

Portfolio Volume 1:

**Family Narratives of Lives Where Both Parent and Child Have Been
Diagnosed with Persistent Physical Symptom (PPS) Conditions.**

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ABSTRACT

There is a limited body of research that explores the lived experiences of families with persistent physical symptoms (PPS) conditions and no qualitative research on the intergenerational component to PPS. Existing research focusing on PPS conditions is often researched from several different, individualised perspectives, such as; the person with PPS (adult or child), their GP or healthcare professional (HCP) or the parents of the child with PPS. Additionally, much of the intervention or treatment research about PPS is based on cognitive behavioural therapy (CBT) or psychodynamic approaches. To the author's knowledge, there is no current research on exploring the storied lives of a family where there are two generations of PPS. The current study aimed to think about the unique life events of a family with multiple generations of PPS from a relational/systemic perspective.

This study employed a qualitative study design, specifically using narrative methodologies to explore the lived experiences of a single family comprising two parents and their three children aged 17, 15 and 12 years old. All the children and their mother have a diagnosis of Ehlers-Danlos Syndrome (EDS) but are specifically afflicted with PPS. The father is in good health. Using narrative inquiry, the family members were interviewed together and then individually. The interviews were audio-recorded, transcribed and analysed following narrative analytic methodologies.

The findings initially presented the storylines of each of the individuals and from the group interview. The main overarching narratives were stories of loss and sacrifice, stories of family unity and a story of collective versus individualistic language. Lastly, the family's negotiation of roles and identities is explored in the context of stigmatised illness. The findings were then reviewed in context. Clinical implications and future research ideas are discussed.

1. INTRODUCTION

1.1 Overview

To take the audience on a journey, I will firstly explain how the research went from inception to production. This chapter explores the researcher's epistemological position and defines key terms and any specialised language used throughout. Then, a broad spectrum of the background literature is explored from several different perspectives. Given the project's personal resonance, a first-person perspective will be employed when reflecting on my own position in relation to the research.

1.2 Epistemological Position

Before starting Clinical Psychology training, I am unsure if "epistemology" was even part of my vocabulary. However, learning about knowledge and moving away from "positivist" notions of reality has allowed me to move towards an understanding of a social reality, co-existing and co-constructed through language and the ways we communicate (Berger & Luckman, 1966; Conrad & Barker, 2010). Gergen (2015) explains that through our social relationships we begin to understand and make sense of our world.

Personally, I take more of a "critical realist" position towards the acquisition and understanding of knowledge, loosely defined by understanding that realities exist however, we understand them through language and co-construction. However, this research is aiming to uncover the way people with PPS story their lives and therefore a social constructionist stance will be employed. Qualitative research is grounded in socially constructed methodologies. Constructionist methodologies, specifically narrative methodologies, are interested in how the individual's narrative and understanding about their social world is constructed within a specific socio-cultural, historical and biographical context (Ashworth, 2015; Gergen, 2015; Liamputtong & Ezzy, 2005).

Applying social constructionist ideas to illness can provide more favourable conceptions to people with PPS as it incorporates the experience of the biological body as well as placing the individual within their social context (Williams, 1999). This cluster of illnesses are socially constructed, for example, the whole concept of illness gets constructed through context and a dynamic linguistic negotiation, rather than having explicit biological origin (Sowińska, 2018). There is nothing innately stigmatising with illness, but the context within which it sits may be problematic, for example, if there is a societal discourse of productivity, someone with fatigue or pain may be classed as lazy (Conrad & Barker, 2010). Given the stigmatised nature of these diagnoses ideas of 'epistemic injustice' are brought to mind (Fricker, 2007). Epistemic injustice discusses how from a social perspective, any deviations from more established biomedical explanation of illness are problematic due to the subjective nature of symptoms (Fricker, 2007; Kidd & Carel, 2017).

1.2.1 Personal Significance of the Research.

I arrived at this project through looking for meaning for some of my own experiences. Having grown up with a chronically unwell parent with a plethora of life-limiting physical health problems, I wanted to understand how the roles of the child change with age and the influence these roles have on the identities of the child. I also struggled with my own health as a child; plagued with outpatient appointments, hospital stays and missing out on aspects of my childhood. While I grew out of my health problems, my experiences navigating an unwell parent detail an undulating journey from child, to friend, to carer and back to child, while navigating hope, fear and uncertainty.

I wondered whether these mirrored fluctuations between health and roles resonated with others and the ways they may have understood and shared their stories. I was curious about how people understood their relationship with their family, with their parents, with themselves and their own health. While pursuing this research project, unfortunately my

parent's health took a significant downturn to the point where I felt the personal, emotional impact for me would be too challenging to immerse myself for my Major Research Project (MRP).

In addition to this, this research focused on people who were in a similar situation to me; adult children of a parent with chronic illness. My supervisor challenged me to think about the logistics of recruitment and how I would find "me", given that I was not presenting in mental or physical health services, to share my experiences. Therefore, after some reflection I changed the scope of the project.

Following this, while remaining close to exploring how people's roles and identities change in the context of illness over time, I became interested in stigmatised illness and the impact this has on sufferers and their family members. Through supervision, anecdotal discussions were had about how, in clinical practice, if a parent or child presents with PPS, then it may also be present in another generation too. This began my MRP journey...

1.3 Stigmatised illness, definitions and a comment on language

Medically unexplained symptoms (MUS) is an umbrella term for a cluster of illnesses (such as Chronic Fatigue Syndrome [CFS], Irritable Bowel Syndrome [IBS], persistent pain, fatigue, fibromyalgia and non-epileptic seizures [NES]) commonly used in healthcare settings (Chalder et al. 2019). These illnesses are stigmatised, misunderstood and lack a biological aetiology (Hart, 2014). MUS and similar diagnoses such as Somatic Symptom Disorder (SSD) are found in the Diagnostic and Statistical Manual of Mental Disorders (DSM) fourth and fifth edition, respectively (American Psychiatric Association [APA], 2000; 2013 a & b). The DSM classification of these illnesses cement the relationship between these conditions and mental health problems. To move away from the more stigmatised, mental health perspective of these diagnoses, the label PPS appears to be the preferred term for these conditions, however, this is not a diagnostic term (Chalder et al., 2019; Marks & Hunter,

2014; Picariello et al., 2015). Throughout the literature it appears that the different acronyms are used interchangeably depending on context or author's preference. While these labels prevail in the literature and the medical world, they do not fit with the social constructionist epistemological position of this research. To honour the research's epistemological position, the favoured terminology will be people living with PPS (PLWPPS) when reflecting on an experiential perspective and PPS for the 'diagnostic' label. Language will deviate from this when reporting on literature to remain authentic to the context in which the research was conducted, therefore language like 'patient' or 'MUS' may be used.

1.4 How do we currently understand PPS from a medical perspective?

PPS conditions are often highly stigmatised. They are defined as “persistent physical complaints, such as dizziness or pain, that do not appear to be symptoms of a medical condition...they last for more than a few weeks, but doctors cannot find a problem with the body that may be the cause” (NHS Choices, 2018). Haller et al. (2015) discussed the prevalence rates for these disorders finding that across 32 studies encompassing 24 countries, 40 – 49% of patients presented in primary care with at least one unexplained symptom. It is estimated that 0.2 – 0.4% (approximately 1 in 250 people) of the UK population have been diagnosed with chronic fatigue syndrome (Hunter et al., 2017; National Institute of Health and Care Excellence [NICE], 2007). In addition to this, PLWPPS cost the UK economy more than £14billion a year because of sick days and poorer quality of life (Birmingham et al., 2010; Chew-Graham et al., 2017).

PPS are among the most common reasons for visiting a GP in the UK (Shraim et al., 2013). Despite ensuring that patients know that these conditions are not 'all in their head', they are often left feeling that they are wasting medical professionals' time (Burbaum et al., 2010; Edwards et al., 2010). PLWPPS often undergo numerous tests, with potentially iatrogenic effects, to be left without a diagnosis and feeling as if they are a nuisance to the

professionals (Moulin et al., 2015a). However, although there is no clear medical evidence, research has highlighted the possible role for a genetic contribution to some PPS conditions such as CFS and Ehlers-Danlos Syndrome (EDS) highlighting intergenerational linkage (Albright et al., 2011; Sobey, 2014). EDS is a connective tissue disorder which also blurs the boundaries between medical and misunderstood diagnoses. Similar to PPS conditions there is no diagnostic testing available but Hypermobile EDS, the most common EDS classification, affecting approximately 10% of UK population, has evidence of autosomal dominant heritability (Sobey, 2014).

It appears that these conditions are misunderstood from a medical perspective, perhaps because there is no clear biological aetiology or testing procedure to *prove* the illness validity. It may be beneficial to understand these conditions and the experiences of sufferers from an alternative, psychological perspective.

1.5 How do we currently understand PPS psychologically?

While several different models have sought to understand PPS from a psychological perspective, research shows that not one model provides a sufficient explanation alone (Brown, 2004). Different psychological models have been used to explain PPS including psychodynamic theories, systemic theories and more recently behavioural and cognitive-behavioural approaches.

Psychodynamic approaches emphasise the contribution and importance of the role of attachment in understanding PPS. The impact of parental illness and attachment has been seen in the literature, highlighting parental illness as a risk factor for poor psychosocial functioning in children and adolescents (Armistead et al., 1995; Barkmann et al., 2007; Stoeckel & Weissbrod, 2015). Research has established a relationship between insecure attachment styles, trauma and later adulthood PPS, specifically highlighting for men, trauma and attachment style independently predict later “somatization” (Waldinger et al., 2006). The

psychodynamic model for PPS follows Freudian principles and suggests that the physiological symptomatology represents unresolved internal conflict, perhaps related to trauma or attachment difficulties, displaced into bodily dysfunction (Mobini, 2015; Stone et al., 2005).

From a behavioural perspective, research highlights the relationship between parental modelling of health seeking behaviour and childhood learning experiences through social learning theory (Bandura, 1977). Levy and colleagues (2001) explained how somatic symptoms can be learned through reinforcement and behavioural modelling and explored that for twins with IBS symptoms, social learning has a greater (or equal) contribution as heredity factors. From a cognitive-behavioural perspective, Salkovskis and Warwick (1986) introduce the health anxiety model to understand PPS. This model proposes that the individual's beliefs and assumptions about the meaning of symptoms play an important role in predisposing people to health anxiety and misattribution of physical symptoms. Models of perception highlight the key role that interpretation of symptoms plays when considering how meaning is ascribed to a symptom (Barksy & Wyshak, 1990; Henningsen, 2018).

There are several systemic perspectives that have attempted to understand PPS. Strategic family therapy, developed by Haley (1973; Madanes & Haley, 1977) poses that physical symptoms act as a form of communication that expresses a problem within a family system, suggesting that faulty communication patterns give rise to challenging symptoms (Roy, 1987). Where psychodynamic theories propose that a symptom is a manifestation of an internal conflict, systemic theory poses that the symptom manifests from an external conflict within the family system (Roy, 1987). Structural family therapy explores how families described as "psychosomatogenic families" demonstrated common characteristics such as enmeshment, avoidance of conflict along with overprotection and overinvolvement between family members (Minuchin, 1970; Roy, 1987). The development and maintenance of

symptomology relies on learning theory and how it moves throughout a family system (Roy, 1987). Minuchin et al. (1974a) explore the role of the family environment in the development of PPS. This systemic model underpinning PPS discusses ideas of enmeshment, poor problem-solving skills and a lack of flexibility in coping when difficulty arises within or external to the family system (Husain et al., 2007). When conflict surfaces within a family, often marital discord, the child (or family member) develops symptoms of illness as an unspoken and unintentional attempt to direct attention away from the problem (Roy, 1987). This maintains the status quo, or specifically the 'sick-role homeostasis' of the family as well as avoiding the conflict (Husain et al., 2007; Roy, 1982).

Rolland (1987) examines the impact of intergenerational illness and family life cycles. He delineates the important role of how a family organises itself in the context of illness and how this shapes the family's relationship to illness. From a systemic perspective, patterns relating to health and illness manifest throughout a family's history, and it is often thought that people adopt family scripts and take up roles within their families (Byng-Hall, 1988). A family script is a shared experience about who plays what role in different contexts; these combine into family stories that are iterative and transformative to an individual's identity within a family system and play an important role in the family's comprehension and management of intergenerational illness (Byng-Hall, 1998).

Although the underlying mechanisms for each of these theories differ, across the theoretical perspectives, the importance of childhood experiences and the familial context are noted. As such, it would be interesting to investigate the family perspective and how families experience these difficulties. The next section of this chapter will explore previous PPS research from different perspectives, as well as considering identity formation within PPS and discuss family perspectives on these conditions (Burbaum et al., 2010; Kozłowska et al., 2012).

1.6 Background Literature – Situating the Research

The literature positions itself from multiple perspectives; from the professionals that work with PLWPPS, the adult or parent (with their own PPS or their child's PPS) and the child (with PPS).

1.6.1 Healthcare professional's perspective of PPS.

Doctors often report that they find this group of patients challenging to work with; describing them as 'heartsink patients' (Stone, 2014a). HCPs often state they lack the correct skills, knowledge, training and time to manage the complexities associated with these clients (Furness et al., 2009; Hinton & Kirk, 2016). Once HCPs have ruled out organic origins of the patient's complaints, they may attribute the symptomology to psychological factors, which can leave the patient feeling dismissed and misunderstood (Hareide et al., 2011). General Practitioners (GPs) often feel that they lack the appropriate language to communicate and manage these patients helpfully which can at times impact on the doctor's identity and understanding of their roles (Stone, 2014b). GPs also report they do not have enough strategies to explain PPS which may mean repeated consultations for patients (olde Hartman et al., 2009). Additionally, their professional credibility may feel under threat as these patients are not clear cut and easy to diagnose (Mik-Meyer & Obling, 2012). This results in both doctors and clients experiencing frustration from their interactions (Newby & Andrews, 2017).

1.6.1.1 Identity formation. The interactional component between PLWPPS and their HCP may indicate that if the professional's identity is impacted, then the client's identity may also be affected. As highlighted above, PPS can call healthcare workers to question their professional credibility and identity but for people with these diagnoses, their own identities can be questioned and / or doubted (Burbaum et al., 2010). The role of attachment, trauma and negative childhood experiences on the development of PPS has been established in the

literature (Adshead & Guthrie, 2015; Maunder & Hunter, 2014; Waldinger et al., 2006).

These factors all influence how individuals form and construct their identities; therefore, it is necessary to understand how people with PPS make sense of their identity in the context of a stigmatised illness. Burbaum and colleagues (2010) discuss conflicting identities in people with PPS, expanding that some people with these diagnoses will identify strongly with the sickness role while others feel part of a community and identify as successful individuals.

Often when people become ill it can impact on the roles they take in society. Ideas around illness careers have been discussed, focusing on the social impact of “legitimate” illness and the opportunities that people are afforded (Freidson, 1988). When patients visit their GP, they use language and tell stories to effectively “convince” the doctor that their illness is legitimate. Japp and Japp (2005) discuss the concept of ‘legitimacy narratives’ and highlight the need for patients to seek medical attention to establish legitimacy for their distress amongst medical professionals and peers. This concept is underpinned by Fricker’s ideas of “*testimonial injustice*” which highlights how pre-existing stereotypes around this type of illness influence the audience’s experience of the narrator’s story (Fricker, 2007; Kidd & Carel, 2017). However, when illness is of unspecified aetiology, people are not given the same social allowances as those with diagnosable conditions (Rossen et al., 2019). This affects the way PLWPPS construct their identities as the legitimacy of their patient identity is questioned. GPs and Psychotherapists also play a role in co-creating an identity for their patients. GPs may struggle to address the specific psychological needs of these patients, while psychotherapists may *only* attribute the symptoms to psychosocial factors (Burbaum et al., 2010). Additionally, research suggests that identity formation, specifically the uptake of a stigmatised identity can be attributed to the responses of family members and HCPs, highlighting that there may be a need for a more systemic, familial approach (Sowińska & Czachowski, 2018).

Diagnoses, specifically psychiatric diagnoses have a way of informing the individual's identities and language often conflates the disorder with the individual for example, you *are* psychotic, rather than, you have psychosis (Rossen et al., 2019). However, specific medical diagnoses provide a certainty that removes the onus of disease e.g. it is not something that is inherent in your identity, rather, something that you have, implying a more societally acceptable identity (Rossen et al., 2019). Illness narratives contribute to how people construct their identities, specifically when they may face a loss of their personal identity when dealing with illness while also fighting for acknowledgement of their patient identity (Charmaz, 1983; Rossen et al., 2019; Sowińska, 2018).

Having explored how PLWPPS construct their identities and their need to have their experiences legitimised by others, the next section explores the person's experience of their illness.

1.6.2 Adult's perspective of PPS.

PLWPPS can find their interactions with doctors may leave them feeling misunderstood, frustrated and needing to convince doctors that they are genuinely ill (Stone, 2014a). PLWPPS describe the fluctuations of their condition as chaotic, laden with uncertainty and a lack of clarity, while also being bothered by ambivalence from HCPs and not being believed or being told that the problems are only psychological (Nettleton et al., 2005). As a result of these experiences, Nettleton and colleagues (2005) discuss how PLWPPS find their symptoms and their interactions with HCPs leave them feeling marginalized from healthcare services.

A daily struggle with social engagement and everyday tasks both inside and outside the house is described by fibromyalgia sufferers, with women finding it particularly difficult to perform the gender roles that they deemed important to their identity (Paxman, 2019).

PLWPPS report as symptoms often change over time, with fluctuating intensity and type,

they develop a greater understanding of their condition (Sowińska & Czachowski, 2018).

PLWPPS discuss the need for their GPs to treat them as an individual within a context rather than just a cluster of symptoms, also highlighting that good quality communication with their GP is integral to feeling understood (Houwen et al., 2017).

1.6.2.1 Parent's perspective of PPS. For those PLWPPS who are also parents it is reported that their parenting lifestyle is impacted (Duryea, 2008; Fisher & Chalder, 2003). People diagnosed with chronic illness report a sense of loss resulting from the illness-free life they cannot live (Duryea, 2008). This sense of loss may lead to guilt, depression and anxiety about their ability to parent and the roles they can no longer fulfil which may subsequently impact on their parenting capacity (Ahlström, 2007; Duryea, 2008). Parenting style also plays a role in development and maintenance of PPS, for example, the relationship between the development of CFS and the concept of maternal overprotection has been established (Fisher & Chalder, 2003, Janssens et al., 2009). The influence of maternal overprotection on the development of negative health beliefs in their children may be due to anxiety surrounding their own illness development and behaviour (Fisher & Chalder, 2003).

Missen et al. (2012) reflect on the financial impact an unwell child has on the family, detailing a loss of parental income and an increase in monthly outgoings, which may lead to a subsequent impact on specifically the mother's physical and mental health and their familial relationships. Evans and Keenan (2007) offer a counter to solely focusing on the mother's contribution to subsequent childhood PPS by reporting that children with fathers with chronic pain exhibited more psychological and physical problems compared to children with healthy parents.

Sieh et al. (2013) discuss that around 10% of children with unwell parents are at a greater risk of psychosocial adjustment problems, which are involved in the development of PPS. They attribute this to the family cluster effect, which highlighted the combination of

genetics and environment (Sieh et al., 2013). Craig et al., (2002) discuss how ‘somatising’ behaviour can be learned intergenerationally and cite difficult parenting relationships and parental unexplained illness as contributory factors to poorer psychosocial development in the children. Rutter (1966) also discusses that somatic illness in a parent can be a risk factor for later mental health problems in the child which increases vulnerability to development of PPS. Having an unwell parent, fluctuations in parental coping ability as well as the child’s age all have an influence on unexplained symptoms in the child and later development of adult PPS (Hotopf et al., 1999; Romer et al., 2002). However, the underlying mechanisms between these relationships are unclear.

Much of the literature references the role of the parent, specifically the mother with an undertone of mother blaming (Craig et al., 2002; Hotopf et al., 1999; Rutter, 1966). Mother blaming is a concept where women are held wholly responsible for the health of their children, even into adulthood, as well as being blamed for their children’s difficulties in a way that fathers often aren’t (Jackson & Mannix, 2004). Vallido et al. (2010) discuss how mothers who experience physical illness have a difficult time negotiating the roles of mother and ‘patient’, while also feeling that they are not sufficiently supported by HCPs. This bias in the literature may seek to reinforce the difficulties that woman and mothers have in being taken seriously when seeking help for their own and their children’s health (Jackson & Mannix, 2004).

1.6.3 Children’s perspective of PPS.

The experiences of children or adolescents with PPS has been established in the literature (Eminson, 2007; Gillelud et al., 2009; Hotopf, 2002; Morris & Ogden, 2012). Parslow et al. (2017) found that children with CFS experience difficulties with being believed and disruption to the sense of self. They discussed that children with PPS find themselves suspended between a state of uncertainty about recovery and a state of hope for the future

(Parslow et al., 2017). Teenagers with PPS discuss how they would prioritise a sense of feeling 'normal' rather than share their distress about their symptoms with parents or friends and risk social exclusion due to their symptoms (Moulin et al., 2015a).

A review of work with children with 'somatisation' found that children's anxieties and health complaints is often an echo of the parent's fears for their child (Garralda, 1996). Konijnenberg et al. (2005) highlighted that children with unexplained pain show significantly reduced school attendance and engagement in social and sporting activities. The fluctuation in school attendance for adolescents with CFS often leaves them feeling lonely, left out and forgotten by their peer groups (Winger et al., 2013).

1.6.4 Family Perspective.

Much of the current research focuses on the individualistic perspective however; PPS conditions induce challenges for the adults (parents), the child, and the HCP so it seems that more of a relational, family perspective is warranted. Family based research is important as it shifts the focus of attention from the individual to understanding the meaning of phenomena at the family level (Gilgun, 2005). Research has highlighted that within families where children have PPS, there is often evidence of illness amongst the family, often (but not exclusively and under researched) in the mother (Garralda, 1996).

Garralda (1992) discuss the role that family functioning plays within the development and maintenance of somatic symptoms in the child, highlighting that problematic relationships with health within the family may relate to development of 'somatic symptomology' in the child(ren). As with parent-held illness beliefs, family-held illness beliefs have an influential role in the management and treatment of PPS further fuelling the need to take a family level perspective (Garralda & Chalder, 2005). Clinically, it is not uncommon for families to reject a psychological perspective for their children's PPS and to reportedly feel let down by medical systems (Kozłowska et al., 2012). Regardless of whether

a child or parent has PPS, Gilleland et al. (2009) consider the relationship between both the roles of 'parental somatic' behaviour and children's emotional functionality, concluding that there is an interaction at both parent and child level indicating a family perspective is advised.

Keeping in mind the relational impact of families with these conditions, Roy (1982) highlights that for families where illness is present there are higher levels of agreeableness between the members and a greater sense of closeness. This suggests that the experience of being unwell brings families together, even if they do not feel understood by others (Garralda, 1996). Rosland et al. (2012) review articles on the impact of family interactions and illness responses concluding outcomes are positively affected by strong family cohesion and self-reliance on the family unit. Lastly, Crix et al. (2012) explored how talk around illness is negotiated within a single-family unit where CFS is present. Their findings draw on Minuchin's (1974b) systemic ideas of triangulation whereby problematic parental relationships are triangulated through child illness. The authors conclude that all family members experienced problems attributed to the illness, but also, with their familial communication processes and negotiation of roles.

Overall, the key medical and psychological explanations of PPS have been explored as well as the multiple perspectives presented in the literature. What is evident is that the relational perspective/ family understanding of these difficulties is important and currently under researched – a systematic review will now be conducted to further explore the evidence base to see what is available.

2. SYSTEMATIC LITERATURE REVIEW

2.1 Overview

This chapter focuses on a systematic review of peer-reviewed literature, detailing how the search was conducted, how the studies were identified and why they may have been included or excluded from the review. Then a summary of the findings and an assessment of quality using Tracy's "big tent" criteria are presented (Tracy, 2010). This quality assessment criteria were chosen as due to its wide applicability and acceptability amongst the research community (Tracy & Hinrichs, 2017). The following questions will be answered with this review to provide a clear and current understanding on this topic:

1. From what perspective do families with PPS currently understand their experiences?
2. What is known about the lived experiences of families where there is PPS?

Lastly, the chapter presents the rationale for the project and aims for the research.

2.2 Search strategy

The search strategy was constructed using a) the synonyms from terms within the project title, b) the systematic review questions and c) key words from abstracts of important articles on the topic included in the introduction.

The initial searches took place on 5th January 2020. Databases searched included PubMed, Scopus and APA PsycNet. An additional hand search of Google Scholar was undertaken which added no extra resources. In total, 313 articles were found using the constructed search strategy (Appendix A). This reduced to 305 after the removal of duplications. These papers were title and abstract screened and at this stage, they did not seem appropriate for the systematic review questions thus the search strategy was re-defined and re-run. The second search was conducted on 15th May 2020 and a third search on 4th June

2020 with the revised search strategy detailed below in Table 1. The inclusion and exclusion criteria and process outcome are detailed in Table 2 and the flow diagram below (image 1).

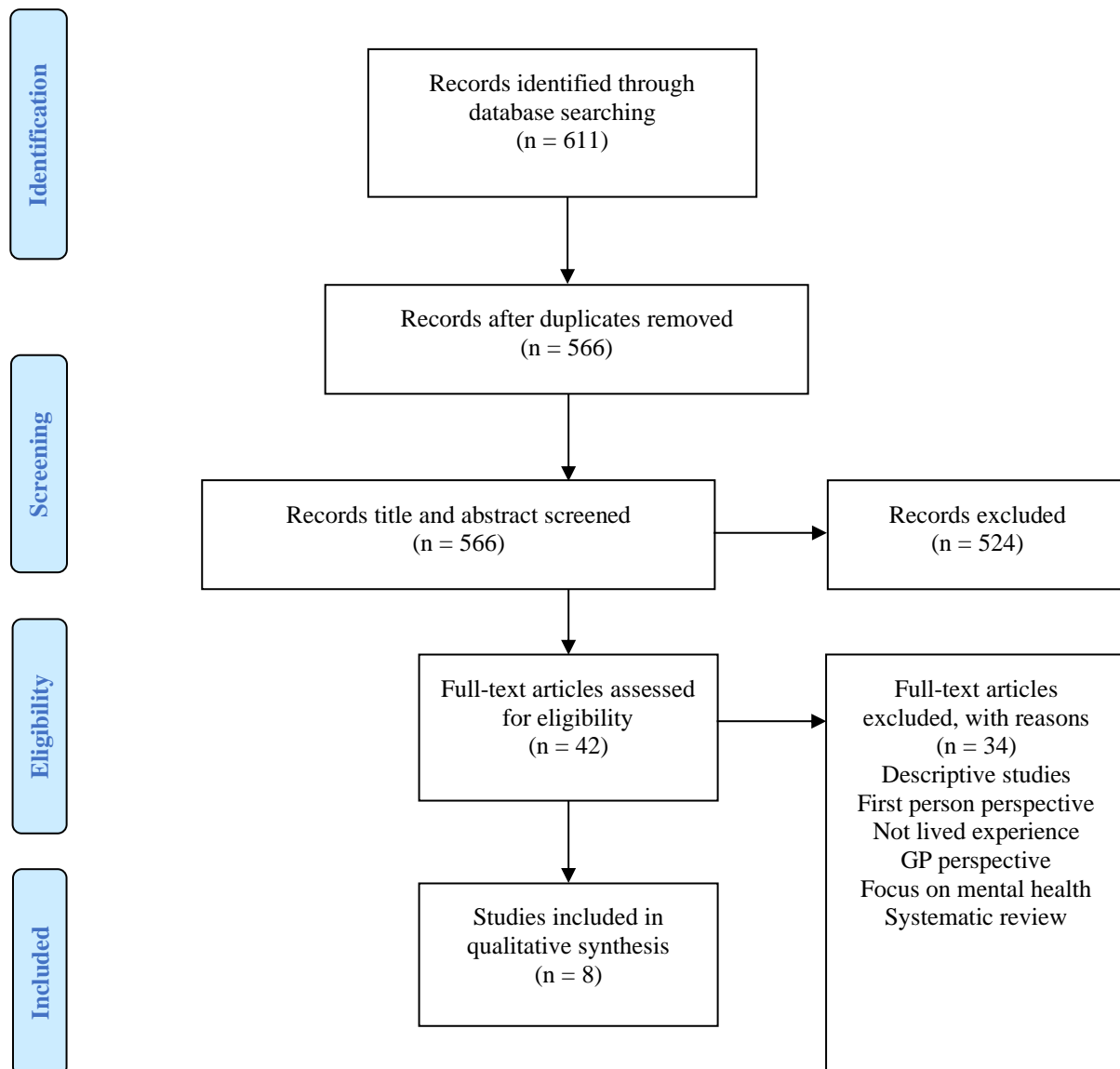
Table 1: Search Strategy

Family	Parent OR Parents OR mother* OR father* AND Child* OR daughter* OR son* OR teenager* OR adolescent* OR family OR families OR “family system” OR “family assessment” OR intergeneration* or transgenerational
Diagnosis	“Persistent physical symptom*” OR "PPS" OR "MUS" OR "medically unexplained symptom*” OR "somatic illness" OR somatisation OR somatization OR “somatoform disorder” OR "functional illness" OR "idiopathic illness" OR "psychogenic illness" OR “conversion disorder”
Methodology	qualitative OR “qualitative research” OR “Narrative*” OR “lived experiences” OR “narrative enquiry” OR “Narrative Inquiry” OR “Narrative Research” OR quantitative OR “quantitative research”

Table 2: Inclusion and Exclusion Criteria for Systematic Review

Inclusion Criteria	Exclusion Criteria
A focus on lived experienced	Non-human subjects
Peer-reviewed original research	Descriptive Studies
Any PPS condition	Grey literature or unpublished work
Focus on a relational or family perspective ¹	Not published in English
	Primary focus on mental health
	From GP perspective
	Only the individual’s viewpoint about their own condition
	Full text not available online
	Wrong population e.g. GPs
	Case studies

¹ Any lived experience of family life is included.

Image 1: Flow chart for systematic review

The initial search conducted (Appendix A) contained the words ‘case study or series’ and did not include any quantitative research. These search terms were originally included because it felt relevant to the current research. However, the findings from this search were largely descriptive case studies which did not fit with the other inclusion criteria for the study (Table 2). The second and third searches were run with the inclusion of ‘quantitative research’ and the removal of ‘case studies or case series’.

The concepts used to construct the search strategy were focussed around variations of family, the diagnosis and a description of lived experiences/methodology. The variations in search terms were combined with Boolean operators (AND, OR, NOT) as well as employing truncation tools to ensure that I gathered variations of specific words. The separate concepts overall were combined with 'AND'. Regarding the 'diagnosis' aspect of the search strategy, specific diagnoses, such as CFS, were not included because the research is interested in the lived experience of stigmatised illness rather than the experience of a specific diagnosis. The "abstract/title/keyword" or "all fields" options were used within each dataset.

When finding appropriate and relevant studies for this review, certain inclusion and exclusion criteria were employed (Table 2). Within the final search there were three relevant reviews; one narrative and two systematic reviews which were subsequently hand searched for appropriate papers (Dunne et al., 2019; Hinton & Kirk, 2016; O'Connell et al., 2020).

2.3 Quality assessment

Tracy's (2010) eight "big tent" criteria were chosen to assess the papers found via the above search strategy. Following comparison with other quality frameworks such as Elliot et al. (1999) and Madill et al. (2000), Tracy's (2010) criteria were chosen as they cover several key ideas that will assess quality and provide a multi-layered approach to evaluating qualitative research. Overall quality ratings of "high" and "low", have been attributed to the studies if they were deemed to have made overall efforts to produce quality research. For ease of rating, studies were given a "high rating" if they met four or more of Tracy's (2010) eight quality criteria.

2.4 Data extraction

Data was extracted and collated by the researcher into a table to summarise the findings (Table 3) which are presented in more detail below.

Table 3: Summary Table for Included Studies in the Systematic Review

Author/Title	Aims	Sample	Method	Key findings
Carter (2002) <i>Chronic pain in Childhood and the medical encounter: professional ventriloquism and Hidden voices</i>	“The purpose was to explore the impact of living with chronic pain from a perspective of children and families”	Recruitment: UK based Children aged seven to 13 years old Three families took part – Each family was interviewed on at least 2 occasions	Qualitative methodology: Thematic analysis Journals and loosely structured interviewed.	1. The quest for a diagnosis and referral fatigue” 2. “Professional judgement and disbelief” 3. “Communication or Ventriloquism?” “Professionals who believe the family
Dennison, Stanbrook, Moss-Morris, Yardley & Chalder (2010) <i>Cognitive Behavioural Therapy and psychoeducation for CFS in young people: reflections from the families’ perspective</i>	“To examine the under researched question of the views of the experiences of patients and families who took part and to elicit participants experiences in the own terms to understand their expectations, therapy experiences”	Recruitment: UK based – KCL hospital based 16 young people (10 females and 6 males – all white) interviewed and 16 parents took part (14 mothers and 2 fathers) Children aged 16 to 24 years	Qualitative methodology Telephone interviews Inductive thematic analysis	30 minor themes from the young people interviews and 31 minor themes from the parent interviews – organised into three themes 1. “Pre-therapy ideas and expectations” 2. “Experiences of therapy” 3. “Perspectives on effectiveness”
Moulin, Akre, Rodondi, Ambresin & Suris (2015) <i>A qualitative study of adolescents with medically unexplained symptoms and their parents. Part 1:</i>	“To explore how these adolescents and their parents experience the condition and its impact on their daily lives and to provide recommendations for HCPs”	Recruitment: Switzerland Based – French language 10 adolescents with different PPS conditions – aged between 12 and 20 years old (7 female and 3 male) 16 parents (11 mothers and 5 fathers)	Qualitative methodology: Focus group – six focus groups and two individual interviews Thematic analysis	Themes identified were: 1. “Disbelief” 2. “Being different” 3. “Hiding the symptoms”. 4. “Adolescent’s health first”

experiences and impact on daily life

Moulin, Akre, Rodondi, Ambresin & Suris (2015)

A qualitative study of adolescents with medically unexplained symptoms and their parents. Part 2: How is healthcare perceived

“To explore how the experiences with, and perceptions of the healthcare of adolescents who have MUS and their parents”

Recruitment: Switzerland Based – French language

10 adolescents with different PPS conditions – aged between 12 and 20 years old (7 female and 3 male)

16 parents (11 mothers and 5 fathers)

Qualitative methodology:

Focus group – six focus groups and two individual interviews

Thematic analysis

Six themes identified:

1. “Needing a label for the symptoms.”
2. “Seeking an aetiology to explain symptoms”.
3. “Negotiating the medical system”.
4. “Medication and treatment – medication, psychiatry and treatment, complementary and alternative medicine”.
5. “Interactions with doctors”.
6. “Inclusion of parents during consultations”.

Karterud, Risør & Haavet (2015)

The impact of conveying the diagnosis when using a biopsychosocial approach: A qualitative study among adolescents and young adults with NES (non-epileptic seizures)

“To explore the impact of using a biopsychosocial approach to explain the diagnosis of non-epileptic seizures (NES)”

Recruitment: Norway based – hospital

11 adolescents and young adults

10 female and 1 male

Aged 14 – 23 years old

Qualitative methodology

Interview

Systematic text condensation

Three key themes identified:

1. “Threatened self-image”
2. “Being believed and belief in oneself”
3. “Getting and explanation

McWilliams, Reilly, McFarlane, Booker & Heyman (2016)

Nonepileptic seizures in the paediatric population: a qualitative

To understand the experiences of young people (aged between 0 and 19 years old) with non-epileptic seizures and their families.

Recruitment:

UK based, Great Ormond Street Hospital

20 participant families – 10 patients (4 females and 6 males) and 29 family

Qualitative methodology:

Focus group

Telephone interview

Six main themes with 3 subthemes:

1. “Upset and Afraid”
2. “Missing out”
3. “Feeling misunderstood”
4. “Confusion and Uncertainty”
5. “Less than epilepsy”
4. “Making sense and moving on”

<i>study of patient and family experiences</i>		members (22 at focus group and 7 by telephone)	Thematic analysis	
Mantilla & Rojas (2018)	“To describe explanatory models (EM) offered to caregivers of paediatric patients with conversion disorder who attended the <i>hospital de la Misericordia</i> ”	Recruitment: Colombian based – outpatient clinic in hospital Nine mothers and one father were interviewed (10 diagnosed cases with conversion disorder [5 female and 5 males], aged nine -17 years old).	Qualitative methodology In-depth interview with parents and caregivers Grounded theory and phenomenological design	<ol style="list-style-type: none"> 1. “Beliefs about the origin of the symptoms” 2. “Experience of symptoms” 3. “Impact on daily life” 4. “Treatment-related aspects”
<i>The visible and less visible in the suffering of a conversion disorder in children and adolescents. A qualitative study of illness explanatory models presented to caregivers and adolescents with conversion disorder</i>				
Hulgaard, Rask, Risor & Dehlholm (2020)	Exploring children with severe functional disorders and their parent’s illness perceptions	Recruitment: Denmark based, CAMHS paediatric department 11 young people (10 female/1 male), aged 11 to 15 years old 16 parents (9 mothers/7 fathers)	Qualitative methodology: Interviews IPA	<p>Three main themes identified:</p> <ol style="list-style-type: none"> 1. “Ascribing identity to the disorder” 2. “Monocausal explanations” 3. “Mutable illness perceptions”
<i>Illness perceptions of youths with functional disorders and their parents: An interpretative phenomenological analysis study</i>				

2.5 Summary and findings

Overall, eight qualitative papers were included and summarised with the details of the participants and the demographics of the studies. The inclusion criteria of 'lived experience' was deemed to be most important for the research question, which meant that quantitative studies were not included in the final review. Overall, the systematic review questions will be answered by addressing the following themes that appear in the literature; the biopsychosocial framework and family perspective or functioning. Following the literature review a separate quality assessment is undertaken.

2.5.1 From what perspective do families with PPS currently understand their experiences?

People with PPS have previously been misunderstood by the medical model, despite families primarily wanting a medical definition (Moulin et al., 2015a). Karterud et al. (2015) propose that young people with NES prioritised taking a biopsychosocial approach above a purely psychological understanding to justify their experiences to others, to legitimise their illness and inhibit any negative impact to their identities and social roles. The authors interviewed 11 young people aged between 14 and 24 years old, who felt that having a link between their symptoms and their personal histories helped to explain and understand their experiences and provide meaning. Despite mentioning the incorporation of personal histories and wider psychosocial influences, Karterud and colleagues (2015) made no mention of the family context, the role of the parents, nor which aspects of a personal history are most helpful for meaning making. This may be problematic given that their participants describe the need to be believed as well as wanting an explanation for their experiences, which fits with a more interpersonal/ relational perspective. Findings from this study should be interpreted with caution as when considering Tracey's (2010) quality criteria, there were

methodological issues and a lack of clarity and credibility that meant that the quality of the paper was rated as “low”.

Mantilla and Rojas (2018) took an opportunity sample of ten children as inpatients, diagnosed with ‘conversion disorder’, aged between six and 17 years old. They interviewed their parents or caregivers about how they make sense of their child’s illness. The parents favoured both a psychosocial and a “magical/mystical” explanation but suggested many coexisting models may provide a succinct explanation. These mixed findings highlight that there is no clarity in how families understand these conditions. Mantilla and Rojas (2018) also shared that there was no clear impact that these symptoms have on daily life for the family and that there is no correlation between the different explanations and the impact it has on the family. The authors’ findings do not provide any further understanding about the current conception related to families with PPS. Additionally, the Colombian based project purports findings such as “magical/mystical thinking” to understand PPS, however, these findings denote a cultural shift that is significantly different to a UK population and therefore may not resonate. This, among a poor description of their ethical processes, the credibility and the resonance of their results meant that this research was deemed “low” in quality.

Moulin et al. (2015a) discuss the differences in how parents perceive their child’s illness in comparison to their adolescent’s own perception of the illness. Having interviewed 10 adolescents with different PPS conditions and 16 of their parents, they suggested that the adolescent’s health comes first, often causing a loss of the voice of the parent about their own perspective and needs. Moulin and colleagues (2015a) also highlighted that the family often felt isolated within their own context as they felt unable to communicate their difficulties; fearing others would not understand. This study however, had large numbers of people who declined to be part of the research, which may be a consequence of the transient nature of the symptomology and participants willingness to engage with talking about their experiences.

Overall, the research was deemed high quality due to attention given to ethical procedures, novel and relevant research, as well as findings that resonate in the wider European context.

Hulgaard et al. (2020) participants favoured a single causal explanation (as opposed to multicausal like the biopsychosocial model). They interviewed 11 young people aged 11 to 15 years old and their parents about their perceptions of the child's illness, finding that once a medical reason had been excluded, they tended to favour a psychological explanation, however, they did not elaborate on this further. Hulgaard and colleagues (2020) also revealed that the families' prior experience with healthcare services influenced the way the families perceive and understand the child's PPS. This "high" quality and very current review of patient's perception of their experience provides a potential insight into the direction that the literature may be moving, for example, moving away from the biopsychosocial model. This study is based in Scandinavia, however, while despite similar demographics, there is a different cultural way of living which may mean that the findings aren't directly comparable with a UK sample.

Carter (2002) reported that the voice of the child gets lost and discusses the ideas of "adult-child ventriloquism" whereby the parents speak for the child and it is the HCPs interpretation of the adult's version of the child's experience that gets noted. They also felt that when interviewing children and their parents, families struggle with managing the illness because of a lack of clarity around diagnosis and treatment. Families also report what they describe as "referral fatigue" after hearing too many differing messages from repeated doctors, tests and referrals. Finally, they suggest that HCPs are unaware of the true impact that PPS has on families and perhaps the subsequent reinforcing element of family life on the symptoms. Carter (2002) highlights the discrepancy between the families' understanding of their own experience and the medical professionals' perspective and the need for a mutual

understanding. This research was deemed high quality due to its strong research rigor, novel and worthy topic as well as providing sincere and credible research (Tracey, 2010).

It appears that there is no sense of clarity on how families with PPS want to be understood, with mixed preferences for the biopsychosocial approach, singular causal approaches or a more individualistic approach. However, little is known about the family's experience from a more systemic perspective and perhaps how the relational aspect fits with how families understand their own PPS. The next review question seeks to further understand what is known about the lived experiences of families with PPS.

2.5.2 What is known about the lived experiences of families with PPS?

As seen, there is a mixed view of what is known about families with PPS and from what perspective is currently favoured, however, it is useful to explore what is known about the experiences of families where there is PPS thus highlighting any gaps in current thinking and research.

Families have reported feeling the need to legitimize their illness. In the case of NES, there was even a plight of not receiving appropriate treatment compared to 'organic' illness i.e. epilepsy (McWilliams et al., 2016). There was also a theme across the literature which highlights that families with these illnesses often feel misunderstood by others: their peers, HCPs and sometimes their own family members (McWilliams et al., 2016). They conducted focus groups with ten young people and 29 family members, and the results showed six main themes that described how families experience life living with PPS. Families reported feeling scared about the future, that they were missing out on life, that they were misunderstood by people around them, as well as a sense of uncertainty and lack of clarity about understanding the illness. The final theme was about meaning making, which the parents felt was pivotal to recovery, however, the children exhibited a sense of indifference. As will be seen below, the findings from this study should be taken cautiously given that it was deemed on the lower

end of the quality scale, as the findings were not wholly novel, nor was there clear evidence of credibility or sincerity (Tracey, 2010).

Some of these themes are echoed by Moulin and colleagues (2015a) who interviewed adolescents and their parents about their experiences of living with MUS where they discuss a sense of disbelief from people around them, which they attribute to people not understanding these illnesses. The adolescents within this group spoke about how in the beginning, the disbelief started with their parents. The second theme of *being different* affected the parents and the adolescents differently. The adolescents reported feeling excluded and rejected from their peers, which was corroborated by their parents. However, for the parents, they did not communicate their needs to their friends which meant that they also felt isolated. They highlighted that there was a stronger reciprocal/co-dependent relationship between the parents and teenagers as noted by both parties. This would allude to the importance of a relational component when trying to understand the experiences of families with PPS. However, the teens differed from their parents in how they appeared to cope with their difficulties, for example, the adolescents wanted to mask their symptoms and pretend that they were 'normal' like their peers. Their parents on the other hand, as a coping mechanism, felt the need to discuss their difficulties with others. This suggests that there may be a generational difference or an experiential difference e.g. a prioritising of the symptom sufferer. Finally, the authors suggest that PPS influences relationships in different ways, this being an important aspect for understanding the illness experience and for treatment. Continuing this idea of treatment planning, Moulin and colleagues (2015b) discuss that for the parents, in order to have a greater understanding and ease through navigating the healthcare system, they often long for a name or a label and on occasion want something pathological for their children. This need for a label often puts parents in a conflictual

position for wanting something recognizable to be *wrong* with their children. Moulin et al. (2015 a&b) were both deemed to be 'high' quality.

Karterud et al. (2015) discuss the importance of being believed by doctors and having to legitimise their illness through their language. This impacted their illness identity which has been shown to affect societal roles, credibility and functioning. However, they do not draw on the role of the family or how the family is impacted by the condition, therefore it is hard to draw any conclusions about the family's experience. Further, given the study was based in Norway, it is not clear how much the findings would resonate with a UK population should they have considered the family in more detail.

Hulgaard et al. (2020) frame their understanding of 'functional' illness with Leventhal's common-sense model (CSM) of self-regulation (Leventhal et al., 2016) which explores illness perception over several elements such as identity, timeline, control, causes and consequences. They discuss the important role that language and communication plays in the way that patients come to terms with their illness and how it informs their identity. The parents of 11 young people with PPS shared that they struggle with the term 'functional' as it seemed to delegitimise their illness identity. The families reflect on the difficulty they feel with the association between the symptomology and a psychiatric explanation and feel that attempts to locate the problem in the family and move away from a medical explanation may feel quite blaming. They highlight the need for future research to focus on the construction of illness perception amongst a family context. A strength of these results may highlight that having a model in mind when formulating PPS conditions provides a useful foundation for a more cohesive understanding for these conditions and it provides a structure when working with complexity.

Moulin et al. (2015b) discuss the important role of parental involvement in medical consultations with their children. They found that parents (and their children) felt it was

important for them to be involved in the consultations despite on occasion the adolescents feeling that their parents monopolised the conversation. This highlights the need to take a more relational/familial approach when working with families who have PPS. Dennison et al. (2010) were also interested in the experiences of patients and their families who had either CBT or psychoeducation intervention for CFS. As part of a wider study they invited 46 adolescents to be interviewed, however only 18 consented to take part, as well as 16 sets of parents. It was not clear why they had such poor uptake which contributed to the low-quality rating this study received. Briefly, they found that the adolescents and their parents both suggested that family involvement was important, specifically with comprehension and practical involvement in the treatment. The families also suggested problems with the rigidity of CBT and psychoeducation, which suggests that a more flexible approach is more suitable to families with PPS.

A summary of all the findings from the review have been presented, a full quality evaluation will be explored below.

2.6 Quality Evaluation

Much of the literature found was from a European/Scandinavian perspective therefore the breadth of conclusions drawn, and the resonance of the findings should be taken with caution. Tracy (2010) offers eight criteria in which you can assess quality in qualitative research, these are; worth topic, rich rigor, sincerity, credibility, resonance, significant contribution, ethical and meaningful coherence. These findings are briefly reported in Table 4, however, full quality assessment notes are found in Appendix B.

Table 4: Quality Assessment table

Paper	Worthy Topic	Rich Rigor	Sincerity	Credibility	Resonance	Significant Contribution	Ethical	Meaningful coherence	Overall Quality rating
Carter (2002)	✓	✓	✓	✓	✓	✓	Maybe	✓	High
Dennisson, Stanbrook, Moss-Morris, Yardley & Chalder (2010)	✓	Maybe	×	×	Maybe	✓	✓	✓	High
Moulin, Akre, Rodondi, Ambresin & Suris (2015a)	✓	×	×	×	✓	✓	✓	✓	High
Moulin, Akre, Rodondi, Ambresin & Suris (2015b)	✓	×	×	×	✓	✓	✓	✓	High
Karterud, Risør & Haavet (2015)	✓	×	Maybe	×	Maybe	✓	×	✓	Low
McWilliams, Reilly, McFarlane, Booker & Hayman (2016)	✓	Maybe	Maybe	×	✓	×	×	Maybe	Low
Mantilla & Rojas (2018)	✓	✓	✓	×	×	×	Maybe	×	Low
Hulgaard, Rask, Risor & Dehlholm (2020)	✓	✓	×	Maybe	✓	✓	×	✓	High

Worthy Topic. For research to be high quality it needs to be considered as novel, relevant and interesting. All eight papers were deemed to meet criteria. They were all novel and interesting for different reasons: focusing on methodology, perspective, or explanatory models. In terms of methodology, Hulgaard et al. (2020) used interpretative phenomenological analysis (IPA) and provided new thinking about the biopsychosocial approach to PPS. Dennison et al. (2010) applied qualitative methodologies to CBT evaluation, novel at the time of the article. Carter (2002), McWilliams and colleagues (2016), Moulin et al. (2015a, b), and Karterud et al. (2015) all spoke from different, novel perspectives such as from the perspective of the family, separate parent and children perspectives, the child or young people's perspectives. Lastly, Mantilla and Rojas's (2018) research offers explanatory models as a novel way of understanding often misunderstood conditions.

Rich Rigor. This theme relates to the appropriateness and complexity of the sample chosen, as well as the authors' detail of the context of the research. This concept also explores the processes involved with data collection and analysis. The findings for this quality aspect were mixed. Studies were deemed to be lacking in research rigor if they did not seem to have appropriate numbers for their chosen analyses, had large participant drop-out rates, required retrospective recollection for the data or if their recruitment strategy was not clear (Dennison et al., 2010; Karterud et al., 2015; McWilliams et al., 2016; Moulin et al., 2015a, b). However, Carter (2002), Mantilla and Rojas (2018) and Hulgaard et al. (2020) were all deemed to have followed high quality rigorous research proceedings when it comes to their sample and how the authors explained their methodology and processes.

Sincerity. When reviewing the quality of research, it is important for researchers to be aware of the subjective nature of qualitative research. Sincerity refers to the concept of transparency and the sharing of the authors' self-reflexive processes embedded in the

research. Most of the research included in this review failed to share their processes with the reader. Mantilla and Rojas (2018) attempted to offer elements of sincerity by documenting the potential influence their decision-making process may have on their research rigor. Carter (2002) mentioned that the study comes from a constructivist position and alluded to the possibility of a more reflective relationship to research, but they do not provide any more detail about how they challenged their own biases and assumptions. Each of the other papers did not share enough information to demonstrate that they had taken a transparent approach to their research.

Credibility. Credible research encompasses a process whereby researchers check their findings with their participants, provide rich detail that guides the reader rather than tells them what to think and lastly, evokes triangulation which promotes the importance of multiple sources of data. Overall, the credibility of the highlighted research was mixed. Some studies attempted to put forward credible research by using multiple researchers to code and analyse the transcribed data and come to a communal consensus of the themes achieved (Hulgaard et al., 2020; McWilliams et al., 2016; Moulin et al., 2015b). There were some papers that did not share any information about how they sought credibility in conducting and writing up their research (Dennisson et al., 2010; Karterud et al., 2015; Mantilla & Rojas, 2018; Moulin et al., 2015a), whereas Carter (2002) demonstrated their credibility by sharing that the interview schedule was informed by diaries from their participants. In addition, the authors gave ample and rich descriptions of the family experience and encouraged the families to provide their feedback on the transcripts before the authors reached their final conclusions. The other seven papers did not include any member reflections within their analyses, limited detail was provided and a lack of multiple voices were present when writing up their research.

Resonance. Through clear presentation, resonance denotes the idea of how ‘generalisable’ the findings are to wider, similar populations. Most of the findings would resonate with the wider audience, largely due to the presentation and the novelty of the research (Carter, 2002; Hulgaard et al., 2020; McWilliams et al., 2016; Moulin et al., 2015a & b). Karterud et al. (2015) showed potential for their research to have a wider impact, due to easily digestible results and processes, however, they had problematic recruitment processes therefore it is hard to draw conclusions about the wider resonance of the findings. Dennisson et al. (2010) based their findings on historical recollections of their participants therefore despite their findings having potential resonance, the quality of the data collection is questionable. They would also have benefitted from providing more detailed examples for their results to guide the reader to make an appropriate decision about the proposed generalisability. Similarly, Mantilla and Rojas (2018) did not provide any details or examples to back up their themes. Their findings related to magical or mystical thinking, which fits more with the Colombian sample, and is less applicable to a UK sample.

Significant Contribution. This concept is about the impact that the research leaves, whether this is from a methodological, conceptual, or practical perspective amongst other things. Carter (2002), Dennisson et al. (2010), Karterud et al. (2015), Moulin et al. (2015 a & b) and Hulgaard et al. (2020) all offered practical contributions that highlight the important role of communication between families and with HCP. Those studies that were deemed to not have offered a significant contribution due to either the recruitment processes or not coming to a helpful consensus with novel findings include Mantilla & Rojas (2018). Similarly, although McWilliams and colleagues (2016) allude to the importance of language and labels with this population, they stated that it was only “*one* of the first studies” suggesting a lack of novelty and significant contribution.

Ethical. Has the research been conducted in an ethical manner? This concept ensures that the research is conducted in an ethical way, from conception to review. It was a challenge to review this aspect of quality because most of the studies merely put in a sentence saying that they achieved ethical approval for their study. This would mean that an ethics board had approved the study and deemed it ethically sound, however, they provided little evidence of the ethical issues they had considered (Dennisson et al., 2010; Karterud et al., 2015; McWilliams et al., 2016; Moulin et al., 2015 a & b). Carter (2002) goes beyond this and discusses the importance of including the children in the decision-making processes about their involvement. Hulgaard and colleagues (2020), however, made no mention about their ethical process, which made it difficult to assess how they'd created ethically sound research.

Meaningful Coherence. Does the research achieve what it set out to do, and have they used appropriate processes and procedures to achieve it? All studies except for Mantilla and Rojas (2018) seemed to meet the aims that they set out to achieve, in an appropriate way and have their results and interpretations fit (or contribute) to the evidence base (Carter, 2002; Dennisson et al., 2010; Karterud et al., 2015; McWilliams et al., 2016; Moulin et al., 2015 a & b). Mantilla and Rojas (2018) did not get the findings that they had hoped for and their methodology may not have been appropriate hence their inconclusive findings.

Overall, the quality of the literature is mixed, with a lack of reflexive, constructionist research that offers a systemic, relational perspective to living with PPS. When thinking about what is currently known about families living with PPS, the evidence base is limited. Primarily, research focuses on the biopsychosocial understanding of PPS and discusses family experiences from the perspective of the parent or the child, normally about the unwell child's experience.

A strength of this systematic review is that it has provided an overview of the current family-based research on PPS, highlighting that research is lacking in this area and there is a gap. A limitation of this systematic review is that, while I had a solid rationale for not including specific diagnoses in the search, it may have meant that papers were missed in the process. Crix et al. (2012) was a relevant paper, however, it did not come up in any systematic or hand searches of the databases. This paper was presented to me via a discussion with a supervisor with a specialist research interest in the area. It was surprising that it did not come up, but it could be related to not including specific diagnoses. In future, I will ensure to include *all* variables in the search strategy.

2.7 Rationale for the research

There is little research exploring the relational component to these conditions and how families make sense and live with PPS from a systemic perspective. The literature alludes to there being an impact to family functioning and family life when living with PPS. However, there is no mention about what happens between family members when there is stigmatised illness, for example, how people's roles change within the family or what happens when more than one person is ill. There is evidence that where PPS is present within a family, it may be present within more than one generation, however this has not been explored qualitatively (Shraim et al., 2013). The evidence base offers that families seek out more of a single causal framework to explain their experiences which as can be seen, results in disbelief and delegitimised self-narratives. This research will explore singular narratives but also aims to see whether the collective narrative offered by a family differs from the constructions of the individuals in how they make sense of their own experiences. This is a complex area, however, there is a gap in the research exploring the following:

- There is clear evidence on how parental health influences child health and vice versa (Evans & Keenan, 2007; Garralda, 1996; Hotopf et al., 1999; Missen et al., 2012; Sieh et

al., 2013) but little on any relational patterns between the two generations with respect to health and an exploration of the 'intrafamilial environment' is useful to understand symptomology within families (Roy, 1982).

- There are many models that look at this area from an individualistic approach, with little recent exploration of this topic from a relational/systemic perspective.
- Lastly, there is little understanding or discussion around how families with PPS may construct collective family identities and navigate different role relations in the context of PPS.

Therefore, the rationale for this project is:

- From both a topic and a methodological standpoint, the research is novel and interesting as narrative inquiry within a family unit has not been explored with PPS, specifically focusing on role relationships and identity formation.
- It will be clinically relevant to address the experiences of these families from a systemic perspective as the current treatment and management for PLWPPS is an individualised, usually CBT perspective (Edwards et al., 2010).

2.8 Research Aim

The focus of interest of this research is thinking about how families (and individuals within those families) construct their identities and navigate role relations within a system, where two generations have PPS.

The following question will be explored through this research project:

'How do families construct their identities and make sense of their role relations when both parent and children are unwell with PPS?'

3. METHODOLOGY

3.1 Overview

The following chapter focuses on how I addressed the research question and the processes involved. I will guide the reader through my journey with clarity and transparency to ensure high research quality. The chapter reports on the specific research design; the chosen methodology and its rationale, as well as ethical issues and proposed resolutions, service user involvement and procedure for data collection and analysis.

The COVID-19 pandemic has had a significant impact on this project, specifically recruitment and data collection. A large proportion of this method section was written prior to the pandemic. As a means of authentically guiding the reader through the fluctuations of my MRP journey I have decided to write about changes to this methodology in a reflexive manner, using italics to comment on either changes to process or my reflections.

3.2 Design

This project used a qualitative methodology. Qualitative interviewing allows access to an individual's social world and a deeper understanding of how people make sense of their inner world, in the wider context (Miller & Glassner, 2011). This project utilises narrative methodologies to explore how we have come to understand the experience of individuals, based on the current socio-political and cultural context within which the research was created (Hunter, 2010).

3.2.1 Why narrative?

A narrative, as defined by Gergen (2015) is a story about events in consecutive order which allows the audience to make meaning. Narratives are co-constructed between individuals and in the context of research, between the researcher and participant (Mischler, 1997; Wells, 2011). Through this analytic framework researchers have the tools to make

sense of stories within their own complex contexts by exploring them from different levels, such as structural, contextual, or performative (Esin et al., 2014). Narrative analysis focuses on co-constructed meaning making, whether it centres on life stories, discursive tools used in storytelling or how people position themselves in relation to their stories (Esin et al., 2014). Riessman (2008) addresses the importance of the local and historical context and draws on the “taken for granted” wider social discourses that inform narrative construction.

The narrative methodologies drawn upon were constructionist narrative analysis (in line with the research’s epistemological position), with some elements of discursive psychology (Edwards & Potter, 2001; Esin et al. 2014). This analytic method permits the exploration of how participants use language to co-construct their stories as well as provide meaning within a socio-political, biographic and historical context (Esin et al., 2014). Aspects of discursive psychology are drawn upon when considering how people use their talk e.g. what rhetoric devices do people use to narrate their stories (Edwards & Potter, 2001). Additionally, this methodology was chosen as the researcher was interested in understanding how individuals make sense of their identities and role relationships in the context of a family with PPS in two generations. Narrative methodologies marry well with enquiring about stigmatised illness because we all story our lives, but narrative analysis provides a space for other stories to be told where a medicalised story is normally triumphed (McAlpine, 2016).

However, there are some limitations to this methodology in that, like many other qualitative methodologies, narrative frameworks are subjective. It can also be time and labour intensive to explore and present people’s life stories authentically (Moriarty, 2011).

3.2.2 Epistemological Position.

Constructionist methodologies are not interested in trying to illicit the “truth” of the individual’s experience, but rather trying to understand the social processes that have informed people’s constructions of the world (Gergen, 2015; Hunter, 2010). Social

constructionist perspectives on narratives take into consideration the biographical, historical, cultural and societal contexts within which the narrative sits (Liamputtong & Ezzy, 2005).

These contexts provide discourses that can be drawn on to make sense of the world.

This methodology not only accounts for the broader context but also for what people are able to do with their ‘talk’, for example, people have agencies to choose some frames of discourse over others, use different rhetorical devices, as well as take social action with their language (Edwards & Potter, 2001). Social constructionist methodologies do not position people as passive within their context and within an interaction (Riessman, 2008). The narratives that people construct are based on what social discourses are available to them and how they use language to construct their “truth” (Riessman, 2008; Wells, 2011).

This project used a single-family case study design. This methodology has been previously used by Crix et al. (2012) and Bamber (2014) to explore family experiences of illness in the child generation. The benefit of this methodology is that it allows for an in-depth exploration of both the unique individual narrative accounts and to see how family narratives are co-constructed in a specific context (Crix et al., 2012; Wells, 2011). To the researcher’s knowledge, this has never been done within the context of PPS in two generations, exploring both the individual and collective narratives surrounding identity formation and changing role relationships.

3.3 Ethical Issues

Recruitment took place in a specialist clinic in a large London Hospital with NHS service users as participants. NHS ethical approval processes were followed using the NHS Integrated Research Application System (IRAS). This involved considering all the possible ethical issues that may occur in a research project and attempting to find solutions to protect against unethical processes (excerpts from this form can be found in appendix C). Once all this information was collated, it was submitted and reviewed by the London NHS Research

Ethics Committee who, after some recommended amendments, deemed the project to be ethically sound (Rec approval reference:19/LO/1697; appendix C). After this, approval and project registration were sought from the Research and Development (R&D) team within the NHS trust. Once this was approved, full sponsorship was requested and given by the University of Hertfordshire Ethics board (Reference number: LMS/PGR/NHS/02942).

The ethical issues relevant to the study are the emotional content of the topic, consent and confidentiality, service user involvement and the consequences of previous literature and current stigmatising processes inherent with these diagnoses. This project explored the personal experiences of a family and how they make sense of their unique life events. Given the nature of the condition, the family may have already had a difficult experience navigating the health care system. When considering how to conduct the research ethically it was important to keep an open-mind and non-judgemental approach for fear of being “another HCP” that left the family feeling judged (Sowińska & Czachowski, 2018).

My planned recruitment strategy was to attend the hospital clinic to facilitate an ease of participation and convenience for the participants. However, given the COVID-19 pandemic this needed to change, and data would be collected digitally/virtually instead. I contacted the REC committee who deemed this a non-substantial amendment because it reduced labour for NHS staff and kept people away from hospitals. The amendment was approved by the local R&D department and university sponsorship (see appendix D). Just before the R&D department decided to stop all non-COVID related research projects, I had tense conversations about the future of the project.

I began to think about how I would manage a group interview over a web-based platform as I was now contending with freezing connections as well as potentially awkward silences. I was concerned with confidentiality in relation to working from home and undertaking the interviews while in a national lockdown. I felt anxious about contending with

multiple voices, not centring myself and attending to different social cues. When in person, it is easier to notice slight changes in people's expressions and body language, however, these nuances of communication are not overtly apparent when working over video. Aafjes-van Doorn et al. (2020) highlight that video working can bring about therapist's professional self-doubt and anxiety about how they come across on screen. This is significantly problematic for therapists who are young, female and those early in their careers (Aafjes-van Doorn et al., 2020). This mirrors some of the anxiety I was feeling about conducting the research interviews over Zoom. However, I followed guidelines from the Division of Clinical Psychology (DCP) on delivering effective therapy via video to help create an effective relationship conducive to producing meaningful research (British Psychological Society, 2020).

3.3.1 Consent and Confidentiality.

Given that the family were recruited from an adolescent medical service, they will have children under 18 years old. The information sheet and any communication provided enough information about any risks, benefits and possible outcomes of study so that participants gave informed consent (Appendix E). The younger children needed to assent to the study if they were under 16 years old and give consent via their parents (Appendix F). A study debrief form was given to the participants after they had participated (Appendix G). The information was accessible for both adults and children to be suitably informed. Confidentiality is an important ethical issue to consider; the interviews were recorded in accordance with university and Trust policy. The family members identities were protected by changing their names and having no other identifiable data in the results.

Pre COVID-19, I would have met the family in person, have gone through the information sheet and the consent forms and addressed any of their concerns. With the current restrictions, I consulted the Health Research Authority (HRA) website on guidance

for how to proceed (HRA, 2020). Townsend et al. (2020) highlight the importance of conducting high quality ethical research in the time of a global pandemic. Despite the restrictions and the necessary adaptations, appropriate consent procedures remained at the utmost importance considering there were multiple consent procedures e.g. under 16 assent and consent. I was aware that my fears about the project collapsing were fuelling my anxiety to explicitly follow all guidance about conducting research in uncertain times.

3.3.2 Other Ethical Considerations.

Additionally, given the parent blaming tone of some of the literature on this topic, mother blaming was avoided through careful use of language, maintaining a non-judgmental stance and having reflective discussions within supervision.

Given a context of avoiding mother-blaming, I was drawn to perhaps overcompensating with my interactions with the mother. I wanted to ensure I spent more time engaging the mother, trying not to offend her or make her feel uncomfortable. I wanted to accommodate the mother's needs, for fear that she had had negative interactions with psychologists and HCPs in the past. I worried about losing these participants if I had not made the mother feel comfortable with the idea of research participation. Additionally, as a woman, who is not yet a mother, I was drawn to how important it was for me to ensure that I did not unconsciously perpetuate mother-blaming narratives by paying attention to the needs of the mother and also by discussing this in supervision (Jackson & Mannix, 2004).

Finally, for a brief time I “entered” this family system during the interviews. I remained aware of the emotional impact of the interviews and signposted psychological support if necessary.

3.4 Sampling

In line with this study's research aims and methodological approaches a purposive sampling strategy was employed (Ames et al., 2019). Based on the background literature, the rationale for the research and the research aims I recruited a family where there are PPS in both the parent generation and the child generation, as well as "health" in both generations.

This study sought to recruit participants from a specialist adolescent service in London, which supports adolescents with complex medical needs and their families. There is no minimum age requirement for patients, but the current youngest service user is five years old. The upper age limit for this clinic is 19 years old. The service users are referred with many diagnoses such as NES, CFS, and fibromyalgia. The team were briefed about the project's inclusion and exclusion criteria. My external supervisor, who is one of the team psychologists, identified families who fit the inclusion/exclusion criteria and once identified, this pool of families were sent the information sheet and were asked to contact the researcher to opt in or to ask for more information. They were given two weeks to respond under ethical approval, otherwise assumed to have opted out. Given the scope of this project, an arbitrary cut-off of a minimum age of 12 years old was decided for the youngest member of the family; this was due to an assumed emotional maturity in a child of secondary school age (Swinson, 2010).

The chosen participant family were the first family who fitted the study criteria and had the availability and willingness to participate. The following inclusion and exclusion criteria (Table 5) were adhered to:

Table 5: inclusion and exclusion criteria for study

Inclusion Criteria	Exclusion Criteria
<ul style="list-style-type: none"> • A family with both a parent/carer and child with symptoms that fall under the umbrella of PPS. 	<ul style="list-style-type: none"> • Children younger than 12 due to a perceived/assumed level of emotional and physical maturity.

- Families where all individuals can sit for approximately 45 minutes
 - English speaking
 - A support system in place.
 - Non-English speaking and non-verbal individuals
 - Any safeguarding issues including severe social care issues.
-

Additionally, given the nature of the content i.e. exploring people's experiences and how they tell their stories, at this stage it is useful for the discussions to be undertaken in English. Esin and colleagues (2014) speak about stories getting "lost in translation" when working in a different language to that of the researcher. I am a native English speaker and therefore fluency in English language is important. Although, it would be interesting and important to explore the conflation of different cultures within the room and the impact this would have on the co-construction of meaning, I am new to narrative analysis and want to ensure the best analysis I can at this stage.

Wells (2011) suggests a participant pool of five is enough for an in-depth analysis using narrative methodology. Thus, a minimum of five "datasets" are required (Baker & Edwards, 2012; Wells, 2011). I have not specified any race, gender or sexual orientation of the family make-up as I was concerned about the availability of these families within the medical team. A discussion was had within supervision about what constitutes a family. The 'traditional' definition of a family according to the Cambridge English Dictionary is "a group of people who are related to each other, such as a mother, a father, and their children" (Cambridge Dictionary, 2020). However, this does not take into consideration the evolving nature of society and context whereby the meaning of family has changed and incorporates many variations of "family" (Sharma, 2013). For that reason, I did not wish to restrict myself by specifying a type of family, however, discussions within the research team were had around the benefit of choosing a more 'traditional, nuclear' family for this research as a 'pilot' as it had not yet been explored.

As COVID-19 started to take hold of London, the research site became a treatment centre to support those most critically affected by the virus. After meetings with the supervisory team it was deemed that my external supervisor would contact specific families that she thought would meet the criteria. She had previously contacted two families who declined to participate for various reasons. As COVID restrictions continued, I didn't want my research to put any extra pressure on staff who had been re-deployed. I reflected on how I would be able to move forward with recruitment and preserve my project, while limiting the disruption to both staff and service users. Saberi (2020) explores the need for research to continue during the time of a global pandemic and discusses the best way to go about this, which calmed my anxieties about being a nuisance to staff.

3.4.1 Participants.

The family information has been summarised in Table 6. To protect the family's anonymity, each member has been provided with a pseudonym. I contacted the family to give them the opportunity to choose their own pseudonyms, but I did not receive a reply.

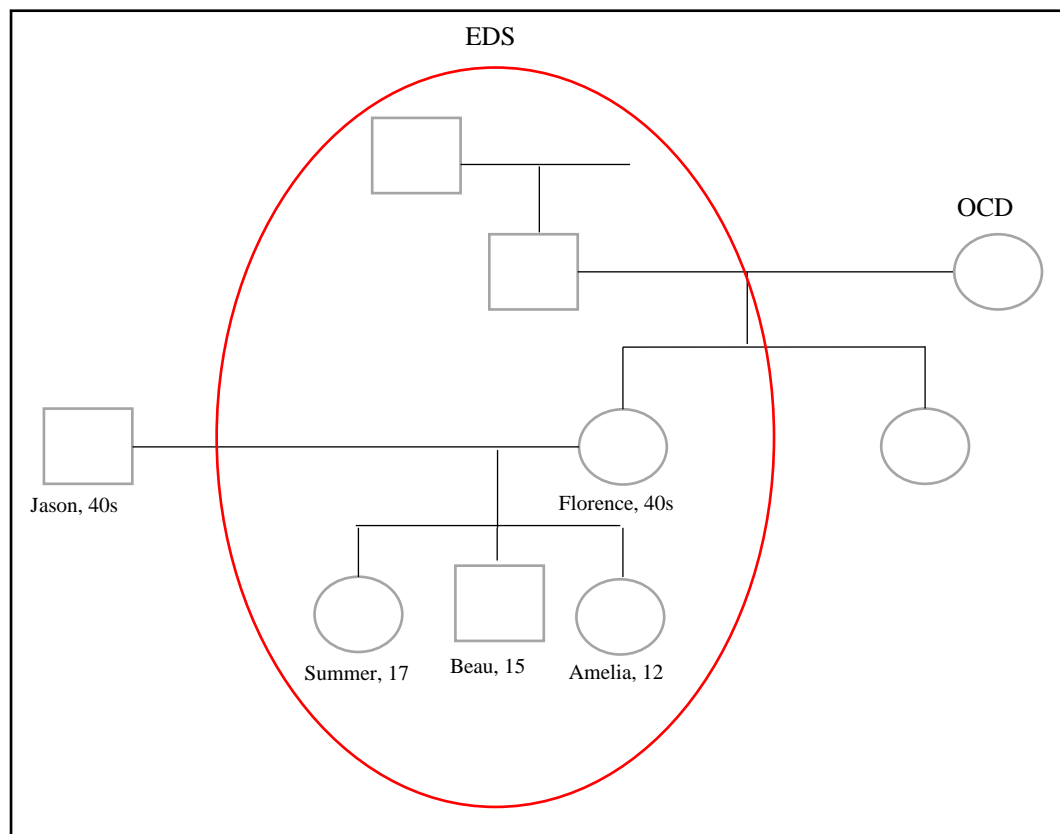
Table 6: Participant demographics

Participant	Age	Employment/Schooling	Diagnosis
Jason (Father)	40s	Business Owner	n/a
Florence (Mother)	40s	Part-Time Senior Nurse Practitioner	EDS (chronic pain, joint hypermobility)
Summer	17	Part-time student (A-levels)	EDS (chronic pain, IBS, fatigue)
Beau	15	Part-time student (GCSEs)	EDS (chronic pain & fatigue)
Amelia	12	Part-time student (Year 8)	EDS (joint hypermobility, frequent injuries, eye/visual problems)

The family appeared to be a White British/Irish middle class family. The mother and all three children have a diagnosis of EDS which is defined by the NHS choices website as “a group of rare inherited conditions that affect connective tissue” (NHS Choices, 2019).

Hypermobility EDS, the most common classification, is characterised by joint hypermobility, fatigue, joint pain, digestive problems and dizziness (NHS Choices, 2019). They were all diagnosed after Beau, who had been very unwell since birth (colic, sickness etc). He had been investigated for two years when he was given the diagnosis of EDS, after which his older sister Summer, his mother Florence along with her father and paternal grandfather were tested and the diagnosis confirmed. Amelia was diagnosed when she was roughly two years old.

The family did not express what their specific EDS classification was and on the children's clinic letters, EDS does not feature as a clinical diagnosis, but rather "chronic fatigue", "IBS" and "chronic pain". As previously discussed, EDS falls within the spectrum of hard to diagnose and often misdiagnosed medical conditions (Gazit et al., 2016). There are no medical tests, or current medical explanations for the syndromes, however, there is some heritability evidence (Genetics Home Reference, 2020). People with EDS and PPS, experience similar challenging journeys to diagnosis, similar stigma in response to their symptoms and similar experiences of being misunderstood (Bennett et al., 2018).

Image 2: Family Genogram

Given that EDS is not strictly a PPS condition I had to weigh up whether this family who all except the father, experience PPS would fit the criteria. Reflective conversations were had with the research team and a decision was made in the context of COVID-19. Although, I had originally wanted a family where one child and one parent had no diagnoses to emphasise any possible differences between the well and the unwell it was clear that my project was in jeopardy. From an ethical standpoint this family were both willing and available to participate and I was aware that excluding the family may cost them the opportunity to reflect on their unique constructions of the world. Amelia, the youngest child, was not yet a patient in the clinic and has subsequently not had any psychological interventions and thus was anxious about being interviewed by a (trainee) psychologist. I wondered if this meant that she could fall more into the role of the 'well child' I had originally planned as her symptoms were explained to me as being more injury based.

However, as the research progressed, this was not the case. Although I had some concerns about the experiences of this family being different to those who have more 'traditional' PPS conditions, with discussion it was decided that there were likely sufficient similarities in the families experience to answer the intended research question.

3.5 Procedure

3.5.1 Service User Involvement.

Service user involvement was a key aspect of the research design. A service user consultant family consisting of a mother (Gemma), and her two children, her son (Greg) and daughter (Georgina) were recruited with support from my external supervisor. Within this family both children have CFS plus other PPS conditions and the mother experiences chronic pain. The consultant family played a crucial role in carving out a meaningful and respectful way of recruiting the participant family, designing consent and information sheets. I met with the family to discuss the project design, most of the discussion was focussed on language and labels. Gemma discussed the derogatory connotations associated with medically unexplained physical symptom diagnoses, often shortened to "MUPS" or "MUPPETS" (Appendix H). This provided a brief insight into some of the experiences of living with PPS and having to navigate the healthcare system, as well as perhaps the social media perspective of stigmatised illness.

When querying the language of the working title, Gemma wanted it to incorporate words like 'chronic' or 'long-term', which while valid and appropriate I felt that they did not encompass the challenges associated with stigmatised illness. However, it did highlight the need for people with these conditions to have their experiences validated. To ensure I am reflexively carrying out this research, I questioned my choices of not wanting to include these terms in the title. I was left feeling conflicted with this use of language as I felt there were

elements of legitimacy seeking when having this linguistic conversation with Gemma. I wondered, given my relationship to chronic and long-term conditions if I felt defensive of my parent's experiences and perhaps an unspoken hierarchy of organic illness. My reflections on these issues remind me of the importance of reflexive research in this context because ethically, when conducting narrative research, the idea of whose story is it, comes to mind (Wells, 2011).

The consultant family were keen to provide biographies about their rationales for involvement in the research and asked for them to be included in the information sheet. However, through the ethics application process it was requested that this information be removed as it was deemed “inappropriate” by the REC committee. This felt uncomfortable to me and thus I have attached it in the appendices (appendix H) to honour the family's contribution to the research. I also did a pilot interview with Gemma and Georgina to review the questions, the timing and the flow of the group interview. They reviewed the language used in the interview schedules (appendix J) and we discussed the rationale for the separate group and individual interviews. I have invited Georgina to be part of any future dissemination of the results to ensure that the research continues to be co-produced (Groot et al., 2020).

3.5.2 Preliminary Stages.

Once participants had read the information sheet and had chosen to opt in via the researcher's email, further contact was made. The participant family were given the option of arranging a meeting to discuss the research further. Following the meeting, the family were taken through the information sheet and any questions were answered. Consent and assent forms were discussed and confirmed with each family member.

Once my external supervisor had given the information sheet to an appropriate family, they contacted me via email, and I arranged to telephone the family to discuss the

information sheet and gather consent. After that I sent the family the consent forms via email and offered another appointment to discuss any queries. Once they had consented, I booked an appointment for the group interview to happen on Zoom. After this, I booked individual interviews for each of the family members. I was concerned about the lack of routine and generalised anxiety that was felt by many during these times of COVID-19 and that the family would no longer wish to participate. As a result, I arranged the interviews at the family's convenience but perhaps slightly inconvenient for me.

3.5.3 Data collection: Narrative Interviewing.

The narrative interview itself creates an interactional context within which the narratives and discourse are drawn upon, or resisted (Andrews et al., 2013; Phoenix, 2013). The data collection involved meeting the family for a minimum of a 45-minute interview. This interview was conducted face-to-face with the family and was based on a semi-structured interview schedule which had been piloted with the service user consultants. Within this time, the family members were encouraged to share stories about their relationship to their PPS, to their roles within the family system and to the way they see themselves within the context of their illness and their family. This created the space for collective narratives to be uncovered about how the family as a unit make sense and meaning of their experiences. It also provided an opportunity to explore how and why the family members have drawn on specific discourses available in our socio-cultural context at the time of the interview. I chose for the group interview to be conducted first so the family felt comfortable and safe in my company. There was also an assumed comfort in speaking about the family experiences and the collective narratives first, to enable the family members to warm up to the process. Discussions were had in supervision about how we felt that it would be better to ask participants to position their individual narratives in agreement or in contrast to the collective narratives rather than vice versa.

As it happened, the interviews took place virtually in the family home due to COVID-19. I wondered and remained aware of what impact the change in the local context will have on the way the family chooses to construct their narratives. While there is an assumed comfort when someone is in their own environment, I was aware that I was 'entering', albeit virtually, their home context and this would likely have some impact on the process.

Following the family interview, each family member participated in an individual interview. The purpose of this interview was to encourage stories that may or may not have been privileged in the group interview and to reflect on any individual perspectives. I wanted to encourage participants to share other untold narratives that the group interview context did not permit to be shared or constructed. Participants were asked open questions that facilitated story telling such as “*can you tell me about a time when illness entered your lives?*”. I asked further questions that prompted more detail, further storytelling and information to aid a deeper level of discussion (Kvale, 2007; Wells, 2011). Following every interview, I provided the family members an opportunity to reflect on their experiences and ask any questions about the process. I encouraged family members to contact me over the coming weeks if they felt they wanted to discuss the process further; they did not take me up on this offer. I thanked all the participants for their time and their generosity in what they shared.

Each interview was intended to last a minimum of 45 minutes. Participants were given the option to have breaks as and when they felt necessary, or whenever parents or I as the researcher deemed breaks would be beneficial. Overall, the interviews lasted 313 minutes in total.

We had some technical difficulties given that the interviews were done online. I worried about what impact interrupting someone's speech would have on the flow of the storytelling. I toyed with how much I should interfere and interrupt the flow of conversation when technology impeded my ability to fully comprehend what a participant was saying. In

the end, I decided it was best to ensure I had understood what the family were expressing so I could respond appropriately, this meant at times I spoke over or interrupted the family members. I was concerned about how this may have disrupted both the research relationship and the way that participants were able to share their narratives. In the original plans, the research would have taken place within a hospital setting which would be associated with certain discourses that may or may not be problematic for the family, such as legitimacy narratives (Japp & Japp, 2005). However, I wondered how the development and construction of narratives were influenced given the contextual change to the family's home environment.

After the interview. Although the interviews were recorded on an audio device, it was important for me to keep field notes of my experiences of being in the interview and my part of the co-constructed experience. I transcribed the interviews as close to the interview as possible to keep the memory of the non-verbal expression alive and as close to the verbal or written experience of the interview.

Transcribing the interview. Due to the layers of processes and understanding involved in narrative inquiry, it was important to me to transcribe the interviews myself and immerse myself in the data. Transcribing the interviews was a challenge, to decide what to write down, and how much of the non-verbal or discursive tools that people employ when communicating to include. I was mindful of aspects of the conversation to which I did not give attention and wonder if video recording the interviews may have mediated what may have been missed. The interviews were transcribed by hand through listening carefully and slowly to the audio recordings (at least four times per transcript) and typed it into Microsoft Word and then imported into NVivo. I started with initial transcriptions whereby I paid attention to the words that were spoken, specifically identifying *what* was said. Then as advised by Esin and colleagues (2014) I had a second phase of transcription whereby I focused on *how* that content was spoken. Appendix K shows the transcription symbols used

to denotes changes in speech. These symbols have been adapted from Jefferson (2004). It was necessary to ensure that the transcripts were readable while trying to maintain the authenticity of the speech, to do so additional information is given by the symbols (Jefferson, 2004).

3.6 Analysis

Given the nature of this analysis, it is hard to de-lineate the processes of data collection and analysis as once the questions are asked, the narratives have already begun their construction. Narrative analysis navigates how the interactional context both informs and is informed by the co-construction of narratives (Andrews et al., 2013). Narrative inquiry does not offer any formal structure or procedure on how to move forward with data analysis, however, Riessman (2008) and Wells (2011) offer guidance with some steps to take. They advise that there are different levels within which a narrative can be interpreted; content, structure, performance and context. The process is guided by immersing oneself in the transcript by repetitious reading, and ensuring the researcher is able to take different ‘lenses’ throughout the process e.g. being able to zoom in or out depending on what layer I was reading from. I used NVivo to make notes and to “code” together themes. The different layers of this analysis will guide me to understand the *what, how, who, when* and *where* that are important to the participant’s narratives.

3.6.1 Reading for Content.

This is the initial layer of analysis, focusing on *what* is being spoken about and asking if there are any patterns or themes in the stories that are being told. It may be that patterns initially noted in one or two transcripts are shaped and transformed by themes found in subsequent transcripts (appendix L shows aspects of content for the analysis). It is important

to note that over numerous readings of the transcripts and listenings of the recording is how these themes may develop.

3.6.2 Reading for Structure.

The transcripts were re-read in order to consider *how* participants construct their stories e.g. what communication tools were used, how they started or ended stories, what they chose to tell and therefore what may have been left out as a result. When reading for structure, it is important to pay attention to “storylines” that flow throughout the participants narratives about themselves and each other that begin to inform ideas about identity. An example of reading for structure is presented in Appendix M.

3.6.3 Reading for Performance and Context.

It was important for me to read each transcript for performative elements too, in doing so, I considered to *who*, *when*, *why* and *how* they narrated their lives. When considering the *who* aspects of the analysis I paid attention to the relational and interactive element of the interview, thinking about who I am to the participant and how did that inform the way they shared their story. This draws on ideas of positioning (Esin et al., 2014), highlighting the ways that the co-constructors position themselves and each other in relation to the narrative. For understanding the *when* and *why* components of the narratives I read for context too and asked myself about the different contexts in which this co-construction sits e.g. the (inter)personal, political, sociocultural, temporal as well as discourses available to the participants. Lastly, performance and context were analysed to think further about *how* participants construct their resulting personal meaning and identities. An example of this is presented in Appendix N.

3.6.4 Presenting the Narratives.

It was a challenging process to appropriately represent the narratives of this family. I wanted to authentically present their experiences as it appears through my own sociocultural and historical lens. Each of the individual interviews (including the family interview) were analysed separately, taking note of how the different storylines and themes were constructed based on the content, context, performance and structure. After laying down the individual narratives and the themes highlighted from the group interview, I compared any similarities or differences across the narratives. Through doing this, it uncovered any overarching narratives from across the interviews and allowed me to think about how the storylines flow throughout (examples of initial stories can be found in appendix O).

3.7 Quality, Validity and Self-Reflexivity

Within qualitative research it is harder to ensure that research is “valid” and “reliable” as more positivist ideas of research would suggest, because of the nature of data collected. Constructs such as reliability and validity are underpinned by a notion of a correct and truthful result which is incongruent with a social constructionist epistemological position. Qualitative researchers, while still wanting to conduct rigorous research, interest themselves in conducting research with transparent processes and clear process rationales.

Given the temporal shift to a greater understanding of epistemological stances from a heavy alignment to the validity and reliability benchmarks of quantitative research, it seems more reasonable to focus on concepts of truthfulness and credibleness (Rose & Johnson, 2020). Trustworthiness and research rigor are also dependent on the quality and depth of the systematic literature review, the theoretical basis of the research and its relationship with the wider socio-political/cultural context (Rose & Johnson, 2020). Furthermore, I will follow structures and processes detailed by Riessman (2008), Wells (2011) and Esin et al. (2014) to further provide clarity and transparency of my intended procedures, the way I understand

how narratives and knowledges are produced and the way in which meaning can be attributed.

As seen above through my use of my service user consultants, I have endeavoured to ensure that I am remaining reflexive throughout. I have kept a diary throughout this process of my MRP to check in with myself and challenge my own biases and assumptions (appendix P). I challenge myself through conversations with peers and supervisors and readings of draft writing with supervisors to ensure that my processes are transparent throughout.

4. ANALYSIS & DISCUSSION

4.1 Overview

In this chapter, I present a summary of the main storylines as told by the family across the interviews. I then present the wider overarching narratives that are drawn on or resisted across the interviews. Finally, I discuss how the family members construct and negotiate the dynamics of their identities and roles within their family system. Throughout this chapter, ideas of positioning and how the family appear to draw on wider social discourses associated with PPS is discussed.

As noted previously, pseudonyms have been used throughout for the family members for example, Florence and Jason, both in their 40s are parents to Summer (17), Beau (15) and Amelia (12).

4.2 The Family Interview

The family interview was my first opportunity to meet these participants. It lasted 70 minutes in total. The interview took place on Zoom with Summer and Jason sitting together on one device, Amelia and Florence together in a bedroom and Beau on his own in his bedroom. These physical pairings mirrored strong relational pairings within the family.

The family engaged me in a journey of humour and sadness, presenting a shared family cohesiveness, with me positioned as the outsider. From early in the interview, this mirrored the stories that they told later of the need for families living with PPS to be strong in the face of potentially difficult interactions with HCPs.

Overall Impression

The interview began with the family orientating themselves to me and vice versa. This was done using humour and comedic facial expressions when Jason introduced his wife and youngest daughter as “*two little girls together (.) they'll do more talk than anybody else*”.

Early in the interview, Florence appeared to position herself as the protector of the family against potential outsider intrusion. In response to a mistake I made when introducing the study, Florence swiftly corrected me, taking an educative position, while protecting her family against the threat of being misunderstood:

K²: “there are certain symptoms that are very specific to EDS but then there are some symptoms that are more (.) sort of (.) chronic fatigue (.) chronic pain (.) type (.) symptoms (.) is that your understanding(?)

F: NO

K: No (2) okay (.) what was your (.) what (.) are kind of some of the common symptoms that you all experience?

F: I would understand the chronic pain (.) comes from the joint issues that comes from the EDS

K: Okay

F: I suppose and then I understand that their fatigue is is caused is part of the EDS”.

² Initials used: F for Florence, J for Jason, B for Beau, S for Summer, A for Amelia, K for Kimberley, Group for ‘group interview’ and Individual for ‘individual interview’

Group: 03.38 – 04.24³

Florence's protective position continued when she took a mocking tone in response to my initial question:

"K: [...] can you tell me something broadly about how you are as a family?"

F: (6) In wha- how are you as a family in relation to (1) WHAT↑ (.) maybe make it more specific to start us off (.) with maybe (.) .Hhaha

Group: 6.59 – 7.24

Her response to my first question may not only be a challenge to an outsider, but also a demonstration of her role in protecting her family. She continued to affirm this position by using humour to protect her family from difficult interactions. After this, a series of responses set a tone of cohesiveness amongst the family unit. Throughout the group interview laughter was shared amongst all family members, except Beau. The family stated and showed their closeness by demonstrating warmth to one another with looks and gestures. Near the start of the interview, however, Beau appears to position himself contrarily and challenges the family unity narrative by stating *"there is a bit of friction though sometimes isn't there"*. He is the only male and the middle child, so he may draw on narratives of marginalisation within his own family when he challenges the family unity narrative.

Later in the interview, soon after he challenged the family unity, the family demonstrated their closeness to me by laughing and pointing at one another when describing who suffered most from tiredness:

J: "the end of school terms always become a little bit more difficult

F: so tiredness is creeping in (.) symptoms get worse when they're tired (.) so they have less=-

³ These numbers denote the timings and the interview in which the quote took place

B: =actually by the end of the day (.)like when we're doing the dishes and stuff like that that's probably when we mainly-

A: [Amelia gestures to Beau, Florence laughs] him(!)

F: HAHHAHA! that's your moment isn't that Beau↑

B: Yep (.)

S: .hahaha (.) and Amelia's(!)

F: [...] Beau's particularly .hah

A: I'm more in the daytime (.) I annoy you guys in the daytime”.

Group: 11.34 – 12.07

Both Jason and Florence drew on narratives of parental protection. They narrate their experiences with their children's schools in the early days of diagnosis and position themselves at the mercy of an ignorant enemy:

“J: there is ignorance was out there (.) to be brutally honest (.) so that was difficult (.) so dealing with (.) education was very hard (2)”.

Jason (group): 20.11 – 20.30

This set the tone for the children to tell stories of their difficult times at school. This was initiated by Summer, who laid the foundations for talking about their struggles. She narrated that she was:

“S: not feeling same (.) level as everyone else (.) so having to do EXtra anyway (.) trying to keep up but to also catch up has been °quite difficult° [...] I would never ask the teachers (1) I kind of don't trust (#) in teachers to help me”.

Summer (group): 23.13

Summer appears to struggle most with friendships which she directly attributes to her illness and specifically the stigma associated with it, she reports that she:

S: *never quite formed (.) a proper friendship (.) erm and as always bouncing between different friendship groups or different people and they're never quite right (.) they've never quite believed↑ that I was ill (.) because when you look at me (.) I look exactly the same as everyone else erm I just don't have anything to show that I'm ill".*

Summer (group): 25.13

Summer appears to draw heavily on narratives around the perceived illegitimacy of PPS conditions here and the consequences on social relationships. After this, Beau followed, when he positioned himself in quite a passive way in relation to friendships:

B: *bouncing through friendship groups is the exact same it's like because you're ill so much it's like (.) you're not there to make those kind of friendship groups (.) so it's like (.) you pretty much just cling on to whoever's gonna talk to you".*

Beau (group): 26.17

As they had begun to speak in age order, Amelia followed her older siblings to explain her experiences with school, she narrated her difficulties with schooling and friendships where she stated:

A: *when I was in school (.) [...] I didn't really talk to many people (.) I DID talk to people (.) but they didn't really talk back or they would (.) they used to ignore me".*

Amelia (group): 26.37

After this, Beau appeared to find it important to move away from friendships and discuss his experiences and his struggle with the school staff too. He reported:

B: *every lunchtime I'd be taken into this little room with the head teacher and the assistant head (.) and I'd almost get interrogated (.) like (.) every lunchtime (.) and that was terrifying (.) I actually remember that (.) I always thought I did something wrong (.) cause I'd just be sitting outside (.) they'd leave you there for ages (.) then it*

was like (.) anytime I did something wrong (.) it's like they went to the nth degree with it [...] so it was almost like there's a bit of bias going on—”.

Beau, (group): 31.24 – 32.53

In amongst Beau's storytelling about difficult times, Florence tells a story about her challenging interactions with the children's school.

F: “we had a big CAF meeting at school erm explaining the condition and what was wrong with the kids and following that admission erm meeting (1) the head reported me to social services for fabricating a medical condition (.) so we were investigated but were basically thinking (.) I had Munchausen's”.

Florence (group): 29.56 – 30.30

She used her talk to show how difficult her situation has been and again drawing on the ignorance of others that Jason spoke to earlier. This fits with a narrative of having to protect and defend her family, while also letting me know I needed to take the family and their stories seriously. Florence told these stories quickly to move away from her experiences of mother blaming and to perhaps shift blame to external parties. She narrates and mimics the authority of the school when the head teacher recommended that Summer “*EAT A BANANA(!) EAT A BANANA(!) YOU'LL GET ENERGY(!)*”. Her experiences echo stories told by other mothers of children with PPS (Morris & Ogden, 2012).

Later in the interview, Florence seemed conscious of her position as a mother and appeared to want to protect her children from hearing about her poor health. She appeared employ humour to protect her family and detract from the stories told of difficult times for the family:

F: I think because of surgery and the pain like makes me quite tired but =yeah (.) literally pain in the arse (.) I have pain in my arse [hahaha]

J: =*pain in the arse* [hahaha]

A: [hahaha]

S: [hahaha]

Group: 37.44 – 38.00

These social difficulties, the ideas of not being believed to be ill and the need to defend and protect one's children draw on wider narratives commonly drawn on by PLWPPS, whereby the lack of understanding associated with these conditions leave PLWPPS having to legitimise their illness with their talk.

4.3 Individual stories

4.3.1 Jason's Story.

Jason is the father of the family. He is in his 40s and a business owner. He spoke of his Irish heritage with warmth and passion and spoke about the importance of hard work. The interview took place in his home. I was struck by the beauty of his home and as he sat in front of large floor to ceiling windows that looked onto a large garden, I was reminded of his role as the successful head of a company and the family provider. He did not appear concerned about confidentiality in what appeared to be a communal space.

Across the interviews Jason appeared to speak freely in front of his children but used his talk in his individual interview to disagree with things said elsewhere. Throughout his interview traditional gender roles were heavily apparent. When booking the interviews with Florence, she made it clear her husband was very busy and I had anticipated that he would not be engaged in the process, yet he appeared to speak thoughtfully and generously throughout. The interview lasted 56 minutes. Across the interviews Jason's use of humour is noted which he attributed to his Irish heritage ("*obviously from an Irish connection .Hehehe*").

Overall impression

When Jason was asked to reflect on the group interview, he drew on his experiences in hospitals:

J: *“I have been involved in similar (.) group (.) therapy (.) discussions in the past with some of the erm hospitals”*

Jason (individual): 3.32

From the outset, he took an educative position informing me of the process of these groups, their benefit and the prestige of being in London hospitals. Jason commented:

J: *“the parents [...] have their defence shields up to some extent (.) not really wanted to give a lot of way (.) but then towards the end it became more open”*

Jason (individual): 3.50

This part of his talk mirrors my experiences of interviewing Jason and Florence; appearing ‘defensive’ at first and more open towards the end. While discussing his experiences with hospitals, he appeared to draw on wider discourses around parental roles in their children’s healthcare, sometimes being positioned as lacking in power and voice, to other times becoming experts in the healthcare system (Peters et al., 1998).

From the offset Jason prioritised a story of busyness which appeared to draw on wider narratives on achievement and masculinity:

J: *I::: like to work out a lot (.) I like to be busy (.) so as long as I can keep busy then I can (.) I can that's much easier to handle”.*

Jason (individual): 8.17

Jason appears to use this talk of busyness, motivation and positivity to move away from the perceived sadness of his children not being able to achieve what he hoped for them:

J: *Summer (.) for example (.) was a really good swimmer-*

K: *Mm-hmm*

J: *and then all of a sudden (.) she (.) you know (.) she was probably good enough to go into competition at one period (.) and then that just stops.*

K: *Yeah*

J: *you know (.) Beau (.) if he was erm healthy (.) he (.) he would be exceptionally good at sport-*

K: *Mm-hmm*

J: *[...] so that can be tough sometimes”.*

Jason (individual): 13.27 – 14.20

Jason talked about his desire to avoid negativity (“*Florence (.) we both of us like to stay or we just be positive*”). He tells a story of doing what he can to avoid inviting negativity into his and his family’s life:

J: *“if anything else comes in the negative (.) negative thoughts or negative feelings (.) they don't do you any good so you have to find a way of just dismissing them as much as you possibly can”.*

Jason (individual): 14,55

While drawing on narratives about the costs of giving into negativity, he explored the idea of future consequences of not keeping busy and motivated, when he said:

J: *“ I think we're becoming more idle I just feel that you'd lose motivation to some extent↑ you'd start erm (.) you could start going down an alley that wouldn't be very nice [...] I'v- I'v- have spent my life dismissing it (.) not going there”.*

Jason (individual): 16.30

Jason told these stories about negativity and losing motivation with apparent discomfort and spoke quickly to move the story along. He appeared to use ‘negativity’ as a euphemism for mental health issues and positions himself as an adversary to them. This draws on old fashioned narratives around mental health being a sign of weakness and

something to be feared and avoided. However, in and amongst this talk, he acknowledges the struggle of being in his shoes by stating:

J: *“when you're alone with kids and a wife that's not well (.) it's very easy for you to start feeling unwell”.*

Jason (individual): 15.59

He appears to be resisting narratives of male strength and gender stereotypes and later validates his own strength towards the end of his interview, when he acknowledges what other men would have done in his position:

J: *“I'm pretty sure a lot of men in my position would probably have got up and walked long time (.) ago hundred percent sure of that”.*

Jason (individual): 51.44

However, while Jason discussed the idea of it being easy to “*start feeling unwell*” and illness in his family, Jason previously narrates himself in a passive position in relation to his own health. His wife and children’s health are prioritised, and his health is either dismissed (“*I've gone to work with colds*”) or attention is diverted back to the other family members:

J: *when you don't feel well, then you understand (.) and then you think yourself (.) JESUS now (.) I understand how YOU DO feel”.*

Jason (individual): 18.47

This is affirmed by the other family members’ view of Jason’s health, for example:

B: *“but when it's my dad (.) it doesn't really make much of a difference”*

Beau (individual)

F: *“Hhhe doesn't really get unwell [...] Jason, Jason, when he got unwell (.) what is he like↑ (I) he had a kidney thing [...] I don't know, it's a difficult question to answer not a lot really”.*

Florence (individual)

The paradox of an individual's health being dismissed juxtaposes the experiences of the rest of the family and puts the unwell family members in a more powerful position, a scarcely felt position by the unwell members in the wider societal context.

Given how he has been positioned within his family, Jason appears to distance himself from the story of illness and prioritises a story of becoming parents as the greatest change in his life:

J: "Do you understand that (?) it doesn't matter whether in (.) erm good health or poor (.) every relationship changes when children come along [...] when kids come into your life and then from there (.) then then then when illness comes along [...] that's the big change for me".

Jason (individual): 23.01

While at times it felt that Jason was taking an educative position as he finished several sentences with "*do you understand that(?)*" but on reflection, it felt that his educative position was more parental, as if he was guiding me, as someone younger than him.

4.3.2 Beau's Story.

Beau, a 15-year-old boy, has a part-time school timetable but does more school hours than his sisters. The interview took place in Beau's bedroom where he didn't appear to have any concerns over confidentiality. During the interviews, Beau appeared talkative and keen to share stories about school, family and illness.

He appeared to speak thoughtfully and generously, as was demonstrated by frequent short pauses and phrases like "*what'd you call it*" in which he appeared to choose his words carefully. He seemed to be more comfortable telling tales of overcoming others and perhaps less comfortable with stories where he was positioned as vulnerable. I noticed that throughout the interview it felt like I was talking with someone older than 15-years-old, however, he also felt like a peer. I wondered if the sense of loss he described due to missing out through illness

reminded me of my own experiences of being hospitalised at 15 years old. The interview lasted 56 minutes.

Overall Impression

Once Beau had been asked if he had any questions about the study, he began his storytelling reflecting on his experiences from the group interview. He talked about the group interview as being a positive experience of reaffirming him and his family's ability to overcome. He expressed:

B: "you (.) kind of realise when you're going through it how much stuff you kinda overcome with your family".

Beau (individual): 1.31

Beau then tells a story of competition in which he compares himself to other children with EDS, his sisters and his peers, positioning himself with increasing favour with each story told. When he compares himself to other people with EDS, he appears to position himself a frightened character who must overcome a powerful force:

B: yeah(!) and it was almost it was almost terrifying because I was like (.) I had to try even harder because I was like (.) if I don't do this (.) then I might end up like them [...] I just knew that (.) you know (.) they had the same problem as me and like (.) almost (.) if I didn't keep up with them (.) then I would almost be in a wheelchair when I was olde- (.) and I was SHITTING MYSELF".

Beau (individual): 18.12 – 18.37

His narration of this difficult encounter with other children with EDS is laden with fear about a prospective future for his illness (Berglund et al., 2000; Bennett et al., 2018). This set the tone for him to continue his narrations of self-comparison, this time with his

sisters, particularly Summer. When asked about his experiences of being unwell with EDS in his family, he appears to position himself in a superior position in relation to his family:

B: *“my (.) EDS isn't as bad as the girls (.) so it's like (.) I try not kick up as much as fuss unless like (.) I'm really bad”*

B: *“erm (.) I almost feel like sometimes my sisters er (.) look up to me (.) sort of because even though I'm not older than Summer [...] I can sort of do more”.*

Beau (individual): 22.43 & 33.37

As his story progresses, he takes an educative position informing me about boom and bust cycles (*“have you ever heard of (.) erm (.) booming and busting[↑]”*). Later, he continues to draw on narratives around competition and masculinity, when he compares himself to his peers:

B: *“when I look at them (.) I don't really see a difference (.) and (.) you know (.) if I ever start comparing myself to them (.) then I think I'll just get myself down [...] .Hheheh it quite surprised me (.) out my friends (.) I'm probably the most active!”.*

Beau (individual): 38.26 – 38.45

It appeared important for him to position himself as just the same, if not better than his peers. He proceeds to tell a story where he is positioned as overcoming challenges and prevailing through his own hard work. He specifically draws on this narrative when he discusses his teacher's expectations of failure. He begins with:

B: *“I always find like struggle with teachers because like (.) I know (.) sometimes I might be ill for a couple of we- (.) sometimes it can be months (.) and they sort of give up on me (.) so it's like I have to (.) almost like SHOW them they SHOULDNT GIVE UP ON ME and I can do as well as anybody else[↑]”.*

Beau (individual): 39.34

He continues to demonstrate his prevailing in the face of adversity when he described a further interaction with his teacher:

B: "my teacher telling me (.) "there's no way you can do this subject (.) if you did this subject you will fail" (.) and because of that (.) that made me want to do the subject even more .Hahah (.) so I did it" [...] I got a level seven which is [...] the equivalent of an A in the old grades (.) I just really enjoyed how shocked he was when he when he was talking to you (.) as "I tell you what if I told myself today (.) I had no idea that you would be it I would tell myself that you'd be getting a (.) a U" (.) and I was like (.) "oh!" [...] there was nothing he could do to say that take credit for what I'd done (.) it was all me!".

Beau (individual): 40.29 – 41.32

In this story he is the unwell child, positioned in a 'one down position' challenging the powerful teacher. This resonates with the underdog tale, when he demonstrates how he has overcome the 'powerful' enemy and proved his teacher wrong. This also draws on wider discourses that people with PPS must navigate in terms of legitimising their experiences, fighting for themselves which in Beau's case was focussed on proving his ability, where healthy students don't have to work as hard.

Overcoming the expectations of others, permits the moving away from the position of a sickly young boy. This story also draws on prevalent cultural discourses of what it means to be an acceptable, successful young man, mirroring themes of prioritising work over health expressed in Jason's individual interview:

J: "I'm so busy in my work [...] work would probably have to get bit of a back step (.) if they were much healthier".

Jason (individual):12.41

In line with wanting to overcome others' expectations, Beau draws on narratives of self-reliance, "*whenever I'm not feeling well (.) I almost want to do it I almost want to overcome it on my own*", which appears to speak to wider narratives around independence for teenagers reaching 16 years old and counters narratives around family unity (discussed later) (Rosland et al., 2012).

4.3.3 Summer's Story.

Summer was 17 years old at the time of the interview. The interview took place in her bedroom, she didn't appear concerned about any issues of confidentiality. Summer appeared more comfortable telling stories about her family, and less comfortable with stories where she was positioned as the protagonist.

Summer appeared engaged in the process of the interview; however, it was the shortest of the interviews, lasting only 36 minutes. While this was the same length as Amelia's interview in duration, it felt shorter as she did not tell lengthy, detailed stories. She provided quite brief answers, which made me wonder whether she had positioned me as a threat. Summer's family told me that she had suffered with her mental health and I anticipated she may have been more open to interacting with a psychologist but perhaps her previous experiences had not been positive.

Overall Impression

When I first met with Summer, she was asked for her reflections on the group interview, she responded similarly to her father:

S: "*we did have a family (.) meeting once with the hospital (.) and so it was kind of similar to that (.) really*[↑]".

Summer (individual): 1.29

From the beginning Summer positioned me as an HCP and I felt some resistance from her throughout the interview. This appeared in response to requests for more qualitative

information about her experiences and her family. She appeared to be protecting herself and her family from perhaps intrusive probes. This draws on narratives pertaining to difficult interactions between HCPs and PLWPPS (Chew-Graham et al., 2017; Stone, 2014a & b).

Like her brother, Summer drew on narratives around (a lack of) achievement, however, she provided a counter narrative to her brother's position of superiority by positioning herself as inferior and not good enough in comparison:

S: *“sometimes I do feel like (#) I'm a (#) bit more um sort of lazy than everyone else in the family [...] like compared to Beau(.) and I always feel bad that he's pushing himself through school to do full days and stuff (.) and I'm not”.*

Summer (individual): 4.14 – 4.36

The need to resist narratives around laziness appear to be more pertinent to Summer. Laziness narratives may be more damaging for woman (especially those with PPS) as they draw on wider discourses about the contribution of unwell people and women in society (Paxman, 2019).

From the offset, Summer positioned herself as an adversary to illness and resisted a strongly held illness identity when she favoured the word “different” rather than illness. She seemed to draw on wider narratives about people with PPS being “different” to healthy children. She stated:

S: *“I really noticed that I was properly unwell (.) not necessarily unwell (.) but kind of more different”.*

Summer (individual): 6.26

The use of this language is consistent across all members of the family. ‘Different’ positions the person as less vulnerable and perhaps incites an element of choice and control over their difference rather than being passively unwell and out of control. Summer goes on

to describe the social consequences of being different, whereby she positions herself as a victim in relation to other teenage girls:

S: *“I think the stress of friendships (.) I wasn't very (.) I wasn'- (.) having a very good time and I did have a girl picking on me (.) but yeah- [dismissive tone]”.*

Summer (individual): 8.10

This appeared to be a challenging time for Summer to reflect on and she doesn't allow the conversation to stay here for very long. Her dismissive tone prompted me to move the conversation on to asking about other experiences of being unwell.

She then considers the importance of age order within her family. She prioritises the story of illness over the story of age order signifying that prior to illness entering her life, her childhood was unremarkable:

K: *can you tell me about another time that you were (.) [...] say (.) different (.) you were unwell*↑

[...]

S: *I was obviously the first child I was and there wasn't really anything much going on with me (.) I don't believe that was really significant”.*

Summer (individual): 8.22 – 9.33

Later, alongside this prioritisation of the illness story, Summer positions herself as responsible in relation to her siblings and illness when she says:

K: *Hmm-mmm (.) and do you notice that there's been any change over time?*

S: *I think it's probably more stressful for everyone (.) because obviously (.) I'm sort of a trial run in a way (.) you know (.) because I'm the oldest so kind of as sort of (.) not that Amelia and Beau will be the same bu- (.) to kind of get a grasp of what may happen”*

Summer (individual): 11.23 – 11.48

Here, she offers a counter narrative to her earlier thoughts around the relative (un)importance of being the first born and perhaps what expectations and losses have come with this position. Summer is then drawn back into a story of family unity when she is asked about her fluctuation in her illness:

K: when your symptoms are bad and feel unmanageable, what's that like to be part of your family?

S: erm (#) I'm not really sure (.) when everyone's having a bad time (.) it does affect everyone else (.) it does have a domino effect (.) maybe not quite so obvious (.) erm but when one person goes down (.) it is notice- (.) is you know it's noticeable (.) erm and I think (.) we all struggle with that”.

Summer (individual): 16.17 – 16.52

She draws on narratives of family cohesion in the face of fluctuating, uncertain illness. While Summer speaks to a family unity, she also narrates the consequences of this closeness in terms of the ‘domino effect’. When she alludes to their struggles, she is resisting narratives around staying positive when things become difficult which seem to be heavily drawn upon by other family members:

J: “You just got to stay up (.) you have to be positive”

F: “You kind of- keep positive and functioning”.

However, after she demonstrates the consequences of being so close as a family, she counters her own story by ensuring that the interview comes to an end on a more positive note:

S: “we (.) I have a very close relationship with the whole family” and “we are quite open with each other so (.) we do kind of talk to each other about things”.

Summer (individual): 31.10

This mirrored her father's need to present the positive. She appeared keen to demonstrate this closeness, perhaps to position me as an outsider, protect her family, and justify her more closed off demeanour in the interview.

4.3.4 Florence's Story.

Florence is the mother of this family. She works part-time as a senior nurse practitioner. She is in her 40s and was interviewed in the kitchen. She did not appear to have any concerns about confidentiality while conducting the interview in a communal area. Florence was diagnosed with EDS when her son Beau was diagnosed at 2 years old. She appeared more comfortable telling stories about her active family lifestyle and appeared less comfortable talking about the challenges of her own health.

Florence appeared engaged in the interview and the process of research, but she questioned whether her answers would be useful. She appeared to be drawing on her experiences of not being taken seriously by HCPs and wider narratives of the invalidation of people with PPS. The interview was 59 minutes in total.

Overall Impression

Florence began her story in response to being asked for her reflections on the group interview. She, like Summer, appeared a little resistant at first and described finding the group interview unremarkable. However, when asked about *her* story of illness, the answers appeared more free flowing which may have been a resistance to her maternal identity as I imagine, most conversations she has are about her children.

From the beginning, like her children, Florence draws on narratives of being different rather than unwell. This appears to draw on wider discourses around the lack of medical acceptance of PPS. She explained:

F: *“I don’t think that I (.) actually realised there was anything wrong [...] obviously I’ve had this all my life and looking back (.) I can see how all the way through my life (.) I have been different”.*

Florence (individual): 1.51

However, as Florence explores this storyline, she highlights that she only knows this difference through hindsight, whereas in the past she narrates her difference as through choice. She describes coming to terms with her illness, however, she explains the turning point in her narrative was when she started to fall.

“F: you just learn to manage around it (.) which I did erm but I was (.) the three kids three pregnancies that did take a toll at that point (.) I was I think Amelia was probably three maybe four at that point (.) and I just thought well this is how it's going °to be° (.) and then I started falling-

K: oh gosh(!)

F: well, actually I fell learning to ski [Hhehehe]

K: Oh, wow, lovely [Hheheheh]

Florence (individual): 4.40 – 5.01

Florence’s talk about falling was for her, indicative of worsening health. It appeared important to Florence that she not be a victim or pitied as when I reacted to her falling, she lightened the mood with a humorous comment about learning to ski. This is consistent with how Florence drew on narratives of humour to lighten the mood in the family interview.

Later, Florence begins to discuss when she first noticed she was different to her peers:

K: you mentioned [...] you noticed you were different what were some of the things that you noticed?

F: [...] doing my a-levels with my friends and we'd all go out to the pub on a Friday night (.) and they would go out on a Saturday and a Sunday afternoon (.) well I'd

pick one [...] I wouldn't- because I was just too tired (.) but I wasn't feeling (.) I was too tired and ° “oh you know” ° (.) I was just thinking “oh (.) well I had a great night last night don't fancy it tonight””

Florence (individual): 6.53 – 7.50

Florence appeared to use voicing of a past self to explain her actions to me, to convince me she wasn't missing out because of illness, it was through choice. I was drawn to and felt saddened by the parallels between her telling this story and stories of Summer, who at the same age, does miss out because of illness. She appeared to be drawing on narratives of maternal protection of Summer by proxy, as demonstrated in the group interview.

As the interview progressed Florence appeared to juggle her professional, illness and maternal identities. She positions herself as knowing what to do when she describes an interaction with a doctor who wanted to place a stoma for Summer:

K: how do you think being a nurse has informed the way you understand what's going on?

F: I talk to a doctor with an ins- in a manner where I'm listened to (.) you know (.) I'm used to I was (.) I'm a senior doc- I'm a senior nurse [...] Summer wasn't getting better (.) and he said to me (.) I think it's time that we give her a colostomy [...] knowing what I know about the condition (.) I just thought (2) “this is not going to be a temporary fix its going to be a permanent colostomy” [...] so I challenged him in a nice↑ way (.) [...] he sat there and he went "Yes (.) yes (.) rather (.) yes (.) I think you're right" [puts hand on chin and strokes it mimicking the doctor][...] at the same time (.) a family (.) who actually live locally [...] (.) the daughter had the same doctor (.) and he went with a colostomy for her (.) she has (.) been peg fed (.) stoma (.) she has a supra pubic catheter (.) she is bedridden and she cannot swallow (.) swallow and everything is gone”.

Florence (individual): 17.22 – 20.26

At this point of the interview, Florence uses several rhetoric devices to counter how she was positioned by the doctor in this narrative in terms of the importance of her being taken seriously. This draws on the lower positioning that she would have faced based on common narratives about women, nurses and mothers. She uses voicing and mimicry to undermine the doctor's taken for granted authority and to reaffirm her own authority as a mother, a medical professional and someone who 'knows'. She goes on to strengthen the narrative that doctors don't always know and validates her own authority when she elaborates about consequences that the other family faced.

Later in the interview, I asked her to tell me about her experiences of being a mother with unwell children. She appeared to take a critical positioning of others, specifically other mothers and friends when she said:

F: *"I do find myself getting irritated with mothers who have kids who don't have (.) health problems"*

Florence (individual):31.31

F: *"I find that I very (.) carefully have (.) picked my friends very carefully over the years [...] when I look back, I think "Yeah (.) you were a bit-"*

Florence (individual):31.39

Within the interactions she described she draws on narratives about the importance of social relationships and how people with PPS who already feel marginalised because of their illness, must choose their social relationships carefully.

4.3.5 Amelia's Story.

Amelia is 12 years old. The interview took place in her bedroom but was set up on Florence's phone. The interview lasted 36 minutes, which was the same length as Summer's however it appeared to flow much more smoothly, felt longer and more relaxed, as she

appeared more willing to share her stories. Amelia appeared quite shy in the group interview, which was also commented on by Beau and Florence, to be out of character. However, during her interview she appeared talkative and engaged in the discussion. Like Beau, Amelia had an air of maturity about her beyond her 12 years, but it appeared that she carried a lot of sadness about her experiences. Amelia was born into illness and I wondered about what choices she had regarding being ill, was she destined to be unwell either way?

Overall Impression

Amelia began the interview by reflecting on her experiences from the group interview, expressing that she was appreciative of how her family see her role:

A: *“yeah (.) um:: (.) I think that was um quite nice (.) to know that I have quite a nice role within the house”.*

Amelia (individual):2.23

Amelia then began her storytelling by positioning herself as knowing no different as she is the only child to be born post-diagnosis. Although not discussed, the context of her talk is associated with challenging wider discourses of parents having children when they are genetic carriers for illness and the consequences this has for the child (Kelly, 2009). She positions herself with having confidence and expertise on the topic given it is all she has known. She stated:

A: *I was kind of when I was kind of born (.) I never really knew any different from having the illness”.*

Amelia (individual): 3.36

She proceeded to detail her relationship to her EDS and how her story of illness is inseparable to her birth. Within her own story, she positions herself in a lesser position than her peers:

A: “so I just found that I was energetic and that would last about (.)half an hour and then I'd just go (.) °flat° and I was tired”.

Amelia (individual): 4.28

She offers a counternarrative to her brother's story of overcoming, whereby, she draws on a narrative of inevitability of succumbing to illness. Not long after, she then appears to reach a point of acceptance that this illness is a part of her life:

A: “it kind of its like so it's like a shadow it follows you everywhere (.) but it not really like a- it's (.) I don't think it's that big part (.) I mean (.) it's just there”.

Amelia (individual):5.50

Amelia draws on narratives of acceptance of a life-long condition, while this is a common experience for people with PPS, it also speaks to Amelia's maturity. She doesn't make any mention of hopes for a better future, which continues to draw on narratives about futurity and uncertainty of the future. The notion of the “shadow” however, applies a dark filter over this acceptance, drawing on narratives of learning to live with but never being free from illness, commonly felt by people with chronic illness (Bluebond-Lagner, 1996).

Despite it being ever-present in her life, she draws on narratives of lack of understanding about her illness, which counters her earlier confidence about understanding EDS:

A: “didn't really understand it (.) so I realised then that I needed to learn more about what I actually had so that I could understand it”.

Amelia (individual): 11.43

The narratives she appears to be drawing on here mirror the wider discourses and lack of understanding about PPS. She spoke of the need to be educated and narratives of professionals not understanding PPS. This appeared to be her positioning me as also lacking understanding and demonstrated the importance of me educating myself to understand her.

This offers a counternarrative of other members of the family taking an educative role with me, whereas Amelia, appears passive in terms of teaching me.

Towards the end of the interview, she continues to maintain the passive position when explained her role in social relationships external to the family. However, she appeared proud when she tells her story of her role in the family as joy-maker:

A: *“I make sure I try to make sure that everyone's quite happy (.) I think I'm quite talkative and I make sure that it's never quiet (.) .hha so if there's like a minute of silence (.) I always find something to do”.*

Amelia (individual): 27.30

However, she narrates how this role comes at a cost as the family struggle when she is not feeling well:

A: *“they usually say that house is too quiet and they're always like trying to find ways to make sure (#) that I get well soon so that I don't um (.) the house is a bit too quiet for too long (.) because they always say it's depressing if I'm not talking around everyone”.*

Amelia (individual): 28.50

While Amelia discusses the important role she has in maintaining the family wellbeing, she positions herself as someone with a lot of responsibility. However, only when she described her relationship with her siblings, did it feel like I was interviewing a 12-year-old child. She commented:

A: *“but then if my brother gets annoying (#) me and my sister get really quite grumpy with him and we're a bit like (.) just ignore him (.) then he will keep on doing it because he finds it funny[↑]”.*

Amelia (individual): 30.08

Having been the youngest sibling in my own family, this resonated with my experiences of feeling like a nuisance and perhaps, meant that I was more drawn into this story. At this point of storying her sibling relationship, Amelia, drew on narratives of being ‘the lucky one’ and having an easier experience growing up compared to her siblings:

A: *“I got it always a bit easier than my siblings when it comes to say for instance when they went to primary school and the teachers were horrible to them and stuff”*

Amelia (individual): 33.09

She appeared to draw on narratives of humility and demonstrates implicit gratitude for benefiting from her parent’s experiences and mistakes. She draws on wider discourses of PLWPPS having it easier than others, or being lazy, due to not meeting societal pressures to be functional like ‘healthy’ people. Positioning oneself as lucky and grateful that their situation is easier than others seeks to contradict common rhetorics about PLWPPS being frequent ‘complainers’ (Dwamena et al., 2009). When she is positioned as needing to be grateful, she appears to respond by taking up the role of bringing joy to her family.

Throughout my time with Amelia she used voicing and reported speech when speaking about her experiences with friends, often changing her tone when describing the way other children have spoken about her and her illness. When she told the story about having it easier than her siblings, her tone was a mixture of sadness and relief, however, I found myself feeling sad for her. Through personal experiences of my own and my parent’s illness growing up, I was aware that I was drawn into similarities between myself and Amelia.

4.4 Wider narratives

So far, I have presented the individual storylines. I now present the wider emerging narratives that are drawn upon across the interviews and present in line with the research aims. These interpretations are presented through the researcher’s lens and are considered

with the local and historical contexts in mind to reflect the researcher's epistemological position. Across the interviews three main collective narratives were primarily drawn on: *"They can't have that type of a life"* – Stories of Loss and Sacrifice; *"The bond between us, it doesn't really falter"* – Stories of Family Unity" and *"We are ill...I can do more"* – Stories of Collective and Individualistic Language.

4.4.1 *"They can't have that type of a life"* – Stories of Loss and Sacrifice.

Each family member draws on the idea of loss and sacrifice in different ways. This story is most frequently drawn upon by Jason and Florence where they appear to lament what their children and family life could have been without illness. However, most of the family talk about loss and sacrifice, to the extent that a counternarrative emerged from Jason (towards the end of his individual interview), who in keeping with his more general tone of wanting to keep things light, emphasised that alongside loss and sacrifice came a lot of gain (*"like anything in life (.) that sacrifice is worth giving (.) then it's worth to you (.) it's worth doing*).

As mentioned, Jason seems to ascribe the greatest change in his life was when he became a parent, however, he narrates the story of loss throughout his interviews. In his individual interview, he reflects on loss in his current life in relation to his own youth:

J: *"I suppose[↑] the only thing I could relate to back (.) come back to my own youth (.) my own childhood and then obviously (.) if I had been in the family (.) we were all reasonably healthy"*.

However, the "reality" of his situation is never far away when Jason tells his story, as he states: *"For our kids(.) they're not (.) so I feel to some extent (.) it's °kind of tough° (.) knowing that they °can't have that° type of a life"*. It is common for PWLPPS to report missing out or suffering loss and sacrifice in their lives due to the limiting nature of their conditions (Tran et al., 2019).

He continues to reflect on stories of loss when he thinks about the sacrifices to her health that Florence made with her pregnancies (Pezaro et al., 2018). He reflects with hindsight that if Florence had known about EDS, she may not have had children:

S: *“without a shadow of a doubt (.) I am 100% convinced if she hadn't got (.) without that (.) if she hadn't got children (.) she'd be much healthier”.*

Halfway through Florence's individual interview, she draws on narratives around loss and the impact health has on the family. She highlights similar instances to Jason such as the children's ability with hobbies and talents:

F: *“you know Summer was FANTASTIC erm (.) she used to play clarinet and she was fabulous at that (.) and then she °had to stop° because of her lungs (.) so stuff like that is a bit that is a bit °frustrating° (.) you think (.) well that's °not fair°.*

This fits with existing literature about PLWPPS who don't reach their full potential due to losses and sacrifices made because of health (Tran et al., 2019). Here, the narratives around loss also appear to be associated with narratives of uncertainty drawing upon the idea that not only is there a sense of what they have currently lost but also, a fear for future losses (Nettleton, 2006; McWilliams et al., 2016).

Beau draws on a different story of loss, specifically focusing on his sadness for his parents' experience because he and his siblings are unwell when he says:

B: *“you really want them to (.) er (.) feel for (.) you know (.) have the same opportunity as they would (.) it's if you know (.) I was of having normal health”.*

He appears to be drawing on ideas of the parentified child whereby he is positioning himself as having responsibilities for parental wellbeing (Chen & Panebianco, 2019).

Towards the end of the group interview Beau proceeded to highlight the sacrifices that his parents, particularly his mother, have made for the children, despite struggling with her own illness. He described:

B: *“Mum's very strong [...] she almost (.) doesn't let it keep her down (.) she just like powers on through it (.) and it's like (.) she makes huge sacrifices for us::=”.*

He appeared to draw on narratives of pride for his parents and the sacrifices they have made, and he mirrors Amelia's earlier position of gratitude.

Beau draws on stories of a loss of opportunity through illness and appears to speak on behalf of his family, when questioned about what happens when illness enters their lives. He responds with:

B: *“(3) I mean (.) I guess we didn't really get to do everything that we normally would do (.) so like it has sort of stunted the (.) erm (.) opportunities that we might have had (.) if we were more normally healthy”.*

Beau, (Group): 13.00 – 13.10

Amelia continues to draw on this narrative in the group interview, she positions herself as removed from her peers and subsequently draws on narratives of difference and missing out when she says:

A: *“I find that I (.) because most kids they fall over sticks (.) they just get a little cut but (.) I would end up getting these really big injuries:: and that was quite erm (.) that's quite hard for me (.) and then I get pain after running around”.*

Amelia, (Group): 35.58 – 36.26

Following her siblings describing their loss and drawing on narratives of missing out, Summer reflected on her own experiences of loss. In response to Beau citing that things had improved for him over time, she drew on counternarratives of things getting harder as she got older. She said:

S: *“especially now during this UCAS period (.) [...] it was like every- was around like “well I can put DofE (.) I've been charity working” this sort of stuff (.) and actually (.) I was there and I was like (.) “don't have anything to °write down°” (.) I think for me*

*(.) I noticed that there is more difference between me and my friends and my peers (.)
I think as I've got on (.) I've struggled more".*

Summer (Group): 42.15 – 42.20

There may be a difference in gender narratives being played out here, specifically the social pressures for girls to be well rounded may be more present than for boys, who can compete in more physical ways. The greatest loss that PLWPPS experience appears to be the loss of social relationships and other social factors external to the family system (Lidén et al., 2015; Winger et al., 2013). While loss and sacrifice has been explored in this family's narratives, they also narrate stories for family unity which is discussed next.

4.4.2 “The bond between us, it doesn't really falter” – Stories of Family Unity.

Throughout all the interviews the family members frequently drew on narratives of family unity. For some family members, they positioned themselves in relation to this narrative as if they have no other options. This finding doesn't fit with previous literature that supports a relationship between family 'dysfunction' and PPS (Dwamena et al., 2009).

Beau initially draws on this narrative early in his individual interview when he explained the importance of coming together as a family in the face of challenging times. When asked how his family copes when faced with a future of worsening health (when he saw wheelchair bound children with EDS) he prioritised family closeness to feel safe in the face of an uncertain future:

B: “we went back up to the hotel room and we just erm (.) we just sat around and we just play cards .hehehe and it was just like (.) it was almost like having that (.) er family moment”.

Beau (Individual): 19.40

For Amelia, like Beau, she finds comfort in the shared family understanding and experience of illness when she says:

A: *“it's nice when we are all ill together because we can all just sort of sit with each other”*

Amelia (individual):20.03

She appears to strengthen this narrative when she talks about how people with PPS are often misunderstood, and therefore feeling understood by your family is beneficial. This has been found in the literature whereby, families coping with chronic illness benefit from feeling understood by their family members, where perhaps, they do not receive it elsewhere (Årestedt et al., 2014). She explained:

A: *“it's also quite nice to know that you've always got some people that understand what you're going through”*

Amelia (Individual): 23.22

Summer, however, appears to offer a counter narrative in her individual interview when she describes her struggle with the lack of escape from illness (*“I mean (.) it can be quite difficult when all three of us (.) you know (.) we're all coping with the same thing”*). Research has demonstrated how people with chronic illness find themselves suspended between attempting to escape the emotional suffering of illness and the perpetual state of enduring that they face (Öhman et al., 2003). This counternarrative was not expressed by anyone else in the family.

In response to Florence describing how Summer's friendship groups had not been *“very thoughtful or caring”*, Summer discussed the consequences of missing out on friendship development and social interactions:

S: *“I think for me (.) when I had no one at school (.) I kind of felt quite sad and quite low (.) I'd come home (.) and I'd be like really kinda of distanced (.) and not very talkative or even you know (.) angry sometimes”*.

Summer (Group): 28.29

Across the interviews family members discuss their responses to Summer's struggle and her subsequent isolation. Conversely, in his own interview, Beau explained that when Summer is feeling well: *"it's almost like the whole morale of the house is up (.) you know (.) we're all (.) we're all having fun (.) m-we're all enjoying ourselves"*.

When Jason, the only well member of the family is asked about his experiences of his whole family being unwell, he continues to strengthen the family unity narrative when he demonstrates that despite illness, they have each other and that is most important:

J: "our life's good in a lot of ways (.) so we know (.) we're all very well connected (.) that's the most important thing [...] as long as we have that (.) then I don't (.) we can get we can get through most of this".

Jason (Group): 47.52 – 48.07

Patterson and Garwick (1994) found that "well-adapted families" with chronic illness, can create a sense of balance between coping and managing the complex demands they face daily. This reliance on each other to be unified and cohesive is further explored by Jason in the group interview when speaking about the consequences of either external pressures or internal flare ups on the family:

J: But if anything else comes through the door (.) then it obviously that then has a big impact (.)

K: [...] is there anyone that perhaps has more flare ups or more kind of becomes more unwell than others in the family?

[...]

J: [...] once somebody's down (.) it's very easy to drag everybody down (.) isn't it↑ you know (.) it doesn't take much for that to happen (.) so it's important that you know that when it does happen (.) we try to stay close in as much as we possibly can".

Jason (Group): 50.10 – 50.41

He both strengthens and resists the narrative of family unity, when he speaks about the how they can lift one another up or drag each other down. This is seen in the literature and draws on the Minuchin Family Stress model (Pardeck, 1989) which proposes that when a family faces ‘extrafamilial’ pressures, strong familial interpersonal skills help the family cope (Pardeck, 1989). Florence continues to strengthen this narrative by describing the importance of teamwork:

K: “I'm getting this real sense of the importance of like teamwork as well-

F: that's it (.) that's exactly what (.) but that's how I think (.) that's how it works (.) so when one goes down (.) but then we all pull in and scoop that one up (.) but then it comes as a consequence (.) to everybody else (1) it is part of how it is I suppose isn't it↑ we all do it for each other (.) and there's no one different in any- you know↑ (.) we all do the same”

(Group): 52.45 – 53.13

Jason and Beau continue to strengthen the narrative of family closeness when they show that no matter what they face, if they remain close, they can take on anything:

J: “I think (.) you know (.) we've dealt with a lot so I don't see why we can't deal anything going forward”.

Jason, (Group): 60.35

B: “It kinda makes you feel like we're strong (.) you know (.) it is there's (.) it's like (.) the bond between us (.) it doesn't really falter that much (.) it's like (.) because of that (.) we do stick together (.) and whenever we are together (.) we work best”.

Beau, (Group): 61.27

The family were also challenged in the group interview to think about if the closeness would be present if illness wasn't part of their lives, and when Summer began to respond with:

S: *“I don't know (.) because when you think about it (.) you know (.) if we were out socialising more–”.*

Summer, (Group): 61.59

Jason interrupts her and speaks with expertise to offer a counter narrative when he says:

J: *“I-I-I think I can I can probably answer that a little bit better because from my own point of view”.*

Jason, (Group): 62.05

He draws on his experiences growing up with a sister he describes as “*mentally handicapped*”. Jason explains that with his sister’s “*handicap*”, his family of origin was always very united (“*the bond in my family's really strong*”). He has a foundation of closeness which he would want in his family, illness or not. His interruption of Summer may have been his way of strengthening his counternarrative protecting the image of family closeness that may not be there if they were healthy. If Summer had finished that sentence, it would have challenged an aspect of his strongly held family identity.

Much of this narrative seems to be strengthened by Beau and Jason and appears to be drawn upon towards the end of each of the interviews. It appears to position the family as united as the interviews are drawn to a close.

4.4.3 “We are ill...I can do more” – Stories of Collective and Individualistic Language.

Another overarching narrative that is frequently drawn upon is the way the family members use language to position themselves in relation to their illness. Most of the family members draw on this narrative throughout, but not unsurprisingly, Jason doesn’t appear to use this narrative, given that he is not unwell. The use of language to explore and explain illness has been established in the literature (Kirmayer, 1999; Risør, 2009). Ware (1999)

discuss how PLWPPS develop a way of talking about illness that is guided by the social processes and discourses occurring around them.

Beau strongly draws on this narrative where he uses first person 'I' to position himself as overcoming adversity, or to establish strength in the face of illness when he says:

B: *"I (.) we always (.) I always try to (.) like (.) do whatever I can on my own [...] I just sort of saw that over time (.) you know (.) the more reliant people are (.) the more likely they are to (.) almost not be as well in health(.)"*

Beau, (Individual): 21.24

Beau appeared to favour more collective language, such as 'we' and 'they' to position himself away from an illness identity. This is particularly evident when Beau talks about the consequences of when he and his sisters start to feel well, he says:

B: *"but then I- when we will feel well (.) there's an overconfidence and it's like (.) they get too conf- they get almost too cocky (.) right↑ so it's like (.) they know that they're well (.) right (.) so they try and do more"*

Beau, (Individual): 27.51

This appears to illustrate that he changes his language to distance himself from the negative consequences of being ill. He then counters this collective narrative when he positions himself as separate to his sisters and in a position of achievement when he states:

B: *"I tried to encourage the girls to do the same as I did (.) but erm (#) they tried it once (.) and they couldn't do it"*

Beau, (Individual): 28.27

Summer's use of language differs to Beau's, she appears to draw on this narrative and use collective language to demonstrate a process of maturation and to align herself with her parents. This is demonstrated when she remembers her brother as an unwell baby. She said:

S: *“yeah (.) so it just kind of I think we kind of noticed with him that it wasn't (.) quite that I- it something really wasn't right (.)”.*

Summer, (Individual): 9.33

She appears to use “we” language to align herself with her parents, positioning herself in the parental subsystem to demonstrate her maturity and move away from narratives around ill children. She continues to use this narrative to demonstrate her maturation when speaking about the sacrifices their mother makes:

S: *“mmm I mean we feel quite bad (.) I mean (.) I don't know about Beau and Amelia actually (.) I probably shouldn't say we (.) but erm I do feel quite bad that she's always asking after for us (.) and we're not asking after her and that we don't do enough for her”.*

Summer, (Individual): 21.38

She appears to move the audience away from the collective “we” position to prioritise her individual opinion as more important compared with the collective illness identity.

Conversely, earlier in her interview, she does use the collective language of ‘we’ to draw on the collective narrative of strength, when they needed to step up when their mother was ill:

S: *“so I think (.) particularly when mum's down (.) it becomes harder (.) because we noticed that she's (.) you know (.) she needs more help and stuff like that (.) and it does get quite hard for us because (.) we're not used to it”.*

Summer, (Individual): 17.42

Amelia uses her language in a different way again, near the start of her interview she begins to draw on this narrative to demonstrate, strength, unity and normality when she says:

A: *“being around when I'm like, (.) if I'm like just around my family [...] because we (.) ba- all basically have it (.) so it's this feels normal”*

Amelia, (Individual): 7.14

Beau uses individualised language to position himself as strong, whereas, Amelia appears to use 'I' language to draw on narratives of self-blame and criticism:

A: *“°they used to say that I was lazy° and [...] that °I couldn't be bothered to do anything° that's why I had to have breaks and stuff (.) but it wasn't (.) it was °because I couldn't manage it°”.*

Amelia, (Individual): 9.58

Amelia also strengthens the narrative of collective language use in a similar way to her sister Summer, when she talks about her experiences of her mother being ill, she said:

A: *“I think that when my mum's unwell it all just goes .hheh really downhill [...] we all kind of realised that even though our (.) mum (.) has the same thing as us but she did like loads of stuff to make sure we were all well (.) then we were ill and she was ill (.) it was quite hard”.*

Amelia, (Individual): 18.15 – 18.55

Florence uses her language in a similar way to Beau as a means of distancing herself from illness. When I asked her to give examples of what happens when the unwell family members have a flare up and she responds with:

F: *I mean (.) it's funny because they've described EDS flare ups happening how awful they are and “blah de blah de blah” (.) I don't (.) I don't know if we really identify when that's happening.*

K: *Yeah (.) really↑*

F: *I think maybe (.) maybe we do have a bad day↑ maybe it's (.) yeah (.) I don't call it- maybe we have the odd (.) you have a bad day (.) “someone's having a bad day”.*

Florence, (Individual): 22.12 – 22.27

Although the question was related to a homogenous group of ‘unwell’ family members, she responded by strengthening the narrative of the use of collective language. She also draws on ideas of normalising the illness with non-medicalised language e.g. “*a bad day*”. Morris and Ogden (2012) discuss mothers’ needs to normalise their children’s illness experience which is demonstrated here by Florence’s move away from medicalised language.

In the beginning of her individual interview she also proposes a counternarrative to normalising the illness, when she says:

F: “this is just EDS (.) “there's nothing wrong with you” (.) its EDS (.) there's nothing (.) nothing wrong there”.

Florence, (Individual): 4.30

While she appears to use this language to normalise her own experience, it may be that she is voicing past experiences of rejection and dismissal from HCPs with this phrasing and had positioned me as a professional in this moment. However, within the group interview, in front of her children, she appears to strengthen the collective language narratives and seems to use her language to normalise the illness: “*but I (.) we are ill in the same way as I've got brown hair*”. Florence’s use of language within the group interview, appears to have a similar function to her use of humour in front of her children.

Fleischman (1999) reflects on how the use of language allows us to apply differential meanings to our experiences, however, there is an interplay between medicalised language that impacts how we ascribe meaning to our own stories. Horton-Salway (2001) also explores how language and rhetorical devices are employed by PLWPPS to formulate aspects of their identity and to counter outward perceptions of people’s experiences of PPS.

4.5 How do families construct their identities?

Throughout the stories told, it appears that the illness identity is present throughout with individual’s either holding this identity strongly or rejecting it. Florence, Beau and

Summer, all reject the illness identity for different reasons, with Florence appearing to favour a more can-do identity, Beau favouring an identity based on strength, and Summer favouring an identity focused on maturation. Amelia, however, strongly draws on the illness identity, as she was born into illness and while she has benefitted from learning from her siblings, she appears to prioritise the illness identity to remain connected to her family.

Additionally, traditional gender identities appear to come through with Jason's role as the breadwinner and provider for the family. Beau, being the only male child, also prioritises his male identity, through comparing himself to his peers and focusing on training and exercise. Florence, who also works, prioritises more traditionalist gender roles of the woman raising the children. Florence's identity as a mother often triumphs above her illness identity, signifying that her illness comes second to her children's (Vallido et al., 2010).

What does come through is how infrequently the children prioritise their child identity. Parents of chronically unwell children report a loss of the child identity in the context of illness (Smith et al., 2013). Amelia appears to reject her child identity when she describes her experiences of interacting with doctors:

A: *"I never really understood because the doctors would ask me questions and I was only like three or four (.) but my mother was answering them as well (.) but then the doctor would always be like "so what do you think about that?" I didn't really understand it [...] I didn't know really know what any of that meant".*

Amelia, (Individual):14.30 – 15.00

I imagine that these experiences are mirrored in her siblings and would demonstrate the need to detach themselves from their child identity to feel more comfortable around doctors and appear more knowledgeable about their condition.

The family members use language, pausing and different rhetoric tools to enact or to reject their identities (Horton-Salway, 2001). There are the individual identities but also the

collective family identity of closeness that gets prioritised when the family are together and when the family is under ‘threat’ from external parties/pressures.

4.6 Role Relationships

When illness enters this family, while there are several different individualised responses, there is also the collective response which dictated how the roles and relationships change within this family system. Amelia describes how each of the family members navigate the sick role:

A: *“if we're feeling well (.) if one of us is like feeling well (.) or we're all feeling a bit ill but we're all okay [...] one person's like really well (.) and we all kind of just feel well (.) so it kind of makes everyone happy (.) yeah (.) and then if we're ill (.) we all kind of feel a bit like (.) ill really”*

Amelia, (Individual): 8.14 – 8.36

Despite it being apparent that illness is ever-present within this family’s life, each of the unwell family members navigate the sick role in different ways. It appears that when the children are ill, the parents move into the role of keeping the mood light. With this, Florence describes Jason as their “*whistling Rufus*” and brings his “*happy soul*” to the family to keep the tone light. He reported in his interview that he does things “*to keep a nice joyfully (.) air around the place*”.

In Florence’s individual interview, she draws on the benefits of Jason’s other role as the provider highlighting that it is his way of coping, however, she didn’t appear to want to discuss these benefits in front of her children:

F: *“he's turned his whole focus in coping with and managing this kind of illness (.) is on earning money and giving the kids security (.) which he's done we've got (.) they've got their own houses now (.) without mortgages”.*

Florence, (Individual): 28.57 – 29.01

Jason works hard so that he can provide for the children's futures, however, this may be drawing upon wider discourses of lack of achievement for PLWPPS as with Jason's fatherly provision, there is an implied assumption that the children won't be successful. However, this is an opportunity that is not commonly had by people suffering with chronic illness and it comes at a cost to the family with Beau commenting in his individual interview:

B: *"it took me a while to actually even (.) like (.) get to know my dad (.) you know (.) [...] I knew that I loved him (.) but then [...] the only reason why he was at work so long was for us".*

Beau, (Individual): 43.01 – 43.50

Summer finds that she worries about her father's health from the resulting stress of the provider role:

S: *"he'd come back at times and he'd be like really pale↑ and you know really like sickly↑ looking and that was really stressful because [...] stress can really affect (.) you know (.) his health".*

Summer, (Individual): 19.21-19.41

She appears to be taking an expert position in relation to her fears about her father's wellbeing and shifted her role from child to parent. When Jason is enacting his provider role, Florence feels isolated when things become difficult, she said in response to being asked who she turns to for support:

J: *"I don't actually (.) there isn't anybody to be turned to particularly because I am on my own with it"*

The negotiation of the parent role seems to be shared between the family members, including the children. The parents' roles are described by the family members in the group interview and it appeared that Florence was keen to share the leadership role, which is a counternarrative to her proposed authority earlier in the interview:

F: I lead (.) I think (.) I lead the lead I do lead s- (.) I do lead I do model I lead (.) I do (.) encourage and keep people up (.) Yeah, I do do that”.

B: But the leadership role does switch because like (.) sometimes Mum will be doing stuff that will be helping us (.) and then other times dad will be doing stuff like [...] like he's kind of (.) he encourages us to do more [...]

S: I'd say he's the rock (.)

F: We say he's 50% of the rock”

(Group) 53.58 – 58.36

However, it appeared evident from all the interviews, the most challenging time for this family was when Florence had her hip replacements, meaning that the “mother role” was temporarily left vacant. It was swiftly filled between the children with them all commenting:

S: “I did the washing and stuff like that[↑] but it was like we tried to keep (.) doing stuff that Mum would do”.

Summer, (Individual): 18.15

A: “would make sure that the dinner was done and I make sure that the- (.) I'd asked my mum to erm (.) tell me like food shops so I could order online make sure that it got put away in the right places and do things (.) I was kind of charge the kitchen (.) my brother would do the bins and the hoovering (.) my sister would make sure the house is really clean because she likes tidying”.

Amelia, (Individual): 29.34 – 29.58

B: “we all had to sort of erm work together to kind of fill the hole when Mum wasn't there”.

Beau, (group): 67.04

Beau reflected that he relished this role change (“*I almost enjoyed it (.) right↑*”), whereas Summer found it challenging (“*it was hard for us to kind of put ourselves (.) in her shoes*”).

While Florence and Jason have clearly defined roles, Amelia describes Beau as “*the joker*”. This appears to be Beau’s way of stepping into his father’s role, perhaps when his father is absent at work. It appears that Beau feels he needs to step into this role to maintain the equilibrium of the household. Summer storied that she negotiates several roles; she is the guinea pig, the troubled teenager and the one that keeps an even temperature in the family. It appeared that Summer felt pressured to be a certain way for her siblings:

S: “*I think Beau and Amelia very much erm (.) look up to me because I’m obviously the oldest sibling (.) it’s kind of the way things normally go (.) so I think they look at what I’ve:: done (.) and they try and do bett::er::*”.

Summer, (Individual)29.07 – 29.20

Summer is the only child to have been reported as having mental health problems. This was attributed to social difficulties at school, but it appears that perhaps she felt her role within in the family was unspecified, and felt pressure being the “*trial run*”. Summer also takes the position of “*a thermostat*” for the whole family, as reported by Florence in the group interview. This is built on by Beau when he explains:

B: “*when Summer’s doing good (.) it’s almost like there’s a uplift to the whole family [...]* Summer’s almost like the catalyst to the right reactions”.

Beau, (Group):55.53 – 56.18

This pressure for Summer to be well to maintain whole family wellbeing is mirrored in Amelia’s role. Summer described her in the group interview as “*the happiness pill (.) the vitamin*”. In his individual interview, Jason described Amelia’s role and alludes to family struggles that may have been going around Amelia’s birth when he says:

J: *“if anything meself and Florence would always say that Amelia came to us at the right time [...] Amelia was the right person at the right time in our family because she was (.) she took that bit of life into the family to some extent [...] she was very important at that point in our lives”.*

Jason, (Individual), 41.02 – 41.37

She appears to take on a rescuer role of keeping the family healthy. However, Florence commented on the consequences of this role:

F: *“when her symptoms get too much there is a complete change in her when °she is very dark° and very (.) very anxious and very upset (.) and that could be very difficult for everybody because you have this lovely ball of rainbows that then suddenly comes quite dark and °quite hard to sort of bring her back round to herself°”.*

Florence, (Individual), 43.21 – 44.15

Årestedt et al. (2014) found that for families living with chronic illness there is a daily negotiation of roles and sharing of responsibilities in a way that may not occur in healthy households. That applies to this family as they discuss how they negotiate several different roles amongst themselves. It is also apparent that the role of time influences the relationship dynamics. While enacting these different roles, the family members also choose to prioritise, or move in and out of certain identities by drawing on different narratives and wider societal discourses.

5. DISCUSSION

5.1 Overview

Before providing an overview of the results, I will re-orientate the reader to the research question set out in chapter 2.

‘How do families construct their identities and make sense of their role relations when both parent and children are unwell with PPS?’

This research was interested in understanding through the application of narrative methodologies how a family with PPS in two generations, construct and negotiate various identities and roles within their relationships. While some of the main themes have been discussed in the previous chapter, I continue to bring together some of the remaining themes, embedding them in existing literature. This chapter then explores some of the methodological considerations relevant to this research such as the strengths, reviewing quality and some of the research’s limitations. Lastly, I consider some of the clinical implications of this research and any future directions that may be indicated.

5.2 Overview of results

This thesis, to the author’s knowledge is the first of its kind to explore how a family with two generations of PPS story their lives and their unique life events from a narrative perspective. Given the parameters of the MRP, areas that did not directly address the research question were unfortunately, not included.

The analysis initially explored how the individual family members told their stories and which narratives they drew on or rejected to jointly construct their stories. The analysis then explored the wider narratives that emerged from the interviews, while lastly focusing on how the family constructed their multiple identities and navigated changing roles in the context of illness.

Within the individual storylines, common narratives drawn upon were stories of ignorance, overcoming adversity, social difficulties, busyness/achievement, humour, lack of achievement/laziness, competition, rejection by services, denial of illness, parental protection, ‘being lucky’, acceptance and sibling relationships. The family members appeared to draw on these narratives to affirm, to reject, or to counter certain preferred identities.

The story of ignorance that certain family members drew on, fits with wider literature about these conditions being misunderstood by people around the sufferer (Edwards et al., 2010; Giroux et al., 2016). There is a large pool of evidence from the perspective of HCPs not understanding PPS (Dirkzwager & Verhaak, 2007; Furness et al. 2009; Giroux et al., 2016; Stone, 2014 a & b). This family discussed the ignorance they faced from school staff. Research has shown that support from teaching staff minimises the distress induced by school for PLWPPS (Shannon et al., 2010). This differed immensely from the experiences of this family and further research should be undertaken to explore the competency and confidence of school staff to understand the complexities of children with PPS.

The family members also drew on narratives of overcoming adversity. This has been explored in the literature, whereby patients draw on different meaning making mechanisms to reject the illness narrative to overcome their challenges (Østbye et al., 2020). The way in which the family members draw on the overcoming illness narrative resonates with the way PLWPPS use their talk to legitimise their illness expression (Harter et al., 2005; Japp & Japp, 2005; Nettleton, 2006).

The story of acceptance was narrated by a few of the family members, with the metaphor of a 'shadow' being used to understand the experiences of living with PPS. Lidén, et al. (2015) found that PLWPPS expressed the need to learn to take care of themselves and accept the current state of their health to help them assimilate PPS into their lives. Bennett et al. (2018) explore the psychosocial implication of living with hypermobile EDS, highlighting that they experience social stigma, specifically being told that they 'look fine', which was expressed by the participants in this study. Bennett et al. (2018) further findings of *rejection from services, social difficulties, lack of achievement, fear of the unknown and the loss associated with a restricted life* are also echoed in the findings from my MRP.

Another theme from the analysis was the idea around the denial of illness, either from denying the importance of Jason's illness or Florence denying the impact of her illness on her childhood to move towards a 'normal' life. Research found that over time PLWPPS make attempts to lead a 'normal' life to protect their family, which may involve hiding their symptoms or rejecting their illness all together (Kornelsen et al., 2016).

The wider narratives presented across the interviews drew on ideas of loss and sacrifice, family unity and the use of language. The family members all experienced loss and sacrifice, largely focusing on a loss of opportunity and social relationships. PLWPPS have reported that they feel that life goes on without them and report significantly greater social isolation and loneliness compared to their healthy counterparts (Dirkzwager & Verhaak, 2007; McWilliams et al., 2016; Winger et al., 2013).

Family unity was discussed by all family members. The overarching sense was that their family unity and cohesion was a strong protective factor from further distress. The family discussed that the times that they felt most overwhelmed was when they faced external pressures, or when something unexpected happened. Research has shown that family cohesion may be disrupted when family rituals are unsettled (Fiese et al., 2002; Santos et al., 2016). Family functioning (often mediated by the family members relationships with their mother) has been shown as a protective factor for healthy psychosocial functioning (Hoffman et al., 2016; Hunfield et al., 2002). Family "dysfunction", however, has been shown to be a common experience in high-utilising primary care service users with PPS (Dwamena et al., 2009).

While this family draws heavily on their family unity, research has shown the family members of PLWPPS report how their lives, roles and relationships are negatively impacted by the unwell person and their illness (Ashe et al., 2017; Liedberg & Henriksson, 2002). The wider evidence base has documented the challenging reciprocal relationship between PPS

and family “dysfunction” (Dwamena et al., 2009; Edwards et al., 2010). This was not the case as expressed by the family who took part in my research and while the consequences of family cohesion were discussed, overall, the benefits of family unity triumphed. This fits with findings from Roy (1982), Garralda, (1996), and Rosland et al. (2012).

This family also frequently used narratives of individual versus collective language to position themselves closer or further away from illness and to narrate themselves into a more or less powerful position within their own lives. Kirmayer (1999) discusses the use of rhetorical tools that PLWPPS use to legitimise their illness and to change their positions within their own context.

The identities prioritised amongst the family members were the illness identity, gendered identities, the rejection of a child identity and the juggle between a parent, a patient and professional identities. The rejection of the child identity is commonly found in children who are chronically unwell. One reason for this may be due to spending much of their time communicating with adults (in hospitals), they are treated like and feel like an adult (Kirkpatrick Johnson & Mollborn, 2009; Smith et al., 2013). The process of maturation demonstrated by the children in this family fits with ideas of life cycle transitions and the requirements of a family to be flexible in the face of stressors such as illness or injury in childhood or an unwell parent (Hamberg & Adams, 1967). When chronic illness enters a family, it can disrupt the natural role transitions and can in fact induce role reversal in some contexts (Johnston, 1990). Jason and Florence appeared to have a strong marriage and worked well as a team, however, Jason’s physical absence from the family through work was noted. It is common within single parent families, there can be a move by the children to step into, or be drawn into the parent role, which mirrors the experiences with this family (Duryea, 2008; Johnston, 1990).

The dynamism of the mother role was another finding that emerged from the analysis. The role of the mother has been established in the literature, but often from a relatively pejorative stance with mother blaming or highlighting the role that maternal overprotection plays in subsequent PPS in the child (Fisher & Chalder, 2003). The novel finding from this research was the way the mother role was centralised for this family and how this was negotiated amongst the family members when Florence's illness worsened. Florence prioritised her children's health needs over her own, which has been seen by Vallido et al. (2010) as they discuss how unwell mothers' prioritisation of their mother role comes at a cost to their own health.

Overall, while there are some findings in this study that were in line with the current research, several novel findings also came from this study relating to the lived experiences of a family with multigenerational PPS.

5.2.1 A reflection on theory.

In chapter one, many theoretical perspectives were explored when trying to understand PPS from a psychological perspective. When reflecting on the findings of this research, it is important to note how they fit with the theoretical perspectives referenced in chapter one, specifically attachment theory and systemic theory. Family unity was deemed to be an important finding when thinking about how families with multiple generations of PPS construct their identities and navigate their roles. Drawing on systemic theory, specifically Minuchin's model of the 'psychosomatic family', this family highlighted their cohesion as a protective factor for their distress however, it could be hypothesised that ideas of enmeshment and familial overinvolvement/overprotection may be also present (Kog et al, 1985; Minuchin, 1970; Roy, 1987). Many of the systemic theories that underpin research on stigmatised illness relate to discord amongst the family members which suggest that symptomology may develop to direct the focus away from the familial problems (Roy, 1987).

With this family, although they referred to some arguments, they were keen to prioritise their closeness in the face of adversity, however, I only met the family on this brief occasion, so it would be useful to explore this finding further from a systemic perspective.

As noted, this family experienced multi-generational illness. Byng-Hall (1998) referenced the idea of the development of family scripts in the context of intergenerational familial illness. In her individual interview, Florence discussed the influence of illness throughout her life, exploring how because of her father and grandfather's relationship to illness and pacing behaviour, she may have cultivated a more "can-do" identity. Over time and generations this may have meant that a family script relating to health and coping may have developed and subsequently may be being enacted in her children.

The findings relating to loss and sacrifice draw upon ideas around attachment theory. The relationship between PPS and attachment and the relationship between loss and attachment have been established in the literature (Adshead & Guthrie, 2015; Shaver & Fraley, 2008). With this family, across the interviews and in the findings, the need for the parents to protect their children from further loss and to help them cope with the sacrifices resulting from chronic illness was ever present. Parental overprotection can be thought about in the context of attachment theory whereby parental overprotection may lead to an insecure attachment in the child, which may contribute to subsequent physical illness (Fisher & Chalder, 2003; Janssens et al., 2009; Maunder & Hunter, 2001). However, for this family, at the point in time I met them, it seemed that their protectiveness of each other was appropriate to the context of discussing their familial experiences with an outsider, rather than *overprotection* per se. Additionally, attachment theory may underpin the finding of the centralised role of the mother too, supporting a relationship between maternal-child attachment and intergenerational manifestation of internalised symptomology (Brenning et al., 2011).

Lastly, without further information it is difficult to link relate the findings to the behavioural/cognitive-behavioural theories because these theories focus on the individualistic perspective e.g. the person's thoughts and beliefs about their own health which doesn't fit with the relational perspective of this research.

5.3 Quality Assessment

To maintain transparency with the process of this research, the strengths and limitations are explored. Table seven shows a quality assessment of the current research using Tracy's (2010) criteria.

Table 7: Quality Assessment for Current Research

Worthy Topic	<i>The research is a novel topic, interesting and provides a timely perspective for the topic. The research filled a gap of looking at EDS and PPS from a relational perspective. To the researcher's knowledge, this has not been done with this methodology and with two generations (plus) of PPS.</i>
Rich Rigor	<i>Tracey (2010) comments that qualitative research benefits from a "researcher who is widely read and provides clarity [...] by which she arrived at findings". To provide rigorous research, I have ensured that I have thoroughly read different theoretical conceptual ways of conducting narrative research. I kept careful field notes and methodological transcriptions. I shared transcripts with other members of the research team to ensure careful and appropriate analysis of the material. Aspects of my transcripts, reflexivity and field notes can be seen in appendices L – O, P and Q, respectively.</i>
Sincerity	<i>This research has been conducted in a manner that has allowed me to remain reflexive and to challenge my own position throughout, where possible I have shared my reflexivity with the audience to maintain transparency and clarity. I have had several supervisory discussions which have challenged my position and changed some decisions I made throughout the course of the research. Examples of a reflexive diary can be seen in appendix P.</i>
Credibility	<i>The research aimed to offer a clear, transparent approach to this topic. Supervision was used to ensure credibility of the findings and the processes</i>

involved. I aimed to provide thick, rich descriptions of the findings that guide the audience to make sense of the narratives through their own lenses. Unfortunately, due to time constraints I was unable to conduct member reflections or multivocality from other families. This is a limitation of the current research.

Resonance *Given the novelty and the extent to which this research is conducted with rigor, I would hope that the research will have resonances with the wider audience. The research's aim is that its implication will be far reaching, and this family's narratives are meaningful for others to explore further. These findings will resonate with other PPS condition. It was an unexpected finding that the analysis resonated with both PPS and EDS literature.*

Significant Contribution *This describes how the research should contribute to a possible four different domains; methodological, theoretical, heuristics and practical. This research aimed to provide a significant contribution to methodological, theoretical and a practical perspective, because it has clinical implications, novel methodology and will add to the theoretical understanding from a systemic perspective.*

Ethical *Conducting high quality ethical research means meeting standards for situational ethics, relational ethics, procedural ethics, and exiting ethics. Each of these have been addressed and thought about throughout the course of the project as evidenced by the ethical approval documents attached in appendices C and D.*

Meaningful Coherence *This aspect of quality research is fundamentally, whether the research achieves what it sets out to do. This research sets its aims out in chapter two and I have demonstrated that the research met its aims with the findings explored above. The research has embedded the current findings in previous literature.*

5.3.1 Strengths of the research

Anderson (2010) suggests that some of the strengths of qualitative research are that issues can be researched in depth, draw on human experience and permit an iterative process to knowledge conception. The main strength of this research is that it provided an opportunity to explore an under-researched area, specifically with PPS conditions and even less

researched with EDS. Systemic research with PPS has not been explored recently, perhaps due to the rise of CBT. CBT (for major depression) has been shown to be more economical than other longer-term psychotherapeutic options, so may be favoured by the NHS (Goodyear et al., 2017). This study provided an opportunity for the family to discuss their unique life events in the context of their illness and puts forward a novel perspective of the narratives of a family where PPS is in multiple generations. The research enabled the family to uncover some of the ways they negotiate their roles and the strengths they have within their family. They expressed they found it beneficial to recognise their achievements, despite having their lives be largely problem focused.

5.3.2 Limitations of the research

While this research puts forward beneficial and novel findings, it is important to reflect on the limitations that are inherent in any research despite attempts to mitigate. In the initial design stages of the project, I had planned to recruit a family with PPS where there was a parent and child with poor and good health, respectively. However, as explained, the research recruitment process was impacted by COVID-19 and this family make up was not available. Additionally, this research focused on PPS diagnoses, and the uncertainty associated with the unique life events of these family members, however, the participant family had a fascinating family history with EDS. EDS is a rare diagnosis which is largely misunderstood by the medical profession (Bennett et al., 2018). The narratives constructed by this family, largely resonate with findings where participants have other common PPS conditions. However, it is important to remember that with all qualitative there is an element of subjectivity.

Another limitation of the research is associated with the dual position of researcher and psychologist. As a trainee clinical psychologist (TCP), I am new to qualitative research but have worked clinically for many years. It remained difficult to hold a neutral stance when

conducting the interviews and not show the same curiosity for my participant's struggle and distress as I would have done had they been my service users on placement. If I had asked questions in the same way, I may have moved away from the authentic and transferable research processes. I wondered how my introduction as a TCP might have informed my positioning and the co-construction of narratives within the interviews. Research has discussed that trainee psychologists often do not privilege their identities as researchers above clinicians which has an impact on their engagement with the research process (Smith & Thew, 2017).

With a single-family case study design, it is not possible to remove the stories of this family, from their specific context and their own individual differences, and their experiences may not sufficiently resonate with other family's with PPS. For this reason, it would be useful to explore the same research aims with a larger participant pool.

The research was conducted online due to COVID-19 and although there is established literature on conducting qualitative research online, a limitation is the lost nuances of not being in person for the research (Salmons, 2015). For the group interview particularly, it would have been beneficial to have the family in one room, in a more neutral context as I was aware of the impact that the context has on the co-construction of the narratives.

Finally, the last limitation of this research was not having the time to do any member checking or co-coding (Busetto et al., 2020). An aspect of high-quality qualitative research is sharing and checking your findings and analytic processes with your participants, however, given the timeline and the scope of this research, it was not possible. Future research would benefit from ensuring that this aspect of the quality assessment is fulfilled.

5.4 Clinical Implications

The epistemological position of this research was employing a social constructionist perspective, the research was not interested in making claims about a material ‘truth’ about the stories of this family. However, as seen, the stories this family told resonated with stories told by other families in previous literature. Therefore, it is important to think about what implications these findings have for the clinicians who work with PLWPPS as well as any wider recommendations.

During the interviews, I found in the beginning my interactions with Florence (and Summer to some extent) were a little bit challenging. It is probable that this mirrors experiences in the clinical world, whereby PLWPPS have had multiple difficult interactions in which they need to legitimise their illness. Clinicians need to be curious and appreciative of the difficult journey patients have had before they reach specialist appointments (Jessop, 2014). Clinicians will also need to be mindful of PLWPPS relationship to health seeking (Reder & Fredman, 1996).

The relationship between how family members juggle their symptoms, roles and identities in the context of misunderstood illness has been discussed. While it is important to prioritise the dynamic relationship between all these factors, in current NHS frameworks, therapeutic and medical interventions are offered from an individualistic perspective. The current favoured treatment paradigm for PLWPPS is often CBT-based treatments. Both the negotiation of illness identity and the dynamism of relationships in the context of illness have been documented within this study. For this reason, along with the intergenerational component, clinicians should start to conceptualise PLWPPS healthcare from a more relational/systemic perspective. This may also signify the need for an MDT perspective when it comes to working with children and families. To acknowledge and legitimise the reality of the symptoms, clinicians must include medical, physical therapy and psychological

perspectives. This may create more room for psychological and systemic interventions to support families as they negotiate multi-generational physical symptoms.

As discussed, PLWPPS experience challenging journeys with the healthcare system, yet what appeared to be novel was that these difficult interactions with professionals begin at an earlier age. For this family, their difficult interactions with professionals (of any kind) began with primary school, or possibly earlier as they were seeking a diagnosis for the first two years of Beau's life. The difficult dynamics between the family and school system did not just focus on the child, the family alluded to the involvement of social services around notions of truancy and child abuse (accusations of Munchausen's by proxy). When a child presents in a paediatric health context, while the child's educational ability and engagement in school might be discussed during an assessment, the relationship the parents have with the school in this context may not have been. If the person's symptoms do not present until later childhood or early adulthood, then these early experiences may not be explored in great depth. However, the stigmatised identities may have already been formed and may already be challenging and disrupting the way PLWPPS use their talk to negotiate their illness within the wider systemic context (Reese et al., 2009). Clinicians should be aware of PLWPPS early interactions with professionals and how this may inform their current interactions.

5.5 Future Research

This research narrated the lives of one family. The participants appeared to be a White, British/Irish, middle-class family, and it would be worthwhile to explore whether this family's experiences and constructions of their world resonate with other families. PPS conditions are a predominantly white illness; consequently, it would be interesting to explore the unique life events of non-white families (Evangelidou et al., 2020). It appeared that with these participants, gender roles were being enacted and thus it would be worthwhile to explore how some of these roles would be impacted with a different family make up such as

same sex families, or families with adopted children. Future research may also benefit from widening out the family context to include multiple generations and to uncover the emerging narratives intergenerationally and across historical contexts.

5.6 Personal Reflections on the Research

The journey to completion of the MRP and subsequently my clinical psychology training qualification has been an undulating journey negotiating difficult terrain with uneven grounds, with some successes but many challenges. There can be parallels drawn between my experiences journeying with my MRP and the experiences of PLWPPS navigating social climates for example; the social sacrifices made, the confusion and lack of understanding involved with the process of conducting research and (at times) the lack of clarity and uncertainty involved with the future. While I have been afforded the opportunity for a beginning, middle and end for my journey, which I understand is not the case for PLWPPS, it perhaps gave me a greater opportunity to empathise with my participants.

While conducting the analysis for this project, there were times when I felt both blinkered by and blinded to my own personal experiences. As I felt I was being positioned as an outsider to this family, I had a challenging discussion with my supervisor about the idea that, of course, I was an outsider to this family so I wouldn't be positioned any other way. This made me reflect on the personal resonance of the project and my pseudo-insider status towards this research and drove me to question my positioning and the subsequent co-constructed findings. I thought about the differences between insider and outsider research, and while my parent and I have not suffered with stigmatised illness, I am somewhat of an insider. Ellis-Caird (2017) discusses the challenges of positioning oneself as an outsider to insider research and although this wasn't wholly insider research as I didn't present myself as an insider, it is important to reflect upon for the future. Similarly, through supervision I was challenged to draw on some of my own experiences of being severely unwell as a child,

which without supervision and the opportunity to reflect, I would not have re-connected with. Several of the participant family's experiences mirrored with my own, particularly with the parents feeling like they are protecting the children and the conflicting desire for maturation in the children. The negotiation of roles amongst this family also resonated with my own experiences and I wonder if adult children with unwell parents, who have experienced their own poor health often feel conflicted about their roles and identities. While my family and I have been lucky to be supported by great friends, I recognised many of the stories told by the children about missing out, feeling misunderstood and some of the anger at the unfairness and injustice of the situation and the system around us.

5.7 Final Conclusions

This research set out to explore how families constructed their identities and made sense of their role relationships when both parent and child were unwell with PPS. To do this, the background literature was examined in depth and the research was situated within its context to ensure there was a gap to be filled. Through researching the background literature on this topic, it appears that PLWPPS experience a lack of clarity, while also feeling misunderstood in many aspects of their storied lives. There is a certain complexity and blurriness to this topic that mirrors the lived experiences of people with PPS or EDS.

This research employed narrative methodologies to understand the unique life events of a single family in which four out of five members were diagnosed with EDS, specifically afflicted by PPS. Family and individual interviews were conducted with all the family members, who had been recruited from an adolescent specialist medical team in a London hospital. The analysis showed how the family drew on a multitude of narratives and wider discourses to co-construct their stories with me. Through the analysis, some of the family's multiple identities were uncovered for example, their relationship to their illness identities, their parent and child identities as well as a discussion around gendered identities. Within the

analysis, an exploration was conducted of how the family navigate and negotiate roles within their family and how this is moderated by illness. Lastly, clinical implications, possible service changes as well further research ideas have been discussed.

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7. APPENDICES

Appendix A – Initial Search Strategy

Parent OR Parents OR mother* OR father* AND Child* OR daughter* OR son* OR teenager* OR adolescent* OR family OR families OR “family system” OR “family assessment” OR intergeneration* or transgenerational AND “Persistent physical symptom*” OR "PPS" OR "MUS" OR "medically unexplained symptom*" OR "somatic illness" OR somatisation OR somatization OR “somatoform disorder” OR "functional illness" OR "idiopathic illness" OR "psychogenic illness" OR “conversion disorder” AND qualitative OR “qualitative research” OR “Narrative*” OR “lived experiences” OR “narrative enquiry” OR “Narrative Inquiry” OR “Narrative Research” OR “case study” OR “case series”

Appendix B – Full Systematic Review Quality Assessment

Quality Assessment table

Paper	Worthy Topic	Rich Rigor	Sincerity	Credibility	Resonance	Significant Contribution	Ethical	Meaningful coherence	Overall Quality rating
Carter (2002)	Yes – described as novel for its time e.g. at the time, need for literature to focus on the voice of the child with chronic pain.	Yes – appropriate data collection and analytic method but very small sample and unclear of the context.	Yes – express that the study comes from a constructivist philosophy – suggests some sense of personal reflexivity.	Yes – interview schedule is based on the diary entries from the participant families. Families commented on the transcripts and were returned to the researchers. Thick description of the family experience and detailed description of procedure	Yes – clear presentation and transferable conclusions drawn. Highlights the importance of giving the child voice and not just stay with the adults’ interpretation (professional or parents)	Yes – some practical contribution e.g. about how professionals should communicate with these families. Also, practical contribution around the ideas of ‘family hurt’	Maybe - Mentioned the idea of the children taking part and their right to be included in decision making	Yes – achieves what it sets out to do in an appropriate way	High
Dennisson, Stanbrook, Moss-Morris, Yardley & Chalder (2010)	Yes - Novel because CBT-based research is often quantitative	Maybe – sufficient/ low numbers for qualitative research but unclear by so many declined to take part, could be to do with time difference between end of intervention. The sample – all white, typical of British sample. However, retrospective recollection of the intervention	No – no clear evidence of self-reflexivity of the authors, however, when describing the iterative analysis process	No – no multiple perspectives, no participant involvement. No credibility checking for this piece of research	Maybe? Finding are questionable because they are based on recollections from more mature children/young adults, compared with younger children. Clear presentation – perhaps more examples in the results would be more beneficial.	Yes – first qualitative evaluation of CBT experiences. some useful practical suggestions for treatment considerations	Yes – ethical procedures were reported. Participants and the researchers had not met before interview Telephone interview for flexibility.	Yes – achieves what it sets out to achieve	High
Moulin, Akre, Rodondi, Ambresin & Suris (2015a)	Yes – this topic is relevant and provides interesting findings. It is novel in the way provides the viewpoint of the	No – numbers for a thematic analysis. Appropriate sample (age and parent-wise). Data translated from French to English – unclear if this translation was checked by anyone?	No - Not clear about how participants were allocated to which focus group or who lead them. No mention of	No – no information given on triangulation, multivocality or member reflections	Yes – interesting approach, clear presentation of results. The findings are generalisable to other European populations	Yes – results embedded in theory (social cognitive theory – focuses on benefits of peer-to-peer support. Methodologically	Yes – mentioned that ethical approval was sought and approved. Recruitment strategy may have been a bit	Yes – sets out achievable aims and seeks to meet them with appropriate methods and procedures and has interpretations and	High

	parents and the children and asked them separately, rather than putting them all in the same group which may leave some members disempowered	Nature of condition mentioned e.g. transient nature so that some declined because not affected and others declined because too severe. No field notes or reflexivity	reflexivity of the authors. Method and procedure were clear however, they gave participants an inventory to complete but gave no information about what the scores meant.			and Practically beneficial in terms of its contribution	invasive? Calling them after a couple of weeks if not opted in for the study. Detailed consent procedures	results that fit within the evidence base.	
Moulin, Akre, Rodondi, Ambresin & Suris (2015b)	Yes – this topic is relevant and provides interesting findings. Same as above. Also, interesting to see to familial viewpoint re service provision.	No – same as above. However, they address the difference between collecting data with the focus group and the two participants that had individual interviews	No - Not clear about how participants were allocated to which focus group or who lead them. No mention of reflexivity of the authors. Method and procedure were clear however, they gave participants an inventory to complete but gave no information about what the scores meant.	No – no information given on triangulation, multivocality or member reflections. However, in this second paper, slightly clearer on how consensus in themes was reached.	Yes – interesting approach, clear presentation of results. The findings are generalisable to other European populations	Yes – results embedded in theory (social cognitive theory – focuses on benefits of peer-to-peer support. Methodologically and Practically beneficial in terms of its contribution	Yes – mentioned that ethical approval was sought and approved. Recruitment strategy may have been a bit invasive? Calling them after a couple of weeks if not opted in for the study. Detailed consent procedures	Yes – sets out achievable aims and seeks to meet them with appropriate methods and procedures and has interpretations and results that fit within the evidence base.	High
Karterud, Risør & Haavet (2015)	Yes – useful to explore the experiences of young people with PNES but not sure if it was novel.	No – strange recruitment process, lack of clarity about reasons as to why they struggled to recruit and changed their recruitment planning. Inclusion of participants with epilepsy? Unclear how this would differ from PNES experience. Systemic text condensation – lack of explanation as to what this is?	maybe – no mention of the possible personal biases or reflexivity as to why the participants came to the themes they did. They were, however, transparent about their methodological process and the steps they took?	No – no mention of sharing their findings with participants or gathering their reflections. Some detail in parts, missing detail in others.	Maybe – the results are easy to read and easily digestible. The results are sensible and may be generalisable of the findings of this population, however, there are problems with recruitment so unsure how generalisable the findings are.	Yes – novel contribution to think about PNES from a biopsychosocial approach. Helped patients to understand the relationship between the body and the mind.	No – ethical approval was sought but no clarity on the ethical issues that were addressed.	Yes – contributes to the evidence base about people with PNES wanting to be believed and have their experiences validated. Their methodology was appropriate, but recruitment problems meant that conclusions drawn are questionable.	Low

McWilliams, Reilly, McFarlane, Booker & Hayman (2016)	Yes – novel, not much known about familial experiences of living with PNES	Maybe – Broad aims, appropriate sample and good UK context, like the ethics – could look up the details of analysis. Sufficient participant size. No field notes, however, interesting questions embedded in theory. lived experience by the research team from being in a family with NES.	Maybe – no self-reflexivity from the authors (except small mention about how the questions were developed) but detailed adaptations when some participants couldn't attend the focus group.	No – multiple researchers assigned the codes to the data and collaboratively they bought all the themes together. Researchers did not consider alternative voices	Only female participants Yes – clear aesthetic presentation. The authors make sensible extrapolations from the data they collected	No – not novel as it is described as 'one of the first studies to...'. But does contribute to evidence that discusses importance of language used with this population as well as service provision	No – project approval info given. No information on ethical processes or thinking. However, mention of adaptations made for children with LD. Inclusion of variety of caregiver.	Maybe – alludes to the idea of a 'rebranding' for people with NES. Achieves broad aims to explore the experiences of families with NES. Thematic analysis inhibits in depth exploration. Embeds the research in previous literature and explains future possibilities too.	Low
Mantilla & Rojas (2018)	Yes – novel in terms of using Explanatory models with conversion disorders but has been explored extensively with other diagnoses. It is also relevant as it highlights the need for greater understanding of these disorders.	Yes – transparency about research processes and methodology. Sample were representative of population however; it was a small sample for the chosen analytic methods and, they were specifically chosen for the research	Yes – authors note their decisions that would affect the rigor of their research and discuss their decision making with recruitment	No- mentioned earlier about wanting to do credible research but then not fulfilling criteria for credible research e.g. no consultation with participants, no member reflections	No – Poor presentation, no detailed examples to back up people's experiences. Some of the results focus on magical-mystical thinking – this is not generalisable to a UK finding	No – not really new findings – struggled to come to useful conclusions	Maybe – some consideration of the ethical processes but nothing about dissemination or sharing the research.	No – didn't necessarily get the findings that they hoped to achieve. Also, says it starts with grounded theory but then describes only thematic analysis, so methods may not be that appropriate as numbers too small, and inconclusive findings.	Low
Hulgaard, Rask, Risor & Dehlholm (2020)	Yes – worthy topic as not been examined from IPA perspective and interesting results about monocausal explanations	Yes - Pilot interviewed conducted with a youth and a parent. Questions embedded in literature – based on the CSM model. Good method with conducting the interviews and reviewing the transcripts.	No – no evidence of self-reflexivity. Not clear 'voice of the researcher'. Could have been more transparent about own biases/beliefs etc.	Maybe? – during the data collection and analysis – it was shared between multiple researchers. Themes reached together but not large amount of detail given about the how results were reached.	Yes – This is the first of its kind. Findings are presented in a clear and readable way	Yes- Practical contribution – highlights the role of communication in management of 'functional' conditions. Also has suggestions for the bringing	No – no mention of ethics processes available.	Yes – sets out aims of research and comes back to what it set out to achieve. The study's method and procedure are appropriate to meet the need of the study	High

However, no mention of fieldnotes.

Not gone back to the participants to check their findings

together of a paediatric perspective and a psychiatric perspective

Appendix C – Original Ethics Approval and Excerpts from IRAS form

A21. How long do you expect each participant to be in the study in total?

In terms of the interviews, my plan is that they take place at the convenience of the participants e.g. when they are already there for treatment. Ideally all interviews will be completed across the treatment period (up to a week). This will be organised at the conveniences of the family members and according to what they feel they are able to manage. In terms of study participation, I will give an overall period of 6 months, in case the family need to be contacted for further information or dates are not convenient. In terms of total involvement in the project as a whole, I would like to keep the family involved to feedback my findings until December 2020.

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

Time and Travel

Participants will be required to travel to and attend an University College Hospital London (where their routine appointments take place). This will involve using participants' personal time as well as finances with regards to travel costs. Not taking into account travel time to and from the research interviews, a maximum of 7 hours of participants' personal time will be required for participation in both the individual and collective interview.

Unfortunately, there is insufficient budget available for this piece of research and so participation is voluntary and travel costs cannot be reimbursed.

Personal and Sensitive nature of interview

The interview will involve asking participants about their identities, how they feel these are constructed in the context of illness and how they navigate different roles and relationships within their family units. People with PPS conditions often have a long and complicated relationship with the healthcare system. As such, the questions asked during interview may trigger difficult emotional responses for the participants.

With this in mind, the research interviewer is a Trainee Clinical Psychologist and she will use the clinical skills to support the participants in an empathic way and will create a supportive environment which may entail signposting to relevant organisations. The Trainee Clinical Psychologist has been working clinically in the NHS for 5 years and has worked with families and children before. Participants will also be able to decline to answer questions if they are too emotionally challenging. Additionally the interviews are taking place where their patient's clinical team are available to follow up, if necessary.

Whilst it is hoped that this will be sufficient in supporting participants through any emotional challenges during the research interviews, the interviewer can signpost candidates to access follow up support if required from the wider team and local counselling services.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

Yes No

If Yes, please give details of procedures in place to deal with these issues:

Topics discussed will be of a sensitive nature including; experiences of services, reflections on own illness and caring roles, loss and grief. As mentioned in the previous section, as a Trainee Clinical Psychologist I have skills in having emotionally challenging and sensitive discussions and am able to provide a space which is sensitive, empathic and containing for the families. During Clinical Psychology Training I have also worked with children and families with mental health difficulties. Additionally before training I have worked clinically with patients in the NHS for 5 years. I can also signpost them to their team psychologist or local counselling services if they do not want to discuss it within the team.

As with anything, there is a small chance of unexpected disclosures from participants about factors which might place themselves or others at risk. In both the participant information sheet and during informed consent the procedure to be followed in such an event will be explained to participants. Specifically where there is immediate risk participants will be supported to access A&E or the emergency services contacted. If risk is not immediate candidates will be encouraged to access support through their health care team. In the unlikely event of concerns regarding the safeguarding of others being raised, then the team around the child will be notified and information regarding any safeguarding concerns can be passed to relevant teams.

A24. What is the potential for benefit to research participants?

The process of the research interview will give the participants the opportunity to reflect on their experiences and lives with a professional who has specific training. It will also be beneficial to the participants to know that they may be involved in research that could become a stepping stone to changing how families experience treatment for PPS conditions. Additionally, it may open up discussions about alternative treatment options for participants.

A26. What are the potential risks for the researchers themselves? (if any)

It may that the researcher will be subjected to experiencing distress from the participant family. The researcher can seek support within the research team or the wider course team along with personal therapy if necessary. The research supervisors may seek support from their supervisors or line managers.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).

Following a discussion with the wider care team, they will identify families who may fit the inclusion/exclusion criteria and once identified, this pool of families will be sent the information sheet (or given it in their routine care appointments, whichever comes first) and will be asked to opt in via email to the research student. The participant family will be the first family who fits the study criteria and has the availability and willingness to participate in the study. Those who have opted in but not been chosen will be emailed thanking them for their interest and explaining the situation to them, with an opportunity to contact the researcher for more information if necessary. Once they have opted in, the research student will arrange a time to meet the family, in the clinic, at their convenience to discuss the study and gather informed consent.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

Yes **No**

Please give details below:

A28. Will any participants be recruited by publicity through posters, leaflets,

advertises or websites? Yes No

A29. How and by whom will potential participants first be approached?

The prospective participants will initially be approached by the clinical team. They will be informed about the study and then given the information sheet. They can then opt in to the study and after that they will be offered an appointment with me to further discuss the project and seek informed consent.

A30-1. Will you obtain informed consent from or on behalf of research participants?

Yes No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

Informed consent will be obtained by the researcher. I will meet with the family, discuss the information sheet and consent and withdrawal procedures with the family before they consent to go ahead with the study. The adults will sign the consent form. I will spend some time with the children to discuss the project and consent and I can ask the parents to explain the study and to consent on behalf of their children if under 16 years old. The Health Research Authority states when seeking consent from children there are a number of aspects that need to be considered. For example, a child or a young person's right to give consent is dependent on their capacity to understand the complexities of the research. I will ensure that the information is presented in a way that meets the child's intellectual ability and capacity. The younger children within the study, for example, the younger sibling, they would need to assent to the study, or give consent via their parents. I would also make the information accessible given that both adults and children will need to be suitably informed to consent to the study. Finally, I will explain that they can stop the interview at any time.

If you are not obtaining consent, please explain why not.

n/a

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

Yes No

A31. How long will you allow potential participants to decide whether or not to take part?

Participants will be allowed up to 2 weeks to decide to be involved in the project. This is because of the time constraints of the project as I may need to seek alternative participants.

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs? (e.g. translation, use of interpreters)

It is part of the inclusion criteria that the participants speak English and are able to adequately understand verbal and written information and explanations. This is a qualitative study which incorporates the specific use of language as a way of communicating meaning and sense-making, therefore it is important for the researcher and participants to be able to have the best chance of understanding each other without complicating this with the use of interpreters or translation services. Therefore, no arrangements have been made.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.

The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.

The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other

research procedures carried out on or in relation to the participant.

The participant would continue to be included in the study.

Not applicable – informed consent will not be sought from any participants in this research.

Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)? (Tick as appropriate)

Access to medical records by those outside the direct healthcare team

Access to social care records by those outside the direct social care team

Electronic transfer by magnetic or optical media, email or computer networks

Sharing of personal data with other organisations

Export of personal data outside the EEA

Use of personal addresses, postcodes, faxes, emails or telephone numbers

Publication of direct quotations from respondents

Publication of data that might allow identification of individuals

Use of audio/visual recording devices

Storage of personal data on any of the following:

Manual files (includes paper or film)

NHS computers

Social Care Service computers

Home or other

personal computers

University computers

Private company computers

Laptop computers

Further details:

Quotes from the data may be used in subsequent publications of the research. Participants will be informed of their right to ask for quotes to be removed from the data set. If quotes are used, these will be anonymised to protect the identity of the participant.

A37. Please describe the physical security arrangements for storage of personal data during the study?

The research student will only receive personal contact details for the prospective participants once they have opted in to participate in the study. They will opt-in by emailing the research student and further information will be shared via email. This information will be held separately from the data set and from any other information about participants. This information will subsequently be deleted following the completion of the interviews.

The research interviews will be audio recorded using a Dictaphone which only the research student has access to. On the same day that the audio recordings are made, they will be anonymised and stored electronically on a password protected laptop in a password protected file. The password chosen will be of high strength, using a combination of letters, numbers and symbols to ensure this. The 4 members of

the research team will be the only people aware of the password. Once stored electronically, the original audio recording will be destroyed.

The audio recordings will then be transcribed. Once transcripts have been completed, the audio recordings will be deleted. A transcription service may be used to transcribe the data. In this event there will be a confidentiality agreement in place with the transcriber.

The transcriptions, which will be anonymised, will be stored in a password protected document on the Research student's computer and the Academic Research Supervisor's university computer. Both computers are password protected with a high strength password.

A38. How will you ensure the confidentiality of personal data? *Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.*

At the recruitment stage, the research student will hold contact details for potential participants who have consented to sharing this information. These details will be stored a password protected word document on a password protected laptop. Once potential participants have been contacted and research interviews arranged and completed, their contact information will be deleted.

At the time of electronic storage the data will be anonymised and all participants allocated a unique pseudonym.

A40. Who will have access to participants' personal data during the study? *Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.*

The research student and external supervisor (Clinical Psychologist working within the service) will have access to the contact information of the potential participants. The external supervisor is embedded in the care team and when the participants opt in we will seek consent for their details to be shared with the research student.

A41. Where will the data generated by the study be analysed and by whom?

The data from the project will be analysed in the research student's home (in a private, lockable room) or in a private study room in the University of Hertfordshire Learning Resource Centre. Additionally, it may be that the data is analysed within the supervisory team's offices. The data will be analysed by the research student but may be viewed by members of the research and supervisory team.

The original audio data generated by the study may be transcribed using a transcription service. In this instance there will be a confidentiality agreement with the transcriber who will not have access to the data once it has been transcribed. If a transcription service is not used the student researcher will complete the transcribing of the data.

A74. What arrangements are in place for monitoring and auditing the conduct of the research?

Oversight of the quality assurance and governance will be provided by the University of Hertfordshire and data management will be undertaken by the research student and the chief investigator. The research student will also evaluate the research process on a day to day basis and seek guidance from the Chief Investigator who will also monitor and oversee this research.

The specific following steps are in place to help monitor and ensure this.

1. The research will be supervised and reviewed by Dr Jenna Harrington (Clinical Psychologist and Senior Lecturer). Dr Harrington works for the organisation (University of Hertfordshire) which has agreed to sponsor this piece of research.
2. The research will be supervised and reviewed by Dr Halina Flannery (Clinical Psychologist). Dr Flannery works in the psychological medicine department at University College London Hospital, the site in which this research will take place. Dr Flannery is able to provide particular support in ensuring that the

appropriate risk procedures are followed if required and that the research is conducted in an appropriate and ethical manner.

3. The research analysis will be supervised and reviewed by Dr Wendy Solomons (Clinical Psychologist and Deputy Senior Clinical Tutor). Dr Solomons works for the organisation (University of Hertfordshire) which has agreed to sponsor this piece of research.

3-2. Please describe the arrangements for seeking informed consent from a person with parental responsibility and/or from children able to give consent for themselves.

The family will be sent the information which has been co-created with a service user family (parent plus two children aged between 16 and 18) to ensure that it is age-appropriate. The glossary was added to ensure more difficult phrases could be understood. I will also meet the family to discuss the information sheet and check if they have any questions before they consent to be part of the study.

4. If you intend to provide children under 16 with information about the research and seek their consent or agreement, please outline how this process will vary according to their age and level of understanding.

There will be children under 16 years old. I have co-created the information with teenage service users so language should be appropriate for children but I will seek to explain the study to them in clear language and check their understanding so that they can provide informed consent to participate. However, this will be done in accordance with their parents in the room to ensure that they can support the child in comprehension of the information and perhaps provide consent on behalf of their child.

Copies of written information sheet(s) for parents and children, consent/assent form(s) and any other explanatory material should be enclosed with the application.



Dr J Harrington and Ms K Friedner
Department of Psychology and Sport Science
School of Life and Medical Sciences

University of Hertfordshire
Higher Education Corporation
Hatfield, Hertfordshire
AL10 9AB

Telephone +44 (0) 1707 284000
Fax +44 (0) 1707 284115
Website www.herts.ac.uk

14 February 2020

Dear Dr Harrington and Ms Friedner

Re: UNIVERSITY OF HERTFORDSHIRE SPONSORSHIP IN FULL for the following:
RESEARCH STUDY TITLE: Family Narratives of Lives Where Both Parent and Child
Have Been Diagnosed with Persistent Physical Symptoms (PPS) Conditions
NAME OF CHIEF INVESTIGATOR (Supervisor): Dr Jenna Harrington
NAME OF INVESTIGATOR (Student): Ms Kimberley Friedner
UNIVERSITY OF HERTFORDSHIRE ETHICS PROTOCOL NUMBER:
LMS/PGR/NHS/02942

This letter is to confirm your research study detailed above has been reviewed and accepted and I agree to give full University of Hertfordshire sponsorship, so you may now commence your research.

As a condition of receiving full sponsorship, please note that it is the responsibility of the Chief Investigator to inform the Sponsor at any time of any changes to the duration or funding of the project, changes of investigators, changes to the protocol and any future amendments, or deviations from the protocol, which may require re-evaluation of the sponsorship arrangements.

Permission to seek changes as outlined above should be requested from myself before submission to the Health Research Authority (HRA) NHS Research Ethics Committee (REC) and I must also be notified of the outcome. It is also essential that evidence of any further NHS Trust Management Permissions (formerly known as R&D Approval) is sent as soon as they are received. Copies of annual reports and the end of study report as submitted to the HRA also need to be provided. Please do this via email to research-sponsorship@herts.ac.uk

Please note that University Sponsorship of your study is invalidated if this process is not followed.

In the meantime, I wish you well in pursuing this interesting research study.

Yours sincerely

A handwritten signature in black ink, appearing to read 'J M Senior', with a horizontal line underneath.

Professor J M Senior
Pro Vice-Chancellor (Research and Enterprise)



**London - Central Research Ethics Committee**

3rd Floor, Barlow House
4 Minshull Street
Manchester
M1 3DZ

Telephone: 0207 1048 007

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

14 December 2019

Dr Jenna Harrington
University of Hertfordshire
College Lane Campus
Hatfield
AL10 9AB

Dear Dr Harrington

Study title:	Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions.
REC reference:	19/LO/1697
Protocol number:	n/a
IRAS project ID:	264193

Thank you for your response to the Committee's request for further information on the above research and for submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report

The latest guidance on these topics can be found at <https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/>.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites listed in the application subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

The favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [UH Professional Indemnity Insurance]		24 July 2019
Interview schedules or topic guides for participants [Interview Schedule]	Version 1	09 August 2019
IRAS Application Form [IRAS_Form_27092019]		27 September 2019
IRAS Checklist XML [Checklist_09102019]		09 October 2019
Letter from sponsor [Letter from Sponsor]		31 July 2019
Other [Response Table with REC recommendations]	Version 1	19 November 2019
Participant consent form [Adult Consent and Debrief form - tracked]	Version 3	19 November 2019
Participant consent form [Consent and Debrief form for 12 - 15 - tracked]	Version 2	19 November 2019
Participant consent form [Assent form for children for 12 – 15 year olds - tracked]	Version 2	19 November 2019
Participant consent form [Parent/guardian consent form for child aged 12 to 16 years old - tracked]	Version 2	19 November 2019
Participant information sheet (PIS) [Participant information sheet - tracked]	Version 2	19 November 2019
Referee's report or other scientific critique report [Peer Review]		11 July 2019
Research protocol or project proposal [Project Proposal]	Version 1	01 July 2019

Summary CV for Chief Investigator (CI) [Chief Investigator CV]		09 August 2019
Summary CV for student [Student CV Kimberley Friedner]		12 August 2019
Summary CV for supervisor (student research) [Supervisor CV Dr Halina Flannery]		12 September 2019
Summary CV for supervisor (student research) [Supervisor CV Dr Wendy Solomons]		12 September 2019
Summary of any applicable exclusions to sponsor insurance (non-NHS sponsors only) [UH Insurance Certificate]	Version 1	24 May 2019

Statement of compliance

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

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at: <https://www.hra.nhs.uk/planning-and-improving-research/learning/>

19/LO/1697	Please quote this number on all correspondence
-------------------	---

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

Yours sincerely



pp



Email: NRESCommittee.London-Central@nhs.net

Enclosure: "After ethical review – guidance for researchers"

Copy to:



Appendix D – Ethics Amendment Documentation

On 27 Mar 2020, at 09:52, SIMON-MODEBE, Eyoanwan (UNIVERSITY COLLEGE

LONDON HOSPITALS NHS FOUNDATION TRUST) <e.simon-modebe@nhs.net> wrote:

Dear Kimberley,

Project ID: 127429 (Please quote in all correspondence)

IRAS ID: 264193

REC Ref: 19/LO/1697

Title: Family Narratives of Lives Where both Parent and Child have PPS

Amendment: NSA

Confirmation of Amendment Capacity & Capability

The UCLH/UCL Joint Research Office (JRO) acknowledges receipt of the above non-substantial amendment.

We have reviewed the amendment and the HRA Approval email dated 13/03/2020.

The JRO has no objections to this amendment and the study may continue at UCLH.

If applicable, you must ensure that you localise all patient facing documentation prior to consenting participants; this will be subject to random audit checks.

Please forward this email on to all relevant parties involved with this study at UCLH.

Please insert a copy of this email in your site file.

Best wishes with your research.

Kind regards,

JRO Amendments Officer

Joint Research Office, Research Management and Governance

From: Research Sponsorship <research-sponsorship@herts.ac.uk>

Subject: JH-KF NS UH Protocol number: LMS/PGR/NHS/02942 NSA1

Date: 20 March 2020 at 12:21:36 GMT

To: "Kimberley Friedner [Student-LMS]" <kf17aat@herts.ac.uk>, Jenna Harrington <j.harrington@herts.ac.uk>

Cc: Halina Flannery <halina.flannery@nhs.net>, Helen Ellis-Caird <h.ellis-caird@herts.ac.uk>, Research Sponsorship <research-sponsorship@herts.ac.uk>

Dear Kimberley and Jenna,

Further to receipt of your completed SP2 form, this is to confirm approval for notification of the amendment, reference NSA1. When you receive acknowledgement of the notification from the Health Research Authority, you must inform research-sponsorship@herts.ac.uk so continued University sponsorship of this research project can be confirmed.

Regards, Ellie

Ellie Hubbard

Research Information and Governance Manager & Deputy REF Manager

Research Office

Appendix E – Information Sheet

PARTICIPANT INFORMATION SHEET

Version 2_19/11/19

IRAS ID: 264193

Title of study

Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions.

Introduction

You are being invited to take part in a study. Before you decide whether to participate, it is important that you have a clear understanding about what the research entails and what your involvement means. Please take the time to read the following information carefully and discuss it with others if you wish. Do not hesitate to ask us anything should you require further information to help you make your decision or something is not clear.

Thank you for taking the time to read this.

What is the purpose of this study?

The purpose of this study is to look at the experiences of family members where both a parent and child have PPS (which is an umbrella term for a selection of diagnoses such as chronic fatigue syndrome (CFS), irritable bowel syndrome (IBS) and fibromyalgia). Within families, members have certain roles which change and develop over time. These roles, amongst other things influence the development of our identity. The study will focus on understanding how family members develop their identities and negotiate their roles and relationships within the family unit, in the context of difficult to explain illness. The study will also seek to explore the conversations which get prioritised and those that don't get discussed within the relationships. It will also be looking at understanding the family's collective experience as well as the experiences of the individuals within the family. This will give people the opportunity to offer any different perspectives they may hold. Shining a light on the family's perspective may offer opportunities for the future.

Do I have to take part?

Participation is completely voluntary, and it is entirely up to you if you decide to take part. If you do, you will be given this information sheet to keep and be asked to sign a consent form. However, if at a later date you decide you do not want to continue you can withdraw up to six weeks after participation, without having to provide an explanation. If you do withdraw from the study during the interviews, then data collected until the point of withdrawal may still be used.

Are there any age or other restrictions that may prevent me from participating?

In order to participate in the study, you will need to be over the age of 12 years old. You will also need to be able to speak English fluently, be living in the UK, and have no current safeguarding issues (such as concerns about safety e.g. risks to self or others or other social vulnerabilities). Lastly, you will need to be able to manage an interview of at least 45 minutes. Additionally, in order to participate, you will need to be part of a family where both a parent and a child in the family unit have PPS conditions. There needs to be a minimum of two parents and two children in the family unit.

How long will my part in the study take?

If you decide to take part in this study, you and your family will be involved in it for one interview which will last a minimum of 45 minutes, and then each family member will have an individual interview lasting up to an hour (or whatever can be tolerated).

What will happen if I agree to take part?

The first thing to take place is that we will arrange a time which is convenient for you and your family to take part in the interview. For convenience, the interview can take place at the hospital on a day you are already visiting for your clinic appointment. We will ensure that it is in a space both private and available at the time that we want to meet.

Firstly, we will meet to complete the family interview. Prior to commencing the interview, I will talk through the structure of the interview, what to expect, and answer any questions you may have. After the family interview, I will interview each family member, perhaps later in the day or on another day, depending on the individual's capacity and tolerance.

During the interview I will ask each family member a number of questions about your experiences, and I will record the responses. The interviews will be audio-recorded so that I can analyse your responses at a later time. At the end of the interview I will provide you with further information about the study and other areas of information and support which you may find helpful, as well as my contact details should you have any further questions.

What are the possible disadvantages, risks or side effects of taking part?

The only possible risk identified is that you may find the process of reflecting on and discussing your experiences generates an emotional response. Although this can be a normal response, some people may find it distressing to experience strong emotions while participating in an interview. Should this occur, I will check with you about whether you feel able to continue, or whether you would like to have a break or to discontinue the interview. You will not be expected to talk about anything that you do not wish to talk about and can choose to stop the interview at any time. After the interview, I will provide information on relevant support agencies which may be beneficial if you wish to access additional support.

What are the possible benefits of taking part?

There is no known benefit to taking part though the participant may find it helpful in reflecting on and exploring their experiences.

How will my taking part in this study be kept confidential?

The interviews will happen in a location which can maintain your privacy, with only the researcher and participant being present. Your personal data will be treated carefully – you will be assigned a participant identification number which will be used instead of your name to maintain confidentiality. Additionally, information with your name included, such as consent forms, will be kept separately from the interview data, so that anonymity is preserved. Hard copies of written data (e.g. signed forms) will be kept in a locked filing cabinet. Electronic data will be held securely in password protected files, on either a password protected computer or saved on an encrypted external hard drive which will also be kept in a locked filing cabinet. These will only be accessible by the researcher. Access to viewing the data will only be available to the researcher and the researcher's supervisory

team. The data will be deleted or destroyed fifteen years after completion of the project, which is in line with recommendations from the Research Ethics Committee.

How will we use information about you?

GDPR stands for the General Data Protection Regulation. In the UK we follow the GDPR rules and have a law called the Data Protection Act. All research using patient data must follow UK laws and rules. Everyone involved in this study will keep your data safe and secure. We will also follow all privacy rules. The personal information used will include your name and email address. The research team will use your personal information to contact you to participate in the research. As detailed above this will involve interviewing you about your illness and your life as a family. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. As discussed above we will keep all information about you safe and secure. Once we have finished the study, we will write our findings in a way that no-one can work out that you took part in the study.

What will happen to the data collected within this study?

The data recorded will be held securely, as stated above, for up to fifteen years following the completion of the project, in line with British Psychological Society Guidelines and recommendations from the London Central Research Ethics Committee. At the end of this period, written materials will be destroyed, and all electronic data will be deleted. Should you wish to withdraw your data from the study, you are required to inform the researcher within six weeks of completing the interview.

The results will be used for the researcher's Doctorate in Clinical Psychology thesis and may potentially be used for journal publications and conference presentations. Any extracts of interview transcripts in the research report or any publications will be fully anonymised.

Feedback on the results of the study will be made available upon request.

It will not be possible to pay any travelling costs involved in taking part in the study.

Ethical approval for this study has been obtained from the NHS Ethical Approval team – REC reference number **19/LO/1697**.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team
- by sending an email to kf17aat@herts.ac.uk or j.harrington@herts.ac.uk

Who can I contact if I have any questions?

If you would like further information or would like to discuss any details personally, please get in touch with me, in writing or by email:

Kimberley Friedner
Department of Clinical Psychology
Health Research Building
University of Hertfordshire
Hatfield
AL10 9AB

kf17aat@herts.ac.uk or k.friedner@herts.ac.uk

You can also find more information at www.hra.nhs.uk/information-about-patients/

Although we hope it is not the case, if you have any complaints or concerns about any aspect of the way you have been approached or treated during the course of this study, please write to the University's Secretary and Registrar at the following address:

Secretary and Registrar
University of Hertfordshire
College Lane
Hatfield
Herts
AL10 9AB

UCLH Patient Advice & Liaison Service (PALS)

Address: PALS, Ground Floor Atrium
University College Hospital
235 Euston Road
London NW1 2BU

Telephone: Main Hospital: 020 3447 3042

Email: uclh.pals@nhs.net

Thank you very much for reading this information and considering taking part in the study

Appendix F – Consent Forms (consent and assent)

ADULT CONSENT FORM

Version 3_191119 IRAS ID: 264193

I, the undersigned [*please give your name here, in BLOCK CAPITALS*]

.....

of [*please give contact details here, sufficient to enable the investigator to get in touch with you, such as an email address*]

.....

hereby freely agree to take part in the study entitled “Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions”.

Please read the following statements before you agree to take part in this study.

1) I confirm that I have read and understood the participant information sheet and I understand what my participation in this study involves.

<input type="checkbox"/>
<input type="checkbox"/>

Yes

No

2) I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason. If I withdraw from the study before six weeks of when the interview has taken place, the data that I have submitted will also be withdrawn at my request.

<input type="checkbox"/>
<input type="checkbox"/>

Yes

No

3) I understand that the information that I will submit will be confidential and anonymous, used only for the purpose of this study

<input type="checkbox"/>
<input type="checkbox"/>

Yes

No

4) I agree that research data gathered for the study may be published and if this occurs all precautions, such as removing any identifiable information (names, ages etc) will be taken to protect my anonymity.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

5) Contact information has been provided should I wish to seek further information from the investigator at any time for purposes of clarification.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

6) I understand that my interview will be audio taped.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

7) I agree to take part in the above study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

Signature of participant: _____

Date:

Signature of (principal) investigator: _____

Date:

(KIMBERLEY FRIEDNER)

CHILD AND ADOLESCENT ASSENT FORM (AGE 12 – 15 YEARS OLD)

Version 2_19119

IRAS ID: 264193

I, *[insert name here, in BLOCK CAPITALS]*,

.....
child of *[insert parent's name here in BLOCK CAPITALS]*,

.....
who can be contacted at *[please give parent/guardian's contact details here, e.g. an email address]*

.....
freely agree to take part in the study entitled "Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions".

Please read the following statements before you agree to take part in this study.

1) I confirm that I have read and understood the participant information sheet and I understand what my participation in this study involves.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

2) I understand that taking part is voluntary and I can stop participating at any time, without giving a reason. If I leave the study before six weeks of when the interview has taken place, the data that I have submitted will also be removed at my request.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

3) I understand that the information that I will submit will be kept confidential and anonymous, used only for this study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

4) I agree that research data gathered for the study may be published and if this occurs all precautions, such as removing any identifiable information (names, ages etc) will be taken to keep my personal information private, confidential and keep me anonymous.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

5) Contact information has been provided should I wish to seek further information from the investigator at any time for purposes of clarification.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

6) I understand that my answers in the interviews will be audio taped and give permission for this.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

7) I agree to take part in the above study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

Signature of participant: _____

Date:

Signature of parent/ guardian: _____

Date:

Signature of (principal) investigator: _____

Date:

(KIMBERLEY FRIEDNER)

CHILD AND ADOLESCENT CONSENT FORM (AGE 12 – 15 YEARS OLD)

Version 2_19119

IRAS ID: 264193

I, *[insert name here, in BLOCK CAPITALS]*,

.....
child of *[insert parent's name here in BLOCK CAPITALS]*,

.....
who can be contacted at *[please give parent/guardian's contact details here, e.g. an email address]*

.....
freely agree to take part in the study entitled "Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions".

Please read the following statements before you agree to take part in this study.

1) I confirm that I have read and understood the participant information sheet and I understand what my participation in this study involves.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

2) I understand that taking part is voluntary and I can stop participating at any time, without giving a reason. If I leave the study before six weeks of when the interview has taken place, the data that I have submitted will also be removed at my request.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

3) I understand that the information that I will submit will be kept confidential and anonymous, used only for this study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

4) I agree that research data gathered for the study may be published and if this occurs all precautions, such as removing any identifiable information (names, ages etc) will be taken to keep my personal information private, confidential and keep me anonymous.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

5) Contact information has been provided should I wish to seek further information from the investigator at any time for purposes of clarification.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

6) I agree to take part in the above study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

Signature of participant: _____

Date:

Signature of parent/ guardian: _____

Date:

Signature of (principal) investigator: _____

Date:

(KIMBERLEY FRIEDNER)

PARENT/ GUARDIAN CONSENT FORM (FOR CHILD AGED 12 – 15 YEARS OLD)

Version 2_19119

IRAS ID: 264193

I, *[insert name here, in BLOCK CAPITALS]*,

.....
parent of *[insert child's name here in BLOCK CAPITALS]*

.....
who can be contacted at *[please give contact details here, e.g. an email address]*

.....
freely agree for my child to take part in the study entitled "Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions"

Please read the following statements before you agree to take part in this study.

- 1) I confirm that I have read and understood the participant information sheet and I understand what my child's participation in this study involves.**

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

- 2) I understand that my child's participation is voluntary, and they can stop participating at any time, without giving a reason. If they leave the study before six weeks of when the interview has taken place, the data that they have submitted will also be removed at my/their request.**

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

- 3) I understand that the information that my child will submit will be kept confidential and anonymous, used only for this study.**

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

- 4) I agree that research data gathered for the study may be published and if this occurs all precautions, such as removing any identifiable information (names, ages or anything obviously identifiable etc) will be taken to protect my child's anonymity.**

<input type="checkbox"/>	Yes
<input type="checkbox"/>	

No

5) Contact information has been provided should my child or I wish to seek further information from the investigator at any time for purposes of clarification.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

6) I understand that my child's interviews will be audio taped and give permission for this.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

7) I agree for my child to take part in the above study.

<input type="checkbox"/>	Yes
<input type="checkbox"/>	No

Signature of parent/ guardian: _____

Date:

Signature of (principal) investigator: _____

Date:

(KIMBERLEY FRIEDNER)

Appendix G – Debrief form

Debrief form - Version 2_19119 IRAS ID: 264193

Dear Participant,

Thanks for taking part in the study “Family Narratives of Lives Where both Parent and Child have been diagnosed with persistent physical symptom (PPS) conditions”. The information that you have provided will be kept confidential and all personally identifiable data will be destroyed after the completion of the research. You can ask to have your contribution removed from the study without giving a reason up to six weeks after participation.

1. What are the aims of the study?

To understand how family members, create their identities and navigate roles and relationships within a family where both parent and child have PPS.

2. What if I have any questions about the study that I would like to ask now?

Please contact the researcher Kimberley Friedner at kf17aat@herts.ac.uk or k.friedner@herts.ac.uk

3. How can I contact the researcher if I have any further questions or if, for any reason, I wish to withdraw my data once I have left?

Please contact the researcher Kimberley Friedner at kf17aat@herts.ac.uk or k.friedner@herts.ac.uk

4. Can I obtain a summary of the results of the study? What form will this summary take?

To obtain details of the results of the study, which will take the form of a written report, please contact the researcher Kimberley Friedner at kf17aat@herts.ac.uk or k.friedner@herts.ac.uk

If the study has raised personal issues that you are not comfortable discussing with the researcher now – what should you do? Please seek advice and support from the following support networks included below.

Your local GP

UCLH Patient Advice & Liaison Service (PALS)

Your local IAPT service

Address: PALS, Ground Floor Atrium

The Samaritans Telephone: 116 123

University College Hospital

The doctors in your clinical team

235 Euston Road

London NW1 2BU

Telephone: Main Hospital: 020 3447 3042

Email: uclh.pals@nhs.net

If you have concerns about this study, or the way in which it was conducted, please contact the Kimberley Friedner (Principal Investigator) at kf17aat@herts.ac.uk or k.friedner@herts.ac.uk

or Dr Jenna Harrington at j.harrington@herts.ac.uk.

Thank you again for your participation and support.

Appendix H – Emails with Georgina and Gemma re ‘MUPPETS’

On 28 May 2019, at 19:58, Gemma Smith <Gemma@Smith.co.uk> wrote:

Hi Kimberley

It was nice to meet you too, and for Georgina and I to feel that our experiences can be used to help others.

Georgina hasn't been too well since coming back from UCLH last week so we'll need a few days to review what you've sent.

I would like to ask though – does the term ‘Medically unexplained symptoms’ have to be used?

A number of families have had this term used in a derogatory way – that their/their child's symptoms are as a result of some mental disorder that they should be able to pull themselves together and recover from, often by ignoring their perceived pain/fatigue and getting back to school/work.

There was also a large sharing on social media of the term when used as shown – being thought of as ‘Muppets’ was very hurtful – an apology was given but the association between the term and the perceived insult still

exists. <0838B8805FBE43C1AB1A0793CA5B2D23.png>

The concern among families is that anyone using that term could be looking for a way to dismiss/downgrade genuine physical medical symptoms – either because they don't have specialist knowledge are reluctant to refer to another specialist or reject the diagnosis made by another specialist if it's a condition that they ‘don't believe in’ or the diagnosis was obtained through private medical care I.e. the patient ‘bought’ the diagnosis.

I would hate people not to take part in your research because a set of words makes them too worried to.

Could an alternative phrase be used such as both parent and child have ‘long term’ or ‘chronic’ illnesses or conditions?

Hope that makes sense!

Kind regards

Gemma

From: [Kimberley Friedner \[Student-LMS\]](#)

Sent: 27 May 2019 22:24

To: Gemma@Smith.co.uk

Subject: Service User Consultant for University of Hertfordshire Trainee Clinical Psychologist.

Hi Gemma,

It was lovely to meet you both last week and thank you once again for being so open and offering you support for the project.

I wanted to briefly summarise our meeting and attach the information sheet and consent form for you and Georgina to review.

I shared with you my rationale for the project and a brief explanation of the what the project will entail. We discussed some of your family's experiences with these diagnoses and the importance of language around diagnosis. I shared with you that I would like your help with ideas around recruitment, discussing at which point in patient's journeys would people prefer to be contacted and we agreed that later was better. I asked for your and Georgina's feedback on the information sheet and consent form which I attach to this email. I would also like to ask what you both think of the wording of the title of the project. My current working title is

"Family narratives of lives where parent both parent and child have medically unexplained symptoms".

Once again, thank you for interest and help with this project. I look forward to working with you on the design of this project and receiving your feedback. If you could do track changes on a word document and add in any comments or questions you have I can try my best to address your suggested changes.

Take care!

Kimberley (Friedner)

Trainee Clinical Psychologist

Appendix I – Service user bios from Gemma, Georgina and Greg**UNIVERSITY OF HERTFORDSHIRE****PARTICIPANT INFORMATION SHEET**

This information sheet has been co-created with my service user consultants; a mother and her son and daughter who all experience persistent physical conditions. They wrote bios to explain their rationale for involvement with the research:

I am a parent of two teenagers who both developed complex long-term health conditions in childhood. When you become a parent, you take on the role of nurturer, teacher, protector and provider. As the parent of children with health conditions that aren't easily explained you can add to your roles those of medical detective, biological researcher, and advocate as well as those of nurse, physiotherapist and counsellor. Having your own health concerns can mean you feel empathy for what your children are experiencing, guilt that you can be limited in the care that you're able to provide as well as worrying that the concern that your children naturally feel for you is adding to their difficulties. Many times, over the years each of us has been helped by support from Psychology services and we hope that research such as this can help families like ours get the most effective support.

[My Daughter] developed a medical condition, with a clear diagnosis and treatment pathway, in 2009 and since then has developed many additional complex conditions which have been challenging to identify and treat. Her experience is that to live the best life with such conditions you need a good support network of medical professionals, friends and family. She has acted as a consultant to this research in the hope that it will help medical professionals understand the impact of these types of conditions on all members of the family, and how best to support such families.

[My son] has had a complicated medical history since 2008. His condition has affected his physical abilities and caused him to experience higher levels of fatigue, along with a variety of

other symptoms. He hopes that this research will benefit people like him and result in a better understanding of the wider effects of these conditions such as those on the family unit.

Appendix J – Interview Schedules

Group Interview schedule

Informed by Wells (2011) and Bamber (2014; unpublished thesis). It is important for a few open-ended questions, followed up by curious, conversational questions.

“I am interested to hear your stories of coming to live with PPS. There is no right or wrong, and I am interested in everyone’s views. Please be aware that we can stop at any time and I would like to ensure that we take breaks at your convenience.

“Before we start thinking about health, tell me something broadly about how you are as a family”.

“Can you tell me how health difficulties became a problem/or entered your lives, in this family?”

“And how did that continue...?”

“Who else got involved?”

“Can you tell me about your experiences of being in a family where both parent and child have persistent physical symptom conditions?”

“When these symptoms first became difficult how did that influence the way you functioned as a family over time? How did that change from before?”

“How do things change when [insert family member] became unwell?”

“Has anything changed since then?”

“Any critical things that have changed?”

“How do you make sense of your own illness?”

“How do you make sense of your own illness in the context of your parent/child being unwell?”

“Can you tell me about the roles that you each take in your family?”

Can you tell me some stories about how your family functions?”

“How has this affected how you’ve come to think of yourself as a family? And how has this changed over time?”

Individual Interview schedule

I am aware I may be asking questions similar to those I asked in the group interview, but I am also aware that you may want to say something when there are fewer people around.

“How was the group interview for you?”

“Were there points when you were listening to [insert family member] telling that story that felt different to how you remembered it?”

“Can you tell me about a time you were unwell in your family?” and another time...

“How did this start? Can you remember any specific things happening at the time?”

Can you tell me about how [insert diagnosis] entered your life?

Can you tell me about a time of when you first noticed something was wrong?

Can you tell me about a time when you were unwell in this family?

- *Can you tell me a time of/What happens when the unwell feel well and vice versa?*

Can you give me examples of times when your parent/child/sibling was unwell?

“How do you describe [your family member]?”

“How do you relate to [your family member]?”

Appendix K – Transcription Symbols

Transcription symbols used (adapted and simplified from Jefferson, 2004)

Transcription Symbols	Examples	
(2) (#) (.)	“I kind of don't trust (#) in teachers to help me from that first school”.	Different numbers in brackets denotes length of pause, (#) denotes
:::colon	B: there's always a lot more er s::::ensitivity	Indicates an elongation of previous letter
Hyphen -	A: (#) and of peep- (.) where we were ill	Indicates a broken off word
.hehe or haha	B: .hehehe so what you talking about the dynamic(?)	Indicates laughter (full stop before indicates in breath before laughter). The more haha the louder/stronger the laughter
CAPITALS	B: in a <u>wheelchair</u> when I was older (.) and I was SHITTING MYSELF	This indicates talk that was louder than the surrounding words
<u>underlined</u>	A: I felt <u>really sick</u>	This indicates emphasis on a word
“speech marks”	A: "Yes (.) yes (.) rather (.) yes (.) I think you're right"	Speech marks indicate when a person is quoting someone else.
(xxx)	A: Have to just (xxx) really	Inaudible speech
[square brackets]	A: [Okay-] B: [But]	Square brackets indicate when speech overlapped between people e.g. different participants or participant and researcher
= equals sign	A: from both sides↑= B: =Yeah	This indicates ‘latching’ when there is not noticeable difference between what one person says and
↑	A: I'm very clumsy↑	Indicates a rising tone of voice
° Degree signs °	B: °its not as bad anymore°	Degree symbols denote a lowering or quietening of speech
(?) (!)	A: Not particularly no(!)	The uses of these bracketed symbols indicate tone e.g. a questioning tone (?)
[text in square brackets]	S: But yeah- [dismissive tone]	Notes from interview: non-speech elements

Appendix L – Examples of Reading for Content

- disappointment
- dismissed by healthcare
- Disruption to schooling
- doctor's advice
- dynamics
- Easier than siblings
- Education
- empathy
- encouragement
- escape
- exercising

F 13:36: we didn't know any that- We didn't know what was going on wasn't right when you speak to health erm visitors and midwives and that they just say "oh your baby's colicky or your baby's just fussy" and there was nothing identified. So we then had Beau, who couldn't, couldn't keep food down and couldn't poo or didn't move normally and screamed all the time and it was a lot of trouble. When we got referred to the paediatric probably= I don't know. I wonder what this was like, in forming their parental identities?

J 14:20: =we tried to get him to eat. content: explicitly talking about experiences of being dismissed.

all 14:22: hahaha

F 14:24: Was it about 10 months, was he about a year old or was not quite yet. He sat up

Amelia

Select options for displaying the references in a node

A 07:27: But if we will go out and then (.) I look at other people and how like they've, they (.) um got like really hyperenergetic and then they're fine it just and (.) then how like my friends (.) um (.) they always (.) um (.) notice one girl who used to like say that, (.) uh (#) because I used to be off school a lot, she, (xxx) "oh you're always off school of are you just like lying" and stuff like because I didn't look unwell!

R 07:56: Yeah

A 07:57: but °I found it quite hard. Children can be mean, a shame to be treated this way - would she have seen how her siblings were treated? It's quite,

R 08:03: Yeah, that's what I mean. Content - speaks to difficult relationships with school children but also difficult times at school.

A 08:07: Yeah.

R 08:08: yeah And can you tell me a story about what happens when you're feeling well in your family? What's that like?

Appendix M – Examples of Reading for Structure

Florence

F : I talk to a doctor... was, I'm a senior... know, when you r... come along going... HHHHHHHH reas... know I was talking... many people that... important. So I fo... London hospital. I... loads of bowel pro... was probably 12 r... and we were unde... to a doctor who's... really good stuff w... that we give her a... chance to repair a... knowing what I kn... fix its going to be

S 07:55: er:::

B 07:55: [close]

A 07:55: [close] [hahaha] ((laugh)). Connected

F 08:02: [hahaha] connected, Ok

R 08:04: Yeah?

S 08:06: yeah

F 08:08: supportive of each other, thoughtful

R 08:11: mm-hmm

B 08:15: there is a bit of friction though sometimes isn't there

F 08:15: was ne worried that it wasn t going to get mentioned.

S 08:15: Structure component: he had mentioned it earlier and it didn't get picked up, he was sure to bring it in again to ensure this aspect of his story is picked up

R 08:15: ch other sometimes. Stuff like that.

Annotations:

- Created: 9 Aug 2020 By: KF
Structural component - the use of mimicry and voicing to continue the story of asserting her authority, which began at the beginning of the group interview when she showed her authority with me as the researcher
- Created: 2 Jul 2020 By: KF
reported speech

Group Interview

S 07:55: er:::

B 07:55: [close]

A 07:55: [close] [hahaha] ((laugh)). Connected

F 08:02: [hahaha] connected, Ok

R 08:04: Yeah?

S 08:06: yeah

F 08:08: supportive of each other, thoughtful

R 08:11: mm-hmm

B 08:15: there is a bit of friction though sometimes isn't there

F 08:15: was ne worried that it wasn t going to get mentioned.

S 08:15: Structure component: he had mentioned it earlier and it didn't get picked up, he was sure to bring it in again to ensure this aspect of his story is picked up

R 08:15: ch other sometimes. Stuff like that.

Appendix N – Examples of Reading for Performance and Context

Coding Detail View MRP2020

Jason

Q (1)

J 01:41: Yes. Well, well, I mean, I think um I think a lot of, okay there's some elements of illnesses that aren't mental health mentally health wise, contractable for there's a lot out there. I believe that is, I think mental mental health has a lot to do with somebody's well being.

R 01:58: Exactly, condition, it's only

J 02:07: Yeah. But different situations

R 02:17: no, exact share a context?

J 02:27: yes,

R 02:27: you're all different individuals. But

J 02:29: yeah, yeah,

Created: 19 Jun 2020 By: KF

What does he think about mental health?

Positioning: Who does he think I am as a mental health professional? Is he talking to me as a psychologist or a res...

Delete

Coding Density

MRP2020

Beau

R 28:21: Yeah,

B 28:22: as I go see that kind of erm, you know, have you ever heard of erm booming and busting?

R 28:26: mm-hr

B 28:27: It's like they all want to the energy to do trying to stop it really bad, if it h started off with l Didn't matter ho

Created: 25 Jun 2020 By: KF

expert positioning - he is educating me about psychological ways of coping.

Delete

you know, it's like, s, they don't have ort of erm feeling of know, even if it gets (.) Last year I ry night you know, I tried to encourage the girls to do the same as I did, but erm (#) they tried it once, and they couldn't do it. And it was like, I remember this one time when Summer. She was feeling like, err really well, and she was getting really confident. And erm she, (.) my auntie, at the time she was preparing for a marathon. So er she was doing this running thingy every day. And then err Summer she thought, she'd tried the same thing. But she started off way too strong on the erm app. She

Appendix O – Examples of Story Construction

The screenshot shows a software interface with a sidebar on the left and a main content area on the right. The sidebar has several sections: CODES, CASES, NOTES, SEARCH, and MAPS. The main content area shows a list of nodes, with one node selected and its details displayed below. The selected node is titled 'Florence' and contains a large block of text with several lines highlighted in green. A tooltip is visible over the text, providing a description of the node's content.

CODES

- Nodes

CASES

- Cases
- Case Classifications

NOTES

- Memos
- Annotations
- Memo Links

SEARCH

- Queries
- Query Results
- Node Matrices
- Sets

MAPS

- Maps

1 item selected

Florence

Q~ "

F: There's a lot of there is a lot of frustration in it because there's, you know, they can't do (. .) that you wish that they could do things they can't (. .) access but you wish that they could access, you know, like, erm Beau, you know, he's a really talented sportsman, but actually he can't like football. He can be (. .) amazing, but he can't play football because obviously he does play football, but he can't play for club because of the, you know, injury- injuries, he picks up and the the fatigue from that full on of level of training would have a knock on effect on other things. So it's things like that they can't just go and do because it will (xxx) You know Summer was FANTASTIC erm, she used to play clarinet and she was fabulous at that and then she had to stop because of her lungs. So stuff like that is a bit that is a bit frustrating, you think, well that's not fair. But then equally, well okay, maybe those things are not fair. They have (xxx) it is what it is. It's frustrating because represent their int because of their a finally getting part top of it all. But the appropriate resou they're not not ab just like, accept the because I can't change it. And if those that have the power to change it aren't willing or able to change it, then you just have to work around it in sort of the more

Created: 9 Aug 2020 By: KF

A story of loss - how Florence describes what her children have lost as a result of illness

Delete

The screenshot shows a software interface with a sidebar on the left and a main content area on the right. The sidebar has sections: Coding, Detail View, and MRP2020. The main content area shows a list of nodes, with one node selected and its details displayed below. The selected node is titled 'Beau' and contains a large block of text with several lines highlighted in green. A tooltip is visible over the text, providing a description of the node's content.

Coding

Detail View

MRP2020

Beau

how we were talking about how you know, whenever you do the dishes, it's like, how one person might not feel well, so they won't do it so then someone else has to fill in. Yeah, well, sometimes, you know, one person might not feel well, but the others don't feel well too you- but they can't say anything because then you know, (#) there won't be enough people to keep functioning you know

R 23:57: and Is th feel like your siste or say I kind of ho

B 24:12: yeah, ye

R 24:12: Um, can felt you weren't al

Created: 25 Jun 2020 By: KF

mentioned that you you can't speak up

Is this part of the story of family unity? There is a sense that there is interdependence within the family system. The collective need is greater than the individual

Delete

B 24:21: Um, that someone actually happened yesterday. And er it was like, they were going out for a walk. And erm (. .) I really wasn't feeling up to it. (. .) And er I just remember like really wanting to say that I REALLY did not feel up to it. And then it was like, as soon as I ac-actually said something, (. .) and then as soon as I said it, I'm out of my my siblings are normally the

Appendix P – Samples of Reflexive Diary Entries

Reflections on the ethics process prior to REC meeting.

The NHS ethics process has been really challenging. I felt quite alone when going through all the steps of filling out the IRAS form, negotiating what must be done between university ethics and NHS ethics and then preparations required for the upcoming REC committee meeting. However, I've learnt lots of skills that will be useful should I want to conduct research in the future. I have used supervision as a supportive space for this difficult process. I am particularly anxious about the REC meeting coming up as now, it may be that the research doesn't get approved from an ethical perspective. I don't know what I'll do if that's the case! My supervisor advised that I start to think about an alternative project, perhaps from staff perspectives but I want to pursue my project for as long as possible. I can only prepare as best as possible to answer all their questions and defend my project.

Reflections on recruitment process

COVID-19! I thought I'd faced all the hurdles with all the challenges with preparing the ethics form, the REC meeting and responding to the REC. Recruitment was always going to be a challenge but I didn't anticipate that I would have to deal with a global pandemic inhibiting my recruitment for my MRP. While we'd sent out the information sheet a couple of weeks before lockdown, two families had declined to participate for various reasons. My anxiety increasing as time ticks on. The meetings we've had as a supervisory team have left me again with fear for the future of my project but also doubts about moving forward with a family with a diagnosis (EDS) that doesn't necessarily fit the inclusion criteria for this study. I had to weigh up whether to move forward with my supervisor's suggestion to go with this family (as well as the ethical issue of not accepting a family that were keen to take part in the research) or wait until a potential family stepped forward that fit a more mainstream example of PPS.

Reflections after Summer's interview

I felt so anxious after this interview, it was very short, and her bluntness left me feeling a bit intimidated. Jason and Beau had been so chatty and had been very willing to answer my questions with story after story. Summer's interview had followed, and her family had spoken about her role as being integral to family wellbeing. I found that it was hard to engage her in some extensive storytelling and she appeared resistant to my questions. I found her hard to contend with and it seemed like she wanted it to be over as soon as possible. I was worried whether I would firstly have enough data from the short interview but also if the rest of the interviews would go the same way. I found it emotionally difficult and it made me feel sad when Summer explained that she was bullied by some girls at school. She was closed off about this, but my curiosity wanted to explore this in a bit more detail. However, I was aware of juggling my researcher and clinician hat where, if I were wearing the latter, I would dig and explore (where appropriate) but I was aware that I didn't know how much to prompt for further information. I will discuss this in my next supervision meeting to think about what I could learn from this experience.

Appendix Q – Examples of Interview Field Notes

Field notes from immediately after Jason's interview:

I noticed that he spoke a lot about his family of origin, perhaps feels more aligned to them rather than current family.

I was aware of being told how 'busy' he is when booking the interview so noticed that maybe I was explaining things to him in a way that showed I felt nervous about wasting his time e.g. giving lots of information about the study.

It was hard to see his facial expressions because of the way he was sitting, therefore it made it hard for me to read non-verbal cues. I noticed that he seemed to use a lot of humour when talking about his experiences which may have related to the important value he places on lightness.

It appeared important for him to talk about work. I wondered if this was his version of talking about illness. It's the main topic of conversation for others but he doesn't have that. He spoke a lot about his job and his role as provider of the family.

Field notes immediately after Beau's Interview:

I noticed he said "what do you call it" a lot – I wonder if this a sense of uncertainty? Intimidation or is it teenage language?

I was surprised by how he was so keen to tell stories and engage with the research.

His confidence and insightful reflections were unexpected. I am not sure how I thought a 15-year-old would be? Perhaps I expected him to be less interested or less willing to tell stories. He came across as much older than 15/16, I felt saddened by the responsibility he carries to keep motivated and drive and do the same for others