**Pervasive Refusal Syndrome (PRS):**

**Understandings and perspectives of treating professionals**

**working with children and young people**

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**ABSTRACT**

Pervasive refusal syndrome (PRS) is a descriptive label given to a constellation of difficulties with predominating features of refusal and rejection of help across several domains - walking, talking, eating, drinking and self-care. The severity of the presentation is often life threatening. It is a contested diagnosis, with debate regarding both its diagnostic specificity and aetiology.

Despite the fact that management almost always requires extensive input from a multi-disciplinary team (MDT) at a tertiary level of care, there is considerable opacity surrounding what constitutes an effective intervention. This research is the first to qualitatively analyse the understandings and perspectives of professionals, from a range of disciplines, who have experience of working with children and young people given a label of PRS. The overarching research question is: what are the understandings and perspectives of treating professionals working with children and young people who have been given the clinical diagnosis of PRS?

Semi-structured interviews were used with eleven participants who had experience of working with presentations termed PRS in an inpatient psychiatric service. Thematic analysis yielded three core themes: Making Sense, Uncertainty and Our way.

The findings expand on previous literature regarding clinical management with this population. They add new insights to the area, contribute towards practice-based evidence and inform service development. Recommendations are clearly outlined with the aim of increasing consistency, coherence and confidence amongst services working with this population. These include the need to facilitate strong therapeutic relationships with both individuals and their families, enable consistent and clear communication amongst teams and establish novel clinical networks. Theoretical and research recommendations, which widen the lens and draw together different frameworks, are also suggested as a means of developing a more holistic paradigm to understand the distress.

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**1. INTRODUCTION**

**1.1. Overview**

This chapter provides an introduction to the presentation termed pervasive refusal syndrome (PRS). Firstly, the search strategy and terminology used are defined and critiqued. The construct is historicised and an overview of clinical management, including challenges and dilemmas encountered by professionals, is then provided. Particular attention is given to the ways in which PRS has been differentiated from other diagnostic categories and the possible implications of this with regard to understandings and perspectives. Lastly, the rationale and aims of the study are clearly stated.

**1.2. Literature search strategy**

I began exploration of the area with the Lask (2004) paper entitled ‘Pervasive refusal syndrome’ which gave a broad overview of the definition, demography, differential diagnoses, aetiology and management of PRS. I also read ‘Managing Pervasive Refusal Syndrome: Strategies of Hope’ (Nunn, Thompson, Moore, English, Burke & Byrne, 1998). This reading developed an interest in the theoretical rationale informing clinical practice and inspired the topic of this research.

I then began an extensive literature review (Appendix A). I was especially interested in searching for accounts from professionals working with children and young people given a clinical diagnosis of PRS. However, there is only a very small body of literature in this area: 28 journal articles or book chapters specifically mentioning PRS were identified. The overwhelming majority of this literature comprises case studies and/or expert opinion. Additionally, there have been two literature reviews (Jaspers, Hanssen, van der Valk, Hanekom, van Well & Schieveld, 2009; von Folsach & Montgomery, 2006) and two papers that have included follow-up data, one of which used standardised questionnaire measures (Forslund & Johansson, 2013; Guirguis, Reid, Rao, Grahame & Kaplan, 2009).

**1.3. Terminology**

1.3.1. Pervasive refusal syndrome (PRS)

PRS is a descriptive label given to a constellation of difficulties with predominating features of refusal and rejection of help across several domains – including walking, talking, eating, drinking and self-care – in the apparent absence of an organic basis. The often life-threatening severity of the presentation results in significant physical disability and an inpatient admission is usually necessary.

PRS is a contested diagnosis, with debate regarding both its diagnostic specificity and aetiology. It is not formally recognised as a psychiatric diagnosis in any major taxonomy, including the DSM-5 (APA, 2013) or ICD-10 (WHO, 2010). However, the term is used as a clinical diagnosis in services (Graham, 1995), and suggested diagnostic criteria have been described (Appendix B). Nunn, Lask and Owen (2014) recently advocated renaming the presentation ‘pervasive arousal-withdrawal syndrome’ (PAWS). The term PRS will be used for the purposes of this study as it is likely to be most familiar to both professionals and the population themselves. Furthermore, no subsequent literature has used the term PAWS.

Demographically PRS has been more commonly reported in females; the male to female ratio in published literature is around 3:1. It has predominantly been described in children and young people aged between 7 and 15 years old (Jaspers *et al.,* 2009). However, the presentation has also been reported in a 4-year-old boy (Taylor, Dossetor, Kilham & Bernard, 2000), and Riaz and Nawab (2014) recently suggested that the distress experienced by a 50-year-old woman with a diagnosis of a learning disability was best characterised as PRS. Due to difficulties inherent in researching this population, its incidence and prevalence are unknown: around 50 individuals have been described to date. Published literature depicts interventions undertaken in the United Kingdom, Australia, Sweden, Germany, Ireland and The Netherlands (Appendix C).

1.3.2. Terminology critique

The dangers inherent in medicalising the term ‘refusal’ are acknowledged. The refusal of food, drink and self-care can be viewed from multiple perspectives, including as acts of defiance or resistance (demonstrated by the suffragette movement or the ‘dirty protests’ of the Irish Republican Army). Additionally, given the contested nature of PRS, it is also important to highlight the propensity for psychiatric diagnoses to become reified through language (Boyle, 2007). Alternatives to psychiatric terminology are used in line with professional guidance (BPS, 2015). However, the use of diagnostic labels has been unavoidable where discussing previous literature and elucidating the differential diagnoses debate.

**1.4. Historicising PRS**

The construction of PRS will directly impact its clinical management. This section will examine the construction of PRS through a historical lens (Foucault, 1972) by tracing its development in a semi-chronological fashion, structured primarily by the evolution of differing aetiological conceptualisations. It is acknowledged that this is only one means of structuring this narrative.

1.4.1. PRS: a variant of PTSD resulting from undisclosed abuse

PRS first appeared in psychiatric journals in the final decade of the twentieth century. The seminal paper (Lask, Britten, Kroll, Magagna & Tranter, 1991) outlined the histories of four British girls aged between 9 and 14 years old with a *“life threatening condition manifested by dramatic social withdrawal and determined refusal to walk, talk, eat, drink or care for themselves”* (p.866).These girls were admitted to a British inpatient psychiatric service[[1]](#footnote-1) where a discrete syndrome of PRS was ascribed. The admissions were lengthy – between 6 and 18 months – but effective. All were reported to be doing well following discharge.

*1.4.1.2. Concept of refusal*

The authors noted an unequivocally *“determined and wilful refusal”* (Lask *et al.,* 1991, p.868). The volitional nature of the refusal was regarded to be the core feature distinguishing PRS from other presentations.

*1.4.1.3. Aetiology*

Whilst Lask *et al.,* (1991) noted a tendency of these children and their families to avoid conflict – pointing to a potential psychological mediating mechanism – the importance of considering the possibility of non-disclosed abuse was very heavily emphasised. Sexual abuse by a family member was disclosed, or strongly suspected, in three of the girls described in the original paper. The authors hypothesised that the severity of the presentation resulted from (i) the experience of sexual abuse, and (ii) an inability to disclose even the existence, let alone nature, of this abuse due to fear of threatened consequences. Lask *et al.,* (1991) were influenced by literature describing avoidance behaviour as a particular feature of post-traumatic stress disorder (PTSD) resulting from sexual abuse (Kiser, Ackerman, Brown, Edwards, McColgan, Pugh *et al.,* 1988). The description of child sexual abuse accommodation syndrome (CSAAS) (Summit, 1983) also influenced the authors. Thus, PRS was initially conceptualised as a variant of PTSD with avoidance behaviour[[2]](#footnote-2). Subsequent publications added weight to the central role of abuse, particularly sexual abuse, in the aetiology (Lask, 1996). McGowan and Green (1998) also reported the history of an 11-year-old girl who presented with a less severe variant of PRS. Disclosure of sexual abuse, which had been silenced by threats, led to an immediate and complete recovery in this instance.

*1.4.1.4. Implications*

Highlighting the aetiological role of abuse, particularly sexual abuse, had clear consequences. Firstly, families were positioned as a potential threat to their child’s wellbeing, meaning their close involvement in the admission was discouraged. Lask (1996) stated that most of the young people would require alternative care upon discharge. Indeed, three of the four girls described by Lask *et al.,* (1991) were not discharged to the family home, and an 8-year-old British girl described by Graham and Foreman (1995) resided in a weekend fostering placement during admission. Secondly, it raised a managerial dilemma regarding the extent to which the possibility of sexual abuse should be investigated before it became abusive in itself (Graham & Foreman, 1995). Highlighting abuse as central to the aetiology also impacted on the way in which others constructed PRS. For example, Nourse, Ryan and McMenamin (1999) reported three instances where PRS was considered as a differential diagnosis, but excluded on the basis that sexual abuse did not appear to be a precipitating factor.

*1.4.1.5. Social, cultural and political context*

It is important to situate the ‘discovery’ of PRS in the early 1990s in context. Firstly, this was a time of increased public consciousness about the potential impact of child sexual abuse (NSPCC Centenary Appeal, 1984; ChildLine launch, 1986). Following the judicial review into the Cleveland Scandal[[3]](#footnote-3) (Butler-Sloss, 1987) pressure was increased on healthcare professionals not to miss signs of abuse, and the Department of Health and Social Security (1988) recommended that odd or unexplained behaviours in a child should arouse the suspicion of sexual abuse. Further, the experience of sexual abuse in childhood was frequently implicated in the aetiology of mental health difficulties (Mullen, Romans-Clarkson, Walton & Herbison, 1998). Brown and Perkins (1992) submitted that sexual abuse was central to the aetiology of three girls aged between 5 and 6 years old presenting with unexplained deterioration in skills, language and social withdrawal; difficulties improved dramatically following the disclosure of sexual abuse. Lask (1996) commented that the greater cognitive maturity of those given the clinical diagnosis of PRS might increase awareness of the potential consequences of disclosure.

A second context is the monolithic rise of diagnostic manuals in the latter half of the 20th century, for cultural, theological, political and professional reasons (Scull, 1979). PRS was introduced as a descriptive term for difficulties that did not ‘fit’ predefined categories (Lask *et al.,* 1991). Graham (1995) states that the term PRS is “*accurately descriptive* (with) *no unjustifiable…assumptions”* (p.651). Two of the most prominent psychiatrists involved in the PRS endeavour (Dr Bryan Lask and Dr Ken Nunn) advocated adopting a multi-axial diagnostic system with a prose style as an alternative to psychiatric classification (Nicholls, Chater & Lask, 2000; Nunn, Nicholls & Lask, 2000).

1.4.2. PRS: learned helplessness resulting from a traumatic experience

Thompson and Nunn (1997) depicted PRS in an Australian context by means of a case note review. This paper described seven children admitted under the auspices of PRS in an inpatient psychiatric unit[[4]](#footnote-4). The review, which detailed the histories of six girls and one boy aged between 7 and 15 years old, used existing features of the presentations to form the basis of inclusion for the review. These were as follows: (i) clear food refusal and weight loss[[5]](#footnote-5); (ii) social withdrawal and school refusal; (iii) partial or complete refusal in two or more of the following domains: mobilisation, speaking, attention to self-care; (iv) angry and active resistance to being helped or encouraged to begin engaging in these activities; (v) no organic condition that accounts for the severity or degree of symptoms and (vi) no other psychiatric illness that accounts for the severity or degree of symptoms.

*1.4.2.1. Concept of refusal*

Again, the distinguishing feature of PRS was taken to be the volitional and purposeful nature of the refusal: *“active, angry conscious refusal”* (Thompson & Nunn, 1997, p.146). The authors clearly advocated for PRS to be regarded as a discrete syndrome, rather than a chance convergence of various co-occurring presentations.

*1.4.2.2. Aetiology*

Thompson and Nunn (1997) foregrounded the experience of traumatic life experiences in the development of PRS. However, in contrast to the British literature, abuse was positioned as one of several types of traumatic life events (including illness, migration, parental conflict) with the potential to precipitate the onset of the difficulties. Nunn and Thompson (1996) implicated loss of personal and vicarious hopefulness in the aetiology of PRS. The authors invoked the attributional reformulation of the cognitive theory of learned helplessness to theorise the development of difficulties (Abraham, Seligman & Teasdale, 1978) whereby traumatic events are hypothesised to be experienced as uncontrollable. Experiences of uncontrollability are then generalised to thoughts regarding the future, provoking feelings of helplessness[[6]](#footnote-6) and hopelessness. Importantly, Nunn and Thompson (1996) highlighted the vicarious and circular nature of these feelings: each family member becomes consumed with the threatened future of the other (Brown & Inouye, 1978; DeVellis, DeVellis & McCauley*,* 1978). However, the reporting of PRS in a 4-year-old Australian boy called this aetiological explanation into question (Taylor *et al.,* 2000), as it is unlikely that such a young child has the developmental capacity to sustain such a cognitive generalisation.

*1.4.2.3. Implications*

Firstly, the application of the learned helplessness paradigm directly informed guidance for a specific and distinct management approach based on the application of hope-promoting principles (Nunn *et al.,* 1998). Secondly, the distancing of abuse as central to the aetiology potentially facilitates a shift whereby families can be viewed as a resource, rather than a potential threat. This is evident in the fact that all of the children described by Thompson & Nunn (1997) were discharged to their family home. However, Nunn *et al.,* (1998) cautioned that families should assume a ‘strictly social role’ and abandon their care providing responsibilities. Finally, this aetiological explanation offers an understanding of the way in which both family members and professionals may vicariously perpetuate distress.

1.4.3. PRS: depressive devitalisation

In the early years of the 21st century, around 2% of children from families seeking asylum in Sweden were reported to have developed a pervasive loss of bodily functions with no organic basis[[7]](#footnote-7). Bodegård (2005a) coined the term ‘depressive devitalisation syndrome’ (DD) to describe this presentation, which at its most severe was perceived to be synonymous with PRS. The sudden rise of a previously undescribed syndrome prompted considerable media attention[[8]](#footnote-8), extensive political debate and significant allocation of psychiatric resources. A commission mandated by the Swedish Ministry of Migration and Asylum Policy reported no new cases of DD following a refugee amnesty in 2006 (Swedish Government Report, 2006). However, Forslund and Johansson (2013) reported that the number of children and young people admitted to Swedish psychiatric services with a diagnosis of DD had continued to increase.

*1.4.3.1. Nature of refusal*

The nature of the behaviour was the source of passionate debate. The widely reported fact that some presentations appeared to improve contingent to the granting of permanent residency permits led one national newspaper to accuse parents of malingering in order to manipulate the Swedish state[[9]](#footnote-9). However, Bodegård (2005a; 2005b; 2006) suggested that the refusal was of a passive nature, and that it was only intermittently punctuated by a ‘panicky’ active refusal.

*1.4.3.2. Aetiology*

Bodegård (2005b) constructed DD as a family psychiatric syndrome underpinned by ‘lethal mothering’ – a psychodynamic concept in which the mother fantasises that the child is dying, and the child conforms by taking up a position of the devitalized infant. The role of previous environmental trauma and learned helplessness was acknowledged; however, a specific trauma model of symptom development was not implicated. A subsequent literature review, which outlined similarities and differences between PRS and DD, suggested common aetiological factors included a history of both child and parental psychiatric problems and precipitating stressful events (von Folsach & Montgomery, 2006).

*1.4.3.3. Implications*

Firstly, constructing the presentation as a family psychiatric disorder has management implications. Several parents of the children and young people described by Bodegård (2005b) were themselves admitted to an adult psychiatric service. Secondly, highlighting the passive nature of DD opened up debate regarding the nature of refusal. Von Folsach and Montgomery (2006) asserted that the ‘passive’ refusal characteristic of DD and the ‘active’ refusal described in PRS could be seen as subtypes of an overarching refusal presentation. In practice, however, DD appears to have been assimilated into the concept of PRS (Jaspers *et al.,* 2009), and no cases of DD have been reported outside of Sweden. Lastly, the strong assertion that severe cases of DD should be viewed through a PRS lens (Bodegård, 2005; von Folsach & Montgomery, 2006) perhaps meant that whilst different aetiological explanations for the presence of such distress in the asylum seeking population were proposed[[10]](#footnote-10) these had limited impact.

1.4.4. PRS: varying forms of expression

Broadening the notion of ‘refusal’ had a significant impact on subsequent literature. In a literature review of 24 published case studies Jaspers *et al.,* (2009) hypothesised that an individual may fluctuate between a refusal-withdrawal-regression spectrum, dependent on their state of mind and context. Importantly, however, the notion of a separate entity of PRS, distinguished by active refusal, is maintained: *“active resistance to any form of help is the distinguishing feature of PRS”* (p.649). Indeed, the authors advocate for the inclusion of PRS in the DSM.

Subsequent literature questioned whether active volition should continue to be regarded as the hallmark of PRS. Conceptualising refusal as a continuum ranging from active refusal to passive resistance was suggested, given the apparent fluctuation within the presentations of three German young people between the ages of 14 and 16 (Jans, Ball, Preiss, Haberbausen, Warnke & Renner, 2011). Indeed, Jans *et al.,* (2011) suggested modifying diagnostic criteria for PRS to reflect the fact that active and angry features are present only some of the time. Forslund and Johansson (2013) also reported a continuum of refusal in five children and young people of Azerbaijani origin aged between 7 and 17 and seeking asylum in Sweden. Both groups of authors agreed with Jaspers *et al.,* (2009) that PRS could be differentiated from other diagnoses.

*1.4.4.1. Implications*

Broadening the notion of refusal directly impacted subsequent literature, the majority of which problematises the centrality of active conscious refusal in the conceptualisation of the presentation (Bodegård, 2014; Lee, Duff, Martin & Barrett, 2013; Nunn *et al.,* 2014; Wright & Beverley, 2013).

1.4.5. PRS: a deadlock in the sympathetic and parasympathetic nervous systems

Most recently, Nunn *et al.,* (2014) suggested a reformulation of the presentation on the basis of clinical experience. The authors posited that the apparent refusal could better be understood as a combination of extreme anxiety, avoidance and behavioural paralysis. A predominantly biological aetiology was hypothesised: in the event of trauma, the severity of the stressor combined with individual ‘sensitivities’ cause a simultaneous hyper-arousal of both the sympathetic nervous system (resulting in anxiety) and parasympathetic nervous system (resulting in withdrawal). This results in a behavioural deadlock. It was postulated that this model accounted for behavioural variability, such as both withdrawal and active resistance. The authors advocated that the term PRS be renamed ‘pervasive arousal-withdrawal syndrome’ (PAWS).

*1.4.5.1. Conceptualisation of refusal*

From a PAWS perspective, implicating volition and intent in the difficulties is deemed inappropriate. This is in stark contrast to earlier literature. It could be argued that by implicating social (a stressor), psychological (individual ‘sensitivity’) and biological (hyper-arousal of nervous systems) factors the PAWS model recognises the emerging, but increasingly dominant, discourse acknowledging the inextricable interplay between mind and body.

*1.4.5.2. Implications*

The extent to which the PAWS model will be embraced is unknown. Bodegård (2014) asserts that the concept has been endorsed by the Swedish Board of Health and Welfare and is to be included in the Swedish ICD-10-SE[[11]](#footnote-11). However, PRS remains a term used in services and literature (Riaz & Nawab, 2014). The PAWS model has clear management implications. Nunn *et al.,* (2014) emphasise the need to greatly reduce pressure on the child or young person to recover as a means of decreasing sympathetic arousal. The potential role of psychotropic and anticholinergic medication to target simultaneous hyper-arousal of the nervous systems is also suggested.

1.4.6. Summary

This section has demonstrated the ways in which, despite its relatively short history as a concept, understandings and perspectives of the difficulties termed PRS have shifted dramatically. Authors have begun to question the validity of underscoring active refusal as the central distinguishing feature of PRS. The transformation of PRS from a purely descriptive term, to a term perceived to embody a discrete entity – with advocates for its inclusion in the DSM-5 and ICD-10-SE, and authors stressing the need for a distinct and specific intervention approach – highlights the way in which psychiatric diagnoses can become reified over time.

**1.5. Clinical management**

An admission for a child or young person given the clinical diagnosis of PRS is costly, both emotionally and financially (Nunn *et al.,* 1998). As shown above, the way in which the presentation is conceptualised has a direct impact on clinical management. However, clinical management is a neglected aspect of the literature. There is a very minimal evidence-base to be drawn upon, comprising case studies and expert opinion[[12]](#footnote-12) and no formalised guidelines.

1.5.1. Setting

The majority of published case studies concern children and young people admitted to an inpatient psychiatric setting (Appendix C). However, successful outcomes have also been reported following intervention in an inpatient paediatric admission (Graham & Foreman, 1995; Edwards & Done, 2004; Lee *et al.,* 2013; McNicholas *et al.,* 2013; Taylor *et al.,* 2000; van der Walt & Baron, 2006). Only two papers (Thompson & Nunn, 1997; Wright & Beverley, 2012) describe management of PRS on a outpatient basis. As such, it is important to acknowledge the bias inherent in the literature towards those who have had lengthy inpatient admissions. It is possible that those who do not receive an inpatient admission may differ in some important respects from those who do.

1.5.2. Prognosis

Jaspers *et al.,* (2009) reported that 67% of those given the clinical diagnosis of PRS made a full recovery and a further 25% made a partial recovery[[13]](#footnote-13). Two groups have collected follow-up data. Guirguis *et al.,* (2009) administered a semi-structured questionnaire focussing on current functioning at 3 to 16 years post-discharge. Of the three British young people who consented to the telephone interview two were reported to have made a complete recovery according to predefined criteria. The other had a diagnosis of anorexia nervosa. Forslund and Johansson (2013) also reported a good prognosis for all five children and adolescents of Azerbaijani origin, aged between 7 and 17 years old, treated in a Swedish inpatient psychiatric service. Data concerning current ability, clinician assessment of functioning and two self-administered questionnaires[[14]](#footnote-14) was collected between 1 and 8 years post-discharge.

Several authors have drawn attention to the fact that length of illness preceding admission appears to strongly correlate with length of admission (Lask, 2004; Nunn *et al.,* 1998). This highlights the need for early identification and intervention.

1.5.3. Overarching management principles

Two papers, both of which comprise expert opinion, have exerted particular influence on the management of PRS in psychiatric inpatient settings (Nunn *et al.,* 1998; Lask, 2004). Both authors advocate the use of milieu therapy (Crouch, 1998) and a structured, yet flexible, multi-disciplinary team (MDT) approach.

Nunn *et al.,* (1998) state that the specific needs of the PRS population necessitate a distinct management approach: one that takes into account the life threatening nature of the presentation, determined refusal and compromised nature of consent. The paper explicitly suggests hope-promoting principles as the basis of the approach, in line with the aetiological hypothesis of learned helplessness (Nunn & Thompson, 1996). For example, the principal goal of the admission is stated as the *“recovery of the patient to become an adequately functioning, developmentally appropriate, hopeful person”* (p.235). Further, the intervention should be rooted in the persistent, non-punitive and realistic promotion of personal hopefulness. The paper suggests approaches relevant to different stages of the admission. For example, at pre-admission and in the early stages of the intervention important principles include: negotiating the management contract, communicating staff expectations and predicting the difficulties of being helped. Nunn *et al.,* (1998) also highlight the need for children and young people to control the pace of recovery throughout the admission. Importantly, the authors suggest that visiting hours are restricted in order to reduce the child’s contact with the ‘enmeshed’ family network, help the young person to focus on ward programme and minimise discord between family and the team.

Lask (2004) offers a somewhat briefer outline of key management principles as follows: the use of the ward milieu; patience, time and sympathy; basic nursing care offered by skilled staff; physiotherapy; individual therapy; and parental counselling or family therapy. Rehabilitation attempts are suggested to be better placed once the admission moves into a recovery phase, as initial attempts commonly result in setbacks.

1.5.4. Profession specific involvement

A limited amount of literature has elucidated the specific roles of professionals working with the presentation.

*1.5.4.1. Nursing*

The literature stresses that the level of nursing provision required for this population is extremely high (Lask *et al.,* 1991). Nunn *et al.,* (1998) suggest that more than one child or young person with a clinical diagnosis of PRS should not be admitted concurrently due to the demands on staff resources, both physically and emotionally. The most fundamental of nursing requirements is ensuring survival – nasogastric tube feeding is usually required – and fostering activity, such as eating and self-care (Nunn *et al.,* 1998). Jaspers *et al.,* (2009) highlight the need for tender loving care, patience and sensitivity in working with this population. The use of the therapeutic ward milieu has been described as a means of providing a safe, consistent and secure therapeutic environment. Guirney (2012) states that, aside from those that are non-negotiable, expectations should be minimised and communicated clearly and simply. Lee *et al.,* (2013) highlight the benefits of using a communication file in terms of increasing communication and consistency between nursing staff. The need for consistency has been described as necessary in order to reduce interpretation and possible manipulation, by both parents and their children (Edwards & Done, 2004; Nunn *et al.,* 1998).

*1.5.4.2. Physiotherapy*

The essential role of physiotherapy has been emphasised (Lask, 2004; Nunn *et al.,* 1998). Nunn *et al.,* (1998) describe three phases involved in intensive physiotherapy, progressing from passive involvement, through to minimal active involvement and finally to active mobilisation, such as walking. Magagna and Bladen (2012), a psychotherapist and physiotherapist respectively, describe their work with a 14-year-old girl in a British psychiatric inpatient context. They portray an approach underpinned by principles of collaboration, curiosity, containment, confidence, creative imagination and surprise. The authors also describe some of the challenges a physiotherapist working with this population may face, such as being perceived as a threat to the child or young person’s ‘need’ to be unwell and feeling isolated from the wider MDT.

*1.5.4.3. Individual therapy*

Nunn *et al.,* (1998) suggest that individual work should aim to strengthen inner perceptions and promote personal coping strategies. If a child is not able to communicate verbally then therapy sessions should comprise a monologue speculating on the child’s emotions and communicating that they are valued. If the child expresses a wish to end the session prematurely this wish should be respected. Magagna (2004; 2012) reflects on her experiences of offering intensive individual psychotherapy to this population, based on a psychodynamic hypothesis that these children and young people lack a strong, benevolent ‘internal parent’ to support them and the psychic structure of their family members is too weak to contain their immense feelings. Play, dream interpretation and drawing are advocated as part of this work. However, Lask (1996) and Thompson and Nunn (1997) suggest that individual therapy is best offered in a cognitive, rather than psychodynamic, framework. A role for individual therapists to facilitate reflection within the MDT (Nunn *et al.,* 1998) and provide individual psychology sessions to parents, both during and post-admission has also been suggested (Lee *et al.,* 2013).

Play and drama therapy (Lee *et al.,* 2013; Taylor *et al.,* 2000) and music therapy (Van der Walt & Baron, 2006) have also been incorporated into an intervention for PRS. Lee *et al.,* (2013) suggest that these professionals may be seen as slightly separate to the rest of the team by the child or young person, and as such they might be somebody with whom they first ‘test out’ reactions to increases in improvement.

*1.5.4.4. Family therapy*

Less has been written specifically detailing the role of family therapy. However, Nunn *et al.,* (1998) suggest that the work should focus on improving family dynamics and extricating enmeshed relationships. Additionally, family therapists may support the family through the process of transferring their care providing role to the team and adjusting to a strictly social relationship with their child (Nunn *et al.,* 1998).

1.5.5. Challenges and dilemmas

Several challenges and dilemmas involved in working with this population from a professional perspective have been reported in the literature.

*1.5.5.1. Child and young person behaviours*

Nunn *et al.,* (1998) list behaviours that are often experienced as challenging by professionals, including targeted mutism and aggressive outbursts. Lee *et al.,* (2013) also highlight the difficulties managing self-injurious behaviour. A pattern of regression and deterioration in the face of praise and the need for thoughtful communication which does not involve open praise or direct requests has also been documented (Edwards & Done, 2004; Anonymous, 2001; Lee *et al.,* 2013). Nunn *et al.,* (1998) suggested that praise should be specific and designed not to fuel the perception of increased expectation.

*1.5.5.2. Family behaviours*

Nunn *et al.,* (1998) described some of the challenges of involving parents in the admission from a professional perspective including: failure to adhere to restrictions, denial and minimisation of difficulties within the family and reluctance to accept a psychiatric diagnosis and care. Parents may also harbour feelings of resentment towards nursing staff for taking away their ability to look after their child, and/or experience them as punitive and inflexible, resulting in power battles. Families may also decline family work offered in paediatric units (Edwards & Done, 2004).

*1.5.5.3. Challenges for the team*

Several challenges for the team have been elucidated, including dilemmas encountered when working with compromised consent (Graham & Foreman, 1995), refraining from positive reinforcement (Lee *et al.,* 2013) and maintaining team cohesion (Nunn *et al.,* 1998). A small amount of literature has made reference to the emotional impact of working with these children. Rejection, frustration and feeling redundant and hopeless are common features in descriptive accounts (Edwards & Done, 2004; Magagna, 2012; Guirney, 2012; Bladen & Magagna, 2012). Such emotions have been described as decreasing empathy and cultivating a desire to ‘retaliate’ (Guirney, 2012). Nunn *et al.,* (1998) highlight the importance of self-reflection in order to ensure treatment is not punitive, particularly given the vicarious nature of learned helplessness. Menzies-Lyth (1960) highlights how professionals can develop social defences, such as depersonalisation, detachment and resistance to change, when faced with overwhelming anxiety, sadness and discomfort. Working with children and young people who ostensibly reject help – despite a significantly high level of need – also challenges engrained institutional, and likely personal, assumptions about helping.

**1.6. Differentiating PRS**

The volitional and pervasive nature of the refusal behaviour has been taken to be the principal distinguishing feature of a separate and discrete entity of PRS (Lask *et al.,* 1991; 2004; Jaspers *et al.,* 2009; Thompson & Nunn, 1997). However, many authors have detailed significant similarities to other presentations of both an organic and psychiatric nature (Lask, 2004; Lee *et al.,* 2013; McNicholas *et al.,* 2013; Nunn *et al.,* 2014; Thompson & Nunn, 1997). Jaspers *et al*., (2009) provide an overview of the main differential diagnoses considered in the process of identifying PRS in their comprehensive literature review. These diagnoses, alongside reasons for exclusion offered by Nunn *et al.,* (2014), can be found in Table 1[[15]](#footnote-15).

1.6.1. Organic differential diagnoses

It is acknowledged that the clinical diagnosis of PRS will necessarily follow the exclusion of an underlying organic cause (Jaspers *et al.,* 2009; Lee *et al.,* 2013). Amongst medical conditions investigated are leukodystrophy, frontal lobe disorder and various types of encephalitis. An inaccurate medical diagnosis can have significant implications for management. For example, Wright and Beverley (2012) highlighted how an incorrect diagnosis of encephalitis lethargica led to a limited and hands-off approach, in direct contrast to the more active management needed to promote gradual recovery in PRS.

1.6.2. Functional differential diagnoses

Many of those who are given a clinical diagnosis of PRS are initially given a different psychiatric diagnosis (Jaspers *et al.,* 2009; Wright & Beverley, 2012). McNicholas *et al.,* (2013) also highlight a case whereby a diagnosis of PRS was superseded by a diagnosis of catatonia *after* an intervention for PRS was not seemingly effective. This has important implications, as different diagnoses may lead to substantially different approaches to intervention.

*Table 1: Differential diagnoses*[[16]](#footnote-16)

|  |  |
| --- | --- |
| Presentation | Reasons for exclusion |
| Depression (54%) | Range and intensity of PRS symptoms exclude diagnosis.  PRS can present without any definite sign of low mood, psychomotor slowing or agitation and there is usually no diurnal variation.  Response to treatment for PRS can occur with a volatility of improvement and deterioration rarely seen in depression. |
| Somatoform disorder (42%) | Range and intensity of PRS symptoms excludes diagnosis.  Refusal in PRS is pervasive to activities in which there is no lack of function unlike that seen in conversion disorder.  Somatoform disorders are not characterised by help rejection. |
| Anxiety (29%) | Range and intensity of PRS symptoms excludes diagnosis.  Help-rejection not usually seen in anxiety. |
| Eating disorder (17%) | Range and intensity of PRS symptoms exclude diagnosis.  Absence of compensatory behaviours and assumed absence of cognitions in PRS |
| Selective mutism (4%) | Range and intensity of PRS symptoms exclude diagnosis. |
| Catatonia (4%) | Absence of unresponsiveness, stereotypies and waxy flexibility in PRS.  Capacity to engage remains intact in PRS in contrast to the automaticity of negativism. |
| Chronic fatigue syndrome (4%)[[17]](#footnote-17) | Fatigue is not central or most prominent in PRS presentation  Those with chronic fatigue syndrome often express frustration with their health condition and a desire to recover. |
| Factitious disorder (4%) | In PRS help rejection, not care eliciting, is prominent. |

*1.6.2.1. Depression, anxiety and selective mutism*

It is acknowledged that there is a considerable comorbidity of presentations termed depression and anxiety with PRS (Lask, 2004). There is an established evidence-base underpinning the management of both these presentations in children and young people. Short-term individual psychological therapy, including cognitive behaviour therapy (CBT), is recommended as a first line treatment for the management of both depression (DeFilippis & Wagner, 2014; National Institute for Health and Care Excellence: NICE, 2009;) and anxiety (Cresswell, Waite & Cooper, 2014). NICE guidelines (2009) also suggest combining individual therapy and fluoxetine to manage depression in young people if necessary. CBT and/or behaviour therapy have also been advocated for children presenting with selective mutism (Cohan, Chavira & Stein, 2006).

*1.6.2.2. Somatoform disorder and factitious disorder*

Thompson and Nunn (1997) report the presence of conversion disorder (in this instance gait disturbance) as a comorbid presentation in three children treated for PRS. CBT, behaviour therapy and self-monitoring techniques are recommended aspects of management for all types of somatoform disorders in children and young people (Silber, 2011). The difference between somatoform and factitious disorder is broadly taken to be one of volition. Jaspers *et al.,* (2009) discuss how factitious disorder was considered a possible differential diagnosis in their work with an 11-year-old Dutch girl given a clinical diagnosis of PRS whose behaviour was described as giving a *“very forced and theatrical impression”* (p.646). Bass and Glaser (2014) assert that in cases where fabrication and exaggeration are suspected, factitious symptoms are likely to be mixed with psychosomatic ones.

*1.6.2.3. Eating disorder*

There are significant similarities between difficulties described as PRS and those described as an eating disorder, to the extent that there are instances of PRS occasionally being described as a subtype of an eating disorder (Lask, 2000). Family-based treatment has been advocated for the management of young people given a diagnosis of either anorexia nervosa (Eisler, 2005) or bulimia nervosa (Le Grange, Crosby, Rathouz & Leventhal, 2007). In a family therapy model, parents are directly involved in the intervention as a means of empowerment and increasing self-efficacy. Multi-family approaches have also been found to be effective (Eisler, 2005). CBT is also recommended for those given a diagnosis of bulimia nervosa (Schmidt, Lee, Beecham, Perkins, Treasure & Eisler, 2007).

*1.6.2.4. Chronic fatigue syndrome (CFS)*

Several children and young people described by Thompson and Nunn (1997) were initially given the diagnosis of CFS. Wright and Beverley (2012) also describe how PRS became the working diagnosis when the presentation of a 14-year-old boy with a diagnosis of CFS deteriorated. Whilst there continues to be much conflicting advice and differences of opinion regarding the management of CFS (Smith & Wessley, 2014), both NICE guidelines (2006) and a systematic review of management in children and young people (Knight, Scheinberg & Harvey, 2013) advocated CBT and graded exercise therapy (GET) as best practice. Garralda and Chalder (2005) highlighted the importance of family engagement in working with young people given a label of CFS.

*1.6.2.5. Catatonia*

Catatonia is acknowledged to be an extremely variable presentation with no standardised diagnostic criteria for children and young people (Takoaka & Takata, 2000). Established treatment for catatonia in adults is medical: benzodiazepines followed by electroconvulsive therapy (ECT) (Taylor & Fink, 2003). A systematic review suggested that a similar protocol is used in the management of catatonia in children and young people (Lahutte, Cornic, Bonnot, Consoli, An-Gourfinkel, Amoura *et al.,* 2008). Fink and Carlson (1995) suggested that Graham and Foreman (1995) inaccurately labelled catatonia as PRS in an 8-year-old girl. The authors cited the effective use of anti-depressants and ECT in another 8-year-old girl with a similar presentation (Cizadlo & Wheaton, 1995), and suggest that this approach would have been more appropriate in the case described by Graham and Foreman (1995). Catatonia was also a diagnosis considered in the treatment of an 11-year-old Irish girl following a lack of response to management for PRS in a paediatric inpatient setting (McNicholas *et al.,* 2013). Consultation with a prominent author in the field of catatonia led to a significant change in management: a benzodiazepine was prescribed and ECT was advocated[[18]](#footnote-18). Abreaction and narcosis[[19]](#footnote-19), which have been recommended as an adjunct to the management of severe somatising disorders, were also used. Their use did not lead to any discernable improvement.

**1.7. Consequences of viewing PRS as a discrete entity**

Previous sections have highlighted the way in which PRS has largely been constructed as an uncommon discrete entity, with volitional refusal as its fundamental differentiating factor [[20]](#footnote-20). This has significant implications for the way in which PRS is perceived, experienced and understood.

1.7.1. Child and young person

There is currently no published literature written from the perspective of an individual who has been given the label of PRS. It is suggested that they may experience feelings of confusion, anxiety and isolation, which may be potentially compounded by the inherent implication that their distress is best described by foregrounding an apparently volitional refusal. This may not fit with their lived experience. Alternatively, there may also be a feeling of specialness or uniqueness given the apparently low incidence of such a presentation.

Research in analogous areas has found that the term given to a difficulty can influence outcomes. For example, adults given the diagnosis of myalgic encephalitis (ME) have been found to have a significantly poorer prognosis than those diagnosed with CFS, despite the two being clinically synonymous (Hamilton, Gallagher, Thomas & White, 2005). The authors hypothesised that the biological connotations associated with ME potentially obscured the use of psychosocial strategies found to be beneficial in managing the presentation. A similar process of nominative determinism may arise in PRS.

1.7.2. Family

The way in which PRS is constructed is likely to have a significant impact on the engagement of the family with the admission. Some parents may fundamentally disagree with the proposed psychiatric explanation of the diagnosis and the implicit implication that their child is actively refusing. This may cause friction in the relationship between parents and the team (Nunn *et al.,* 1998). This conceptualisation may also increase concern amongst parents regarding the way in which their child is perceived and understood by professionals. For example, Chris, the father of an 8-year-old British girl given a label of PRS, explained how he felt compelled to reiterate to staff how different his daughter’s behaviour was compared to her ‘normal’ behaviour (Lee *et al.,* 2013). Additionally, Chris mentioned his concern that staff may have suspected sexual abuse on the basis of the Lask *et al.,* (1991) aetiological formulation. Conceptualising the presentation as extremely uncommon can also be seen to reduce parental confidence and increase deference to ‘experts’ (Anonymous, 2001; Sakeld, 2013).

The fact that the presentation is widely seen as uncommon has implications for resources. Newspaper reports suggest that the parents of an 11-year-old British girl given a diagnosis of PRS had to fight against the system to get the physical and emotional needs of their child met within a general paediatric unit given the lack of available psychiatric beds (Allen & Allen, 2002). The scarcity of specialist resources in some localities has been highlighted in both published literature (McNicholas *et al.,* 2013) and the media (Allen & Allen, 2002) and has led to fundraising efforts in order to meet costs for private consultations (Sakeld, 2013).

PRS has been depicted in the media as extremely rare. For example, a British tabloid newspaper recently erroneously reported a 10-year-old girl to be only the third British child to develop PRS. She was described as a *“screaming caged animal”* and *“possessed”* This has implications for the way in which PRS is understood in an everyday context and is likely to increase stigma (Allen & Allen, 2002; Wright & Beverley, 2012). This may be perpetuated by the fact that online newspaper articles are likely to be much more accessible to many families and concerned others searching for information than academic journals.

1.7.3. Professionals

Firstly, the long tradition of papers distinguishing ‘wilful’ refusal as the hallmark of PRS suggests professionals may perceive those given a diagnosis of PRS to have a degree of volition regarding their behaviours. This is difficult to substantiate in the absence of literature regarding professional perceptions. However, individual agency appears to have been attributed by McNicholas *et al.,* (2013), who described how an 11-year-old Irish girl seemed to almost *choose* to recover in order to avoid transfer to a British psychiatric service. The perception of agency is important given previously outlined challenges of working with this presentation: it may decrease empathy (Guirney, 2012) and possibly make ‘punitive’ treatment more likely (Nunn *et al.,* 1998).

Secondly, positioning PRS as an uncommon and discrete entity may potentially disempower and disconnect professionals from their experience and erode confidence ultimately leading to a paralysing affect on the system. A lack of experience and confidence in working with this presentation has led to professionals consulting external ‘experts’ which may further diminish individual and team perceptions of their own self-efficacy and competence (McNicholas *et al.,* 2013). This may be amplified in a context of different terminologies and conceptual understandings.

Thirdly, the perception of PRS as an uncommon and discrete entity may heighten anxiety and concern within the system. Professionals may be concerned that either an organic diagnosis may be missed or that they may be blamed for not identifying a ‘correct’ psychiatric diagnosis. Instances of both can be found within academic literature (Forslund & Johannson, 2013; Wright & Beverley, 2012) and newspaper reports (Carvel, 2009; Robothon, 2004). Anxiety may lead to what has been termed ‘abnormal treatment behaviour’ (Singh, Nunn, Martin & Yates, 1981). When applied to PRS such behaviours may include: repeated damaging and extensive medical investigations in an attempt to find an organic cause for the difficulties; refusal to acknowledge *both* physical and psychological factors as relevant; and giving potentially iatrogenic advice, such as bed rest (Thompson & Nunn, 1997). This is important given the anecdotal suggestion that such behaviours are contraindicated: Thompson and Nunn (1997) suggest a negative correlation between medical over investigation and chronicity of the presentation.

1.7.4. Services

Publically funded services, such as the National Health Service, need to be accountable to a wide variety of stakeholders for their activities. As such, it is important to be able to clearly communicate a theoretically justified rationale for choice of intervention approach. This need is heightened in the current climate of austerity in the NHS (The NHS Five Year Forward View, 2014). Specific admission costs for those with a diagnosis of PRS are not available. However, a rough calculation based on available data highlights the demand on hospital resources: a psychiatric admission may cost in the region of £250,000[[21]](#footnote-21). The discourse of evidence-based practice has come to be seen as synonymous with quality healthcare and best practice (e.g. NICE; Cochrane Collaboration). Wider policy initiatives also explicitly stress the importance of rooting professional practice in the evidence base (Harper, Gannon & Robinson, 2013). The fact that there are no evidence-based guidelines relating to best practice in PRS may therefore be seen as a cause for concern for some, not least clinical commissioners. A complementary, if less established, paradigm of practice-based evidence provides a means for practitioners to generate an evidence base grounded in routine clinical practice (Margison, Barkham, Evans, McGrath, Mellor-Clark, Audin *et al.,* 2000; Mellor-Clark, & Barkham, 2003).

**1.8. Rationale for study**

Despite the fact that a clinical diagnosis of PRS usually necessitates a lengthy and costly admission the delineation of effective management has been neglected. There is a clear rationale for focussing on treating professionals’ understandings and perspectives, particularly with regards to perspectives of clinical management and challenges and dilemmas encountered:

1. A lack of literature focussing on the clinical management of PRS may be seen as concerning given the potentially life threatening nature of the presentation, the financial and emotional cost, the complexity of challenges which may be faced by the staff team and the partial or incomplete recovery reported in 33% of the population (Jaspers *et al.,* 2009).
2. Professionals are increasingly required to provide a theoretically reasoned justification for choosing one treatment approach over another. Practice-based evidence provides a means of contributing to this.
3. The two main papers in this area (Lask, 2004; Nunn *et al.,* 1998) predate significant changes in the aetiological conceptualisation of the presentation. These changes have been shown to have implications for clinical management.
4. No qualitative research has focussed on the experiences of professionals working with this population.

**1.9. Aim and research question**

The guiding aim of this study is to qualitatively analyse the understandings and perspectives of treating professionals, from a range of disciplines, who have experience of working with children and young people given a label of PRS. The study offers an opportunity to explore the ways in which professionals understand PRS and the rationale underpinning the way in which it is managed. It also provides an opportunity to expand on the limited amount of previous literature depicting challenges and dilemmas faced by professionals working in this area.

It is hoped that the findings of this study will have several practical applications: firstly, to contribute towards practice-based evidence ultimately benefiting individuals and their families who receive a clinical diagnosis of PRS; secondly, to inform service development and approaches to staff support; and, thirdly, to provide the basis for further clinically relevant research, which will facilitate a greater understanding of effective management.

With this in mind, the central research question is:

**What are the understandings and perspectives of treating professionals working with children and young people who have been given the clinical diagnosis of PRS?**

Specific attention will be given to:

1. perspectives on clinical management;
2. challenges and dilemmas

**2. METHOD**

**2.1. Overview**

This chapter outlines the epistemological position taken in this study and the theoretical rationale that guided choices in methodology. Recruitment procedure, data collection and data analysis are discussed alongside the considerations taken to ensure that the research upheld ethical principles. A statement of researcher reflexivity is provided.

**2.2. Epistemological position**

Approaches to research can be differentiated by their underlying epistemological position: the types of claims that can be made about the nature and status of knowledge. Clarity regarding the position taken by the researcher is essential, as such positions determine assumptions held about the relationship between the collected data and the world (Harper, 2011). Researchers can take a number of different positions regarding the extent to which data are seen as reflecting reality. At extremes of the spectrum are the naïve realist and radical social constructionist positions. In a naïve realist position, the researcher assumes a relatively uncomplicated and direct relationship between data and reality. In contrast, a radical social constructionist position is primarily concerned with the social construction of knowledge through discourse and context and the consequences of these constructions for those who are positioned by them. Willig (2012) has described the difference between the two as being the difference between the researcher positioned as a ‘detective’ or ‘architect’.

A critical realist epistemological position was taken for this research. This position shares with social constructionism the requirement that concepts be examined in the context of the social and historical conditions which allowed them to emerge. However, in a critical realist account it is not reality itself that is seen as socially constructed, but rather our theories of reality and the methods we use to investigate them (Pilgrim & Bentall, 1999). As such, the analysis will be guided by the underlying assumption that although data can inform us about an objective reality, it does not do so in a self-evident, unmediated fashion. Rather, real entities can only be partially apprehended. This position allows for the content of the data to be considered, whilst at the same time acknowledging multiple perspectives and the subjectivity of knowledge production (Willig, 2008). In taking this epistemological stance it is recognised that personal influences will inevitably shape the analytical process. However, by clearly stating these (see 2.8) the reader can consider the analysis in context.

**2.3. Methodology**

2.3.1. Qualitative methodology

A qualitative method was chosen on the basis of the research aim and question and the epistemological position outlined above. The goal in qualitative research is not to generate and test predictions of outcomes, but rather to provide a means of attempting to open up enquiry through allowing for rich descriptions of experience to be heard (Willig, 2008). As such, a qualitative methodology is especially appropriate for exploratory objectives (Yardley, 2000), and lends itself well to exploring sensitive and complex issues (Barker, Pistrang & Elliott, 2002). As a result, any insights generated on the basis of qualitative analysis tend to be considered to be context specific.

As previously highlighted (see 1.8) there is no existing qualitative research in this area. However, research into clinicians’ experiences of working with analogous presentations have used qualitative methodology to provide new insights (Horton, Poland, Swati, de Lourdes,de Varvalho & McArthur et al., 2010; Long, Wallis, Newman, Arcelus & Meyer, 2012; Ryan, Malson, Clarke, Anderson & Kohn, 2006). It is hoped that a qualitative methodology will allow for the emergence of a rich description of clinicians’ experiences from which themes can be generated.

2.3.2. Thematic analysis

Various approaches to thematic analysis have been described (Boyatzis, 1998; Guest, MacQueen & Namey, 2012; Joffe, 2011), with debate as to whether it is best viewed as a tool applicable across qualitative methods (Boyatzis, 1998) or a method in its own right (Braun & Clarke, 2006). Braun and Clarke (2006) suggest that the hallmark of thematic analysis is its theoretical flexibility, as it is not confined by a theoretically informed framework.

Thematic analysis was chosen as the most appropriate method for this research for several reasons. Firstly, it is compatible with the research aims and questions; it enables an inductive approach with regards to the identification and analysis of patterns of meaning in an under-researched area (Braun & Clarke, 2006). As such, it is an appropriate method to use when mapping out the range of ideas and concepts within the terrain, and lends itself well to the emergence of unanticipated findings. Secondly, it has been referred to as a method that *“works both to reflect reality and to unpick or unravel the surface of ‘reality’’’* (Braun & Clarke, 2006, p.81). Using thematic analysis in this way fits well with the critical realist position outlined above, and can be seen as a ‘contextualist’ method sitting between the two opposing epistemological positions. Lastly, it has been used to offer unique insights into professionals’ experiences of clinical management in analogous areas such as CFS (Horton *et al.,* 2010) and eating disorders (Long *et al.,* 2012; Ryan *et al.,* 2006).

Alternative research methods were considered. Interpretative phenomenological analysis gives primacy to how participants make sense of their experiences at an individual case analysis level rooted in a phenomenological framework (Smith, Flowers & Larkin, 2009). This method was discounted, as the present research does not share this idiographic focus or epistemological underpinning. Grounded theory aims to generate a plausible theory of phenomena that is grounded in the data set (Glaser & Strauss, 1967). Again this was discounted, as this research does not seek to build a theory.

**2.4. Procedure**

2.4.1. Recruitment strategy

*2.4.1.1. Service*

Participants were recruited from a Tier 4 inpatient Child and Adolescent Mental Health Service (CAMHS) situated within a paediatric hospital in London (referred to henceforth as the service). This service was chosen as it has extensive experience of working with children and young people given a clinical diagnosis of PRS. Indeed, the original case series described young people at this service (Lask *et al.,* 1991). Very few inpatient CAMHS services are known to have significant experience of working with PRS.

The service accepts referrals from children and young people aged between 7 and 15 years old. The multi-disciplinary team has particular experience of working with children and young people given a diagnosis of an eating disorder, CFS, somatoform disorder and/or a mood disorder. The overarching framework for the service is underpinned by principles of a therapeutic milieu (Crouch, 1998). This model is grounded in psychodynamic principles and emphasises individual strengths and coping strategies through facilitating strong relationships with staff and peers. This model has been explicitly recommended for use with a PRS population (Nunn *et al.,* 1998; Lask *et al.,* 2004). A précis from a recent case note review of children and young people given the clinical diagnosis of PRS (McNicholas & Nicholson,2014) is presented as a means of situating the service’s experience of working with this population and characterising the children and young people on whose experiences the participants drew (*Table 2*). As can be seen, the experience of the service is very similar to that reported by Jaspers *et al.,* (2009).

*Table 2: PRS Case Note Review 2003[[22]](#footnote-22)-2013[[23]](#footnote-23)*

|  |  |
| --- | --- |
| Number of children and young people given a label of PRS | 15 (11 female; 73%, 4 male; 27%) |
| Average age | 12 (range 10-15 years) |
| Average admission duration | 46 weeks (range 16-69 weeks) |
| Prognosis | 9 (60%) full recovery  4 (26%) partial recovery  2 (13%) met criteria for PRS at discharge |
| Predisposing factors[[24]](#footnote-24) | 9 (60%) school stress  9 (60%) family stress |
| Precipitating factors | 12 (80%) viral illness  3 (20%) injury |

McNicholas and Nicholson (2014) divided the population into two distinct subtypes of PRS: (i) angry and active or (ii) withdrawn. A relationship between subtype and recovery time was reported; the nine young people categorised as presenting with angry and active resistance had shorter admissions than those whose presentation could be characterised by withdrawn behaviour. Further, a subjectively poor therapeutic alliance between parents and the team, and clear pre-morbid separation anxiety, correlated with a poorer outcome.

*2.4.1.2. Recruitment procedure*

A letter of invitation was sent to all professionals working in the service (Appendix D). This was accompanied by an information sheet, which detailed the overarching research aims and procedure (Appendix E). Participants were requested to email the researcher at a designated email address to register their interest, stating an estimate of the number of children and adolescents with a diagnosis of PRS they had experience of working with. Those who met inclusion criteria were contacted to arrange a convenient time and location for the interviews.

2.4.2 Inclusion and exclusion criteria

In order to participate in the study, participants were requested to have clinical experience of working with at least two children and young people given the label of PRS (as deemed by the service). Eligibility was checked before an interview was scheduled.

2.4.3 Sample

*2.4.3.1. Sample size*

The sample size of 8-12 participants was set in accordance with evidence-based recommendations (Guest, Bunce & Johnson, 2006). The size of a sample is ideally established inductively until ‘saturation’ has been achieved and there are no further observable themes in the data. However, prior stipulation of participant numbers was required in order to gain permission to commence data collection. As such, data saturation was not a feasible determinant of sample size for this research.

*2.4.3.2. Participants*

Thirteen professionals expressed an interest in taking part in the research. Two potential participants were eligible but were either unable to commit to a time or did not respond to further correspondence. Another potential participant did not meet the inclusion criteria.

As part of the ethics application process, permission was sought to contact former staff members should there be an insufficient response from current staff members (see 2.5). A physiotherapist who had previously worked with the team was invited to participate after it became apparent that the current physiotherapist working in the service did not meet eligibility criteria. It was felt to be essential that the profession was represented given the crucial role of physical rehabilitation in clinical management (Lask, 2004; Nunn *et al.,* 1998). Although this participant was perhaps not drawing from as recent examples as some of the other participants, her account largely corresponded with others and there was no obvious difference between the content of the interviews.

The final sample comprised eleven clinicians from various professional backgrounds. A profile of participants can be found in Table 3. Demographic data was sought in order to contextualise the findings. Seven participants were female and four were male. Ages ranged between 28 and 48. Six participants identified as White British. One participant identified as White Other. Four participants declined to give information about their age and ethnicity.

**2.5. Data collection**

2.5.1. Semi-structured individual interviews

Individual face-to-face interviews were chosen as the method of data collection. Alternative methods, including focus groups, were considered; however, it was considered that individual interviews afforded a greater degree of confidentiality and opportunities for depth than many alternatives.

A semi-structured interview schedule (Appendix F) formed the basis of the interview. This format enabled a focus on the research questions whilst retaining flexibility to explore and follow up on matters arising in the interview context. The interview schedule was developed based on existing literature and feedback from a presentation to psychologists working in both the inpatient and outpatient CAMHS service within the hospital.

*Table 3: Profile of participants*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Pseudonym** | **Profession** | **Years worked in service** | **Number of young people with label of PRS worked with in current service** | **No. of units worked at where PRS label used** |
| Sean[[25]](#footnote-25) | Family Therapist | 2 | 4-5 | 3 |
| Max | Consultant Paediatrician | 3 | 1[[26]](#footnote-26) | not stated |
| Sarah | Mental Health Nurse | 3 | 4 | 1 |
| Emily | Systemic Psychotherapist | 4 | 3 | 1 |
| William | Consultant Psychotherapist | 4 | 6 | 2 |
| Katie | Therapeutic Care Worker | 5 | 8 | 1 |
| SR | Mental Health Nurse | 5 | 10 | 2 |
| Sunshine[[27]](#footnote-27) | Paediatric Physiotherapist | 5 | 8-10 | 1 |
| Bobby | Senior Nurse | 7.5 | 10 | 1 |
| Bella[[28]](#footnote-28) | Mental Health Nurse | 11 | 7-8 | 2 |
| Andrew | Consultant Psychiatrist | 12 | 20+ | 1 |

The interview schedule was piloted with the field supervisor, who is an experienced clinical psychologist working in the service. Several aspects of the interview schedule were consequently amended. For example, a question about the personal impact of the role was added given that many of the experiences described were very emotionally provoking. It was also fed back that it was difficult to recall experiences ‘on the spot’. As such, participants were explicitly requested to reflect on their experiences with this population prior to the interview. If this had not been possible, additional orientating questions were added at the beginning of the interview. This piloting process raised my awareness of respondent burden, meaning I subsequently ensured that the questioning was clear and transparent and that participants were aware that they could take a break at any time. The data from this pilot interview was not included in the analysis; however, its content inevitably influenced my reading of the accounts.

2.5.2. Interview procedure

Participants were offered the opportunity to choose the location of the interview; the only stipulation was that privacy must be assured. The vast majority of interviews took place within the outpatient CAMHS clinic space or the participant’s office. One participant was interviewed at home.

Each participant was given a copy of the information sheet and asked to read and sign the consent form (Appendix G). Participants were asked to choose their own pseudonym and complete a participant information sheet prior to the commencement of the interview (Appendix H). This formed the basis of Table 3. Questions concerned their profession, the number of children and adolescents with a label of PRS worked with and the number of inpatient services worked at where the role involved working with children and adolescents given the clinical diagnosis of PRS.

The semi-structured interview schedule was used to guide questioning. In order to enhance transparency regarding the research process, and reduce possible anxiety, the schedule was introduced as a template for a conversation guided by each participant’s knowledge and experiences. I also informed participants that I had no direct clinical experience with this population. Each participant was informed of his or her right to withdraw at any point without consequence. They were also offered the opportunity to take a break at any point if this felt useful. Participants were given the opportunity to ask questions and seek clarifications before the recording device was switched on.

A digital voice recorder was used to audio record all interviews. Interviews ranged in length from 42 minutes to 75 minutes with an average of 55 minutes.

Following the interview, participants were again offered the opportunity to ask questions or raise concerns. They were also offered the opportunity of a debrief.

**2.6. Ethics**

2.6.1. Ethical approval

Following registration (Appendix I), ethical approval was obtained from the University of East London (Appendix J). The study was registered with the research and development department of the participating service (Appendix K). Due to the recruitment of hospital staff, approval from the service’s Clinical Research Advisory Committee (CRAC) was also required (Appendix L).

2.6.2. Ethical considerations

*2.6.2.1. Informed consent*

In order to obtain informed consent, participants were given an information sheet at the point of recruitment and again before the interview. Participants completed a comprehensive consent form prior to commencement of the interviews, which informed them of their right to withdraw their consent to participate in the study at any point. They were given the opportunity to ask questions.

*2.6.2.2. Confidentiality*

To uphold the principal of confidentiality transcripts were anonymised with regards to names and place. The anonymised transcripts will be held on a password-protected USB memory stick for ten years before being erased.

*2.6.2.3. Anonymity*

Participants were clearly informed that whilst all names and identifying service information would be anonymised in transcripts, thesis extracts and any resulting publications, it remained a possibility that contributions might be identifiable by readers within the service (colleagues and service-users) and also, potentially, by readers outside the service. Participants were informed that this risk would be minimised by careful and considered selection of quotations.

*2.6.2.4. Distress*

Discussing experiences of working with distressed children and young people has the potential to bring to the fore some uncomfortable feelings. Participants were offered the opportunity to debrief their experiences of the interview afterwards. It was also made clear to each participant that they could request a break should they so wish. The above risk was deemed to be small and was balanced against the potential benefits of participation such as offering an opportunity to reflect, share thoughts, experiences and knowledge and contribute to practice-based evidence.

*2.6.2.5. Dissemination*

There are ethical aspects of dissemination, such as ensuring that the results are fed back to those who participated. The research findings will be presented to the service. It is hoped that a synopsis will be electronically circulated to an online professional forum for clinicians working in inpatient CAMHS settings (Quality Network for Inpatient CAMHS: QNIC) and that it will be accepted for publication in a peer-reviewed academic journal.

**2.7. Data analysis**

2.7.1. Transcription

Each interview was transcribed verbatim in a timely manner in order to ensure familiarity with the interaction and content. Punctuation inevitably influences meaning (Poland, 2005). Whilst transcripts were punctuated in order to increase readability, significant attention was given to ensure the intended meaning of the extract was retained. Transcription was considered to be a core initial process in the analytical process (Bird, 2005). Transcribing conventions can be found in Appendix M.

2.7.2. Analysis

In line with the suggestions of Braun and Clarke (2006), the analysis comprised six stages: familiarising self with data; generating initial codes; searching for themes; reviewing themes; defining and naming themes; and producing the report.

Transcripts were read and re-read several times in varying orders in order to increase familiarity with the accounts. Initial codes were generated for each individual transcript. These were organised into a coding frame structured by data segments from across the data set (Joffe, 2011). This process has been critiqued on the basis of being reductionist and enabling the abstraction of meaning from the surrounding context (Salmon, 2003). I was mindful of this critique and was careful not to prematurely reduce the richness of the data. An example excerpt of the coding process for an individual transcript can be found in Appendix N. An example of a code comprising extracts across the data set can be found in Appendix O.

Initial codes were collated into possible themes through an ongoing recursive process (Appendix P). A theme was defined based on Braun and Clarke’s definition: *“a theme captures something important about the data in relation to the research question”* (2006, p.82). Whilst a theme should denote a level of patterned response or meaning within the data set, a higher prevalence of a pattern does not necessarily equate to increased significance (Buetow, 2010). The themes were continually reviewed to ensure the coded extracts fitted with the entire data set. This process also decreased repetition and therefore increased the distinctiveness of the final themes. Themes and subthemes were defined and named though this recursive process.

A combined inductive (‘bottom up’) and deductive (‘top down’) approach to thematic analysis was used (Fereday & Muir-Cohcrane, 2006). The themes identified were strongly rooted in the data. However, the research question, my reading of existing literature and my personal and professional experiences also influenced the way the data was understood (see 2.8). In line with the social constructionist epistemological position taken, the analytical process progressed from analysing semantic content to the interpretation of the deeper latent meaning that encompasses broader contextual factors (Joffe, 2011).

**2.8. Researcher reflexivity**

From a social constructionist perspective meaning is always ‘given’ to data and not simply identified within it (Willig, 2013). My own experiences have inevitably informed my reading and presentation of this research. I consider the following professional experiences, interests and influences to be particularly pertinent:

1. I have no direct experience of working with a child or young person given a label of PRS. However, I undertook a BSc placement year within a specialist outpatient CAMHS team attached to the participating service eight years ago. In this role I observed an assessment of a young girl where the diagnosis of PRS was queried. She was subsequently admitted to the inpatient service, and I was able to identify her from some of the participants’ descriptions. Whilst I did not disclose this experience to participants, I was aware of my curiosity to know more about her particular outcome. I also recalled how I had felt confused, anxious and hopeless whilst observing the assessment. I tried to be mindful regarding the ways in which this may have impacted on my questioning.
2. I undertook my third DClinPsych. placement within the same specialist outpatient CAMHS team. Whilst I did not know any of the participants personally, some of their names and faces were familiar. I reflected on how my prior knowledge of the inpatient service may have informed the questions I asked. I was careful to explicitly state that I was no longer working in the outpatient service; being perceived as an internal staff member might have impacted on the way in which participants related to me and potentially changed the content and nature of the interviews. I also tried to attune to possible assumptions that may be made about me based on my gender, appearance and status as a fellow health professional and how these may have influenced responses.
3. Prior to training, I was involved in designing and implementing a day service programme within a specialist outpatient CAMHS Eating Disorder Service in the context of a scant evidence-base. Clinical management decisions were often shrouded in ambiguity. It was important for me to maintain a curious position to avoid imposing on participants and the data aspects I personally had experienced as helpful when working in this context (which included cohesive and supportive teamwork and the need to develop a clear and explicit rationale for intervention).
4. Through training I have developed a particular interest in both systemic models of working and critical psychology approaches. The importance of theorising power (Foucault, 1977; Hagan & Smail, 1997), collaboration through exploring multiple positions and co-constructions of difficulties and solutions (Anderson & Goolishian, 1988) and redressing social injustice through clinical practice and social action (Patel, 2003) have had particular impact. I considered how this might have led me to focus on privileging certain issues, both in the interviews and the analytical process. For example, I noticed my particular interest in the ways in which the label of PRS was used by different participants and tried to remain curious and open minded about the impact of this.

From a personal perspective, I identify as a White British woman in my late twenties. I feel it is important to invite the reader to know about an aspect of my own developmental history. Though I have no clear memories of it, my parents tell me that when I was four years old I regressed developmentally to the extent that I became unable to talk for several weeks. My difficulties quickly and completely resolved after, following advice, my parents talked to me about family stresses they had been trying to protect me from. I believe that growing up hearing this story shaped my understanding of how inextricably linked mind and body are, as a counterbalance to the clear legacy of Cartesian dualism evident in dominant discourse that enables a distinction between physical and mental health.

I sought to ensure that I remained curious and as open-minded as possible through the use of a research journal (Ortlipp, 2008). I found documenting my reactions following research interviews particularly useful and continued to detail my thoughts during transcription and analysis. An example excerpt can be found in Appendix Q.

**3. RESULTS**

**3.1. Overview**

The main research findings are presented in this chapter. The analysis is structured by themes and subthemes representing the way in which the data was understood.

**3.2. Summary**

The analytical process identified three main themes and nine subthemes. These are represented in Table 4.

*Table 4: Themes and subthemes*

|  |  |
| --- | --- |
| **Theme** | **Subtheme** |
| Making sense | Similar yet different |
| Pervasive. Refusal. Syndrome? |
| Frameworks for understanding |
| Uncertainty | The impact |
| Negotiating uncertainty |
| Our way | Re-humanising relationships |
| A family alliance |
| Clear expectations |
| Censoring talk |

**3.3. Making sense**

This theme refers to the variety of ways in which the concept of PRS was perceived and understood. The large majority of participants spoke about the complexity of the meaning-making process.

*“…[[29]](#footnote-29)I find it sort of completely baffling’’.*

SR. Line 12.

*“…it’s just really complicated”.*

Sunshine. Lines 25-26.

*“…it’s a puzzle”*.

Sean. Line 107.

Complexity was reflected in the way some participants spoke in a longwinded or circuitous manner. Katie reflected on this directly.

*“…I think possibly my ramblings or not cloudy but complex answers and not being able to give you straight answers just represents it* (PRS)[[30]](#footnote-30) *itself”.*

Katie. Lines 878-881.

3.3.1. Similar yet different

This subtheme highlights difference amongst participants with regard to whether or not a discrete category of PRS was warranted.

Many participants compared and contrasted PRS to other, more familiar, presentations. These included psychiatric diagnoses (anxiety, depression, obsessive compulsive disorder, oppositional defiant disorder, eating disorder), medically unexplained symptoms (CFS and chronic regional pain syndrome), neurodevelopmental disorders (autism spectrum disorder) and various organic presentations, including leukodystrophy. A particular emphasis was given to similarities between PRS and CFS and/or medically unexplained symptoms.

*“…I think there’s a lot of overlap with chronic fatigue…and to be honest the symptoms are pretty much the same, how I’ve seen it”.*

Katie. Lines 494-499.

*“…he wasn't really* (PRS) *although he had many* (features)*, he didn't actually have a formal diagnosis. It comes and goes with chronic fatigue but he presented kind of with PRS.”*

Bobby. Lines 1146-1150.

*“…I think they fit within the box of medically unexplained”.*

Max. Lines 80-81.

Many participants appeared to suggest that given the degree of overlap, dimensional means of conceptualising were more helpful and accurate than categorical means. For example, many participants could be seen to conceptualise the difficulties as falling towards one end of a spectrum or as part of a broader constellation of difficulties.

*“…if you think of Venn diagrams there’s overlap between them so chronic fatigue syndrome would be one, pervasive refusal syndrome might be another, pervasive withdrawal PAWS might be another, somatising illness, anorexia certainly, eating disorders”.*

Andrew. Lines 136-141.

Where PRS was perceived to be a distinct entity, participants appeared to suggest that its main differentiating features were the pervasiveness of ‘active’ refusal (extending to all areas of function, rejection of help and resistance to ‘recovery’) and/or the extent of ‘inconsistencies’ in function. At times, this construction of PRS appeared to be used heuristically in order to differentiate between presentations: the ostensibly genuine inability inherent in CFS was compared to the volitional nature of the refusal characteristic of PRS.

*“…I think there's a question mark about how much is chronic fatigue and how much is pervasive refusal, so there's a question mark about how tired he is fatigued or unable to do, versus what he is actively refusing to do”.*

Sean. Lines 261-265.

Beth: “*Do you think it makes sense to have a separate category of PRS?”*

Sarah: *“Yea…in the sense of like that element of resistance to wanting to get better which you don’t really get with a lot of other disorders”.*

Lines 896-899

*“…with some of them they have a diagnosis of pervasive refusal but yea they’re more chronic fatigue in the fact that they don’t do anything because they find it too tiring, and then you’ll get ones of the diagnosis of pervasive refusal where it seems to fit a lot more because there are all these inconsistencies in what they can and can’t do”.*

SR. Lines 256-263.

3.3.2. Pervasive. Refusal. Syndrome?

This subtheme relates to the ways in which the term given to the presentation impacted on the process of making sense, at both an individual and collective level. It also highlights potential consequences of using the term with regard to family engagement.

Many participants suggested that the terminology either accurately described the main clinical features or was useful in giving a sense of what the main difficulties may be. This is interesting given recent debates regarding nomenclature.

*“…I think it* (the label) *does what it says on the tin”.*

Sunshine. Lines 494-495.

Beth: *“I wondered if you felt the term PRS was a helpful way of conceptualising the presentation?”*

Bella: *“(5) I think when you stop and think about the words and what they actually mean I think it’s quite right, because I think the refusal to receive help and to let people in is actually all pervading. So I think if you break it down and actually think about what it means then I think it’s a fairly good description”.*

Lines 559-566.

*“…it gives you an instant picture of what you’re expecting to see in the room”.*

William. Lines 595-597.

However, there was a strong sense amongst participants that the term PRS was not highly valued. Some participants appeared to question whether the label aided the process of understanding. The way in which some participants expressed uncertainty regarding whether some of the individuals they discussed had formally been given a diagnosis of PRS also suggests the label was not seen as clinically useful. Many discussed their tendency to avoid using the term, preferring instead to describe the main features without using the word ‘refuse’.

*“…I don’t think it adds anything other than naming stuff which is what we all like to do”.*

Max. Lines 180-182.

*“…I think it’s much more helpful to kind of name and being able to say out loud what the actual behaviours are and the experiences, rather than actually labelling it”.*

Bobby. Lines 222-226.

*“…labelling and giving a diagnosis isn't always the most helpful thing to do…if I was talking to a young person about it I would use the description I did earlier that it seems like things have got really hard and you've kind of opted out of life”.*

Bella. Lines 626-627.

The term PRS was widely perceived to be a potential obstacle to the engagement of families and young people. Most participants suggested that the word ‘refusal’ implied a degree of active control. This was seen to apportion a degree of blame, and infer a level of individual agency at odds to the way in which children and young people and their families often make sense of the presentation.

*“…I personally don’t think the word pervasive refusal helps, although I think that’s what it is”.*

Katie. Lines 795-797.

*“…I think parents can sometimes feel that it sounds as if the child is being blamed and perhaps they are being blamed and I think blame is very unhelpful”.*

Emily. Lines 635-638.

SR: *“…she was quite upset when she got a diagnosis of pervasive refusal,* *I think they think we think they have a lot more control over it, which isn’t necessarily the case”*

Beth: *“Does that then have an impact on the engagement?”*

SR: *“Yea it can do, in that particular case I think she felt a bit betrayed by us”.*

Lines 528-536.

A small minority of participants used the term ‘pervasive withdrawal syndrome’ (PWS) as an alternative to PRS. This shift in terminology appeared to have been relatively recent. For these participants, emphasising withdrawal seemed to offer a more satisfactory means of making sense and provide a more neutral basis upon which to engage families.

*“…I don’t like refusal…it’s actually withdrawal”.*

Sarah. Lines 976-977.

Beth: *“What do you feel the impact of giving this label* (PRS) *to families is? What’s the impact on the therapeutic relationship?”*

Andrew: *“Um I think it varies, I think families often don’t like the word refusal so I prefer to say pervasive withdrawal syndrome on the whole”.*

Beth: *“Ok so would that now be the term that you would be using?”*

Andrew: *“More likely, more likely”.*

Lines 182-190.

This change in terminology did not appear to have been discussed and agreed as a team. Many participants did not mention the term PWS at all, whilst for others the shift in terminology added to the challenge of making sense.

*“…in recent cases we’ve had others that have presented very similar, but they’ve been given pervasive withdrawal which is quite confusing to us, well to me anyway, because we never really had this pervasive refusal and nobody has really told us what the big difference is”.*

SR. Lines 536-561.

*“…I don’t know whether PWS is superseding PRS do you know?”*

William. Lines 606-607.

3.3.3. Frameworks for understanding

This subtheme highlights how, in the absence of a clearly delineated construct of PRS, participants drew on various theoretical frameworks to aid understanding.

Contradiction regarding the nature of the refusal was a significant feature of the accounts; the task of delineating intentional from unintentional behaviour appeared to be particularly challenging for participants. Whilst the overwhelming majority of participants stressed their understanding that the young person had no agency in their apparent refusal and rejection of help, many simultaneously conferred a degree of volition. This was most clearly demonstrated through the choice of language (such as “won’t” as opposed to “can’t”), and the way in which agency was often implied when discussing apparent inconsistencies in function. At times, some participants spoke in ways that implied they felt both a volitional and non-volitional explanation for the behaviour might be accurate.

*“…I don’t know if that’s how I would describe it* (PRS) *but yes I suppose I would say unable or unwilling to get out of bed”.*

Emily. Lines 130-136.

Contradiction was also compounded by the fundamental lack of consensus regarding the meaning and nature of the word ‘refusal’. For example, some participants thought it was possible to unconsciously refuse: *“…they have no conscious decision to refuse”.* (Sarah, Lines 849-851). Others disagreed: *“…refusal is something you choose to do. You don’t by accident refuse”* (Bobby, Lines 1530-1531). Divergent understandings and perspectives regarding the nature of refusal added to the complexity of making sense on a collective level.

*“…the crux of the issue* (is) *there's a real disparity of understanding among everyone about how much is active choice and how much is unconscious behaviour”.*

Bobby. Lines 630-634.

Although participants rarely explicitly situated their understandings of PRS within a theoretical framework, they could be seen to largely draw on one of two overarching frameworks. Firstly, some participants described PRS as akin to a developmental functional regression: the child or young person has regressed in terms of their developmental ability. From this perspective, the child or young person tended to be constructed as helpless, dependent and analogous to a baby.

*“…when you first have them they’re just infantile, they really are, foetal position, lying on the bed, eyes closed, mute, breathing, not reacting to your voice”.*

Sunshine. Lines 13-15.

*“…it’s a bit like the first year of life where you kind of come into the world…it takes a baby about a year to be able to verbalise, start to be able to eat solids, start to be able to take its first steps, it’s kind of the same process”.*

Bobby. Lines 273-282.

Alternatively, other participants implied that the presentation was best described a mental state where the child or young person lacked full conscious awareness. A couple of participants with additional training in psychodynamic theory explicitly drew on the concept of the unconscious in the process of meaning-making, through drawing on the theory of a psychic retreat (Steiner, 1993)

*“…it’s a kind of unconscious retreat when things get too difficult”.*

Sarah. Lines 856-857.

*“…the patient needs to withdraw to go into that retreat, to stop to non-exist disconnect completely from their bodies and their mind and the world around them”.*

William. Lines 615-618.

Others spoke about how the young person appeared to have gone to a ‘safer place’ in the mind but did not appear to be specifically referring to the theory of a psychic retreat. This description evoked an analogy of PRS as akin to a state of reduced consciousness or even death. At the same time, however, language used (such as ‘abandonment’ or ‘opting out of life’), implicitly inferred a degree of agency.

*“…(it’s) kind of a complete yea an abandonment of their life”.*

Katie. Lines 50-51.

*“…I think about it as them opting out of life…the only option available to them was to just choose, I mean the opposite of choose life, choose not life”.*

Bella. Lines 97-103.

The fact that there often appeared to be no significant or unique precipitating factors, outside the realm of those commonly experienced in late childhood and early adolescence, added to the challenge of understanding the life-threatening severity of the presentation. Again, participants drew on different frameworks when making sense of the development of the difficulties. Some participants appeared to foreground individual vulnerability as a particularly significant factor. This was interpreted as akin to the stress-vulnerability model described by Zubin and Spring (1977).

*“…often there’s a normal life event that most other people could manage, but something about their personality or their family or their support system or whatever means that they don’t have the capacity to manage, and so they end up with pervasive refusal”.*

SR. Lines 132-138.

*“…we had a few that when they came out suddenly it was quite evident that maybe they were even on the autistic spectrum, so maybe the difficulties that had been there in the past you know bullying whatnot that then spiralled was because they weren’t able to manage their learning in those environments”.*

Sunshine. Lines 508-514.

Others appeared to implicitly invoke the systemic concept of circularity in the development and maintenance of the difficulties. The way in which changed dynamics within the system were foregrounded in this process resonates with the systemic theory of problem-maintaining feedback loops (Hayes, 1991).

*“…the whole family almost sort of just reverts back, something is fulfilled that wasn’t being fulfilled before”.*

Sarah. Lines 209-211.

*“…sometimes there’s secondary gain to being bed bound especially if you’ve got loads of attention from your family or presents or your parents are by your bedside while your other siblings are at home”.*

Andrew. Lines 394-398.

**3.4. Uncertainty**

This theme highlights the way in which the lack of a cohesive unifying framework heightened a perception of uncertainty amongst the team. This could be seen to discern a considerable impact on clinical management. The ways in which participants appeared to negotiate uncertainty will be explored.

3.4.1. The impact

Uncertainty could be seen to have a detrimental impact on the collective confidence of the team. Some participants compared confidence levels when working with this population to the greater levels of confidence experienced when managing presentations with a more established evidence-base.

*“…I think it (.) means we are more vulnerable (.) to erm sometimes a collective lack of confidence or clear sense of direction umm a bit sort of working blindly sort of feeling”.*

William. Lines 455-459.

*“…compared to other things I think there'd be less confidence, less certainty”.*

Sean. Lines 226-227.

Participants took up different positions in relation to this uncertainty and lack of collective confidence. For many, uncertainty accentuated the need to adopt a curious and tentative approach to the work: “*you have to be so patient, you really don’t know”* (William. Lines 93-95). Whereas for others uncertainty unhelpfully increased anxiety, which was seen to have an insidious impact on the confidence of all those involved in the admission:*“…I think confidence* (is helpful) *and if you don’t have it then you pretend to have it”* (Bella. Lines 129-131).

In the absence of consensus, ostensibly rather modest differences in opinion were interpreted as becoming amplified and polarised. This was most notable in the way in which some participants highlighted the distinctive approach taken by the service and/or compared the service model to alternative approaches. The extracts below demonstrate the way in which stressing the differences between approaches could be seen to result in the formation of an ‘us’ and ‘them’ dynamic.

*“*(the other view is) *basically you leave people and you do minimum care and…you don’t do any more than that, it’s very much that’s what they want you to do, that’s not what we do”.*

Sean. Lines 194-197.

*“…at* (hospital) *we’ve got* (service). *That’s very different from everywhere else in the world”*

Max. Lines 319-320.

Beth: “*What’s the impact of that* (conflicting opinion)*?”*

Sarah: “*I think it makes people believe in our approach more”.*

Lines 524-527.

In some ways this dynamic appeared to be perpetuated by the very widely held perception amongst participants of a weak and inadequate evidence-base. Existing literature was widely regarded to be of poor quality on the basis of being ‘unscientific’ in contrast to more rigorous and systematic evidence such as that obtained from randomised control trials.

*“…certainly if we were going to grade evidence it ain’t up there is it? Level 1 or level 2 in fact it’s almost anecdotal, so it’s poor quality evidence anyway which I wouldn’t tend to follow”.*

Max. Lines 442-445.

*“…I say well there’s not a lot of literature and they* (families) *often say well I've read this and that and I say read it again because it’s not actually research, it’s opinion”.*

Sean. Lines 438-441.

Many participants appeared to distance themselves from existing literature by suggesting that it contradicted the service model and/or did not reflect their perception of what was helpful. Many participants thought the service was particularly well placed to conduct research as it was the ‘best’ place for an admission and had the greatest amount of relevant experience. Some expressed frustration that they had not prioritised publishing their own literature in order to provide an alternative perspective.

*“…there’s nothing out there to say what we do… I really feel like we should have been doing* (research) *and I’ve been kind of cross that we haven’t”.*

Bobby. Line 1694-1704.

*“…there’s nothing out there, there’s no guidelines on how to work for it and for me its so frustrating that somebody* (can publish) *loads of stuff and there’s nothing to go against that so what’s out there is accepted because its published and you know you think if a paper is published then* (they) *must know more but there’s nothing else out there to contradict it so why would anyone believe anything different?”*

Sarah. Lines 931-940.

Lastly, uncertainty exerted an impact on the relationship between professionals and families. Many participants reflected on the challenges they had encountered when trying to work with families in this context. For example, trying to ensure families placed trust and faith in the approach, whilst, at the same time, acknowledging that the team were unable to offer any certainty, both in terms of the nature of the presentation and outcomes.

*“…they* (family) *want to know what it is so if you tell them you don’t know what it is it’s quite difficult”.*

Sean. Lines 146-147.

*“…it’s hard because you don’t want to give them* (family) *false hope or dash their hopes either when you don’t know what’s going to happen”.*

SR. Lines 547-549.

*“…there have been times when families have said what’s the research base that you’re working from? What’s your statistics? What’s your proof that this approach works? And we are kinda asking them to take a bit of a leap of faith”.*

Bella. Lines 222- 226.

The challenge of engagement also appeared to be compounded by the dominant perception of parents as anxious, very familiar with PRS literature and quick to seek a second ‘expert’ opinion. Many participants suggested that second opinions often contrasted with those advocated by the service. This was interpreted as feeding into a vicious cycle: uncertainty leads to divergent opinions, leading to confusion, which then raises anxiety and increases the likelihood that clinical authority will be challenged. This in turn results in covert conflict with other services, perpetuating an ‘us’ and ‘them’ dynamic which blocks the potential for collaboration, thereby maintaining uncertainty.

*“…I think quite often if they don’t agree with the way we are working they’ll refer to other articles that say well children should be left”.*

Emily. Lines 414-417.

*“…other places across the country have different views and it’s becoming a bit of a…battle”.*

Bobby. Lines 1452-1453.

3.4.2. Negotiating uncertainty

This subtheme relates to the way in which participants negotiated the impact of uncertainty. This could be seen in three main ways: turning to practice-based evidence, tolerating uncertainty and minimising differences.

Some participants spoke about how the absence of a strong evidence base meant newer staff members often learnt vicariously from more established team members. There was a sense that practice became established on the basis of it being ‘tried and tested’.

*“…I think a lot of what how people learn or their knowledge base comes from anecdotal information from other staff”.*

Bella. Lines 214-216.

“(I) *really had to follow the expertise of the team and each other and kind of learning as you went”.*

Sunshine. Lines 129-130.

*“…I’ve worked here so long that that’s the way we’ve done things and so you learn it that way”.*

SR. Lines 208-210.

For most participants, learning from the knowledge and skills of more experienced colleagues appeared to be unproblematic, effective and largely sufficient. However, others questioned whether such a strategy was adequate, possibly in light of the evidence-based discourse that pervades psychiatric services.

*“…we don’t have any real evidence about what’s going on outside of our own experience, and what kind of evidence is that?”*

William. Lines 660-661.

A second means of negotiating uncertainty was through tolerating uncertainty regarding the cause of the difficulties. The decision around when to stop the search for an organic explanation appeared to be delegated to the paediatrician. However, most participants seemed to have reconciled themselves to the fact that aetiological certainty was not likely to be a realistic prospect. Time and experience seemed to be particularly important in helping participants to take this position. Importantly, there was a sense that it was important for the team to model tolerating uncertainty in order to facilitate a shift towards engagement in a rehabilitation model amongst the young people and their families.

*“…the most helpful thing is…to recognise when it’s time to stop doing more investigations…that’s incredibly important because it allows people to get onto a rehabilitation model”*

Max. Lines 185-196.

*“…what I learnt was it’s not worth looking for that starting trigger it’s more about how we manage what’s happening now and how to move forward”.*

Sunshine. Lines 26-29.

*“…when I first started working with these sorts of children I always thought oh there must have been something that happened…but actually after a couple of years working with them, often there isn’t”.*

SR. Lines 127-132.

A final way in which uncertainty appeared to be negotiated was through minimising differences between a PRS management approach and the approaches taken for other presentations often seen within the service. Several participants appeared to suggest that the overarching framework used to guide other interventions was equally applicable to this population.

*“…you know sometimes you might get into a debate about is it ME or is it pervasive withdrawal or whatever, the treatment is very similar…so actually it’s a little bit academic in that sense”.*

Andrew. Lines 216-225.

*“…I think pragmatically we treat this like we treat other medically unexplained stuff”.*

Max. Lines 453-454.

*“…it’s the same treatment whether you’ve got CF or PRS or whatever, gradual rehabilitation (.) usual stuff, what else do you do?”.*

William. Lines 468-469.

**3.5. Our way**

This theme captures aspects of clinical management perceived to be particularly helpful in this work. Most participants explicitly stated that the approach is not clearly delineated: *“…the treatment isn’t absolutely cut and dried”* (Andrew, Lines 333-334). However, the name of the theme aims to reflect the fact that common strands could be pulled together. The word ‘our’ refers to the previous interpretation that uncertainty at times appeared to promote an ‘us’ and ‘them’ dynamic amongst the staff team (see 3.4). All participants strongly expressed the need for a psychiatric inpatient admission including multifaceted input from a comprehensive multi-disciplinary team and a fully adapted environment in order to meet physical care needs.

3.5.1. Re-humanising relationships

This subtheme highlights the way in which the overwhelming majority of participants perceived strong therapeutic relationships to be the bedrock of the approach.

Strong therapeutic relationships with the child or young person were perceived to be fundamentally important. The overarching service model of a therapeutic milieu was viewed as facilitating an environment whereby relationships were highly valued. Empathy, patience and kindness were widely seen as necessary qualities to build trust and strong relationships. Interpersonal qualities tended to be spoken about as more useful than specific therapeutic techniques. Implicit in accounts was a suggestion that the biggest task for professionals was to bear witness to distress through simply ‘being with’ the young people.

*“…the therapeutic factors when they do meta-analysis they're always the same so the type of therapy is fairly small, it doesn't matter what you do as long as you listen to people and be nice to them and they like you and stuff like that. I feel a bit the same”.*

Sean. Lines 426-231.

*“…I felt that the most important thing was the therapeutic relationship with the child or the young person so sometimes they wouldn’t really be looking at you but essentially they can’t really cut off their ears so it would just be talking and letting them know I was there”.*

Sunshine. Lines 56-61.

*“…just being present and listening…because even if they’re not communicating they’re still listening”.*

Sarah. Lines 395-397.

As highlighted previously (see 3.2.3.) descriptions of the presentation frequently invoked analogies of the young person as akin to a baby or somebody with significantly reduced conscious awareness. This could be seen to exert a tangible impact on the behaviour of professionals. For example, some participants described a tendency for people to talk in front of the young people on the assumption that they can’t hear. Strong relationships were interpreted as providing an antidote to this, through a process of re-humanising.

*“…it’s really easy in these sorts of cases to forget who the patient is, because everything else is going on around them and they’re not actually doing anything, so it’s actually important to talk to them* (the young person) *on the assumption they can hear you”.*

Max. Lines 218-220.

*“…I think what can happen when you have someone that presents as a baby…is that idea out there in the world of oh they don’t know, they didn’t hear that, but you forget that the 15-year-old they absolutely know what’s going on, they’re observing all the time, that’s the only skill that they actively do if they open their eyes”.*

Katie. Lines 612-619.

Many participants, particularly those from the nursing team, gave examples of the ways in which this process of re-humanisation could be established through relationships. Examples included: the use of de-psychologising language (*“…just using normal words not using therapy words”*. Bella, Lines 163-164); telling humorous personal anecdotes; involving young people as much as possible in all aspects of their care; joking with colleagues in front of the young person; and showing empathy, kindness and honesty wherever possible. For Katie, this approach was powerfully reinforced by unexpected comments one young person made upon discharge.

*“…I just try and make sure that you develop something* (a relationship) *and I think that’s stuck with me from when we had a girl…she* *was wailing but one of us* (staff) *was in front of her trying to read her stories whilst the other was brushing her hair...when she left she said I’ll always remember you were reading to me*…*I was really like wow she remembered that”.*

Katie. Lines 354-359.

Several participants discussed how parents occasionally shared photos or videos of their child as healthy and happy with the team. This was seen to be important in nurturing hope amongst professionals for the young person’s future. There was also a sense that professionals found remembering those with whom they had previously worked extremely rewarding, and that cherishing and sharing these memories was embedded in the team culture. Such practice appeared to foster alternative analogies reflecting the re-humanising process, such as the child as a butterfly.

*“…we always mention you know Jamie or Jim[[31]](#footnote-31) and wow she’s doing this now and well it’s akin almost to bringing someone back from the dead”.*

William. Lines 716-719.

*“…it’s almost like in that time some of them were like little butterflies you know they cocooned and they gradually emerged as a new individual…it was brilliant and it made it all worth it and helped you to take a deep breath for the next one”*

Sunshine. Lines 448-556.

Developing the young person’s ability to verbalise unexpressed emotions through facilitating emotion recognition was also seen as an integral part of the re-humanising relationship. This was given particularly high regard due to the widely shared view that these young people, and in some cases also their families, had ‘poor psychological mindedness’ and, as such, found it challenging to voice interpersonal issues and conflict. Helping the young person to name their emotions and experiences in order to increase their understanding of themselves, and experience a sense of connection to others, was widely perceived to be a core task of the admission. Encouraging this facilitation appeared to be a role all members of the MDT were invited to take up, rather than the domain of a particular profession.

*“…we encourage them to talk about emotions and naming what they think is going on for someone else and yea that’s kind of like an everyday piece of work”.*

Sarah. Lines 303-306.

For several participants, developing an ability to verbalise was central to promoting therapeutic change.

*“Gradually over time the child starts to put things into words more and I think that’s part of the process of getting better”.*

Andrew. Lines 285-287.

However, it should be noted that a minority of participants questioned the degree of emphasis placed on verbalising and naming, particularly in terms of its ability to facilitate behavioural change. This suggests that the emphasis given to naming is not unanimously agreed within the team.

*“…I haven't really ever seen someone say I was worried about this, I was anxious about this or I’m scared about this and its translated to movement in their condition…other people might have but I can't remember, well none of the ones that I’ve seen”.*

Sean. Lines 742-748.

*“…I don’t in principle object to that* (emphasis on talking about emotions), *there’s nothing intrinsically wrong with that, but I suppose given that these children aren’t talking I wouldn’t particularly prioritise it”.*

Emily. Lines 260-266.

A potential challenge to the development of re-humanising relationships appeared to be the strength of emotions these children and young people often evoked. Many participants referred to powerful emotions of frustration, sadness and rejection. Others also discussed an urge to protect and ‘mother’, which challenged professional boundaries. Emotions appeared to be particularly heightened at times when individuals perceived themselves as unable to meet the child’s needs (*“…as a human you want to fix but you can’t”* Sunshine, Line 357), or when providing intimate care in the context of apparent ‘refusal’.

*“…I’ve just been shocked at how powerful it is, like I’ve never felt like that with a patient before”.*

Sarah. Lines 732-733.

*“…it stays with you a bit more than the other children I think because the rawness of their distress seems more in some cases”.*

SR. Lines 600-602.

*“…there’s something that feels quite provocative when you're doing intimate cares with a child that age, especially if you're coming from the mindset that it’s an active refusal… it stirs something up in you”.*

Bobby. Lines 1125-1142.

One means of managing the emotional demands of the caring role appeared to be through seeking distance. This could be overt, such as asking somebody else to take over, or a covert, largely unconscious, process, such as expending significant effort on detailed care planning which resulted in less time to process difficult emotions.

*“…it’s really hard work…I’m like I’m just going to step out of the room like I’ve done that once and I just can’t do it anymore, like someone take over”.*

Sarah. Lines 735-738.

*“…the temptation…is to put plans in place and plans are essential…but that is part of the issue”.*

Bobby. Lines 100-106.

Participants also reflected upon how best to facilitate relationships in a context of predominating ‘refusal’ and ongoing medical risk, which can change the focus of the intervention.

3.5.2. A family alliance

This subtheme recognises the importance of families and the team working in partnership in order to promote recovery.

Working with parents was seen to be extremely important, not least due to the voluntary legal status of the admission and the perceived importance of the young person experiencing professionals and parents working together in ‘alliance’.

*“…the alliance between the family and the parents in particular is very, very important”.*

Andrew. Lines 335-337.

*“…staff and parents working together that’s a big one, a really big one”.*

SR. Lines 215-216.

*“…the others that I can think of that did go well…it always felt like everyone was on the same page or singing from the same song sheet”.*

Bella. Lines 304-310.

Participants overwhelmingly concurred that a division between the team and parents was a poor prognostic factor. Differences of opinion between families and professionals, be they concerning the aetiology of the presentation or the most appropriate treatment approach, were perceived to be a challenge to ensuring a strong alliance.

*“…where we haven’t been so successful is where we have had a very difficult and complicated relationship with the parents…prognostically that’s an important point”.*

William. Lines 498-500.

*“…I think sometimes we haven’t been successful because the parents have just not been on board.”*

Emily. Lines 409-411.

A particular dilemma concerned strengthening engagement with the family through remaining open to their views and opinions, whilst also providing a clear and consistent approach perceived to be in the child or young person’s best interests. The challenge was interpreted as difficult to reconcile. It could be seen to be negotiated based on individual circumstances. Important factors appeared to include whether the family had previously agreed with the approach or whether they were likely to be litigious.

*“…you have to stay on board with the family but then again that is really difficult I mean do you hold your ground and say this is what we are doing, this is what we believe in, this is what we believe is right, or do you shape your response to the family?”*

Sean. Lines 374-378.

3.5.3. Clear expectations

This subtheme highlights the importance of clear expectations, both within the context of a graded rehabilitation model and underpinning the admission as a whole.

There was consensus amongst participants that the most appropriate intervention approach was underpinned by principles of graded rehabilitation. This appeared to be a very established aspect of the intervention and not one that was particular to a PRS admission. Participants understood the aim of scaffolding to be gradually increasing independence and responsibility over time.

*“…so say for eating we would start by getting our hands helping their hands to do it, and slowly over time they would take more and more responsibility until they’re doing it by themselves, and that works, for us that works really well in the children I’ve seen”.*

SR. Lines 191-195.

Consistency was frequently highlighted as an essential ingredient of success. However, a significant challenge concerned finding an optimum balance of expectations; participants agreed there needed to be enough expectations to provide momentum but not enough to unduly place pressure on the child or young person. In the context of a lack of protocol for pacing, this appeared to be negotiated by constant monitoring and evaluation.

*“…I don't really think there’s a recipe for that* (pacing) *I think it is a bit you do something reflect on it see if it works”.*

Sean. Lines 467-469.

All participants agreed that it was most appropriate to grade expectations in line with ability: too high expectations were perceived to potentially undermine confidence in the intervention and lead to a ‘slippery slope’ of decreasing expectations. However, participants varied in their views regarding how firmly expectations should be adhered to. Bobby reflected that rigidity amongst the team could mirror the prominent features of the presentation itself.

“…(you need to be) *able to stop and feel ok to change your plans because that’s the other way you can go, you can stick and dig your heels in and be just as actively refusing”.*

Bobby. Lines 404-407.

For many participants, the emphasis given to graded rehabilitation was perceived to differentiate the approach from other services working with the same population.

*“…the difference of emphasis can be sometimes how much you encourage them to move forwards and set goals and like I say we don’t force anything…but on the other hand we’re not completely hands off, and some people might say to work in that way”.*

Andrew. Lines 583-589.

Families and young people were perceived to benefit from having clear expectations of what to expect prior to the admission. Some participants expressed a view that staff, families and young people should all expect to tolerate a degree of distress in pursuit of increased function. A handful of participants felt that it would be helpful for this to be conveyed more explicitly to families, particularly prior to admission.

*“…it would be easier if we were able to say to parents this is what we find helpful, but we would anticipate that your child will become very distressed, agitated, won’t want to co-operate. We find that it’s helpful nevertheless to still try and push through with that”.*

Emily. Lines 381-386.

*“…I don’t think a child should be admitted unless…they’re* (parents) *all on board because what ends up happening is...care stops, all the adults don’t know what to do…and the conflicts are (.) they’re not managed. So pre-admission planning is really important”.*

Bobby. Lines 1368-1381.

However, at the same time, there was significant reflection on the difficulties of presenting a very clear and consistent expectation to young people and their families. Challenges included the possible ramifications should a young person not be admitted given difficulties negotiating expectations (as they may then not receive a service) and/or potential difficulties in maintaining therapeutic relationships.

*“…I think we need to be more clear on our programme…the difficulty is that lots of the patients that we have…have nowhere else to go, so if we would be really strict…then we have a risk that we could be isolating them…it’s a very fragile thing…*(but) *if we start making exceptions from the beginning, I don’t think that helps”.*

Katie. Lines 677-682.

3.5.4. Censoring talk

This subtheme highlights the way in which reflecting on what was and was not likely to be helpful to verbalise to the child or young person was perceived to be an integral element of the management approach. This was particularly the case for the nursing team.

There was a sense from many participants that it was necessary to regulate their speech with regard to what was and was not likely to be helpful for young people and their families to hear. For example, despite their frequency and prevalence, participants concurred that naming inconsistencies was likely to be unhelpful – particularly in the earlier stages of an admission. Participants appeared to have learnt to censor talk on the basis of past experience; many gave examples of times when naming inconsistencies resulted in deterioration in the child or young person’s functioning. It should be noted that some participants highlighted how this approach was taken when working with other presentations (such as conversion disorder) and that not naming was not specific to a PRS admission.

*“…it is very tempting to say “oh but I’m having to help you to put your hand to your mouth but I’ve just seen you reach over there for that why can you not do that?”…but point it out and they will go backwards to show to you that they can’t”.*

SR. Lines 245-250.

*“…I think that drawing attention to it…potentially puts pressure on the young person to do that again, to do it all of the time and improve, so I wouldn't comment on it”.*

Bella. Lines 387-392.

*“…I’ve knocked on someone’s bedroom door because they are bedbound and literally I mean hearing them walking the room and they’re like “just a minute”…then hearing the pitter patter, and then after walking in you just have to keep going”.*

Katie. Lines 130-136.

Participants appeared to create a context where children and young people would feel safe enough to test out progress and know that this would not be explicitly reacted to. This could be seen to be developed by overlooking progress, letting the young person know increased function would not unduly raise expectations and stressing that progress was not perceived to equal recovery.

*“…I remember like she snuck a cornflake into her mouth and nobody talked about it, which then meant that she would carry on doing it”.*

Sarah. Lines 752-755.

“(you need to) *let the patient have the understanding that you realise that although they’ve increased their function things are still bad”.*

Max. Lines 381-384.

Censoring talk concerning inconsistencies was observed to have significant ramifications. Firstly, the notion that this conversation topic was ‘off limits’ appeared to set a precedent for professionals to evade other types of talk. For example, the majority of participants appeared to be reticent to discuss precipitating factors for the distress directly with the child or young person, even at the point of discharge, for fear of causing distress or impairing progress. The implications of this are two-fold. Firstly, the child or young person may not be given the opportunity to develop a coherent narrative around their difficulties. Secondly, avoiding these types of conversations appeared to perpetuate the notion that this population can be characterised by ‘poor psychological mindedness’.

*“you don't want to take them back there cos you're worried and you don't want to distress them. You're not sure how able they are to go and think about that. I actually wonder if we do them a disservice by not giving them some kind of narrative around their experience”.*

Bobby. Lines 993-1000.

*“…with these kids they never formulate stuff like that. They just sort of get back into life”.*

Sean. Lines 812-813.

*“…you asked a very important question about what can patients verbalise when they’re not in that state…the child themselves find it very hard to describe – they really don’t have the language”.*

William. Lines 649-654.

Beth: “*Have you ever had a sense from those who’ve got better what was going on?”*

Max: *“They don’t talk about it”*

Beth: *“Do you mean you don’t talk about it with them or they don’t talk about it full stop?”*

Max: *“They don’t talk about it, I don’t think”.*

Lines 539-545.

Secondly, the avoidance of certain conversations with young people appeared to be mirrored in the way in which some ‘difficult’ conversations with families appeared to be circumvented.

*“…it can be quite difficult with those families in terms of having the conversations around what’s going on in the room, like almost like the elephant in the room that doesn’t quite get named but is talked around”.*

Sarah. Lines 353-354.

*“…I think um there should perhaps be an opportunity for the children and family to have perhaps more difficult conversations around some of the potential triggers to the difficulties”.*

Emily. Lines 237-240.

Finally, censoring talk presented an emotional challenge for participants. The need to remain composed, and not react, could be seen to stir up strong emotions, particularly frustration.

*“…probably the biggest challenge would be frustration …because sometimes you'll see them do things that they say they can't do when people aren't watching”.*

Bella. Lines 341-344.

*“…I can get really frustrated I can get to a place where I’m like “they’re so in control, this is so irritating”. You can see the inconsistencies”.*

Katie. Lines 127-129.

The potential for overwhelming emotions to be evoked was seen to underline the importance of staff practices that promote open and unrestricted expression. Many participants appeared to highly value formal and informal spaces, such as process meetings, supervision and team debriefs. There was a widely shared view that the effectiveness of these approaches lay with their ability to promote effective communication and a reflective space within which participants could talk honestly and without fear of judgement.

*“…I think what really helps me is… to sit in a room that’s safe with people that trust my practice and won’t judge me if, excuse my French, if I’m like “this kid’s driving me fucking insane” ”.*

Katie. Lines 170-173.

**4. DISCUSSION**

**4.1. Overview**

This chapter opens with a discussion of the way in which the findings of the present study elucidate the research questions. Recommendations are then made in light of the findings. A critical review outlining issues relating to factors affecting the quality of the research is discussed, followed by a reflexive review. Conclusions of the research will then be drawn.

**4.2. Findings in relation to research aims**

The guiding aim of this study was to qualitatively analyse the understandings and perspectives of treating professionals who have experience of working with children and young people given a label of PRS. The research aspired to contribute towards practice-based evidence, inform service development and approaches to staff support and provide the basis for further clinically relevant research. Thematic analysis was used to explore the following research question:

**What are the understandings and perspectives of treating professionals working with children and young people who have been given a label of PRS?**

Specific focus was given to:

1. perspectives on clinical management;
2. challenges and dilemmas

The research question will be discussed in light of the findings. Implications will be drawn and additional literature will be discussed where relevant in order to add new insights.

Whilst there is debate regarding the role of generalisability in qualitative methodology, Willig (2001) contends that once a given experience has been identified, it is also likely to be found in other contexts. As such, I consider that the findings of this research provide insights that are potentially of interest and relevance to other teams working with this population, and also professionals working with other presentations where the nature of the diagnosis is contested.

4.2.1. Making sense: contradictory understandings and perceptions

This theme highlighted the variety of ways in which participants perceived and understood the presentation termed PRS. That PRS was widely regarded as confusing and challenging to understand is not surprising given the complex and contested nature of the presentation. However, divergent understandings and perspectives amongst participants were interpreted as a significant challenge to the development of a consistent, integrative and shared framework for understanding. Many of the findings mirrored existing schisms reported in the literature.

*4.2.1.1. PRS: a discrete entity?*

Participants differed over whether or not they thought a discrete category of PRS was warranted. Understandings appeared to be situated through comparing and contrasting to other diagnoses. That particular discussion concerned similarities and differences between PRS and CFS is unsurprising given the context of the service (Fiebelman *et al.,* 1990), and the fact that many children and young people who receive the label of PRS have previously been given a diagnosis of CFS. This finding expands on previous literature that has debated whether or not PRS should be seen as a discrete entity (Jaspers *et al.,* 2009; Jans *et al.,* 2011; McNicholas *et al.,* 2013; Nunn *et al.,* 2014; Nunn & Thompson, 1997).

An interesting finding was the way in which participants who had worked in the service for a similar period of time could be seen to give quite disparate estimations regarding the number of children and young people with a diagnosis of PRS they had worked with (*Table 3*)[[32]](#footnote-32). It may be inferred from the degree of discrepancy that participants may use their own heuristic criteria to differentiate PRS. An alternative explanation may be that estimations varied due to the unreliability of memory. A possible means of mitigating against this might have been to share the list of names collated for the case note review (McNicholas & Nicholson, 2014), and ask participants to reflect on their experiences of their work with each named individual prior to the interview. However, this is likely to have reduced the richness of this clinically important data.

A dimensional means of formulating the difficulties was perceived by some participants to be a helpful way of making sense of the presentation. This perhaps reflects a growing trend towards problematising a categorical model of psychiatric diagnosis (Widiger & Samuel, 2005; Esterberg & Compton, 2009). However, other participants justified the existence of PRS as a separate diagnostic entity. The way in which degree of active refusal was highlighted as a differentiating factor accords with previous literature (Jaspers *et al.,* 2009; Lask *et al.,* 1991; McNicholas *et al.,* 2013). However, the focus on inconsistencies in function, as a second distinguishing feature of PRS, has not received the same degree of attention in published literature (see *Table 1*).

*4.2.1.2. PRS: volitional?*

Delineating intentional versus unintentional behaviour in the presentation appeared to be particularly challenging for the majority of participants to reconcile. Many strongly expressed a view that implicating volition was unhelpful. However, it was inferred – from both subtleties of language, and the way in which active volition and inconsistencies in function were highlighted as means of differentiating PRS ­– that the population were perceived to have a degree of agency by professionals. This finding mirrors similar contradictions in the literature (Jaspers *et al.,* 2009; Nunn *et al.,* 2014) and perhaps represents the legacy of earlier literature that clearly distinguished a discrete category PRS on the basis of a wilful and volitional refusal (Lask *et al.,* 1991; Nunn & Thompson, 2007). This implies that there may be a role for psycho-education in future training programmes for professionals working with this population.

*4.2.1.3. PRS: a useful term?*

Participants were divided regarding the appropriateness of the term PRS. Some felt that it accurately described the main clinical features, whereas others felt it implied blame and incorrectly presumed agency. Two participants appeared to suggest PRS would be better termed pervasive withdrawal syndrome (PWS) – a term not described previously in the literature[[33]](#footnote-33) – however this did not appear to have been mutually agreed within the team. A more consistent finding was the way in which the term PRS was widely devalued on the basis of potentially causing friction in therapeutic relationships. This lends support to those advocating a shift in terminology away from foregrounding ‘refusal’ (PAWS: Nunn *et al.,* 2014). The finding that the term PRS appeared to be rarely used in direct clinical work mirrors findings in other areas where diagnosis is contested. For example, clinicians working with adolescents given the diagnosis of borderline personality disorder have also been found to prefer an approach that describes the presentation without invoking labels (Koehne, Hamilton, Sands & Humphreys, 2013). Whooley (2010) describes this as a ‘workaround’.

*4.2.1.4. PRS: different theoretical understandings?*

Participants described the presentation in markedly different ways. Some appeared to understand the difficulties as a functional developmental regression akin to a functional equivalent of a neurological regressive syndrome. This description resonates to some extent with the conceptualisation of a refusal-withdrawal-regression spectrum suggested by Jaspers *et al.,* (2009). However, unlike current findings, Jaspers *et al.,* (2009) contended that PRS fell within the refusal end of the spectrum, and that regression represented a far more ‘serious’ presentation. Other participants implied a lack of full conscious awareness in their descriptions of the presentation, akin to a state of reduced consciousness. This has not previously been reported in the literature. There were also differences in the degree to which participants implicated individual vulnerabilities or wider systemic dynamics in the development of the difficulties. Interestingly, participants seldom appeared to draw on previously proposed theoretical understandings of PRS in order to aid the process of understanding, such as learned helplessness (Nunn & Thompson, 1997), depressive devitalisation (Bodegård, 2005) or nervous system hyper-arousal (Nunn *et al.,* 2014).

*4.2.1.5. PRS: different subtypes?*

In their case note review of the children and young people with a label of PRS worked with in the participating service McNicholas and Nicholson (2014) grouped the population into subtypes (either a predominantly angry *or* withdrawn presentation). This distinction was not found in the current study[[34]](#footnote-34): participants did not appear to naturally group the population into these categories. I was only party to this data following completion of the interviews, meaning I did not have an *a priori* assumption and enabled participants to describe their understandings and perspectives freely[[35]](#footnote-35). The use of static categories in the case note review raises interesting questions – particularly as the case note review did not reference the term PWS used by participants in this research. It would be particularly interesting to explore what the authors were drawing on to inform this categorisation, and whether subtypes of an angry or withdrawn presentation within the overarching umbrella term of PRS is experienced by professionals as accurate or useful.

*4.2.1.6. Implications*

These findings suggest that a significant challenge for the future is the development of a coherent, integrative and shared framework for understanding in order to help participants make sense of the presentation. Such an endeavour may benefit from considering the following:

* Should the presentation termed PRS be seen as a discrete entity?
* If so, what are its defining features?
* What should the presentation be called, if anything?
* What is the utility of clinical diagnosis, and for whom?
* Which frameworks should guide clinical practice?
* How applicable are these findings to those working with other contested diagnoses?

4.2.2. Uncertainty: negotiating uncertainty at an individual, group and intra-group level.

This theme highlighted the ways in which the lack of consensus regarding the nature of PRS increased uncertainty, and presented a significant challenge to confidence and consistency in clinical management. The way in which participants could be seen to try and negotiate uncertainty can be understood at different levels of explanation.

*4.2.2.1. Individual level*

Cognitive dissonance theory (Festinger, 1957) holds that individuals have a tendency to seek consistency amongst their cognitions. In the event of dissonance, an individual may either modify a conflicting cognition or aim to reduce its importance. Participants in this research could be seen to negotiate uncertainty through similar means. For example, dissonance appeared to be reduced through minimising the importance of finding aetiological ‘answers’, emphasising the need for pragmatism, and/or focussing on similarities between an intervention for PRS and other presentations.

*4.2.2.2. Group level*

The finding that many participants thought ambiguity decreased confidence at a group level is not surprising. Similar findings have been reported in literature that has focussed on the experiences of professionals working in analogous areas such as medically unexplained symptoms (Furness, Glazebrook, Tay, Abbas & Slaveska-Hollis, 2009) and CFS (Chew-Graham, Dowrick, Wearden, Richardson & Peters, 2010). Again, participants could be seen to negotiate this through valuing pragmatism. Whilst a pragmatic focus may increase group confidence, to do so at the expense of pursuing a conceptual understanding of the presentation appears limiting. For example, it may perpetuate uncertainty in the longer term through hindering group discussion regarding the nature of PRS, and obstructing the development of a shared rationale that could be used to promote consistency in management.

An interesting finding was the way in which, in the context of uncertainty, the group appeared to position evidence in relation to clinical practice. Rather than acknowledge the interrelated nature of both evidence-based practice and practice-based evidence, the two seemed to become dichotomised. For example, several participants bemoaned the absence of robust evidence-based practice to guide clinical management due to the lack of systematic and rigorous research. This concurs with research suggesting that certain forms of evidence and methodology occupy a lower status on a proclaimed hierarchy (Ramchandani, Joughin, & Zwi, 2011). However, at the same time, practice-based evidence was implicitly highly valued in the way in which many participants explained the way they learnt vicariously from more experienced team members. The uncritical way in which some participants described this process resonated with the way in which knowledge acquisition has been compared to the inheritance of conformity: knowledge can become valued for its apparent authority, rather than as an idea or tool to guide understanding (Main, 1990). On reflection, I wondered whether individuals might find it challenging to question established beliefs and whether this may stifle service development. This finding implies that more needs to be done in order to bridge the perceived gap between evidence-based and practice-based evidence.

*4.2.2.3. Intra-group level*

At a wider macro level, uncertainty and lack of consensus regarding clinical management could be seen to exert a significant impact on intra-group processes. In particular, relatively modest differences in approach (mostly relating to the emphasis given to graded expectations) appeared to become amplified and polarised. This was interpreted as resulting in an ‘us’ and ‘them’ dynamic in relation to other services and existing literature. Bion (1961) proposed that when groups face uncontrollable anxiety they may operate from within unconscious basic assumptions. It is suggested that one of these basic assumptions – a ‘fight-flight’ assumption, in which the group acts as if its main task is to fight or flee from some common enemy – appears to be of relevance here. This finding has important implications, as it suggests that services may find collaboration and dissemination of best practice more difficult. It is also important given that participants reported that differences of opinion regarding management between services increased the challenge of engaging families.

4.2.3. Our Way: strong relationships

This theme highlighted perspectives concerning the intervention approach. Participants unanimously agreed that a multifaceted intervention in a psychiatric setting including specialist input from a comprehensive MDT and a fully adapted environment were necessary pre-requisites for a good prognosis. This is in line with previous literature (Lask, 2004; Nunn *et al.,* 1998). However, in direct contrast to previous literature, PRS was not seen as necessitating a *“separate distinct management approach”* (Nunn *et al.,* 1998, p.328). Rather, it was interpreted that each aspect of the intervention ultimately related to the task of forming a strong consistent therapeutic alliance at an individual, family and team level.

*4.2.3.1. Relationship between professionals and child or young person*

In many ways, the finding that a strong therapeutic relationship with the child or young person was considered the bedrock of the intervention is unsurprising. Firstly, the service model is embedded in the concept of a therapeutic milieu that gives significant importance to relationships (Crouch, 1998). Secondly, previous research in this area has emphasised the need for strong relationships (Guirney, 2012; Lask, 2004; Nunn *et al.,* 1998) and a strong relational bond with a play therapist was perceived to be central to the recovery of an 8-year-old girl given the label of PRS in a paediatric service setting (Lee *et al.,* 2013). Lastly, the importance of the therapeutic relationship has been elucidated in qualitative research into the perspectives of professionals in analogous areas, including medically unexplained symptoms (Furness *et al.,* 2009), CFS (Horton *et al.,* 2010) and eating disorders (Reid, Williams & Burr, 2010; Ryan *et al.,* 2006). The role of relationships in facilitating change has also been found to be a robust finding across presentations and psychotherapy modalities when using meta-analytic methodology (Norcross & Wampold, 2011) including in children and adolescents (Shirk & Karver, 2003).

The focus on therapeutic relationships as a means of re-humanising has not been reported in previous literature. Whilst earlier papers emphasised relationships as a core hope-promoting strategy (Nunn *et al.,* 1998; Lask, 2004) participants in the current research did not appear to draw on the same theoretical framework of learned helplessness. The focus on re-humanising relationships through ‘being with’ appears to resonate with Rogers’ (1961) humanistic assertion that individuals need certain environmental conditions as a pre-requisite for personal growth: genuineness (openness), acceptance (unconditional positive regard) and empathy (being listened to and understood). The importance of relationships that bestow a sense of identity in relation to others has been recognised in other areas where functioning is compromised, such as in work with those whose cognitive functioning abilities are challenged. In his pioneering work with adults given the label of dementia, Kitwood (1997) offered an alternative to the dominant medical discourse. The alternative paradigm emphasises putting the person first through underscoring ‘personhood’: *“a standing or status that is bestowed upon one human being, by others, in the context of relationship and social being”* (p.8). Principles of promoting personhood (such as recognition, collaboration and containment) correspond with the way in which participants in the current study described the task of developing strong relationships.

Participants agreed that the primary task of the admission should be everyone working together towards an agreed goal. The finding that clear expectations are regarded to be a core management strategy has been discussed in previous literature (Nunn *et al.,* 1998; Lask, 2004; Lee *et al.,* 2013). In the current study, goals appeared to be operationalised by scaffolding expectations in line with current functioning. Such practice corresponds with the developmental theory of the zone of proximal development (Vygotsky, 1978), which stresses that social interaction is the basis for learning and growth. This approach was also perceived to be a core means of facilitating peer integration, which is an aspect of social development widely perceived to be compromised in this population (Jaspers *et al.,* 2009). Participants discussed the challenge of finding an optimum balance between setting enough expectations, whilst at the same time not placing too much pressure on the child or young person. In the absence of a clear protocol, this appeared to be negotiated through continually evaluating the impact of expectations on the therapeutic relationship and taking steps to amend where necessary.

The current study suggested a subculture of censoring talk, particularly with regard to not naming apparent inconsistencies in function. This was understood as a means of not compromising relationships. However, the findings suggest that it may become harder to puncture reality as time goes on, and that participants may become complicit in the avoidance of potentially distressing conversational topics. A potential impact of not addressing these issues may be the perpetuation of the perception of this population as having ‘poor psychological mindedness’. This finding resonates with Hare-Mustin’s (1994) depiction of the therapy room as having walls lined with mirrors that reflect back only the discourses that the therapist and individual bring to their interactions. It could also be seen that comparatively fewer resources appeared to be dedicated towards helping the child or young person build a coherent narrative around their experiences. This may hinder the development of self-identity and increase the risk of relapse.

*4.2.3.2. Relationship between professionals and parents*

At a wider level, the finding that participants considered close collaboration with parents to be desirable separates the current findings from previous literature that clearly advocates substantial separation between parents and their child (Nunn *et al.,* 1998; Bodegård, 2005a; Jaspers *et al.,* 2009). The overwhelming consensus amongst participants regarding the importance of a positive and strong family alliance is to be expected: working in partnership with parents is stipulated in both law and CAMHS policy (Children Act, 1989; National Service Framework: children and young people, 2004). It also mirrors a wider trend in services towards recognising family as a potential resource. The existence of a strong relationship between the treating team and parents was widely perceived to be predictive of a good prognosis for the young person and vice versa. This supports the findings of the case note review (McNicholas & Nicholson*,* 2014) and a comprehensive meta-analysis (Shirk, Karver & Brown, 2011). Some participants discussed the challenge of being perceived as an ‘expert’ by families given the level of uncertainty surrounding effective management. A collaborative and curious stance, underpinned by systemic principles, was widely regarded to be an effective means of building a strong alliance. Whilst this may be helpful for many families, others may potentially experience a ‘non-expert’ stance as lacking attunement to their need for certainty and authority (Allen & Allen, 2002). Professionals may therefore need to find a means of validating the family’s hopes and anxieties and providing containment, whilst not colluding with the impression that professionals necessarily have the ‘answer’. Considering multiple contexts of conversation (e.g. Coordinated Management of Meaning: Cronen & Pearce, 1982) offers a means of remediating disjunctures in interaction.

Participants appeared to suggest that outlining clear expectations to the family, particularly in advance of the admission, facilitates strong alliances. The desirability of devising a management contract prior to the work commencing has been outlined in previous literature (Nunn *et al.,* 1998) and echoes a core standard for practice outlined by the CAMHS Tier 4 Steering Group Report (2014). Clear expectations were perceived by participants to be a means of providing a containing and consistent environment, rather than a means of preventing manipulation of plans by parents as previously described (Nunn *et al.,* 1998).

Participants concurred that whilst building alliances with families is fundamental, there are significant challenges to be negotiated. Such challenges in inpatient CAMHS teams have been well described by Gross and Goldin (2008). Current findings build on previous research by elucidating the ways in which a widespread lack of consensus amongst those working with this presentation can potentially amplify tension between the family and team. Furthermore, the practice of censoring talk appeared to set a precedent for more challenging conversational topics to be circumvented or minimised in family and team interactions, such as challenging potentially unhelpful dynamics between parents and their children. Structural systemic therapists consider it necessary for professionals to first ‘join’ the family system in order to understand the invisible rules that govern its functioning, before a therapist can usefully intervene (Minuchin, 1972). However, current findings imply a tendency for participants to get stuck in the static position of censoring talk (which may be helpful initially) and consequently find it difficult to navigate other possibilities.

*4.2.3.3. Impact on team dynamics*

A subculture of censoring talk appeared to have developed – particularly amongst the nursing team – in part as a means of developing strong relationships with the children and young people. This could be seen to exert an impact at an emotional level, particularly in terms of evoking frustration. The findings extend the previous description of frustration when working with this population (Guirney, 2012) by highlighting the way in which frustrations may be intensified by contradictory beliefs regarding the nature of ‘refusal’ and difficulties in providing consistency between professionals. Previous literature has suggested that both paediatric nurses (Edwards & Done, 2004) and parents (Anonymous, 2001; Lee *et al.,* 2013) find not being able to talk about certain conversation topics emotionally challenging. This research has highlighted the fact that this is also found amongst professionals working in a psychiatric service context. The anxiety aroused by the task of providing care could be seen to have the propensity to form collective defences as a means of guarding against difficult emotions. A particular example was the way in which some participants described care plans becoming the focus of a great deal of attention and energy, to the extent that participants could be distracted from the task of ‘being with’. This resonates with Menzies Lyth’s (1960) description of ritual task performance as a social defence formed by groups of professionals in an attempt to eliminate anxiety.

On the basis of previous literature, I had expected to find some suggestion that management occasionally erred towards becoming punitive (Nunn *et al.,* 1998). It appeared that in-built structures for staff reflection and discussion, such as clinical supervision and peer support, largely safeguarded against this possibility. Indeed, the degree of agreement regarding the importance of professionals talking openly and freely with each other was impressive in the context of such divergent understandings.

*4.2.3.4. Implications*

Findings concerning the centrality of strong alliances to the work need to be highlighted for several reasons:

* Competency frameworks usually preface core competencies, such as the development of a strong therapeutic alliance, as a foundation for other competencies (e.g. Competency Framework for CAMHS, 2011). However, this message can sometimes be overshadowed by a drive towards the acquisition of modality specific skills. Current findings suggest that building strong relationships should be regarded as the principal task of the work.
* In the current socio-political context policies increasingly emphasise the need for evidence-based practice in health care settings. Operationalising the process of therapeutic change through relationships can be challenging, and quantitative research paradigms often aim to exclude or control for such ‘confounding’ variables.
* The rhetoric of austerity authorises service cuts. This inevitably results in increased demands on staff time, potentially reducing opportunities to build strong relationships.

**4.3. Recommendations**

Given that these findings have contributed new insights into working with this population, there are several recommendations to be made across different levels of practice. In light of the perceived similarities between PRS and other presentations, it is suggested that some of these recommendations may also be helpfully applied to those working with differential diagnoses (see 1.6) and professionals working with other presentations where diagnosis is contested.

4.3.1. Facilitating strong therapeutic alliances with individuals and families

The findings suggest the importance of facilitating strong individual and family alliances. Services may usefully consider means of enabling and empowering professionals to simply ‘be with’ the child or young person and perceiving this to be core to the therapeutic work. Findings also suggest that it may be useful to encourage families to share videos, photographs and other keepsakes with the team in order to promote the development of relationships between individuals and professionals. Recruiting reflective professionals, who embody qualities of empathy, patience and warmth is also likely to be beneficial.

Professionals may also find it helpful to consider ways in which to develop the young person’s own understanding of their experiences as a means of strengthening their self-identity. This appears to be particularly important given the interpretation that professionals may be complicit, to some extent, in evading such conversations by censoring talk. Narrative therapy techniques have much to offer in this regard (White & Epston, 1990). Facilitating creativity and adapting practice to meet the needs of the population is likely to be helpful. For example, using digital applications (e.g. ‘iDraw’ or ‘BookMaker’) may be experienced as more acceptable by the child or young person, at least initially, than traditional talking therapy. Clinical psychologists are particularly well placed to facilitate such work, and oversee the transition to outpatient settings in order to reduce the risk of relapse.

Attending to the development of strong family alliances is also likely to be beneficial. Reflecting upon the relationship to help of all members of the system (including professionals) (Reder & Fredman, 1996), and actively listening to possibly disparate views and connecting this difference to find a shared pathway through a process of collaboration, are likely to be prerequisites of strong relationships. Further, the use of systemic therapeutic techniques – such as reflecting team discussions in which the family and young person are invited to bear witness to team discussions (Andersen, 1987) and externalising the problem (Tomm, 1989)[[36]](#footnote-36) – may reduce unhelpful dynamics. The implementation of ‘family friendly’ practices, such as pre-admission outreach visits undertaken by the team, are also likely to create a context for a strong alliance through empowering parents (Gross & Goldin, 2008).

4.3.2. Facilitating consistency and clear communication amongst teams

Service development may usefully prioritise practices that promote consistency and clear communication. The use of a communication book (Lee *et al.,* 2013) or diary (Beverley & Wright, 2013) may provide a practical means of sharing information, knowledge and experiences amongst professionals.

Professionals may also benefit from collaboratively reflecting upon their understandings and perspectives of working with those given the label of PRS. In particular, it is suggested that professionals discuss the practice of censoring talk. If, as appears to be suggested, it is agreed to be helpful to censor talk at the beginning of the admission this might helpfully reduce cognitive dissonance for clinicians and enable them to focus on developing strong bonds. Professionals should also be aware that such practices have the potential to promulgate a general reticence to broach certain conversational topics. Ongoing team reflection and discussion about the tendency to become stuck in this position may increase confidence in creating the frame for difficult conversations and facilitate a more dynamic process. A shared agreement regarding when and how to start making inroads to naming is likely to be experienced as more containing for professionals and helpful in terms of promoting consistency.

The present study highlights the ways in which effective use of both formal and informal means of supervision can be experienced as helpful, particularly in terms of mediating the significant emotional challenges often experienced in this work. Utilising an external facilitator for at least some of this supervision, as is the case in the participating service, may help to avoid role conflict and increase open and honest communication (Appleby, 1987). Further, Guirney (2012) suggests the use of a named ‘back-up’ helper amongst the nursing team in order to ensure that support is available when needed.

4.3.3. Establishing novel clinical networks

It is important that services work together towards consensus. Conflicting advice and opinions is likely to decrease congruency amongst services and increase confusion for service-users. It is suggested that the Delphi method may offer a means of exploring or achieving consensus on areas of dispute, such as terminology. This method draws together existing knowledge and pinpoints areas of agreement and disagreement through a series of cycles of anonymous written decision and argument, managed by a facilitator. The iterative approach means arguments can be refined and tested until they are robust and fully considered. As participants do not need to meet, it enables group communication that otherwise might have been impossible due to geography, time or other constraints (Stone, Fish & Osborn, 1992). This method has been used to develop clinical practice guidelines regarding the definition, diagnosis, assessment and intervention in developmental communication disorder (European Academy of Childhood Disability, 2011).

Secondly, it is important that services are empowered to share best practice and research findings. There are several existing means of communication amongst inpatient psychiatric services. The Quality Network for Inpatient Clinicians (QNIC) aims to provide a single channel for the effective dissemination of information and co-ordinate information sharing between psychiatric units. The Forum for Inpatient Child and Adolescent Psychology Services (FICAPS), a member network within the British Psychological Society, has a similar remit. It is highly recommended that their use should be capitalised upon in order to enable services to learn from one another and work together.

Clinical networks bringing together professionals working in psychiatric and paediatric services are warranted. Such networks may also be promoted in the context of the NHS Five Year Forward View (2014), which emphasises the need for closer integration between traditionally divided services in future models of care. One such means may be the establishment of a Practice Research Network (Campbell, 2003), a large network of clinicians or teams who collaborate to collect and report data gathered in the ‘real world’ over long time periods (rather than data collected for specific orchestrated clinical trials). Virtual teams are able to come together, exchange ideas and develop projects. A useful first step may be to assess the estimated national prevalence of presentations termed PRS (including PWS and PAWS), which will provide useful clinical information alongside data for clinical commissioners and resource allocation.

4.3.4. Theoretical recommendations

Given the apparent divergence of perceptions and understandings of PRS widening the lens, re-examining theoretical assumptions and drawing together different frameworks would appear to be timely. Working towards a more holistic and integrated paradigm to understand the distress may facilitate consistency, coherence and confidence within services working with this population.

Developing a holistic bio-psycho-social formulation may provide one means of opening up understandings, particularly given that these findings suggest many participants appeared to have implicitly internalised a mind-body schism. Thompson and Nunn (1997) also noted a tendency for those working with this population to see the distress through either a psychological *or* biological framework. By viewing the difficulties through a wider bio-psycho-social lens the following observations around normative child development become useful to consider:

* Neurobiological changes in early adolescence[[37]](#footnote-37) such as synaptic pruning and myelination that enables faster neural connections (Sowell, Siegel & Siegel, 2011).
* The unique tasks of development in adolescence, including negotiating autonomy, individuation and separation (Hall, 1904; Havinghurst, 1953; Erikson, 1959) alongside changes in cognitive development (Piaget, 1936).
* Sociocultural, economic and political factors which may increase the challenges of negotiating adolescence in a modern democratic society (Mead, 1928). Furthermore, families may become stuck in terms of re-negotiating relationships between developmental stages (Carter & McGoldrick, 1994), and concern within the system may perpetuate distress through vicarious reinforcement (Brown & Inouye, 1978).

A possible implication of widening the lens may be demonstrating the potential utility of liaising with a wider range of health professionals, such as clinical neuropsychologists. This could potentially facilitate more comprehensive understandings through bridging the mind-body schism previously highlighted. It may also bring alternative frameworks and policies to the attention of services. For example, the National Service Framework for Long Term Conditions (2005), (which includes paediatric neurological presentations), suggests guidelines for best practice that may usefully be applied to services working with PRS. Furthermore, the International Classification of Functioning, Disability and Health – Children and Youth version (ICF-CY: WHO, 2007) offers a common language and terminology for difficulties involving functions of the body. Moving towards the use of holistic and integrated frameworks is one means of bridging the existing gap between paediatric and psychiatric services, both of which have experience of successfully working with this presentation (Appendix C).

Opening up theoretical understandings may also enable a far greater focus on difference within the population, such as explaining the preponderance of females given this label. It may also highlight the fact that ethnicity is seldom discussed as potentially pertinent. Reflecting on the impact of difference may also be clinically useful; considering the social GRRAACCEESS[[38]](#footnote-38) acronym (Burnham, Palma & Whitehouse, 2008) may increase awareness of the way in which different aspects of identity are important, interrelated and constitutive of a person’s experience. This may lead to improved consideration of the possible impact of difference. For example, the experiences of a 15-year-old boy, who will be in the minority with respect to both gender and age, is likely to be very different from the experiences of a 12-year-old girl, who may be perceived as fitting within the usual ‘norms’ (*Table 2*: McNicholas & Nicholson, 2014).

4.3.5. Future research

Service-user perspectives, particularly regarding experiences of the admission, are much needed and may contribute new and valuable insights. For example, qualitative research in analogous areas has found differences between aspects of the work valued by service-users and those valued by professionals in the field of eating disorders (de la Rie, Noordenbos, Donker, & van Furth, 2007). Anecdotal reports suggest that those who are given the label of PRS may be less inclined to partake in research. Services may helpfully consider discussing consent to be contacted for research purposes with each family throughout the admission. This may potentially provide an opportunity to negotiate the best means of contact – should consent be given – in order to empower individuals to take an active role in decisions concerning research.

Findings of this study suggest that a lack of outcome data in the area has the potential to increase tension between families and services. Routine follow-up, with standardised quantitative and qualitative measures, may be implemented in order to provide useful outcome data. Additionally, the assertion that psychiatric services are best placed to cater for the significant needs of these young people needs exploring, particularly as difficulties termed PRS may be successfully managed in paediatric settings across the UK (Edwards & Done, 1994; Lee *et al.,* 2013). It is argued that a comparison of specialist and non-specialist care pathways may be helpful in eliciting outcome data. For example, a London-wide study of care pathways in eating disorders found specialist intervention was over twice as effective as non-specialist pathways (House, Schmidt, Craig, Landau, Simic, Nicholls *et al.,* 2012).

Research underpinned by different epistemological positions may help to illuminate some of the questions that have been raised by this research and triangulate findings. For example, employing a constructionist methodology may permit a greater exploration of discourses underlying the management of PRS and their impact in a clinical setting. Such research may also offer an interesting insight into the role of language in constructing societal perceptions of the presentation. Findings suggest that there are additional emotional challenges faced when working with a population where inconsistencies in function are perceived to be common; phenomenologically oriented research may helpfully explore this further. Lastly, research combining quantitative and qualitative research paradigms, such as Q factor analysis methodology, may also be usefully used to explore understandings as a means of working towards consensus[[39]](#footnote-39).

Cross-cultural research is indicated, as diagnostic conceptualisations are known to vary cross-culturally. For example, differences in the way in which clinicians in the UK and US interpret ‘mania’ in younger children have been reported (Dubicka, Carlson, Vail & Harrington, 2008). Given that the term PRS appears to have been used in only a handful of countries, it is highly likely that the same types of difficulties, and perhaps the notion of refusal itself, are thought about and understood differently across cultures. Such differences become particularly important where different conceptualisations permit potentially harmful interventions, such as ECT (Cizadlo & Wheaton, 1995) and the prescription of barbiturates (McNicholas *et al.,* 2013).

Future research may benefit from utilising research designs that enable multiple comparisons within the data. These findings appeared to suggest a strong unity of views amongst certain professional backgrounds, particularly nursing. It may be useful to compare and contrast understandings and perspectives across professional backgrounds, and perhaps other variables such as level of experience and number of services worked in where the label of PRS is given. Expanding data collection across psychiatric and paediatric services would also widen the lens of current research.

Lastly, research in the field of medical sociology has demonstrated that the name given to a presentation has the potential to change the way in which it is experienced (Link, Cullen, Struening, Shrout, & Dohrenwend, 1989) and have a detrimental impact on prognosis (Hamilton *et al.,* 2005). It would be interesting to research whether a similar process of nominative determinism applies in this area. For example, do those who are given the label PRS have a different prognosis to those given the label PWS or PAWS?

**4.4. Assessment of research quality**

Various means of assessing the quality of this research were considered (Braun & Clarke, 2006; Smith *et al.,* 2009; Spencer & Ritchie, 2011). Yardley (2000) suggests the following principles, which have been assessed to fit with the critical realist epistemological stance taken.

4.4.1. Sensitivity to context

Sensitivity to context was demonstrated in the introduction chapter through situating the development of PRS in a sociocultural context and identifying gaps in the literature. The collection of data relating to participants’ demographics and experiences (*Table 3*), alongside data contextualising the service’s experience of working with PRS (*Table 2*), highlights sensitivity to context. Careful selection of illustrative extracts that represent the full range of perspectives, and the inclusion of questioning in the extracts where possible also provide a means of increasing sensitivity to context.

4.4.2. Commitment and rigour

Commitment and rigour were demonstrated through various means including piloting the initial interview schedule (to ensure the interview questions were inline with research aims) and ensuring a careful and thorough analysis that adequately addressed the variation and complexity observed. Rigour was also achieved through supervision, which helped me to remain committed to the epistemological underpinnings of the research. Furthermore, the research question and epistemology were coherent with the method of investigation and analysis undertaken. The problematic features that can befall thematic analysis were considered (Salmon, 2003) and the data was interpreted rather than summarised.

4.4.3. Coherence and transparency

To increase coherence, a list of all quotes for individual codes and each subtheme were read through by my supervisor and discussed in supervision as part of the analytical process. Transparency in the analysis was achieved by detailing every aspect of the data collection and analytical process, and by presenting excerpts of the data in which readers can themselves discern the patterns identified in the analysis. A detailed account of researcher reflexivity, and details of the research journal kept to document reflections, was provided in the methods chapter as a means of upholding transparency.

4.4.3. Impact and importance

It is suggested that this area is important to research for various reasons including: the lack of existing literature; the potentially life-threatening nature of the presentation; and the emotional and financial cost of a PRS admission. Focusing on the clinical management of the presentation offers an opportunity to draw some important conclusions, which, it is to be hoped, will directly inform clinical practice, service development and future research. The findings will be discussed in relation to relevant literature in order to make a novel contribution to the area (see 4.2).

**4.5. Critical review**

As the criteria proposed by Yardley (2000) provide only one means of evaluation, it is considered necessary to critically evaluate this research paying particular attention to the way in which methodological choices may have impacted on the quality of data and confidence in the findings.

As previously mentioned, service user perspectives are absent in this area of literature. In hindsight, this study may have amended its design in order to incorporate such perspectives. For example, service users could have been asked which questions they would have liked to have been included in the semi-structured interview questionnaire. Alternatively, professionals may have been asked to reflect on the questions they thought service-users might have wanted to ask during the interview.

Findings should be considered in the context of the particular characteristics of those who participated. The sample, though self-selecting, was representative of the wider professional configuration in the service. It is suggested that the diversity of participants with regard to clinical experience and professional affiliations added to the richness of the data. However, the sample is limited in three important respects:

1. Understandings and perspectives are limited to those of participants working in one service setting. There may, as suggested by some of the participants themselves, be significant differences between services. This limits the ability to generalise from the findings.
2. Professionals with experience of working with fewer than two children or young people with a diagnosis of PRS were excluded from participating in this research. This is an important limitation as it may be that those with less experience have different understandings and perspectives. An unintended consequence of the inclusion criteria was that the locum clinical psychologist was unable to participate.
3. Some participants who met eligibility criteria chose not to participate[[40]](#footnote-40). The fact that the sample inevitably comprised only those who wanted to discuss their experiences, or saw no obstacles to participation, may have biased the findings. For the purposes of this study, the understandings and perspectives of the service’s social worker, teacher and dietician may have enriched the analysis. In particular, their views may have expanded upon previous suggestions that those perceived by the young person to be less closely integrated to the team can be the professionals with whom the young people first ‘test out’ reactions to progress (Lee *et al.,* 2013; van der Walt & Baron, 2006). In hindsight the research design may have benefitted from initially presenting the research to the service, as this may have increased participation.

A second consideration concerns the timing and location of the interviews. All participants, except one, were interviewed within service space (either clinical rooms or their office). Offering flexibility in interview location was intended to ease participation. However, familiar surroundings, close proximity to clinical activity and external pressures constraining time for the interview possibly constrained what participants felt able to say. Furthermore, several interviews were disturbed by interruptions, which disrupted the flow of the conversation. Participants were offered a choice of interview location, and it is perhaps unsurprising that the vast majority chose to be interviewed at the service. However, it is contended that the quality of the data may have been enhanced if interviews were held in a more neutral space, away from competing demands.

Individual interviews afforded the opportunity for a greater depth of information to be amassed. However, they have been critiqued on the basis that they do not capture naturally occurring interaction (Potter & Hepburn, 2005). Analysing naturally occurring speech in everyday interactions, such as during ward rounds, or an MDT meeting, may therefore have increased confidence in findings. Alternatively, a focus group format may have provided an insight into the operation of social processes in the articulation of knowledge.

Finally, confidence in the conclusions drawn from this study is potentially hindered by the lack of inter-rater reliability. A list of all quotes for some individual codes and each subtheme was carefully discussed in supervision as part of the analytic process. However, I was persuaded by Braun and Clarke’s (2013) assertion that the use of systematic inter-rater reliability is underpinned by the realist assumption that there is an accurate reality in the data and that coding can capture it. From a social constructionist perspective coding is understood to be an active and reflexive process that inevitably bears the mark of the researcher. In a similar way, some researchers have proposed that the integrity of data is ensured through validation from participants (Williams & Morrow, 2009). Whilst I intend to feedback the results to participants, I consider that potential differences will provide additional data rather than invalidate the analysis presented. I have endeavoured to uphold principles of reliability by ensuring the process of analysis is as transparent, recursive and thorough as possible.

**4.6. Reflexive review**

I reflected on the ethical issues raised in relation to participant concerns about what they said being reported to, or known about by, others. This was highlighted by the way in which being recorded potentially constrained what some participants seemed to feel able to say. Some participants expanded on previous comments once the audio-recorder had been switched off. This led me to reflect on the ways in the research was influenced by wider contexts, such as uncertainty regarding the potential impact of the extracts on the existing literature.

The data cannot be extracted from inevitable attempts from participants to favourably represent themselves and the service. I also queried whether the particular professional affiliation of the interviewee may be a factor, for example those affiliated with the nursing team (who comprised the largest subgroup) may have felt less constrained than others who were the sole representative of their profession, and as such potentially more identifiable.

Throughout the analytical process I was extremely wary about the possibility of participants being identifiable, particularly by others within the service. I reflected that my own anxiety to get the analysis ‘right’ and wariness regarding the possible impact of the analysis (particularly in the context of divergent opinions) may partly have been amplified through a process of transference. Regular supervision and the use of a reflective journal were integral to considering how my perception of this may have impacted the analysis. I have endeavoured to provide a balance in quotations and be as transparent about the analytical process as possible in the hope that this will enable extracts to be read and understood in context.

During the interviews and analytical process I was surprised at the way in which the approach taken by the service appeared to be contrasted to other approaches. I reflected on the fact that my interpretation of the literature had formed my belief that the approach described by participants was not substantially different to that previously described (Lask, 2004; Nunn *et al.,* 1998). I tried to remain curious about this, and reflected on the fact that perhaps the way in which this research was designed and implemented did not allow this difference to be sufficiently captured.

I reflected on my questioning style throughout the interview process. I noticed my curiosity to ask more about certain young people who had been vividly described, and my tendency to pursue questions relating to the diagnostic process and the utility of labelling. As such, I paid particular attention to making explicit my own assumptions through the process of supervision and a reflective journal, so as to minimise the impact of these on the interview process. I also reflected on my phrasing of certain questions in the first few interviews. For example, I noticed how I had occasionally introduced an area of questioning by asking a dichotomous question (such as whether something was helpful or unhelpful) rather than asking a broader open-ended question. Opening up the style of questioning facilitated different avenues of response.

**4.6. Conclusions**

The study offered an opportunity to explore the understandings and perspectives of a treating team working with children and young people given a label of PRS. Qualitative analysis of interview data yielded three key themes: Making sense, Uncertainty and Our way.

Findings from this study expand previous literature concerning clinical management (Nunn *et al.,* 1998; Lask, 1994). The many and varied challenges and dilemmas faced by professionals working with this presentation have been elucidated. The study aimed to contribute to practice-based evidence, inform service development and approaches to staff support and provide the basis for further clinically relevant research. It is suggested that these aims have been met.

It is hoped that through dissemination these findings will be made meaningful to professionals working in this area, to those who are given a label of PRS and their families and to other professionals working in areas where diagnosis is contested.

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World Health Organization. (2010). *International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10).* Geneva: WHO.

Wright, B. & Beverley, D. (2012). Pervasive Refusal Syndrome. *Clinical Child Psychology and Psychiatry, 17*(2), 221-228.

Yardley, L. (2000). Dilemmas in qualitative health research. *Psychology and Health, 15,* 215-228.

Zubin, J. & Spring, B. (1977). Vulnerability - a new view of schizophrenia. *Journal of Abnormal Psychology, 86*(2), 103-124.

**APPENDICES**

**Appendix A: Literature search strategy**

EBSCO Electronic databases (Academic Search Complete, CINAHL, psycINFO, and psycARTICLES) were searched in August 2014. Given my awareness from initial reading of the limited literature in this area, I narrowed my initial search to papers and books mentioning ‘pervasive refusal syndrome’. No limits or parameters were used in order to give a comprehensive overview of the literature. This search returned 40 unique results.

Papers were selected by screening the abstracts. They were included if they reported either case studies or discussion relating to pervasive refusal syndrome or comment on published papers (25 papers). Papers were also included if they referred to PRS in the context of another presentation; eating disorder (4 papers), conversion disorder (1 paper) and catatonia (1 paper). Case conference abstracts (1 paper), papers not published in English (1 paper), book reviews (2 papers) and papers of no perceived relevance to PRS (5 papers) were excluded. Four instances of PRS were found in book chapters. These were included in the literature review. Given the variety in terminology, I also searched for ‘pervasive arousal-withdrawal syndrome’ and ‘depressive devitalisation’. This did not add any unique results.

I repeated the search using the electronic database SCOPUS, which added three further papers relating specifically to PRS and two relating to PRS in an eating disorder context. Lastly, a hand search from key papers found nine further references. Of these, two were related to depressive devitalisation and one was a comment on a previous paper. The others were excluded (3 papers not published in the English language and 3 conference paper abstracts).

Newspaper articles were searched for using the electronic database Nexis. A search for ‘pervasive refusal syndrome’ returned 35 results. The search terms ‘depressive devitalisation’ and ‘pervasive arousal-withdrawal syndrome’ did not return any matches.

Given the significant gaps identified in the literature, and the nature of the research, further search terms related to previously suggested differential diagnoses were added. This search was limited to review papers in the first instance in order to establish consensus on management guidelines for children and adolescents. These were initially searched for under the parameter of children and adolescents, however given the paucity of research in some fields the parameters were opened up. The search was also focused on professional’s perspectives and understandings of working with the differential diagnoses.

**Appendix B: Suggested diagnostic criteria for PRS**

Thompson and Nunn (1997) suggested the following criteria on the basis of a case note review.

1. Clear food refusal and weight loss.
2. Social withdrawal and school refusal.
3. Partial or complete refusal in two or more of the following domains: mobilization; speech; attention to personal care.
4. Active and angry resistance to acts of help and encouragement.
5. No organic condition to account for the severity of the degree of symptoms.
6. No other psychiatric disorder that could better account for the symptoms.

Jaspers *et al.,* (2009) suggested that the criteria of food refusal or weight loss should be subsumed within other domains of refusal. The authors also suggested diagnostic criteria should reflect the fact that the endangered state requires hospitalization.

Jans *et al.,* (2011) suggested modifying diagnostic criteria for PRS to reflect the fact that active and angry features are present only some of the time.

Riaz & Nawab (2014) suggested that the criterion of social withdrawal and school refusal be amended to ‘social withdrawal and refusal to engage in educational, supported, domestic or vocational activities’.

**Appendix C: Published journal articles describing individual cases[[41]](#footnote-41)**

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Authors** | **Country** | **No. of cases** | **Ages** | **Sex** | **Country of origin** | **Context of admission[[42]](#footnote-42)** |
| Lask *et al.,* (1991) | UK | 4 | 9-14 | Female | UK | Psychiatric |
| Graham & Foreman (1995) | UK | 1 | 8 | Female | UK | Psychiatric |
| Thompson & Nunn (1997) | Australia | 7 | 10-15 | 6 Female  1 Male | Australia | 6 Psychiatric  1 Outpatient |
| McGowan & Green (1998) | UK | 1 | 11 | Female | UK | Psychiatric |
| Taylor *et al.,* (2000) | Australia | 1 | 4 | Male | Australia  (Chinese parents) | Paediatric |
| Anonymous (2001)[[43]](#footnote-43) | UK | 1 | 8 | Male | UK | Psychiatric |
| Lask (2004) | UK | 1 | 14 | Female | UK | Psychiatric |
| Magagna (2004) | UK | 1 | 17 | Female | China | Psychiatric |
| Edwards & Done (2004) | UK | 1 | 11 | Female | Not stated | Paediatric |
| Bodegård (2005a) | Sweden | 5 | 7-10 | 3 Female  2 Male | Former Soviet Republic states | Psychiatric |
| Bodegård (2005b)[[44]](#footnote-44) | Sweden | 16 | 7-12 | 6 Female  10 Male | Former Soviet Republic states | Psychiatric |
| Van der Walt & Baron (2006) | Australia | 1 | 11 | Female | Not stated | Psychiatric |
| Jaspers *et al.,* (2009) | Netherlands | 1 | 11 | Female | Not stated | Psychiatric |
| Guirguis *et al.,* (2011) | UK | 4 | 12 | 1 Male  3 Female | Not stated | Psychiatric |
| Jans *et al.,* (2011) | Germany | 3 | 14-16 | 2 Female  1 Male | 2 German  1 Ukraine | Psychiatric |
| Wright & Beverley (2012) | UK | 1 | 14 | Male | Not stated | Home treatment[[45]](#footnote-45) |
| Forslund & Johansson (2013) | Sweden | 5 | 7-17 | 3 Female  2 Male | 5 Azerbaijan | Psychiatric |
| Lee *et al.,* (2013) | UK | 1 | 8 | Female | Not stated | Paediatric |
| McNicholas *et al.,* (2013) | Ireland | 1 | 11 | Female | Not stated | Paediatric |
| Riaz & Nawab (2014) | UK | 1 | 50s | Female | Not stated | Psychiatric Learning Disability Service. |

**Appendix D: Letter of invitation**

**SERVICE LOGO**

Dear [clinician name]

You are invited to participate in a research study entitled ‘Pervasive Refusal Syndrome (PRS): understandings and perspectives of treating professionals working with children and young people’.

Research into the clinical management of PRS is a particularly sparse area of the literature. It is hoped that focusing on professionals’ experiences will help to inform practice-based evidence and service development whilst also providing a springboard for further research.

In order to participate it is required that you are currently, or have previously been, a member of the [service]. It is also a requirement that you have had experience of working with two or more children or adolescents given a clinical diagnosis of PRS.

If you may be interested in participating, please refer to the information sheet enclosed for more information. Please register your interest by emailing **pervasiverefusalstudy@gmail.com** before **[date].** It would be very helpful if you could give an indication of your level of experience with this population and your preferred means of contact in this email.

Thank you in anticipation. I look forward to hearing from you,

Beth Coombs

Trainee Clinical Psychologist

University of East London

**Supervised by:**

[Name] Dr Jenny Jim

Clinical Psychologist Clinical Psychologist

[Service] University of East London



**Appendix E: Information Sheet**

**SERVICE LOGO**

**Principal Investigator**

Beth Coombs

Trainee Clinical Psychologist

**Project Title**

Pervasive Refusal Syndrome (PRS): understandings and perspectives of treating professionals working with children and young people.

**Project Description**

The aim of this study is to explore multidisciplinary experiences of the clinical management of children and adolescents given a clinical diagnosis of PRS. Research into the clinical management of this population is a particularly sparse area of the literature. It is hoped that the results will inform practice-based evidence and service development whilst also providing a springboard for further research.

This study is being conducted as part of a Clinical Psychology Doctorate at the University of East London.

**Procedure**

You will be contacted in order to arrange a time and date for the interview. You are welcome to choose a location to suit you, however it must be somewhere that confidentiality can be assured. It is anticipated that the interview, which will be audio-recorded, will take no longer than one hour. You will be asked to sign a consent form and complete a short information form. This will ask questions about (i) profession, (ii) length of time working on the MCU, (iii) number of children and adolescents with a clinical diagnosis of PRS treated, (iv) number of units worked at where your role involved treating children and adolescents who were given a clinical diagnosis of PRS.

The interview will be guided by a semi-structured questionnaire. Questions will be focussed on your experiences of working with this population including your perspectives on management, and challenges or dilemmas you may have encountered.

You will be able to request a break at any point should you so wish. You will also be able to withdraw your consent to participate at any point without being obliged to give a reason. It is possible that reflecting upon your experiences may bring up some uncomfortable feelings. You will be offered the opportunity to debrief following the interview.

**Confidentiality and Anonymity**

Confidentiality will be upheld at all times. The interviews will be transcribed and anonymized. These will be saved on a password-protected USB memory stick for 10 years. Whilst names and places will be anonymized, it is possible that quotations used in thesis extracts and other forms of dissemination may be identifiable by others. This will be minimized by careful selection of quotations.

**Benefits to participation**

Participation in this study will provide an opportunity to reflect upon practice and share thoughts, experiences and skills. It will also be a means of contributing to the knowledge base that informs ways of working with this population.

**How to participate**

In order to participate it is required that you are currently, or have previously been, a member of the [service]. Experience of working with two or more children or adolescents with a clinical diagnosis of PRS is also required.

If you would like to participate, please register your interest by emailing **pervasiverefusalstudy@gmail.com** by **[date]**. It would be very helpful if you could include an indication of your level of experience with this population and your preferred means of contact in this email. Please also direct any queries to this email address. The principal researcher will contact all those who respond.

Please retain this information for future reference.

Thank you in anticipation.

Beth Coombs

Trainee Clinical Psychologist

University of East London

**Appendix F: Interview schedule**

**SERVICE LOGO**

**Interview Schedule**

**Introduction**

* Ask participant to read information sheet, sign consent form and complete participant data sheet.
* Ask participant to choose a pseudonym.
* Share hope that interview can be a conversation guided by the schedule but also their own thoughts and experiences.
* Inform participant about the digital recorders and reiterate steps taken to ensure confidentiality and anonymity.
* Assure that ok to ask for clarification or request a break at any point.
* Ask whether they were able to recall previous experience prior to the interview.as requested. If not then offer more space in the warm up to ensure experiences are sufficiently recalled.

**Warm up**

* Can you tell me a little bit about your experiences of treating children and young people given the label of PRS?

*Prompts:* What first comes to mind? Are there any experiences that particularly stand out?

**Main interview**

* How do you make sense of the needs of these young people?

*Prompts*: Is there anything you feel is unique to the presentation? Is there anything that you draw on to inform your understanding?

* Which aspects of the treatment do you feel are the most helpful?

*Prompts:* Who finds what most helpful? What do you think the intervention should comprise?

* Are there any aspects of treatment you consider to be less helpful?

*Prompts:* Which frameworks or models inform management? There is a lack of literature in this area – does this have an impact?

* What do you think helps these young people make steps towards recovery?

*Prompts:* What do you consider recovery to be? How successful do you think the service is with this population?

* Can you tell me about any challenges or dilemmas you have encountered specific to working with this presentation?

*Prompts:* Challenges specific to young people, family or staff team

* What are your thoughts about terming the presentation ‘PRS’?

*Prompts:* How do you feel different people within the system perceive the label? Is there an effect on engagement?

* Has working with these young people had a personal impact on you and if so in what way?
* Why did you decide to take part in this study?
* What has it been like talking about your experiences today?

**Closing**

* Ask if participant feels anything has been missed or has any questions.
* Offer opportunity to debrief.
* Explain how and when research findings will be made available,
* Thank participant for agreeing to take part and for their time.

**Additional prompts**

Can you tell me more about X? What is your experience of X? What do you think about X? Can you give me an example?

**Appendix G: Consent form**



**SERVICE LOGO**

**Consent to participate in a research study**

Pervasive Refusal Syndrome (PRS): understandings and perspectives of treating professionals working with children and young people

I have the read the information sheet relating to the above study and have been given a copy to keep. The nature and purposes of the research have been explained to me. I have had the opportunity to discuss the procedure and ask questions about this information. I understand what is being proposed and the procedures in which I will be involved have been explained to me. I understand that I am not obliged to take part in this study.

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

I freely and fully consent to participate in the study. Having given this consent I understand that I have the right to withdraw from the study at any time without disadvantage to myself and without being obliged to give any reason. I also understand that should I withdraw, the researcher reserves the right to use my anonymous data in the write-up of the study and any future analysis.

Participant Name (BLOCK CAPITALS) ……………………………………..……...…

Participant Signature…………………………………………………………………...

Researcher Name (BLOCK CAPITALS)………………………………………….….

Researcher Signature……………………………………………………………………

Date: ……………………..…….

**Appendix H: Participant information sheet**

**SERVICE LOGO**

**Participant Data Sheet**

Pseudonym:

Profession:

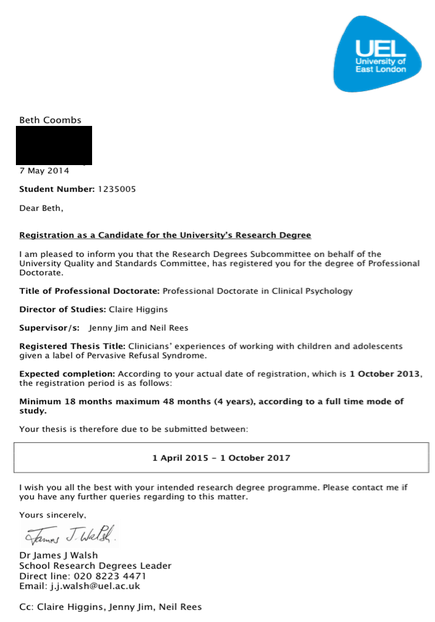
Length of time on MCU:

Number of children and adolescents with a label of PRS treated:

Number of inpatient services worked at where your role involved treating children and adolescents given the clinical diagnosis of PRS:

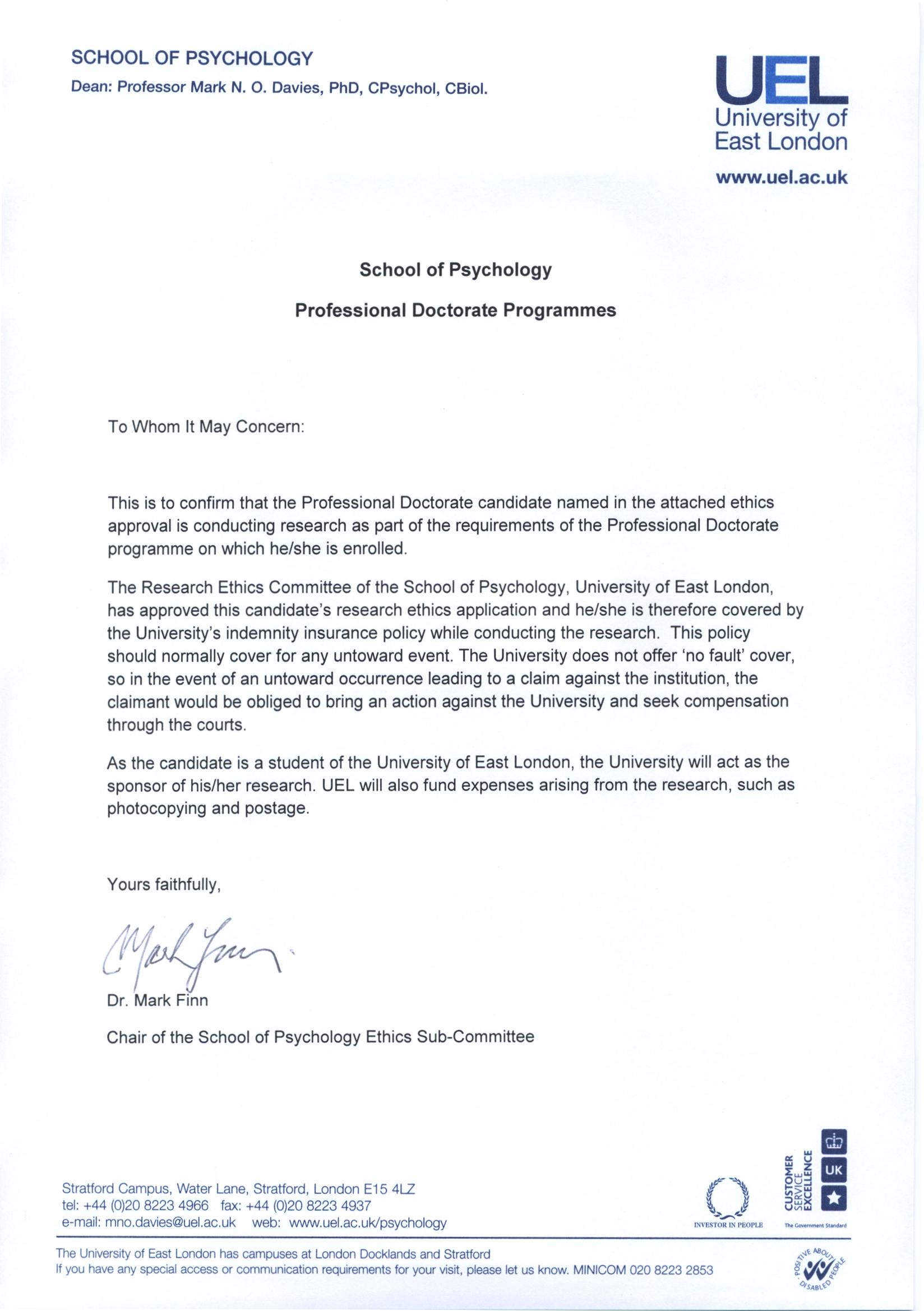
Thank you for your participation.

**Appendix I: UEL Registration**

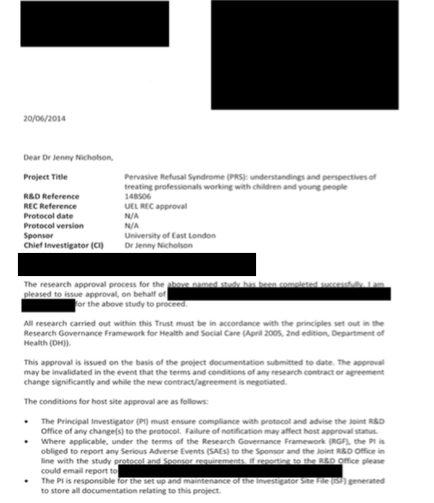
****

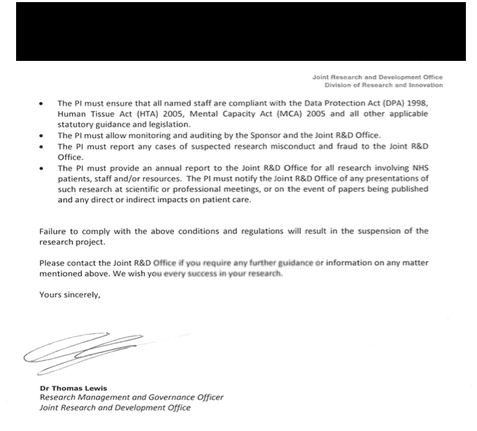
****

**Appendix J: UEL ethics Approval**



**Appendix K: Research and development approval**

****

****

**Appendix L: Clinical Research Adoptions Committee (CRAC) approval**

****

**Appendix M: Transcription conventions**

(.) Pause.

(5) Timed pause.

[inaudible] inaudible section of recording.

[laughter] laughter during the interview.

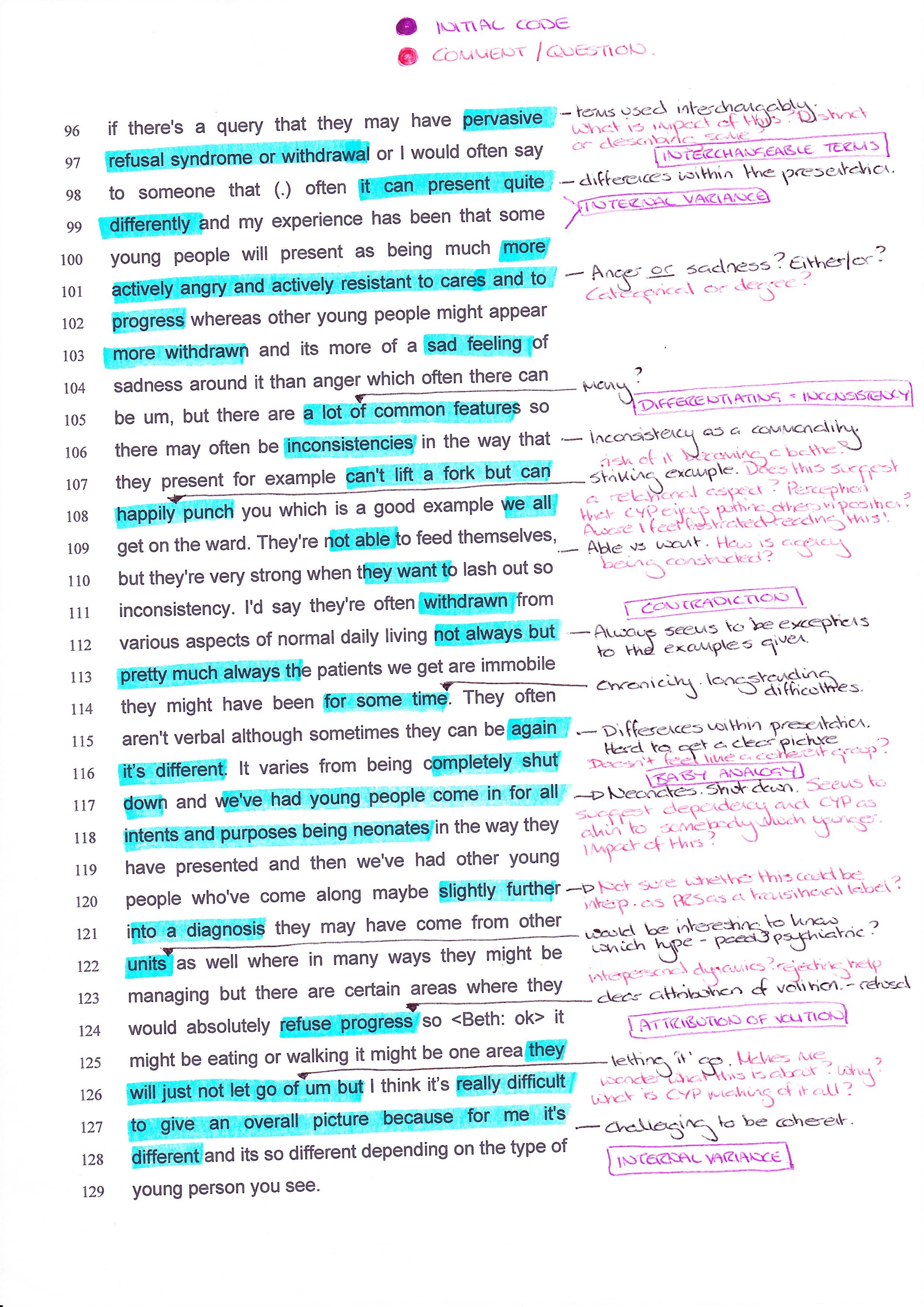
<speech> denotes brief interjection by other speaker

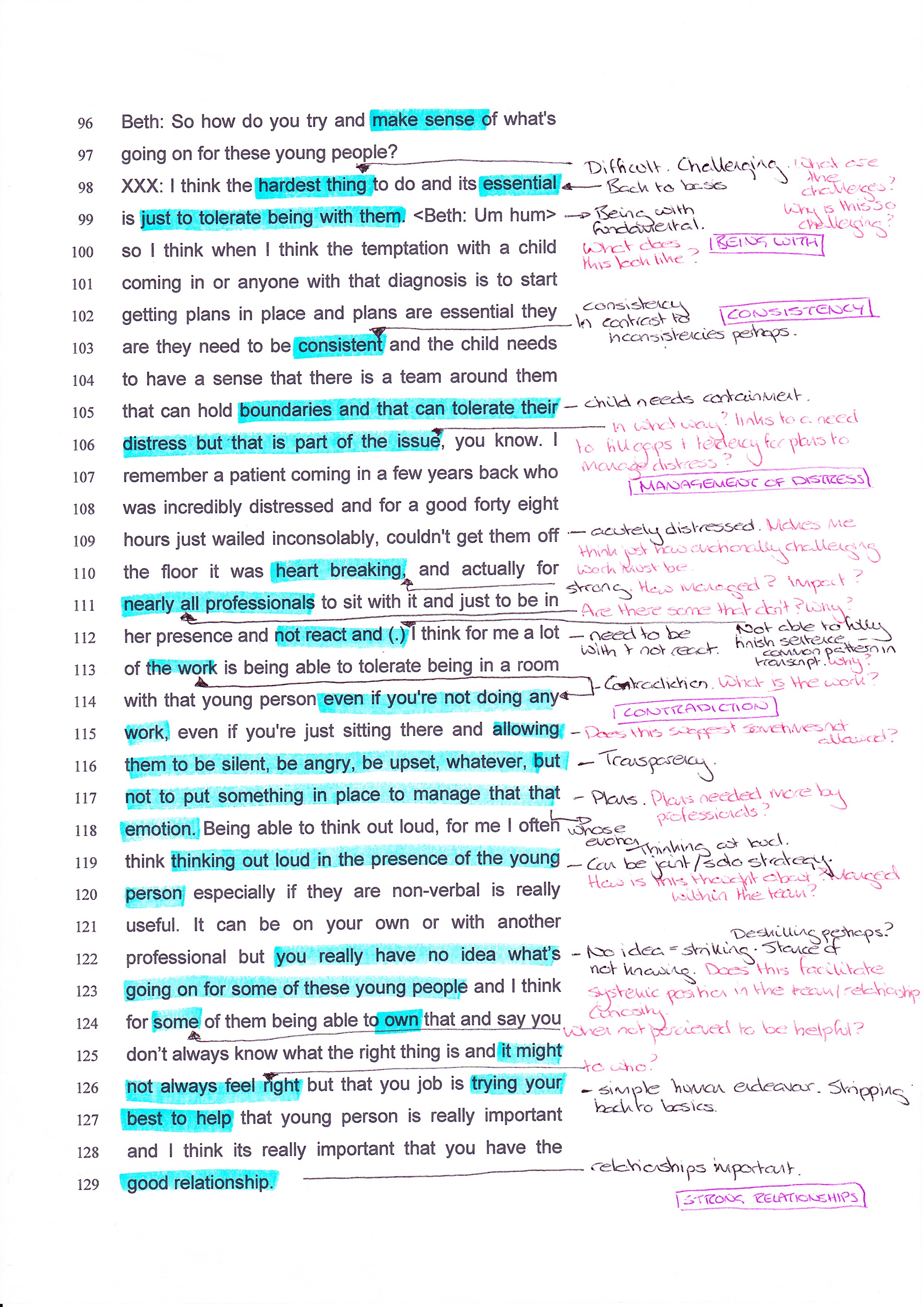
Sounds such as “mm” and “er” have been transcribed phonetically, as have colloquialisms, abbreviations, stutters and half-said words.

Wide margins and numbered lines aided the analytical process.

Adapted from Malson (1998)

**Appendix N: Example excerpt of coded transcript**



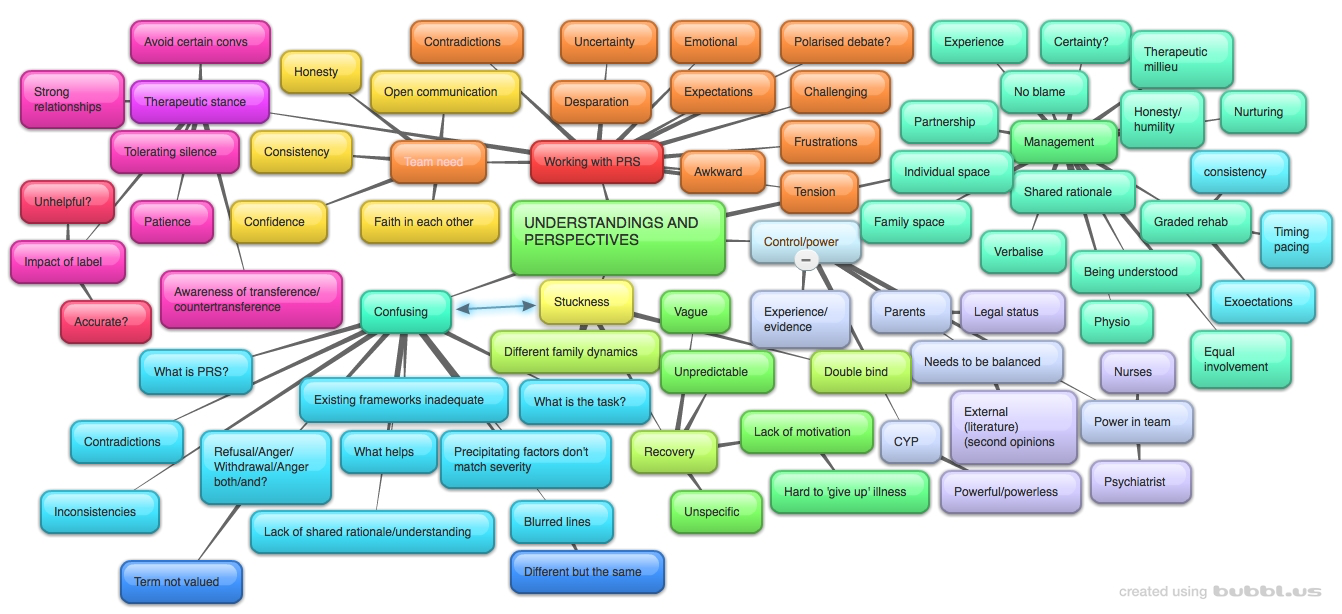


**Appendix O: Example of extracts across the data set coded**

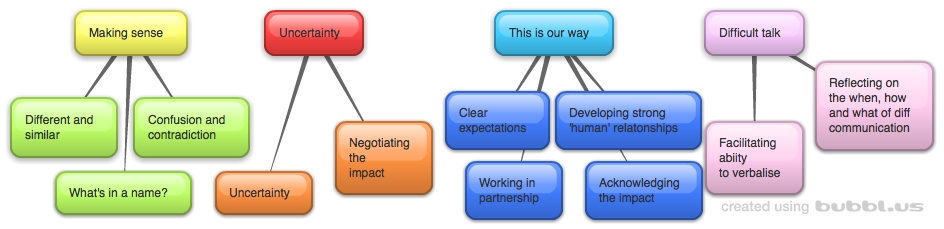
*Code: Graded expectations*

|  |  |  |
| --- | --- | --- |
| **Participant** | **Lines** | **Participant** |
| Andrew | 298-301 | *I think it’s counterproductive to force them, and I think you can cause problems by pushing them too hard, so err on the side of not pushing them too hard is my view.* |
| Andrew | 304-307 | *you can, not all the way through, but after the first few months, you can gently encourage them to start doing more and you can you just do it in a graded way* |
| Bella | 200-206 | *The expectation would be that a young person with this condition would participate in whichever parts they could when they're admitted. That might be two minutes in a group, or in a mealtime, but the expectation is that that then gets built up to the point where they are fully integrated into the community* |
| Katie | 521-524 | *I think its better to build up than to have high expectations from the beginning and then start going down, because I think that’s a very very slippery slope* |
| Emily | 544-547 | *should you go down the path of making them do something despite their distress, or whether we should actually back off and you know try and focus on something they can do?* |
| Max | 355-360 | *I think what works well is a multi-disciplinary approach, where you have a graded increase in what someone’s able to do. The tricky thing is what pace do you go at, and I think that’s the hardest thing and that’s the thing people tend to get wrong* |
| Sarah | 237-242 | *I think there needs to be an element of challenging but not pushing so you have to find some sort of balance between pushing but not pushing too fast, so like gentle encouragement, I would have said that’s probably the best word* |
| SR | 191-195 | *so say for eating we would start by getting our hands helping our hands to do it and slowly over time the would take more and more responsibility until they’re doing it by themselves and that works, for us that works really well in the children I’ve seen.* |
| Sunshine | 69-  75 | *I would start to set a bit of an expectation like…when I come to see you next it would be really great if maybe you could squeeze my hand or if you could let me know you’re listening in some way* |

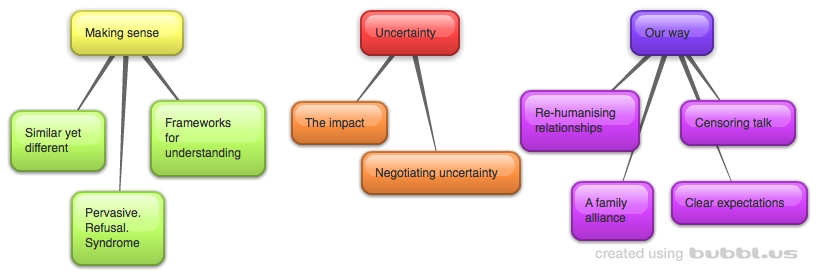
**Appendix P: Theme development**



*Initial themes*

**

*Final themes*



**Appendix Q: Reflective journal extract**

*Example excerpt of entry following interview[[46]](#footnote-46)*

Feeling pretty exhausted following the interview, although time seemed to fly by. There’s so much to try and process all at once. I’m anxious I may be unintentionally closing down important avenues by asking certain questions and not others. I keep asking multiple questions in one as though that’s an effective strategy! I feel like I’m hearing a lot about the family through these interviews but the views of the child or young person aren’t coming across strongly at all. I wonder what they’d make of my questioning?

My initial impressions were that X seemed quite reserved. For example they were asking a lot of questions about confidentiality before I switched the recorder on. I also wondered if I had got them at a particularly stressful point in the day as they stressed their time commitments several times. I noticed how I felt under increased pressure to try and keep the questions moving and not dwell for too long on any one question. At the same time though once we got going they just kept talking and seemed to be very open and I was surprised at the level of self-reflection. I wondered if they were presenting a very positive picture of the service but as I’ve wondered this from many participants perhaps the service is as good as it is being made out to be.

I’m noticing how emotionally exhausted I can feel following the interviews – its clear that participants are struggling with some really big emotions and big questions but also there are so many contradictions and different opinions – where to start?! This interview also raised my awareness of aspects I hadn’t considered previously like the child or young person potentially becoming traumatised by medical tests. I don’t think I’ve thought about the care pathway closely enough.

I felt a little bit startled by a comment X made once the recording was switched off. I wondered what this was about and what might be meant by it. It’s becoming clear that being recorded changes to some extent what people able to say. Wondering if there is a bigger political context to this, which makes me feel a bit uneasy with regards to the analytic process. X also mentioned they were a bit nervous before hand so I wonder if I might have done something to help allay nerves.

*Entry following preliminary stages of analysis*

Feeling completely overwhelmed – there is so much to consider! I feel like I am missing so much out and that there is pressure to get it ‘right’. I’m really noticing how I feel really compelled to use lots and lots of quotes, including many from the same few participants. This has made me really go back to other accounts and try and engage with them again to ensure I represent a balance and the full spectrum of participant’s understandings and perspectives. On some points it seems like hardly anything is agreed on but I do feel there are some commonalities at the same time.

I’m really noticing how my own preconceptions could easily exert an impact. I really found some of the contents of the interviews emotionally challenging – am I expecting to find this in the data and therefore attending to certain participants accounts more than others? Really hard to balance, I want each participant to have a voice and not privilege one more than others. Also wondering how much I have been affected by the order of coding as its inevitably slightly harder to maintain curiosity as much as before. I feel like I have hardly any space in the write up to do justice to all of the findings identified.

Supervision has been really helpful to reflect about my fear of stirring something up in the team – this is a concern that has been on my mind throughout the research process. For example, I really don’t want participants to feel they have been misrepresented or to unwittingly create factions through presenting quotes highlighting differences in the team. Jenny and I spoke about the process of accurately reflecting my own interpretations of the data and giving sufficient evidence for claims made. Thinking about implications and possible recommendations has also felt useful.

1. The service had significant experience of treating young people without an identifiable organic aetiology (Feibelman, Lask & Williams, 1990). [↑](#footnote-ref-1)
2. PTSD diagnostic criteria are continually revised (Summerfield, 1999). In DSM III (1980) an established catastrophic stressor outside the range of usual human experience was a diagnostic requirement. Lask *et al.,* (1991) state it was difficult to establish all criteria were fulfilled. [↑](#footnote-ref-2)
3. The Cleveland Scandal refers to the mass diagnosis of 121 cases of child sexual abuse in Cleveland, UK, by two paediatricians using controversial diagnostic practices. At least 70% of the diagnoses were incorrect. [↑](#footnote-ref-3)
4. One child was treated on an outpatient basis at a local service with a modified treatment programme. [↑](#footnote-ref-4)
5. This was likely separated from the other refusal domains due to the authors’ experiences with children and young people with a diagnosis of an eating disorder. [↑](#footnote-ref-5)
6. Nunn (1996) asserts that the concept of helplessness is better understood as the loss of hope. [↑](#footnote-ref-6)
7. The families overwhelmingly originated from minority backgrounds within former Soviet states. These countries were not recognized as being prima facie dangerous by the Swedish state. [↑](#footnote-ref-7)
8. Cases were termed PRS in international media, and ‘apathetic’ in Swedish media. [↑](#footnote-ref-8)
9. Serious illness is a humanitarian ground for a Swedish residency permit. [↑](#footnote-ref-9)
10. Aronsson, Wibery, Sandstedt and Hjern (2009) implicated the stress of the visa application process. Hacking (2010) hypothesised a process of ‘imitation and internalization’; the behaviour of those who managed to access resources is imitated before becoming internalized over time. [↑](#footnote-ref-10)
11. The syndrome will be identified as a subtype of depression with the specification ‘withdrawal’, and the additional code of ‘a problem concerning refugee state and asylum application’. [↑](#footnote-ref-11)
12. A majority of papers offer a very brief overview of the intervention drawing on principles cited in Lask (1994) and Nunn *et al.,* (1998). [↑](#footnote-ref-12)
13. Recovery criteria are not stated. [↑](#footnote-ref-13)
14. Clinicians assessed functioning according to the Global Assessment of Functioning (GAF) scale. The questionnaire measures comprised a measure of depression (Montgomery–Asberg Depression Rating Scale) and PTSD (PTSD Checklist – Civilian version). [↑](#footnote-ref-14)
15. DSM-IV diagnoses are used by both authors [↑](#footnote-ref-15)
16. It is interesting that there is no reference to PRS as a variant of PTSD given its history and the continual widening of the PTSD diagnosis. Jaspers *et al.,* (2009) identify trauma as an aetiological factor in 79% of reported cases. [↑](#footnote-ref-16)
17. The statistic reported by Jaspers *et al.,* (2009) is inaccurate. Five children and young people, rather than one, were initially given the diagnosis of CFS in the Thompson and Nunn (1997) case note review. [↑](#footnote-ref-17)
18. This was declined by her parents. [↑](#footnote-ref-18)
19. Abreaction involves reliving a ‘repressed’ experience aided by hypnosis or medication. Narcosis involves examination and suggestion whilst mildly sedated with intravenous amylobarbitone (colloquially known as truth serum). [↑](#footnote-ref-19)
20. A recent move away from implicating volition is acknowledged (PAWS: Nunn *et al.,* 2014). However, given that this is a recent shift it is therefore hypothesised that its impact will be limited. [↑](#footnote-ref-20)
21. This calculation is based on the assumption of an average duration of a stay in a Tier 4 inpatient CAMHS service (389 days; Jaspers *et al*., 2009) and the average nightly cost of Tier 4 inpatient admission (£652; Department of Health, 2012). [↑](#footnote-ref-21)
22. Earlier data was not available. [↑](#footnote-ref-22)
23. Ethnicity was not reported. [↑](#footnote-ref-23)
24. No single common predisposing factor was identified. A full range of predisposing and precipitating factors were not identified. [↑](#footnote-ref-24)
25. Names have been changed to protect anonymity. Participants were given the opportunity to choose their own pseudonym. [↑](#footnote-ref-25)
26. Max has worked with five other children and young people with a label of PRS in paediatric services. [↑](#footnote-ref-26)
27. Sunshine left the service in 2011. [↑](#footnote-ref-27)
28. Bella left the service in 2014 immediately before data collection. [↑](#footnote-ref-28)
29. ‘...’ denotes words cut to ensure that excerpts are concise and clear. Care has been taken not to alter participants’ intended meaning. [↑](#footnote-ref-29)
30. Parentheses have been used to provide clarity. [↑](#footnote-ref-30)
31. Names changed by William. [↑](#footnote-ref-31)
32. Participants were requested to reflect on their experiences prior to interview. [↑](#footnote-ref-32)
33. It is interesting that participants did not suggest PAWS to be a more accurate term. [↑](#footnote-ref-33)
34. Participants described PRS as encompassing a wide range of behaviours, including active refusal, anger and withdrawal. A couple of participants suggested that a difference between refusal and withdrawal might be reflected in new terminology (PRS vs PWS), however this was not a consistent finding. [↑](#footnote-ref-34)
35. I was aware of literature suggesting that children and young people may move *between* an angry and passive presentation (Jans *et al.,* 2011). [↑](#footnote-ref-35)
36. I would like to acknowledge that two participants suggested these techniques as possible recommendations for future clinical practice. [↑](#footnote-ref-36)
37. This is hypothesised to be particularly relevant given the average age of admission (12 years old: McNicholas & Nicholson, 2014). [↑](#footnote-ref-37)
38. Gender, Race, Religion, Age, Ability, Class, Culture, Ethnicity, Education, Sexuality and Spirituality [↑](#footnote-ref-38)
39. In a Q sort participants are asked to sort items relative to one another along a dimension such as ‘agree’ or ‘disagree’. [↑](#footnote-ref-39)
40. Two participants expressed interest in taking part but were either unable to commit to a time or did not respond to further correspondence. Several participants also mentioned members of staff who had expressed an interest in the research but had not contacted me. [↑](#footnote-ref-40)
41. This table is limited to papers that specifically describe individuals where a label of PRS has been ascribed. It therefore does not include papers written by Danish and American authors who have commented on the cases. It also does not include authors who have contributed conference papers or papers not written in the English language, such as those from Turkey, France and Denmark. Lask (1996) states that he has consulted on over 20 cases of PRS but insufficient detail is described to warrant inclusion. [↑](#footnote-ref-41)
42. Most paediatric admissions involved extensive psychiatric liaison. [↑](#footnote-ref-42)
43. This paper is written from a parent’s perspective. [↑](#footnote-ref-43)
44. This paper includes the 5 cases discussed in Bodegard (2004a) [↑](#footnote-ref-44)
45. This admission involved extensive psychiatric support five days a week. [↑](#footnote-ref-45)
46. No identifying information given to uphold confidentiality. [↑](#footnote-ref-46)