DOCTOR OF CLINICAL PSYCHOLOGY (DCLINPSY)

Doctorate in Clinical Psychology: Main Research Portfolio

1) The lived experience of children and adolescents with a rare disease. A meta-study; 2) he experience of service users/people with personal experience being interview panel members for DClinPsych selection interview; 3) The transition from paediatric to adult healthcare with Oesophageal atresia/trachea-oesophageal fistula (OA/TOF). A qualitative study exploring the experiences of young people, adults and parents.

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Doctorate in Clinical Psychology

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Impact of COVID-19 on my Research

Detailed in the following brief commentary is the impact of the COVID-19 pandemic on my doctoral research, and how I responded to these challenges.

I had already planned for my main research project to be conducted online, so when the pandemic started it felt like there was not as much impact on the study as there could have been. Though on reflection, I wonder whether the possible stress that was caused by the pandemic may have impacted the number of potential participants, i.e., completing an online survey may not have been high on people's list of priorities from March 2020 onwards.

The pandemic had an impact on the face-to-face meetings that I had been having with my research supervisors, though we were able to adapt fairly swiftly and commence online meetings. Apart from the occasional technical hitch, these meetings continued to be helpful and, being virtual meant that it cut down on travel time. Other meetings with co-researchers were also conducted online. For some of these meetings we struggled initially with connecting online and had to resort to speaking on the phone and emailing. Whilst I have been grateful that research meetings were able to continue despite the global pandemic, the lack of human contact was challenging throughout the various lockdowns. I feel that I made good use of supervisory support, but this was also more virtual contact. I am reminded, as we emerge from lockdown, just how important human face-to-face contact is for maintaining mental health.

The uncertainty throughout the pandemic has taken its toll in terms of my own stress levels and I noticed, especially during the first lockdown, that I was more emotionally labile than I would normally be, which would sometimes mean that I was not fully "present" and able to engage in research tasks such as analysis or writing. I also noticed that my concentration span, particularly early on in 2020, was very much reduced. During lockdown it was especially challenging to be doing everything in the same office, studying, research, lectures, placement. I found that it was difficult to switch between different tasks and sometimes found that I would get muddled between various tasks, often working on one project and realising that I was actually thinking about a different project and then having to start all over again. In responding to these particular challenges, I soon realised that I would need to have more frequent, short breaks throughout the working day. I also tried to make sure that I came away from my

computer screen regularly and engaged in “self-care” whenever possible (baths are a particular favourite of mine!). I have also made use of my supervisory team and tried, whenever possible, to be honest when I have been struggling.

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Service Improvement Project paper: 5027

Main Research Project: 8089

Executive summary: 997

Reflective narrative: 2362

Total: 23858

Please note: all word counts exclude abstracts, figures, tables, quotes, foot notes and references.

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Abstracts

Literature Review: The lived experience of children and adolescents with a rare disease. A meta-study. The purpose of the current meta-study was to identify and synthesise the available qualitative research evidence across rare disease conditions, to establish common themes related to childhood lived experience of rare diseases and to understand the challenges posed by rare diseases. This meta-study draws together findings from 12 qualitative studies that represent the lived experience of a large sample of children (n = 652) with rare diseases from a variety of countries. Children's lived experience of rare disease can be understood as feeling different and being treated differently; a narrative of restriction; distress and uncertainty; coping by drawing on internal and external sources of support. This meta-study highlighted that children's experience was formulated with a lack of reflexivity and drawing on a mix of child and adult experience. Theoretical reference across the studies was lacking, though frequent use of the term "psychosocial" suggested a grounding in the biopsychosocial paradigm. In addition, this meta-study found that the understanding of children's lived experience of rare disease is still in its infancy and reflects a majority western world perspective. Future research could perhaps diversify to include the experience of children in non-western countries. The clinical implications of this meta-study are that clinicians working in the area of rare diseases need to be aware that children may struggle with feelings of difference, restriction, distress and uncertainty. Children with rare diseases would also benefit from peer support from others with rare diseases.

Service Improvement Project paper: The experience of service users/people with personal experience being interview panel members for DClinPsych selection interview. The following Service Improvement Project presents a piece of work that was commissioned by the University of Bath, Clinical Psychology Doctoral (DClinPsych) programme admissions committee. This work firstly aimed to explore the experience of users of psychology services (known as People with Personal Experience, or "PPE") being part of the DClinPsych trainee admission interview process and secondly provide recommendations regarding the admissions process the following year. A focus group was conducted with 4 of the PPE representatives and transcribed verbatim. A version of Interpretative Phenomenological Analysis (IPA) adapted for focus groups was used to analyse the data. Four themes reflected the experience of the PPE as part of the interview process, these were: Challenges; A Unique Perspective; Connectedness; Finding a Voice.

The themes intersect multiple levels of experience. Results are discussed in the context of previous research in the area. Recommendations and the corresponding response from the programme admissions committee are presented.

Main Research Project: The transition from paediatric to adult healthcare with Oesophageal atresia/trachea-oesophageal fistula (OA/TOF). A qualitative study exploring the experiences of young people, adults and parents. The overall aim of this study was to explore the experience of healthcare transition from paediatric to adult healthcare for adults' born with OA/TOF and parents. A particular focus was how the different paediatric and adult services were experienced, whether parents and adults experienced a change in their roles during healthcare transition and also to identify challenges faced as part of the transition. OA/TOF is a rare chronic condition that can require lifelong medical support. There is evidence to suggest that transitioning from paediatric to adult healthcare can be problematic with OA/TOF, a finding that appears to be true in both rare and chronic conditions. The literature on healthcare transition is saturated with a common chronic illness narrative and there is a suggestion that the experience of transitioning with a rare condition is worse than transitioning with a common chronic condition. We currently don't clearly know why this might be the case. A qualitative, cross sectional, survey-based study was carried out. Unrelated parents (n=23) and adults (n=21) were recruited through a UK based OA/TOF charity. Data were collected through an online survey. Data from six open ended questions were analysed using a six-stage hybrid approach combining elements of inductive and deductive thematic analysis (Fereday & Muir-Cochrane, 2006). Five themes were constructed reflecting the experience of parents and adults transitioning from paediatric to adult healthcare: Thrown into the unknown; A cultural shift; Stepping back and stepping up; "No transition as such"; Living with uncertainty. The findings suggest that a formalised, managed healthcare transition is practically non-existent for people born with OA/TOF. We recommend a formalised healthcare transition process in OA/TOF. This should involve a period of preparation to get adolescents and parents ready for transition. It would be beneficial for those working in adult services to receive additional training regarding issues that adults born with OA/TOF may struggle with. It would also be helpful for families to have a named keyworker to support them through the process of transition.

List of Abbreviations

BABCP	British Association for Behavioural & Cognitive Psychotherapies
BPS	British Psychological Society
CASP	Critical Appraisal Skills Programme
CHD	Congenital Heart Disease
CQC	Care Quality Commission
DClinPsych	Clinical Psychology Doctorate
DOH	Department of Health
EJP	European Joint Programme on Rare Diseases
HCPC	Health and Care Professions Council
HCT	Healthcare Transition
IPA	Interpretative Phenomenological Analysis
JIA	Juvenile Idiopathic Arthritis
NICE	National Institute of Clinical Excellence
OA/TOF	Oesophageal Atresia/Trachea-Oesophageal Fistula
PPE	People with Personal Experience
TOFS	Tracheo-Oesophageal Fistula Support
UN	United Nations
VACTERL	Vertebral abnormailites; Anal atresia; Cardiac defects; Tracheal abnormalities; Esophageal atresia; Renal and Radial abnormalities; other Limb abnormalities.

Literature Review:

The lived experience of children and adolescents with a rare disease. A meta-study

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May 2021

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Abstract

The purpose of the current meta-study was to identify and synthesise the available qualitative research evidence across rare disease conditions, to establish common themes related to childhood lived experience of rare diseases and to understand the challenges posed by rare diseases. This meta-study draws together findings from 12 qualitative studies that represent the lived experience of a large sample of children (n = 652) with rare diseases from a variety of countries. Children's lived experience of rare disease can be understood as feeling different and being treated differently; a narrative of restriction; distress and uncertainty; coping by drawing on internal and external sources of support. This meta-study highlighted that children's experience was formulated with a lack of reflexivity and drawing on a mix of child and adult experience. Theoretical reference across the studies was lacking, though frequent use of the term "psychosocial" suggested a grounding in the biopsychosocial paradigm. In addition, this meta-study found that the understanding of children's lived experience of rare disease is still in its infancy and reflects a majority western world perspective. Future research could perhaps diversify to include the experience of children in non-western countries. The clinical implications of this meta-study are that clinicians working in the area of rare diseases need to be aware that children may struggle with feelings of difference, restriction, distress and uncertainty. Children with rare diseases would also benefit from peer support from others with rare diseases.

Introduction

The UK government Department of Health defines rare diseases as “life-threatening or chronically debilitating, requiring special, combined efforts to enable patients to be treated effectively” (DOH, 2021). The European Commission’s (2019) definition of a “rare disease” is a disease affecting less that 5 in 10,000 of the general population, the current number of known rare diseases stands at 8000 and 80% of these have a genetic origin (Eurodis, 2005). For consistency, within the current study the term “rare diseases” will be used throughout, however it is acknowledged that “rare diseases” include both “diseases” that are diagnosed based on symptoms, such as Addison’s disease, Achard Thiers Syndrome, Chronic Hiccups and Papillitus; and also “conditions” that people are born with, such as Cystic Fibrosis, Hemophilia A & B, Holt-Oram Syndrome and Jejunal Atresia. It is estimated that approximately 3 million people in the UK are affected by rare diseases (DOH, 2019), 75% of whom are children (Rare Disease UK, 2018).

In recent times rare diseases have been prioritised internationally as a key area of research. In 2017 the United Nations General Assembly resolution on Global Health and Foreign Policy highlighted the commitment made to empower those who are vulnerable by virtue of physical and/or mental health needs (UN, 2017), and in 2019 the UN declared their commitment to ensure universal access to integrated quality health services for the treatment of neglected and rare diseases (UN, 2019, art. 22). The European Union launched the European Joint Programme on Rare Diseases (EJP RD) in January 2019 (EJP RD – European Joint Programme on Rare Diseases, 2021). The EJP RD included in their call for proposals 2021 for research into the psychological and social impact of rare diseases. Since the UK officially left the European Union at the end of 2020 there has been a lack of clarity regarding involvement with the various EU rare disease reference networks, with some expressing concern that there will be a detrimental effect on individuals living with rare disease in the UK (TischKowitz et al. 2020). 2013 saw the launch of the UK Strategy for Rare Diseases (DOH, 2013) followed by an implementation plan in 2018 (DOH, 2018), setting out a shared vision for improving the lives of patients and families affected by rare diseases and aimed for “no one to be left behind just because they have a rare disease” (DOH, 2013. pg 3). Building on the commitments of the UK strategy and implementation plan, in 2021 the DOH published the UK Rare Diseases Framework with the broad aim to “set out a community-led vision to build upon existing UK strengths and improve areas of

weakness to deliver health outcomes and improve the lives of those living with rare diseases".

People with rare diseases face "unique, multidimensional challenges" (Eurodis, 2005) that are different to the challenges faced by those with common diseases (Bryson et al. 2019). A systematic review of lived experience studies found that adults with rare disorders face several challenges, including lack of information; a lack of available knowledge of the specific condition; social consequences; lack of appropriate quality healthcare; high cost of the few existing drugs and care; inequalities in availability of treatment and care (Von der Lippe et al., 2017). Importantly, these challenges are different from those experienced by people with common chronic conditions (Von der Lippe et al., 2017; Rare Disease UK, 2018; 2019).

Whilst the lived experience of adults with rare diseases is clear, the understanding of children's experience of living with a rare disease is less clear (Rare Diseases UK, 2018). This is surprising because rare diseases tend to be diagnosed at birth or during early childhood (Jaffe, A. et al 2010; Rare Disease UK, 2018), rare diseases also disproportionately affect children (DOH, 2021). Research into childhood experience of rare conditions does exist, however a limitation commonly reported in individual qualitative studies of rare diseases is that of small sample size (Brewer et al. 2008; Bruns and Foerster 2011; Lammens et al. 2011; Pelentsov et al. 2015; Picci et al. 2015; Tabolli et al. 2010; Somanadhan & Larkin, 2016), which in turn limits the knowledge development in this area (Hews-Girard et al. 2019). This problem is difficult to overcome since, by their nature individual rare diseases are "rare", however since 1 in 17 people will suffer from a rare disease at some point, rare diseases have been termed "individually rare, collectively common" (Onepoll, 2016). Therefore, understanding the lived experience of children represents a gap in the rare disease literature which the current review aims to address.

To understand children's lived experience of rare diseases a methodology is required to combine and synthesise qualitative research findings. Meta-synthesis is an appropriate methodology as it allows for both depth and breadth of examination of meanings, experiences and perspectives (Lachal et al. 2017). Meta-synthesis involves "bringing together and breaking down of findings, examining them, discovering the essential features, and, in some way, combining phenomena into a transformed whole"

(Schreiber et al. 1997, pg. 314). Furthermore, findings from a meta-synthesis can ultimately help to shape health care policy (Finfgeld, 2003). A variety of meta-synthesis approaches exist (Barnett-Page & Thomas, 2009) and were considered, meta-ethnography (Noblit and Hare, 1988) in particular, is an approach to bring together the separate parts of qualitative studies to form a "whole" that is "greater than the sum of its parts" (Barnett-Page & Thomas, 2009. Pg. 2). However, the meta-study approach (Paterson et al. 2001) was ultimately chosen for the current synthesis because it is a rigorous and comprehensive methodology offering "a critical, historical, and theoretical analytic approach to making sense of qualitatively derived knowledge" (Paterson et al. 2001, pg 2) and can provide a more "intriguing and complex" account of qualitatively derived knowledge than "more aggregate meta-synthesis approaches" (Patterson et al. pg 39). The process of "making sense" through a meta-study involves three analytical components (Barnett-Page and Thomas, 2009; Paterson et al., 2001); meta-method, meta-theory and meta-data-analysis (Table 1.3). Considered together, these three components form a meta-synthesis. Paterson et al. (2001) resist providing a structured procedure to meta-synthesis, rather they state that the process should be a "dynamic and iterative process of thinking, interpreting, creating, theorizing and reflecting" (Paterson et al, 2001, pg. 111) and can be presented as insights that have been produced by the three meta-methods (Thorne et al. 2002).

Because of the collective commonality coupled with the unique challenges that rare diseases can pose, the objective of this meta-study is to identify and synthesise the available qualitative research evidence across rare disease conditions, to establish the common themes related to childhood lived experience of rare diseases and to understand the challenges posed by rare diseases. Table 1.1 presents the two questions that the current meta-study aims to answer.

Table 1.1

Questions to be Answered by the Current Meta-Study

1	What theories and methods are used to formulate children's lived experience of rare disease within qualitative research literature?
2	What is children's lived experience of rare disease?

Method

We conducted a meta-study, following the guidelines proposed by Paterson et al. (2001). The protocol was pre-registered. (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=167922).

Selection criteria

Studies were included if the main focus was on child and/or adolescent (aged <18 years of age) lived experiences of a rare condition/disease; a disease that affects no more than 5 in 10,000 of the general population; were English language and used qualitative methods in a standalone or mixed methods study. Studies were excluded if they only used quantitative methods, if they focused exclusively on adult experience of managing a rare condition/disease and/or if the disease in question was not sufficiently rare. An additional criterion for exclusion was if a study scored below 5 on the CASP quality appraisal tool.

Search and data sources

Databases used for the initial literature searches were: Embase, PsycINFO, Web of Science and PubMed. Grey literature was searched using SIGLE, ETHOS and Conference Proceedings Citation Index-Science (CPCI-S) via the Web of Science. The search strategy was refined and finalised with assistance from the Education and Psychology Librarian at the University of Bath. Particular words relating to participants, populations and methodology relevant to the review question, such as “child”, “rare diseases” and “interviews” were used as search terms, alone and in combination (Table 1.2). See Appendix B for the full search strategy.

Table 1.2

Key Search Terms

Search terms	Combinations
rare disease	Rare diseases OR Rare AND disease
orphan disease	Orphan disease OR Orphan AND disease
lived experience	Lived AND experience OR experience OR experiences
child	Child OR adolescent OR teen OR juvenile Child AND adolescent AND teen
adolescent	
teen	
juvenile	
interviews	Interview(s) OR survey(s) OR questionnaire(s) Interpretation OR interpretations OR interpreters OR interpretable Theme OR themes OR themed
survey	
questionnaires	
Interpretation	
theme	

Study selection

Titles and abstracts of all identified papers were uploaded to the online systematic review management software RAYYAN (Ouzzani et al. 2016). All studies were double screened by two reviewers independently, at title/abstract stage and at full text stage. Disagreements were resolved through discussion.

Data extraction

For each study, the following data were extracted: authors' disciplinary background; country of origin; sample size; participant demographics; study methodology; first and second order constructs (participant quotes and author generated themes); reported limitations and a concise summary of the study findings. A bespoke data extraction form (Appendix C) was used by two reviewers, the first reviewer extracting data from all of the studies whilst the second reviewer extracted data from 50% of the studies.

Data Analysis

For the current study, the three meta-methods of the meta-study methodology were carried out concurrently and were used to answer the research questions: "What theories and methods are used to formulate children's lived experience of rare disease within qualitative research literature?"; "What is children's lived experience of rare disease?". Components of the meta-study and details regarding the application of these components are presented in Table 1.3.

Meta-Method

The meta-method component involved a critical appraisal of methodological rigour. The reviewer used the Critical Appraisal Skills Programme (CASP) checklist for qualitative research (Appendix D). The meta-method component also involved an examination of various methodological aspects of the included studies. The particular aspects that were focussed on were informed by the published guidelines for conducting a meta-study (Paterson et al., 2001) and were as follows: examination of the research questions, the aims and the methodology and assessing if an adequate "fit" exists between these; identifying the disciplinary backgrounds of the researchers and setting of the research, assessing use of reflexivity; reflecting on the sampling procedure and how this may have influenced the findings; identifying which data collection techniques were used, in particular reflecting on "style and skill" of the researchers; assessing whether the conclusions that reached by the researchers seem to be in line with the original aims.

Meta-Theory

The meta-theory component firstly involved a thorough reading of each individual study and noting which theories are referred to by the authors. As advised by Paterson et al (2001), in the absence of overt theoretical references across studies, key terms used by authors throughout the text were used to hypothesise the theoretical underpinning of the research. The historical context of the theories was also considered.

Meta-Data-Analysis

For the meta-data-analysis component a meta-ethnographic approach (Noblit & Hare, 1988) was used. First-order constructs were identified (direct quotations from study participants) that had been ascribed to child participants (<18). All first-order constructs were entered into an excel spreadsheet and a process of interpretation commenced whereby common themes were identified across the dataset and through an iterative process the first-order constructs were grouped into third-order constructs. The lead author constructed four, third-order constructs that describe the lived experience of children with rare diseases (Table 1.5). The third-order constructs are described below with illustrative examples of first-order constructs, labelled with the author. To increase the credibility and trustworthiness of the meta-data-analysis, a second reviewer went through the process separately and the findings from the first and second reviewer were compared. The third-order constructs that the second reviewer constructed bore a striking similarity to the lead authors third-order constructs.

Table 1.3.

Components of the Meta-Study and Details Regarding the Application of These Components

Questions posed by the current meta-study	Component of Meta-Study	Process of application for each component
What theories and methods are used to formulate children's lived experience of rare disease within qualitative research literature?	Meta-Methods	Appraisal of the methodological quality of the individual studies using the CASP checklist. Identifying specific elements of the individual studies that relate to the methodology. Considering trends, themes and assumptions are represented across the studies, and what is neglected.
What theories and methods are used to formulate children's	Meta-Theory	Identifying theories and the paradigms that underpin those theories.

lived experience of rare disease within qualitative research literature?		Examination of the historical evolution and context of the theories.
What is the lived experience of children with rare disease?	Meta-Data-Analysis	Utilising the meta-ethnography approach developed by Noblit & Hare (1988) Identifying the first (participant quotes) and second order (researcher generated themes) constructs from the individual studies that relate specifically to children and adolescents. Inputting first and second order constructs to an excel spreadsheet.

Results

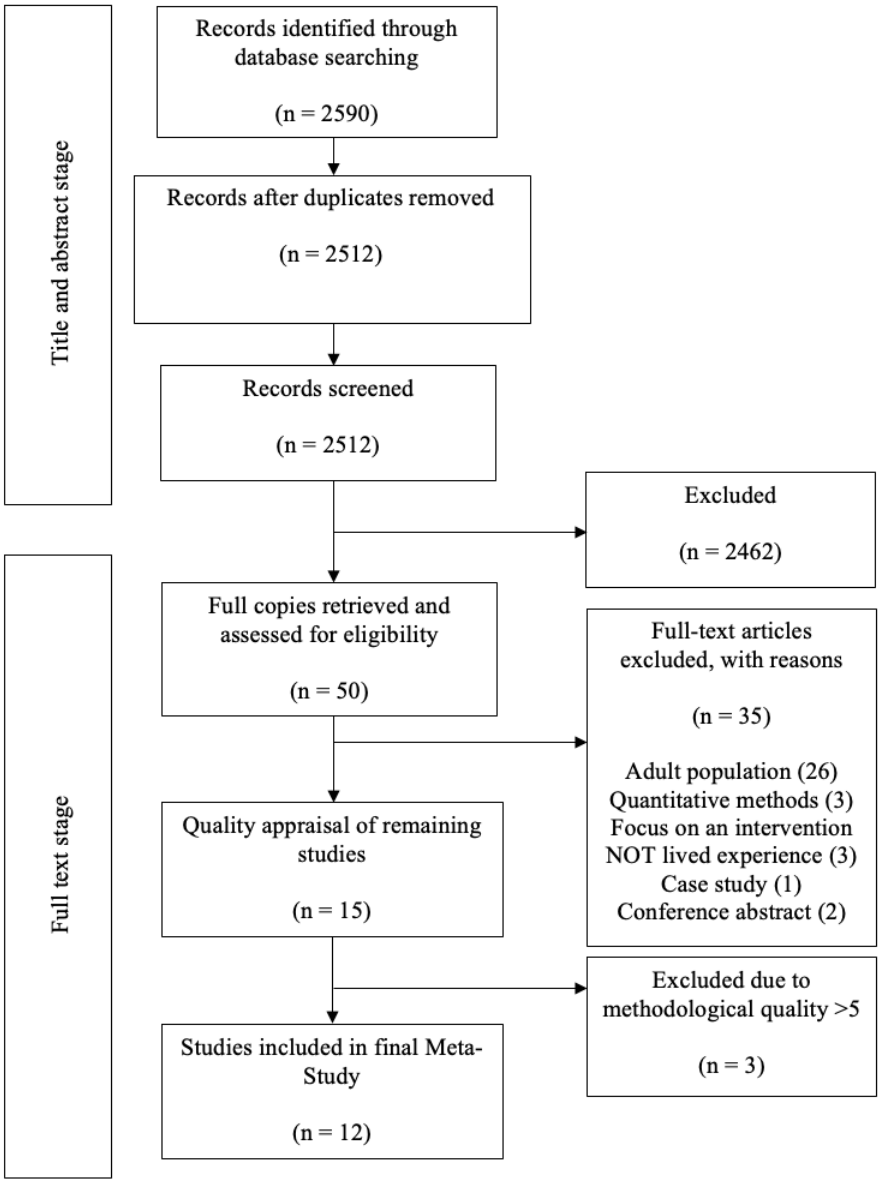
Included Studies

The initial search produced 2512 studies after duplicates were removed (Figure 1.1), 2462 studies were subsequently excluded at the title and abstract stage, reasons for exclusion at this stage included non-medical research and non-human research. Full texts for 50 studies were assessed for inclusion eligibility, of which 35 were excluded (see Figure 1.1 for reasons).

In total, 12 studies involving 652 children and adolescents aged 4 – 21 were included in the meta-study (Table 1.4). Age was not reported in 3 studies and sex was not reported in 2 studies. Of the studies that did report sex and age data, 91 males and 82 females. 10 countries were represented across the studies, 9 of which were “Western” countries.

Figure 1.1.

PRISMA Flow Chart Showing Data Screening Process (Moher et al. 2009)



Description of Included Studies (meta-method)

A high level of agreement between the two reviewers was attained on the CASP scores (87%). The CASP scores across the studies varied from 5 to 10 (Table 1.4) which means that the quality of the included studies was variable. 3 studies that scored below 5 were removed from the meta-study (Figure 1.1). The included studies presented clear aims

that were appropriately addressed by qualitative methodology. Most studies (n = 9) lacked reflexivity. With the exception of Paz Lourido et al. (2020) and Livermore et al. (2019), it was not clear whether the authors had a dual role of researcher and clinician. It would have been useful to know this information particularly because this may have had an impact on recruitment.

Across the studies the disciplinary backgrounds reflected medicine (n = 5), nursing (n = 4), psychology (n = 3), general academia (n = 1) and research and development (n = 1). The samples were drawn from treatment centres (n = 7), a treatment centre and a conference (n = 1), a conference (n = 1), a patient information day (n = 1), a support group (n = 1) and a charity (n = 1). Sampling methods employed were either purposive (n = 9) or convenience sampling (n = 3). In most studies, apart from both Livermore studies, the sample was reported to be a mix of adults and children; studies included adults with a rare condition (n = 3), parents (n = 9), grandparents (n = 1), teachers (n = 1) and siblings (n = 1). This meant that the conclusions that were made, i.e., regarding the experience of *children*, actually represent many stakeholders, rather than just children. This has implications for understanding children's lived experience of rare diseases, particularly the meta-data-analysis element of the current meta-study. Often, when conducting a meta-ethnography, one would identify and use first and second-order constructs (participant quotes and researcher generated themes), however, the second-order constructs in most of the papers (n = 9) represented an amalgamation of child and adult experience, therefore it was decided to use only first-order constructs in the meta-data-analysis.

Various data collection techniques were used including: A focus group (n = 1); 1 to 1 interviews (n = 5); focus groups plus 1 to 1 interviews (n = 3); questionnaires (n = 1); and published creative writing (n = 1). Analysis methodologies employed across the studies also varied: thematic analysis (n = 2); content analysis (n = 3); grounded theory (n = 1); interpretative hermeneutic phenomenology (n = 1); interpretive description (n = 1); qualitative case study methodology (n = 1). Epistemological position was reported in one of the studies as "deductive".

Table 1.4.

Included Studies

Author, year & journal	Country	Authors' disciplinary background	Rare condition/setting	Participants	Age range	Sex	Sampling	Data collection	Analysis	CASP quality score
Currier & Zimmerman (2019) Nephrology nursing journal	USA	Nursing, Psychology	Paediatric Kidney Failure; Dialysis treatment centres	500 children and young people	5 – 18.	Not reported	Purposive	Published creative writing	Content analysis (deductive)	9
Dell et al. (2016) Annals of the American Thoracic Society	USA, Canada, UK and Ireland	Medicine, Psychology	Primary Ciliary Dyskinesia; treatment centres at hospitals and a patient conference	Focus groups: 9 children, 7 parents of children under 12, 7 parents of adolescents Interviews: 20 children; 20 adolescents; 29 parents.	Not reported	Interviews: Children: 8 Male 12 Female Adolescents 12 Male 8 Female	Convenience	Focus groups and open-ended interviews.	Content analysis	7
Dogba et al. (2013) BMC Pediatrics	Canada	Medicine	Osteogenesis imperfecta; Specialist paediatric orthopedic hospital.	12 patients 8 parents	15 – 21	Children: 9 female 3 male Parents: 8 mothers 4 fathers	Purposive	One to one semi structured interviews	Interpretative description	9

Erbis et al. (2018) Pediatric Rheumatology	Germany	Medicine (Rheumatology)	Autoinflammatory diseases; A patient "information day".	48 AID patients (14 children, 9 adolescents, 25 adults), 35 family members	5 - 18	18 female 30 male	Convenience sampling	focus groups and questionnaires	Mixed methods including Mayrings Qualitative Content Analysis	8
Henderson et al. (2009) American Journal of medical genetics	USA	Medicine, Psychology	Neimann-Pick disease type B; annual Niemann-Pick Disease Family Conference.	8 patients (4 adults, 4 adolescents), 9 parents	13 - 18	3 male 5 female	Convenience sampling	Semi-structured interviews	Qualitative case study methodology	7
Livermore et al. (2020) Journal of poetry therapy Livermore et al. (2019) Paediatric rheumatology	UK	Nursing	Juvenile Dermatomyositis; clinics at a specialist tertiary hospital in London.	15 participants	8-18	6 males, 9 females	Purposive sampling	Interviews (including body mapping, time-lining, comic book design)	Interpretative hermeneutic phenomenology	9 (2019) 6 (2020)
Paz-Laurido et al. (2020) Health and Quality of Life Outcomes	Spain	Nursing	"rare diseases" Patient charity.	Participants (n= 28) included children with rare diseases (n= 8), parents (n= 12) and school staff (n= 8).	Age not reported	Sex not reported	Purposive sampling	Focus groups (n=3) and semi structured in depth interviews (n = 8)	Discourse Analysis	10

Roborel de Clemens et al. (2020) Ophthalmic Genetics	USA and France	R&D international healthcare company	type 1 Usher syndrome ; Ophthalmology clinics	18 Patients (7 USA, 11 France): Children = 5 adolescents 4 adults age 18 and over = 9 mothers = 7 (2 USA, 5 France)	6 -17	11 female 7 male	Purposive sampling	Semi-structured Interviews	Thematic Analysis	8
Thomas et al. (2020) Haemophilia	India	Nursing	Haemophillia; Heamophilia treatment centre	19 Children = 9 mothers = 7 fathers = 1 grandmothers = 2	4 – 12	9 male	Purposive sampling followed by theoretical sampling.	Semi-structured interviews	Grounded Theory	5
Sneller et al. (2014) International journal of paediatric dentistry	UK	Institute of Work, Health and Organisations, Uni of Nottingham	Amelogenesis Imperfecta; Paediatric unit of a dental hospital	8 Adolescents = 4 Parents = 4	11 - 16	3 male 1 female	Purposive sampling	Semi-structured focus groups	Thematic analysis	9
Zeltner et al. (2016) Journal of Inherited Metabolic Disease Reports	Germany, Austria and Switzerland	Medicine	Intoxication-type inborn errors of metabolism; Metabolic centres	19 paediatric patients and 26 parents	8 – 18	10 male 9 female	Purposive sampling	Focus groups and interviews	Mayrings Qualitative Content Analysis	9

Description of Included Studies (meta-theory)

Most studies (n = 8) did not directly refer to any theoretical basis of their studies. Two studies were explicit about their theoretical basis; Paz-Lourido et al. (2020) identified the social-critical paradigm and Henderson et al. (2009) referred to Erikson's theory of psychosocial development. Both imply that the authors considered children's experience of rare disease to be a dynamic and subject to change; A social critical perspective suggests an interest in social change related to power and inequality (Alderton, 1998), whilst the theory of psychosocial development conceptualises a process whereby an individual passes through various stages over time as they develop and change (Orenstein & Lewis, 2020). The concept of change relating to a theoretical perspective is further evident in two further studies, Dogba et al. (2013) describe a theoretical positioning that emphasizes a "temporal approach" to the rare condition, that the experience of having the rare health condition does not remain "frozen in time". Thomas et al. (2020) use of grounded theory to create a "theory of therapeutic metamorphosis", suggests that therapeutic input is transformative and can fundamentally change the individual in a positive way.

Across the studies, frequent use of the term "psychosocial" was evident (i.e., psychosocial stress; psychosocial impact; psychosocial difficulties; psychosocial burden; psychosocial adjustment; psychosocial needs; psychosocial effects; psychosocial aspect; psychosocial issues). It has been suggested that the term "psychosocial" refers to "everything and nothing in particular" (Martikainen et al. 2002, pg 1091). Within the included studies it was not always clear what the authors use of the term meant, however Livermoore et al.'s (2019) definition provides some clarity: "pertaining to both psychological and social, encompasses a multitude of variables, such as social and emotional support, mood and anxiety" (pg 1). Henderson et al.'s (2009) direct reference to Erikson's theory of psychosocial development sheds further light on the term, the theory that an individual goes through 8 stages of development through life that are influenced by biological, psychological and social factors (Orenstein & Lewis, 2020). Furthermore, the concept of health-related quality of life (HRQOL), which is directly referred to in some of the studies (n = 8), helps to give more of a theoretical structure to the term "psychosocial". Zelner et al. (2016) provides the following definition: "HRQOL is a multidimensional construct that represents a patient's perception of the impact of disease and treatment on functioning in a variety of dimensions, including physical, psychological and social domains"

(Varni et al. 1999, p 126). In this context, frequent use of the term “psychosocial” to refer to the interaction and interrelation of psychological and social factors would seem to suggest that the studies are rooted within the biopsychosocial paradigm.

The biopsychosocial perspective was first proposed by George Engel (1977) as a response to the traditional biomedical paradigm. Engel was influenced by the general theory of systems developed by Von Bertalanaffy (1968), who proposed that organisms can be understood as organised systems with multiple parts, interrelated and constantly interacting. Therefore, underpinning the biopsychosocial model is the idea that, as an organised system, an individual’s wellbeing is influenced by an interaction between biological, psychological and social factors (Wade & Halligan, 2017). The model is widely used in research, though in clinical practice less clarity exists as to its usage (Suls & Rotham, 2004), there is evidence of health professionals’ negative attitudes regarding the importance of psychosocial issues (Astin et al. 2006) and health services tend to be built around the biomedical model (Suls & Rotham, 2004). The biopsychosocial model has been criticised for omitting spirituality (Sulmasy, 2002; Edge & Lemetyinen, 2019), for not fully considering the social domain (Lehman et al. 2017) and not adequately addressing cultural variation (Hatala, 2012). Lehman et al. (2017) have suggested a dynamic biopsychosocial model drawing on Brofenbrenner’s ecological systems theory (1979, 1986). Lehman et al. discuss how Brofenbrenner’s concepts of Microsystem, Mesosystem and Exosystem can help to “elaborate interpersonal dynamics in social environments” (pg. 1), and how the concept of the Macrosystem help to understand how “contextual dynamics”, such as culture and spirituality, impact on health and shapes the relationships between the biological, social and psychological factors. The influence of Bronfenbrenner’s theory can be seen in Henderson et al. (2009), coping with rare disease is described as being influenced by layers of context.

Meta-Data Analysis

Table 1.5 presents the distribution of the four, third-order constructs across the 12 studies. Each of these will be discussed in detail, with illustrative quotes from the primary studies.

Table 1.5.

Distribution of Third-Order Constructs Across Studies

Author (year)	Sense of self defined through difference	Daily life informed by a restriction narrative	Intra and Interpersonal distress in a context of uncertainty	Coping with the lived reality of the rare disease
Currier & Zimmerman (2019)	X	X	X	X
Dell et al. (2016)	X	X	X	
Dogba et al. (2013)	X	X		X
Erbis et al. (2018)				X
Henderson et al. (2009)	X	X	X	
Livermore et al. (2019)			X	X
Livermore et al. (2020)	X		X	
Paz-Lourido et al. (2020)	X	X	X	X
Roborel di climens et al. (2020)		X		X
Thomas et al. (2020)		X		X
Sneller et al. (2014)			X	X
Zeltner et al. (2016)	X		X	

Sense of self defined through difference

This third-order construct describes how children with rare conditions' sense of self is defined, both through an internal sense of feeling different and an external sense of being treated differently by those around them. Across the studies, this sense of difference changed depending on the age of the child; young children might report experiences of being ignored/overlooked whilst older children reported experiences of being actively excluded. A felt sense of difference was sometimes due to the types of medical treatment that children require, for example Currier (2019) explained that a child reported feeling "different because I need dialysis for kidney problems." Sometimes a felt sense of difference was because a child might be the only person with the condition, for example one child explained that "it's a weird feeling because no one else has got it. I'm the only one that's got it." (Dell et al, 2016)

Feeling different could sometimes lead children to conclude that they are not "normal"[1], Zeltner et al. (2016) describe that the most frequent "fondest wish" of the children was "not to have the disease" and be "more like normal kids". Sometimes the felt

[1] The term "normal" is directly quoted from Zeltner et al (2016). It is acknowledged that this is a subjective term.

sense of being different was described in more tangible ways, for example a child might describe looking unusual compared to their peers, Livermore et al. (2009) provided the following quote from a child illustrating this point, “I hated the way that I looked so I wouldn’t leave the house without make up on. My face was literally bright red and swollen, and I just looked horrible, like a baboons bum.” Sometimes the felt sense of being different was not so obvious, even being “invisible” to others, for example another child explained that their chronic illness “is hidden, it’s more inside the body, it’s not like a visible disability with a wheelchair all the time, so just because you can’t see it, it doesn’t mean it’s not there.” (Livermore, 2019).

In addition to *feeling* different, children and young people also experience being *treated* differently by others, particularly their peers, which could be a consequence of looking different or experiencing obvious symptomology. One child explained that their peers “don’t want to talk to me because I’m short” (Currier, 2019), whilst another child felt like they were “being judged by other people because I constantly sniff and cough” (Dell, 2016)

Although being treated differently can sometimes be experienced as negative, this is not always the case. Children sometimes perceived benefits to being different, almost like having a sort of social cache which helped them to reframe being “different” as being “special”: “It turned out that having the illness has good moments as well because it’s so rare people took an interest. I got invited to this party for people with bad illnesses, so you do feel kind of special.” (Livermore, 2019)

Children with rare conditions feel different and experience being treated differently because of their condition, the experience of being different can be negative and positive. In a sense this third order construct can be encapsulated by a dialectic between not being “normal” whilst also being “special”.

Daily life informed by a restriction narrative

This third order construct captures the sense that a child’s daily life is restricted by their rare condition. Restriction in this case can be understood as multifaceted and refers not only to symptoms being experienced as restrictive, also how children with rare conditions can experience functional restrictions, physical restrictions and educational restrictions. Children talk about not being able to engage in certain types of activities:

> *"I can't play badminton. Badminton: for me, that's impossible. Even if you put colours on the shuttlecocks... Um, when it comes to basketball, That's okay, that's okay. Sometimes, if someone throws a ball really hard, I can't follow the ball anymore, actually, I lose sight of it."* (Roborel de Climens, 2020)

Children also talked about their condition sometimes getting in the way of attending school, for example one child explained that "I cannot go to school everyday as it is too much for me. I need to rest and I also have other treatments. I can attend a few subjects and that is enough." (Paz-Lourido, 2020)

The various restrictions that rare diseases impose on children can be experienced as challenging and inconvenient, particularly when treatment takes up a lot of time and gets in the way of engaging in "normal" life. For example, one child described how "annoying" their treatment was "because I have to do it for 10 minutes, it takes time out of my life that I want to go off and do something else instead." (Dell, 2016)

Intra and Interpersonal distress in a context of uncertainty

This third order construct reflects the distress that the children with rare conditions experience. Distress can be a feeling that is within the person (Intrapersonal), understood as an emotional burden that saps energy and motivation, one child describing how "all of [their] energy" is "wasted" and leaves them feeling like they "don't want to wake up in the mornings" (Dell et al. 2016). Another child simply described how they "feel useless to all the world" (Currier, 2019). Children's distress is also presented as being experienced between themselves and others (Interpersonal). Some children reflect on being ignored or not being believed by the adults around them:

> *''I understand more what the doctors are saying. I wish they'd just tell me. Sometimes I overhear, like when they measure my stomach or liver and spleen. I worry they'll get really big. Sometimes I overhear them say I need to get an MRI and I don't like those.''* (Henderson, 2009)

Another example of not being believed by the adults around them is provided by a child who explained that "for most of us with rare diseases, physical education is the most difficult subject. The teacher told me that my illness is not true, if nobody can see it, it doesn't exist." (Paz-Lourido, 2020). Even when others *do* understand the experience that the child is going through, a sense remains that they do not fully understand, which may increase distress:

> *‘‘Me and [my brother] can relate really well about it. And [my other brother], he knows all about it, but he doesn’t have it so he doesn’t know what it’s really like. He’s okay about it, but some issues he doesn’t really understand.’’* (Henderson, 2009)

As it seems that children with rare diseases commonly experience being misunderstood, not believed or ignored it is unsurprising that also reflected in the research is a sense that children and young people may be living with a condition but not being so open about it because of the risk of others misunderstanding and perhaps even bullying them. Distress can also be caused by a lack of trustworthy information about the rare condition, which can lead to a feeling of uncertainty as to what the future may hold. This feeling of uncertainty fuels a child’s desperate search for answers, coupled with a fear that they are being problematic; wasting others’ time:

> *“Is it psoriasis? Is it eczema? Is it scarlet fever? Why are you such non-believers? Is it Lupus? Is it growing pains? Please help me, use your brains? Shall we google it? There’s got to be something that fits? We’re made to feel we’re wasting their time, the examining couch too high, I can’t even climb, My face bright red, The weight too heavy to lift my head, I now am very weak, An answer I seek, Help me get a name, Please help me, stop the pain.”* (Livermore, 2020)

It cannot be guaranteed that the feeling of uncertainty will be abated, as even medical professionals are unable to provide answers to the questions that children have, for example one child explains that “my GP (General Practitioner) said he didn’t know what was happening to me, so he referred me to my local hospital, but they didn’t know what was wrong with me.” (Livermore, 2019)

Coping with the lived reality of my rare disease

Children and young people with rare diseases are often living lives that are beset with challenges, this third order construct concerns the ways in which children and young people cope with these challenges. Ways of coping can be broadly defined as either intrapersonal or interpersonal. Children are sometimes presented as drawing on internal resources, i.e. intrapersonal coping, seeing themselves as strong and engaged in a fight with their rare condition, exemplified by one child who stated that “this is me, and this is my life. I will never let anyone, anything, any disease, any rejection, get the best of me. I am a survivor and a fighter” (Currier, 2019). The use of the terms “survivor” and “fighter”

speaks to a wider narrative of strength and resiliency that seem to be particular threads that are woven through these studies. Part of this strength and resiliency seems to be an ability to live in the moment and connecting with others, for example one child states that they have to "live day to day, [and] never take anything for granted" (Dogba, 2013). In addition to living in the moment, some children and young people with rare conditions have developed a stoicism that perhaps helps them to cope. Livermore presents an account from one child as follows: "You get over it, because you don't have a choice, that's the thing that I learnt fairly quickly, that it's fine to be sad about it, but if you're going to sit there and be sad about it, you're going to let it stop you living your life, because it's not going to go away." (Livermore, 2019). Another child displays their stoicism by saying, "when I see somebody who had a minor break and will be down for a month. Really this is nothing, I mean really nothing." (Dogba, 2013).

In addition to drawing on internal resources, children described things that are external to the self and how these could help them to cope with their rare disease. This can be understood as interpersonal coping. Throughout the studies children made reference to specific objects or people who would help to perform difficult treatment regimens or be able to 'step in' more often to support with education. For example, a child refers to their dialysis machine, which they call "Vanessa": "Oh Vanessa you keep my blood so clean. Three days a week I have a date with you at noon, until the sun sets and over the building comes the moon... Beep! Beep!" (Currier, 2019). Whilst many children rely on machines, they also rely on other people, as is evident in this child's statement: "I have an assistant who helps me with everything, knows when I am tired, when I am not feeling well, calls my mother if necessary and also helps me when I cannot hear the teacher." (Paz-Lourido, 2020). More often than not children rely on both "things" and people:

> *"I think, for me, it is just the little things and not so much the big things. For instance, reaching something or relying on other people to do certain things for you. Everything for me, like personal care, or being in the kitchen, is pretty hard."* (Dogba, 2013)

A key point that comes across from the data is that the "others" that one connects with should have a shared experience of the particular condition: "Would be great to learn how others cope with this. Perhaps they can talk to someone. How they handle living with the symptoms, the pain." (Erbis, 2018). One could imagine that this may be challenging

when a condition is rare, though this may be less of a challenge if connections are made via the internet, something that Sneller et al. (2014) explore in their research.

Discussion

This meta-study synthesised the research findings from the lived experience of a large sample of children (n = 652) with rare diseases from various geographical locations. A variety of methods were used across studies to formulate children's lived experience of rare diseases. Methodological quality of the included studies varied (10 – 5), children's experience was formulated with a lack of reflexivity and drawing on a mix of child and adult participants. Reference to theory across the studies was lacking, though frequent use of the term "psychosocial" led to a hypothesis that the studies were broadly positioned within the biopsychosocial paradigm, suggesting that children's experience of rare disease is formulated as multidimensional, multi-layered and influenced by various different factors. The meta-data-analysis found that children with rare diseases feel different and experience being treated differently; they feel restricted in a variety of ways by rare disease; feelings of distress and uncertainty are common; children describe ways of coping with the challenges of rare diseases by drawing on internal and external sources of support.

Findings from the current meta-study are consistent with findings from the adult rare disease literature. Feeling different and being treated differently by others, restriction of daily life and uncertainty are all highlighted in research focussing on adult's experience of rare diseases (Brugalle et al. 2019; Von der Lippe et al. 2017; Bailey et al. 2017; Garrino et al., 2015; Jaeger et al. 2015; Lyn et al. 2020; Huyard, 2009; Bryson et al. 2020; Nunn, 2017). In addition to being consistent with the adult rare disease literature, there are also commonalities with the current findings and research into adolescents' experience of living with common chronic illnesses. Adolescents' living with common chronic illnesses also report that they feel different and are treated differently by others (Taylor et al. 2008). Difficulties related to education are reflected in the review by Taylor et al. (2008), particularly bullying, lack of support and lack of understanding, all things that are highlighted in the current meta-study. The theme "coping with the lived reality of the rare disease" bares a similarity to a main theme identified by Taylor et al. (2008), "Being normal/getting on with life". In the current study, stoicism was highlighted as a way of coping with rare disease, this is something that is also reflected in the adult rare disease literature, for example, Brugalle et al (2019) report that adults reject "wallowing in self-

pity" and sometimes reflect that their rare disease increases their resiliency. The importance of connection, meeting others with rare disease is something that is also highlighted in an adult focussed rare condition qualitative literature (Huyard, 2009; von der Lippe, 2017; Doyle, 2015; Meade et al. 2018; Crowe et al. 2019). However, comparing the current findings to those of Taylor et al. (2008), uncertainty is not talked about in their review of adolescent experience of more common chronic conditions. This is perhaps understandable when one considers that common chronic conditions by their nature are possibly better understood, meaning that greater clarity exists around their development and progression. In addition to this, specific treatments, interventions and support groups exist for people with common chronic conditions (Doull et al. 2017; Embuldeniya et al. 2013), whilst comparative support may be fragmented or non-existent for people with rare conditions (Anderson et al. 2013; Kesselheim et al. 2014; Jaffe et al. 2010; Currie & Szabo, 2018; Stoller 2018), further impacting on a sense of uncertainty. Indeed, the lack of medical knowledge for many rare diseases is highlighted frequently in the adult literature (Huyard, 2009; Von der Lippe et al., 2017; Jaeger et al. 2015; Kesselheim et al. 2015). Therefore, someone with a common chronic condition may not experience the same sort of uncertainty that is far more common with rare conditions.

Whilst there were similarities with the adult rare disease literature, there were also differences. Isolation is often reported in adult literature as being central to the lived experience of rare disease (Petersen 2006; Barlow et al. 2007; Frank et al. 2007; Brodin et al. 2015; Jaeger et al. 2015; Kesselheim et al. 2015; Palareti et al. 2015), but this was not something that was evident from children's experience in the current meta-study. Reasons for this divergence could be that children were not asked about feeling isolated and therefore it is not reflected in the data, alternatively it could be that a coping strategy, relying on others, means that they do not feel isolated. Interestingly, *parents* of children with rare diseases frequently report feeling isolated (EURORDIS, 2005; Brewer et al. 2008; Anderson et al. 2013; Somanadhan & Larkin, 2016; Baumbusch et al. 2018; Carpenter et al. 2018), so perhaps parents act as a buffer from children feeling isolated.

An additional finding of the current synthesis is that the "experience" reported in the included studies is not exclusively child related, rather the second-order constructs are an aggregate of a variety of perspectives. For the current meta-study to reflect the lived experience of children, only the first-order constructs were considered at the meta-data-

analysis stage. This is of particular importance because it has been highlighted in previous research that children sometimes feel that they are being ignored or decisions are made for them without consultation (Sartain et al. 2000), this is also something that was evident in the children's accounts in the current meta-study, some of the studies highlight that the experience of children differs throughout childhood and that younger children may actually feel "silenced" (Currier and Zimmerman, 2019; Dell et al. 2016; Livermore, 2019). Future research could focus primarily on child experience, particularly because this meta-study highlights that children with rare diseases feel that they have not been listened to, or even "silenced", furthermore children and their parents' experience of rare disease might differ and therefore considering their accounts separately may provide different insights.

Strengths and limitations

Across the studies a variety of methodologies were represented with varying analytical rigour, for example some second-order constructs were presented without any illustrative first-order constructs (Erbis et al. 2018; Henderson et al. 2009; Thomas et al. 2020; Zeltner et al. 2016) and in some cases the analysis process was not presented with sufficient information (Erbis et al. 2018; Henderson et al. 2009; Livermore et al. 2020). Of central importance to quality and trustworthiness of findings is reflexivity (Clarke et al. 2015) and is integral to conducting good qualitative research (Newton et al. 2011). The current meta-study found a lack of reflexivity across all the studies. Only two studies considered the impact of the disciplinary background of the researchers (Paz-Lourido et al. 2020; Livermore et al. 2019). A lack of reflexivity in qualitative health research has been reported elsewhere, Newton et al. (2011) provides one potential explanation which is the restrictions that journal editors place on manuscript length, suggesting that whilst research may be conducted in a reflexive manner, reporting of reflexivity may be a casualty of strict word limits, this is supported more recently by Levitt et al (2018). Braun and Clark (2020) recommend that, in order to enhance reflexivity and improve practice, journal editors provide greater word limits for qualitative papers. The lack of reflexivity could also be related to the epistemological tradition of the type of journals that publish the research. Although a mix of quantitative and qualitative research can be found in medical journals, suggesting both positivist and interpretivist paradigms, the dominant framework within medical education and research continues to be that of positivism (Bunnis & Kelly, 2010). From a positivist perspective reality is objectively observable and exists independently

from the researcher (Alderson, 1998) and as such overt reflexivity may be viewed as radically different (Ferguson, 2018). Therefore, lack of reflexivity in the studies could be due to editorial decisions that are directly informed by a positivist tradition (Bunniss & Kelly, 2010).

The data presented in the current meta-study mainly represents western countries, with the exception of Thomas et al. (2020) which was based in India. It has been suggested that western countries healthcare systems tend to follow a biomedical trajectory (Suls & Rothman, 2004) and it is known that the biopsychosocial model was proposed as a response to the biomedical model (Engel, 1977). The findings of this meta-study therefore represent a majority western view, it would be interesting to compare the findings to future research in non-western countries.

There was a lack of clarity with regard to the theoretical underpinnings of the included studies and this limited the meta-theory component. It has been suggested that, to ensure clarity and integrity of scientific thinking, researchers should be explicit about the theoretical framework on which they base their research (Tudge et al. 2009). Other meta-studies have commented on changes in theoretical positioning over time (e.g. Thorne & Patterson, 1998). This was not possible in the current study, perhaps because of the relatively short time period represented by the studies, ie. 11 years (2009 – 2020), reflecting a more homogenous theoretical context. The short time period also suggests that understanding of the lived reality of children with rare diseases is still at an early stage.

A critique levelled at individual qualitative studies is that, because of small sample size, results cannot be generalized, and knowledge development is limited (Hews-Girard et al. 2019). When one considers rare diseases, it is perhaps unavoidable that a qualitative study focusing on a single rare disease will have a small sample size, though when considered together rare diseases are “collectively common” (Onepoll, 2016). Being a combination of various qualitative studies, the current meta-study benefits from a large sample size and also reflects a variety of rare diseases. However, it is important to note that 500 participants were drawn from just one of the studies (Currier & Zimmerman, 2019). Never-the-less, a strength of the current meta-study is that it presents a view of childhood experience of rare disease that has depth and breadth.

Whilst the questions posed by this meta-study regarded children’s experience, the studies themselves presented an aggregate of child and adult perspectives. Steps were

taken to only represent children's experience in the current study, i.e., only using first-order constructs to inform third-order constructs. Despite this, it could be argued that the findings do not exclusively represent children's experience, and this could be considered a limitation.

Previous researchers have reported on the process of conducting a meta-study as challenging (Aagaard et al. 2017). Thorne et al. (2002) reflect on the complex nature of the methodology and suggest that the endeavour "should not be undertaken by the inexperienced or faint of heart" (pg 449). The data provided by the included papers was perhaps not best suited to some elements of meta-study methodology. In particular, the meta-theory section posed a challenge because it was unclear what the theoretical underpinning of the research was, and in addition the relatively short time-period that the included research covered indicated that there was less theoretical heterogeneity. It has been stated that meta-study is most appropriate when there has been a paradigm shift in the approach to research in a specific area (Radnitzky, 1972). Meta-study methodology seems to be suited to a wide timeframe, the timeframe of the current synthesis is limited to 10 years, making it harder to provide a meta commentary of theoretical and methodological changes and trends. On reflection, the complexity of meta-study method might also suit a larger number of studies. Paterson et al. (2001) do not state exact numbers for a meta-study, though a greater number would increase the variety of methodologies/data/theories and increase the quality of findings.

Conclusion

The purpose of this meta-study was to explore the theories and methods that are used in qualitative research to formulate children's lived experience of rare disease and to gain an insight into children's lived experience of rare disease. A variety of methodologies are used, though a weakness tends to be a lack of reflexivity. Research tends to be presented with a lack of clarity regarding theoretical positioning. It would be helpful if future research could address these shortfalls. In addition, this meta-study highlighted that the understanding of children's lived experience of rare disease is still in its infancy and reflects a majority western world perspective. Future research could perhaps diversify to include the experience of children in non-western countries. Clinical implications of this meta-study are that clinicians should be aware that children with rare diseases will have a felt sense that they are different from others, even if it is not immediately obvious from

the outside, this difference may not always be perceived in a negative light, perhaps supporting a child in seeing their difference as positive could be helpful. A child's everyday life is restricted by rare disease, this may mean that practical solutions need to be found, perhaps drawing on the expertise from occupational health and physiotherapy colleagues. The experience of distress, uncertainty and being ignored that is highlighted in children's experience of rare disease suggests that, at the very least, steps should be taken to allow children to be heard and perhaps offer psychological support should this be needed. Finally, it has been highlighted that children with rare diseases would benefit from peer support from others who also have experience of rare disease, ideally the same rare disease. Use of technology, i.e., the internet, can help to facilitate this.

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Service Improvement Project Paper:

The experience of service users/people with personal experience being interview panel members for DClinPsych selection interview

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Abstract

The following Service Improvement Project presents a piece of work that was commissioned by the University of Bath, Clinical Psychology Doctoral (DClinPsych) programme admissions committee. This work firstly aimed to explore the experience of users of psychology services (known as People with Personal Experience, or "PPE") being part of the DClinPsych trainee admission interview process and secondly provide recommendations regarding the admissions process the following year. A focus group was conducted with 4 of the PPE representatives and transcribed verbatim. A version of Interpretative Phenomenological Analysis (IPA) adapted for focus groups was used to analyse the data. Four themes reflected the experience of the PPE as part of the interview process, these were: Challenges; A Unique Perspective; Connectedness; Finding a Voice. The themes intersect multiple levels of experience. Results are discussed in the context of previous research in the area. Recommendations and the corresponding response from the programme admissions committee are presented.

Introduction

Since the 1980's the UK government has increasingly supported a shift from an expert medical model of healthcare, towards a model that places service user involvement front and centre of all aspects of healthcare. This is reflected in governmental policy, particularly in terms of education of mental health professionals (Department of Health [DOH], 2005, 2007, 2009, 2010; Involve, 2004, 2007); all training programmes that are approved by the Health and Care Professional Council are required to have some element of service user involvement (HCPC, 2014). In terms of service user involvement in clinical psychology, the British Psychological Society (BPS) first made this a mandatory requirement of training accreditation criteria in 2002 and it remains in place to this day (BPS, 2017).

Various studies from the areas of psychology, nursing and social work support the hypothesis that the involvement of service users positively impacts on student learning, which may in turn affect the provision of services in several important ways. For example, there is evidence that service user involvement increases students' communication, partnership and advocacy skills (Curran 1997, Wood & Wilson-Barnett 1999, Beresford & Croft 2004, Tew *et al*. 2004, Simons *et al*. 2007, Duffy 2008); challenges professional orthodoxies and power (Harper 2002, Beresford & Croft 2004, Tew *et al*. 2004, Rush & Baker 2006); supports reflexivity in practitioners with regard to treatment implications and which approaches they use (Wood & Wilson-Barnett 1999, Khoo et al. 2004; Repper & Breeze 2007, Townend *et al*. 2008). In addition to benefits for students and services, service user involvement in the education of mental health professionals can also have a positive impact on the service users themselves, benefits such as feeling more empowered, having increased self-esteem, and seeing problems in different ways have all been reported in the literature (Walters *et al*. 2003, Barnes *et al*. 2006, Repper & Breeze 2007).

Clinical Psychology Doctoral training

Clinical Psychology training is highly competitive, in 2018 only 15% of applicants were successful in gaining a place on one of the 30 Clinical Psychology Doctoral (DClinPsych) courses in the UK (Clearing House website data, 2018). Whilst guidance exists about admissions to DClinPsych training programmes (BPS: Standards for the accreditation of Doctoral programmes in clinical psychology, 2017; HCPC: Standards of education and training, 2017), a universal model of admission to DClinPsych courses does not exist (Table

2.I) and there is a paucity of evidence regarding the validity of selection processes (Phillips, Hatton & Gray, 2004; Roth & Leiper, 1995).

Table 2.I.

Admissions Process (Information gathered from Clearing House website accessed February 2019)

Course	Number of Interviews	PPE sit on interview panel	Group exercise/discussion	Written task	Role play/client related exercise	Presentation	Video exercise
Bangor	1	X					
Bath	1	X					X
Birmingham	3	X	X				
Cardiff	2	X		X			
Cov and War	2	X	X		X		X
Edinburgh	2			X	X		
Essex	1		X	X			
Exeter	1		X				
Glasgow	2				X		
Hertfordshire	1		X	X			
KCL	1	X					
Lancaster	1	X	X		X		X
Leeds	2	X				X	
Leicester	1			X	X		
Liverpool	2	X					
Manchester	1	X					
Newcastle	1	X					
Nottingham	3						
Oxford	1		X				
Plymouth	2	X					
Royal Holloway	3	X					
Salomons	2	X					
Sheffield	1	X	X	X			
Southampton	2	X			X		
Staffordshire	2						
Surrey	3	X		X			
Teeside	2	X					

UCL	1		
UEA	2	X	
UEL	2		X

Whilst there is a lack of clarity regarding a standard admissions procedure, the BPS and HCPC is clearer about involvement of "service users" or people with personal experience of mental health difficulties, also known as People with Personal Experience (PPE) in DClinPsych training courses. For example, the HCPC state that "service users and carers must be involved in the programme" (Section 3.7). The BPS recommend that training programmes have collaborative working relationships with service users "to identify and implement strategies for the active participation of these stakeholders in the programme" (section 8.2.6).

Service users and DClinPsych training

Across the 30 UK clinical psychology training programmes service users are involved in a variety of ways, as part of advisory groups/committees, delivering teaching and supporting research. In 2018, 18 programmes had service user involvement on interview panels (Table 2.I). As service user involvement in clinical psychology training increases it is vital that this involvement is not tokenistic (BPS, 2012). Service user involvement should be "meaningful, with expectations and responsibilities clearly defined, understood and respected by all parties" (Matka et al. 2010, pg. 2153). To ensure that service user participation is not tokenistic, Browne et al (2015) recommend that the power on interview panels is equally shared between health-care professionals and people with personal experience. Furthermore, the BPS recommends that, to enable service users to offer meaningful input, they are consulted in the review of admission processes (BPS guidelines, 2008).

Background

At the university of Bath there has been involvement of PPE from inception of the DClinPsych. To be a PPE one needs to have either personal experience of mental health difficulties or be a family member/carer of someone who has had mental health difficulties. PPE help to deliver teaching, consult with trainees on research studies, and sit on various committees. There is currently a PPE committee that is attended by 20 people. The

committee is made up of academic staff, trainees and PPE. The committee meets every 3 months to discuss research, teaching, conferences, placements and admissions.

In 2019 there were 2 major changes to the University of Bath's DClinPsych admissions process (Table 2.2). With these two changes the course team wanted to assess the PPE involvement, with a view to utilise their feedback to continue to improve and streamline the admissions process.

Table 2.2.

Changes Made to Bath DClinPsych Admissions Process in 2019

Change	Description of change
PPE on interview panels	PPE representation on admission interview panels. PPE involvement in admissions had previously been limited to the admissions committee and as part of a presentation to the trainee candidates during interview days.
One interview instead of three	Streamlining the admissions process so all candidates are seen by one interview panel. The interview panel consists of three of the major stake-holders: a member of the course staff, a regional supervisor and a PPE. Prior to 2019 candidates were interviewed twice, by a different panel each time (consisting of a member of the course staff team and two regional supervisors).

Aims and objectives

This project aimed to explore the PPE experience of being part of the DClinPsych interview process and make recommendations for the admissions process the following year.

Method

All PPE representatives (n = 6) were invited to take part in a focus group, conducted by the lead researcher, to talk about their experiences. 4 of the 6 PPE took part in the focus group, the other 2 contributed via email, giving written feedback/responses to the focus group schedule. The focus group took place 15 days after the admission interview process.

Data analysis

The focus group was audio recorded using a digital recorder. The audio recording of the focus group was transcribed verbatim and analysed using the qualitative research methodology, interpretative phenomenological analysis (IPA). Although IPA was initially developed to understand experience from an individual perspective, the approach has been adapted and used with focus group data (de Visser & Smith, 2007; Dunne & Quayle,

2002; Flowers, Duncan, & Frankis, 2000). It has been suggested that whilst using IPA for focus group data can add complexity to the analytic process, the result can be an enhanced understanding of participants' accounts (Phillips et al. 2016). The four-stage approach for using IPA with focus groups by Phillips, Montague and Archer (2016) was followed (Table 2.3). In order to enhance the trustworthiness of the research (Korstjens & Moser, 2018) a second researcher, who was not involved in the DClinPsych, took part in the analysis process. The second researcher familiarized herself with the focus group transcript and worked through the 4-stage process (Table 2.3.) independently from the primary researcher. Interpretations were then compared and discussed; a high level of agreement was found.

Table 2.3.

Four-Stage Approach for Using IPA

Stage of analysis	Description of stage
Stage 1: Looking for "groupness"	Searching through the transcript for examples of participants responding as a group compared to individual accounts.
Stage 2: Clustering reoccurring group interactions	Pulling together patterns of data identified in stage 1 that reflect "re-occurring group interactions while also contextualising individual experiences within broader patterns of relationships." (pg 294)
Stage 3: Identifying interactional relationships	Linking the clusters of data together to create a "conceptual model of the relationships between the different types of interactions that occurred in the focus group" (pg 295)
Stage 4: Incorporating group elements into an analysis	Using the conceptual model to "add to our knowledge if the topics under investigation, to develop analytic themes, and to link to psychological concepts" (pg 296)

Recommendations were also extrapolated from the data, which were then fed back to the university interview committee. The summary of recommendations and subsequent responses from the service can be found in Table 2.6.

The Researcher's Perspective

The principal researcher

At the time the focus group was conducted the principal researcher was 39 years old. The researcher is a Caucasian British male from a middle-class background. The researcher was a qualified Health Psychologist who was also a trainee on the DClinPsych. During the project the researcher had a dual role of "trainee" on the doctoral course and "evaluator" of an aspect of the DClinPsych. It is recognized that there was a possible power imbalance between the researcher and the PPE, it is therefore probable that this had an impact on the data, both from what was shared by the PPE and also in terms of the particular lens/lenses through which the data was subsequently understood. In an attempt to mitigate the possible bias that this may have on the results a second researcher was consulted who carried out a secondary analysis of the data, which was then compared to the main analysis. The second researcher had no affiliation with the clinical psychology course at Bath and was an assistant psychologist in the local area with a particular interest in PPE research.

The second researcher

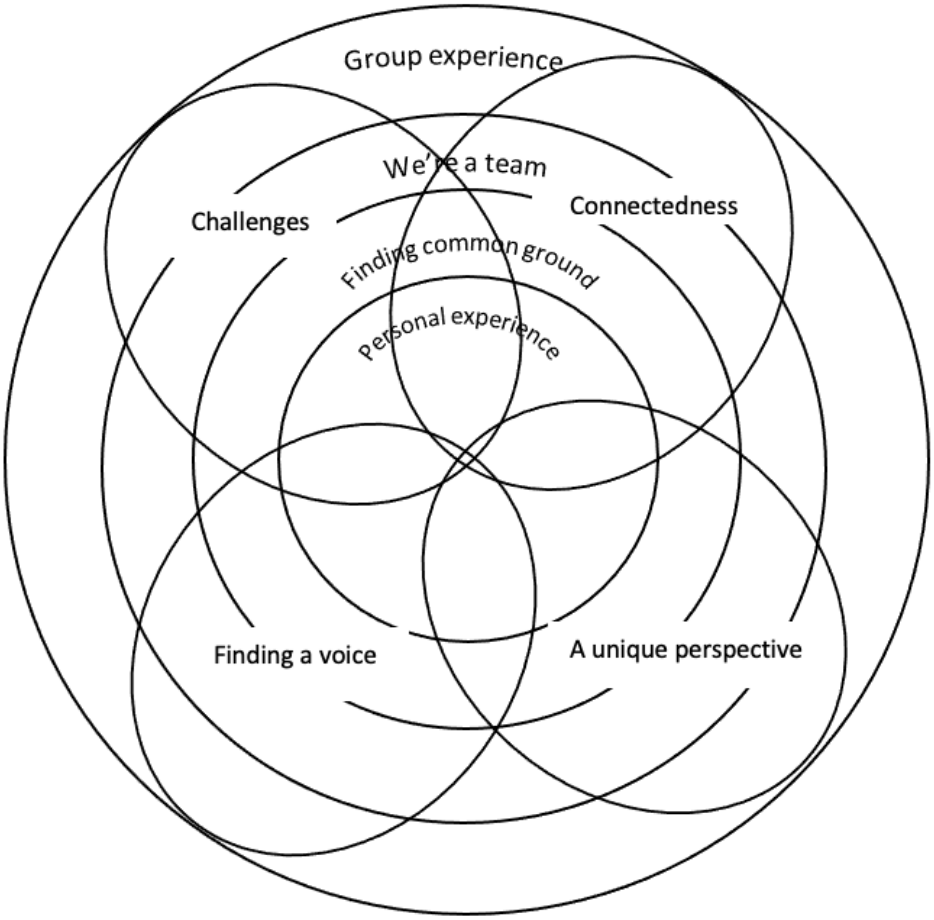
The second researcher is a Caucasian British female in her 30's. She was an assistant psychologist in a local mental health context. She had an interest in qualitative methodology and PPE advocacy, having been involved in previous research in this area.

Results

Four themes reflect the experience of the PPE as part of the interview process. The themes intersect multiple levels of experience (Figure 2.1). These "levels of experience" are distinct from the themes and should be considered as lenses through which one can understand and view the themes.

Figure 2.1.

Themes with Intersecting Levels of Experience



Levels of experience

Because the focus of the research was on lived experience, an IPA methodology was used. Over the course of the focus group there was a subtle shift from personal experience to group experience, during the analysis process this became understood and labelled as "levels of experience" (Table 2.4). "Levels of experience" is not a theme, it would be better understood as a framework that the themes are built around and it fits with the idea of "groupness" that Phillips, Montague & Archer (2016) refer to in their 4-stage approach to IPA. Levels of experience are differentiated by the use of pronouns such as "I", "me", "we" and "you"; it was also characterised by participants agreeing with each other's points and complimenting each other.

Table 2.4.

Levels of Experience

Levels of Experience	Description
Personal experience	Exemplified by the use of "I" pronoun and comments such as "I think", "I felt/feel" and "for me".
Finding common ground	The level of experience that links personal experience to shared experience. This can perhaps be interpreted as participants recognising themselves in each other. Commonalities in individual experience are highlighted by participants' frequent use of agreement and expanding on each other's points.
We're a team	The level of experience described as "We're a Team" is typified by the participants' use of the pronoun "we", denoting group membership. The "in-group" of the PPE is also contrasted by use of "them" and "they" to refer to the "out-group" of the trainees.
Group experience	Characterised by the use of the pronoun "you", suggesting that the participant is speaking for the group, and as the group.

Themes

The four themes are; Challenges; A Unique Perspective; Connectedness; Finding a Voice. The following is an exploration of each of these themes, along with illustrative quotes from the participants.

Challenges

The PPE talked about the challenging aspects of being a PPE. They acknowledged that being a PPE is not easy but were all able to reflect on the challenges. Challenges were talked about on a personal level exemplified by the use of "I" pronoun, for example Esther said the following, "I had no preconceptions because I was terrified", whilst Claire said:

“the first day I was so nervous, so like, anxious, and thinking this is like people’s future, [B: yeah] and it’s kind of like I’m in that decision to [B: yeah] you know I could sort of, help somebody or now, you know, and that was sort of a bit nerve-wracking!” (Claire)

The impression given is that the PPE are already experiencing a heightened level of anxiety coming into the interview process, in addition to this the candidates’ anxiety also permeates, further increasing the PPE’s anxiety. Beth’s experience exemplifies this, “you're managing people's nerves a lot you know, and there's a lot of anxiety and emotion and I think just absorbing that is very very draining”. Beth widens the narrative by using the pronoun “you”, suggesting that challenges are a shared experience. To make further sense of the feeling of heightened anxiety that the PPE reflect on within the focus group, the discomfort expressed by Claire can be considered, “for me it felt a bit uncomfortable and I wondered why that was and thought about it and thought “oh yeah it’s just a bit exposing”. The following quote from Beth is also helpful in understanding the heightened anxiety experienced:

“the feeling of feeling a bit exposed and kind of thinking ‘you know an awful lot about me, but I don't know anything about kind of your personal stuff’ and it's, it's just very weird dynamic and it's quite, because you have to be very vulnerable with, you know, your psychologist” (Beth)

Beth and Claire are describing the experience of having one’s personal life laid bare when seeing a psychologist. Of note is that when talking about this experience Beth again uses the pronoun “you”, indicative of a group experience, which can also be interpreted as a “stepping back” from the process and reflecting on experience in the moment. This also speaks to the strong bonds and group identity that seems to have developed.

The PPE talked about involvement in the PPE process as part of the journey moving forwards with recovery. There was a sense that being a PPE was an achievement and a way to make sense of negative experiences in the past. There is also a narrative about being in the “right place” in the journey process.

“It is a journey, to be able to reflect, whereas if I'd come into this and I was still in my fight to get the therapy that I badly needed six years ago it would be very natural to feel anger at "the service" (Esther)

Esther's use of the word "fight" and her reflection on how there might be a sense of anger if the PPE was still engaged in that fight, gives the impression that the provision of therapy is by no means a forgone conclusion, rather it is something that has to be worked hard for, even fought for. This gives the sense that the PPE feel that they are coming from a place of vulnerability, they have felt disempowered and marginalised, both by society and their own self-belief. The following quote encapsulates this, and perhaps also hints to the PPE now being in a place where they have built up resilience:

> *"You're coming from a place where you, you do struggle with your mental health and you, there's a lot of stigma that goes along with that, but when you're actually able to use your experience in a way that's helping others it's just so, it's so, it's so amazing."* (Claire)

A Unique Perspective

The PPE reflected on their own experience of mental health services and being able to appreciate that in a unique way, in effect they have an "insiders" view because of their lived experience. For example, Esther said the following: "professional psychologists who have changed my life, it has been that powerful, as Beth said again it's sort of a ripple effect".

The PPE reflected on how the right choice of psychology trainee could have a positive "ripple effect" in the future, as highlighted by Beth: "The impact that person could have in their career, in their lifetime, they would see you know, probably hundreds, possibly even thousands of people in their kind of lifetime and impact all those people".

The PPE also talked about finding themselves in a position of power whereby they can decide who does and doesn't get through to clinical training; Delia refers to how the impact of this process can cause a cultural shift in clinical psychology:

> *"there's less sort of 'them and us…if people feel that they're having an input and having a voice, I think those, we will start learning together and moving on together and there will be less hopefully feeling of 'them and us'"* (Delia)

The PPE know what they want in a psychologist, they know what feels right, and they talk about personal responsibility; they can make or break the future of these trainees and they are wanting to get it right. Claire reflected on the weight of this responsibility: "I think 'ooh, did I make a mistake?' or whatever, that sort of responsibility was quite, yeah, it was quite big for me". The PPE talked about an innate sense of who would be the "right"

sort of trainee. This was often referred to as a sort of sixth-sense that those who have been on the receiving end of psychological support had: “I felt the benefit of the PPE system, I would just say it brings that, and it was really simplifying it, it brings that very human level, that sort of intuition” (Esther).

Connectedness

This theme gives a sense of the PPE feeling that they have strong ties to each other and also felt that there was a connection between the other interviewers, who were primarily clinical psychologists. The theme of connectedness came through very much in terms of how the participants would often agree with each other about their experience, for example Beth said, “I would agree with kind of both you and Delia and Esther just being blown away by the breadth of work people are doing”. By agreeing within the focus group, there was a sense that the PPE had found common ground with each other. This can perhaps be interpreted as participants recognising themselves in each other. Commonalities in individual experience are highlighted by participants’ frequent use of agreement and expanding on each other’s points:

E: I think that would be for everyone as well, specialists as well, perhaps somebody who comes in presenting what seems, you know, quite an innovative idea in say child psychology and they happen to be a child psychologist [multiple: yes] I think they would have to also perhaps restrain themselves professionally from enquiring further [B: yeah] you know because it would be a professional interest [B: yeah] I think that’s probably the same for everyone, not just yourself.

A sense of connectedness was also expressed through the use of compliments, suggestive of a group identity typified by mutual respect and support:

C: so coming to do this was my first ever experience of PPE involvement in Bath

B: Yeah that was really brave! Really brave!

E: Well I was absolutely terrified, and it’s also the first time since I had to stop working so it was quite a milestone for me

C: mmm, same here.

Compliments show that the PPE value each other. By valuing each other, by having this group process, they are also giving themselves value and allowing themselves to recognise their value and group.

The “Connectedness” theme also refers to the sense of connectedness that participants feel by having a PPE identity, Esther reflected on this saying “It's really reassuring that you know you're not out there on your own!” Participants also talked about feeling connected to the other interviewers, for example Delia said “I thought we jelled very well as a team actually”. There was a sense that the participants felt a feeling of equality during the interview days, exemplified by the following quote from Esther, “they all stood around chatting at lunchtime, that was very levelling, it's things like that, so very simple things, but incredibly meaningful”. There was a sense of connectedness when the participants began talking from the “we’re a team” level of experience. Beth described how “we’ve been through the criteria, you know thinking it through for, you know, years, and you know changing it a little bit as we went along”.

There are two distinct “we”: firstly, participants talk about working together in subgroups (the interview panels), in this case they are together with psychologists in an unusual situation as they may not have experienced this previously, they may have felt more like “us and them”. Secondly the participants also talk about the “we” of being the PPE.

Finding a Voice

This theme is typified by the sense that being a PPE is empowering, gives value and is life changing. Participants often talked about the PPE process as being key to them reclaiming their “lost voice”. For example, Claire spoke of her experience “from a service user point of view it's empowering and being able to have a voice as well in who's coming through on the system”. Esther was positive about the experience saying that “it was incredibly inspiring, as someone who's never really, no one's really taken any value in my views”. Similarly, Beth said the following, “I think for me, again having it valued and having my voice heard and my experience seen as equal, that was utterly transformative to me”. Delia reflected that, “if people feel that they’re having an input and having a voice, I think those, we will all start learning together and moving on together and there will be less hopefully feeling of ‘them and us’ ”.

Esther’s and Beth’s comments touch on a wider narrative, woven through the focus group, regarding the disempowering and silencing nature of mental illness. In the following quote Claire refers to a societal stigma that relates to mental illness:

“I lost my job and thought "I'm on the scrap heap now" you know, there's still an element of that but you know going forward it's this impact that I'm making, turning my experience into a positive experience but also knowing that I still have value, I mean I've lost my job but that's not the end of the story… You’re coming from a place where you do struggle with your mental health and there’s a lot of stigma that goes along with that, but when you’re actually able to use your experience” (Claire)

For Claire there was a sense that she considered her professional life to be over, that she had been discarded on a “scrap heap”, a powerful image evoking thoughts of mental health problems breaking you in some way. Claire now feels she is at a point where she acknowledges the loss of her previous job and she can also have a sense of optimism; this is shown by her saying her “story” is not yet over. Claire also refers to being able to “use your experience”, which seems to be about the PPE finding value in what has often been very challenging and difficult experiences.

The participants talked about the empowering effect of being part of the PPE process, that it allows for the lived experience, which may have been very challenging, to be used in a positive way.

“to feel like you can use that to help other people and help try and minimise the awfulness of their illnesses [C: and also yourself as well] and suffering it massively impacts on how you feel and as you say it’s really empowering” (Beth)

Of note here is the use of the pronoun “you”, suggestive of a shared experience, the fact that the word “empowered” is used by multiple PPE also strengthens the sense of a shared experience.

Service Recommendations and Response

Based on themes presented in the previous section, a summary document was produced which outlined what the PPE felt went well and what would be recommended for the following year's interview process (Tables 2.5 & 2.6). This was fed back to the admissions committee and their responses are presented in Table 2.6 in red type.

Table 2.5.

PPE feedback regarding what went well

What went well
Having plenty of time to read through the application forms in advance of the interview day.
The interview training day was very well prepared and comprehensive.
The whole process of deciding on which candidates would be offered a place felt very democratic and the PPE's felt that "everyone's voice was heard". This was important and links to an overall finding from the research that reflects the experience of "having a voice and feeling valued".
The PPE felt very welcomed by the course team throughout the process. There was a sense that they felt "held" and supported by the course team and their individual panels.
Being told by the chair of the interview panel that the PPE view was just as important as the views of the other panel members. The PPE felt that there was a real sense of "levelling" in the panels – that they were all equal.
Being given the option to collect candidates from the "holding area"
Feeling like an important part of a wider process. Picking the "right" sort of candidates and thus having a knock-on effect for the future of psychological practice in the UK.
Daily debriefing was seen as helpful and essential.
The admin team's "amazing" organisational skills with all the paperwork – this was particularly helpful in reducing overall stress/anxiety levels.
Being able to have short breaks outside

Table 2.6.

PPE Feedback Regarding Changes, Recommendations Made and Service Response

What did the PPE think could be changed/adapted?	
Suggested changes/feedback	**Recommendations on how this could be addressed/implemented and response from service (in red)**
The PPE felt that it would be helpful to have a brief introduction of the PPE at the beginning of the day. This would be particularly helpful for the PPE who will be circulating throughout the day.	Perhaps a member of the course team could provide a brief introduction on each interview day, explaining who the PPE are and that they are more than happy to talk with candidates through the day, and answer any questions. **Not possible this year due to online process but can definitely be taken forward for next year. We can also make this clearer in the interview info sent out to candidates.**
The PPE felt that it would be helpful if they were able to have a list of the acronyms and initials that are used for various therapies, services, posts and positions that the PPE could refer to when looking through applications and also on the interview days (e.g. DBT = dialectical behaviour therapy; HCA = Health Care Assistant; AP = Assistant Psychologist; CAMHS = Child and Adolescent Mental Health Service).	Could the PPE be provided with a sheet of common acronyms and initials on the interview training day. Also make it clear that this list is by no means exhaustive and encourage the PPE to ask the course team and/or chair in the interview room if there are any terms that they are unsure as to the meaning. **Yes, I think one has been drawn up for PPE generally. I will make sure this is re-circulated and discussed to update**
More guidance on how to make notes from the candidate application forms, particularly in terms of what is permitted (e.g. that highlighting or making notes on the application forms is ok).	Could this be explicitly talked about as part of the interview training day, or be in written guidance in the training packs? **This was discussed in interviewer training, although paper forms were not able to be provided this year.**
Being a part of the interview process can be physically and emotionally draining, the PPE all talked about feeling overwhelmed at various times. It was felt that more opportunities to debrief would be helpful.	Debriefing for interview panel members already occurs, but it would be beneficial to open this up to the PPE members who are circulating during the interview days. Even having opportunities for more "informal" chats through the day with course staff could be beneficial. **During this cycle we added a 'reflective slot' for PPE interviewers at the end of the day, which was facilitated via teams by our lead PPE. This lasted 1-2 hours each day and was well-received.**
The PPE reflected on the issue that during the interview training and interview days there is a real possibility that they could come across either their own psychologist, or a psychologist who knows or supervises their psychologist. It was commented that this could be uncomfortable and/or feel exposing.	Is this something to discuss further at the PPE committee? Perhaps the PPE could be informed that, should they have any worries or concerns, they can speak with a member of the course team at any point during the training and interview days. **We now make this very clear to PPEs and they have the opportunity for additional debrief or to speak to the admissions team at any point.**

Discussion

Despite there being PPE involvement in various aspects of the doctoral admissions process at all the UK universities that offer clinical psychology doctoral training, there are only a small number of research studies that look at the experience of PPE in this process (e.g. Smith et al. 2020; Matka et al. 2009; Vandrevala et al. 2007), the majority which comes from nursing. The current research, therefore, expands on the limited amount of research into PPE's experience of being part of the admissions process for trainee clinical psychologists. The aim of this study was to explore the PPE experience of being part of the DClinPsych interview process and make recommendations for the admissions process the following year. The themes were: Challenges, A Unique Perspective, Connectedness and Finding a Voice.

The BPS recommends that PPE should be enabled to offer meaningful input to the selection process of clinical psychology trainees (BPS Guidelines, 2008). The PPE who took part in the current research certainly seemed to have felt that they were able to offer meaningful input as reflected in the themes A Unique Perspective and Finding a Voice. Smith et al. (2020) reported similar experiences regarding service users feeling that they had been given a voice through the process. In the current study, the sense that the PPE had found their voice through the process was powerful, and perhaps this process continues and is also part of this research; the focus group itself could be seen as furthering the sense of giving voice to a marginalised group, the impetus is to make sure that PPE involvement is not tokenistic and ideally the message conveyed to the PPE is that their involvement and their input is important and appreciated. As part of the process of conducting the current study the PPE feedback was fed back to the course team with the hope that the recommendations would be acted on.

The feelings of empowerment and being valued reported by the PPE in the current study supports the assertion made in the Good Practice Guide by Adamson and Howe (2015) that involvement in recruitment increases PPE "confidence and self-esteem through taking a meaningful role in the process" (pg 6). Increased self-confidence, feelings of empowerment and a sense of being valued are also experiences that are reported by PPE in previous research (Masters et al. 2002; Forrest et al. 2000; Rhodes & Nyawata, 2011; Vandrevala et al. 2007; Matka et al. 2009; Smith et al. 2020). Similar to the current research, Smith et al. (2020) in their study looking at both trainee and PPE experiences of

being involved in the Leeds clinical psychology doctoral course, report that PPE felt appreciated and supported by the doctoral course, as was being able to break down “us & them” attitudes. Similar sentiments were shared by the PPE in the current study, “Giving back” is reported, which is similar to how the current PPE talked about the ripple effect of participation. Matka et al. (2009) sought views of PPE, practitioners and students regarding the involvement of PPE on interview panels for social work and clinical psychology trainees. They report a view of professionals on interview panels reporting that PPE involvement helped to confirm their own views about the suitability of candidates and also add the “personal perspective”, this is echoed in the current research within the theme “A Unique Perspective”.

It is clear that the PPE’s personal perspective plays a pivotal role. Rhodes & Nyawata (2011), in their research looking at PPE involvement in trainee nurse recruitment, report that PPE’s talked about having a unique perspective on what would make a ‘good nurse’ and reflected that this was due to having had many years’ experience receiving healthcare. The PPE in the current study also talked about being at the right point in the recovery journey, there are similarities in this finding with previous research. Ingham (2001) explored the PPE perspective in nursing education and found that PPE felt that an individual would need to have “come to terms” with their condition and “not be angry” in order to make the most productive contribution. This is echoed in the current study by Esther’s comment about being angry when she was engaged in a fight for treatment.

Similar to the findings of the current study, Smith et al. (2020) also report on the demands that PPE discuss with regard to their involvement. It is reported that participants in that study felt the pressure of making the “right” decision as it was perceived that this would have a large impact on the interviewees’ futures. Smith et al (2020) also describe a feeling of vulnerability reported by some participants, which is echoed in the current study. Though in contrast to the current study, Smith et al (2020) report that some of the PPE felt “ignored or patronised” by the interviewees, this is not something that the PPE in the current study talked about. Rhodes & Nyawata (2011) found that PPE felt that preparation for the interview process was “rushed and not detailed”, in contrast to the PPE in the current study who felt that everything had been done to help prepare for the process. These contrasting findings may be because the researcher was a current trainee on the course, and the PPE knew that the findings would be “reported back” to the course,

however there were also comments that were more of a constructive critique. It therefore felt like PPE were able to be open and honest about their experience.

Matka et al. (2009) reported that although PPE involvement was seen as generally positive, however it is noted that amongst practitioners, there was some concern as to differences of opinion regarding candidates and it being unclear how this could be resolved. In the current study differences in opinion were mentioned by the PPE, though this seemed to be positive in terms of being able to reach a resolution. In the current study the clinical psychologists on the interview panels were not consulted and therefore their view on this matter may be different, though this is perhaps something to pursue in future research.

Limitations

There were some limitations to the current study, not all the PPE who were involved in the interview process were able to attend the focus group, though they did subsequently provide some written feedback based on the focus group topic guide. This feedback echoed what had been discussed in the focus group. The PPE were all female, including the 2 who were unable to attend. It would be interesting to see if a mixed sex cohort, or even a male cohort would express the same sentiments.

The researcher's position as a trainee on the course could have had an impact on the particular narrative that was shared in the focus group. The participants may have felt that they could not be completely candid as the researcher would be feeding back to the admissions committee. With this in mind it may be helpful for PPE to have the opportunity to provide anonymous feedback in the future.

Conclusion

The purpose of this study was to explore the experience of PPE as they became part of the DClinPsych interview process at the University of Bath. An adapted form of Interpretative Phenomenological Analysis (IPA) was used to analyse the responses of the 6 PPE representatives who took part in the focus group. The PPE described challenges of being part of the process, they offered a unique perspective, they highlighted the connectedness of the process and they explained that they felt they had "found their voice" through being a PPE. Recommendations were made to the course team regarding how the interview process could be changed and/or adapted, based on the responses from the PPE. The course team responded positively to these recommendations. In terms of implications for future research, there are further questions that the current research has brought up in terms of

the themes, in particular further research could explore in more detail the unique perspective of PPE, and what aspects of potential trainees may be identified by involving PPE in selections, that might be missed if only clinical psychologists are on the panel.

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Main Research Project:

The transition from paediatric to adult healthcare with Oesophageal atresia/trachea-oesophageal fistula (OA/TOF). A qualitative study exploring the experiences of young people, adults and parents.

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Abstract

Aims and objectives

The overall aim of this study was to explore the experience of healthcare transition from paediatric to adult healthcare for adults' born with OA/TOF and parents. A particular focus was how the different paediatric and adult services were experienced, whether parents and adults experienced a change in their roles during healthcare transition and also to identify challenges faced as part of the transition.

Background

OA/TOF is a rare chronic condition that can require lifelong medical support. There is evidence to suggest that transitioning from paediatric to adult healthcare can be problematic with OA/TOF, a finding that appears to be true in both rare and chronic conditions. The literature on healthcare transition is saturated with a common chronic illness narrative and there is a suggestion that the experience of transitioning with a rare condition is worse than transitioning with a common chronic condition. We currently don't clearly know why this might be the case.

Design

A qualitative, cross sectional, survey-based study.

Methods

Unrelated parents (n=23) and adults (n=21) were recruited through a UK based OA/TOF charity. Data were collected through an online survey. Data from six open ended questions were analysed using a six-stage hybrid approach combining elements of inductive and deductive thematic analysis (Fereday & Muir-Cochrane, 2006).

Results

Five themes were constructed reflecting the experience of parents and adults transitioning from paediatric to adult healthcare: Thrown into the unknown; A cultural shift; Stepping back and stepping up; "No transition as such"; Living with uncertainty.

Conclusions

The findings suggest that a formalised, managed healthcare transition is practically non-existent for people born with OA/TOF.

Relevance to clinical practice

We recommend a formalised healthcare transition process in OA/TOF. This should involve a period of preparation to get adolescents and parents ready for transition. It would be

beneficial for those working in adult services to receive additional training regarding issues that adults born with OA/TOF may struggle with. It would also be helpful for families to have a named keyworker to support them through the process of transition.

Introduction

Oesophageal atresia (OA), which can present with or without trachea-oesophageal fistula (TOF) are rare conditions whereby a baby is born with a food pipe that does not connect to their stomach and/or the lower part of the food pipe is connected to their windpipe. OA/TOF occurs in approximately 1 in 3500 live births (Losty & Thursfield, 2020), there are no known gender or ethnicity differences (Beasley 2016). Once a condition with a high infant mortality rate (Deurloo et al. 2005), in the 1940's advances in surgical techniques meant that infant survival became much more likely (Bae et al. 2005) and survival rates into adulthood are now estimated to be >97% (Losty & Thursfield, 2020). For individuals born with OA/TOF, infancy and early childhood is characterised by complex hospital-based care (Zamiara et al. 2015). Health related issues can continue to impact on an individual's quality of life throughout childhood and adolescence (Peetsold et al. 2010) requiring frequent review and investigations (Roberts et al. 2016; Rintala et al. 2009). Once reaching adulthood, perception of general and disease specific quality of life is generally reported to be positive (Ijsselstijn et al, 2013; Ure et al. 1998; Koivusalo et al. 2005). However, some will continue to experience negative consequences from their OA/TOF (Deurloo et al. 2005; Connor et al. 2015; Roberts et al. 2016; Ten Kate et al. 2020). The most common health related issue is dysphagia (difficulties swallowing) (Connor et al. 2015; Mousa et al. 2019), which means that eating is often challenging (Caplan, 2013; Rozensztrauch et al. 2019; Ten Kate et al. 2020). Gastro-oesophageal reflux disease (stomach acid passing into the food-pipe) is also common, as are recurrent lung infections (Dingemann et al. 2014; von Allmen, 2017; Koivusalo et al. 2005; Connor et al. 2015; Rintala et al. 2009; Mahoney & Rosen, 2017; ten Kate et al. 2020) and respiratory symptoms (Gatzinsky et al. 2011), all of which require ongoing management (Smith 2014). There is also evidence that adults born with OA/TOF and their family members can struggle with ongoing psychosocial difficulties throughout life (Love and Morice, 2012; Caplan 2013; Faugli et al. 2009; Rozensztrauch et al. 2019; ten Kate et al. 2020; Losty and Thursfield, 2020). Given the evidence that, for some, OA/TOF related difficulties will continue through

adolescence and into adulthood, transitional care and ongoing adult follow-up is imperative (Dingemann et al. 2017; Ijsselstin et al. 2016; Roberts et al. 2016; Connor et al. 2015; Svboda et al. 2017; Krishnan et al. 2016; Mousa et al. 2019; Koumbourlis et al. 2020). However, transition to adult healthcare is typically experienced as problematic. Perhaps, in part because comprehensive paediatric care is followed by simple follow-up by a GP. This becomes problematic and might be experienced as a lack of support, particularly when GPs have little or no training in such a rare condition (Ten Kate et al. 2020). With the exception of the recent paper by Ten Kate et al. (2020) there is a lack of research that explores transition issues in people born with OA/TOF (Krishnan et al. 2016).

Background

Healthcare transition has been defined as "the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-oriented health-care systems… to provide health care that is uninterrupted, coordinated, developmentally appropriate, psychologically sound, and comprehensive." (Blum et al. 1993. Pg 570). Interest in healthcare transition has increased over the past 30 years (Straus & Brown, 2019), the main reason being that survival rates of many life-limiting and life-threatening conditions have improved (Fraser et al. 2012). Increased numbers of adolescents are therefore "graduating" to adulthood and consequently healthcare transition is a "global health and social care issue" (Kerr et al. 2020). Indeed, the importance of healthcare transition has been highlighted in the NHS Long Term Plan (2019). In particular, it is stated that there is an impetus to "move towards service models for young people that offer person-centred and age-appropriate care for mental and physical health needs, rather than an arbitrary transition to adult services based on age not need" (NHS, 2019. pg 55).

Life transitions are challenging (Rapely and Davidson, 2010) and this is no less true in the transition from paediatric to adult care. This particular transition typically occurs during the time when an adolescent strives for autonomy from parental control. This process is described by Carter and McGoldrick as a normal stage of the Family Life Cycle theoretical framework (Carter and McGoldrick, 1989). Carter and McGoldrick describe family stress during times of transition as "emotional disequilibrium". The interaction of various systems and how they work together to support or impede one another are the key determinants of how well the family will manage transition (McGoldrick et al. 2013).

Additional stress can be caused if individuals are unable to adapt and achieve the expected development between stages.

Research literature regarding common and rare chronic diseases have highlighted the importance of formal healthcare transition (HCT) programmes (Crowley et al. 2011; Fegran et al. 2014; Dellon et al. 2013) and reported positive outcomes (Schmidt et al. 2020). It is recommended that transition programmes are flexible, individualised and developmentally appropriate (Rosen et al. 2003; Dellon et al. 2013). The level of patients' knowledge and skills should be assessed and supported, autonomy enhanced and independence and patient responsibility encouraged (Rosen et al. 2003; Staa et al. 2011; Lugasi et al. 2011; Dellon et al. 2013) whilst not undermining parental involvement (Staa et al. 2011). In the absence of formal healthcare transition, communication between families and healthcare providers can be disjointed (Dellon et al. 2013) and transition failure can risk disengagement in services (NHS long term plan, 2019), non-adherence to treatment, loss to follow-up (Lugasi et al. 2011; Dellon et al. 2013), adverse social and educational outcomes (DOH, 2006) and further consequences for the whole family (Waldboth et al. 2016). The Quality Care Commission has identified that good examples of positive transition planning exist in the areas of cystic fibrosis, diabetes and cardiac care (CQC, 2014). However, despite some positive examples, healthcare transition tends to be "poorly planned, poorly executed and poorly experienced" (Cornish, 2015), often leading to increased distress in both young people and parents (Waldboth et al. 2016; Coyne et al. 2019; Fegran et al. 2014). Research into healthcare transition tends to lack quality and there is a suggestion that the field is within the exploratory stages (Betz et al. 2016). A key recommendation made by the Care Quality Commission in their 2014 report is that commissioners "must listen and learn from young people and families" with regard to transition (pg 6).

The lived experience of healthcare transition in chronic conditions has been the subject of a number of systematic reviews and meta-syntheses (Lugasi et al. 2011; Fegran et al. 2014; Waldboth et al. 2016; Coyne et al. 2019; Lerch and Thrane, 2019). Transition to adult services has been described as a "culture shock" (Coyne et al. 2019), an unsettling experience (Fegran et al. 2014) characterised by feelings of alienation (Lugasi et al. 2011). Transition planning varies within and across conditions (Coyne et al. 2019; Lugasi et al. 2011; Fegran et al. 2014; CQC, 2014). Patients and families often report a lack of

information regarding transition process and disease management post transition (CQC, 2014; Coyne et al. 2018, 2019; Rutishauser et al. 2014; Iversen et al. 2019). Patients and families also express concern that the frequency of consultations post transfer will be drastically reduced (Fegran et al. 2014). There is an expectation that, when moving to adult services, responsibility will shift from parent to young person (Waldboth et al. 2016; Coyne et al. 2018). However, it has also been reported that lack of clear role expectations of young people and parents through the transition process exists (Clarizia et al. 2009).

Patients and families frequently report strong emotional attachments to paediatric services and subsequently healthcare transition is often experienced as a loss (Coyne et al. 2019; Lugasi et al. 2011; Fernandez et al. 2014; Fegran et al. 2014; Rutishauser et al. 2014; Gray et al. 2017). However, in a recent systematic review by Waldboth et al (2016) transition as loss is not reported, suggesting that this may not be a universal experience. Waldboth et al. (2016) focused on rare diseases, where transition out of pediatric care might be less likely to engender feelings of loss and abandonment if those patients already, during childhood, experienced a minimal level of reliable ongoing care from specialist services.

The lived experience of transition differs with regard to parents and young people (Coyne et al. 2019; Betz et al 2016; Lewis and Noyes, 2013; Walsh et al. 2017). Parents' experiences of healthcare transition tend to be characterized by difficulty relinquishing control, worries regarding the perceived readiness to transition to adult care, feelings of loss and abandonment (Peeters et al., 2014; Fernandez et al. 2014; Akre and Sulis, 2014; Heath et al. 2017; Gray et al. 2017; Lerch and Thrane, 2019). Young people often want to be more independent, particularly because they might have felt infantilised by paediatric care (Lugasi et al. 2011). However, young people also report feeling unprepared and needing guidance and support regarding transition (Lugasi et al. 2011; Fegran et al. 2014). Differences in transition expectations can be challenging for both young people and parents, particularly when some parents might struggle to "let go" (Fegran et al. 2014). It has been suggested that parents have a fear of ''letting go'' because of limited confidence that their child can take care of themselves (Peeters et al., 2014).

In a recent review of systematic reviews, Hart et al. (2019) identified that common chronic conditions tend to be the focus of transition research, in particular type 1 diabetes. Rare conditions therefore represent a gap in the literature. Of the reviews referred to here,

only one focusses exclusively on what would be termed “rare” diseases (Waldboth et al. 2016), with the others including both common and rare diseases. The problem with including rare diseases in a review of common conditions is that they become subsumed into a broader narrative about chronic illness healthcare transition more generally. This risks losing sight of the specific challenges that may be unique to rare conditions (EURORDIS, 2009) such as diagnostic delays, lack of information, social isolation and stigma, healthcare service access difficulties (Anderson et al. 2013; Dwyer et al. 2014; Zurynski et al. 2008; Jager et al. 2015; Kole & Faurisson, 2009; Nutt & Limb, 2011; Molster et al. 2016; Rare Disease UK, 2010). A key finding from a report produced by Rare Diseases UK (2014) was that transition is better for those affected by more common conditions. Furthermore, there is a lack of qualitative research exploring experiences of living with a rare disease (Von der Lippe et al. 2017).

To summarise, OA/TOF is a rare chronic condition that can require lifelong medical support. There is evidence to suggest that transitioning from paediatric to adult healthcare can be problematic with OA/TOF, a finding that appears to be true in both rare and chronic conditions. The literature on healthcare transition is saturated with a common chronic illness narrative and there is a suggestion that the experience of transitioning with a rare condition is worse than transitioning with a common chronic condition. The gaps in the literature regarding the experience of transitioning with a rare condition and the experience of transitioning with OA/TOF will be addressed by the aims of this study.

The present study aims to investigate the experience of transitioning from paediatric to adult healthcare services from the perspective of adults with OA/TOF and parents of adults with OA/TOF. The objectives of this study were as follows:

- To explore parents’ and adults’ experiences of the different services during the healthcare transitioning process.
- To investigate whether parents and adults experience a change in their role during the process of transition from paediatric to adult healthcare services.
- To identify the challenges that are faced as part of the transition from paediatric to adult healthcare.

Method

Design

A qualitative, cross sectional, survey-based study was carried out. Qualitative methodology was deemed appropriate because of the focus on lived experience and the lack of research into transition issues in this group. An online survey was used to gain an insight into the experience of transitioning from paediatric to adult healthcare with OA/TOF. Survey research has been described as "a useful and legitimate approach to research" (Ponto, 2015). Survey methodology allows participants greater flexibility of when and where they take part, it is also possible for participants to be assured complete anonymity (Braun & Clarke, 2020).

Recruitment and participants

The study was advertised via the TOFS charity (Registered charity no: 327735. Registered company no: 2202260) closed Facebook group using a text advertisement and a short video. Purposive sampling was used to recruit Parents/carers of individuals with OA/TOF (>16) and adults with OA/TOF (>16) who were either in the process of transitioning to adult healthcare services, or who had already transitioned to adult services. Inclusion and exclusion criteria are presented in Table 3.1. A minimum sample size of 20 is recommended for survey based qualitative research (Braun and Clarke, 2020). A total of 23 parents and 21 adults born with OA/TOF were included in the study. Sample characteristics are presented in Table 3.2.

Table 3.1

Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria

- Parent/carer of an individual with OA/TOF
  o Individual with OA/TOF is 16 or over, who is currently, or has already transitioned from paediatric to adult healthcare services
- Young person with OA/TOF
  o Aged 16 or over and who is currently, or who has already transitioned from paediatric to adult healthcare
- The participant lived in the United Kingdom during the period of time that the child with OA/TOF transitioned into adult healthcare services.
- Ability to read and write in English
	- Parent/carer of a child with OA/TOF who has not yet begun to transition from paediatric to adult services
	- Young person who is under the age of 16.
	- Young person who has not transitioned from paediatric to adult services
	- Non-English writing

Table 3.2.

Demographic information for parents and adults with OA/TOF

Demographics	Parents (n = 23)	Adults with OA/TOF (n = 21)
Age		
16-25		11
26-34		4
35-44	2	1
45-54	10	4
55-64	8	1
65-74	3	
Sex		
Female	21	18
Male	2	3
OA/TOF sub-type		
OA/TOF	10	12
Long-gap OA/TOF	6	5
VACTERL	6	3
Other*	1	1
Child's age		
16-25	16	
26-34	4	
35-44	3	

Note = Other = Long gap and VACTERL, colonic interposition*

Data Collection

The survey was piloted with three parents of young people with OA/TOF, and one young person with OA/TOF. Data were collected through an online survey comprising 11 demographic information questions, one quantitative measure for anxiety and depression and six open ended questions. The six open ended questions were influenced by the the Family Life Cycle theoretical framework (Carter and McGoldrick, 1989) and were designed to reflect issues reported in the healthcare transition literature (Shultz, 2013; Zhou et al. 2016; Timmer et al. 2017; Heath et al. 2017). The six open ended questions are presented in Table 3.3. Data collection lasted for four months.

The lead researcher was a 40-year-old Caucasian male. He was a qualified health psychologist and current trainee clinical psychologist, with prior experience of various qualitative methodologies, particularly thematic analysis. He had no knowledge of OA/TOF prior to starting the research but had previously worked for 10 years in the area of Paediatric Health, specifically chronic fatigue syndrome. His previous doctoral research thesis explored the impact of chronic illness on families and was influential in his choice of current research topic.

Table 3.3.

Open Ended Questions, Parent and Adult Variations

Qualitative Questions				
Parent version	Adult version	Total number of words	Range of words	Mean number of words
If your child has already transitioned to adult healthcare, what differences have you noticed between paediatric and adult services?	If you have already transitioned to adult healthcare, what differences have you noticed between paediatric and adult services?	2853	7 - 334	65
Did you notice any changes in your role in managing your child's health condition as your child transitioned into adult healthcare services? What were these changes?	Did you notice any changes in your role managing your health condition as you transitioned into adult healthcare services? What were these changes?	1928	1 - 138	44
What are the challenges you faced as your child transitioned into adult healthcare services?	What are the challenges you faced as you transitioned into adult healthcare services?	2072	1 - 178	47
Please describe what has been helpful for you as a parent (or as a family) when coping with the challenges related your child transitioning to the adult healthcare services?	Please describe what has been helpful for you (or your family) when coping with the challenges related your transition to the adult healthcare services?	1367	0 - 150	31
Is there anything that you think would have been a helpful addition to the transition process?	Is there anything that you think would have been a helpful addition to the transition process?	1770	0 - 206	40
Is there anything else you would like to share about your experience of transitioning your child from paediatric to adult healthcare?	Is there anything else you would like to share about your experience of transitioning from paediatric to adult healthcare?	1358	0 - 177	31

Ethical considerations

Ethical approval was obtained from the University of Bath Psychology Research Ethics Committee (REF number 20-059; Appendix H). Full consent was gained; all data was anonymised at source by setting Qualtrics to "anonymise responses" mode (no identifying data and no IP address collected). Upon completion of the online survey, participants were presented a debrief page comprised of information regarding additional support via their GP or other support services, as appropriate. As no personal data was collected, it was not

possible to provide any additional individual aftercare other than the automated debrief information.

Data Analysis

The aim and objectives of the current research were to tap into and understand the lived experience of parents and adults with OA/TOF, as such the lead researcher decided that the research epistemology would be broadly essentialist/realist reporting the experiences, meanings and the reality of the participants (Braun & Clark, 2006). Research questions focussing on lived experience can be addressed using Grounded Theory (Strauss & Corbin, 1998), however the following reasons (Braun and Clarke, 2020) were considered as to why thematic analysis was more appropriate than grounded theory for the current research: the aim of the research was not that of theory generation, the goal of the research was to identify and describe patterns in the data and the sample was relatively small. Data from the six open ended questions were analysed using a six-stage hybrid approach combining elements of inductive and deductive thematic analysis (Fereday & Muir-Cochrane, 2006). A hybrid approach was chosen because, whilst there is a lot known about healthcare transition, there is very little known about healthcare transition experience in OA/TOF. For further details of the analytic process see Table 3.4.

A code manual (examples of code definition is presented in Table 3.5, the coding manual can be found in Appendix G) was developed with individual codes based on the Healthcare Transition Systems Dynamics Model (Hamdani et al. 2011). Twenty deductive codes were defined and described fully by the lead researcher. The use of a code manual provides a clear trail of evidence, increasing validity and allows for inter-rater reliability (Roberts et al. 2019). It is recommended that for testing code reliability 10-20% of the data should be assessed by an independent coder (Joffe, 2012), therefore the lead researcher and the secondary researcher coded the same selection of responses independently, then the coding was compared. The lead researcher then fully coded the whole dataset using the code manual. Inductive codes were created from the data when it was clear that there was divergence from the deductive codes, a further 14 codes were created in this way.

Analysis was undertaken by the primary researcher. In order to increase trustworthiness a subset of three parent and three adult survey responses were independently analysed by the second researcher. The early codes and themes were discussed, compared, and agreed, and the remaining transcripts analysed by the primary

researcher to explore common or divergent themes. The lead researcher coded the survey responses in Microsoft Word, the coded sections of text were then cut and pasted into Microsoft Excel where it was possible to sort the data according to code name. The lead researcher used the online tool Miro (Miro.com) to organise the grouped codes into a thematic map. To further enhance trustworthiness of the research findings the lead researcher kept a reflective diary, and the complete analysis and themes were discussed and clarified in research supervision meetings.

Table 3.4

Stages of the Thematic Analysis and Application of the Stages

Analysis Stage	Brief Description	Process
1. Developing a code manual	Production of initial codes.	The primary researcher produced a list of 20 codes, based on the Healthcare Transition Systems Dynamics Model. The codes were defined and described.
2. Testing the reliability of the code	Checking that the codes describe what they are meant to describe through a process of inter-rater reliability.	The primary and secondary researchers coded 10-20% of data, findings of both then compared.
3. Summarizing data and identifying initial themes	Summarizing the participant responses.	The primary researcher Made summary notes for each participant transcript regarding the key information. Initial ideas for themes were noted.
4. Applying template of codes and additional coding (mixing deductive and inductive coding)	Deductive coding using coding manual, indictive coding from the raw data.	The primary researcher used the coding manual to identify meaningful units of text within the raw data. The primary researcher was mindful of information which did not fit with the a priori deductive codes, leading to the assignment of new inductive data-driven codes.
5. Connecting the codes and identifying themes	Coded data grouped in order to construct the themes.	The primary researcher produced a thematic map by grouping together thematically similar codes.
6. Corroborating and legitimating coded themes	Data audit: making sure the raw data corroborate the themes.	The primary researcher checked the final themes against the participant responses to ensure that parents and adult experience had been accurately reflected.

Table 3.5.

Example of Code Definition from Code Manual

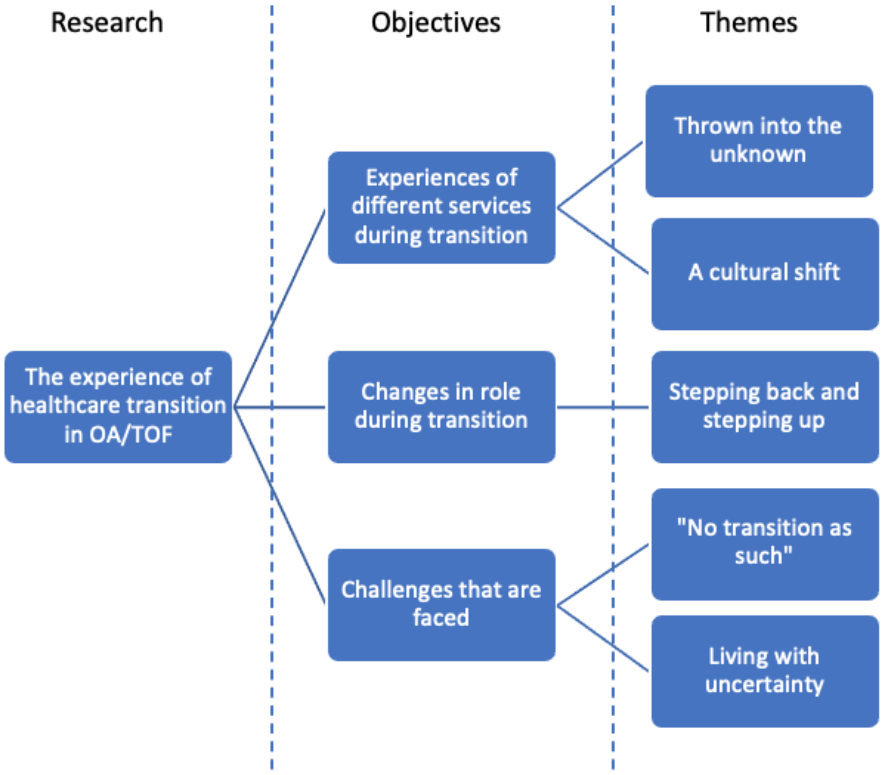
Code label	Definition	Description	Examples
Discharge from paediatric health services	An "official" exit from paediatric healthcare that is orchestrated by the healthcare provider.	The participant talks about being discharged from paediatric healthcare, or not – either way the word "discharge" is used.	*"he has actually been discharged from the paediatric hospital" (parent 3)* *"No specialist support after discharge from children's hospital care when my daughter became an adult." (parent 5)*
Communication and continuity of care	Communication between paediatric and adult services, and a level of care continuing from paediatric to adult healthcare.	The participants describe communication (or lack of) between healthcare providers. The participants refer to the care they received in paediatric and what happened to this when they moved to adult care.	*"No link/database across the country" (parent 1)* *"Now, my daughter's consultants are at two different locations and there is no communication between them" (parent 2)*

Results

The current study found, with the use of hybrid deductive/inductive thematic analysis, five themes were constructed reflecting the experience of parents and adults transitioning from paediatric to adult healthcare. The Five themes are as follows: Thrown into the unknown; A cultural shift; Stepping back and stepping up; "No transition as such"; Living with uncertainty. The themes are presented in relation to the research objectives in Figure 3.1.

Figure 3.1.

Themes Relating to Research Objectives



Thrown into the unknown

The first theme encapsulated the experience of leaving the familiarity of paediatric healthcare and entering the unknown world of adult healthcare. Parents and adults described the process as a sudden swift shock, from stability to instability, often leading to feelings of abandonment and fragmentation of care. The respondents had felt held by paediatric services, who they would have continued to see since birth. The feeling of being held by paediatrics ends at discharge and, particularly for parents, is experienced as abandonment. Respondent 16 (parent) explained that “we really felt we were abandoned by paediatric care.” This perhaps reflects the strong bond that parents have built up with paediatric services as their child has grown up. Feelings of abandonment are compounded by the sense that what comes next, i.e., adult healthcare, is experienced as woefully inadequate. For example, respondent 17 (parent) said “you feel abandoned as paediatric care is fantastic but adult care is zero”, whilst respondent 32 (adult) shared that they “didn't find helpful things whilst transitioning and we felt truly alone.”

There was a real sense that, for parents, leaving paediatric services was particularly difficult, perhaps even a wrench. For adults born with OA/TOF, there was less evidence of the type of emotive language used by parents. Adults tended to describe discharge from paediatric services in a more "matter of fact" fashion, for example respondent 33 (adult) stated that "I was just discharged by the paediatric surgeons and that was it." And respondent 40 (adult) said "I was discharged at 16 and haven't had any other medical care".

The move from paediatric to adult healthcare is frequently described by parents and adults with the use of the word "sudden", suggesting that the move to adult care was quick and there was little time for preparation, this is clear from the experience shared by respondent 24 (adult) "I felt it was a challenge going into that environment as it felt rather sudden". In a metaphorical sense, parents and adults feel like they have fallen off a cliff when they are discharged from paediatric health services, indeed respondent 1 (parent) describes the process using this very metaphor, "like falling off a cliff - from great specialism, experience & care in Paed into the mainstream/general of the NHS system". Respondent 7 (parent) shared a similar experience, "[…] and we felt as though we'd jumped off a cliff/crossed to another world". Also highlighted in these quotes is the sense that paediatric and adult services are completely different worlds, this is explored further in the next theme, "A cultural shift". Having "fallen off the cliff", participants described feeling that the "new world" of adult healthcare felt fragmented and thus led to confusion as to who bore responsibility for various aspects of their healthcare. An example of this comes from respondent 37 (adult) who stated that "you change to adult services and you're suddenly left with a GP who doesn't know you and none of those regular appointments either." Similarly, respondent 29 (adult) highlighted that there was a "lack of clarity around who should be responsible for my care." Respondent 24 reflected that they "would have benefitted from some private conversation with my consultant but not sure that's down to the service...more family relationships."

A cultural shift

This theme reflects the perceived difference between paediatric and adult healthcare. Parents and adults typically presented a positive view of paediatrics and a negative view of adult services. Exceptions did exist, though these were in the minority. Adults tended to describe a continuity of care under paediatric services, and consequently

felt that their needs were met. For example, respondent 39 (adult) explained “when I was seeing the paediatrician, I knew who I would go to if I started experiencing difficulties again.” The respondents expressed that, under paediatric care, they had been confident that support would be available should they need it. The same could not be said for the experience once in adult care. Contact with adult services was typically reported as significantly reduced in comparison to contact with paediatric services. Respondent 25 (adult) stated that “I have had nothing as an adult, was visiting paediatric once a year.”

A particular concern expressed by both parents and adults was the lack of knowledge of the condition amongst health professionals in adult services, particularly amongst GPs, who would typically be the adult’s main or only point of contact in adult services. This is highlighted by the experience of respondent 12 (parent) who explained “I have also found that GPs have no knowledge of the condition and yet the only way to get an appointment with a consultant is through a GP.” The perceived lack of OA/TOF knowledge amongst health professionals can be seen as part of a wider experience of lack of resources in adult care. Parents and adults described their experience of adult healthcare as typified by having no support, no point of contact. Some respondents went as far as to say that they had not found anything helpful in their experience of adult healthcare.

> *“There has been no change, I have been fobbed off all my life and passed from pillar to post seeing many different doctors, having to explain my condition as most have never heard of it and as I don't look sick there has never really been any support.”* (Respondent 43, Adult)

Respondent 23 (parent) provided a further example of lack of support, “there has been no real adult care for my TOF son, we rely solely on the understanding and knowledge of the condition from GPs -some seem to understand, some don’t.” Similarly, respondent 16 (parent) shared that “we have no specialist team to turn to when we have problems or concerns.”

Parents’ and adults’ descriptions of adult healthcare painted a picture of fragmentation. Respondent 20 (parent) referred to the “lack of specialism and joined up support” in adult services, whilst respondent 32 (adult) lamented that “the two departments need to openly talk and integrate and have dialogue with each other!” Parents’ and adults’ sometimes felt ignored or dismissed by health professionals, an example of this is from respondent 6 (parent) who explained that “I had to request a

camera [for an endoscopy], and at first was refused. My concerns were not important". Respondent 38 (adult) shared the following experience, "I was referred to a respiratory specialist after frequent chest infections aged 25 (now 28) but they suggested my TOF/OA was irrelevant."

Stepping back and stepping up

This theme reflects how parents and adults experience a shift in their roles when they move from paediatric to adult healthcare. Parents talk about taking a step back in their role managing their child's health, which they see as necessary but also challenging to do. Respondent 21 (parent) described this as having to "take a back seat", whilst respondent 14 (parent) described how "stepping back and trying to let the child take over is hard when they are less able to express feelings, needs and medical history". Although taking more of a back seat, it is evident parents continue to provide support to their child, should this be needed. For some families, roles are renegotiated so that both child and parent manage health issues, leading to a team approach, for example respondent 2 (parent) described how they were "always available to be a sounding board to [daughter]". Respondent 7 (parent) described how "my daughter and I have always discussed and dealt with everything together." Respondent 27 (adult) talked about how "mum and I much more responsible for knowing what symptoms are and the impact of them".

For adults with OA/TOF, a big shift in their role was being given responsibility for their own health. They described being in charge of their own health condition, to which varying degrees of confidence were expressed. For example, respondent 33 (adult) talked about being "in charge" in adult services and that this had been helped by their parents "already allowing me to make a lot of my own decisions." Whilst respondent 41 (adult) stated that they felt they were "really not good at managing my own medical issues." Being more responsible was generally seen in a positive light by the adult participants, though they also reflected on having to become the expert in their condition. Becoming an expert in OA/TOF is necessary because of medical professionals' lack of knowledge of the condition, referred to in the previous theme. Respondent 38 (adult) explained "to have my TOF/OA considered as a factor in my adult health, I have had to research myself and take this to the GP."

“No transition as such”

This theme reflects the finding that, with OA/TOF, adults’ and parents’ experience is typically that there is no formal transition from paediatric to adult healthcare. In some cases, there might not even be an official referral to adult services. For example, respondent 42 (adult) stated that “there was no ‘transition’ as such” and respondent 43 (adult) explained that “you can’t comment on something that didn’t exist”. Respondent 22 (parent) stated that “the main challenge was that there was no real transition”. Respondent 21 (parent) shared their frustration that they were “not being referred from paediatric [care]. I think we should have been referred so we had a point of contact.” This is not to say that adults born with OA/TOF are not able to access adult healthcare, more that the managed process of “transition” seems to be at best, rare, and at worst, non-existent. There was evidence that, in some cases, attempts at transition planning had failed.

> *“We had been told that there was a care plan for transition the year before it was due to happen, but for some reason they didn't action it and we got paperwork in the post, with no explanation, almost at the end of the transition.”* (Respondent 2, Parent)

Respondent 24 (adult) shared their experience of an attempt at transition planning, “I feel like some sort of transition to adult care book was provided but I don't really remember any of the information going in...to be fair...I was sixteen and probably had slightly more exciting things to be doing!” When one considers the previous themes, it is perhaps unsurprising that the process of leaving paediatric care is so traumatic if one considers that there is no managed transition process. Rather than transitioning to adult healthcare it seems that, for some, leaving paediatric care feels like the end of a journey. One parent believed that their child’s notes were “destroyed” prior to the move from paediatric to adult healthcare. More often than not the move from paediatric to adult care is reported as unsuccessful, with little to no planning in the run up to discharge. For example, respondent 15 (parent) shared the following experience, “No named consultant/point of contact. No regular check-ups. No one who knows about TOFs. No parental input allowed. Very poor transition experience.” Respondent 22 (parent) highlighted that “there is much to be done improving the transition to adult health care”, while respondent 32 (adult) stated that their “transition” was “a complete shit show.”

Living with uncertainty

Characterised by worries regarding living life with a chronic illness, this theme reflects the sense that the move from paediatric to adult healthcare presents additional concerns and uncertainties. For parents, there was uncertainty as to whether their adult children would have the skills to manage outside of paediatric care. Respondent 1 (parent) had particular concerns that their 17-year-old child "doesn't have, or take, much responsibility about his general life/care as yet." Respondent 21 (parent) explained that their child "didn't understand full history, date of ops, complications. He also didn't know how to describe or tell a good history, so wasn't getting correct treatment." Having had to become the expert in OA/TOF whilst raising their child, parents were also concerned that their child might not have the same level of knowledge about the condition that they have. An example of this concern is shared by respondent 17 (parent), "[…] yes as obviously now they contact my daughter. She doesn't know a lot of her background. She also struggles to explain things". Often parents expressed particular concerns about their adult child's ongoing health issues related to OA/TOF, primarily physical health issues but also mental health issues. This was a concern even if their adult child was currently in good health. There is a sense that there is something lurking in the background that raises parental anxiety. Factors such as lack of confidence or knowledge mean that parents have ongoing concerns about their child, and subsequently express protectiveness. For example, respondent 12 (parent) described how they "worry that my son may develop serious oesophagus damage which won't be picked up, which could lead to cancer." Whilst respondent 15 (parent) said the following, "especially when your teen is also depressed, suicidal, won't look after his health or get help for his health both physically and mentally." Respondent 11 (parent) added that they found that they were "constantly checking, asking, worrying" about their adult child.

For adults, living with uncertainty meant that they could not be sure that their health concerns would be addressed or even taken seriously in adult services. Experiences were shared whereby OA/TOF related symptoms have been "ignored" and "dismissed", leading to an increased sense of frustration. Respondent 30 (adult) described how "many consultants have completely ignored my symptoms for about 15 years which led to severe frustration." Whilst respondent 39 (adult) shared the following experience, "When

suffering a chest infection, some GPs dismiss my chest symptoms and say that my back hurts from coughing a lot (even when I hadn't been coughing much as it wasn't 'productive cough).”

Uncertainty regarding ongoing healthcare support means that adults have found themselves in a position whereby they have had to work hard for the right support, for example respondent 32 said “I had to fight for treatment and was responsible for completing treatment etc.”, similarly respondent 37 (adult) mentioned having to become “pushy” and “push” for the right care. Respondent 29 (adult) even expressed concern that some people may fall through the gaps and not receive any support at all, “I am concerned that people with less knowledge about healthcare (my mum and I are both healthcare professionals) will easily get lost”.

Discussion

The current research aimed to investigate the experience of transitioning from paediatric to adult healthcare, from the perspective of adults with OA/TOF and parents of adults with OA/TOF. Findings highlighted that both parents and adults felt that the process of leaving paediatric care represented a shift from stability to instability, from what was known to what was unknown. Paediatric care was typically reported as being comprehensive and supportive, whilst the opposite was true for adult care. Parents and adults experienced a shift in their roles during the process of moving from paediatric to adult care. Parents had to “step back” and relinquish responsibility, in some cases with reluctance, and adults “stepped up”, taking on more responsibility in their healthcare. It was common for parents and adults to express a sense of ongoing uncertainty related to living with OA/TOF. A key finding of this research is that the majority of parents and adults reported that there had been no formal healthcare transition.

An important finding of this research is that people born with OA/TOF do not experience a managed transition from paediatric to adult healthcare, even though it is advised that healthcare should continue into adulthood (Ijsselstijn et al. 2013, 2016; Krishnan et al. 2016). The finding that there is no formal healthcare transition is concerning, particularly because previous research has highlighted the negative outcomes resulting from an absence of formalised healthcare transition (Dellon et al. 2013; NHS long term plan, 2019; Lugasi et al. 2011; DOH, 2006; Waldboth et al. 2016). In common chronic conditions such as congenital heart disease (CHD), diabetes, epilepsy and juvenile

idiopathic arthritis (JIA) formalised healthcare transition processes have existed for some time (Betz et al. 2016; Chu et al. 2015; Kerr et al. 2017). Benefits of formalised healthcare transition are positive health outcomes such as increased self-reported quality of life, increased independence and self-care (Schmidt et al. 2020). The experience of healthcare transition for people with rare conditions is generally worse than the experience of those affected by common chronic conditions (Rare disease UK, 2014). In the current research even the word "transition" was unfamiliar to many of the respondents. It seemed that the experience of suddenly being discharged from paediatric services with little to no planning was common.

The current study found that the experience of moving from paediatric to adult healthcare was problematic, particularly for parents, as it was typically a swift, unexpected change from stability to instability and coupled with feelings of loss and abandonment. Previous research into transition experiences highlight feelings of loss experienced by parents and adults with common chronic conditions (Coyne et al. 2019; Lugasi et al. 2011; Fernandez et al. 2014; Fegran et al. 2014; Rutishauser et al. 2014; Gray et al. 2017), though not with rare diseases (Waldboth et al. 2016). The current study provides contrasting evidence to the findings by Waldboth et al. (2016). Parents experience of loss could be because, from the moment their child is born with OA/TOF, they developed a particularly close connection with paediatric services during a time that is often intensely traumatic (Caplan, 2013; Le Gouez, 2016; Rabone and Wallace, 2021). This suggests that children and young people born with OA/TOF and their parents receive a good service in paediatric care. A similar experience is described by Lugasi et al. (2011) who found that paediatric services are typically experienced as places of safety, eliciting feelings of familiarity and strong attachments when patients and families have usually spent a lot of time there.

Also highlighted in the current study was that parents' experience of leaving paediatric care was akin to falling off a cliff. A similar characterisation of healthcare transition is reflected in the wider literature, where the "falling off a cliff" metaphor is often used by parents of children with rare conditions as they transition to adult services (Rare diseases UK, 2014). Emotive language used by parents to describe leaving paediatric care is also reflected in research into other conditions. In their systematic review of cystic fibrosis, congenital heart disease and diabetes, Coyne et al. (2019) report that parents describe being "absolutely devastated" and being "dumped in adult services". Similarly,

Schultz (2013) reported a parent's description of leaving paediatric services as being "kicked to the curb". The quality care commission in their 2014 report "from the pond to the sea" describe how parents feel "abandoned" by health services.

The current study found that parents and adults experienced a profound cultural shift in the move from paediatric to adult healthcare. Cultural shift has been reported in previous research (Coyne et al. 2019, 2018, 2017; Lugasi et al. 2011; Heery et al. 2015; Garvey et al. 2012; Sheehan et al. 2014; Heath et al. 2017; Fegran et al. 2014). Patients sometimes report feeling "shocked" by the difference between paediatric and adult healthcare services (Garvey et al. 2012). The "warm and cozy familiarity" (Staa et al. 2011) of paediatric care is often contrasted with a much less "hands on" approach (Sheehan et al. 2014) of adult services which offer "vague, indifferent" (Iverson et al. 2019) and impersonal treatment (Coyne et al. 2018). The cultural shift from paediatric to adult healthcare is compounded by the fact that there is a perceived lack of physical resources and knowledge base in adult care, leading to fragmentation of care in adult services. This is in line with recent research findings that people born with OA/TOF experience a lack of follow-up and condition specific expertise in adult services (Ten Kate et al., 2020; Rabone and Wallace, 2021). A lack of resources in adult services and fragmentation of care is also commonly experienced in other rare diseases (Rare Disease UK, 2014; Coyne et al. 2017; Ariceta et al. 2016) as well as common chronic conditions (CQC, 2014) such as congenital heart disease (Heery et al. 2015), juvenile idiopathic arthritis and epilepsy (Burke et al. 2018).

Adults and parents in the current research reported that they experienced a change in their roles with regard to managing the OA/TOF, characterised by a shift in responsibility with parents "stepping back" and adults "stepping up". This shift in responsibility, which has been identified as particularly challenging for parents in the current study, is also reported in previous research (Learch and Thrane, 2019; Heath et al. 2017; Kaye et al, 2016; Coyne et al. 2017). Parents find it challenging to be excluded from consultations in adult services (Coyne et al. 2019). Fear, rejection and uncertainty have been reported elsewhere as common parental experiences as a young person transitions to adult services (Schultz, 2013). Although undoubtedly challenging for parents, shifting responsibility to young people has been identified as an important aspect of successful transition (Carter and McGoldrick, 2013; Coyne et al. 2019). A successful and structed shift in responsibility is a

huge step in ensuring successful self-management of conditions, and better health outcomes (CQC, 2014; Schmidt et al. 2020).

The current research found that parents and adults expressed differing views with regard to healthcare transition; parents seemed to express greater worry and concern about the process than adults. This has also been highlighted in previous research (Heath et al. 2017; Coyne et al 2017; Boyle et al 2001; Craig et al. 2007; Iles & Lowton, 2010; van Staa et al. 2011; Anthony et a;. 2009; Moons et al. 2009). Parents often report specific concerns about their child's readiness for transfer, whilst young adults often seem more "laid-back" (Heery et al. 2015; Heath et al. 2017), Ariceta et al. (2016) reported that parental overprotection and a sense that the patient may not be "mature" enough to cope with adult care are seen as barriers to transition. It is clear that "letting go" can be challenging for all parents and is particularly a common experience across chronic conditions (Reed-Knight et al. 2014; Wright et al. 2016; Heath et al. 2017), for example in congenital heart disease (Burstrom et al 2018), diabetes (Sheehan et al. 2014) and epilepsy (Schultz 2013). The process of relinquishing and renegotiating responsibility for healthcare is something that parents have trouble adjusting to (Betz et al 2016; Lewis & Noyes 2013; Walsh et al. 2017; Burke et al. 2018). Therefore, it is of vital importance that transition planning involves preparation regarding shift in responsibility (Coyne et al. 2019).

The theme "living with uncertainty" has parallels with the wider research literature. Parental uncertainty in particular is something that is highlighted in various conditions such as congenital heart disease (Burstrom et al. 2018) and cystic fibrosis (Coyne et al. 2019). Uncertainty can be fuelled by a lack of information. Unfortunately, gaps in transitioning education is a common experience across conditions (Fernandez et al. 2014) including juvenile idiopathic arthritis and epilepsy (Burke et al. 2018); diabetes (Iverson et al. 2019; Huang et al. 2011); congenital heart disease (Bratt et al. 2017); cystic fibrosis (Bregnballe et al. 2017; Craig et al. 2007; Iles and Lowdon, 2010; Huang et al. 2011) and inflammatory bowel disease (Huang et al. 2011). Burstrom et al. (2018) found that being well prepared and informed helped parents to feel more secure in the process of transition. Greater knowledge of conditions and thus greater confidence in self-management has also been reported elsewhere (Heery et al. 2015). Evidence suggests that if an individual has increased knowledge in their condition, they will feel more confident in their ability to self-

manage and separate from parental care, which may also help parents to “step back” (Clarizia et al. 2009).

Limitations

This study had a number of limitations. Whilst the study gives a good indication of the experiences of women born with OA/TOF and mothers transitioning in the UK, the sample is not representative of all parents and adults with OA/TOF and may not be generalizable to other countries. There was also no measure of ethnicity, so it is not possible to know if the sample is representative of differing communities. In addition to this the respondents were self-selected and majority female. There are no known gender or ethnicity differences in this condition (Beasley 2016), however as the sample was majority female and mothers, the experience may not be representative of males born with OA/TOF or fathers. Future research could perhaps address this gap. As an anonymous online survey, it was not possible to expand on or explore further the parent and adult responses, also parents and adults were being asked in retrospect about their experiences of healthcare transition the data could be subject to recall bias.

As participants were recruited via one UK charity, it could be argued that the sample was biased in this respect. Future research could address this by expanding recruitment to include healthcare centres, possible via paediatric consultants, thereby allowing for those who may not access charitable support to still take part.

The age range of the sample meant that the lived experience expressed in the current research included that of people who transitioned from paediatric to adult healthcare between 20 to 30 years ago. It is possible their experience may not be comparable with the experience of young people currently going through the healthcare transition process, however, there were commonalities in the experiences shared by younger respondents and parents of younger adults.

A further limitation regards to the use of the word “transition”. Despite piloting the survey and liaising with several PPE, halfway through the data collection process it became clear that the word “transition” was an unfamiliar term. Several respondents were being filtered out of completing the survey because they gave a “no” response to the question “have you/has your child transitioned to adult services?”. The wording of the filter question was thus changed from “have you transitioned to adult services?” to “have you been

discharged from paediatric services?". It is possible that the survey would have garnered more respondents had this question been used from the beginning of data collection.

Whilst the current study had a number of limitations, the data nevertheless provided a rich and detailed account of the experience of moving from paediatric to adult healthcare with OA/TOF.

Conclusion

To date there has been a paucity of research that explores healthcare transition in adults born with OA/TOF. This survey based qualitative study provides an insight into the lived experience of moving from paediatric to adult healthcare from the perspective of people born with OA/TOF and their parents. The findings expand on recent research into adult's experience of living with OA/TOF (ten Kate et al., 2020; Rabone and Wallace, 2021) and suggest that a formalised, managed healthcare transition is practically non-existent for people born with OA/TOF. Participants reported that they experienced changes in their roles with regard to managing the OA/TOF and parents found this particularly challenging. The challenges of moving from paediatric to adult healthcare as reported in the current study are similar to those experienced in more common chronic conditions. In contrast to other common conditions, whilst problems are still evident in the transition process, a formalised "process" still exists. In the case of OA/TOF many adults and parents are in the position whereby the word "transition" is simply not in their vocabulary.

Relevance to clinical practice

This current study supported the finding from the wider literature that people born with OA/TOF may continue to require healthcare support throughout adulthood. In the absence of formalised healthcare transition in OA/TOF there is an argument for an OA/TOF transition working group to develop shared transition guidelines. These guidelines should address the issues that are highlighted in the current study, including the shock of moving from paediatric to adult healthcare, the challenges of shifting parental responsibility, communication and coordination between services and an increase of condition specific knowledge in adult services.

The current research highlights that moving from paediatric to adult healthcare is experienced as a huge shock and a cultural shift, therefore it would make sense for there to be a preparation period prior to transition. Preparation is key to successful transition (Blum et al. 1993; Burke et al. 2018) and the move from paediatrics to adult healthcare

should be discussed and planned years in advance (Chesshir et al. 2013) ideally between the ages of 12 and 14 (White et al, 2018; CQC 2014; NICE 2016). The process of healthcare transition in OA/TOF should involve scaffolding partnership working and help transferring knowledge and responsibility at the right pace, it should not be expected that this will happen post transfer, unsupported. Healthcare transition in OA/TOF should be tailored to families and individual characteristics, needs and wishes, this would help to reduce the experience of confusion and uncertainty that is reported in the current study.

Clear from the current research is that adults born with OA/TOF and parents experience a change in their healthcare/health management role during transition. It would therefore be helpful for the process of transition to include an element of teaching/training, supporting parents in "stepping back" whilst encouraging adults born with OA/TOF to "step up" to self-management. The professionals providing this support should be skilled at communicating with young people and families. A variety of professionals at different levels could take this role, which may also take the burden off the over-worked core nursing/medical team.

The responses given in the current research suggest that paediatric services offer expert care that is comprehensive and consistent, therefore a case could be made for paediatric services to work in partnership with adult services, providing training and joint working to all health professionals (including GPs) likely to work with adults born with OA/TOF. There is evidence that this approach can be successful in other conditions such as cystic fibrosis (Okumura et al. 2014). Also evident from the current research is that participants identified that having a named keyworker through the process of transition to adult services would have been beneficial. Whilst it is acknowledged that GPs have a huge workload, if they could be supported to be more involved in the process of healthcare transition, possibly even pre-transition, as recommended by the CQC (2014) and in the NICE guidelines (2016), they could serve as the named keyworker, spanning the "two worlds" (Betz et al. 2015) of paediatric and adult healthcare.

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Executive Summary

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Clinical Tutor: Dr Elizabeth Marks
Word Count: 997

Meta-Synthesis

Rare diseases have been identified both internationally and nationally as an importance area of research. People with rare diseases face “unique, multidimensional challenges” (Eurodis, 2005) that are different to the challenges faced by those with common diseases (Bryson et al. 2019). Whilst the lived experience of adults with rare diseases is clear, the understanding of children’s experience of living with a rare disease is less clear (Rare Diseases UK, 2018). To understand children’s lived experience of rare diseases a meta synthesis was carried out, using a meta-study methodology (Paterson et al. 2001) to combine and synthesise qualitative research findings. This meta-study aimed to answer the following questions: What theories and methods are used to formulate children’s lived experience of rare disease within qualitative research literature?; What is children’s lived experience of rare disease? In total, 12 studies involving 652 children and adolescents aged 4 – 21 were included in the meta-study. A variety of methodologies were used across the studies and a lack of reflexivity was evident. It was unclear in the majority of the studies what the theoretical underpinnings of the research were. Four third order constructs (themes) were created by the lead author, these described children’s lived experience of rare diseases. Recommendations made from this meta-study are that clinicians should be aware that children with rare diseases will feel different from their peers, but that this difference may not always be experienced as negative. Children with rare diseases experience a restriction of their daily lives and therefore may benefit from practical solutions. The experience of distress, uncertainty and being ignored may be common for children with rare diseases and therefore psychological input may be required, particularly for children to feel that they are being heard. It is also recommended that children with rare diseases be supported in making connections with peers within the rare disease community.

Service Improvement Project

Over the past 40 years the UK government has supported a shift from an expert healthcare model, to a model that places service user involvement front and centre of all aspects of healthcare. In is mandatory for there to be an element of service user involvement in the training of both health and mental health professionals. Research suggests that the involvement of service users in the training of health professionals positively impacts on learning and in turn improves service provision. The University of Bath

have had involvement of People with Personal Experience (PPE) in the clinical psychology doctoral training course from the inception in 2008. In 2019 the course made 2 major changes to the admissions process: PPE would be represented on the admission interview panels for the first time; Candidates would be seen by one interview panel rather than three. This service improvement project aimed to explore the PPE experience of being part of the DClinPsych interview process and make recommendations for the admissions process the following year. All PPE representatives (n = 6) were invited to take part in a focus group, conducted by the lead researcher, to talk about their experiences. 4 of the 6 PPE took part in the focus group, the other 2 contributed via email, giving written feedback/responses to the focus group schedule. An adapted version of interpretative phenomenological analysis (IPA) was used to analyse the data. Four themes reflected the experience of the PPE as part of the interview process. The themes intersected multiple levels of experience. Recommendations made from this service improvement project were as follows: The PPE to receive an introduction from the course team on the interview days; the PPE to be provided with a list of common acronyms and initials on the interview days; guidance to be provided regarding note taking from candidate applications; informal "debriefing" to be provided at the end of interview days for all interviewers, including PPE; to be clear to new PPE that being part of the interview process may mean that they cross paths with a psychologist who could have treated them in the past.

Main Research Project

Oesophageal atresia (OA)/trachea-oesophageal fistula (TOF) are rare conditions whereby a baby is born with a food pipe that does not connect to their stomach and/or the lower part of the food pipe is connected to their windpipe. Since the 1940's the condition has been treatable with surgery shortly after birth, meaning that babies survive through infancy, childhood, adolescence and into adulthood. Ongoing health issues can be experienced by people born with OA/TOF and it is recommended in the research literature that ongoing healthcare should be available throughout childhood and adulthood. There is evidence to suggest that transitioning from paediatric to adult healthcare can be problematic with OA/TOF, though there is a paucity of research that examines the transition issues from the perspective of adults born with OA/TOF and their parents. The present study aims to investigate the experience of transitioning from paediatric to adult healthcare services from the perspective of adults with OA/TOF and parents of adults with

OA/TOF. The objectives of this study were as follows: To explore parents' and adults' experiences of the different services during the healthcare transitioning process; To investigate whether parents and adults experience a change in their role during the process of transition from paediatric to adult healthcare services; To identify the challenges that are faced as part of the transition from paediatric to adult healthcare. A qualitative, cross sectional, online survey-based study was carried out. Data was analysed using a six-stage hybrid approach combining elements of inductive and deductive thematic analysis (Fereday & Muir-Cochrane, 2006). Five themes were constructed reflecting the experience of parents and adults transitioning from paediatric to adult healthcare. The Five themes are as follows: Thrown into the unknown; A cultural shift; Stepping back and stepping up; "No transition as such"; Living with uncertainty. A key finding of the current research is that parents and adults born with OA/TOF do not experience a managed transition from paediatric to adult healthcare. Recommendations are made with regard to what a transition process should entail.

Appendices

Appendix A: Instructions for authors - Journal of Paediatric Psychology

The *Journal of Pediatric Psychology* is an official publication of the Society of Pediatric Psychology, Division 54 of the American Psychological Association. *JPP* publishes articles related to theory, research, and professional practice in pediatric psychology.

Author Guidelines

We would like to inform our authors that we now detect plagiarism easily. *JPP* employs the CrossCheck plagiarism screening system. By submitting your manuscript to this journal you accept that your manuscript may be screened for plagiarism against previously published works.

Journal of Pediatric Psychology will not consider papers that have been accepted for publication or published elsewhere. Copies of existing manuscripts with potentially overlapping or duplicative material should be submitted together with the manuscript, so that the Editors can judge suitability for publication. The Editors reserve the right to reject a paper on ethical grounds.

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including title page, references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Review articles should not exceed 30 pages. Invited commentaries should be discussed with the Editor. The *Journal of Pediatric Psychology* no longer accepts brief reports but will accept manuscripts that are shorter in length.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (7th ed.). There are two exceptions:

The academic degrees of authors should be placed on the title page following their names, and a structured abstract of not more than 250 words should be included. The abstract should include the following parts:

1. Objective (brief statement of the purpose of the study);
2. Methods (summary of the participants, design, measures, procedure);
3. Results (the primary findings of this work); and
4. Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch).

Informed consent and ethical treatment of study participants: Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Diversity, Equity, and Inclusion Considerations

Reporting and interpreting data related to Race and Ethnicity, including use of Bias-Free Language: Race and ethnicity are social constructs couched within a sociopolitical framework. Race and ethnicity are not genetic or biological categories. Care should be taken in the methods used to characterize samples in regard to race and ethnicity, reporting of this information, and interpretation of findings related to race and ethnicity categories.

Reporting of race and ethnicity (and associated intersectional factors such as culture, social structures, etc.) in the manuscript may vary across countries, languages, and cultures. Authors should provide sufficient rationale and justification for their data collection and reporting of race and ethnicity of their sample to be understood and appreciated by an international readership.

1. **Terminology.** Authors should follow the APA Style Guidelines on Bias-Free Language. The guidelines contain both general guidelines for writing about people without bias across a range of topics and specific guidelines that address the individual characteristics of age, disability, gender, participation in research, racial and ethnic identity, sexual orientation, socioeconomic status, and intersectionality.
    - Terms used to describe racial and ethnic groups (including spelling and capitalization) should adhere to bias-free language for Racial and Ethnic Identity
    - Similarly, authors should use systems centered language, showing awareness that disparities are due to inequities or deficiencies in social structures, systems, and processes rather than individual weaknesses or choices; for example, rather than stating that a population is “vulnerable” or “at risk”, identify the harms or social structures that drive oppression and racism (see psyarxiv.com/6nk4x/ for further details).
2. **Source used to identify race and ethnicity.** Clearly state the categories used to collect race and ethnicity data (e.g., Census data categories, funding agency categories) and the source of this information (e.g., participant self-report, electronic health record). Please indicate why those sources/categories were chosen (e.g., specified by the

funding agency). For example, an author may state: "Reporting race and ethnicity in this study was mandated by the National Institutes of Health, consistent with the Inclusion of Women, Minorities, and Children policy".

3. **Reporting race and ethnicity for sample description.** Race/ethnicity of the study population should be reported in full in the Results section and/or in a participant characteristics table, as applicable. All race and ethnicity categories represented in the sample should be reported individually rather than collapsing data across groups (e.g., "Other"). Note: This reporting requirement does not dictate how race and ethnicity categories are used in analyses – authors may conduct statistical analyses with race and ethnicity variables combined as appropriate to their study goals and methods, with appropriate rationale.
4. **Interpretation of race and ethnicity findings and recognizing limitations.**
    - Avoid making assumptions and conclusions that whiteness is the norm; for example, do not assume White comparison groups are needed or that racial differences found in one group are abnormal in comparison to White individuals. For further details, see https://psyarxiv.com/6nk4x/
    - Consider the structural effects of racism, and histories of exclusion, mistreatment, and exploitation on the populations included in the research and/or in relation to the findings. Authors should avoid making conclusions that may be interpreted as placing blame on minoritized populations. As it relates to interpreting the study findings, racism should be named. Authors are encouraged to identify the form (interpersonal, institutional, systemic), the mechanism by which it may be operating, and other intersecting forms of oppression (such as based on sex, gender, sexual orientation, age, regionality, nationality, religion, or income) that may compound its effects. For further details,
      see: https://www.healthaffairs.org/do/10.1377/hblog20200630.939347/full/
    - Authors should clearly acknowledge limitations of samples due to their lack of racial and ethnic representation (e.g., limited generalizability due to homogeneity of the sample). If data on racism or discrimination were not collected, authors should acknowledge this limitation in the interpretation of their findings. For further information on available scales of discrimination see https://scholar.harvard.edu/files/davidrwilliams/files/measuring_discrimination_resource_june_2016.pdf

Special Instructions for Types of Manuscripts

Manuscript types include:

- Original research
- Review articles
- Topical reviews
- Systematic reviews
- Invited commentaries

Original Research

Randomized controlled trials: JPP is committed to enhancing the transparent reporting of all intervention studies. (1) All Randomized Controlled Trials (RCTs) must be registered at or before the time of first patient enrollment in any primary registry of the WHO International Clinical Trials Registry Platform (ICTRP) or in ClinicalTrials.gov, which is a data provider to the WHO ICTRP. Provide the registry name and registry number in the cover letter and methods section. (2) You are required to submit the CONSORT checklist and a flowchart of

your research showing the steps found in the Consort E-Flowchart on this checklist for RCTs. You can use CONSORT checklist extensions for different designs and types of data beyond two group parallel trials. Please clearly indicate the page numbers where each checklist item is reported in the manuscript. Please upload this checklist as supplementary material when you submit your manuscript for consideration. Meeting these basic reporting requirements will greatly improve the value of your trial report and may enhance its chances for eventual publication. (3) If you are submitting a secondary data analysis from an RCT, please clearly indicate that it is a secondary data analysis in your manuscript and refer readers to the primary publication of outcomes. Consult with the editorial office if there are questions about reporting.

Pilot and feasibility trials: JPP is interested in publishing high-quality pilot and feasibility trials, which are randomized or non-randomized studies in advance of a future definitive RCT. The primary aim of the pilot or feasibility trial, however, is to assess feasibility of conducting the future definitive RCT not to evaluate efficacy. There are some key differences in pilot and feasibility studies from standard randomized or non-randomized trials, particularly in the type of information that needs to be reported and in the interpretation of standard CONSORT reporting items. Therefore, if you are submitting a randomized pilot or feasibility trial to JPP, you are required to follow the reporting elements of the CONSORT extension for randomized pilot and feasibility trials, and to use this checklist . If you are submitting a non-randomized pilot or feasibility trial, following recommendations, we would also like authors to use the CONSORT extension on pilot and feasibility studies as the main reference document for reporting, but ignoring items related to randomization and adapting other items as applicable.

Please clearly indicate the page numbers where each checklist item is reported in the manuscript. Please upload this checklist as supplementary material when you submit your manuscript for consideration.

Non-randomized trials: If you are submitting a non-randomized trial to *JPP*, you are required to follow the reporting elements of the TREND statement and to use this checklist for non-randomized trials. Please clearly indicate the page numbers where each checklist item is reported in the manuscript. Please upload this checklist as supplementary material when you submit your manuscript for consideration.

All intervention studies (RCTs and non-randomized trials) will undergo an additional review for transparent reporting conducted by the *JPP* Assistant Editor for Transparent Reporting. Review comments will be provided on the corresponding checklist. Authors will be required to address any identified reporting issues prior to publication.

Authors are also encouraged to visit the Equator Network for additional information on transparent reporting of all manuscript types.

(2) *Single Subject Studies*: As a journal that encourages submission of intervention studies, the Journal does accept, and encourages submission of, well-conducted single subject studies (N-of-1 designs). Case studies and narrative reports of special cases that are more descriptive will not be considered for review. It is important to note that rigorous single subject designs are considered logical equivalents of Randomized Controlled Trials and include control conditions that support conclusions of causality. Previously published examples can be found in this journal including: Bernard, Cohen, & Moffett (2009); Powers et al. (2006). Authors considering submissions of case reports adopting N-of-1 methodology should consult the following sources within this journal: Cohen, Feinstein, Masuda, & Vowles (2014); Cushing,

Walters, & Hoffman (2014); Rapoff & Stark (2008); Case reports that adopt formal N-of-1 methodology should not exceed 20 pages.

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(3) Measurement development and validation articles: For additional guidance please read, Holmbeck, G. & Devine, K. (2009) Editorial: An Author's Checklist for Measure Development and Validation Manuscripts.

(4) Historical Analysis in Pediatric Psychology: This is a special series of papers devoted to the history of pediatric psychology. Authors interested in submitting a paper for this series should contact the Editor of *JPP* to discuss potential papers prior to submission. There is no deadline for these papers (they may be submitted anytime). All submissions will be peer reviewed and should comply fully with the *JPP* Instructions to Authors. Papers in this series should be tightly focused contributions that expand our understanding of the roots, evolution, and/or impact of pediatric psychology as a discipline. Manuscripts may focus on the influence of individuals, published works, organizations, conceptualizations, philosophies or approaches, or clinical and professional activities. Successful papers should articulate a clear purpose/question and develop a compelling argument for the topic. Contributions should include a breadth of coverage, such that contradictory data are included and potential biases acknowledged. Historical analysis is more than a recounting of the "facts" and should include a thoughtful and scholarly interpretation of the subject matter. Papers should rely on primary sources and must be clearly and appropriately referenced. Supplemental materials to accompany the article may be posted online.

Review articles:

(a) *Topical Reviews:* Topical reviews summarize contemporary findings, suggest new conceptual models, or highlight noteworthy or controversial issues in pediatric psychology. Topical reviews are not intended to provide short data summaries or syntheses. Rather they are intended to foster new ways of thinking about a topic area and provide a direction for future

research or practice. They are limited to 2,000 words, contain no more than 2 tables or figures, and have an upper limit of 30 references. Supplementary online material (e.g., additional tables) may be considered on a case by case basis.

(b) *Systematic reviews and Scoping Reviews:* Reviews should not exceed 30 pages. Authors are required to attach the PRISMA checklist or extension for scoping reviews (PRISMA-ScR)and flow diagram as supplementary material for each submission. Authors can find the PRISMA checklist and flow diagram in downloadable templates that can be re-used. Authors of systematic reviews that do not include a meta-analysis must provide a clear justification in the manuscript explaining why such an analysis is not included for all or relevant portions of the report.

Please consult this editorial (New Guidelines for Publishing Review Articles in *JPP*) which further describes guidelines for review articles, and the Checklist for Preparing and Evaluating Review Articles.

Invited commentaries

- Commentaries are invited on all topics of interest in pediatric psychology, and the page length and scope should be discussed with the Editor. Un-invited commentaries will not be considered.

Additional Guidance

The following links provide additional guidance for authors and reviewers: Editorial Policy, Authors' Checklist, Guidelines for Reviews, Suggestions for Mentored Reviews, "People First," NIH policy, Replication of research, Duplicate and redundant policies, Conflict of interest.

See the following articles for detailed guidance concerning preparation of manuscripts: Editorial: Thoughts in Improving the Quality of Manuscripts Submitted to the *Journal of Pediatric Psychology*: How to Write a Convincing Introduction; Methods: Editorial: How to Report Methods in the *Journal of Pediatric Psychology*; Results and Discussion: Editorial: How to Write an Effective Results and Discussion Section for the *Journal of Pediatric Psychology*.

Funding

Details of all funding sources for the work in question should be given in a separate section entitled "Funding." This should appear before the "Acknowledgements" section.

The following rules should be followed:

- The sentence should begin: "This work was supported by . . ."
- The full official funding agency name should be given, i.e. "the National Cancer Institute at the National Institutes of Health" or simply "National Institutes of Health," not "NCI" (one of the 27 subinstitutions) or "NCI at NIH" (full RIN-approved list of UK funding agencies)
- Grant numbers should be complete and accurate and provided in parentheses as follows: "(grant number xxxx)"

- Multiple grant numbers should be separated by a comma as follows: "(grant numbers xxxx, yyyy)"
- Agencies should be separated by a semi-colon (plus ‘and’ before the last funding agency)
- Where individuals need to be specified for certain sources of funding the following text should be added after the relevant agency or grant number "to [author initials]."

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- Internet Explorer 9
- Internet Explorer 10
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Crossref Funding Data Registry

In order to meet your funding requirements authors are required to name their funding sources, or state if there are none, during the submission process. For further information on this process or to find out more about the CHORUS initiative please click here.

Appendix B: Full search strategy.

Database	Search terms
Web of science (Web of Science Core Collection; BIOSIS Citation Index; Current Contents Connect; Data Citation Index; KCI-Korean Journal Database; MEDLINE; Russian Science Citation Index; SciELO Citation Index)	(TS=(rare AND disease*) OR TS=(orphan AND disease*)) AND TS=(lived AND experience) AND (TS=child* OR TS= adolescent* OR TS= teen* OR TS= juvenile) *Databases= WOS, BCI, CCC, DRCI, KJD, MEDLINE, RSCI, SCIELO Timespan=All years Search language=Auto*
APA PsycNET (PsycINFO; PsycARTICLES; APA Books; PsycTESTS; PsycEXTRA)	**Keywords**: rare disease *OR* **Keywords**: orphan disease *AND* **Any Field**: child* *AND* **Any Field**: teen* *AND* **Any Field**: adolescent*
Pubmed	(("rare diseases"[MeSH Terms] OR ((((("rare diseases"[MeSH Terms] OR ("rare"[All Fields] AND "diseases"[All Fields])) OR "rare diseases"[All Fields]) OR ("orphan"[All Fields] AND "disease"[All Fields])) OR "orphan disease"[All Fields])) AND (("child"[Title/Abstract] OR "adolescent"[Title/Abstract]) OR "teen"[Title/Abstract])) AND (((((((("interview"[Publication Type] OR "interviews as topic"[MeSH Terms]) OR "interviews"[All Fields]) OR (((((("survey's"[All Fields] OR "surveyed"[All Fields]) OR "surveying"[All Fields]) OR "surveys and questionnaires"[MeSH Terms]) OR ("surveys"[All Fields] AND "questionnaires"[All Fields])) OR "surveys and questionnaires"[All Fields]) OR "survey"[All Fields]) OR "surveys"[All Fields])) OR ((((((("questionnair"[All Fields] OR "questionnaire's"[All Fields]) OR "surveys and questionnaires"[MeSH Terms]) OR ("surveys"[All Fields] AND "questionnaires"[All Fields])) OR "surveys and questionnaires"[All Fields]) OR "questionnaire"[All Fields]) OR "questionnaires"[All Fields])) OR (("theme"[All Fields] OR

	"themed"[All Fields]) OR "themes"[All Fields])) OR ((((((((((((("interpret"[All Fields] OR "interpretability"[All Fields]) OR "interpretable"[All Fields]) OR "interpretating"[All Fields]) OR "interpretation"[All Fields]) OR "interpretation's"[All Fields]) OR "interpretational"[All Fields]) OR "interpretations"[All Fields]) OR "interpretative"[All Fields]) OR "interpreted"[All Fields]) OR "interpreter"[All Fields]) OR "interpreter's"[All Fields]) OR "interpreters"[All Fields]) OR "interpreting"[All Fields]) OR "interpretive"[All Fields]) OR "interpretively"[All Fields]) OR "interprets"[All Fields])) OR (("experience"[All Fields] OR "experience's"[All Fields]) OR "experiences"[All Fields]))
Embase	('rare disease':ab,ti OR 'orphan disease':ab,ti) AND ('child'/exp OR 'child'/de OR 'child':ab,ti) AND ('adolescent'/exp OR 'adolescent'/de OR 'adolescent':ab,ti) AND ([embase]/lim OR [embase classic]/lim)

Appendix C: Bespoke data extraction form

Title of Paper:

Year of Publication:

1st Author:

1st Author's disciplinary background:

Journal:

Please complete the following details:

Inclusion/exclusion criteria	
No. participants	
Age range	
Participant's sex info	
Country	
Diseases/conditions	
Focus of the research (i.e. construct under investigation)	
Type of analysis (the exact words used to describe this)	
Any theory reported that informs the analysis?	
Data collection	
Recruitment	
Exact quotes reported in paper (cut and paste verbatim)	
Analysis findings (e.g. the themes)	
Limitations reported? (what were they)	
Take home message (concise summary of the research findings)	

Appendix D: Critical Appraisal Skills Programme (CASP) checklist for qualitative research

Section A: Were the results valid?				
Q1. Was there a clear statement of the aims of the research?	Yes	Can't tell	No	Comments: HINT: Consider • what was the goal of the research • why it was thought important • its relevance
Q2. Is a qualitative methodology appropriate?	Yes	Can't tell	No	Comments: HINT: Consider • If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants • Is qualitative research the right methodology for addressing the research goal
Q3. Was the research design appropriate to address the aims of the research	Yes	Can't tell	No	Comments: HINT: Consider • if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)
Q4. Was the recruitment strategy appropriate to the aims of the research?	Yes	Can't tell	No	Comments: HINT: Consider • If the researcher has explained how the participants were selected • If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study • If there are any discussions around recruitment (e.g. why some people chose not to take part)

Q5. Was the data collected in a way that addressed the research issue?	Yes	Can't tell	No	Comments: HINT: Consider • If the setting for the data collection was justified • If it is clear how data were collected (e.g. focus group, semi-structured interview etc.) • If the researcher has justified the methods chosen • If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide) • If methods were modified during the study. If so, has the researcher explained how and why • If the form of data is clear (e.g. tape recordings, video material, notes etc.) • If the researcher has discussed saturation of data
Q6. Has the relationship between the researcher and participants been adequately considered?	Yes	Can't tell	No	Comments: HINT: Consider • If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location • How the researcher responded to events during the study and whether they considered the implications of any changes in the research design
Section B: What are the results?				
Q7. Have ethical issues been taken into consideration?	Yes	Can't tell	No	Comments: HINT: Consider • If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained • If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the

				study on the participants during and after the study) • If approval has been sought from the ethics committee
Q8. Was the data analysis sufficiently rigorous	Yes	Can't tell	No	Comments: HINT: Consider • If there is an in-depth description of the analysis process • If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data • Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process • If sufficient data are presented to support the findings • To what extent contradictory data are taken into account • Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation
Q9. Is there a clear statement of findings?	Yes	Can't tell	No	Comments: HINT: Consider whether • If the findings are explicit • If there is adequate discussion of the evidence both for and against the researcher's arguments • If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst) • If the findings are discussed in relation to the original research question
Section C. Will the results help locally?				
Q10. How valuable is the research?	Comments: HINT: Consider • If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research based literature • If they identify new areas where research is necessary			

	• If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Appendix E: Instructions for authors - British Journal of Clinical Psychology

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If the authors cannot or do not intend to make the data publicly available, a statement to this effect, along with the reasons that the data is not shared, must be included in the manuscript.

Finally, if submitting authors have any questions about the data sharing policy, access the **FAQs** for additional detail.

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Authors are reminded that the *British Journal of Clinical Psychology* adheres to the ethics of scientific publication as detailed in the ***Ethical principles of psychologists and code of conduct*** (American Psychological Association, 2010). The Journal generally conforms to the Uniform Requirements for Manuscripts of the International Committee of Medical Journal Editors (**ICJME**) and is also a member and subscribes to the principles of the Committee on Publication Ethics (**COPE**). Authors must ensure that all research meets these ethical guidelines and affirm that the research has received permission from a stated Research Ethics Committee (REC) or Institutional Review Board (IRB), including adherence to the legal requirements of the study county.

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As part of the journal's commitment to supporting authors at every step of the publishing process, the journal requires the submitting author (only) to provide an ORCID iD when

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Author Guidelines updated 14th October 2019

Appendix F: Instructions for authors - Journal of Clinical Nursing

1. SUBMISSION

Thank you for your interest in the *Journal of Clinical Nursing*. Note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium. See Cover letter in Section 4 **Preparing Your Submission** for further details.

Once you have prepared your submission in accordance with the Guidelines, manuscripts should be submitted online at https://mc.manuscriptcentral.com/jcnur

The submission system will prompt you to use an ORCID iD (a unique author identifier) to help distinguish your work from that of other researchers. **Click here** to find out more.

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We look forward to your submission.

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The Journal of Clinical Nursing (JCN) will consider for review articles previously available as preprints. Authors may also post the **submitted version** of a manuscript to a preprint server at any time. Authors are requested to update any pre-publication versions with a link to the final published article.

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This journal expects data sharing. Review **Wiley's Data Sharing policy** where you will be able to see and select the data availability statement that is right for your submission.

Data Citation

Please review **Wiley's Data Citation policy**.

2. AIMS AND SCOPE

The Journal of Clinical Nursing (JCN) is an international, peer reviewed, scientific journal that seeks to promote the development and exchange of knowledge that is directly relevant to all spheres of nursing practice. The primary aim is to promote a high standard of clinically related scholarship which advances and supports the practice and discipline of nursing. The Journal also aims to promote the international exchange of ideas and experience that draws from the different cultures in which practice takes place. Further, JCN seeks to enrich insight into clinical need and the implications for nursing intervention and models of service delivery. Emphasis is placed on promoting critical debate on the art and science of nursing practice.

JCN is essential reading for anyone involved in nursing practice, whether clinicians, researchers, educators, managers, policy makers, or students. The development of clinical practice and the changing patterns of inter-professional working are also central to JCN's scope of interest. Contributions are welcomed from other health professionals on issues that have a direct impact on nursing practice.

We publish high quality papers from across the methodological spectrum that make an important and novel contribution to the field of clinical nursing (regardless of where care is provided), and which demonstrate clinical application and international relevance.

Topics include but are not limited to:

- Development of clinical research, evaluation, evidence-based practice and scientific enquiry;
- Patient and family experiences of health and health care; illness and recovery;
- Nursing research to enhance patient safety and reduce harm to patients;
- The nature of nursing need, intervention, social interaction and models of service delivery;
- Clinical nursing leadership;
- Examination of clinical decision-making;
- Exploration of organisational or systemic factors that enhance or impede the provision of effective, high-quality nursing care;
- Application and dissemination of clinical knowledge and theory;
- Role development and inter-disciplinary working, exploring the scope and changing boundaries of clinical nursing; and
- Cultural comparisons and evaluations of nursing practice in different health sectors, social and geographical settings.

Useful Resources
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*If you are presenting a paper from a study from which publications have already been drawn, or are planned, please carefully read our **guidance pertaining to multiple publications from a single study.***

3. MANUSCRIPT CATEGORIES AND REQUIREMENTS

i. Original Articles

Pilot studies are not suitable for publication as original articles.
Word limit: 8,000 words maximum (quotations are included in the overall word count of articles, and abstract, references, tables and figures are excluded).

Abstract: 300 words maximum, no abbreviations. Structured under the sub-headings: Aims and objectives; Background (stating what is already known about this topic); Design; Methods (for both qualitative and quantitative studies state n); Results (do not report p values, confidence intervals and other statistical parameters); Conclusions (stating what this study adds to the topic); Relevance to clinical practice. Trial registration details (if required).

Main text structure: Introduction (putting the paper in context - policy, practice or research); Background (literature); Methods (design, data collection and analysis); Results; Discussion; Conclusion; Relevance to clinical practice.

References: 50 maximum
Impact Statement: should contain 2-3 bullet points under the heading 'What does this paper contribute to the wider global clinical community?'

Research Reporting Checklist: May be required. Please see **Section 5**.

ii. Review Articles

Literature reviews on any area of research relevant to clinical nursing are welcomed. We encourage authors to prospectively register their reviews with a registry such as PROSPERO (**https://www.crd.york.ac.uk/prospero/**) or the Joanna Briggs Institute (**https://joannabriggs.org/ebp/systematic_review_register**).

Word limit: 8,000 words maximum (quotations are included in the overall word count of articles, and abstract, references, tables and figures are excluded).

Main text structure: Review Articles should be structures, under the sub-headings: Introduction, Aims, Methods, Results, Discussion, Conclusion, and Relevance to Clinical Practice.

References: 50 maximum

Research Reporting Checklist: Required. Please see **Section 5**.

iii. Discursive Articles

Word limit: 8,000 words maximum.
Main text structure: Aims; Background; Design (stating that it is a position paper or critical review, for example); Method (how the issues were approached); Conclusions, Relevance to clinical practice.

iv. Special Issue Articles

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Journal of Clinical Nursing is now considering submissions of Registered Reports. Registered Reports are a new form of empirical article in which the methods and proposed analyses are pre-registered and reviewed prior to research being conducted. For more information please refer to our **Registered Reports guidelines**.

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The Journal accepts two types of commentaries, with the first being preferable:

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- Expert opinion from one or more people (who may agree or disagree) on a current understanding/status of an area, or how practice should be undertaken. No abstract; limit references to 5 or less; 2,000 words maximum.

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a. Reserved for discussion about published papers.
b. No abstract; four or less references.
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f. Letters by article authors in response to Letters to the Editor disputing their articles are usually accepted for publication after the same type of review described above in e.
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viii. Editorial
To convey an opinion, or overview of an issue, by the Editor or someone invited by the editor. No abstract; limit references to four or less; 1,500 words maximum.

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Parts of the Manuscript
The manuscript should be submitted in separate files: title page; main text file; figures.

Title Page:
The title page should be submitted separately to the main file and contain:

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ii. A short running title of less than 40 characters
iii. The full names of the authors
iv. The authors' institutional affiliations at which the work was carried out
v. Corresponding author's contact email address and telephone number
vi. Acknowledgements.
vii. Conflict of Interest Statement
viii. Funding or sources of support in the form of grants, equipment, drugs etc.
ix. Clinical Trial Registration Number (if applicable)

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Main Text File and Figures
The main text file should be presented in the following order:

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ii. Main text;
iii. References;
iv. Tables (each table complete with title and footnotes);
v. Figure legends;
vi. Appendices (if relevant).

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Tables should be self-contained and complement, not duplicate, information contained in the text. They should be supplied as editable files, not pasted as images. Legends should be concise but comprehensive – the table, legend, and footnotes must be understandable without reference to the text. All abbreviations must be defined in footnotes. Footnote symbols: †, ‡, §, ¶, should be used (in that order) and *, **, *** should be reserved for P-values. Statistical measures such as SD or SEM should be identified in the headings.

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Legends should be concise but comprehensive – the figure and its legend must be understandable without reference to the text. Include definitions of any symbols used and define/explain all abbreviations and units of measurement.

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Although we encourage authors to send us the highest-quality figures possible, for peer-review purposes we are happy to accept a wide variety of formats, sizes, and resolutions. **Click here** for the basic figure requirements for figures submitted with manuscripts for initial peer review, as well as the more detailed post-acceptance figure requirements.

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Authorship
The list of authors should accurately illustrate who contributed to the work and how. All those listed as authors should qualify for authorship according to the following criteria:

1. Have made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data;
2. Been involved in drafting the manuscript or revising it critically for important intellectual content;
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9. EDITORIAL OFFICE CONTACT DETAILS

For queries about submissions, please contact **JCN@wiley.com**

Appendix G: Coding Manual

CODE 1	
LABEL	**Communication and continuity of care**
DEFINITION	Communication between paediatric and adult services, and a level of care continuing from paediatric to adult healthcare.
DESCRIPTION	The participants describe communication (or lack of) between healthcare providers. The participants refer to the care they received in paediatric and what happened to this when they moved to adult care.
CODE 2	
LABEL	**Discharge from paediatric health services**
DEFINITION	An "official" exit from paediatric healthcare that is orchestrated by the healthcare provider.
DESCRIPTION	The participant talks about being discharged from paediatric healthcare, or not – either way the word "discharge" is used.
CODE 3	
LABEL	**Preparedness for transfer**
DEFINITION	A process of getting ready for the move from paediatric to adult healthcare.
DESCRIPTION	The participant talks about a process within paediatric care that they were made ready for an eventual transition to adult care, or they talk directly about how this did not occur.
CODE 4	
LABEL	**Referral to adult healthcare**
DEFINITION	An official referral to adult healthcare, typically made by a clinician within Paediatric healthcare.
DESCRIPTION	The participant either mentions the word "referral" or they talk about being referred to adult healthcare and what this was like.
CODE 5	
LABEL	**Co-ordination of transfer to adult health services**
DEFINITION	The management of the transition process from paediatric to adult healthcare.
DESCRIPTION	The participant talks about the process of transition and how it was managed, they might use the word "transfer", "transition" or "move".
CODE 6	
LABEL	**Evaluation of needs for adult health services**
DEFINITION	A process whereby an individual enters adult services, and their needs are evaluated.
DESCRIPTION	The participant talks about having their healthcare needs assessed in some way whilst they are under adult services, or they explicitly say that this didn't happen.
CODE 7	
LABEL	**Allocation of resources for adult health**
DEFINITION	Following evaluation of needs in adult healthcare, there is an allocation of resources that aim to meet the needs to the individual.

DESCRIPTION	The participant talks about, or makes reference to, an allocation of resources within adult healthcare.
CODE 8	
LABEL	**Health service needs met**
DEFINITION	An individual's specific health needs being met by the particular service that they are seeing.
DESCRIPTION	The participant talks about their health needs being met by either paediatric or adult healthcare services.
CODE 9	
LABEL	**Self-management skills**
DEFINITION	The personal resources that an individual has in order to manage their health condition without having to rely on external resources.
DESCRIPTION	The participant talks about, or makes reference to, personal resources that are drawn on to manage the health condition, or alternatively they talk about a lack of personal resources to manage the health condition.
CODE 10	
LABEL	**Self-determination and confidence of young people**
DEFINITION	A sense of a young person being proactive and confident in their daily life.
DESCRIPTION	The participant talks about, or makes reference to self-determination and/or confidence of themselves (if they are the person with OA/TOF) or their child (if they are a parent)
CODE 11	
LABEL	**Need for transition planning**
DEFINITION	An identification, either by the individual, family or healthcare provider, that there is a need/desire for planning the move from paediatric to adult healthcare.
DESCRIPTION	The participant makes some reference to a transition plan/transition preparation.
CODE 12	
LABEL	**Health factors**
DEFINITION	The factors that relate to the individual's own health e.g., condition severity, activity limitations, visibility and stability.
DESCRIPTION	The participant says something that relates directly to their health/health condition.
CODE 13	
LABEL	**Personal factors**
DEFINITION	The factors that relate to the individual's personhood e.g., developmental readiness and perceived independence, autonomy and control.
DESCRIPTION	The participant says something that relates directly to who they are as a person.
CODE 14	
LABEL	**Successful/unsuccessful transition**

DEFINITION	A move from paediatric healthcare to adult healthcare that is considered to have been successful/unsuccessful.
DESCRIPTION	The participant mentions/refers to their move from paediatric to adult healthcare in a positive/negative way.
CODE 15	
LABEL	**Quality of life**
DEFINITION	A multidimensional term that refers to the level at which an individual feels that they able to engage in everyday activities
DESCRIPTION	The participant either uses the term “quality of life” or a variation of this.
CODE 16	
LABEL	**Co-ordination**
DEFINITION	The interconnection between family and healthcare, can both refer to physical factors and communication.
DESCRIPTION	The participant talks about either communication (or lack thereof) or some other sort of interconnected exchange between family and healthcare.
CODE 17	
LABEL	**Shift in responsibility**
DEFINITION	A process whereby the responsibility for the health condition shifts from the parent to the child
DESCRIPTION	The participant talks about responsibility moving from parent to child
CODE 18	
LABEL	**Family and peer support**
DEFINITION	The help and assistance provided by one’s own family and friends. The concept of support is multifaceted and can include physical support, emotional support, monetary support and spiritual support to name just a few.
DESCRIPTION	The participant talks about being supported, helped or assisted by a family member or a friend.
CODE 19	
LABEL	**Parental protectiveness**
DEFINITION	The level of protection that is provided by a parent of a child.
DESCRIPTION	The participant talks about a parent being “protective” or “overprotective”.
CODE 20	
LABEL	**Resources in health and social services**
DEFINITION	The resources (or lack thereof) that are available in health and social care.
DESCRIPTION	The participant refers to healthcare resources (or lack thereof) and/or describes what these resources are.

Additional codes from data:

Code 21: Falling off cliff
Code 22: Importance of charity

Code 23: Differences between Paed and adult/comparison between
Code 24: Uncertainty
Code 25: Lack of knowledge
Code 26: Being the expert
Code 27: Lack of resources
Code 28: Fragmented care
Code 29: No transition
Code 30: Emotional impact
Code 31: Abandoned
Code 32: Sudden change
Code 33: Ongoing struggle
Code 34: Being ignored

Appendix H: Ethical Approval Confirmation Email

psychology-ethics
Tue 01/09/2020 20:00
To: Andy Haig-Ferguson

Dear Andy

PREC reference number: 20-059

Thank you for letting us know about this amendment. I am happy to confirm that you have received a favourable ethical opinion, via Chair's Action. Your file has been updated to include these changes.

Your application has received a favourable ethical opinion. However please be aware that a researcher (or supervisor in the case of UG or Masters students) is responsible for ensuring full GDPR compliance. Please seek further advice from dataprotection-queries@lists.bath.ac.uk if you have any concerns.

Best of luck with your research.

Please make sure you quote your unique PREC code, 20-059, in any future correspondence.

Rebecca Wise
On behalf of Psychology Research Ethics Committee