuses on relationship between SCD and self-concept	Focuses only on physiological impact of SCD
Paper in the English language	Focuses on psychological aspects of SCD without considering self-concept, i.e. psychiatric conceptions only, experience of transition without reference to self-concept
Paper available via University of Hertfordshire or the BL	Relates to the experiences of family or professionals in relation to SCD, not the person themselves
Paper about children, young people or adults	Focuses on genetic research /animals
Primary research	Papers that include SCD in combination with other significant physical illnesses/disability
	Review papers

Initial Search N = 376	
Duplicates removed N = 164	
Titles screened N = 49	

Initial Search N = 376	
Abstracts screened N = 42	Papers without abstracts were excluded at this stage alongside those that did not fit inclusion criteria.
Full copies assessed N = 33	Papers whose full-text was unavailable in English through the access methods noted in the inclusion criteria were also excluded at this stage.

The papers that were successfully included after the stages of checking are listed in Appendix 1.

2.4 Review of Collated Papers

After summarising the types and quality of paper, the key areas that the papers addressed will be described.

2.4.1 Types of Paper

Of the 33 papers, 29 used exclusively quantitative methodology, 1 used mixed methods and 3 used exclusive qualitative methodology. Of those using quantitative methodology, approaches to analysis included correlation and multiple regression, inter-group comparison using parametric or non-parametric testing, and one study using structural equation modelling. Of those using qualitative methodology, one used grounded theory (constant comparative method), one used a form of thematic analysis, and the last used Bardin content analysis/thematic categorical analysis. The mixed methods paper used analysis of variance and multiple regression for its quantitative analysis and appeared to use a form of thematic analysis for its qualitative analysis. Most of the research was conducted in the USA and/or the UK – exceptions to this were Jamaica (2 papers), Brazil (1 paper), Iraq (1 paper) and Iran (1 paper).

2.4.2 Quality Assessment

The tool used for assessing the quality of the papers was the Mixed Methods Appraisal Tool (MMAT) (Hong et al, 2019). This is a tool that has been developed for systematic literature reviews where the literature contains papers that are quantitative, qualitative and mixed in their methodology. The advantage of using this approach is that similar frameworks developed by the same researchers can be applied to these differing sorts of research, rather than comparing two or more different tools that may not have a great deal of conceptual overlap. The MMAT suggests that scoring not be applied purely numerically. Therefore, Appendix 1 includes the responses to each of the questions, with an at-a-glance colour coding to give a quick impression of quality. Any form of assessment of this type relies on the assumptions of the tool and the subjective way in which it is applied by the reviewer. The questions for the quantitative papers especially seem to rely on the positivistic epistemology generally associated with such research. Nevertheless, this does not make such a tool of no use, but instead invites the caveat of not taking its outcomes as True, and that there may still be valuable meaning in research that is not considered high quality by these metrics.

Although advantages of using the MMAT have been described above, it could have been useful to use a tool specifically focused on qualitative research. One alternative could have been the CASP checklist for qualitative studies, CASP (2018), which includes more criteria, for example, an item on the relationship between the researchers and the participants which may have provided a deeper assessment of the quality of the reflexivity and power dynamics of the papers reviewed. Another alternative could have been the Big Tent Criteria for Qualitative Quality (summarised in Tracy & Hinrichs (2017))). Even more so than the CASP checklist, the Big Tent Criteria look in the widest sense of the value of the research,

and the many dimensions within qualitative research that are not always considered in quantitative research, or indeed in tools like the MMAT which, although inclusive of qualitative methods, seem to focus and originate in quantitative work.

2.4.3 Comparisons between those with SCD and those without

A total of thirteen papers involved research where comparison was made between people with SCD and at least one other group who did not have SCD, either controls without chronic illness or people with a different chronic illness. These papers made comparisons of notions such as mental health symptoms, self-esteem and self-efficacy. Each of these will be highly dependent on the researchers' conceiving of them as a construct, and the ways in which the researchers thought it appropriate to measure them. The participants' construing, both of the measures and of the constructs they imagine the measures to be measuring, will also impact on the research.

Two of the papers compared people with SCD to those with other chronic illnesses (Seigel et al (1990) and Thompson et al (1998)). Both of the papers suggested similar mental health difficulties between those with SCD and those with other chronic illnesses, such as asthma. Although Thompson et al (1998) measured self-esteem, they did not directly compare the people with SCD with those with cystic fibrosis. Seigel et al (1990) reported similar levels of self-esteem between those with SCD, those with diabetes and those with asthma. The remainder of the studies that made comparisons, compared people with SCD with people without chronic illness.

Six papers suggested that people with SCD had similar level of self-esteem to those without a chronic illness (Lee et al (1997), Gentry et al (1997), Cepeda et al (2000), Midence

et al (1996), Brown et al (1993b) and Forrester et al (2015)). Two papers reported that people with SCD had lower self-esteem than those without chronic illnesses (Kumar et al (1976) and Seigel et al (1990)). One further paper (Forrester et al (2015)) reported adolescents with SCD had a level of ambivalence about their self-concept, which may link to fear of death, decreased physical activity, concealing their illness and feeling over-protected by family. Each of these papers showing differences in self-esteem were of very good quality, whereas many of the papers that suggested there were no differences were of lower quality, lacking clarity on the representativeness of their sample and on managing potential confounding factors. Forrester et al (2015), one of the high quality papers, gave evidence suggesting a balanced position of both similarities and differences in self-esteem – suggesting a reasonable conclusion may be that there are differences, especially in certain areas, but that these should not be over-estimated.

One paper presented findings of no differences in self-efficacy between people with SCD and their unaffected siblings, and no difference from normative scores for the measure (Gold et al (2008)). This paper was of a lower quality, again being unclear on the representativeness of the sample and the management of confounds. It is the only paper in the review that compared self-efficacy between groups, suggesting tentative evidence that it is similar between those with SCD and those without.

Two papers reported differences in the specific area of self-concept in relation to physical function and ability (Lee et al (1997) and Panepinto et al (2005)). Two papers (Cepeda et al (2000) and Brown et al (1993b)) suggested that there were no differences, making this is a contested area. Comparison of the quality of the respective papers suggested that Cepeda et al (2000) is of a good quality, lacking only in making certain that its

comparison group did not have chronic illnesses; it was the highest quality of the concerned papers. Therefore, it may be more reasonable to conclude that the evidence does not support these differences.

Two papers addressed differences in self-perception of acceptance by peers. One described no differences in perception of social acceptance or competence (Lemanek et al (1994)). One paper reported differences in acceptance by peers – that those with SCD viewed themselves as being less accepted than those without SCD (Brown et al (1993b)). Brown et al (1993b) is a paper of a lower quality due to issues with representativeness and confounding variables, whereas Lemanek et al (1994) is a paper of very good quality, therefore again it is reasonable to conclude from the available evidence that there are not differences in perceived self-acceptance.

Four papers reported differences in the mental health of those with SCD (Seigel et al (1990), Bhatt-Poulose et al (2016), Kumar et al (1976) and Thompson et al (1998)). In Seigel et al (1990) and Bhatt-Poulose et al (2016), these were reports of higher ratings of depression than controls, in Kumar et al (1976) lower levels of anxiety than controls, and Thompson et al (1998) suggested that people with SCD had less oppositional/attentional problems compared to those with cystic fibrosis. A further paper reported no differences in anxiety or depression (Simon et al (2009)) All of these papers were of a higher quality. It is difficult to draw firm conclusions, especially given that the direction of difference varies in these papers where a difference is found. However, these differences are less important than findings described later regarding how mental health is impacted by self-concept.

Finally, two papers had findings on the topic of comparison of body image between those with SCD and those without. Bhatt-Poulose et al (2016) reported differences between those with SCD and those in the general population in relation to body image. They found that people with SCD had both poorer body image and less accurate weight estimation. Cepeda et al (2000) reported no differences in body image between those with SCD and those without, despite there being differences in physical development. This paper and Cepeda et al (2000) were rated as being of higher quality based on the MMAT. Given that the research was done with different populations – Cepeda et al (2000) with children in the USA and Bhatt-Poulose et al (2016) with teenagers in Jamaica, this could reflect developmental differences or cultural ones. Bhatt-Poulose et al (2016) explicitly argue that due to their larger sample size, they believe their results should be taken to supersede those of Cepeda et al (2000). Given the quality assessment of both papers and that they are studying differing populations, it may be reasonable to conclude that in some contexts there can be significant differences between those with SCD and those without on their body image, but that this is not a universal phenomenon. Cepeda et al (2000) explained their results through their finding that the children studied, whilst significantly physically less developed than those without SCD, were still within the usual range of development. It could therefore be suggested that as development progresses, the physical divergence and thus the body image comparison increases.

2.4.4 Associations between self-concept and other factors in those with SCD

A total of seventeen papers did research that did not compare between groups, but looked instead at the correlations between self-concept facets and other relevant factors. This was in samples wholly composed of people with SCD.

Four papers, all of a good quality, presented evidence showing that self-efficacy was associated with reduced physical symptoms (Clay & Telfair (2007), Goldstein-Leever et al (2020), Anie & Telfair (2005) and Edwards et al (2001)). Edwards et al (2001) created a measure of disease-specific self-efficacy that was used in a number of the studies in this part of the review – so the construing of self-efficacy and its measurement may be closely focused on managing SCD, which may make some sense of the findings. In these four papers, increases in self-efficacy correlated with decreases in physical symptoms of the illness, and in the majority of the papers, this was the assumed direction of causation. These papers may reflect a different direction of influence – that disease severity correlating negatively with self-efficacy may reflect that the more severe the illness is, the poorer the sense the person may have of their capacity to cope with it. Anie & Telfair (2005) suggested that this could be the case, though they acknowledged that direction of causality was difficult to ascertain, so this interpretation may be due to their choices regarding analysis. Given these findings, the possibility of bidirectionality between self-efficacy and disease severity could be explained by a cyclical process where physical symptoms impact self-efficacy which in turn determine the likelihood of physical symptoms in future. Because none of the papers in the review included an intervention to improve self-efficacy and at the same time measured physical symptoms, it is not possible to state with certainty whether one direction is better supported than the other, but only that they are clearly associated.

Two papers reported an association between self-efficacy and pain (Edwards et al (2001) and Adegbola (2015)) – again, this was a negative correlation, where greater self-efficacy and lower pain were related. Two papers investigated the link between self-efficacy and self-care (Matthie et al (2015) and Clay & Telfair (2007)). Both papers suggested that greater self-efficacy and greater activity of self-care were associated.

Three papers looked at the relationship between self-efficacy and healthcare use (Matthie et al (2015), Edwards et al (2001) and Cronin et al (2019)). Edwards et al (2001) described an association between higher self-efficacy and lower healthcare utilization, but Matthie et al (2015) suggested that neither self-efficacy nor self-care predicted healthcare use, but only being of a lower income. Cronin et al (2019) reported that self-efficacy predicted activation (a related concept that overlaps both with self-care and self-efficacy), and that activation predicted reduced healthcare use. This paper was of a slightly lower quality than the other two. Given the other two papers were assessed as being of a good quality, the differences in their findings may be due to different ways of measuring healthcare usage – Edwards et al (2001) measured number of times seeing a doctor in a year, and Matthie et al (2015) measured attendance at hospital. This would likely give different sensitivity to healthcare usage, but with the weight of evidence being somewhat more suggestive of a link between self-efficacy and reduced healthcare utilization. The findings of Matthie et al (2015) also highlight that such situations do not occur in a vacuum – that whilst self-efficacy is important, socioeconomic factors are also very important and may overshadow the different aspects.

Two papers reported trait factors that had associations with self-efficacy in those with SCD. Khudhair (2019) reported that self-efficacy was higher in men, younger people, those with less education, and those who were single, though it is worth noting that this paper was of an especially low quality and appearing to have aims of looking at associations but only reported descriptive statistics – which may be due to translation issues or the article not having been published correctly. Jenerette & Valrie (2010) reported that perceived care from

one's mother in childhood predicts self-efficacy. This suggests that there is a developmental process regarding the development of self-efficacy and why it might differ between people.

One paper described a way of changing self-efficacy: Dobson (2015) found self-efficacy improved by teaching guided imagery and is a rare piece of research that experimentally compared the impact of an intervention. It was assessed as being of a lower quality due to issues around control of the variables, but still suggests that self-efficacy is not static and there are interventions that might be useful for improving it.

Self-efficacy was reported to correlate with a variety of psychological factors. Adegbola (2015) described a negative relationship between sleep quality and self-efficacy. Two papers reported a relationship between self-efficacy and psychological symptoms of distress (Clay & Telfair (2007) and Edwards et al (2001)). Once again, this relationship was a negative one. Three papers suggested links between self-efficacy and quality of life (Goldstein-Leever et al (2020), Ahmadi et al (2018) and Jenerette & Murdaugh (2008)) – these were in the positive direction, with greater self-efficacy and greater quality of life being associated. One paper (Treadwell et al (2015)) found positive correlations between self-efficacy and stress – higher self-efficacy and higher stress being associated, and self-efficacy predicting readiness for transition, education, and independent living. The authors stated that the correlation with stress was because those who faced more stressors then had to develop coping in response to this. The two papers describing a negative relationship with psychological symptoms of distress are of higher quality than Treadwell et al (2015), which did not make clear whether the sample was representative or how they accounted for participants not responding. Therefore, the positive link between self-efficacy and stress put forward by Treadwell et al (2015) may be an anomalous finding.

The majority of the papers that did not make comparisons with groups without SCD were concerned with self-efficacy. However, there were also a small number of papers that instead investigated self-esteem. Four papers showed a relationship between self-esteem and psychological symptoms of distress (Thompson et al (1998), Burlew et al (2000) and Wilson et al (1997) and Simon et al (2009)) – lower self-esteem and greater mental health difficulties being correlated. It is worth noting that the constructs of self-esteem and of mental health and adjustment may share significant overlap, in which cause these correlations would be unsurprising.

In conclusion, self-efficacy was associated with reduced physical symptoms, pain, psychological symptoms of distress and increased self-care. It was influenced by distal factors such as maternal care, and proximal factors, such as training in coping strategies. Self-efficacy and another facet of self-concept, self-esteem, were associated with mental health, where the greater the self-efficacy or self-esteem, the better the adjustment. Nevertheless, for the most part, causal relationships, whether unidirectional or bidirectional, were unclear.

2.4.5 The role of the body within self-concept

Four papers specifically investigated the role of the body within self-concept (Bhatt-Poulose et al (2016), Reddy et al (2011), Cepeda et al (2000) and Tanabe et al (2010)). The disagreement between Bhatt-Poulose et al (2016) and Cepeda et al (2000) has already been discussed, with the conclusion that it may reflect developmental and cultural differences. Tanabe et al (2010), a paper of good quality, reported that self-awareness and body-awareness were important contributors to self-care and self-management, which may add detail to the links between different aspects of self-concept and self-care.

Two articles reported gender differences in relation to body dissatisfaction (Reddy et al (2011) and Bhatt-Poulose et al (2016)). Reddy et al (2011), perhaps due to a small sample size, did not compare the groups, but found that body dissatisfaction in men correlated with higher stress, whereas with women it correlated with higher stress and also feelings of ineffectiveness and interpersonal mistrust. However, Reddy et al (2011) is a paper of lower quality, as it inappropriately derived many of the conclusions from non-significant results as if they were significant. More useful findings therefore come from the higher quality paper of the pair: Bhatt-Poulose et al (2016) also reported that body dissatisfaction appeared to operate in different ways for different genders – young women wanted to be thinner, and young men wanted to be larger. This pattern regarding young women with SCD was similar to the pattern described for young women without SCD. However, the young men with SCD were reported to desire to be significantly bigger in comparison to men without SCD. A similar pattern was described in weight estimation – that young men with SCD perceived themselves to be underweight far more than young men without SCD. Even despite this, the overall SCD group was suggested to be more likely to over-estimate their weight. This is perhaps because of the SCD group having a significantly lower weight than the control group, but similar levels of believing oneself to be overweight in young women between those with SCD and those without. In the regression models, the authors report that weight over- and under-estimation were both significant predicted by having SCD, that negative body image was significantly predicted by having SCD and by having a distorted view of weight. They also reported that having depression was significantly predicted by having SCD and having a perceived body weight that was above or below what it should be, and that suicide attempts were significantly predicted by having SCD and by negative body image. Cepeda et al (2000)

did not report gender differences or lack thereof because of the matched pairs design used in that study.

This evidence supports that body image is a relevant part of self-concept in relation to SCD and may have a connection to some of the factors already discussed, such as psychological symptoms and self-esteem.

2.4.6 Relationship between SCD, self-concept and areas of identity

Two papers, both of a good quality, were concerned with how specified areas of identity influenced SCD in relation to self-concept (Royal et al (2011) and Costa et al (2018)). Royal et al (2011) was concerned with the role of race. They reported that the less positive regard they felt that others had for their race the more they felt that they were treated differently because of SCD. They also reported that the more that race is central to one's self-concept, and the more they viewed their own race as unique and separate, the more they thought that their experience with SCD was influenced by race. In the qualitative part of the research, they constructed themes from the data of lack of knowledge, being treated as disabled, being linked to drug addiction, and the double (intersecting and multiplicative) impact of Blackness and SCD.

Costa et al (2018) was concerned with gender, since their study was with men with priapism. They described that these men struggled with loneliness, self-image especially related to masculinity and embarrassment, and sexual difficulties. They reported that many of them were not well-informed about how to manage priapism and would use counter-productive management techniques. The researchers suggested that the self-esteem impact around fragility as opposed to masculinity was especially acute in the Brazilian context.

In conclusion, these studies demonstrate that aspects of identity, and their incorporation into self-concept, have an impact on how people believe their SCD is treated, as well how they respond to their SCD themselves.

2.4.7 The Qualitative Papers in Greater Depth

Given that this research project itself is qualitative in nature, it could also be valid to pay particular attention to the qualitative papers found in this systematic review. Four papers found in the systematic review utilised qualitative methods – three solely, and an additional one as part of a mixed methods approach. These papers were Forrester et al (2015), Tanabe et al (2010), Costa et al (2018) and Royal et al (2011).

Forrester et al (2015) completed interviews with Jamaican adolescents, conducting a qualitative analysis with a particular interest in the changes in their development due to sickle cell and how this in turn impacted their identity. As a group, the researchers worked together to develop themes. One theme was that of positive self-concept, as well as feeling that they were living a normal life, that they had ways of coping with SCD (especially spiritual means), and that they did what they needed in order to manage their health. By contrast, other themes included having a strong fear of death, decreased physical activity, avoiding sharing that they had SCD and having an overprotective family. The researchers therefore suggested that the self-concept of the participants had some ambivalence, even if it leaned to the positive.

Tanabe et al (2010) conducted a qualitative study in self-management techniques. They gathered participants for a workshop discussion, selecting those who were thought by

their peers to use a good range of self-management skills. They then used approaches

informed by content analysis, within a grounded theory framework, to construct themes. The

principal theme, which they based on frequency of mention, was self-awareness. This

included knowledge about behaviours that individually led one into difficulties with one's

health, as well as the development of body awareness – being attuned to one's body and

following its lead.

Royal et al (2011) investigated the place of race in one's identity for people with

SCD. They used measures of racial identity that looked at its centrality in people's lives, their

ideological orientation towards it and their pride in their own racial identity. They also asked

about the person's experience of the intersection of race and sickle cell, and gave space for

open responses. The participants were principally African-American or Latinx-American. In

general, participants had a positive view of their own race, viewed their race as similar to

other races in the US as well as the world in general. Those participants whose parents were

born outside the US had a greater sense of their race being an oppressed minority.

Participants endorsed the view that they were treated differently because of SCD, and that

their race influenced this – the more centrally they held their racial identity, and the more

they expressed the uniqueness and separateness of their racial identity, the more they

endorsed this. In the qualitative findings, themes constructed included that people lack

knowledge about SCD, that people with SCD are treated as being disabled, that SCD is

linked to race and drug addiction (especially in how people are viewed by others), and that

being Black worsens the difficulty of having SCD. Some participants contended that race was

not a factor in their experience of SCD.

Costa et al (2018) used qualitative methods to explore how men with SCD who suffered from priapism conducted self-care. They completed this in an area of Brazil with a significant number of people of African descent, and hence also a significant incidence of SCD. Their qualitative analysis was one informed by a pre-existing model of self-care, which emphasised that self-care incorporates universal demands, developmental demands and health deviations. Universal demands are the basic needs to be met by any person regardless of health status, developmental demands are those required for self-care of a condition, and health deviations are the ways in which a condition interferes with self-care. As already described, they found that the men struggled with loneliness, masculine self-image, shame and sexual dysfunction. Their self-care was impacted by a lack of knowledge of the best approaches, as well as shame and stigmatisation. The main health deviation found was that of significant pain. As already referred to, the specific culture and context of Brazilian men was described as important by the researchers to understanding this research.

Considering the specifically qualitative research landscape, there are certain strengths but also some clear gaps. The impact on self-care and self-management of the illness, in common with many of the quantitative studies, was a focus of these studies. Thus where Tanabe et al (2010) described the importance of listening to one's body, the interest seemed more in this as a process and what it led to, as opposed to how it was experienced, and indeed how different views of the body might impact on what it means to listen to said body. Although ideas of identity and self-esteem have been investigated in these papers, the body-specific nature of this identity has not been directly studied. Costa et al (2018) arguably came closest to this, but their study focused on one particular element of the body in relation to one particular, significant, symptom of SCD. There is room, therefore, to explore beyond priapism and beyond the role of the body in masculinity. Intersectionality was an important

feature of Royal et al (2011). This is an element that will be important to take into any further qualitative work.

Whilst overall self-image and self-concept has seen some research, and some elements of relating to the body have also, it can be seen that an area that remains unstudied by qualitative approaches is the relationship with the body in the more conceptual or experiential sense. As well as being a valuable investigation into an area where little has been studied, this sort of research could be a supplement to this existing research, informing, for example, about some of reasons for the differences in self-care, or whether it underlies differences in self-concept or if other features predominate.

2.5 Implications for This Project

2.5.1 Summary

The collated literature showed that the area of self-concept has been studied in various manifestations in people with SCD. Self-efficacy was a particularly emphasised area, which is not surprising considering the findings that it could significantly impact upon physical and psychological symptoms, in part by facilitating self-care, and also being impacted by them. This apparent two-way relationship makes conceptual sense, and may also apply to other areas of self-concept. It appeared that self-efficacy was improved both by early experiences of care and being equipped with specific techniques. Self-efficacy was also an important determinant of quality of life and one's independence and readiness for adult life. Much of the research into self-esteem did not find group differences in self-esteem for those with SCD, but did find it (as with those without SCD) to be a significant factor in one's psychological well-being. Much of the research was conducted with children and adolescents, and researchers often referred to the participants' ages to account for not finding differences

in self-esteem or self-concept more widely. However, research such as Costa et al (2018) that looked closely and related their research to complications common to SCD (and completed the research with adults) did find that self-esteem could be particularly impacted by SCD, at least in certain cultural contexts. The findings of Royal et al (2011) are therefore important: That where race was especially salient to a person's identity, though they viewed their own race positively, they believed that their SCD was treated differently due to their race.

The facets most relevant for this study, body image and body awareness, had only a small number of studies. Earlier work suggested that body image may not be significantly impacted by SCD, though it may have a link (at least in women) with stress and other negative self-views. More recent work, conducted with a larger sample size but also in a different national context (Jamaica rather than the USA) did find significant differences in body image due to SCD, and links between this and difficulties in mental health. For adults experienced in self-care, one part that they had developed was body awareness. With apparent differences in research with children and adolescents compared with adults, it seems that there may be a developmental process – firstly of the negative aspects of SCD and the body becoming salient, and then a positive and self-caring response to it developing in response.

2.5.2 Rationale

Although the wider area of self-concept in SCD has been researched in a number of studies, many of these have focused in on the understandably valuable topic of disease-specific self-efficacy. Research that has specifically looked at the body and how people relate to it has been far less apparent in the extant literature, and much of it has utilised both pre-existing concepts and pre-existing measures, often focusing on eating disorders. It is therefore

possible that whole areas of body image and how people relate to their bodies are not being understood. This is highlighted by the richness that was achieved for example by Costa et al (2018) where cultural ideas were interlinked with the health experiences of just one of the myriad complications possible with SCD.

A qualitative investigation of this area would therefore be valuable in understanding the specific nature of body image as it relates to those with SCD. Using an approach that is exploratory could endeavour to identify and develop concepts that are more specific to SCD. For this reason, the research questions below are chosen to be open, wide and to engender a spirit of exploration.

2.5.3 Research Questions

How do people with Sickle Cell relate to their bodies?

What is the nature of their body image?

Chapter 3: Method

3.1 Design

The design of the study was a qualitative design. Data was collected from online blogs written by people with Sickle Cell Disease (SCD), accessible online in 2020, written between at least 2010 and 2020. The data was analysed using the Reflexive Thematic Analysis approach, with an inductive, constructionist positioning. The choices of method of this project had to adapt to some of the practical difficulties faced by the emergence of the COVID-19 crisis. The key change was in the data collection, from being from interviews with people with SCD in London, to being blog posts from around the world. The next sections will lay out in more detail these various aspects of the research design and the rationale behind them. First, the choice of methodology and analytic method will be explored – the process of how it is put in practice is described later in the section:

3.2 Methodology Selection

The research question was exploratory in nature. The rationale for the research was that there is very little theorising specific to SCD on the topic. That which has been done has not looked at the understandings that come from those people with SCD. It was therefore preferable to use qualitative methods rather than quantitative methods. Qualitative methods are well-suited for expanding and developing the understanding of a topic (Queirós et al (2017)), especially in relation to the richer complexity of human experience and all that cannot be reasonably reduced to quantitative data. Qualitative research contains the powerful possibility of generative moments, where the life of participants, researchers and ideas meet and spark further life (Carlsen & Dutton (2011)). This life of the ideas generated in

qualitative research can pass from the hands of the researcher and go in unexpected and vital directions.

The choice of an analytic method was one informed by the epistemology, methodology, and pragmatics of the research (Harper (2011)). It was also important that one relate all of those things back to the purpose of the project.

Thematic Analysis was a good analytic method for exploring or characterizing the range of ideas that may be held on the topic (Harper (2011)). Thematic Analysis has been suggested to be an analytic method that fits with the critical realist epistemology (Harper (2011)). However, the major proponents and codifiers of Thematic Analysis, Braun & Clarke (2006) contend that an advantage of Thematic Analysis is that it is very flexible, both in terms of how it can fit with different epistemological positions found in qualitative research, and in the way one approaches the data. Since that seminal work, Braun and Clarke have developed their ideas on Thematic Analysis into a specific approach that they title Reflexive Thematic Analysis (e.g. Braun & Clarke (2019)).

Reflexive Thematic Analysis (RTA) was considered a suitable approach for analyses engaged with exploring what ideas might be present or in operation about a topic (within a particular group or community). In this evolved version, there is flexibility enough to include my epistemology – it thus seemed a good fit for this research. Braun & Clarke (2019) suggest that thanks to their origins in a deeply qualitative environment and their commitment to avoiding assumptions, reflexivity and 'knowingness' (their conception of not assuming) are necessary for using RTA. They suggest that other types of Thematic Analysis tend to have more realist bases, and become concerned with features such as codebooks and inter-rater

reliability, which are analytic approaches informed by a different epistemological and methodological foundation. Given the epistemological commitments that I made, this was a possible choice that I believe can fit well for this project.

This does not entail that RTA was the only possible analytic approach. However, it was not that there is a single correct answer (Braun & Clarke (2020a)). Instead, it was a matter of coherence – that something selected is the best fit, or is chosen as the best fit – not that there is a metric to always have a 'correct' approach.

This project was concerned with how a particular group of people experience a psychological phenomenon, that of relating to their bodies. Interpretive Phenomenological Analysis (IPA) is the main structured method that is used for phenomenological research. In IPA one tends to work with each data sample, usually an interview, to generate codes and themes based on one individual, before then looking at what themes one can craft across the whole dataset; RTA may be a better choice for larger and more heterogeneous datasets that consist of data other than interviews or other specifically in-depth accounts (Braun & Clarke (2020a)). This project was using this sort of data, and was open to the purely phenomenological but also the cultural and social, which may be easier to encompass using the RTA approach (Braun & Clarke (2020a)).

An approach that is more frequently used with wider ranges of data, and given that this project was interested in an area where there has been limited development of theory, is Grounded Theory (GT). Pragmatic considerations were one reason why I believed RTA is preferable to GT for this work. Though some GT proponents contend that pragmatic concerns are myths (Timonen et al (2018)) where a full theory development is undertaken, with

appropriate integrity to the method, this can rely on a great deal of analysis and a large

amount of collected data. This project was not aiming at full theory development because it

was more exploratory – there was not yet enough understanding of body image in SCD to

make the leap to theory development. Where these proponents suggest alternatives, especially

for doctoral projects such as this one, what they describe is similar to Thematic Analysis.

Given this, I believed it was better to do a robust Thematic Analysis, than a take on Grounded

Theory described as non-ideal (Timonen et al (2018)). Again, the flexibility of the approach

of RTA supported its use over GT – the data in this project was not just likely to be

heterogeneous, but to vary in its relevance to the research question. Where GT emphasises

intensive coding, and even literal line-by-line coding, this may have reduced the impact of

what is feasible to analyse in the data (Braun & Clarke (2020a)).

My epistemological position was influenced by critical approaches, and was one of

social constructionism. Closely associated with such positioning is Discourse Analysis. One

notable influence on this research was Davenport (2017) – who studied online blogs on fat

acceptance using Discourse Analysis. However, the context of this research and my

relationship to those being researched being different, I believed that my understanding of the

discourses may not be as strong as needed. Whereas in other analytic approaches, especially

RTA, one can approach analysis in a more inductive fashion, Discourse Analysis is most

coherent and useful where there is an existing awareness of the discourses external to the data

(Parker (2002)). Given my position as an outsider to the group of people being studied in this

research, I may have been able to draw upon hegemonic discourses, but would have been less

equipped to identify discourses relevant to people with SCD, people with chronic physical

illnesses, or Black people. The more bottom-up approach that RTA can employ, therefore,

was a better tool for analysis given the current stage of the research in this area and my

identities as a researcher.

3.3 Ethics

Ethical considerations are connected with every part of research, and they cannot be

confined to a single section. However, it is important to acknowledge both the procedural

elements of these ethical considerations (e.g. ethical approval from a responsible body) and

the elements regarding principles (e.g. maintaining confidentiality in internet-based research,

involving service user consultation). Twining et al (2017) give some initial suggestions on the

particular areas of interest in considering ethics for qualitative research as part of their guide

on designing a qualitative study. They suggest that vital issues include confidentiality, data

protection, cultural sensitivity and transparency.

3.3.1 Ethical Approval

Ethical approval was sought from the University of Hertfordshire Ethics Committee

For Studies Involving The Use Of Human Participants. This was granted on Thursday 6th

February 2020, and approved for amendments on 3rd March 2020 and 8th July 2020. The

relevant protocol numbers are aLMS/PGR/UH/04079, aLMS/PGR/UH/04079(1) and

aLMS/PGR/UH/04079(2). The full notifications can be found in Appendix 3.

3.3.2 Consultation

I worked with a service user consultant in the early stages of the project. We discussed

the concept of the project and what factors to consider which I would be unlikely to be aware

of. We also discussed the draft interview schedule and the way the project should be

presented to possible participants. Issues raised included the roles of skinniness/appearing well-fed, the way in which one's body can be seen as more part/property of the community than the individual and the cross-over of body image with choices about dress, for temperature regulation and to control/conceal one's appearance.

When the service-user consultant and I were no longer able to work together due to the pandemic, I agreed with my supervisory team that it was essential that we incorporate the views of someone with expertise in SCD, even if that person could not be someone with lived experience. I researched academics and contacted them. Through this Antoinette Bardon-Gooden became part of the project, and provided very helpful insight on issues around race and nationality, the potential impact on healthcare professionals, and the evolution of talking about SCD over time and the role of technology in that.

3.3.3 Consent and Confidentiality

Consent and confidentiality in internet research are different to but not disconnected from these concepts in other forms of research. Some argue that the creators of online documents are not active participants, and the documents being used in the research are publically available and therefore informed consent is not required (Wilkinson & Thelwall (2011)). Others take a view that people using the internet do not necessarily see it as a totally public space, but perhaps as a semi-public space akin to a coffee shop (Robinson & Schulz (2010)). Different forms of web media have expectations of privacy. Closed social media accounts, open social media accounts, and blogs do not have the same expectation of being accessed. Blogs have been treated as public in various forms, for example, being compared to journalism (Lasica (2003), Nelson & Fernheimer (2003)). On this basis, I did not opt to seek consent from the bloggers from which data was collected. This is partly given the next point

regarding anonymity, and the selection criteria focusing on the publically accessible, many from well-known public blog sites or SCD organisations.

Anonymity is vital in the ethics of online research. Many participants will be using usernames and more obviously artificial identities, with no clear link to their offline identities. Contrariwise, it can be possible to find information about a person just with some quoted phrases (Robinson & Schulz (2010)) – this applied in trial searches that I completed, both of quotations in published literature, and searching from the data in this study to see if I could find the original blogs. Paraphrasing and elision can be used to maintain the anonymity of the participants. This is a principle that I will follow in my research, with the key aim of preventing a link between a person's words and ideas about SCD and their identity. Beyond the general ethical imperative noted from the publications about ethics in research and in internet research, there is also a need for specific awareness of the sensitivity of being known to have SCD. Alongside healthcare provider stigma against people with the disease, there is also broader stigmatization in the general public (Bulgin et al (2018)). Therefore there is additional reason to ensure that anonymity is preserved. In order to illustrate the themes, I will use paraphrased quotations (similar to, e.g. Ball & Nicolle (2015)). I followed the British Psychological Society (2017) guidelines on internet research. Whilst recognising that there is a risk of losing scientific quality of the research through paraphrasing, this has to be balanced with the risk of harm to those whose data has been used. When in doubt, in such ethical questions, and given the afore-mentioned test searches, I have chosen to ensure greater anonymization. Given also the candid expressions about themselves, their families and their doctors in the data, it is my view that it is proportional to paraphrase the quotations in this research. In order to do this, I used my own judgement and the WordTune AI paraphrasing

tool, to try to preserve as much of the original meaning as possible and not introduce too much interpretation (though recognising that that would always occur to an extent).

3.3.4 Data Protection

Due to the data collection method, sensitive personal information was not collected. Nevertheless, the 'raw' data – i.e. website captures, was stored on password protected computers and a password-protected cloud service.

3.3.5 Cultural Sensitivity

The process of remaining culturally sensitive is, as with being ethical in research as a whole, not something that can be confined to one section of a manuscript, nor is it a part that is 'done' or 'finished'. I engaged in the consultation described above to improve the cultural sensitivity of this research, which may have been impaired without the voices consulted with. I described in the Introduction section my position in relation to the research and the people most likely to be the participants of the research. It has been my endeavour to ensure that each part of the ethics and practice of the project features cultural sensitivity. This sensitivity has needed to be on the various axes wherein it is necessary in the relationship between myself as the researcher and the people who are the research participants, especially race and chronic illness.

3.3.6 Transparency

Following Ponteretto (2010) I aimed to be transparent about the following features: That in constructivist research, the journey of knowledge production has an uncertain destination that comes about through co-creation, and therefore one cannot be certain of where it will go and therefore forewarn participants with any certainty of potentially

distressing areas. That one necessarily requests participation on the basis of benefit to marginalised communities, but one cannot make a promise that this will happen – my intention was to be clear about the hoped-for clinical utility without suggesting that the path to it is a straight line. However, whilst these were commitments I continued to hold, the forum to be transparent about them changed from being face-to-face with participants to places such as this thesis, the research article, and in dissemination.

Ponteretto (2010) also notes that study design only has a certain amount of utility in ensuring ethical qualitative research. Instead, they contend, one must commit to collaboration, grow in multi-cultural competence and develop self-awareness.

3.4 Assuring Quality

Braun & Clarke (2020b) proposed a quality framework for research using Reflexive Thematic Analysis, in response both to existing frameworks for quality in qualitative research, plus common mistakes and confusions that they saw in those purporting to use RTA. This quality framework was completed for this study and is included as Appendix 7.

3.5 Data Collection

3.5.1 Determining Amount to Collect

Many papers that use blogs as data sources do not specify their inclusion and search processes. Within those that do, the information provided may be difficult to use as a guide for data collection due to differences in methodology or analytic method. For example, Gargiulo & Margherita (2019) collected over 600,000 words from 70 blogs selected from 150. Their inclusion approach was straightforward – accessible blogs that were active, Italian and discussing self-harm. However, as their analytic approach was a psychodynamic content

analysis, the amount of data collected may be much more than would be constructive to collect for a more qualitative approach. Caxaj & Berman (2010), who used a discourse analysis approach, chose 25 blogs to analyse, alongside pre-existing transcripts. However, they did not describe their approaches to search or inclusion strategies. Braun & Clarke (2019b) state that for RTA, a predetermined 'amount' of data is not an apt way of considering if the data is 'sufficient', but that this has to be reflexively informed – one cannot say when there will be 'enough' data for coding, 'enough' codes for themes, or 'enough' themes for the research on the basis of amount. Instead, this is better determined by quality – which they say can be seen as the original intention of the concept of saturation in Grounded Theory – than as a parallel of sample size and power in quantitative research. This is especially important, they argue, when one is using RTA and when one is not using positivistic epistemology: If the themes are pre-existing within the data, then the amount of data needed to find them and consider that search exhaustive may be calculable. If the themes are constructed in a reflexive interaction with the data by the researcher, then such an idea is not tenable. Of course, this does not mean that there is no need to collect an amount of data that may be seen as a 'good amount', but that this should be driven by the desire to have enough data to engage with in depth, develop codes and themes that are meaningful and make the endeavour worthwhile. They suggest that if analysis is really taking the interpretive, generative big-Q Qualitative approach (as they discuss in Braun & Clarke (2019a)), then it is never truly finished, because there is no limit to the possible ideas that can be created and considered from the data. The endpoint, then, is when one judges, as best and as reflexively as one can, that one has sufficient data to answer the research question.

3.5.2 Inclusion Frameworks

To determine which blogs were to be included in the data collection, I created an inclusion framework, based on a similar approach used by Davenport (2017) in her research into fat acceptance blogs. However, Davenport's focus was on multi-user blogs, and so the inclusion framework for this project focused more on ensuring the blogs had the requisite content for the analysis to address the research question:

Initial Blog Inclusion Framework
The blog must have an identifiable author or authors
The author must be an adult (if not explicitly stated, then on best likelihood)
The author must have lived experience of having Sickle Cell Disease
The blog must concern the topic of SCD
The blog must be publically accessible

Subsequent Blog Post Inclusion Framework
The blog post describes, in some way, living with sickle cell
The blog post is not (solely) a clinical or research update
The blog post is not (solely) a campaigning post
The blog post is not (solely) an effort at organising the SCD community towards a particular goal.

The search strategy was to utilise both a straightforward Google search and searched on popular blog hosting sites, such as Medium, Wordpress and Blogspot. "Living with sickle cell" was the key search term – preliminary investigation suggested this provided a good number of blogs that focused on the experience of the people with SCD, satisfying the inclusion framework for blog posts.

If a blog appeared to meet the initial inclusion framework, I would then look through each of the blog posts and see if they met the subsequent inclusion framework. I used the software tool NCapture for Chrome to save these posts in PDF form, preserving them as they appeared at the time of access, which could then be imported in NVivo (Version 11) for analysis. Those posts from a blog that did not meet the subsequent inclusion framework were

not downloaded, unless it was ambiguous or unclear. I continued with the search strategy and checking against the inclusion frameworks until a large amount of blog posts had been downloaded. As addressed in Section 3.5.1, it was not by a single, calculable measure that the decision to stop downloading further blog posts was made, but through the combination of – having a large number of types of blogs and different authors, having a significant number of posts, and those posts constituting a large number of words (approximately 500,000 – though many words that are counted in a whole blogpost are not really the content of the blog per se).

3.5.3 Features of the Blogs

Approximately 500 documents were collected, from 20 different blogs, some of which were a single writer writing over an extended time period, and others which featured a range of writers writing one blogpost each. The authors, where stated, were from a range of countries in Africa, the Caribbean and the North Atlantic. The table below gives some details of the blogs. What can be seen from what information was available is a mix of multi- and single author blogs, with a range of available and suitable (per the subsequent inclusion framework) blog posts. There were authors from a range of nations, though a significant proportion were from the USA or the UK – which may suggest some of the experiences, when joined in themes, might lean towards those shaped by so-called developed (or colonial metropole) nations. Many of the authors alluded to religion, either explicitly Islam or Christianity, or general invocations of God, heaven etc. There was a mix of gender of authors also, without being overly weighted to male or to female (genders beyond male and female were not specifically identified by the authors).

Blog Type (and number of authors)	Entries Downloaded	Nationalities (where identified)	Genders (where identified)	Religious backgrounds (where referenced)
Multi-author, 7	63	British, Dominican,	Female,	Muslim, Monotheistic,
Single	37	USA, St. Lucia	Female	Christian
Multi-author, 2	64	British,	Female	Monotheistic, Christian
Single	1	British, Gabonese	Female	
Multi-author, 11	32	Zambian, Cameroonian, Kenyan, Tanzanian, Nigerian, British, Ugandan,	Female, male	Christian, Muslim
Single	1	USA	Female	
Single	174	USA	Female	Christian
Single	33	USA	Male	Christian
Single	5	USA	Female	
Single	3	USA	Female	Monotheistic
Multi-author, 11	1	USA	Male, female	Monotheistic
Multi-author, 7	12	USA, British	Female, male	Monotheistic
Single	34	USA,		Monotheistic
Single	5	Nigerian	Female	Monotheistic
Single	5	Haitian, USA	Male	Non-religious
Single	3	British	Female	Christian
Single	25	USA	Male	Monotheistic
Single	6		Male	Muslim
Single	1	Kenya	Female	Monotheistic
Multiple, 2	2	Nigerian	Male, female	

Blogs do not always provide significant identifying information, and indeed, what information is provided is determined by the author and does not necessarily refer to how they might identify themselves in other contexts. Similarly, the identifications above were not necessarily exclusive of each other, but reflect what identifications or references were made in the blog posts. Gaps in the table show where no clear references were present, and for some of the multi-author blogs, although a number of identifications are made, this does not

mean that all of the authors made such things clear. Nevertheless, from what information was available, and assuming the predominance of earnest and honest self-description, this suggested that the authors whose blog posts comprised the data set were diverse in their national background, shared a tendency towards Abrahamic religion, and engaged in blogging in different ways and for different purposes.

3.6 Analysis

3.6.1 Analysis Selection and Structure

The analytic method selected was reflexive thematic analysis. Thematic analysis, which over time became Reflexive Thematic Analysis, is a flexible and accessible approach to qualitative analysis (Braun & Clarke (2006)). The aim of using this type of approach was that it allows one to develop, explore and describe the themes within the data; arranging the data to be more comprehensible, and beginning the process of interpretation. Thematic analysis can focus on a more inductive approach to the data, but can also bring theoretically-driven questions to the process of codifying themes.

This project aimed to use an inductive approach to the analysis. The reasoning for this was to put greater attention on the way in which the participants make sense of their experience. However, it would be incorrect to suggest, then, that the analysis did not contain anything from the researcher, or that the researcher was unaware of some of the background theory that has inspired the research. Meaning is always co-constructed, but in the process of construction one can choose to put more weight on the words of the participants and less on the pre-existing theory. There have previously been implications that inductive analysis and constructivist epistemology were not a comfortable fit, but this is based on too rigid a view of thematic analysis and one absent of 'knowingness' (Braun & Clarke (2019a)). The flexibility

of the approach means choices are on a spectrum – but not an unlimited one. Flexibility means that the approach can be used with a variety of underlying assumptions. It does not mean that these assumptions can be applied inconsistently, or one can proceed without 'knowingness' of what said assumptions are. Furthermore, to be Reflexive Thematic Analysis, in their conception, one must recognise the centrality of the researcher in the production of knowledge. In my view, the sense of qualitative methodology that I took in this research accords with the big-Q Qualitative, reflexive, researcher subjectivity-owning heart of Reflexive Thematic Analysis (Braun & Clarke (2019a)).

Thematic Analysis involves the following basic steps, as laid out in Braun & Clarke (2006): Data collection, data familiarisation, generation of initial codes, organisation of codes into draft themes, reviewing these themes on the basis of the codes they contain and on the basis of the data as a whole, identifying the key aspects of the themes and thereby giving them names, and giving a report of these themes with apposite examples.

Themes in RTA are not simply data summaries, but an explanation or account being told about a pattern one has identified (or constructed) in the data. They are generated by the researcher, and are the result of the choices made by the researcher (Braun & Clarke (2019a)). Through memoing and reflective journal keeping, the influences on this process were noted, reflected upon and where appropriate made explicit in this manuscript – for example, in Appendix 6. I was drawn to the metaphor of a solar system, found in Braun et al (2019) – where the theme is the solar system as a whole, the parts, such as subthemes, that make it up, are the planets, and the sun is the uniting concept that holds the theme together. This was my view of the themes and subthemes in this project – that themes encompass the large but coherent idea, and subthemes are major parts of that idea, that are valuable to

understand in more depth, but the theme is not simply the addition of subthemes. To go back to the solar system metaphor – if a subtheme is a major planet, that does not mean that the theme is simply the major planets – it contains the uniting ideas as its sun, and related but perhaps less individually significant ideas as its minor planets, asteroids etc.

3.6.2 Process of the Analysis

3.6.2.1 Data Familiarisation

The first step of the analysis was data familiarisation. This involved reading of the articles both during the data collection phase and after being collected. This process of familiarisation then gave me guidance on which parts of the data to re-read in more detail. I made sure to read blog posts that originated in a wide range of the blogs, and of the types of blogs, encouraging both breadth and depth in this reading, being sure to read carefully a number of the entries in the long single author blogs. Therefore, when I came towards the next step, I had a sense of the blogs that might be the richest starting points for coding, but also a sense of some of the spread of perspectives in the data.

3.6.2.2 Initial Coding

Within NVivo, I used the PDF feature, and a coding strategy that looked specifically to code lines or passages directly relevant to the research question. This approach had to be more selective, and therefore was more contingent on my choices and awareness at that stage than if the data had come from interviews or focus groups. I gave particular attention to comments regarding the person's relationship with their body, how others viewed their bodies, and how they understood and made sense of their illness. This was somewhat based on the data familiarisation stage, where certain entries stood out as having significant

'density' of these comments, as well as the ongoing examination of the data, to ensure that the codes were not overly based on particularly rich or evocative blog posts.

For example, the passage

"It was always encouraged from a young age that I keep my diagnosis under wraps. Only if they needed to know that I had sickle cell would anyone know."

was coded with the initial code *Needing to keep quiet or hide something about the body*.

As can be seen in the example, the coding aimed to be as direct a summary as possible of the data, with any thoughts about interpretation put into memos rather than codes at this stage. Given the large volume of data, the focus of this stage was on reading and initially coding as many different blogs as possible, trying to ensure the widest range of ideas and concepts were surveyed. This was done until a significant number of these initial codes had been developed, and I believed that there was a good coverage of the different blogs. An example page of initial codes is shown in Appendix 4.

3.6.2.3 Code Organisation

The next part of the process looked to develop links and associations between the initial codes. These initial codes and the content for which they coded were reviewed, checking the fit of the coding but also to aid the search for similarity. Although codes can be organised hierarchically as one way as developing from codes into themes, in this case, it was not my view that this was possible. This was because of the variety of different codes representing different aspects of the data and my understanding of it. I therefore made connections between pairs of codes at a time, either that they seemed to be linked by similarity, dissimilarity, or in some rare occasions, causation or influence. This utilised the

Relationships feature in NVivo, which allows codes, memos and other items to be connected, with types of relationship specified. For example, the code *Being small and weak as a child* was associated as being similar to *Early health vulnerabilities coming from SCD*; the code *Body as prison, disease as death sentence* was marked as being dissimilar to *Defying the deadline given by doctors*; the code *Diagnosed very young* was recorded as seeming to lead to the code *Feeling that SCD is a narrative that encompasses one's life from the beginning.* An example page of code relationships is shown in Appendix 5.

Using the Relationships feature enabled the use of visualisation features. At the smaller scale, the Explore Diagram feature allowed for quickly observing what relationships an individual code had with other codes, and what documents contained data coding by that item, and therefore to review the code itself and where other relationships could be recorded (e.g. the third part of a triangle, where a code was associated with two other codes, but those two codes were not yet associated with each other). At the larger scale, the Project Map feature was used.

Figure 1 shows a screenshot of part of the project map tool of various codes, once a significant number of relationships between the codes had been recorded. (Due to the size of the project map, only an exemplar snapshot is included, which means certain parts are cut off). The project mapping was not an attempt to engage with a more quantitative process, but to help think about and consider patterns – to take my perspective to a wider level and encompass in a glance multiple concepts. Where (through the relationships that had been formed) codes were clustered together, this would prompt me to consider whether this reflected that there was a uniting or organising idea that might be the beginnings of a theme.
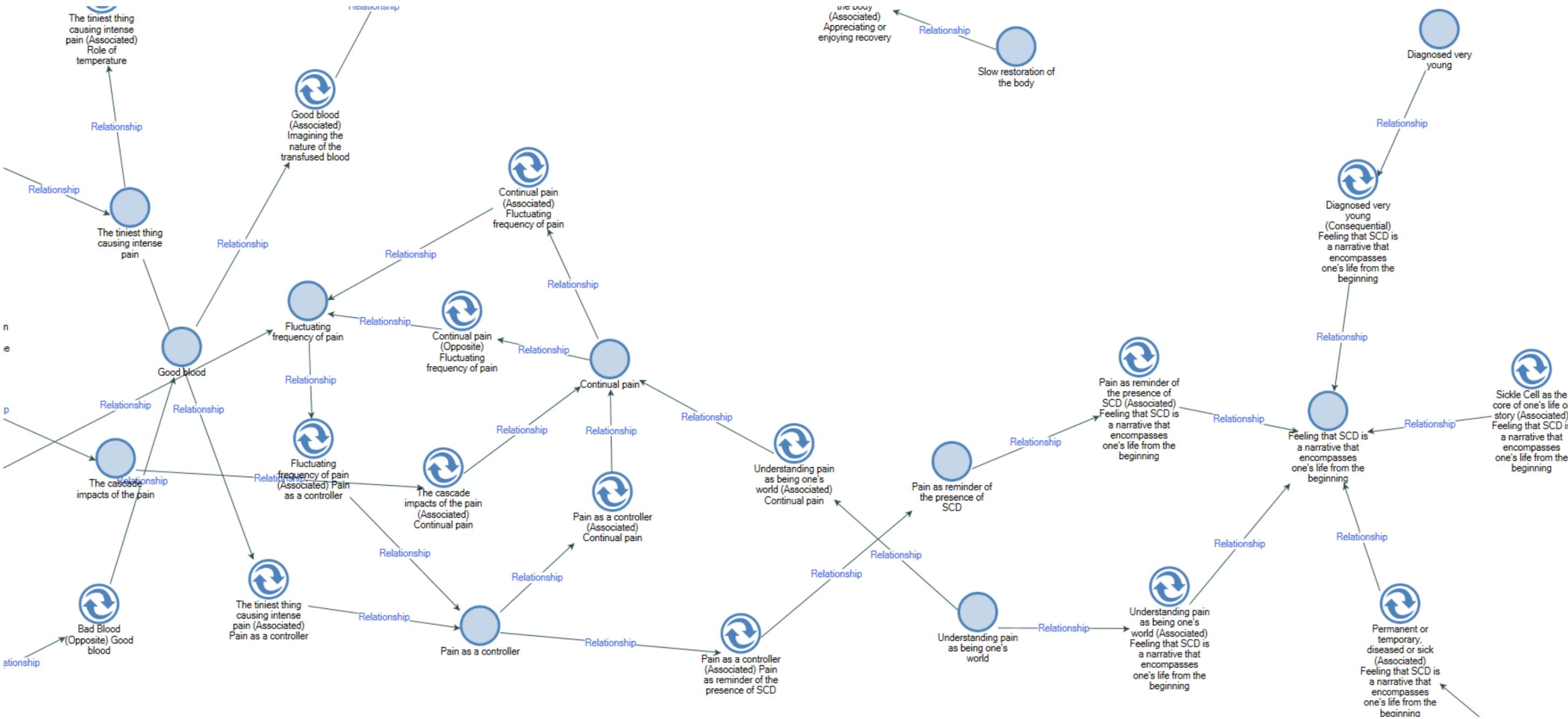
Figure 1

Seeing this area in of the map (shown in Figure 2, again cropped) containing these codes then prompted my thinking about what might conceptually encompass them together, developing some of the ideas that would come together as subtheme 1A. At the same time, if there were codes or collections of codes which seemed significant but were separated from other clusters, I would look to see if there were any appropriate links between them that I had not yet made.
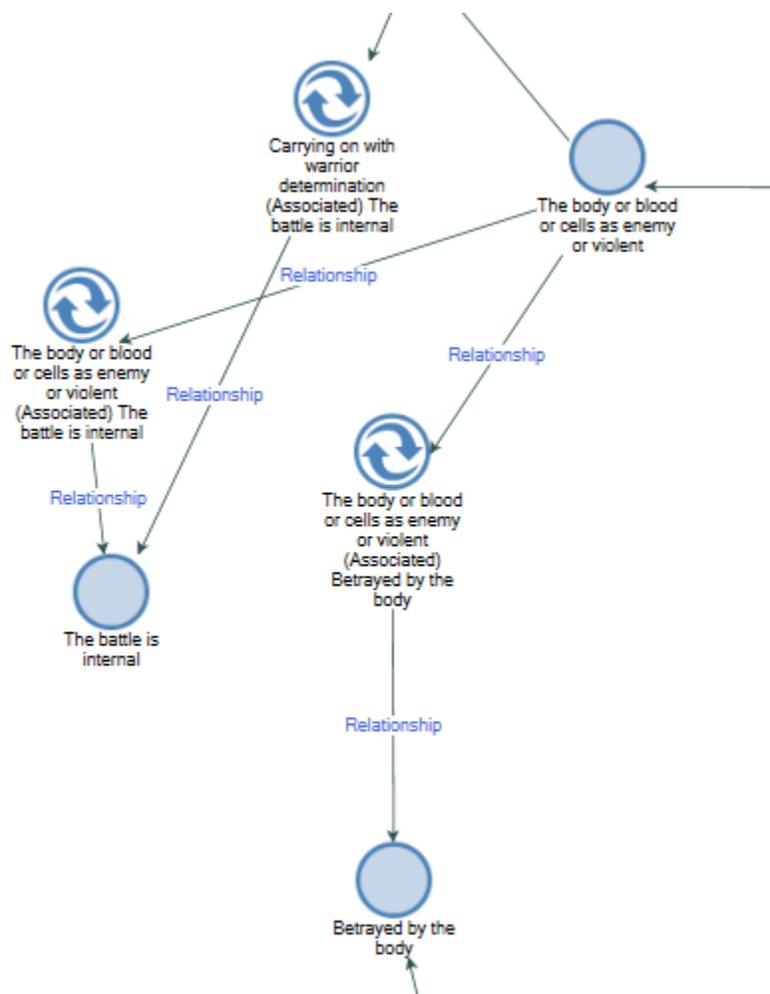
Figure 2

Through this process, I would note down ideas of potential themes or other thoughts, and ongoing reflections about the impact of the project and the analysis on me personally (see Appendix 6 for another excerpt). Some of that took a form closer to poetic, for example in this memo:

"We know our bodies

We know them because we experience them

Every bit of them, every creak and failure and pain, every momentary relief,

We're conscious of it all

So when I tell you that I need blood

I'm speaking a language deeper than numbers

When I say that something is about to go wrong

I have listened – so listen to me"

I then read and re-read the data to see if the links were viable, whilst seeking for and noting down ideas that would help create the narratives of the theme. This was a form of going 'back and forth' between the maps level of a broad view and the text level of a close view, trying to hold both in mind as best possible alongside opening up to ideas that related to the more than the individual parts. This included using the search function to locate any additional examples that supported the links being made, that may not have yet been included, to ensure that initial approaches to reviewing and coding data did not exclude valuable parts. Braun & Clarke (2019a) emphasise that themes in Reflexive Thematic Analysis are not just summaries of the content of a domain, but look to express shared meaning. Furthermore, they describe them as being an endeavour that requires the creativity of the researcher, standing at the meeting point of the data and their own knowledge and subjectivity. It was in this way that I constructed the themes – in close connection to the data but open to the interpretative possibilities that might craft a narrative that united these parts of the data.

3.6.2.4 Reviewing Themes and Theme Naming

These two steps, reviewing of the themes and naming them, which also involved conceptually clarifying them, were a process that overlapped and could not be separated into different steps. My approach to RTA has been to not take everything written as a mechanistic and prescriptive procedure, something supported by its developers (Braun et al (2019)).

When I had constructed some possible themes, I wrote a summary of them and created a diagram (an evolved version of which is shown in Figure 3). I discussed these with my supervisor and project consultant, a process which led to some evolutions in the nature of the themes. Whilst duality was identified as a significant part of the findings, it became de-emphasised as being one of the themes, since this occluded the idea that there is a process of development from enmity towards love and care, as well as collapsing multiple experiences of duality into a single one. Another significant change at this stage was that the theme and subthemes related to identifying with the body had originally been stated more strongly – in essence, "I am my body." Looking more closely at this and discussing it led to moving it from being totalising – that the individual finds their body is the sum of them, to being centralising – that the individual finds their body is more and more important to them and core to their identity, for good or for ill. The theme names were altered to reflect these clarifications, aiming to convey a clear and brief overview of their contents.

3.6.2.5 Reporting the Themes

The report of the themes is the next chapter. Having determined the nature, structure and naming of the themes and subthemes, quotations were identified and selected by looking at

passages marked with codes particularly salient to the themes, with an attempt to draw upon as many different authors as possible whose writings supported the theme.

Chapter 4: Analysis

In this section, the themes and subthemes will be described along with quotations.

4.1 Themes

Through the thematic analysis process, the following themes were constructed:

Theme 1 was titled 'The Battle with my body, and how to win it'. Theme 2 was titled 'My pain-filled body is unique and central to my identity'. Each theme contained two subthemes. Within Theme 1 was Subtheme 1A – 'Body, blood, cells, genes as enemy' and Subtheme 1B – 'Victory through loving one's enemy'. Within Theme 2 was Subtheme 2A – 'Identification with the body strengthened by SCD' and Subtheme 2B – 'Pain maintains the awareness of SCD'.

These themes will now be described – firstly in each instance as whole themes and then as subthemes.

4.1.1 Theme 1 – The Battle with my body, and how to win it

This theme incorporated the many associations with battle found in the data. A familiar description in Sickle Cell Disease (SCD) is that of being a warrior in a battle. In this data, the battle idea could shift to being a battle that the person with SCD has with their own body. Sometimes this could be portrayed as an impossible struggle, but there are means of victory. These means included attitude and practical choices of self-care. Winning, then, could be seen as a movement from resentment and antagonism to co-operation and even appreciation. For some, this came more easily than for others – not everyone appeared to go to the fullest extremes of alienation and warfare or of self-love, but many seemed to relate to their bodies in a dynamic

way along that spectrum. Not being a narrative analysis, it was not possible to distinctly establish if this were a linear process, an oscillating one or some other pattern. It could be that people progress from the recognition of a battle, to seeing it as with their body, to reconciling that 'winning' is reliant on self-love and self-care, though this is not a one-off and final victory. It could be that dependent on the level of pain one is in, the relationship with the body one has established, one's general capacity for compassion and care, and myriad other factors, that one moves back and forth between resentment and opposition to one's body, and appreciation and love for it despite the illness.

This first theme started from the language of being a warrior or in a battle which is frequently used by people with SCD. It was also seen frequently in the data:

"Survivor, fighter, warrior, soldier – you are all of these!" Blogger 1

"Whoever has never had anyone checking on you or even saying hello, or even telling you that you're beautiful? These people are strong! They woke up today! They are a fighter, are a warrior; you can conquer all that you want to by the help of Christ." Blogger 2

"Due to sickle cell, I have been limited in many aspects of my life – work, school, relationships and having children. My body has been through war." Blogger 19

Whilst the understandable expectation would be that this refers to battling the disease, it seemed that this could often overlap with or evolve into one's body being the target of the battle. This is expressed in more detail in Subtheme 1A – 'Body, blood, cells, genes as enemy'. Whether through the stigma, the limitations the illness often brought, or simply the difficulties in separating oneself from a disease that is genetic (of which more in subtheme Subtheme 2A –

'Identification with the body strengthened by SCD'), bloggers could describe coming to see their body as opposed to them:

"The body would also be seen by me as an enemy rather than a friend." Blogger 3

"My body volatile and indecisive

Trying not to panic, panic will take me down

New blood has done no good

Messed around with by my blood anyway

A scream, an urge at least

Doesn't make things any easier

This rollercoaster is still going too fast

And I don't know what's coming next

Except this thought:

"Body, are you betraying me?"" Blogger 9

Betrayal was a common framing of this enmity by the bloggers – that the body is something that *should* be on one's side, but in the way it brings pain, weakness, frailty and limitation, it is breaking that promise and abandoning its duty:

"In the worst way, my entire body betrays me." Blogger 9

"I've been trying to hold back the pain, but now I cry. The idea that my body can betray me and not allow me to look after myself makes me cry in disbelief. In relief, I cry as an ambulance is coming." Blogger 3

Whilst a key current in the way the battle is won was made particularly explicit by Subtheme 1B – 'Victory through loving one's enemy', more generally ideas of endurance, persistence and patience were also present within the theme:

"My potential will not be limited by sickle cell. I have become more focused on what I want out of life. As long as I am consistent and endure, my options are endless." Blogger 3

I'll be honest with you, I wish I always embodied the best of my job, but the more tired and in pain I am from the Sickle Cell, the harder that is. Staying patient is a challenge, but it's one I'm trying to get better at. Blogger 35

In general, the battle appeared to be understandably seen as lifelong and sometimes this could be expressed in terms of taking an interminable and exhausting time:

"Eventually, sickle cell would win. Every time. Always. That was my life – worn down by fighting. Exhausted by the exhaustion, sick to death of my illness, ready to simply give in."

Blogger 1

The bloggers described how this fight did not give the option of pausing or stopping:

"Almost gave up there, but I kept fighting. I had to." Blogger 15

"No breaks... Sickle cell means you never get breaks from your illness!" Blogger 14

However, there was the sense that it was also through fighting that the capacity for triumph was developed, including the afore-mentioned endurance and patience:

"You can be made to feel different, don't worry, I know that! But I see that being different is also special. Why is it special? Because you have to be a fighter. Because you have to be strong

on the inside. Because you come through struggles, serious ones. That's all special. So be

proud of being different." Blogger 10

And the idea of fighting also entailed having comrades-in-arms:

"My first thought was that I was the only one fighting this battle, but when I found out that

there were others out there fighting the same fight I was, it gave me peace of mind." Blogger 7

This may have led to a mutually reinforcing idea around fighting, since it is such a

prevalent metaphor in the community of people with SCD.

Although the key form of 'victory' is elaborated on particularly in the second subtheme

of this theme, there were also related parts that were more practical in nature. For example,

particular techniques of self-care and ways of managing a crisis:

"A common sickle cell symptom is fatigue. The sickled cells do not carry as much oxygen than

non-sickled red blood cells. The fatigue comes from not getting enough oxygen in our body.

But there are three things I do find helpful: 1. Keep hydrated 2. Make sure you sleep as well as

you can 3. Have a healthy diet." Blogger 3

"You cannot just expect things to work out without doing anything. Be aware and keep away

from the things that can trigger a crisis – being too hot, being too cold, being stressed – in the

body or the mind – not drinking enough water, et cetera et cetera." Blogger 32

The means of winning the battle were not static. It was not described as being the case

that once one had reached a position of love for the self and the body, everything was sorted. For

example, at times the writers described feeling frustrated with the times they did not act in accordance with self-care:

"This week was tough. Overdoing things at work especially was a mistake. What makes me treat myself like this? Even though I know I have a problem, I take things for granted and act as if everything will get better even if I exert myself too much." Blogger 10

Across the themes and subthemes, there was a duality at play, both of the severity and difficulty of the battle as presented by the bloggers, but also that it was through struggle that they spoke of finding growth.

4.1.1.1 Subtheme 1A – Body, blood, cells, genes as enemy

The first subtheme of the first theme was concerned with the ways in which the body, either overall or some subdivision of it, seemed to be positioned as bad or dangerous. These subdivisions were usually those most directly related to the condition – blood, cells, or genes.

For some, it is the cells who they called the aggressor:

"Inside the person suffering from sickle cell, the cells are faulty and they're doing the worst they can. They call this a 'crisis'. The cells, the opposite of smooth, scrape and crash inside my blood vessels, creating the most intense pain. This pain is like metal grinding on metal, an unbearable agony right in my skeleton." Blogger 4

"What are we fighting? Early mortality. What are we trying to handle? Bones that are broken – cells that are broken too. Strokes and organ damage. And pain enough to suffocate."

Blogger 5

For others, it was the blood that was described as not to be trusted:

"Just gotta hope my blood count doesn't go down again. Sure, it can look good, but so long as there's pain there's time enough for treason. God – keep the count up and keep me strong."

Blogger 9

At other times, the body as a whole was portrayed as the opponent:

"The fact that we are having a fight with our bodies on the inside is bad enough; it would be nice if someone would create something to support us as we make our way to life instead of giving up on us like so many of us have." Blogger 9

At still others, a part of the body commonly affected by the illness was the part described as a villain:

"Take a look

Meet my eyes

Yet again betraying me

Another time, and another" Blogger 9

The reasons that the bloggers suggest for why they feel their body is an enemy could relate to the restrictions that the body with SCD brings:

"The fact that I have sickle cell limits my activities in many ways. Strenuous exercise triggers pain in my body, so I cannot participate in it. Others do many things that I would like to be able to do, but I am not able to." Blogger 18

For some, this enmity, or at least resentment, dated back in their personal story to childhood:

"My mother wondered what was happening to me. She fed me everything nutritious, she took

every bit of advice from the post-natal clinic, and applied it thoroughly. In spite of it all, my

body was ungrateful". Blogger 7

As elaborated in Theme 2 – 'My pain-filled body is unique and central to my identity', people also strongly identified with their bodies. This was another significant duality that I perceived in the data, since one might anticipate a contradiction between distrust or hostility and close identification. Instead, this could mean that the battle is expressed as a civil war:

"My body is at war

Fighting itself

I just want freedom

From pain

So why is it that fighting

Is the only thing here?" Blogger 9

A part of this forcible and battling embodiment appeared to connect to how it limits how a person can be seen, how they can build their own identity. Although such aspects were not widely described, perhaps compared to my initial expectations, they were touched upon:

"I do not get any choice or say in the sort of blackness it determines for me. I, like many first

generation black Brits, want to define what being black means to me. I don't want to be

pushed into one way of being understood, simply because of my uncontrollable blood."

Blogger 4

Although expressed at the various levels shown above, the cells or the blood, though intrinsically connected to the physical nature of SCD, did not appear to be seen as different or separate from the body. Instead, they seemed to be a synecdoche, metaphorically representing the body as a whole. Each come across as ways of expressing the difficulty of the relationship with the body.

4.1.1.2 Subtheme 1B – Victory through loving one's enemy

The second subtheme of the first theme described 'winning' – which is to say, feeling less overwhelmed by the struggle, and living as well as one can. This seemed to come through the evolution of the relationship of antagonism already discussed into one of attunement, respect and even love. As already mentioned, not every writer described traversing the whole spectrum from seeing the body as a total enemy to being totally beloved, but within the data there were a range of positions. For this reason and the many religious allusions made by the writers, this subtheme's title reflected the more dramatic expression of the idea.

It appeared that bloggers described love and acceptance as the vital ingredient in being able to overcome the strength of the illness and resolve the battle with their bodies:

"It is only through acceptance that we can win" Blogger 6

"Love is the antidote to all illness and all sickness" Blogger 29

Although this form of analysis did not permit more precise accounts of the process, various aspects of how the development of the self-love and self-acceptance occurs were described by the bloggers. For example, part of this development may have come from reframing, and changing one's perspective:

"The focus should shift from what we can't do due to our health to all the things we have accomplished despite it. Those things are to be celebrated." Blogger 3

Another part of the development of self-love may have emerged from having appreciation for the parts that have come only through having had SCD:

"Having Sickle Cell causes me to thrive some days. Because of it, I have an emotional maturity beyond my age, and I practice meditation." Blogger 33

"Sickle Cell is the uniting theme of my life, of every part of me – I'm learning, now, that that includes my character. Living with an illness has meant I've had to grow. This growth has meant some positive attributes have developed in me. Warmth, compassion and loving fun are a few. It is because of these things that I am who I am." Blogger 13

"It's not obvious that sickle cell can have a good side, and I'm not saying it isn't difficult. But this is what it has given me: a strong will, patience, kindness, love, and approachability."

Blogger 15

Self-knowledge, especially in regard to the body, seemed to be another important part of this position of self-regard and positive self-relating:

"The hospital staff, they thought everything was fine. The blood count was normal, they said. But I knew that there was something. It is something that you just know, when there is something starting to go wrong in your body." Blogger 9

"Even in their 20s, others (like myself) have to learn how their bodies work... Since before I was born, I have had Sickle Cell Disease. But everyday there is still something new to learn – about the disease and about my body." Blogger 12

There were various ways of describing self-knowledge and listening to one's body – practical, emotional or spiritual, but each appeared to rely on having an openness to and regard for the body:

"Your body can let you know what's happening, weird as that may seem. You just have to learn how to tune in, make sense of what it's saying – and with a lifelong condition, it's vital to. Of course medical knowledge is important too, but sometimes health professionals can be arrogant whilst sharing it. And they can't be an expert in your unique body – only you can do that. Nobody has more experience with your body than you do, because it has been yours for the whole of your life." Blogger 11

"And not just what the body is saying, but the things you sense and subtly feel. Maybe it's the voice of God, or your own voice. Maybe it's spiritual, or intuitive, or wise. Many times, the instincts and reflections you have will be your most helpful guide." Blogger 11

"Listening to your body, and learning how it communicates when you may be about to have a crisis, is the best way to manage sickle cell disease." Blogger 22

One area that seemed to be deeply significant is the attitude that the person then takes to one's body and the positive possibilities that that opens up:

"[To see one's body as the enemy] is a deeply ineffective and even detrimental view... So when I started to see my body as being on my side, I was able to be more hopeful. I was able to be courageous – not just making assumptions that my body was going to shut down whatever I wanted to do. My openness with others increased too – this new viewpoint helped me see the value of telling people about my life with sickle cell, that I could even help other people – and I've carried on doing that." Blogger 3

This positioning appeared to be expressed as part of an attitude of love towards oneself in general, with the body as a part of that. This seemed to be described as being an active process, and having consequences in action:

"It is the perfect time to rediscover yourself during this time of quarantine.

Get to know yourself in a way you never had the chance to before.

You do not need to feel or be lonely because you are not alone.

"We All Gotta Love Ourselves"" Blogger 2

"It was then that I realized what self-love is all about. Nobody will love and care for you unless you do it yourself first. Focus on what you need, put yourself first, and everything else will follow. Despite the fact that it is difficult, self-love is absolutely necessary to excel in life. Every now and then, I have to remind myself that it is a work in progress. After realizing that, I began doing things that made me happy again." Blogger 14

In this particular quotation, there were suggestions of the impediments to this self-love, which in this account seemed to have a lot to do with the body, and refinding acceptance of it, despite how challenging it can be:

"When did I fall out of love with myself? How did I stop loving me? I don't know, but I know that I did. As for why – these changes in my body – made me stop doing everything I had to, to be good to myself. I had accepted the battling, but that acceptance was fading away. After all, with scars like these, so dramatic, doesn't it seem that part of me must have bled out through them? Embracing the war wounds, that what it takes for me to love me. Doesn't mean I don't care about the fighting I had stopped seeing them as a matter of pride.

So, to reset, to love again, let me tell you about them. The war isn't over, I'll have to battle on. But I can't wait for peace to get started on loving myself. To move forward, leave behind the past and all the visible parts I didn't like. This is my advice – to me, and to you – you see the scars, you see the war, and it makes it hard to love, to even accept yourself. Instead, be proud! Scars mean strength. Scars mean courage. Scars are honourable. You have earned many medals for your bravery – show them off! Don't be ashamed that you were hurt in battle – you are a survivor! Survivors deserve to be proud, and they deserve love and to be looked after. You can be you, just you, without shame or apology." Blogger 2

In this quotation, there was the suggestion that this attitudinal shift is the foundation on which other things, such as the more practical measures mentioned in Theme 1 – 'The Battle with my body, and how to win it' were based:

"You must love yourself first before anything else and as long as you do that, the rest will take care of itself. As you travel through life, you should focus more on the journey than how fast

you make it to your destination. You are in charge of your life, and it is valuable. Be kind to

yourself, be aware of your worth, take care of yourself, don't take anything for granted, but

most of all, love those people who love you." Blogger 14

There were various examples of how this apparent attitude shift could potentially relate to

greater appreciation of the body and its various aspects, that were described as opponents in

Subtheme 1A – 'Body, blood, cells, genes as enemy'. For example, even the sickled cells

themselves could appear to be appreciated:

"Crescent moon cells, sickle moon cells.

The moon shines its light

The cells give us life

What beautiful cells! Beautiful like the moon!

My bones wail with the pain you bring,

My organs hit by the problems you cause

You are still beautiful." Blogger 30

It seemed that there were multiple ways that the attitude of loving oneself and one's body

could have been influenced. It could perhaps have come from following the example of those

who loved them:

"My mother sometimes told me, "don't bother yourself with negative thoughts, everybody can

be unwell, even the strongest people get ill." I held onto those words in this lonely world,

embraced them as my motto, and went about my life as any other person would." Blogger 7

Age and experience may have been another potential factor that affects the capacity of self-love:

"Over the years, I have realized that I need to preserve my body – physically, nutritionally and how I care for myself. The effects of sickle cell attacking you are many and can be almost impossible to cope with, which is why I do what I can to care for myself as best as I can."

Blogger 1

Thirdly, the recognition of the lifelong nature of the illness and that there might be limited other options if one wants to live well may have been another possible influence:

"Due to the fact that sickle cell will remain with me forever, I have chosen to accept it as part of who I am." Blogger 17

As has already been noted, it was not correct to imply that any development of self-love and regard was an absolute or singular experience. Instead, there seemed to be depicted a mature and flexible position, recognising both what one needed to do for the best and also that it is not always possible to enact this, and thus to not criticise oneself for this:

"I was determined to get in shape and make myself stronger, and I succeeded. I radically reduced my pain relief, and I just felt stronger in my body. When you have sickle cell anaemia, making your body strong is crucial, but on those days, on those painful days, you don't have to go to the gym or go out, since I know how hard it is to even get up from bed on those days."

Blogger 1

"Get to know your body and take note of what it responds to, either positive or negative, so that you can make better life choices. In this way, you ensure that your body is protected — at least as far as is in your power to." Blogger 13

Perhaps, therefore, it could be suggested that reckoning with failure and difficulty in self-care and carrying on in spite of them, was a part of the extension of love to oneself. For example, there was this description that it is not always easy, even for those who had recognised it as being necessary:

"I decided to become selfish for New Year. I'm not going to be selfish in the sense that I won't share chocolate or take an extra seat on the bus, but selfish in that my own health comes first for me. It's only been 3 weeks, and I've failed already...

My sickle cell warriors – please prioritise your health. If you already do, great! Keep up the good work! You should not wait until you can't stand the pain before seeking medical advice, as I tend to do. This won't help, it'll just keep you in pain for longer and raise your chances of experiencing further problems." Blogger 28

4.1.2 Theme 2 – My pain-filled body is unique and central to my identity

This theme was about both the increased identification with one's own body and the way in which the chronic pain of SCD is a key cause of this. This encompassed the entanglement of SCD into one's personal narrative and life experiences, that it appeared to be something firmly established in their body and in their life. The salience of the body, as a limiting aspect of self, as a definer of one's identity, was described, both in terms of inner and outer identity. The body, and its differences due to SCD, seemed to create a distance and difference with others which could be present in a variety of contexts and was hard to avoid or surmount. Although this identification

and sense of uniqueness was partly due to the attempts to avoid crises, and the various changes in body function due to SCD complications, the part that was seemingly most present and recurrent in people's experience is pain. The suggestion was that pain is the continuing reminder of SCD, of one's connection to the body and, often, how one is constrained by it.

A description that seemed to recur in the data was the portrayal of oneself as different from others, and that SCD was the reason for this.

"We who have sickle cell are different, no matter how much we want to convince the world that we aren't; we are." Blogger 37

In this quotation, for example, the blogger seemed to describe the separation of one's bodily experience from others, but that they still have to share something of the uniqueness. This was perhaps because the consequences of SCD, as hard as they are to communicate, were also unavoidably present.

"I could spend my entire life trying to describe the pain of sickle cell anemia, but those descriptions would mean nothing to anyone who doesn't have sickle cell... Even so, I still have to explain how sickle cell works to everyone, even friends, family members, and employers... It is a very isolating disease, and the way it makes a person feel is unimaginable." Blogger 3

In this quotation, it appeared that although the person's parent tries to reassure them otherwise, they saw themselves as being diseased – in other words, more inherently unwell, with the possible implication that it is therefore more central to their identity:

"Me: It would be stressful to have a child who is diseased. I can't imagine the stress.

Mum: You aren't a diseased child! No matter when or where, I didn't think of you as diseased. Occasionally you became ill, sometimes even very ill, but you always recovered." Blogger 4

In another blog featuring narratives from both a parent and a child, the parent apparently explained how their sense-making from very early in the person's life was impacted by the knowledge of the illness and the way they viewed or imagined the pain. With the other ideas in this theme, it is possible that these early life experiences helped in bringing SCD into the heart of one's sense of self:

"Even though we weren't sure what to expect when faced with a crisis, the early newborn screening test was helpful since we already knew he had sickle cell disease. Had we not known, we would have never taken him to the hospital during his first painful sickle cell crisis. He probably would have only been given a bottle because we thought he was just crying. Instead, we recognised that something had to be severely wrong and took no chances."

Blogger 16

After the early life experiences, it was also educational experiences that seemed to reinforce the idea of the person being defined by their body in sickness and fragility. This again appeared to have an effect of feeling different:

"During my high school years, I experienced a lot of hurtful words. My classmates would say to me, straight to me "you are the walking dead". Though I pretended to not care, it really hurt. The fact that I would die is clear, but did you have a right to keep reminding me of it?"

Blogger 25

"I can still remember, just like that, when I first really 'got' that I had Sickle Cell – and got that my life was not going to be normal. I was in the fifth grade when one of the school administrators came into class. Her plan was to find students who the teacher thought were good, and arrange a field trip for them. I was pointed out by an admin, and when the teacher saw me, straightaway she told her no because: "It's not that she isn't a good student, but it's her health. What if we take her on a trip and she dies?"

I cannot adequately express how I felt at that time. Sometimes I was ill (back then, it wasn't common for me), so I missed class and a couple of field trips, as it happens. To be honest, before then, I had never known about how this disease would affect me, let alone all the limitations I would have to endure, because of having Sickle Cell. My childhood, at least, the innocent, playful, unworried childhood, ended that very year." Blogger 26

The presence of pain in the words of the bloggers was unsurprisingly widespread. Pain could be described in terms of being trapped or as a prison, a metaphor for the all-encompassing nature of the illness. It could lead to an extension from pain to SCD to the body as being the prison:

"I am unable to describe how painful my crises have been. There were many times when I wished for death simply to end the agonising pain that had engulfed my whole body... It had become my whole life – it had become the only thing I knew at all – just the crises, the pain of Sickle Cell Disease." Blogger 10

"The hardest thing to deal with, with sickle cell is that there is nowhere to hide or run. In many ways, it feels as if my body remains a prison, preventing me from escaping." Blogger 1

"I sometimes feel trapped within a narrow tunnel, the light so far away. Physically, mentally, and emotionally, the treatment at the hospital left me traumatised and scarred. Sometimes I feel so heavy inside carrying all that hurt and pain. It makes me so nervous, so anxious to know a crisis is just around the corner. Once upon a time, I was afraid of going into crisis because I knew what was coming, yet more of the war that I would have to mentally prepare for. If there is no end in sight to this darkness, then everything seems hopeless. Despite all the suffering I have been through, I don't know if I will ever be free of this burden. It's the toughest thing I've ever faced." Blogger 1

The use of carceral metaphors could even extend to the idea of living under a death sentence. Once again, this may have implied the extent of isolation, and its significance for one's identity, as in the second quotation here where it seemed to subsume a person's whole being:

"Receiving the Sickle Cell diagnosis was like receiving a death sentence." Blogger 4

"I had no label other than the label of death. My father disowned me, claiming he did not create children who were sick. He said to my mother that I might look alive, but I was already dead, just still moving around. He saw no value in my life at all because I would die." Blogger 27

The bloggers' desperation with this situation, of the body filled with pain and feeling trapped in that, could reach particular depths. At this point there could be depicted a vision of freedom from one's body in death itself:

"I can tell you that I had many dark thoughts, really dark. I would beg to die, beg that death would take me so at least I would be free at last from my body. I couldn't keep going weighed down by my body – because it was my captor, my dungeon." Blogger 1

"The complications I had from sickle cell are so many, and the same with difficult medical treatments: Transfusions, exchanging red blood cells, surgery, mini-strokes, chest pain, ulcers on my legs, priapism – and more still. These things become day-to-day reality – because there was something worse. I would not be able to sleep for days, even weeks. My family would all do their best to help, staying awake in shared desperation – massaging my legs, keeping pressure where it was needed, propping me up, getting hot water for the hot water bottle where it might make a difference. They were all doing their best to help me sleep but it was for nothing. You reach a certain point and words fail you, trying to describe the level of pain. All I can say is it was enough that I asked my doctor to put me out of my misery – either amputate my legs so they couldn't hurt anymore, or give me enough drugs to get the eternal sleep."

Blogger 29

This sense of imprisonment – isolating and consuming of self – could perhaps be something that progressed with a greater number of crises and hospitalisations. In this quotation, for example, the blogger seemed to suggest that this was reinforced by how their caution and expectation of fragility increasing the sense of trappedness shaped their behaviour:

"Sickle cell can at times make your body seem like a prison. For example, if your mind has a plan but your body cannot do it – not a hard plan, nothing complex, but normal stuff... It seems that the more often I became ill and the longer I stayed in the hospital, the illness rolling on through years and years, I became more restricted, and I limited myself." Blogger 1

As with the idea in Subtheme 1A – 'Body, blood, cells, genes as enemy' of the resentment going back into childhood, so too did the general sense of trappedness and suffering, connected to the pain and SCD. In this quotation, there was again the significance to their personal narrative of the pain-filled life:

"I have this feeling – when a baby is born, they take their first breath, and they cry. I think that first cry says something about the rest of their life. For me, it was a deep lament. A lament for everything that was coming in my life from that moment on." Blogger 7

The uniqueness of the pain-filled body seemingly did not always lead to the negative, drawing on ideas perhaps from Theme 1 – 'The Battle with my body, and how to win it':

"Sickle Cell pain itself is the proof of your strength and your survival." Blogger 21

4.1.2.1 Subtheme 2A – Identification with the body strengthened by SCD

The first subtheme of the second theme was focused on the way in which the presence or knowledge of the illness impacts upon people's identity and sense of who they are. This could potentially lead to heightened importance of the person's body as a centre of their story or selfhood, through comparison with others or the senses of what it cannot do. Many of the examples seemed to mark out the bodily differences due to SCD as the salient cleavage between themselves and others.

The sense of self may have been entangled deeply with SCD, as seems to be expressed in this quotation:

"There is a foundation on which all the different levels of my life are built. It was there at the

beginning, but it's part of every bit of the edifice. And so it will be until the very end of my life.

I have an illness: sickle cell disease." Blogger 13

As has already been described, this could be something that went back to early in their

life. For example, the knowledge of SCD was described by some as occurring in young

childhood. Additionally, the second quotation made reference to the separation and uniqueness

described in Theme 2 – 'My pain-filled body is unique and central to my identity':

"Around the age of 5, I started to understand what it meant for me personally to be diagnosed

with sickle cell." Blogger 21

"The day my parents explained the meaning of the how my life's suffering, the near-constant

pain, that was the most momentous day. I was young – only seven, but they started to let me

know that I was different, that I was not like all the other people." Blogger 18

Although this analysis was not in a position to assess change over time, in the various

accounts of the bloggers, there seemed to be depictions of a role of others in developing this

identification, especially earlier in life. This may have included an identification made of being

different, and one's physical state being why. That could be the advice of parents, and the

contrast with the experiences with one's peers as a child:

"What bad luck! Sickle Cell Disorder is enough of a curse, but needlephobia is a really

annoying extra. I did my best to be brave, but that's not how you want it to be as a child. Other

kids could play, didn't have to worry about things, whereas my parents did not allow me to do

even the littlest thing. They wouldn't let me get wet, for fear of getting sick. Of course, I

managed to do it once, but I learned my lesson – the crisis was a bad one." Blogger 7

Another possible part of the development of one's identity being shaped by the illness could be the attacking jibes of non-understanding classmates:

"Some of the children thought I had demons in me, others figured I must have epilepsy, and

others saw me as fragile and vulnerable, permanently sickly and ill. Bullies saw that and

picked on it." Blogger 7

"I would have liked to join in sports and games, just like the other children in school, but I

didn't understand why I was singled out and couldn't. I got special treatment because of

having sickle cell – more breaks, but not doing any activity that was too strenuous. I'm very

thankful that my teachers got told what to do, about my sickle cell, because my dad made sure

to tell them." Blogger 15

Even the intended-to-be-helpful words of a medic could appear to add to the sense that one's identity was tied up with SCD, separate from others, and as with the other examples, the body was the determinant:

"As a child, what it meant to have sickle cell was still a mystery, especially all the knock-on

problems it can cause. My doctor tried to explain it in a way I would understand – saying "if

we were a sandwich, everyone else has jelly inside and you have peanut butter." And you

wonder why I might have issues about that? Being told I was not like everyone else, that I

wasn't normal – he wasn't wrong, I am different." Blogger 1

Hospitalisation was another potential driver of people experiencing the intrusion of SCD into their life, such that their choices and actions can be changed by it, and they may find that it is in the body that they differ from others:

"When I was 6, I had loads of health-problems. I was in hospital a lot of times in a row, because of crises. And because I wasn't allowed to do certain things, that made me different from the other children. There was judgement and prejudice, people seeing me as dangerous and unpredictable, that something was going to go wrong, it was just a question of when. I didn't like not being a normal kid, so I just acted like I was. Which doesn't work, because my health would fall apart. Instead of being normal, I was super-normal, because my cells had their special shape." Blogger 6

The multi-faceted physical (and beyond) impacts, especially of the severe and varied complications, were another possible part wherein one's sense of self was changed by SCD, and where the nature of the body was significant to one's sense of self:

"It has been said by me before that sickle cell anemia can have devastating effects on your body outwardly, but it also has effects on your internal systems. Among the organs affected by sickle cell disease are the liver, kidneys, lungs, spleen, heart, bones, and joints. I was only 19 and already my body felt wracked with age. It terrified me, and still does, because if that's how I felt then, what is my body going to be like in the future?" Blogger 1

Alongside the complications, the multiple and repeated reminders of mortality appeared to be another contributor to the bloggers' sense of self and identity being determined by their bodies:

"There is a constant reminder to sickle cell patients how short their life expectancy is. We are reminded both by literature and by our daily experiences. My life is fragile and every time I am hospitalized or put into intensive care, I am reminded of that." Blogger 13

Even if spoken with humour, this identification could contain the elements of frustration and antipathy described in Subtheme 1A – 'Body, blood, cells, genes as enemy':

"Come on, body of mine! Tell me what you want! You need me and I need you – we're linked, remember – so let's try and figure out how to help each other." Blogger 31

The closer identification with the body was reflected not just in the afore-mentioned limitations and sense of difference, but also in descriptions that suggested that many of the other parts of one's identity or sense of self were contingent on what happened to the body due to SCD:

"I literally suffered a decay of my body, which in turn stole my inner happiness." Blogger 1

"Because of having my period, I had a severe pain crisis every single month. You can manage to prevent some of the others, but not that one. So I missed a lot of school. I stigmatised myself because of this – a new level of problem. I was just angry at life and just being here, and I got really depressed... I just wanted a break from sickle cell, a permanent sleep seeming like the only way." Blogger 36

Whilst identification with the body was emphasised, it was not at the expense of other parts of being, but rather that the physical part may affect or intrude upon all the other parts of life:

"I told you before, about how for me, even if it's not normal for other people, there's a part of

my life that's the foundation of all the rest. I suppose, because of that, people only see the

foundation and nothing else about me – or they completely miss the links between sickle cell

and the other things in my life. I have sickle cell disease, and it really does impact on every bit

of my life. You cannot just see it as a blood problem all on its own. It changes everything. The

pain would be significant all by itself – but mobility is affected, energy levels are affected,

mental health is affected. It is by no means easy to have one's life built on such a thing."

Blogger 13

4.1.2.2 Subtheme 2B – Pain maintains the awareness of SCD

The second subtheme of the second theme regarded the role of pain in being the most powerful reminder of the presence of SCD in a person's life. Though, as already discussed, there appeared to be numerous reasons why a person's awareness of SCD and incorporation into their identity occurs, pain was deeply significant. SCD pain, which was depicted in powerful and evocative ways by the bloggers, seemed to be of such an intense character. This, perhaps, went some way towards explaining why it kept the presence of SCD close at hand. Though frequently invisible from the outside, the writers suggested that it was near-impossible to lose sight of it or forget it in their inner experience. Because of this, and because it appeared to be connected by the writers with SCD, it may have been an experience that brings one's sense of illness back into consciousness again and again.

Although part of the close identification with the body could come from the intellectual awareness of SCD, the individual experience of pain was a huge part of what kept it present. This

quotation gave this suggestion, and connection with the overall theme of Theme 2 – 'My pain-filled body is unique and central to my identity':

"I could never escape knowing that I was abnormal. That my genes were anomalous. That was what the pain reminded me. That was why I hated it." Blogger 7

The vividness in which the pain was described gave some impression of how difficult it could be to ignore. In this quotation, another element that seemed to be relevant is that that has been true since the person with SCD was in childhood:

"There are different pain descriptions for sicklers, but here are some of the most common: being stabbed again and again, or being clubbed in the same spot over and over with something heavy. When I was still just a kid, I would explain how much it hurt by saying that it was like having knives forcing their way out of me. In her description of her pain, my younger sister, who has the disease, said it felt like a spike was roughly piercing her body."

Blogger 8

The ease with which pain was triggered made it seem powerful and the body, and the person themselves, by comparison fragile and vulnerable. That such easily prompted triggers, which one might encounter, or have to try to avoid, every day, evoked SCD pain, may be another way in which the person's awareness of the illness was maintained:

"My body would shiver with significant bone pain at the mere touch of a drop of rain."

Blogger 32

Similarly, the capacity of pain and of the SCD crisis to overtake every part of oneself, could possibly have reinforced the sense of the presence of the illness. This in turn may have impacted how one behaves, with the pain and its level of control being an apparently important factor:

"During a pain crisis, your body and mind are completely taken over, as is your ability to function. Your world goes dark, and every bit of you is disabled... My focus has gradually shifted to getting done all I can do, and then making sure I stop when I tell I've reached my limit – the point where the pain has fully taken over." Blogger 10

However, where one might be tempted to avoid the pain or pay less attention to it, there was the imperative from others or from experience, that one has to respond to it, for fear of worse suffering. In this way, it seemed, even minor pain raised the issue of the illness, since it may herald the more significant pain of crisis or other complication:

"You cannot sit back and not pay attention to sickle cell pain, no matter how minor. If you sense even a little bit of pain in your body, and don't act, it'll become a bigger and bigger problem." Blogger 21

Even with efforts to forget, these rarely succeeded. Alongside the wisdom of responding to pain in order to keep it from worsening, there may also have been a role for the intensity of the experience. In this quotation, the blogger seemed to identify that the long-lasting pain, and the hospitalisation whilst in that level of pain, are parts that made it both desirable and impossible to forget.

"Pain and suffering, that's what I remember. And not just brief spells of pain, but memories that stretch over a long time. I would fight so hard to not feel that long-lasting suffering again... I try and wipe it from my mind – the time spent in hospital especially, the days on end that are stuck in my head. I wish all of it could be forgotten." Blogger 34

Those writing could appear to portray that the disease was in control and they were not, and that the pain was closely linked to this:

"Sickle Cell had control of my life. At all times. It was agonising pain, the worst you could imagine, so bad I just wished I was dead." Blogger 10

The pain might possibly have predominated in part by diminishing or outright taking away parts, identities and experiences of the self. These were elements from which one might otherwise be able to build a life less dominated by SCD:

"My childhood was robbed by the pain of sickle cell disease." Blogger 24

"Pain and low energy are obstacles I need to endure. A traditional career is impossible for me... All areas of my life have been impacted or limited by sickle cell disease. In view of my health condition, I cannot see my own future with confidence, affecting my mental, emotional, and physical health. My faith is strong and I am not influenced by statistics, however my body is deteriorating. Through life, there have been so many times when I've wanted to give up."

Blogger 38

Although one cannot have an awareness of the illness before having the diagnosis, it seems that the constancy of the pain could give a continual sense of being sick. This sense of

unwellness could then eventually be given a name, but it appeared that even without a specific

medical name, the experience of a significant illness made clear by pain could be similar to those

who had known their diagnosis for some time:

"When I look back to my childhood, there was this continual, but mysterious pain. Awful pain

that I didn't understand – obviously I knew something was wrong, but not that it was Sickle

Cell. It was only as I grew up more that I found out about the illness, and what you can do to

keep going. But because it wasn't obvious from the outside that I was unwell, it was like it was

hidden away." Blogger 20

The latter part of the quote also touched upon how the awareness appeared to be an inner

awareness – that, potentially, it was inescapable for the writer to know they had SCD whilst at

the same time it was very difficult for this to be seen externally. For the above writer, this

perhaps led to feeling isolated. For others, such as in the next quotation, this could appear to put

the lie to one's attempts to seem well, and threaten the hiddenness that one has adopted to

attempt to manage. This may also have related back to the wider theme – that other aspects of

SCD, as well the pain, might induce being seen in terms of one's body, in terms of one's illness,

regardless of how one wants to be construed or understood:

"People who are chronically ill try their best to appear well, and if that's not possible, conceal

the illness as much as possible. Sure, if it's been completely unavoidable, I've let people know

about the misery of my body... There is one part that can never be fully hidden. My family –

even my husband, now he's learnt how – see the secret code in my eyes, the yellow that warns

of danger ahead. I have been walking down the street, taking the tube, and some doctor or

nurse or whatever will look at me and I will know that they know. Sometimes they will actually

talk to me, thinking that I might not know how bad things are." Blogger 4

4.2 Linkages and Visual Representation

Through the theme development process, although distinct themes and subthemes were developed which deal with conceptually separate areas, linkages were made between the themes that may add to the overall understanding of the research. This is also visualised below in Figure 3.

For both of the themes, the subthemes described parts of the wider experience, but one can be suggested to precede the other. In Theme 1, the realisations identified in Subtheme 1B tend to come after the positioning (or recognition of positioning) in Subtheme 1A. In Theme 2, the identification with the body described in Subtheme 2A tended to be a consequence of the experience of persistent and pervasive pain. Finally, the possible cross-linkage between the themes is that strongly identifying with one's own body, but in a way driven by pain and other difficulties, may be a contributor to seeing one's body as an enemy. A heightened sense of difference from others due to the body might stoke resentment, the multiple impacts into all sorts of parts of one's life, and the body's role in that frequent limitation, may increase the sense that it is something to be battled and overcome – at the very least, mistrusted.

Theme 1 –
The Battle with my body, and how to win it

Theme 2 –
My pain-filled body is unique and central to my identity

Subtheme 1A – *Body, blood, cells, genes as enemy*
I am fighting a battle – struggling against my own body as it constrains me

Subtheme 2B – *Pain maintains the awareness of SCD*
Because of the often lifelong experience of extreme pain, there is no point at which SCD is not salient to my life and my experience

Subtheme 1B – *Victory through loving one's enemy*
However, as I go on fighting, I discover that to conquer, to win, I have to listen to my body, respect it, show it love.

Subtheme 2A – *Identification with the body strengthened by SCD*
And this strengthens my identification with my body – how can I ignore it? Pain+SCD+the body can become synonymous with the self.

Figure 3

Chapter 5: Discussion

5.1 Summary of findings

This research aimed to look at the body image of people with Sickle Cell Disease (SCD) and way in which they related to their bodies. Two themes, each with two subthemes, were constructed in response to this.

Theme 1 – 'The Battle with my body, and how to win it' encompassed the duality of relating to the body – that people could come to see their bodies as enemies, as working counter to their well-being, but then could also come to see the way to overcome this was to treat themselves and their bodies with love. Subtheme 1A – 'Body, blood, cells, genes as enemy' and Subtheme 1B – 'Victory through loving one's enemy' focused more deeply on these respective sides of the duality. In this theme, then, the body image of people with SCD could be seen to be dynamic, not so easily expressed as lower than or the same as those without SCD. Instead, it was better seen as journey of relating to the body, with peaks and troughs, of times of antagonism and times of affection.

Theme 2 – 'My pain-filled body is unique and central to my identity' provided a different viewpoint of the question – that for good or bad, body image is very important for those with SCD. Subtheme 2A – 'Identification with the body strengthened by SCD' suggested that the condition of SCD leads one to have the body as a significant determinant of one's identity and Subtheme 2B – 'Pain maintains the awareness of SCD' suggested that the significance of the body, one's view of it and how one relates to it, was influenced to a great extent by the extreme chronic pain of the illness.

In relation to the research questions, these themes offered some answers to the question of how people with SCD relate to their bodies. The themes showed a complex relationship with dualities between enmity and close identification, battling whilst also loving, with the experience of pain being an important part of how it appeared that the relationship to the body is influenced. Ideas that have some parallels with body compassion and body shame could be seen in the themes. In considering the research question about the nature of the body image of people with SCD, there were suggestions of how people with SCD can perceive, think about and feel towards their bodies, and some of the possible reasons why. Although, perhaps due to the data coming from blogs which are likely to convey a number of inner feelings and thoughts, the themes aligned more with cognitive-behavioural conceptions of body image, there were parts of the themes that can speak to the socio-cultural conception, especially in terms of close social relationships such as family relationships. It could be suggested that just as the relationship with the body is a complex one, with the potential for deep negatives and positives, the nature of body image explored in this study was also one that incorporated negative and positive aspects, as the wider field of body image has increasingly done. Ideas around enmity whilst simultaneously being closely identified may be relevant in the body image of other people with other chronic illnesses, or with other reasons that their body is (made) salient.

Through the review of the literature, it was noted that there had been limited research conducted with people with SCD in relation to body image. This research had tended to rely on narrow ideas of body image that had been developed outside people with SCD. This research began engagement with the parts of body image and ways of body relating that may be

particularly significant for those with SCD. Later in this chapter, there are suggestions for further research that builds on this research and might continue to populate the gaps in this topic.

5.2 Links back to initial literature

5.2.1 Links with the initial literature on chronic illness

In this research, there were findings suggestive that one's body became an especially significant part of one's identity, and that pain was very important in this. Łakuta et al (2016) described mechanisms of chronic illness impacting mental health in their study of psoriasis. One was an increase in salience of one's appearance and its appraisal. The present research provided a description of this occurring and some of the ways how this might occur for people with SCD. Edwards et al (2005) suggested that there were changes in self-concept experienced by people with SCD, which they related to the impact on the body, especially in adolescence. For Edwards, they ascribed this to delays in physical growth and development, citing evidence for greater concern about this than social and family concerns, frequently a feature of adolescent life. In this research, pain was an important, though not exclusive part of keeping these concerns at the front of one's mind. This, therefore, could apply at any time in a person with SCD's life, though it is understandable that it is observed at a time like adolescence when the body, already shown by this research to be made salient by SCD and its pain, is additionally salient. This research also gave a possible account, therefore, why concerns about the body are expressed over other concerns common to adolescents – that the illness and the pain it causes have heightened one's identification with the body. If one is more closely identified with the body, then concerns about it are likely to be prioritised. It may then be that this obscures identification with more

predictable or secure aspects of one's life, such as family or friends, this could be one of the causes of mental health difficulties described in Edwards' study.

This research described a relationship between people with SCD and their bodies, with many dualities – between seeing the body as an enemy and learning to love it, of identifying closely with that body whilst also finding it as an antagonist, of suffering deep pain that makes one feel trapped in one's body whilst also seeing oneself as having developed positive qualities impossible to find without having had SCD. There were many ways to respond to SCD, and different people at different times experienced different emotional and psychological reactions. It was interesting to compare this to Edwards et al (2005), who suggested there was a cyclical relationship between the physical and psychological parts of pain in SCD, and argued for the condition overall being seen as psychological and social as much as it is physical, because of the complex interaction of these factors. Edwards et al (2005) draw upon ideas about emotion and pain that come from general psychological research, though noting that they seem especially pronounced in people with SCD. However, it may be that this is due to the impact of the meaning-making is associated with the pain. For example, based on this research, the pain's effects on how closely connected to (or trapped by) the body a person feels, could also have an impact on mood. Edwards et al (2005) described the pain that tends to cause depression, anxiety and other mental health difficulties as being 'unremitting'. The ideas seen in this research about the pain evoking inescapable memories of lifelong suffering, of feeling like one is a prisoner in one's body, is out of control and ruled by SCD, all of these could be thoughts and sense-making that understandably leads to a drop in a person's mood.

Additionally, if as subtheme 1A indicated, one sees oneself as at war with one's own body, at moments of feeling defeated, and when one has not developed the features of self-love and self-care that can be the resolution to said war (subtheme 1B), then in these moments we would anticipate a person to be low in mood and unlikely to take the practical steps to manage pain. Łakuta et al (2016) said that there were negative emotional reactions about one's body in those with psoriasis. This research both reflected this possibility in SCD, whilst expanding it by encompassing the complexity of the relation, as the relating to the body is not wholly negative or positive, but dynamic, especially over the course of a person's life. There were difficulties in coping with pain highlighted by Edwards et al (2005), both in terms of how practical strategies to help might be appraised, and in the expectation of pain recurring and life being limited. Again, this present research gave an account of a complex relationship, where a person may see helping themselves with the condition as fighting a battle, loving themselves or both at once. It showed one's close identification with the body may indicate feeling a prisoner of pain or a person with a strong identity shaped by their journey through SCD. Different positions on this journey may be useful ways of understanding some of the difficulties described in past research.

This research found that one part of way in which the way a person related to their body intrapersonally was based on how others related to it interpersonally – in positive and negative ways. For example, some drew on their parents' example in learning to love and care for their body; others took on board their parents' rejection of them, because of the illness and its effect on their body. A similar distinction was seen in Olsson et al (2016), researching those being treated for blood cancer. They found that the internal or perhaps intrapersonal sense of the body seemed to be impacted in different ways to the external/interpersonal sense – the significant

changes they found focused on the latter, though they acknowledged this may be due to the instrument used. This may be a different experience for lifelong conditions as opposed to cancer and other illnesses that do not begin at the start of one's life, relate to the duration of the illness, or apply to some illnesses more than others. In Olsson et al (2016), the changes in body image reverted when the person was some time after the cancer treatment. SCD, by contrast, is a recurrent and lifelong illness, and treatment can leave lasting changes to the body – such as scarring, or joint replacement. This research described experiences that were reflective of that, of a longer journey with the illness and with it being far more central, and needing to be integrated and made sense of in a way that cancer or other more time-limited illnesses perhaps do not.

This research provided a way of viewing body image for people with SCD as a dynamic process, one that has space for contradictions and differences, one that is unique to individuals in timescale and in whether they reach the place that might be considered most constructive. This may offer an alternative view to the contested view held in the existing literature: Cepeda et al (2000), contrary to much other research, put forward that body image and subsequent low mood was not significantly different in those with SCD than those without; Alao et al (2003) and Bhatt-Poulose et al (2016) for example suggested the opposite. Instead of only reflecting differences in validity of measurement or statistical power, these studies might, in light of the present research, indicate some of different ways of conceiving of one's body and relating to it. This could suggest that results that differ across differing samples could be due to the ideas of body image tapped into, the ways of measuring mood and the relationship between those two aspects, which may capture different parts of the wider experiences of body relating that are occurring. The participants in the present study described a range of influences (parents, peers, pain, and more)

that increased the resentment towards and the regard for their body, as well its importance to their sense of self, and it is reasonable to conclude that the individuals making up a sample group will differ from each other on these facets, in ways that may be difficult to measure quantitatively.

This research showed how people with SCD articulate numerous positions regarding themselves (especially their bodies), some focused on endurance and patience, some feeling defeated, some feeling fragile in some ways, others feeling strong in others. It was not simple to be a person living with SCD – one is neither wholly the living corpse as seen by some, or the unstoppable warrior seen by others, but can feel like both and every possibility in-between. Forrester et al (2015) concluded from their research that the adolescents they studied showed positive self-concept, if looking at the overall picture. However, this did not preclude frustrations and difficulties, especially in relation to pain crises. In that study, the participants reported that they did not like to disclose that they had SCD, and that talking about it more frequently caused them to feel more negative. In this study, SCD, often regardless or against the person's wishes, became important to one's identity and made the body an important part of that. This could create tension for the person – if it is quite central to a person's identity but they also do not like to think of it. These sort of struggles could be the sort of struggles this study described, for example, in developing enmity with the body.

In subtheme 1B, a person's positive motivation towards self-care and their actions of self-love were highlighted as vital factors in how a person with SCD can respond to their body. In this and in Theme 1 as a whole, there was a development of attitude and of technique in self-love

and self-care. This could be a plausible exploration of some of the connections between disease-specific self-efficacy with self-care ability and actions, for example in the model first described in Jenerette & Murdaugh (2008). Modelling of, or giving of, love, was something that the participants in subtheme 1B also identified; it was therefore intriguing that Jenerette & Valrie (2010) found that having had the experience of being cared for by a parent had an impact on self-efficacy in adulthood. In this research, parental love, rejection, overprotection and many other reactions were important elements described by the participants in shaping their relation to their body and their sense of identity.

5.2.2 Links back to initial literature on Body Image

5.2.2.1 Sociocultural View

Of the key sources of influences in body image suggested by Van Den Berg et al (2002), family was the most prominent in this research. For example, in Subtheme 2A – 'Identification with the body strengthened by SCD', parents were highlighted as being important to the formation of the relationship between the person and their body. Peers were also reported to be important, especially in the formative years at school. The internalisation process described by Tiggemann (2012) fits with the descriptions of the importance of family and peers in developing one's own sense of body image in this study.

However, in contrast to a strong emphasis of the sociocultural view of body image, media portrayals were not something that were referred to in this study. This may be because the bloggers came from a variety of different cultures around the world, and thus there might not have been significantly shared media ideas of the body. However, hegemonic ideas are

influential internationally, so it may be that their influence is more implicit – for example, in creating images of bodies seen as healthy, or communicating ideas (both factual and otherwise) about the causes and meanings of illnesses.

5.2.2.2 Cognitive-Behavioural View (including body compassion)

The core concept of the cognitive-behavioural view of body image – that people have beliefs and feelings in relation to their body which influence each other, as well as body-related behaviour – was seen in this research. For example, Theme 2 – 'My pain-filled body is unique and central to my identity' could be seen as showing pain as an activating event. The frequency of this activating event and the schemata described, for example of separation from others or the importance of the body, could then contribute to the distress experienced by the authors.

Theme 1 – 'The Battle with my body, and how to win it' could be argued to show how changes in the beliefs one has about the body, and the feelings one directs towards it, lead to changes in behaviour. Given that Cash (2012) suggests that potentially unhelpful coping strategies parallel those found in conventional CBT, it could be that enmity with the body is a form of over-generalisation or emotional reasoning. These may then reinforce beliefs around hostility through the ongoing pain being seen to confirm said hostility, and through the impact of these beliefs on reducing self-care behaviour meaning that pain does increase or endure.

Arguably, the findings of this research were a still better fit with the concepts of body compassion. With its links to Compassion Focused Therapy, the ideas then parallel the negative and positive positioning towards the body seen in this research. The respective activation of

threat and soothing systems (Gilbert (2013)), specifically towards the body, could be another way of making sense of Theme 1 – 'The Battle with my body, and how to win it'. Subtheme 2B – 'Pain maintains the awareness of SCD' may give a reason why the threat system is likely to be especially active in those with SCD. Being frequently in pain and it maintaining the awareness of the significant illness could understandably prompt a sense of threat, especially given this occurs from one's early years. In the body compassion literature, both in general (Berry et al (2010)) and specifically in those with disabilities (Smith (2013)), recognition of the body as unique and having responsibility for it have been identified as key features. In this research, these features were highlighted especially in Subtheme 1B – 'Victory through loving one's enemy'. In that subtheme, even SCD itself could be seen as having value – in other words giving a unique quality which does not need to be framed critically. Connectedness and ownership, and the consequent response of looking after and tending to the body and its particular needs, were also exemplified in this subtheme.

5.2.2.3 Feminist/Objectification View

As with the role of the media, in the sociocultural view, specific reference to views of the body being strongly shaped by the operation of power and objectification were not found in this study. Again, similarly, their presence can be suggested implicitly, especially when considering the source of the discourses drawn upon by peers and family members who treated the bloggers badly. Both general ableist/eugenic ideas about the value of people who are chronically ill and more culture-specific ones, such as evil spirits from Christianity and Islam, or àbíkú in the Yoruba culture, are alluded to by the bloggers. However, there is not an explicit discussion of this, there is no identification of the powerful observing objectifier.

More generally, the role of race was only infrequently referred to by the bloggers in this study. Whilst elsewhere in their blogs, some wrote about anti-Black racism in a wider sense, links between this and SCD were not explicitly made in almost all cases. This was also the case for other aspects of identity where power and discrimination operate – such as gender.

5.2.2.4 Positive Body Image View

The positive body image view is a viewpoint that was certainly reflected in this study. The descriptions in Tylka (2012) of uniqueness and of listening to one's body both were exemplified in Subtheme 1B – 'Victory through loving one's enemy'. Whilst there were a large number of difficulties described with comfort and interpreting the body in a positive light, the spectrum depicted in Theme 1 – 'The Battle with my body, and how to win it' showed that the positive aspects of body image were present in the experiences of body image analysed in this study. Acceptance of one's body was a feature that appeared to be particularly important, though understandably significantly challenged by the difficulties connect to SCD.

Indeed, many of the blogs appeared to have the purpose of sharing experiences with other people with sickle cell, with encouragements for continuing to live with the illness, including some of the positive attitudes in Subtheme 1B – 'Victory through loving one's enemy'. This idea of mutual support of a positive body image was also a process identified by Tylka (2012).

5.2.3 Notable differences from the initial literature

Certain factors identified as relevant to body image, especially in chronic illness, were surprisingly not particularly discussed in the data. It is important to review this and make some consideration, although tentative, as to why this might be.

Attractiveness was one area identified as important to body image in people with chronic illnesses. Holzer et al (2014), especially, reported that lower limb amputees had lower evaluation of their attractiveness, whilst remaining as invested in it as the control group. What material there was that related to relationships (more commonly referenced than sex), tended to refer to the risks of transmitting the illness by having children with a partner whose SCD genetic status was unknown. Whilst outside the scope of this research, qualitative studies into these attitudes have been reviewed by Smith & Aguirre (2012). Kralik et al (2001) found that sense of attractiveness changed in middle-aged women with chronic illnesses, but that this in their study was identified as a change due to the emergence of an illness. Their participants questioned the prioritization of sexuality in their life, which may be one possible explanation as to why attractiveness was not referred to in detail. It may be that the participants wrote more of the day-to-day experience of pain and managing their condition, and body image in relations to this, as this preceded consideration of attractiveness in importance or in their conceptualizing. This research may differ from the results of Holzer et al (2014), where the importance of appearance and attractiveness was consistent with those without a chronic condition.

Athleticism and physical functionality was another area identified in the introductory literature on body image in chronic illness (for example, Holzer et al (2014), Olsson et al (2013)

in other chronic illnesses; Lee et al (1997), Panepinto et al (2005) in children with SCD). The capacity of the body to do certain tasks, including physical exercise, was referred to somewhat in Subtheme 1A – 'Body, blood, cells, genes as enemy', but often as part of the wider sense of the body letting one down. Again, this may be a comparison with illnesses or acquired disabilities that have an onset later in life, where there is a contrast to be drawn with one's own past function. Athletic activities, especially but not limited to the aquatic, are activities that people with SCD were often kept from throughout their lives. In that sense, this factor could be said to be present in the background, but perhaps not as prominently

Gender is an aspect of identity closely entwined with the development of concepts of body image, as noted in Grogan (2016). However, in the initial literature, gender was raised especially in reference to self-image in SCD. Bhatt-Poulose et al (2016) described gender differences in the body image of young people with SCD. Of particular note was that young men with SCD aspired to be larger in body size, bucking the trends of men without SCD and both groups of women. Costa et al (2018) focused on priapism, which is a problem suffered by cisgender men as well as some transgender women and non-binary people. In their study, their participants all identified as men and related the difficulty to self-image especially in relation to masculinity. Therefore, gender would be an area that one would particularly expect to be addressed by the blog authors. However, this was not the case despite the blog authors including male and female writers.

Theme 2 – 'My pain-filled body is unique and central to my identity' was the theme that most related to the idea of identity. Referred to mostly in this theme was the sense of separation

and difference that SCD had led to. It may be that, when writing spontaneously, rather than being asked about issues determined by the researchers, that the relationship between gender and SCD in identity is not as salient. Schüssler Fiorenza (1992) described that although every person's identity is an intersection of facets, that different contexts make one or other the lens through which the others are filtered at that moment. Perhaps in this research, where the data collected was focused on 'living with sickle cell', the chronic illness/disability facet was the primary lens of identity. It may also be that this particular research did not have a larger number of participants or an approach that was suitable to making comparisons between genders. Finally, the predominance of pain, as Subtheme 2B – 'Pain maintains the awareness of SCD' gave some examples of, may mean that other symptoms, which a person might connect more with their gender, were not in the forefront of their description of life with SCD.

Race is perhaps the most significant aspect of identity that is related to SCD. Atkin & Ahmad (2001), for example, suggested that it was a unique feature of SCD in comparison to other chronic illnesses. Royal et al (2011) found those who felt particularly discriminated against because of race, and those whose identity had their race at the centre, were more likely to feel that their SCD was treated differently due to race. As mentioned in Subtheme 1A – 'Body, blood, cells, genes as enemy', there was a small amount of discussion of race, and how SCD might restrict the possible expressions and constructions of race a person might be able to access. This was far less present than I had expected. It may be that the experience of racism relevant to SCD was less about self-self relating and more about other-self relating – the examples given by Atkin & Ahmad (2001), for example, were education, healthcare and employment. It is also of note that in Royal et al (2011), a substantial minority described that race did not have any impact on their

experience of SCD, and a very small number even suggested that it made camaraderie easier through having other shared characteristics.

As with gender, this may be one aspect of qualitative research where the creation of the data is not as guided by the researcher. Both Costa et al (2018) and Royal et al (2011) collected rich qualitative data regarding gender and race, respectively, by using interviews and questionnaires. It may also be that a research question that more specifically considered race or gender, and collected internet-based data, may find more to address questions about their role in SCD. Gabriel (2016) conducted a qualitative study of Black British women who blogged, suggesting that they were less anonymous than other bloggers, as a move towards authenticity. She found that these bloggers imagined an audience similar to themselves, and only secondarily considered readers who were unlike themselves. Finally, she suggests that these bloggers act to fill gaps in understanding and counter misinformation. In considering how her paper might parallel this research, it may be that when one has an imagined audience similar to oneself, that certain focuses or emphases are sometimes less likely. The blogs studied in this research often expressed that they were offering support and encouragement to other people with SCD. However, her idea regarding standing up to false or negative narratives might suggest that at times things taken for granted – for example shared experiences of racism, as well as the specific interaction with disability discrimination – would be made clear. Again, as has already been suggested, it could be that having SCD is the area of difference – when the blog posts in this research did seem to make an appeal to an external audience, it was often people who shared all other characteristics save SCD – such as the person's local community. In conclusion, it remains an area to be explored in future research.

5.3 Strengths and Limitations

A quality assessment for the research was completed and is included in Appendix 7. In part through using that framework, particular flaws that can occur in Reflexive Thematic Analysis were averted. Therefore, each of the facets of this research that are identified as limitations here also offer a strength. This duality is probably present in all research, but especially qualitative research where the researcher's choice is more clearly acknowledged as shaping the research. Therefore, each limitation and strength reflect a decision made, due to circumstance, experience, or intuition, which necessarily curtail some area of the research whilst opening up another.

A key limitation of the work was that due to the need to change the data sources as a result of the pandemic, the data was not data that came into being specifically for the purpose of the project. Data that comes from interviews can be made deeper or broader as needed during the process of the interview, and by virtue of the interview schedule and the intervention of the interviewer, is more directly addressing the research question. This can mean that the analysis is then based on data that is more cohesive in its content. Areas that might be addressed fleetingly by data collected in the way this project did can be dug into, where their contribution might otherwise be lost. Data collected online has different limitations on collection to interview data – for this project, the raw amount of data collected was vast, and required searching and filtering, even with the inclusion criteria used, to find the relevant blogs and sections within blogs, to address the research question. Because of the available amount of data having the potential to exceed the capacity of any project to adequately encompass it, decisions had to be made about

when enough data was collected and then of that data, how much being analysed was sufficient. As already referred to in the Method section, there are not defined points to draw these lines within the methodology used in this research. However, the choices I made were based on some specific factors. Firstly, an estimate of my capacity to read and review regarding the data collected. Secondly, when it came to analysis, trying to read as many different writers as possible, and read a mix of long multi-post blogs made over a sustained time period and single blog posts by different writers on multi-user blogs. Choices to collect more or less data, and to read more or less of the data that was collected, and the way in which I prioritised which parts of the data to read, will have impacted the conclusions of this research. This was a limitation, one that stems from the time and project size constraints as much as the nature of the data and the analytic method. However, it was a limitation that is likely to occur with Reflexive Thematic Analysis, and thus one I accepted with knowingness as to what felt the best, and possible, choice.

However, this difference in data source was not purely a limitation. It leads to drawing upon the writings of people with SCD who are writing for their own audience – often including others with SCD. The advantage was that the data is not then constrained by the questions asked or unasked by the interviewer, by the routes that the interviewer pursues further, at the necessary expense of others. The prioritisation was done by the people of the group being studied, and is structured by their frames of reference. It could head off in a range of directions entirely unpredicted by the researcher. Though epistemologically I did not believe anything is 'unmediated', it may be that it was less influenced by certain hegemonic forces, and engaging with it may have broadened the possible range of conceiving for the wider audience.

Another limitation with data being collected in this way was the implications for sampling. Reflexive thematic analysis is not reliant on seeking a sample that is relatively homogeneous, in the way that approaches like IPA do. However, collecting data online means that details of the identities of the authors were not always totally clear and the authors of the blogs were from different nations and backgrounds. The selection criteria did address this to some extent, but did not aim to reduce heterogeneity, for fear of missing out on data. Whilst this research drew upon the similarities and shared ideas that are in the data, it was not therefore able to answer questions about contrasts between different groups, different genetic statuses or severity. These are comparisons that have often been considered in prior research, usually that of a quantitative methodology. However, what can be said is that the research drew upon voices from the USA and the UK, where much of the published SCD research is done, plus nations in the Caribbean and across Africa. People in this research had differing access to healthcare, which would likely differ with an interview sample recruited in this country. However, missing from this research would be the voices of the less technologically literate, or with limited access to the internet because of income or infrastructure. This breadth of context and perspective contributed to the richness of the research, and may mean that commonalities regarding SCD itself are brought to the fore, or else the broad range of possible response in the large and varied world population who have the disease. However, it is also important to note that there were many authors whose posts did not indicate specifics about their background (such as nationality or religion). This proviso must be made when discussing the diversity of the authors. It was unknown whether people from certain backgrounds might be more likely to not include this information. It may be that certain of the multi-author blogs emphasised sharing this, because of the purpose of the blogs. In addition, although the strength of the varied backgrounds of the

authors can be pointed to – some authors provided far greater numbers of blog posts than others, so it could be an exaggeration to suggest that all of the findings of this research are equally based upon the writing of authors of each background. However, it is my view that large number of blog posts did not necessarily lead to a greater influence on the theme development, since as mentioned in Section 3.6.2.2, some posts were especially 'dense' with expression, whereas others, being written for other purposes or in different styles, contributed smaller amounts of relevant material across a number of entries.

This connects to the area of representativeness. Representativeness is not evaluated in the same way in qualitative research as in quantitative research, as the determination is based on the informativeness and valuable knowledge of the participants. A larger number of different participants was possible through the blog-based approach compared to interviews, but at the costs already described of the depth of the data and its homogeneity. This may have facilitated a more wide-ranging set of ideas being explored and used in the theme construction, but perhaps also put those themes on a frailer foundation than more targeted research that generated large amounts of data that was shaped towards the research question directly. It was not possible to establish whether those with SCD who write blogs, and those with SCD who participate in research interviews are more similar to the overall group of people with SCD. Indeed, each may be quite different in their own way. However, it was probable that they are different from each other, so by completing research that draws data from online sources, an additional layer is added to the overall reflection that may be possible of the population with SCD.

This research took a different approach to past research in the area. This was the aim from the beginning, and thus this research could offer something novel and valuable to the research literature. Even with limitations, it may have highlighted new areas and avenues that are of interest to researchers, clinicians and people with the illness, through taking this different angle. The other side of this is that the connections with other parts of the extant literature were made, as seen above, through similarity and analogy, rather than by directly drawing upon the area of interest and the pre-existing important constructs (e.g. self-efficacy). Whilst, as shown above, there was dialogue between this research and past research, this research was not placed to serve as confirmation or rebuttal of those studies directly.

5.4 Reflections

In the reflexive process, the part that most took me by surprise was the emotional connection I felt with the combined descriptions of chronic pain and religious faith. Being Christian myself, as many of the participants appeared to be, created a link that I had not fully anticipated prior to the research. I found myself challenged by theodicy (religious accounts of why suffering happens) that differed from my own. Most frequently, the blog writers described their sense of there being a plan for them that included their illness, and certainty that they were more blessed than afflicted. God may have allowed their pain, but for the greater good, and the more they trusted in Them, the better things would be. For me, the cause and the remedy of chronic pain, in relation to God, was quite different – that the pain was not God's will or plan, but a symptom of flawed and incomplete humanity, and one's difficulty with it did not 'track' with one's faith or trust in God. The surprise, for me, was the consistency of people describing

confidence in their faith more than doubt, and the will of God enacted in their illness more than either spiritual attack or a flawed world which God willed to be different.

At times it was hard, and challenging, to see the confidence and positivity held by the blog writers, both in this regard and more broadly, despite their substantially more painful situations and deeply more serious physical difficulties. At times I wondered, and memoed as such, whether the positivity reflected that there was no space to have the negativity; at the same time, as shown especially in the first theme, it was clear that the thoughts and conclusions came through earnest journeying and had made a significant difference in people's lives. Faith understandably influenced one of the subthemes, where it drew upon Christian language – perhaps if I were of a different faith or belief, then it may have been framed differently. However, the presence of faith, especially Christianity and Islam, and of God's will and protection, was woven through much of the data, and so it did not feel like an alien concept to draw upon.

At times, when my own chronic pain was especially bad, I felt deeply emotionally moved and connected – both in positive ways, feeling a pride in doing research that might have value for those with far deeper experiences of chronic pain, and in negative ways, feeling a resentment for the coping and perseverance that some of the participants described, when it was something I myself, with far fewer challenges, could struggle to find.

Throughout, there has been a tightrope of wanting to make sense of, express and share my responses to the data, and being wary of speaking over or over-imposing my own narrative.

This came both from the similarities, as noted above, but also the differences that were noted in the introduction. This tension may relate to some of the strengths and limitations, and the nature of Reflexive Thematic Analysis, where one is aiming to add something, not just summarise the data, but to add something that is based on a sincere and close relationship to the data. In reflecting on this, and in research supervision, it is my view that this itself is part of the process of this sort of qualitative work – that to proceed with no questions or tension is to proceed recklessly and without 'knowingness'.

5.5 Suggestions for Application

The first theme, with its subthemes seeming to form a sequence, could be the starting point for informing some psychological interventions. This could start, for example, from a place of identifying where a person with SCD is in their struggle, and what they see themselves as fighting against. It could then be based on supporting them in having the tools for that struggle – ways of developing the love and regard for self that are the ways identified from the data for a person to 'win' in the battle. In general, the overall findings suggested the greatest merit in a strengths-based approach – the second theme showed how a deficit- and difficulty-orientated consciousness is particularly hard to avoid, but the first theme suggested simply trying to counter this without cultivating love for the self may just keep one stuck in the struggle. This may overlap with ideas from Acceptance and Commitment Therapy and Narrative Therapy. For example, the thickening of subjugated narratives of self-love and strength could be a powerful intervention. Helping a person move from a conceptualised self to a self-as-context, or the link between struggle and suffering in relation to pain could also be very useful.

Throughout the data, there were often instances of patience and persistence being part of what a person had developed as a result of the illness, and working with a person to find their own individualised elements that they see as valued strengths that have resulted from their life with SCD could create a useful foundation to develop further into the 'winning' strategies of self-love and tenderness to their body. Just as Dobson (2015) found that equipping people with psychological pain management techniques improved self-efficacy, it could be that self-compassion and psychological self-care tools could aid those with SCD, both in moving forward from pure struggle and in bolstering the existing strengths and resources that they show. For example, expressive writing has been studied in relation to body compassion (Ouwens et al (n.d.), Sherman et al (2019)). Expressive writing, with prompts based on the themes of this research such as learning to make friends with my body, or having compassion for my pain, could be valuable both to try in clinical practice, or to research. More general self-compassion techniques, such as those from Compassion Focused Therapy, could also be used, with some tailoring to help the person bring the compassionate and soothing feelings to their body.

Another therapeutic approach which may have parallels with some of the ideas identified in this research is the relational approach of Barry Mason (For example, Mason (2003) and Mason (2016)). He highlighted some of the important facets of working with chronic pain in this approach – the role of meaning, stories and beliefs. In the qualitative research he described, the idea of relationship between a person with chronic pain (or members of their family system) and the pain itself was described as being primary or secondary. The primary relationship referred to the pain, or the condition that causes it, being prominent in the person's life, and reducing their connectedness to others. Indeed, as paralleled in this research, the relationship with the illness

could be consuming and hostile, and could cause many negative effects in one's life. A secondary relationship indicated that pain would not be absent from a person's life, but could be less prominent if other relationships were facilitated to retake the prime place. Intervention suggested by Mason includes applying a variety of systemic techniques to elicit and change the relationship with the pain and beliefs about the pain throughout the whole family system. This could be a valuable approach standing alone, or as a supplement to some of the more individual approaches that have also been suggested.

Guidance and awareness for physical health clinicians could also be drawn from this research. The research literature suggested that these clinicians are motivated to facilitate self-care, but it may be that they miss how they can reinforce ideas of difference and alienation that impact on the people with SCD. This research would suggest that getting deeper into the perspective of the people living with SCD, finding out about an individual's own relating to their body, their enmity and their love, their degree of identification and how pain influences this, could give valuable understanding for the clinicians in agreeing care plans, supporting self-care, and managing complications.

It may also be there are implications for the campaigning and support organisations. Battle metaphors are frequently employed by these organisations, often even in their names. If the battle of SCD is indeed (at least to an extent) against oneself, then it could be that those organisations would want to create spaces to reframe the battle metaphors they draw upon, such that it against the disease, or the stigma, or the striving to better treatments or to live well, or else to create a focus on that the ways of battling are those of positivity and compassion. They could

create information for their service users based on this research that helps inform and explain about close identification with one's body and help people reflect on whether this is an identification they find helpful or not.

Finally, there is a socio-political angle. Self-love as a form of liberation (for example, Baszile (2017)) is established in radical Black tradition as well as other revolutionary and liberatory frameworks. Whilst the situating of the body in its Blackness was not continually emphasised by the participants in this research, it was present for some, and other research has connected the ableism and racism that those with SCD experience in an intersectional way. The voices of the writers in this research often made declarations to their fellow people with SCD. There is an invitation to take the individual journey of self-love, but to accept such an invitation may challenge the societal and structural ways in which people with SCD experience the opposite of love. It may be that organisations fighting racism, ableism, unequal healthcare access or the consequences of colonialism could employ some of these ideas – that one can move from struggling with the self to embracing the self, following the example and leading of a group that is significantly and intersectionally marginalised.

5.6 Future Research

It could not be said at the time of writing that the pandemic was over. However, society was changing and adapting to its presence, and endeavours to manage and mitigate its risk had progressed. On this basis, possibilities for extending and deepening this research start, in my view, with exploring the same research question with a different data collection method. Interviews or focus groups, with questions prompted by the themes constructed from using the

internet, could allow for the validation, elaboration or clarification of some of the constructs made in this research. One area that could be explored is looking at the process of change between antagonism towards the body to care for the body.

It could be possible to explore these areas quantitatively as well – for example, creating a measure based on the concepts described in this research and giving it to a large sample, or using it for comparison with those without SCD or those with other chronic illnesses. Quantitative measures could also look at factors that predict differences in one's progression through the journey – for example, experiences of parental care, severity of the illness, attachment style.

As mentioned in the application section, if guidance for clinicians is developed based on this research, further research could ensure that tools or techniques provided for clients to increase body compassion or self-love and self-care are a good fit for people with SCD. This may include existing approaches and any original ones that are developed specific to SCD. Further evaluative research could also assess what techniques to engage physical health clinicians in having a better understanding of the complexities of body relating in people with SCD.

Given that self-efficacy, especially disease-specific self-efficacy, was one of the most frequent concepts identified in the systematic review, it could be valuable for future research to look at the relationship between the findings of this research and that construct. In addition, the other concepts such as self-esteem and self-concept could also be considered. This could be explored with qualitative methodologies – asking participants for their views on body relating,

self-efficacy and how the two interact and intersect, or on their self-esteem and the centrality or otherwise of their relationship to their body within it. With quantitative approaches, one could look at correlations between the factors and see if there is any part that is predictive or mediating of the relationships between them, and in relation to outcomes such as pain, mental health or healthcare utilisation.

This research would suggest that there is good viability for qualitative projects that draw on people's own existing accounts in chronic health research. As described in the strengths and limitations of this study, this may mean that a different section of the population with the particular health difficulty might be reached, who would not be as likely to be offered the chance to participate in research or to take it up. These could make use of a range of qualitative analytic approaches, or techniques such as content analysis.

5.7 Conclusion

This study aimed to find out more about the body image and body-relating of people with SCD. The findings described a dynamic relationship where people could seem to be in a battle with their body, but also found the only way to win that battle is through embracing self-love and care. Body image was found to be an important part of one's identity, as SCD (and especially its pain) kept the illness salient, and encouraged closer identification with one's body. The clinical implications were that intervention based on these ideas could be helpful – therapeutically, in increasing positive relating to the body and drawing on existing strengths; in physical treatment in understanding the dynamic relationship whilst planning care; in advocacy and support, seeing if these are ways of thinking that are valuable or need to be reframed. The last implication was

the invitation offered by the people with SCD to each other, but perhaps also to all people, to learn self-love, and in so doing resist the forces that commodify, denigrate, objectify, exploit and hate our bodies, especially those bodies that in whichever way, as SCD does, do not conform to the hegemonic ideal.

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Appendices

Appendix 1

Key to MMAT Type and Questions	
Types that appear in the systematic review: 1 – Qualitative 3 – Quantitative – Non-randomised studies, including cohort, case-control and cross-sectional analytic studies 4 – Quantitative – Descriptive studies 5 – Mixed Methods (specify types from above)	Each type has its own set of questions. For Type 5 studies, there is a set of questions and one completes the appropriate questions for the type of study that constitute the quantitative and qualitative parts of the mixed methods.
Questions for Type 1 1.1. Is the qualitative approach appropriate to answer the research question? 1.2. Are the qualitative data collection methods adequate to address the research question? 1.3. Are the findings adequately derived from the data? 1.4. Is the interpretation of results sufficiently substantiated by data? 1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?	Questions for Type 3 3.1. Are the participants representative of the target population? 3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)? 3.3. Are there complete outcome data? 3.4. Are the confounders accounted for in the design and analysis? 3.5. During the study period, is the intervention administered (or exposure occurred) as intended?
Questions for Type 4 4.1. Is the sampling strategy relevant to address the research question? 4.2. Is the sample representative of the target population? 4.3. Are the measurements appropriate? 4.4. Is the risk of nonresponse bias low? 4.5. Is the statistical analysis appropriate to answer the research question?	Questions for Type 5 5.1. Is there an adequate rationale for using a mixed methods design to address the research question? 5.2. Are the different components of the study effectively integrated to answer the research question? 5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted? 5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed? 5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?
Responses Y = Yes – there is evidence in the paper that the research DID achieve this standard of quality C = Can't Tell – there is insufficient evidence in the paper to determine whether or not the research did achieve this standard of quality N = No – there is evidence in the paper that the research DID NOT achieve this standard of quality	
Although the MMAT is not based on mathematical scoring, for an at-a-glance estimation of quality, the responses are highlighted in a colour spectrum based on total number of yes, ranging from red where no questions are answered yes, to bright green, where all the questions are answered yes.	

Table of papers for systematic review

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
Journal of Advanced Nursing	Lee E.J., Phoenix D., Brown W., Jackson B.S.	A comparison study of children with sickle cell disease and their non-diseased siblings on hopelessness, depression, and perceived competence	1997	Lee et al (1997) compared children with SCD with their siblings who did not have the illness. They used measures of depression, hopelessness and perceived competence. The construct of perceived competence included their self-esteem as well as their perception of their academic, physical and social abilities. The only significant difference in these areas found was in physical abilities, which they suggested was a natural result of having SCD.	3	CT	Y	Y	N	Y
Journal of Pediatric Psychology	Lemanek K.L., Horwitz W., Ohene-frempong K.	A multiperspective investigation of social competence in children with sickle cell disease	1994	Lemanek et al (1994) studied social competence and self-perception of younger children with SCD, comparing them to a control group. They did not find any differences in the self-perception of competence or the self-perception of acceptance between the groups. They suggested this may be due to positivity biases in younger children, though they acknowledge that it reflects what is found in older children with SCD.	3	Y	Y	Y	Y	Y
Journal of the National Medical Association	Tanabe P., Porter J., Creary M., Kirkwood E., Miller S., Ahmed-Williams E., Hassell K.	A qualitative analysis of best self-management practices: Sickle cell disease	2010	Tanabe et al (2010) conducted a qualitative study in self-management techniques. They gathered participants for a workshop discussion, selecting those who were thought by their peers to use a good range of self-management skills. They then used approaches informed by content analysis, within a grounded theory framework, to construct themes. The principal theme, which they based on frequency of mention, was self-awareness. This included knowledge about behaviours that individually led one into difficulties with one's health, as well as the development of body awareness – being attuned to one's body and following its lead.	1	Y	Y	CT	Y	Y
The Journal of Pediatrics	Kumar S., Powars D., Allen J., Haywood L.J.	Anxiety, self-concept, and personal and social adjustments in children with sickle cell anemia	1976	Kumar et al (1976) compared children with SCD with matched controls on a number of factors. Their assessment of self-concept included many dimensions regarding the children's sense of themselves in terms of behaviour, physical appearance, social status, psychological well-being and academic ability. They found that the scores on this measure were significantly lower for those with SCD than controls. Those with SCD did however score lower on anxiety.	3	Y	Y	Y	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
Journal of African American Studies	Reddy S.D., Edwards C.L., Wood M., O'Garo K., Morgan K., Edwards L., Wellington C., McDougald C.S., Feliu M., McNeil J., Whitfield K.E.	Body Image in a Sample of Adult African American Males and Females with Sickle Cell Disease (SCD)	2011	Reddy et al (2011) studied adults with SCD in relation to their body image. To do this, they had them complete a measure of body image developed for eating disorders, as well as stress. They found that for men, ratings of body dissatisfaction negatively correlated with stress, whereas for women, stress was positively correlated with body dissatisfaction, ineffectiveness and interpersonal distrust. They drew a number of conclusions from correlations that did not in fact reach significance, but they suggested that this gender difference would be important to study in future research, especially in how people with SCD related to idealised bodies.	4	CT	CT	Y	CT	N
Perceptual and Motor Skills	Gentry B.F., Varlik L., Dancer J.	Children's perceptions of psychosocial factors related to sickle cell disease	1997	Gentry et al (1997) assessed a small group of children on their perception of a variety of psychosocial issues, finding that they on average were happy with their level of self-esteem.	4	CT	CT	CT	CT	N
Journal of Adolescent Health Care	Seigel W.M., Golden N.H., Gough J.W., Lashley M.S., Sacker I.M.	Depression, self-esteem, and life events in adolescents with chronic diseases	1990	Seigel et al (1990) looked at depression, self-esteem and life events in adolescents, of which a subgroup had SCD. They compared different chronic illnesses as well as a control group. The participants with chronic illnesses had higher depression and lower self-esteem than those without, and those with SCD did not differ from those with other chronic illnesses.	3	Y	Y	Y	Y	Y
Journal of Pediatric Hematology/Oncology	Goldstein-Leever A., Peugh J.L., Quinn C.T., Crosby L.E.	Disease Self-Efficacy and Health-Related Quality of Life in Adolescents with Sickle Cell Disease	2020	Goldstein-Leever et al (2020) studied the self-efficacy, severity of illness and quality of life of people with SCD between the ages of 13 and 22. In the multiple regression they completed, both severity of SCD and self-efficacy were significant independent predictors of quality of life, whilst self-efficacy was also negatively related with disease severity. From this, they suggested that self-efficacy not only helps prevent complications, but also affects the way one views one's quality of life even when the illness is particularly severe (or indeed, for those with lower self-efficacy, when it is comparatively mild).	4	Y	CT	Y	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
Child Neuropsychology	Clay O.J., Telfair J.	Evaluation of a disease-specific self-efficacy instrument in adolescents with sickle cell disease and its relationship to adjustment	2007	Clay & Telfair (2007), as well as evaluating the psychometric properties of the SCSES for adolescents, took the opportunity to assess the relationship between self-efficacy, self-care behaviours and physical and psychological symptoms. They found significant correlations between self-efficacy and ensuring they drank enough fluids, and inversely between self-efficacy and psychological and physical symptoms.	4	Y	Y	Y	Y	Y
Journal of Pediatric Psychology	Burlew K., Telfair J., Colangelo L., Wright E.C.	Factors that influence adolescent adaptation to sickle cell disease	2000	Burlew et al (2000) measured a number of factors in adolescents with SCD, including their self-esteem as part of psychosocial measures. They compared the psychosocial measures with biomedical measures in predicting the adjustment of the adolescents in multiple regression models, finding that the biomedical measures did not significantly predict adjustment, but a variety of the psychosocial measures did, including self-esteem.	4	Y	Y	Y	Y	Y
Iranian Journal of Blood and Cancer	Ahmadi M., Rohani C., Beiranvand S., Matbooei M., Poormansouri S.	Health-related quality of life and predictive role of self-efficacy in Iranian patients with sickle cell disease	2018	Ahmadi et al (2018) appraised the self-efficacy and quality of life of patients with SCD in Iran. They also assessed the physical complications that the participants had experienced due to SCD. Using multiple regression, they found that both physical and psychological elements of quality of life were significantly predicted by self-efficacy, and that it was one of the most important contributors to variance in quality of life ratings. The other important contributors were complications that reflected the severity of the disease, or the degree of need for blood transfusion.	4	Y	CT	Y	CT	Y
British Journal of Haematology	Panepinto J.A., O'Mahar K.M., DeBaun M.R., Loberiza F.R., Scott J.P.	Health-related quality of life in children with sickle cell disease: Child and parent perception	2005	Panepinto et al (2005) compared parental and child ratings of quality of life for children with SCD. Their measure of quality of life included physical, social and emotional elements, of which one was self-esteem. They also compared the scores with established norms for the measures based on healthy children. The children's self-ratings were significantly lower than these norms for physical function, pain, general health and being limited by their physical condition. Parental and child ratings of self-esteem, amongst other factors, were significantly different, although moderately correlated, with parents reporting lower self-esteem for the children than the children rated themselves.	4	Y	CT	Y	CT	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
Journal of Clinical Psychology	Thompson Jr. R.J., Gustafson K.E., Gil K.M., Godfrey J., Bennett Murphy L.M.	Illness specific patterns of psychological adjustment and cognitive adaptational processes in children with cystic fibrosis and sickle cell disease	1998	Thompson et al (1998) compared children with SCD with those with cystic fibrosis on their mental health, as well as measuring self-esteem, self-efficacy and perceived stress level. They found that both groups had a significant proportion of anxiety diagnoses, whereas those with sickle cell had lower degrees of oppositional or attentional difficulties compared to those with cystic fibrosis. They completed a multiple regression, showing that for children with SCD, mental health symptoms were predicted by higher stress and lower self-esteem.	3	CT	Y	Y	Y	Y
Pain Medicine (United States)	Cronin R.M., Dorner T.L., Utrankar A., Allen W., Rodeghier M., Kassim A.A., Jackson G.P., Debaun M.R.	Increased Patient Activation Is Associated with Fewer Emergency Room Visits and Hospitalizations for Pain in Adults with Sickle Cell Disease	2019	Cronin et al (2019) explored the relationship between self-efficacy and a related concept – activation. They suggested that activation was wider than self-efficacy because it also incorporated knowledge and skills, as well as confidence, which is perhaps more overlapping with self-efficacy. They also collected data on both regular and emergency healthcare usage. They found significant positive correlations between activation and self-efficacy, and negative correlations between activation and number of hospital visits. The only outliers, with high activation but also high healthcare usage had additional diagnoses of substance use problems – the authors suggested that seeking opioids was the likely reason for this.	4	Y	CT	Y	CT	Y
Pediatric Blood and Cancer	Bhatt-Poulose K., James K., Reid M., Harrison A., Asnani M.	Increased rates of body dissatisfaction, depressive symptoms, and suicide attempts in Jamaican teens with sickle cell disease	2016	Bhatt-Poulose et al (2016) compared adolescents with SCD and those without on their body image, perception of their weight and depressive symptoms (including past suicidal attempts). They also recorded the actual weight and height of the participants. The measures of body image and weight assessed whether a person's body shape was the same between desired and perceived, and whether their self-report of being underweight, healthy weight or overweight correctly corresponded to their BMI. Adolescents with SCD had significantly poorer body satisfaction. Young women with SCD wanted to be thinner, and young men with SCD wanted to be larger. Adolescents with SCD also were significantly more inaccurate in estimating their weight, and scored higher on depressive symptoms. Using logistic regression, they found that the risk for depressive symptoms was higher when a participant perceived themselves to be overweight. They also found that those with negative body image satisfaction were at higher risk of having	3	Y	Y	Y	Y	CT

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
				previously attempted suicide. They suggest that these findings challenge Cepeda et al (2000) especially given the larger sample size. Additionally, they note that young men with other chronic illnesses have also been found to want to be larger, and that this may lead to risky behaviours.						
Ethnicity and Health	Royal C.D., Jonassaint C.R., Jonassaint J.C., De Castro L.M.	Living with sickle cell disease: Traversing 'race' and identity	2011	Royal et al (2011) investigated the place of race in one's identity for people with SCD. They used measures of racial identity that looked at its centrality in people's lives, their ideological orientation towards it and their pride in their own racial identity. They also asked about the person's experience of the intersection of race and sickle cell, and gave space for open responses. The participants were principally African-American or Latinx-American. In general, participants had a positive view of their own race, viewed their race as similar to other races in the US as well as the world in general. Those participants whose parents were born outside the US had a greater sense of their race being an oppressed minority. Participants endorsed the view that they were treated differently because of SCD, and that their race influenced this – the more centrally they held their racial identity, and the more they expressed the uniqueness and separateness of their racial identity, the more they endorsed this. In the qualitative findings, themes constructed included that people lack knowledge about SCD, that people with SCD are treated as being disabled, that SCD is linked to race and drug addiction (especially in how people are viewed by others), and that being Black worsens the difficulty of having SCD. Some participants contended that race was not a factor in their experience of SCD.	5 (4, 1)	Y, Y, Y	Y, CT, CT	Y, Y, CT	Y, CT, Y	CT, Y, Y
International Journal of Adolescent Medicine and Health	Anie K.A., Telfair J.	Multi-site study of transition in adolescents with sickle cell disease in the United Kingdom and the United States	2005	Anie & Telfair (2005) assessed a range of factors, both physical and psychological, amongst adolescents approaching transition to adult care. These included self-efficacy, again measured using the scale from Edwards et al (2001). They used multiple regression, which showed that self-efficacy was predicted by physical symptoms rather than other factors measured.	4	Y	Y	Y	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
Applied Nursing Research	Dobson C.	Outcome results of self-efficacy in children with sickle disease pain who were trained to use guided imagery	2015	Dobson (2015) provided children with training in guided imagery to manage pain, and after this intervention assessed their pain patterns, skill at guided imagery and self-efficacy (using the SCSES). Following the training, the children's self-efficacy scores had significantly increased. Dobson therefore suggested that having a tool for managing pain could have a significant impact on self-efficacy for people with SCD, in line with laboratory-based artificially induced pain studies.	3	CT	Y	Y	N	CT
Journal of the National Medical Association	Cepeda M.L., Allen F.H., Cepeda N.J., Yang Y.-M.	Physical growth, sexual maturation, body image and sickle cell disease	2000	Cepeda et al (2000) assessed children and adolescents, comparing those with SCD with matched controls on physical developmental characteristics. They assessed their self-concept as well as body image, which the measure they used conceptualised as satisfaction with particularly body parts or body functioning. The control group was taller, heavier and further through sexual development. However, there were no differences in ratings of body image or of self-concept, even in post-hoc analysis of subscales.	3	Y	Y	Y	Y	CT
British Journal of Clinical Psychology	Midence K., McManus C., Fuggle P., Davies S.	Psychological adjustment and family functioning in a group of British children with sickle cell disease: Preliminary empirical findings and a meta-analysis	1996	Midence et al (1996) assessed children with SCD on a variety of areas including self-esteem, compared to controls. They found there were no significant differences between the groups on self-esteem. They completed multiple regression on some of the factors studied, but did not report any correlations regarding self-esteem.	3	CT	Y	Y	N	N
Journal of Behavioral Medicine	Gold J.I., Mahrer N.E., Treadwell M., Weissman L., Vichinsky E.	Psychosocial and behavioral outcomes in children with sickle cell disease and their healthy siblings	2008	Gold et al (2008) administered questionnaires measuring self-efficacy (focusing on social interaction), social support and coping to children with SCD and their siblings, finding no significant differences between them, and that the scores all lay in the normative range – in other words, that there were no differences between children with SCD, healthy siblings, and children in families without chronic illness.	3	CT	Y	Y	N	CT
J Am Acad Child Adolesc Psychiatry.	Brown RT, Kaslow NJ, Doepke K, Buchanan I, Eckman J, Baldwin K, Goonan B.	Psychosocial and family functioning in children with sickle cell syndrome and their mothers.	1993	Brown et al (1993b) compared children with SCD to their siblings who did not have the disease. Amongst a range of other measures looking at coping and attributional style, completed by parents and teachers as well as the children, they had the children complete a measure of self-perception focused on self-esteem. The overall measure did not show differences between those	3	CT	Y	N	CT	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
				with SCD and their siblings, but subsequent post-hoc analysis of the subscales showed that those with SCD scored significantly lower on acceptance by peers and on school ability.						
Pain Management Nursing	Matthie N., Jenerette C., McMillan S.	Role of Self-Care in Sickle Cell Disease	2015	Matthie et al (2015) studied young adults with SCD on a similar range of factors to Jenerette & Murdaugh (2008) that they expected to influence self-care. They again found that self-efficacy was a significant predictor of self-care. However, they did not find the expected relationship between self-care and hospital visits – the only factor that influenced this was having a lower income.	4	Y	CT	Y	Y	Y
Indian Journal of Public Health Research and Development	Khudhair S.H.	Self – Efficacy of sickle-cell anemic adult patients at bloodisease wards in baghdad teaching hospitals	2019	Khudhair (2019) conducted research into the self-efficacy of patients with SCD in Iraq. Their scale of self-efficacy looked both at management of the condition but also at various life activities, both day-to-day and longer term. They contrasted self-efficacy in different demographic groups, finding that men, younger people, those with less education, and those who were single, each had higher self-efficacy. They did not, however, hypothesise as to why this might be.	4	CT	CT	Y	CT	N
Revista brasileira de enfermagem	Costa D.O., Araújo F.A., Xavier A.S.G., Araújo L.D.S., Silva U.B.D., Santos E.A., Ferreira S.L.	Self-care of men with priapism and sickle cell disease	2018	Costa et al (2018) used qualitative methods to explore how men with SCD who suffered from priapism conducted self-care. They completed this in an area of Brazil with a significant number of people of African descent, and hence also a significant incidence of SCD. Their qualitative analysis was one informed by a pre-existing model of self-care, which emphasised that self-care incorporates universal demands, developmental demands and health deviations. Universal demands are the basic needs to be met by any person regardless of health status, developmental demands are those required for self-care of a condition, and health deviations are the ways in which a condition interferes with self-care. They found that the men struggled with loneliness, self-image especially related to masculinity and embarrassment, and sexual difficulties. Their self-care was impacted by a lack of knowledge of the best approaches, as well as shame and stigmatisation. The main health deviation found was that of significant pain. The researchers suggested that the self-esteem impact	1	Y	Y	CT	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
				around fragility as opposed to masculinity was especially acute in the Brazilian context.		Y	Y	Y	Y	Y
International Journal of Adolescent Medicine and Health	Treadwell M., Johnson S., Sisler I., Bitsko M., Gildengorin G., Medina R., Barreda F., Major K., Telfair J., Smith W.R.	Self-efficacy and readiness for transition from pediatric to adult care in sickle cell disease	2015	Treadwell et al (2015) researched adolescents and adults on the topic of transition to adult care for SCD. They measured self-efficacy, stress and a scale that measured various aspects of readiness for transition. They found that self-efficacy significantly predicted the overall readiness for transition and the subscales related to education and independence in living. They also found a large correlation between stress and self-efficacy – they argued that this could be due to the need to increase one's own capacity to manage stressors – though there would be equally plausible explanations for the reverse finding, that increased stress reduced one's self-efficacy.	4	Y	CT	Y	CT	Y
Psychosomatic Medicine	Edwards R., Telfair J., Cecil H., Lenoci J.	Self-efficacy as a predictor of adult adjustment to sickle cell disease: One-year outcomes	2001	Edwards et al (2001) completed a cohort study comparing a baseline with a one year follow-up of adults with SCD. They used a measure of self-efficacy specific to coping with sickle cell called the SCSES, which they had developed and will be used in a number of subsequent studies in the review. They then surveyed the pain, physical and mental health symptoms and healthcare utilisation of the participants. They found that the scores on the measures were consistent across the time points. They found through correlation that self-efficacy predicted all of the outcome measures at baseline, and all but healthcare usage at follow-up, and also that self-efficacy at baseline and changes in self-efficacy predicted changes in the outcome measures. This was of the pattern that high or increased self-efficacy predicted reduced pain, physical symptoms or mental health symptoms.	3	CT	Y	Y	Y	Y
Cognitive Therapy and Research	Wilson J.J.	Self-evaluation, coping, and depressive affect in African American adults with sickle cell disease	1997	Wilson et al (1997) had adults identify the extent to which they used upward comparison – seeing some people as better-off than them and downward comparison – seeing themselves as being better-off than some people. They also had these adults rate their level of depression. They then used regression analysis and found associations between the self-evaluation approaches and depression, even after controlling for pain, disease severity and demographic features. Those	4	Y	CT	Y	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
				who used more upward comparison showed more depression, and those who used more downward comparison showed less depression; the former was the stronger effect.						
Journal of National Black Nurses' Association : JNBNA	Adegbola M.	Sleep Quality, Pain and Self-Efficacy among Community-Dwelling Adults with Sickle Cell Disease	2015	Adegbola (2015) asked adults with SCD to report their sleep quality and their amount of pain, as well as complete the SCSES to assess self-efficacy. They completed a multiple regression model, finding a significant positive correlation between self-efficacy and sleep quality, a significant negative correlation between pain and sleep quality, and finally a significant negative correlation between self-efficacy and pain. Whilst causal relationships cannot be determined, it can be hypothesised that self-efficacy is increased by having less pain and better sleep, but also could facilitate these things, perhaps through its influence of self-care behaviours.	4	Y	CT	Y	N	Y
Child psychiatry and human development	Simon K., Barakat L.P., Patterson C.A., Dampier C.	Symptoms of depression and anxiety in adolescents with sickle cell disease: the role of intrapersonal characteristics and stress processing variables.	2009	Simon et al (2009) assessed adolescents with SCD and their siblings who did not have the condition, having them complete measures of coping style, hope, attribution style, and a measure of self-esteem and personal inadequacy. They also measured symptoms of anxiety and depression, aiming to use the previous measures as predictors of these symptoms. They found that self-esteem and inadequacy were predictors of depression and anxiety for the teenagers with SCD, though their scores on anxiety and depression did not differ significantly from the normative scores for the measure, nor from their siblings. They completed multiple regressions to assess for mediation, but could not find any models where mediation was supported. They suggested that further research may be needed to find the ways in which self-esteem and inadequacy led to psychological symptoms in those with SCD.	3	Y	Y	Y	Y	CT
Research in Nursing and Health	Jenerette C.M., Murdaugh C.	Testing the theory of self-care management for sickle cell disease	2008	Jenerette & Murdaugh (2008) measured a range of variables which they theorised related to the ability to self-care of adults with SCD. This included self-efficacy, again using the SCSES. They then used structural equation modelling – and the results of the model suggested that self-care management resources, including self-efficacy, social support, assertiveness and self-care ability, each had a significant impact on health	4	Y	CT	Y	Y	Y

Journal	Authors	Title	Year	Summary	MMAT Type and Question Responses					
				outcomes and quality of life, but did not mediate the relationship between the number of crises and complications and those outcomes.						
Journal of Family Nursing	Jenerette C.M., Valrie C.R.	The influence of maternal behaviors during childhood on self-efficacy in individuals with sickle cell disease	2010	Jenerette & Valrie (2010), following on from the previous study, assessed adults with SCD on their level of self-efficacy using the SCSES as well as having the adults rate their view of their mothers' behaviour towards them in childhood. The ratings of the mothers' behaviour focused on being either caring or over-protective. A multiple regression model showed that mothers being (perceived as) caring significantly predicted self-efficacy. They suggest that caring behaviours from parents may engender more general self-efficacy and emotional development, which may in turn facilitate illness-specific self-efficacy.	4	Y	N	Y	N	Y
International Journal of Qualitative Studies on Health and Well-being	Forrester A.B., Barton-Gooden A., Pitter C., Lindo J.L.M.	The lived experiences of adolescents with sickle cell disease in Kingston, Jamaica	2015	Forrester et al (2015) completed interviews with Jamaican adolescents, conducting a qualitative analysis with a particular interest in the changes in their development due to sickle cell and how this in turn impacted their identity. As a group, the researchers worked together to develop themes. One theme was that of positive self-concept, as well as feeling that they were living a normal life, that they had ways of coping with SCD (especially spiritual means), and that they did what they needed in order to manage their health. By contrast, other themes included having a strong fear of death, decreased physical activity, avoiding sharing that they had SCD and having an overprotective family. The researchers therefore suggested that the self-concept of the participants had some ambivalence, even if it leaned to the positive.	1	Y	Y	Y	Y	Y

Appendix 2

<u>Interview schedule</u>

Tell me a bit about your day.

We'd like to know about how you feel about your body?

What do you think has been important in making you feel that way?

Do you think that SCD has played a role? Could you tell me how?

Are there particular ways in which SCD has affected your body physically? Which do you think has been the most important?

What kind of impact does the way you feel about your body have on the way you feel in general?

Can you talk me through how that then affects your life?

Is there anything else that has been on your mind as we have talked, that you would like to be heard?

Appendix 3

Confirmation of ethical approval

University of Hertfordshire UH

HEALTH, SCIENCE, ENGINEERING AND TECHNOLOGY ECDA

ETHICS APPROVAL NOTIFICATION

TO	Timothy Swann-Roberts
CC	Dr Wendy Solomons
FROM	Dr Simon Trainis, Health, Science, Engineering & Technology ECDA Chair.
DATE	06/02/2020

Protocol number: **LMS/PGR/UH/04079**

Title of study: How does Sickle Cell Disease affect body image in the people who have it?

Your application for ethics approval has been accepted and approved with the following conditions by the ECDA for your School and includes work undertaken for this study by the named additional workers below:

Dr Nicholas Troop

General conditions of approval:

Ethics approval has been granted subject to the standard conditions below:

Permissions: Any necessary permissions for the use of premises/location and accessing participants for your study must be obtained in writing prior to any data collection commencing. Failure to obtain adequate permissions may be considered a breach of this protocol.

External communications: Ensure you quote the UH protocol number and the name of the approving Committee on all paperwork, including recruitment advertisements/online requests, for this study.

Invasive procedures: If your research involves invasive procedures you are required to complete and submit an EC7 Protocol Monitoring Form, and copies of your completed consent paperwork to this ECDA once your study is complete.

Submission: Students must include this Approval Notification with their submission.

Validity:

This approval is valid:

From: 06/02/2020

To: 30/09/2020

University of Hertfordshire UH

HEALTH, SCIENCE, ENGINEERING AND TECHNOLOGY ECDA

ETHICS APPROVAL NOTIFICATION

TO	Timothy Swann-Roberts
CC	Dr Wendy Solomons
FROM	Dr Simon Trainis, Health, Science, Engineering & Technology ECDA Chair
DATE	05/03/2020

Protocol number: **aLMS/PGR/UH/04079(1)**

Title of study: How does Sickle Cell Disease affect body image in the people who have it?

Your application to modify and extend the existing protocol as detailed below has been accepted and approved by the ECDA for your School and includes work undertaken for this study by the named additional workers below:

Dr Nick Troop

Modification: Detailed in EC2

General conditions of approval:

Ethics approval has been granted subject to the standard conditions below:

Original protocol: Any conditions relating to the original protocol approval remain and must be complied with.

Permissions: Any necessary permissions for the use of premises/location and accessing participants for your study must be obtained in writing prior to any data collection commencing. Failure to obtain adequate permissions may be considered a breach of this protocol.

External communications: Ensure you quote the UH protocol number and the name of the approving Committee on all paperwork, including recruitment advertisements/online requests, for this study.

Invasive procedures: If your research involves invasive procedures you are required to complete and submit an EC7 Protocol Monitoring Form, and copies of your completed consent paperwork to this ECDA once your study is complete.

Submission: Students must include this Approval Notification with their submission.

Validity:

This approval is valid:

From: 05/03/2020

To: 30/09/2020

Please note:

Failure to comply with the conditions of approval will be considered a breach of protocol and may result in disciplinary action which could include academic penalties.
Additional documentation requested as a condition of this approval protocol may be submitted via your supervisor to the Ethics Clerks as it becomes available. All documentation relating to this study, including the information/documents noted in the conditions above, must be available for your supervisor at the time of submitting your work so that they are able to confirm that you have complied with this protocol.

Should you amend any aspect of your research or wish to apply for an extension to your study you will need your supervisor's approval (if you are a student) and must complete and submit a further EC2 request.
Approval applies specifically to the research study/methodology and timings as detailed in your Form EC1A or as detailed in the EC2 request. In cases where the amendments to the original study are deemed to be substantial, a new Form EC1A may need to be completed prior to the study being undertaken.

Failure to report adverse circumstance/s may be considered misconduct.
Should adverse circumstances arise during this study such as physical reaction/harm, mental/emotional harm, intrusion of privacy or breach of confidentiality this must be reported to the approving Committee immediately.

HEALTH, SCIENCE, ENGINEERING AND TECHNOLOGY ECDA

ETHICS APPROVAL NOTIFICATION

TO	Timothy Swann-Roberts
CC	Dr Wendy Solomons
FROM	Dr Rosemary Godbold, Health, Science, Engineering and Technology ECDA Vice-Chair
DATE	08/07/2020

Protocol number: **aLMS/PGR/UH/04079(2)**

Title of study: How does Sickle Cell Disease affect body image in the people who have it?

Your application to modify and extend the existing protocol as detailed below has been accepted and approved by the ECDA for your School and includes work undertaken for this study by the named additional workers below:

Dr Nick Troop

Modification: Amendment to a different data source for an existing ethics approval – See EC2 for further details

General conditions of approval:

Ethics approval has been granted subject to the standard conditions below:

Original protocol: Any conditions relating to the original protocol approval remain and must be complied with.

Permissions: Any necessary permissions for the use of premises/location and accessing participants for your study must be obtained in writing prior to any data collection commencing. Failure to obtain adequate permissions may be considered a breach of this protocol.

External communications: Ensure you quote the UH protocol number and the name of the approving Committee on all paperwork, including recruitment advertisements/online requests, for this study.

Invasive procedures: If your research involves invasive procedures you are required to complete and submit an EC7 Protocol Monitoring Form, and copies of your completed consent paperwork to this ECDA once your study is complete.

Submission: Students must include this Approval Notification with their submission.

Validity:

This approval is valid:

From: 08/07/2020

To: 30/09/2020

Please note:

Failure to comply with the conditions of approval will be considered a breach of protocol and may result in disciplinary action which could include academic penalties.
Additional documentation requested as a condition of this approval protocol may be submitted via your supervisor to the Ethics Clerks as it becomes available. All documentation relating to this study, including the information/documents noted in the conditions above, must be available for your supervisor at the time of submitting your work so that they are able to confirm that you have complied with this protocol.

Should you amend any aspect of your research or wish to apply for an extension to your study you will need your supervisor's approval (if you are a student) and must complete and submit a further EC2 request.
Approval applies specifically to the research study/methodology and timings as detailed in your Form EC1A or as detailed in the EC2 request. In cases where the amendments to the original study are deemed to be substantial, a new Form EC1A may need to be completed prior to the study being undertaken.

Failure to report adverse circumstance/s may be considered misconduct.
Should adverse circumstances arise during this study such as physical reaction/harm, mental/emotional harm, intrusion of privacy or breach of confidentiality this must be reported to the approving Committee immediately.

Appendix 4

Examples of initial codes

The body external to the self

The body holding power

The body to be listened to

Guided to do less physical things

Unpredictability

Relationship to Sickle Cell – who's in charge

Attending to the body

Being afraid of or for the body

The body as or at extremes

Having to use the body when it seems impossible

The body or the self as weak

The body or blood or cells as enemy or violent

The body as a non human or inorganic object

Direct physical impact of sickle cell

Reaching the lowest point or losing the battle

Bad Blood

Needing to keep quiet or hide something about the body

Visibility of the body or blood could invite more pain

Is this a state or something you own

Body woes to be hidden or only entrusted to a few

Impossibility of desired invisibility

Betrayed by the body

The blackness of the body and the perceived blackness of the body

Feeling targeted by the disease

The body constraining and defining one's blackness

Contrast of needed perfection with bodily imperfection

Body as prison, disease as death sentence

The body holding limited time

Passing the expiration date of the body

Appendix 5

Examples of code relationships developed in the code organisation phase

From Name	From F	Type	To Name	To Fold	Direction
Coding\Altitude as a trigger	Nodes	Associated	Coding\Role of temperature	Nodes	—————
Coding\Bad Blood	Nodes	Opposite	Coding\Good blood	Nodes	◄———►
Coding\Body as prison, disease as death sentence	Nodes	Opposite	Coding\Defying the deadline given by doctors	Nodes	◄———►
Coding\Body as prison, disease as death sentence	Nodes	Associated	Coding\The body holding limited time	Nodes	—————
Coding\Bad Blood	Nodes	Associated	Coding\Blood count suddenly changing	Nodes	—————
Coding\Blood count suddenly changing	Nodes	Associated	Coding\Enjoying when the blood count is higher	Nodes	—————
Coding\Enjoying when the blood count is higher	Nodes	Associated	Coding\The value and positive effect of transfusion	Nodes	—————
Coding\The value and positive effect of transfusion	Nodes	Associated	Coding\Imagining the nature of the transfused blood	Nodes	—————
Coding\Imagining the nature of the transfused blood	Nodes	Associated	Coding\Seeing transfused blood as one's own	Nodes	—————
Coding\Inability of others to reconcile the sickness and the wellness - the chan	Nodes	Associated	Coding\Unpredictability	Nodes	—————
Coding\Good blood	Nodes	Associated	Coding\Imagining the nature of the transfused blood	Nodes	—————
Coding\Appearing not to have an illness or disability	Nodes	Associated	Coding\Being seen as bodily the same to others without SCD	Nodes	—————
Coding\Appearing not to have an illness or disability	Nodes	Consequential	Coding\Trying to act normal only ends up in brokenness	Nodes	———►
Coding\Appearing not to have an illness or disability	Nodes	Associated	Coding\Being bigger than expected in build	Nodes	—————
Coding\Appearing not to have an illness or disability	Nodes	Opposite	Coding\Fragility and sickness as vulnerability to others	Nodes	◄———►
Coding\Fragility and sickness as vulnerability to others	Nodes	Associated	Coding\Visibility of the body or blood could invite more pain	Nodes	—————
Coding\What makes the pain inside the body visible to others especially those	Nodes	Opposite	Coding\Appearing not to have an illness or disability	Nodes	◄———►
Coding\Appearing not to have an illness or disability	Nodes	Consequential	Coding\Body woes to be hidden or only entrusted to a few	Nodes	———►
Coding\Being small and weak as a child	Nodes	Associated	Coding\Being able to distinguish a baby's crisis from other crying	Nodes	—————
Coding\Being small and weak as a child	Nodes	Associated	Coding\Early health vulnerabilities coming from SCD	Nodes	—————
Coding\Being small and weak as a child	Nodes	Associated	Coding\Facing multiple hospitalisations at a young age	Nodes	—————
Coding\Facing multiple hospitalisations at a young age	Nodes	Associated	Coding\Knowing what the body has already gone through	Nodes	—————
Coding\Knowing what the body has already gone through	Nodes	Associated	Coding\Life improving over time	Nodes	—————
Coding\Knowing what the body has already gone through	Nodes	Associated	Coding\Memories of the illness	Nodes	—————
Coding\Life improving over time	Nodes	Associated	Coding\Being less affected as one gets older	Nodes	—————
Coding\Being rejected because of the diagnosis	Nodes	Associated	Coding\Being seen as dangerous or unpredictable	Nodes	—————
Coding\Being rejected because of the diagnosis	Nodes	Associated	Coding\Dissociation from family identity becaues of the illness, assumed reflection on the parent	Nodes	—————
Coding\Braving it all and coming out the victor	Nodes	Associated	Coding\Carrying on with warrior determination	Nodes	—————
Coding\Appreciating or enjoying recovery	Nodes	Associated	Coding\Appreciating the relief	Nodes	—————
Coding\Adventuring together	Nodes	Opposite	Coding\Experience differing between people with SCD	Nodes	◄———►
Coding\Adventuring together	Nodes	Associated	Coding\Learning from peers	Nodes	—————
Coding\Diagnosed very young	Nodes	Consequential	Coding\Feeling that SCD is a narrative that encompasses one's life from the beginning	Nodes	———►
Coding\Permanent or temporary, diseased or sick	Nodes	Associated	Coding\Feeling that SCD is a narrative that encompasses one's life from the beginning	Nodes	—————
Coding\Anticipation of a complication being the biggest fear	Nodes	Associated	Coding\Being worn down by SCD	Nodes	—————
Coding\Being worn down by SCD	Nodes	Associated	Coding\Big problems starting with something small	Nodes	—————

Appendix 6

Example of reflective writing made during analysis phase:

I think I am realising what gives me the reaction to the spiritual content that I am finding – that it is all the Evangelical / Pentecostal talk of struggle and triumph, but not lament. Not negotiation. There's David but no Job. There's Third Isaiah, not Lamentations. It's Friday-Sunday, not Saturday.

And whilst I believe that every tear will be wiped from every eye – in the now, I believe in the right to our sorrow, and the right to our anger. Whilst the sincere faith in the face of something that could easily be seen as a divine cruelty is admirable, and hopefully not in a patronising way, it smacks (and there was even a code for this) of the lack of space for real pessimism, real distress, in a way that has to be denied to the self. And maybe that is essential – one person wrote about how depression made the sickle cell worse (got a code for that too somewhere) – but it still feels jarring somehow.

Example of more poetic memoing in relation to the data:

It's got to have a reason, it's got to have a purpose

How could it be borne if it didn't have a reason?

If I couldn't say – this has forged me, this has made me, and better than if I were without it?

Then the only thing left would be the burden,

The only part remaining would be loss, restriction,

the world of pain.

The same fire that purifies, burns down,

And we want to believe that

We were impure metal

Else

We've simply been burnt alive.

Appendix 7

Ascertaining Quality

Question	Response
1. Do the authors explain why they are using TA, even if only briefly?	Yes, see Section 3.2
2. Do the authors clearly specify and justify which type of TA they are using?	Yes, see Section 3.2
3. Is the use and justification of the specific type of TA consistent with the research questions or aims?	Yes, see Section 3.2
4. Is there a good 'fit' between the theoretical and conceptual underpinnings of the research and the specific type of TA (i.e. is there conceptual coherence)?	Yes, see Sections 1.2 and 3.2
5. Is there a good 'fit' between the methods of data collection and the specific type of TA?	Yes – there do not seem to be any barriers to the form of data collection being used in RTA – see Section 3.5
6. Is the specified type of TA consistently enacted throughout the paper?	Yes, see Sections 3.2, 3.5, 3.6, 4.1 and 4.2
7. Is there evidence of problematic assumptions about, and practices around, TA?	No – the research does not treat Thematic Analysis as a homogeneous or unified approach, has ensured compatibility of philosophy and procedure, has not confused themes with data summaries, has not imported grounded theory procedures, has not assumed that Thematic Analysis is realist or atheoretical, and has not assumed that additional methods are needed.
8. Are any supplementary procedures or methods justified, and necessary, or could the same results have been achieved simply by using TA more effectively?	Supplementary methods are not used, as were not justified.
9. Are the theoretical underpinnings of the use of TA clearly specified (e.g. ontological, epistemological assumptions, guiding theoretical framework(s)), even when using TA	Yes, see Section 3.6.1

inductively (inductive TA does not equate to analysis in a theoretical vacuum)?	
10. Do the researchers strive to 'own their perspectives' (even if only very briefly), their personal and social standpoint and positioning? (This is especially important when the researchers are engaged in social justice-oriented research and when representing the 'voices' of marginal and vulnerable groups, and groups to which the researcher does not belong.)	Yes, see Sections 1.2.2, 3.3.2, 2.4.56 and 5.4
11. Are the analytic procedures used clearly outlined, and described in terms of what the authors actually did, rather than generic procedures?	Yes, see Section 3.6.2
12. Is there evidence of conceptual and procedural confusion?	No – see Sections 3.2 and 3.6
13. Do the authors demonstrate full and coherent understanding of their claimed approach to TA?	As best possible – see Sections 3.2 and 3.6
14. Is it clear what and where the themes are in the report? Would the manuscript benefit from some kind of overview of the analysis: listing of themes, narrative overview, table of themes, thematic map?	Yes – see Sections 4.1, 4.2 and 5.1
15. Are the reported themes topic summaries, rather than 'fully realised themes' – patterns of shared meaning underpinned by a central organising concept?	No – see Section 4.1
16. Is non-thematic contextualising information presented as a theme?	No – see Section 4.1
17. In applied research, do the reported themes have the potential to give rise to actionable outcomes?	Yes – see Section 5.5
18. Are there conceptual clashes and confusion in the paper?	No – see Section 3.5.1 and 5.3
19. Is there evidence of weak or unconvincing analysis?	No – the number of themes follows on from the analysis and how the collected data can answer the research question – choices about this are described and justified in Section 3.5 and 3.6

	Two theme levels appears to be a suitable number of theme levels. Analytic claims – through discussion with research supervisors, Number of data extracts – desire to reflect the amount of data and variety of voices Theme overlap – the Themes and Subthemes are related but deal with different areas. See Section 3.6.1 for a discussion of the composition of themes and subthemes and how themes are not just the addition of their subthemes. See Section 4.2 for a description of how the themes might be linked rather than overlap.
20. Do authors make problematic statements about the lack of generalisability of their results, and/or implicitly conceptualise generalisability as statistical probabilistic generalisability?	No – see Section 5.3, which uses qualitative framings of the strengths and limitations of the project.