The Psychosocial Impact of Living with Cloaca: The Young Person's Experience

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Abstract

Introduction: Cloaca is a congenital condition where a female's vagina, urinary tract, and anus merge into a single channel, affecting approximately 1 in 50,000 births (Wood et al., 2019). The condition necessitates early and complex medical interventions, with the potential for lifelong implications for bowel, urinary, and gynaecological health. Despite the enduring and complex nature of the condition, there is limited qualitative research on the experiences of individuals with cloaca and the psychosocial impacts, particularly as they transition from adolescence to adulthood. Therefore, this study focuses on the individual experience of young adults living with cloaca to build a more comprehensive understanding of the potential psychosocial impacts.

Method: One-to-one interviews were conducted with nine participants. Eight of the participants had personal experience of cloaca (aged between 16-40 at interview) and one participant was a caregiver. A qualitative design was used, underpinned by Constructivist Grounded Theory.

Results: Three themes emerged from the data. The first, sense of the bodily self, reveals how treatment and symptom management can impact individuals' perceptions of their bodies and the development of a sexual identity. The second, medical complexity, highlights the challenges of cloaca's complexity and rarity, impacting interactions with medical professionals, self-management, and increasing vulnerability to medical anxiety and trauma. The third, growing up with cloaca, explores how the condition is integrated into identity, involving the decision to disclose or withhold information. Connecting with others with cloaca offers comfort, normalisation, and a shared language to understand and discuss experiences.

Discussion: This study highlights the challenges that young people with cloaca can face regarding their psychological well-being and identity formation. It proposes a model to understand how growing up and living with the condition and the experience of treatment impacts these areas, emphasising the importance of integrating an understanding of patient and care experiences with other routine clinical outcomes.

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Introduction

Congenital conditions

Congenital conditions are defined as structural or functional anomalies that are present at birth following development in utero. Depending on the condition, some congenital anomalies can be detected prior to birth, at birth or even later in life. The World Health Organisation estimates that 6% of babies are likely to be born worldwide with a type of anomaly which results in a congenital disorder (World Health Organisation, 2023). There is a wide range of congenital disorders, each with its own unique set of symptoms and challenges. They vary from those that impact normal biological functioning such as cystic fibrosis and sickle cell anaemia, to those affecting mobility such as spina bifida. Others may impact the normal development of organs such as heart defects and others may also impact appearance, such as cleft palate. There is also variation in the life-threatening nature of the conditions and the associated prognosis. Although each congenital condition has its own specific features, children and their families can share many similar experiences. For instance, one distinct aspect of all congenital disorders is the fact that the condition is present at birth, which can be an unexpected adjustment for families. Moreover, treatment often involves ongoing medical or surgical intervention, frequent hospital visits, and involvement with many healthcare professionals which could extend from a child's earliest years into adulthood.

The literature shows that congenital disorders can have a significant impact on the psychological well-being of both children and their families. For example, children with congenital conditions such as spina bifida are reported to be at higher risk of experiencing anxiety and depression (Essner et al., 2011), with research suggesting that children are likely to be more isolated, with less positive peer and school experiences (Essner et al., 2011). Moreover, a study exploring the prevalence of depression and anxiety in patients with cystic fibrosis found that psychological symptoms were two to three times higher than community samples (Quittner et al., 2014). The literature also highlights the associated challenges for families of children born with congenital disorders. Studies have found that parents of children with a congenital heart anomaly report greater levels of stress, anxiety, and depression in comparison to the general population (Kaugars et al., 2017). Authors of this study link these elevated rates to the life-threatening nature of the condition, particularly at birth, and the ongoing caregiving needs that are often required. A similar pattern is noted in families of children with spina bifida, with lower levels of cohesion, difficulty of family roles and associated family stress (Kelly, 2008). However, it is important to note that the opposite can also be seen, with the impact of spina bifida providing opportunities for family resilience and cohesion to be developed over time (Kelly, 2008). Important protective factors for families with children with congenital conditions appear to be greater social support and financial security (Lee et al., 2007), with a lower gross household income associated with lower resilience (Delaney et al., 2022). The bi-directional relationship between parental and child psychological health is well-established in the literature, with parental stress likely to have an effect on children with congenital heart defects (Soulvie et al., 2012). Moreover, in cystic fibrosis, a concordance can be seen between parental and child reporting of stress, anxiety, and depression (Quittner et al., 2014).

Other psychological challenges associated with congenital conditions, as highlighted in the literature, include difficulties with self-concepts such as body image and self-esteem. For instance, in cystic fibrosis, the literature emphasizes themes of a felt sense of difference, where individuals experience looking and feeling different from their peers (Darukhanavala et al., 2021). Unintended consequences of treatment that places emphasis on diet and ideal body weight can inadvertently increase the risk of developing eating difficulties or general challenges with body image (Darukhanavala et al., 2021; Petropoulou et al., 2022). Moreover, body image perception may also be influenced by other factors such as scars, tubes, or a need for supplementary oxygen supply (Petropoulou et al., 2022). In the literature relating to spina bifida, functional difficulties and associated social impacts of the condition are seen to impact on global self-worth and self-esteem (Shields et al., 2008). However, an interesting relationship exists here with severity of ‘disability’ not always equating to a more negative concept of the self. In fact, lesser degrees of ‘disability’ have been found to be linked to lower ratings of self-esteem (Minchom et al., 1995). This is a finding commonly replicated in the literature, often labelled as the ‘disability paradox’, whereby greater psychological stress is experienced by those with less severe condition (Albrecht & Devlieger, 1999). Important contextual factors such as parental stress, self-concept and social support are highlighted as potential mediators. Therefore, it appears crucial to consider an individual born with a congenital condition within their social context and external environment, before making assumptions about their experience based only on medical characteristics.

Anorectal malformation (ARM)

One group of congenital conditions which have arguably received less research attention from psychologists are those involving an anorectal malformation (ARM). ARM is an umbrella term that comprises a group of malformations where the anus and rectum do not develop properly (Max’s Trust, n.d.). Different types of ARM involve a variation of anatomical characteristics. They are often categorised as either ‘low’ or ‘high’, depending on the location in the digestive tract where the malformation occurs. A ‘low’ malformation refers to the anus being closed over, in a different position or narrower. A fistula to the skin, which is an abnormal connecting passage, may also be present. On the other hand, a ‘high’ malformation refers to a closure in the bowel at a high level, where it does not connect with the anus, or a fistula may also be present that connects it to the bladder, urethra, or vagina (Great Ormond Street Hospital (GOSH), 2019). In terms of prevalence, ARM is reported to occur in approximately 1 in 4000 to 5000 live births and are slightly more common in males (Wood & Levitt, 2018). As outlined above, these conditions similarly involve extended stays in hospital, ongoing medical and surgical intervention, and involvement with a number of medical professionals.

Despite advances in medical intervention over the last 30 years, the variability of presentation and success of surgical repair, a large proportion of individuals with ARM are likely to require lifelong medical interventions to maintain faecal and urinary continence (Judd-Glossy et al., 2019). There is a focus within the literature on the impact of this incontinence on social, occupational and educational areas of life (Mantoo et al., 2013). From a clinical perspective, achieving better functional outcomes such as continence appears to be seen as an important pathway to a better quality of life. Some studies demonstrate the direct impact of disease-specific factors such as constipation and soiling on quality of life (Goyal et al., 2006; Grano et al., 2010). Other studies have highlighted that despite ratings of significantly worse functioning in physical symptoms, ratings of overall quality of life do not significantly differ from age matched controls (Wigander et al., 2019). Interestingly, it has been suggested that overall quality of life in ARM is less influenced by colorectal outcome than by psychosocial functioning (Hartmann et al., 2004; Stenström et al., 2014). A qualitative systematic review has identified the range of possible psychosocial challenges for individuals and families. Consideration of challenges across age-groups has included financial and social strain for parents, difficulties at school and behavioural problems for young children, low self-esteem and body image concerns for adolescents, and worries about sexual function for young adults (Sventanoff et al., 2022). Moreover, this review highlights that an ARM is associated with a range of difficult emotions for both patients and their families. Some examples include fear that can be experienced by both patients and their families when it comes to hospital stays and surgical procedures and the overwhelm that parents can experience in relation to the responsibilities they held in providing ongoing care to their children, which can be experienced as unpleasant and invasive (Sventanoff et al., 2022). Moreover, Hamid et al (2007) attempted to assess the long-term outcome of surgery for ARM and administered a questionnaire to assess both psychological and clinical outcomes and similarly highlighted the difficult emotional experience associated with ARM. They reported a high prevalence (80% of child participants between the ages of 3-20 years) of anxiety, embarrassment, and low mood. Despite this, 62% of adolescents (12-20 years old) and 71% of children below 12 years of age in the study reported above average hopefulness for the future, although potentially important mitigating factors such as coping strategies or protective factors were not explored. Furthermore, whilst this study aimed to capture a family perspective and also included a separate questionnaire for adolescents, it did not account for the potential variance in reporting between children and parents between participants or the influence parents might have on the responses of the adolescents. It is therefore difficult to discern the child voice within this study.

Thus far, the research suggests that certain psychological and social factors play a significant role in determining a person's quality of life. These factors can have a significant impact in reducing the impact of disease specific factors of ARM on a person’s life (Hartmann et al., 2004). Specifically, the existing evidence base highlights social support and self-esteem as two of these psychosocial factors. Additionally, an increased sense of self-efficacy has also been found to contribute to the reduction of emotional distress experienced by physical experiences such as incontinence (Grano et al., 2018).

Cloaca

Among the different types of ARM, cloaca is considered to be the most severe and complex (Vilanova-Sanchez et al., 2020). Cloaca occurs when a female baby’s vagina, urinary tract, and anus do not separate from one another and instead open into one common channel for the passage of stool, urine, and menstrual flow. In common development, these three structures form distinct and separate openings, allowing for the independent function of the digestive, urinary and reproductive systems. Cloaca is reported to occur in 1 in 50,000 births (Wood et al., 2019). For a baby born with cloaca, this failure of separation creates significant anatomical and functional complications; life threatening difficulties subsequently arise soon after birth, and the first years of life are marked by invasive and complicated procedures (GOSH, 2019). The cloacal repair, which involves the creation of three separate channels, is considered to be one of the most challenging repairs in paediatric surgery (Svetanoff et al., 2023).

Figure 1. Illustration of cloaca in comparison to female common anatomy.

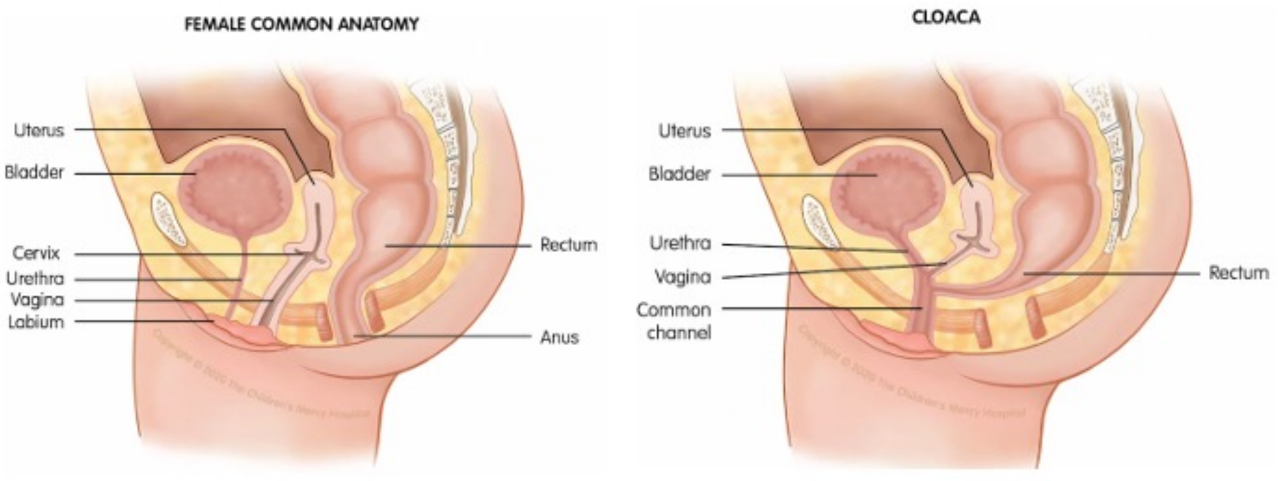

Image taken from Children’s Mercy Hospital. (2024).

While cloaca is classified under the umbrella of anorectal malformations (ARM), it stands apart as the most severe and complex due to the unique anatomical challenges and the subsequent impacts of these. Unlike most ARMs, which primarily affect the digestive tract, cloaca impacts the urological, gastrointestinal, and gynaecological systems simultaneously, creating significant complexity in both reconstructive surgery and long-term care. In fact, the condition's impact on the gynaecological system introduces challenges related to sexual function and fertility that are issues not relevant to other ARMs. Since cloaca exclusively affects female infants, these concerns are directly related to biological sex. However, gender-specific challenges and subsequent societal expectations are likely to play a crucial role in shaping how patients experience and navigate the impact on these three systems, providing a key aspect of consideration inherent to cloaca, as opposed to other ARMs.

As an individual ages, surgical and medical intervention is likely to continue, with a focus on bowel and urinary control and gynaecologic function. The specifics of interventions depend on the severity of the cloaca and the specific organs and structures affected; however, it is likely that the majority of individuals will experience long-term impairments in one or more of these areas (Versteegh et al., 2016). In addition, associated anomalies related to the spinal cord or nervous system can cooccur with cloaca that require further medical intervention and management.

Due to the enduring and complex nature of the condition, living with cloaca involves long-term involvement with a multi-disciplinary team of medical specialists including paediatric surgeons, urologists and gynaecologists.

Moreover, cloaca may present alongside other congenital malformations that can similarly occur in the early stages of foetal development. A diagnosis of VACTERL Association may be given to an individual with two additional anomalies related to the vertebral, cardiac, trachea-oesophageal, renal or limb area. VACTERL Association is reported to affect around 1 in 10,000 to 1 in 40,000 of the general population (Schwarzer et al., 2024). There is presently a small amount of literature which explores the rates of association. For example, one study suggests that up to 40 to 70% of patients with ARM have associated congenital malformations that classify as VACTERL association (Stoll et al., 2007). Another study, conducted by Ahmad et al (2023) involved 834 patients with ARM undergoing VACTERL screening and reported that VACTERL association was seen in 45.6% of patients with ARM. However, these studies do not specify how many diagnoses of cloaca are included under the umbrella of ARM and there is a lack of focused attention on the relationship between cloaca and VACTERL association in the literature more generally.

In terms of treatment specifics for cloaca, the main goals are similar to other types of ARM; namely to improve function and manage medical or psychological difficulties that may arise. There have been significant advances in the medical treatment of cloaca in recent years. For example, techniques such as the PSARP/PSARVUP (Posterior sagittal anorectal vaginal urethral plasty), and the TUM (total urogenital mobilization) have been developed for the complex surgical reconstruction of cloaca (Versteegh et al., 2015). These techniques have dramatically improved management and are generally associated with better cosmetic and functional outcomes (Levitt & Pena, 2010; Rintala, 2016). In addition, the length of the common channel can influence type and complexity of the intervention. The average length of the common channel tends to be 3cm although it can vary from 1 to approximately 10cm (Levitt et al., 2011). The shorter common channels (1-3cm) are most common and are often associated with a better prognosis. For example, individuals with a common channel longer than 3cm usually require more complex surgical procedures and have a greater likelihood of experiencing urological problems (Levitt et al., 2011). Due to advancements in treatment, it is expected that a baby born with cloaca today will generally have a more positive experience of medical and surgical treatment than those born 50 years ago. Although, the constellation of characteristics that can incorporate cloaca means that an individual's experience of medical and surgical intervention is likely to be unique, even among those born with the condition today.

The majority of existing research explores the postoperative functional outcomes for patients with cloaca, with a focus on bowel and urinary control and gynaecologic function. Firstly, in terms of bowel control, studies suggest that ongoing issues with bowel control are a reality for those with cloaca. A systematic review carried out by Versteegh et al (2013) highlighted a high incidence of soiling and constipation (71% and 51% of patients respectively). Just over half (57%) were able to have voluntary bowel movements with oral laxative or enema bowel management programs identified as strategies commonly used to assist bowel movement. In fact, an average of 17% of patients across the studies opted for a permanent colostomy due to the low quality of bowel control. However, it is important to note that this systematic review mainly included reported case series, deemed to be of low quality, with no control groups for comparison. Moreover, it is noted that all studies used different methods to report outcome with varying levels of detail. Therefore, the conclusions that can be drawn are somewhat limited. However, similar results appear to be replicated in different studies. For example, Davies et al (2010) highlighted that in comparison to other types of ARM, patients with cloaca were most likely to need assisted defecation, with only 27% of the participants able to achieve unassisted bowel function. Interestingly, other studies have highlighted bowel function as more favourable in adolescents and adults, with constipation and soiling becoming less common with increasing age (Ortqvist et al., 2022). It is proposed that as an individual ages, they adapt and develop strategies to keep clean such as dietary modifications or effective use of medications (Rintala, 2016). There is a similar picture when it comes to bladder function. It is apparent that maintaining bladder function is of critical importance, as deterioration of renal function is a significant risk for patients with cloaca and can be life-threatening (Rintata, 2016).

The same study as above by Davies et al (2010) highlighted that the cloaca group were most severely affected when it came to urine continence, with only 20% of the participant population (>18 years) fully continent. Ortqvist et al (2022) reported that girls aged 4-7 experience a significantly higher frequency of urine tract infections in comparison to age matched controls.

Gynaecological function is a physical outcome that has typically received less attention, although it does appear to be a developing area in the literature. Patients born with cloaca have a high incidence of gynaecological problems, with the majority likely to have a Müllerian anomaly (Svetanoff et al., 2023). Müllerian structures are typically understood and defined as those that are found in the female foetus that eventually develop into the reproductive organs. These structures fuse together during typical development to create the uterus, fallopian tubes and majority of the vagina (Chandler et al., 2009). It is reported that a large proportion of patients are likely to have some form of separation or duplication of the gynaecological structures (Svetanoff et al., 2023). Müllerian abnormalities can lead to a number of challenges such as obstruction to menstrual flow and increased menstrual pain (Pradham, 2018). Difficulties can remain asymptomatic until puberty when menstruation or sexual activity begins and it could be at this point that further investigations and interventions are required (Warne et al., 2003). Although, in some cases, complications do not always arise at the onset of puberty and so continuous and cautious observation tends to be required throughout a young person's development and even later into their life.

In some cases, reconstruction of the vagina may take place in the early years of an individual's life and the extent of the reconstruction needed is likely to depend on the physical manifestation of the condition and choice of intervention will depend on factors such as the length of the common channel. However, there is an increasing amount of insight and guidance about the importance of considering a number of psychological factors alongside surgical feasibility when making decisions about the timing of such procedures. For example, aftercare is likely to involve regular vaginal dilations which have the potential to cause a significant amount of discomfort, and emotional maturity to cope with this procedure is reported to be an important consideration (Svetanoff et al., 2023).

A retrospective review of outcomes for women aged 13-35 was conducted by Couchman et al (2015). They found that almost half of the sample were identified as having Müllerian anomalies. Nine women, out of a sample of 19, required further reconstructions to facilitate sexual activity or menstruation. Eight of the sample were sexually active, with one woman reporting difficulty with penetration. In terms of fertility, two of the three in the sample who had attempted to conceive were undergoing fertility treatment. However, the small sample size and broad age range means that conclusions that can be made from this data are limited. For example, the mean age of the sample was 22 years of age, which means there would be many women in this sample for whom not trying to conceive would not be unexpected for their age, which reduces the sample size for fertility outcomes even further. Huibregtse et al (2014) carried out a systematic review to ascertain obstetric outcomes, specifically fertility. Whilst the study included other types of ARM, cloacas were able to be studied as a separate group. 15% of the sample had children of their own which, in comparison to 'healthy' controls was found to be a lower childbirth rate. However, childbirth as the only measure of fertility is problematic. It does not account for other psychosocial factors that may influence sexual activity such as body image concerns, sexual anxiety or even desire for children in the first place. Assessment of fertility rates by comparison with sex, age, and social status matched control group was only evident in two of the studies within the systematic review, both with small participant groups.

A retrospective review of female patients with a diagnosis of ARM to ascertain fertility outcomes was conducted by Reppucci et al (2022). Whilst the study focuses on outcomes for complex ARM patients, it is difficult to extrapolate the outcomes that pertain specifically to those with a diagnosis of cloaca. However, in general, the study highlighted that whilst pregnancy was reported within the sample, due to the level of complexity, assisted reproduction techniques such as in vitro fertilisation and caesarean delivery are likely to be needed in presentations such as cloaca and other types of complex ARM. Importantly however, the study highlighted additional complications that require careful consideration during pregnancy, such as the increased risk of urinary tract infections following reconstruction, the potentially harmful effect of senna, a common bowel management intervention, and even difficulty in diagnosing pregnancy, with patients who have had experience of bladder reconstruction likely to have a 57% false-positive rate with a urinary pregnancy test (Reppucci et al., 2020). Moreover, it is possible that extra assistance throughout delivery might be needed, as the review highlighted two patients in the sample that required extra assistance from colorectal surgeons during their caesarean delivery to ensure the safety of both mother and baby. The growing evidence about gynaecological function indicates a complex picture and an evolving understanding of the importance of considering how physical and psychological factors interact in this context. However, at present the literature acknowledges and highlights how little guidance there is for recognising and managing gynaecological concerns past the neonatal period (Svetanoff et al., 2023).

Due to the often small and varied samples within the studies, it is difficult to generalise findings about the postoperative functional outcomes for individuals living with cloaca. However, these findings do highlight that individuals are likely to experience complex difficulties which will require ongoing medical intervention. In terms of psychosocial impact, it is reasonable to suggest that individuals born with cloaca might experience a range of challenges, as highlighted in the existing research around ARM and other congenital conditions. Therefore, a review of the literature was conducted to ascertain the existing evidence base in relation to the experience of living with cloaca and to explore possible psychological challenges.

The review of the literature aimed to get a sense of the evidence base in terms of psychological impact, looking beyond outcomes focusing solely on postoperative functionality (i.e. bowel or urinary continence). In reviewing the literature, it was apparent that a small amount of existing research focused specifically on cloaca, especially in comparison to the existing literature base that explores the impacts of living with other types of ARM. Some studies that focused on ARM more broadly did include cloaca as a distinct category of ARM within the wider participant group and considered cloaca as a separate group with within analysis, results and discussion. Other studies did not necessarily make this distinction for cloaca. Therefore, the review of the literature attempted to capture studies that did explicitly name cloaca and the outcomes related to cloaca, whether the overall focus of the study was on cloaca or more generally on ARM. However, despite this, the pool of existing evidence remains small and underdeveloped at present.

The review of the literature was conducted through the following databases: PsycINFO, Ovid, MEDLINE, Google Scholar and Cochrane Library. The search terms used were: “cloaca”, “cloacal malformation”, “persistent cloaca”; “psychosocial”, “psychological”, “quality of life”; “anorectal malformation”, “anorectal anomalies”. These terms were searched as keywords, titles, and abstracts. This search aided a review of the literature and contributed to the following narrative account and review of the existing literature.

In terms of the psychological impact, the review of the literature highlighted a few studies that used various measures to assess quality of life for individuals diagnosed with cloaca. For example, a study focusing solely on participants with cloaca showed comparable quality of life scores to a European normative data set (Örtqvist et al., 2022). Interestingly, challenges with urinary and bowel function appeared to have a limited impact on reported quality of life. In fact, significantly higher scores which indicate a more positive rating were shown in the social support, peers, and school environment domains for the participant group with cloaca. The findings of this study are interesting to consider in light of the following factors. Firstly, questionnaires were sent to the participants and their caregivers; therefore, it is unclear to what extent parents or caregivers may have been involved in the completion of the questionnaires across the sample, especially given the young age of the participant group (median age of 9.5) which could introduce a level of bias. Also, QoL was measured using ‘KIDSCREEN-27’, which is defined as a generic questionnaire for both healthy and chronically ill young people aged 4 to 18. It is possible that this measure may not have been sensitive enough to capture the specific experience of living with cloaca and difficulties may have been missed. Moreover, it does not account for the fact that problems may change over time. It is also important to consider that European normative data was used to compare the

QoL scores of the cohort of patients with cloaca from Nordic countries. Culturally specific factors might be at play that support a well-maintained quality of life in the Nordic countries which continuously rank highly in worldwide scales on factors including, and not limited to, life satisfaction, social cohesion, felt safety, equal distribution of income and social support (Martela et al., 2020). Moreover, studies have shown that European QoL means are known to be lower, particularly in comparison to Swedish QoL means (Berman et al., 2016). Therefore, it is important to consider the findings of the study in light of these contextual factors. It would be interesting to compare the QoL of these cohort of patients with normative data from the same population as it is possible that the patient cohort may have a better ‘resting’ quality of life that may have obscured findings. In addition, there may be important contextual factors that are facilitating this better quality of life that would be important to capture.

Similar findings were reported by Davies et al (2010) who administered the SF-36 (The Short Form 36 Health Survey Questionnaire) to participants over the age of 16 (25.6 mean age). The SF-36 is a measure of health-related quality of life using eight scales that include physical, social, and emotional functioning. Apart from the general health subscale which showed the normative population group as having significantly higher scores, the study found non-significant differences on the other subscales. This indicates that individuals with cloaca can achieve an adequate quality of life, despite the considerable challenges they may face, which is a hopeful and encouraging finding. However, it is interesting to note that the effectiveness and suitability of the SF-36 as a measure for capturing the experience of individuals with cloaca and other types of ARM has been questioned. For example, Stenstrom et al (2014) suggested that the measure is not sensitive enough to capture the reality of an individual’s experience of living with an ARM. In addition, it does not account for the accommodation to a lifelong condition that is likely to have occurred. Therefore, conclusions about quality of life using this measure could, arguably, not be seen as accurate or representative. Furthermore, it is important to note that this study included cloaca among other types of ARM. Although further analysis did separate types of anomalies and found no differences in the cloaca group as compared to the control group, it could be argued that nuances involved in living with cloaca might not have been captured if sufficient consideration was not given to cloaca as a distinct group in the study design.

Interestingly, Versteegh et al (2016) compared quality of life scores for patients with cloaca and compared them with scores for patients with less medically complex ARM such as those born with a rectoperineal (RP) or a rectovestibular fistula (RV). Quality of life scores were gathered via patient self-report for individuals aged 8-13 (median age 12 years) and parent-proxy report for individuals aged 5-12 (median age 8 years) For the self-report group, no significant overall differences were found between the cloaca and RP/RV group. Although, parents of patients with cloaca did report significantly lower scores on the school performance subscale. In this study, it is important to note that the majority of participants born with cloaca had short common channels (range 1.0-3.0 cm) which are generally associated with better functional outcomes that could be more comparable to patients with a RP/RV. This could have an impact on QoL scores as the study highlights those participants who suffered from poor bowel control reported significantly lower scores on the psychosocial health and school performance subscale, compared to individuals who had less difficulty with bowel control. However, when patients with cloaca were compared to the healthy reference group, QoL scores were again not significantly different, though parents of these children again reported significantly lower scores on the subscale of school performance and also lower psychosocial health. The authors of the study suggest that patients with cloaca have developed a set of coping skills that have facilitated an improved quality of life, in the face of a significant and unforeseen diagnosis at birth. However, the design of this study does not allow for deeper exploration of individual experience and possible supportive and protective factors which means that limited conclusions can be drawn in this respect.

In contrast to the above findings, Danielson et al (2017) compared quality of life outcomes with population-based sex and age-matched controls and across types of ARM. The study found that participants had lower quality of life ratings compared to controls and suggested driving factors such as persistent faecal incontinence, constipation, and sexual problems. Moreover, the study highlighted that the type of malformation was closely related to functional outcome and quality of life, with the worst functional and quality of life outcomes seen in the group of participants with cloaca. The participant group was made up of individuals who were diagnosed with cloaca from 1961 to 1993. However, there were only three participants in the cloaca group and the study does not state the age of these participants. Therefore, the small sample size for cloaca participants and the uncertainty in terms of their age, especially considering significant differences in treatment approaches that might have been experienced, are two important factors to consider.

Similar findings were reported in a recent study that compared females with cloaca with females with non-cloacal ARM found that ratings of quality of life were significantly different between the two groups. In fact, those with cloaca reported lower overall quality of life compared to those with other types of ARM (Ahmad et al., 2022). Whilst this study did not include control data in order to compare the scores with the general population, it remains an interesting contrast to the above findings and demonstrates the inconsistencies and uncertainties present in the developing literature base. It is apparent how, in this research area, researchers face similar challenges with sampling error and bias that are associated with the smaller sample sizes due to the rare, intimate, and unique nature of these conditions.

When it comes to other psychological factors, the study by Davies et al (2010) as described above also considers body image and sexual wellbeing. Lower sexual wellbeing and body esteem scores in comparison to the normative population were reported. Also, there was a linear association between sexual wellbeing and functional difficulties, with the more severe the urinary and bowel incontinence, the greater the sexual anxiety. Again, the authors specify that no significant difference was seen between types of ARM. However, considering the severity of functional outcome often seen in cloaca and as evidenced in this study, it follows that specific consideration should be given to this issue in cloaca alone. It is also important to consider the interacting effects of gender that are likely to be specific to cloaca. For example, in this study, Davies et al (2010) highlight significant gender differences on the items of sexual esteem and sexual assertiveness, with women having lower scores than men.

Moreover, the study by Ahmad et al (2022) referenced above also explored body image and sexual wellbeing. Firstly, over half (54%) of females in the cloaca group reported they felt either 'a little', 'quite a bit' or 'very much’ less physically attractive because of their condition. This is compared to 38% of females in the ARM group, although the difference is statistically non-significant. Neither were there any significant differences between the cloaca and the ARM group when it came to feeling less feminine or self-consciousness about scars. However, almost half (40%) of the cloaca group reported feeling ‘a little’ or ‘quite a bit’ less feminine and 61% of the cloaca group responded as feeling self-conscious about their scars. Aside from the statistically non-significant differences with the other types of ARM, these findings are still worth noting as potentially important features of living with cloaca and suggest areas for further exploration.

For sexual activity and wellbeing within the same study, similar rates of engagement with sexual activity were reported between the two groups. However, the response rate for both the cloaca and non-cloaca ARM group was relatively low, particularly for the ARM group, which meant that no statistical analyses could be carried out. No control group means that conclusions are somewhat limited, however one interesting finding is that despite the limited number of individuals reporting penetrative sexual intercourse in both groups, the frequency of reported climax was higher. This could indicate that sexual wellbeing and satisfaction requires a wider consideration of forms and factors other than penetrative forms of intercourse. Additionally, in terms of femininity, this is an assumed area of importance for sexual wellbeing; it was not apparent whether the study considered whether feeling feminine actually held any meaning for the individual participants, or whether this has, in fact, been impacted on by their condition. This suggests there are a range of nuances to consider, explore and understand when it comes to this area, considering the range of sexual orientation, identity development and potential ways in which individuals may have adapted to their condition.

A qualitative study - one of the only qualitative studies found in the review of the literature - that begins to explore the above areas of complexity in relation to the sexual and also reproductive challenges was conducted by Yu et al (2022). The participant group spanned 18 to 53 years of age and included 17 participants who identified as women and 3 as non-binary or genderqueer. Focus groups and semi-structured interviews captured various barriers to intercourse and intimacy the participant group experienced such as pain, lack of sensation and incontinence. Themes such as self-image were also identified as an important factor. Interesting, there was consensus among participants how transition to adult services was challenging due to having to find clinicians who could support them navigate gynaecological implications associated with their conditions.

Another qualitative study identified in the review of the literature aimed to gain insight into the health experiences of young women diagnosed with cloacal anomalies, focusing specifically on continence management (Liao et al., 2023). The study involved one-to-one interviews with six women aged 16-24, conducted in the UK at a tertiary care centre specialising in congenital anomalies of the urogenital tract. The findings highlighted that uncertainties regarding the ability to maintain continence and the effectiveness of continence methods on any given day were reported as the most stressful aspects of living with cloacal anomalies, presenting a significant challenge to psychological well-being.

The study noted an overall appreciation from participants for their life-saving care and involvement in treatment decisions. However, interestingly, it also suggested that this gratitude might have interfered with the participants' honest communications with medical professionals during consultations. With this in mind, it is perhaps also important to consider the influence of the context and environment in which the study took place. The fact that it was conducted in a clinical setting, potentially where some participants had received their treatment, might have impacted the nature of their participation in the study, in a similar way to the context of the consultation.

The study concludes that informed decision-making regarding continence management is an important facet of an individual’s experience when it comes to the management of their condition and symptoms. Moreover, this can be enhanced by communication approaches that are based on a nuanced understanding of the individual experience, perspective and the complex interventions that are put forward. While this study specifically focuses on continence outcomes, it is among the first qualitative studies to explore the individual experience of living with cloacal anomalies and the potential psychological impacts. The study also introduces an important focus on factors such as the communication between medical professional and individual. Overall, it underscores how the treatment and management of the condition, along with the condition itself, can be interacting and influencing factors on the experience of living with the condition.

It is interesting to consider how age might interact with the psychosocial experience of cloaca. For example, Sventanoff et al (2022) conducted a systemic review and highlighted how different psychosocial challenges might be experienced at different times in the individual and families life. This included financial and social strain for parents, difficulties at school and behavioural problems for young children, low self-esteem and body image concerns for adolescents and worries about sexual function for young adults (Sventanoff et al., 2022). However, it is important to note that only 3 out of 63 studies in this systematic review contained the search term ‘cloaca’ and only one of which focused specifically on the condition in its own right. The other conditions in the study fall more generally under the umbrella of ARM.

In terms of how quality of life ratings differ across age groups, a study by Grano et al (2010) compared quality of life ratings for children with ARM, including cloaca, under the age of 16, to adults over the age of 16 (mean age 24.0 years). They found that adults reported significantly lower levels of QoL on particular subscales such as body image and emotional functioning as well as for physical symptoms. The authors questioned whether quality of life does in fact decrease over time and what factors could be responsible for this or, whether this difference could be due to improvements in surgical and medical techniques. It is important to note that although this study did feature participants with cloaca, they only accounted for 20% of participants, with the rest of the sample made up of those with other types of ARM. The study also highlights the difficulty in capturing the specific experience of younger participants, with parents in this study completing the questionnaires on behalf of the children under 16 years of age. However, this finding remains important to consider alongside similar findings that have been demonstrated in other studies. For instance, a study focusing solely on ARM and not including those with cloaca, found that the rate of depression was higher in children aged 12 to 16 years compared to those aged 7 to 11 years (Funakosi et al., 2005). Alongside this, rates of anxiety in mothers were reported to decrease as their children aged, with mothers of preschool children demonstrating the highest levels of anxiety. This suggests an interesting trend, that could be speculatively understood as a shift as an individual ages, and they begin to develop more awareness of their condition or take on more responsibility.

Considering this indication that difficulties, or perception of difficulties, increases as an individual ages, it is interesting to consider the period of adolescence which is widely acknowledged as a period of increased vulnerability, characterised by substantial changes in biological, psychological, and social functioning (Larson et al., 1996; Lerner & Foch, 2021; Steinberg & Morris, 2001). There can also be a significant amount of reorganisation in the family system as adolescents begin to negotiate for increased independence (Branje, 2018). From a clinical perspective, it is also the point where transition to adult services may start to happen. In the existing research base, even less is known about the experience and outcomes of individuals with cloaca as they move through adolescence and transition to young adulthood (Svetanoff et al., 2023). It is therefore interesting to consider how normative development and strives for more independence, might interact with an experience of cloaca, particularly around the move to young adulthood from a biological and social perspective.

Summary and research aims:

Overall, the existing research base that focuses specifically on cloaca and the experience of living with this condition suggests that an individual’s quality of life, in terms of their physical, social and emotional functioning may be impacted. However, the evidence is mixed and inconclusive; it relies mostly on a small amount of quantitative studies where quality and robustness is variable, with most studies having small sample sizes and with control group data not always available.

Therefore, despite these initial indications that the experience of living with cloaca could impact on the psychosocial outcomes of individuals and their families, limited conclusions can be drawn at present. Considering the mixed picture, more qualitative research is needed that can start to unpick and explore the impact of the condition and the potential complexities in this area and suggest important areas for further consideration and research. The existing qualitative research is similarly limited, albeit developing, with existing research starting to draw attention to some specific challenges in relation to sexual and reproductive factors (Yu et al., 2022) . Therefore, more qualitative research exploring the individual experience of living with cloaca is needed to build a more comprehensive understanding of the potential psychosocial impact.

As highlighted above, research is especially limited regarding the outcomes and experiences of individuals with cloaca during adolescence and young adulthood (Svetanoff et al., 2023). This period encompasses significant biological, psychological, and social changes, and is critical for establishing identity, independence, and social relationships. It is also the point at which the transition from paediatric to adult services occurs. Therefore, this study aims to explore the individual experiences of young people living with cloaca to inform care pathways and support offered to individuals and families on their cloaca journey.

Method

Design

Considering the small but developing evidence base on the psychological and social impact of living with cloaca, an exploratory, qualitative design was used. The study aimed to contribute to a developing understanding of how young people experience this condition and the potential impact on their lives, moving towards a model of living with cloaca in order to inform care pathways for individuals and families.

The research design, including data analysis, has been underpinned by Constructivist Grounded Theory (CGT). The epistemological position of Constructivist Grounded Theory is congruent with the researcher's own epistemological position which is defined as moderate social constructivism. The researcher holds the position that knowledge is socially constructed to some degree. Moderate social constructionism is considered by the researcher to be a particularly relevant position when it comes to exploring the individual experience of a physical health condition, as is the aim of this research study. Through this epistemological lens, there is a distinction between the 'diagnosis', an objectively definable phenomena, and the subjective experience of the 'illness', which is filtered through social and cultural frames. This position is in contrast to the traditional iteration of the medical model, which holds that disease is universal and unchanged across time, space, and place (Conrad & Barker, 2010).

When considering the experience of living with cloaca, the symptoms of this condition can impact on areas that are foundational to an individual's social sense of self, such as sex, procreation, and digestion. Interestingly, these areas are often subject to moral or societal judgements when any abnormality is present, in function or behaviour. For example, society can scrutinise sexual practices and orientations that diverge from the norm, namely those that exist outside of heteronormativity which includes monogamy (Herek, 2007). Females can also be subjected to narrow-minded standards regarding procreation. Those who do not have children, who have children later in life, or who use methods other than traditional conception often face judgement (Cook & Dickens, 2014). Additionally, issues related to incontinence can carry significant stigma, as incontinence may be perceived as a sign of being unclean or having inadequate self-care. Such judgments are often accompanied by assumptions of poor lifestyle choices or a lack of effort in managing one's health. (Elstad et al., 2010; Rantell, 2023) It follows, therefore, that the social context will be intrinsic to any individual sense making of their condition and how it might impact on how life feels and the amount of 'illness' experienced.

Whilst every person born with cloaca will have objective differences in anatomical structure that permit a diagnosis, according to this epistemological position, no objective truth can be discoverable for how it is experienced. Instead, as Constructivist Grounded Theory posits, there are multiple realities and research provides an interpretative representation of those realities, as opposed to an exact reproduction (Charmaz, 2006).

Ethical considerations and approval

Confidentiality of participants

Due to the rare nature of cloaca, it is possible that participants could be identifiable, based on the specific details that were shared, to others who also have experience of this condition. Therefore, it was important to consider the necessary steps to maintain the confidentiality of participants. Firstly, descriptions of the participants were kept to a minimum, and only generic participant information, such as the age range of all participants, was included instead of specific ages of each participant. Secondly, interview data included in the research was anonymised, including direct quotes from participants' transcripts used to directly illustrate themes identified in the analysis.

It was considered whether participants could be given aliases or numbers to attach to the direct quotes included in the research. However, it was determined that this could increase the likelihood that individual participants being recognised by others in the cloaca community. Therefore it was deemed important not to do this in order to maintain confidentiality. All participants were made aware of the steps that were taken to maintain confidentiality in the participant information sheet and consent form. It was also important to highlight the limits of confidentiality, specifically that, despite these steps, there remained a possibility that participants could be identifiable. Therefore, at the end of the interview, participants were asked if there was anything they shared that they would prefer not to be included in the research. They were also informed about the period during which they could withdraw their participants. This information was also outlined in the participant information sheet and consent form.

Moreover, participants were informed about the circumstances that might require confidentiality to be broken, as well as the subsequent protocol that would be followed. If a participant disclosed harm to others, they were made aware that details would be discussed with the supervisor to determine the most appropriate course of action, which might include breaking confidentiality. If a participant disclosed harm to themselves, they would be encouraged to contact relevant support services. If they would not be willing to do this, they would be informed that the details will be discussed with the supervisor to determine the most appropriate course of action, which might involve breaking confidentiality, of which the participant would be informed.

Potential for distress

It was acknowledged that participants could become distressed during the interview when speaking about their experiences of living with cloaca. Therefore, participants were informed that they could take a break from the interview or withdraw from the study at any point, up until two working days after the interview had taken place. During the interview, signs that the participants were becoming distressed were monitored and the opportunity to pause or stop the interview would have been made available if required. The researcher who conducted the interview was clinically trained and had the appropriate skills to manage distress in the interview should it have arisen.

Ethical approval

Ethical approval was sought from the University of Leeds School of Medicine Research Ethics Committee (reference: MREC 22-087) and granted on 2nd August 2023. An application was made for an amendment to the application and ethical approval for this amendment was granted on 16th October 2023. No further ethical approval was required.

Eligibility

Initially, participants were eligible for this study if they identified as having a diagnosis of cloaca and were between the ages of 16-35. Participants were also eligible for this study if they were a caregiver of an individual with a diagnosis of cloaca.

During recruitment, the researcher was contacted by potential participants outside of the upper age limit. Therefore, after further consideration, an application for an amendment was made to the ethical review board to extent the upper age limit from 35 to 40 years of age. This was deemed to be important to enable more voices and experiences to be captured in the study without a compromise to the focus of the study.

There were no exclusion criteria for geographical location. Participants with other congenital malformations (such as other types of ARM), in the absence of cloaca, were excluded from the study. However, there were no exclusion criteria for other medical conditions that co-occurred alongside cloaca.

Recruitment

Participants were recruited with the support of Max’s Trust, a UK based charity that provides support and information to adults, children and families affected by ARM.

Recruitment information that highlighted the inclusion and exclusion criteria, was circulated via posts on social media and emails to all contacts who had signed up to their mailing list and had previously agreed to receive information from the charity. Information about the study was also advertised on the website of the charity. The recruitment information included a link to an online form where interested participants could enter their contact details, if happy to be contacted by the lead researcher. The online form also included a copy of the participant information sheet.

Alongside this recruitment channel, it became apparent that as recruitment progressed, participant-led recruitment via 'word of mouth' was also taking place. The first few participants who took part shared the details of the study to online groups and forums that were not easily accessible or visible to people without the condition, or a personal connection to it. These channels had reach across an international community.

Procedure

One-to-one semi-structured interviews were conducted with participants. Considering the geographical spread of participants, interviews were conducted via video conferencing platforms such as Zoom and Microsoft Teams. Participants were sent the participant information sheet and the consent form by email prior to the interview. Before the commencement of the interview, participants then had opportunity to ask any questions or air any concerns and their consent was gained. The interviews were recorded and automatically uploaded to the University's secure online drive. The average length of the interviews was 1 hour and 7 minutes.

The interview schedule was loosely structured into three sections: looking back, the present and looking forward. All participants were asked broad, open-ended questions and were encouraged to speak freely, with care taken to notice and explore any underlying assumptions and implied meanings. This was facilitated through prompts such as ‘can you tell me more about that’ and ‘can you tell me what you mean by …’. Table 1 provides an example of some of the questions asked to all participants, ensuring that there was space within the interview for spontaneous descriptions and follow up questions that remained close to the participant and their experiences. For the caregiver interview, questions asked with a slight change in positioning i.e. substituting ‘your experience’ for ‘your daughter’s’.

Table 1. Example of interview questions

Looking back *Can you tell me about your early experiences with cloaca?* *What was it like when you became aware of cloaca?*
Present day *Can you tell me about your current experience of cloaca?* *How do you view cloaca now? Has that changed?*
Looking forward *Can you tell me about your hopes for the future; do you anticipate any challenges?* *How do you think you have grown or changed as a person as a result of these experiences?*

CGT methodology encourages self-reflexivity in the research process, ensuring that the researcher remains mindful of what they bring to the study in terms of experiences, biases, and perspectives and how this might influence the data collected and conclusions made (Charmaz, 2006). Therefore, following each interview, the researcher engaged in a period of reflection that was facilitated by the writing of memos. This provided an opportunity to reflect on the initial impressions of the interview which included the processes at play, any felt sense of emotion within the interview and a consideration of the relationship between participant and researcher, including how the research may have been perceived by the participant and what influence this might have had on process. An opportunity for reflection in research supervision alongside the writing of the memos was regularly utilised. Figure 1 is an example of an excerpt from a memo written following an interview.

Figure 1. Example excerpt from a memo written following an interview

I wonder about the language used, the level of medical detail and the time spent talking about the medical/physical detail in this interview (I felt a little squeamish at some points!), the participant had a really impressive grasp on the medical stuff and was keen to describe things to me in more detail than I've heard before. In parts it almost felt as if she could be joining with the medical team, and I wonder about the effect of this, almost as if she was separate from her experiences, her own body? A mix of interest, way of coping, being in control of the narrative perhaps… reflective of the complexity, how much there is to get your head around about your own body, and how used you could become to those medical appointments and the way you might hear language being used about your body. I am interested by "my own body", as opposed to just "my body".

The memos and research supervision also provided an opportunity to reflect on the questions and topics discussed. In keeping with the iterative process of CGT, the researcher was therefore able to create a trail of informal analytic notes about the data, and use them to progressively refine the focus of subsequent interviews; for example, decisions were made to invite more discussion on specific areas in later interviews such as the participant's relationship to sex, experience of vaginal reconstruction and experiences of medical anxiety or trauma using questions to invite disclosure such as 'some people have spoken about their experiences with intimate relationships/sex/their reconstructed vagina], have you experienced something like that?'.

Analysis

As noted above, data were analysed using CGT methods, which involves an iterative, reflexive, and constant comparative approach to develop a theory that accounts for the perspectives and experiences of the participants. Analysis began after the first interview. Data were transcribed and the process of analysis began in between each interview. The transcription and coding were conducted manually by the researcher; this was deemed to be important to support the researcher's immersion in the dataset, in keeping with the methodology which promotes the researcher as an active part of the process, as opposed to an objective, more detached observer (Charmaz, 2017; Mills et al., 2006).

Initial line-by-line coding was the first step of analysis, which required close attention to the data, as the researcher named each line of the data as an action, using '-ing' words. The use of gerunds at this stage of analyses, as is recommended by Glaser (1987), supports the researcher to stick closely to the data and helps with the detection of process. Table 2 is an example excerpt of initial coding from an anonymised transcript.

Table 2. Excerpt from an interview transcript demonstrating line by line coding

I had surgery scars on my stomach as well. So that was another thing. I don't want anyone to even see me naked or anything like that at all. I was embarrassed. So, it was more so that honestly, like I don't want you to look at it, don't look at my vagina.	Being aware of her scars Not wanting other people to see her naked Feeling embarrassed of her naked appearance Anticipating judgement about her genitalia

The second stage, focused coding, involved the development of codes that were less closely attached to the data but instead encapsulated larger segments of the data. At this stage, decisions were made about codes that made the most analytic sense and best represented what was happening in the data. Throughout analysis there was a constant comparison between and across interviews as possible similarities, differences, patterns, and relationships emerged; data was re-examined and compared to the developing focused codes, helping to refine them. Codes then began to be raised to themes. Themes encapsulated overarching concepts in the data, bringing together common aspects of experience and patterns evident in several codes. A fundamental part of this process was the use of memo writing. The researcher used memos as an opportunity to treat focused codes as tentative categories; exploring their potential as a theme by writing narrative statements that experimented with definition of the category, the properties, conditions of occurrence, maintenance, changes, and the relationship with other themes. An understanding of the young person's experience of cloaca then began to emerge.

Axial coding, a type of coding proposed by Strauss and Corbin (1998), is a strategy used to categorise data based on relationships, as different codes are examined, organised into categories and their properties and dimensions defined, centring around an 'axis'. Charmaz (2006) highlights that, within Constructivist Grounded Theory, axial coding is an optional process. An important consideration is whether the pre-set, structured frame offered within axial coding could in fact be limiting. Therefore, in this analysis, less emphasis was placed on the strict adherence to axial coding. Instead, a flexible approach was adopted, allowing greater room for uncertainty, and facilitating the fluid development of themes as they emerged from the data.

Results

Participants

Nine participants were recruited to take part in the research study. Due to the rare nature of the condition, it is possible that participants could be identified by other members of the cloaca community, therefore a group profile of the participants was included to highlight the key demographics of the participant group whilst taking care to maintain the anonymity of the individual participants as much as possible.

Eight of the participants had personal experience of cloaca and one participant was a caregiver. The mean age of the participants with a diagnosis of cloaca was 26.25 years, with the youngest being 20 and the oldest 38 years at the time of interview. Of the entire sample, three of the participants lived in the United Kingdom, two participants lived in the United States, one participant lived in Australia, one participant lived in Canada and one participant lived in Central Europe. All participants, except the caregiver, had been diagnosed with cloaca, and three participants disclosed associated congenital anomalies under the umbrella of VACTERL association, which is defined as a group of anomalies that appear together more often than would be expected by chance, and in the case of VACTERL includes other congenital abnormalities that affect the vertebral, cardiac, trachea-oesophageal, renal or limb area (Schwarzer et al., 2024).

Findings

Three major themes emerged from the data. Each of the themes included several sub-themes, as detailed below.

Table 3. Table of themes and subthemes with number of associated quotes and participant distribution

Theme	**Subtheme**	**Number of quotes**	**Number of participants**
Sense of the bodily self	*From the medical to the sexual body*	43	9
	The body as public and medical property	25	7
Medical complexity	*Invisible burden work*	36	8
	Relationships with medical professionals	24	9
	Medical anxiety and trauma	30	7
Growing up with cloaca	*Navigating disclosure*	26	8
	What's been lost, what's been gained: who am I now?	37	8

Theme 1: Sense of the bodily self

1a. From the medical to the sexual body

For those with cloaca, the boundary between the medical and sexual body often feels blurred. Participants described experiences of examination, intervention, and symptom management, and how these have unintentionally contributed to the medicalization of their intimate areas. This has subsequently impacted their experiences of sex, their ability to connect with their sexual self, and the development of a sexual identity.

For example, from a very young age, participants described how their personal and intimate areas were the focus of conversation and intervention. In medical contexts, life-saving surgeries involving their private parts were followed up by ongoing examinations in a medical context, particularly if complications or infections arose. In the hospital, participants described how being looked at or showing their private parts to doctors can be commonplace, to the extent that a doctor approaching this differently was particularly memorable to one participant:

"She came the day before on her free day to meet me because we hadn't met each other and she said she wouldn't want anybody to look down there if they were a stranger either"

Moreover, symptom management interventions may continue outside of the hospital environment, perhaps initially led and supported by parents, such as dilation, whether that be anal or vaginal, or bowel management techniques such as flushing which involve the sitting of the child on the toilet, as their bowels empty.

In the present day, all participants highlighted the extensive day-to-day management of cloaca symptoms, including lengthy bowel management, concerns about flatulence and bladder incontinence. Moreover, participants highlighted struggles with painful periods and unforeseen and unexplained medical experiences such as nausea and infections. These everyday realities of living with cloaca were felt to be barriers to engaging in sexual relationships.

From a practical standpoint, one participant explained:

"How do you even have the time when you're spending so much time in the bathroom, or you're doubled over in bed trying to figure out what's going on?"

From a psychological perspective, symptom management often involves dealing with bodily matters that do not feel conducive to feeling sexual; another participant noted that:

"It's not very sexy stuff."

Moreover, it appears that this fear of how others may see them is sometimes based on personal experience; some participants have experienced judgment from others about their sexual identity and how the symptom management involved in living with cloaca impacts on this. Some participants describe how colostomy bags at certain points in their lives so far, through choice or necessity, have attracted attention and judgement from others. This was not always from other young people or the public - one participant described the negative opinion from a doctor, following her decision to opt for a colostomy bag:

"He told me that I should get my colostomy reversed because it's the only way I'll find a husband to get married"

Within this statement, there is an implicit assumption that necessary symptom management interventions, such as a colostomy bag, has a detrimental impact on sexuality and attractiveness, and consequently, the medical and the sexual body cannot coexist. Regardless of the fact that, for this participant, they felt that a decision to have a colostomy bag has significantly improved their quality of life in other areas.

All of the participants spoke about the medical procedures that they underwent in order to achieve a functioning vagina, from a menstrual and sexual perspective. These procedures included reconstructive surgeries and a process called dilation, which involves the regular stretching of the vagina. Participants spoke about the medical advice they received to undertake the dilation daily, or as close to daily as possible. However, for the majority of participants this process is experienced as uncomfortable, sometimes painful, and time-consuming:

"I did it for a little bit, but I just couldn't like I just felt so uncomfortable, I just couldn't do it… it's not nice and it hurts."

Some participants described significantly distressing experiences with dilation in a medical context:

"I had a gynaecologist basically who messed up the dilation really badly, when I was telling her to stop, it was terrible – I got a bad tear and I had to be admitted to hospital with a bad infection, it was just a really awful process"

Other participants recalled being a younger child and experiencing dilation being done to them by a parent:

"I would say the most traumatising part was the stretching of the vagina from my mother at home while I was already able to remember this stuff"

These medical interventions, regardless of intent, could be experienced as painful and invasive and have, for some participants, creating distressing associations between the intimate parts of the body and medical interventions that they have endured. Consequently, some participants expressed a felt sense of disconnection from these areas of their body and saw this as a psychological defence.

"I'd just rather not think about my intimate parts and what happened to them… that area doesn't exist in my head"

The medicalisation of their intimate parts can make it difficult to transition from the medical to the sexual context; one participant described a desensitisation to sexual touch, finding it difficult to distinguish between medical and sexual touch and therefore finding it difficult to experience sexual touch as enjoyable or stimulating.

"*I think like because I'm touched so often and so regularly in medical settings, you're so turned off, you're so disconnected to then being in a romantic situation where someone is touching you, yes obviously they are touching you in a different way, but how is it really that different?*

In fact, touch in a sexual context could sometimes be experienced as unsafe; transporting individuals to the medical setting where touch has meant something very different, functioning in a similar way to the process of a trauma memory. One participant described that for sex and intimacy to be safe and fulfilling, they need an emotional connection with that person

"…otherwise it almost feels like threatening and scary and I'm just very disconnected"

Additionally, one participant described how being looked at in a sexual context can have a similar impact to touch, highlighting how the medical and sexual sense of the body can become inextricably linked, or perhaps how powerful the sense of the medical body is and how difficult it can be to develop and connect with the sexual self.

"If they are looking at me too much, at my reconstruction, down there, if they are looking too much then I kind of flick into oh this feels like an examination, and it gets in my head a bit too much"

Understandably, this provides barriers to the extent that participants feel free and able to explore their sexual self and experience sexual freedom. The majority of participants describe long-term relationships as the first time they have been able to progress on their journey to bodily acceptance and even start to experience pleasure from sex.

1b. The body as public and medical property

Participants described how growing up with cloaca has meant that their bodies have enduringly been something to be examined and interacted with, resulting in a general feeling that their bodies are not their own. Participants described how their bodies can be the subject of others' curiosity; with experiences of their bodies being commented on, questioned, and interacted with in both a medical and non-medical setting.

"My body isn't mine, it's always something to be examined by someone else"

In a social and education setting, participants described that, from a young age, they experienced an invasion of their personal space, with others demonstrating a right to physically interact with their bodies. For example, some participants reflected on experiences with peers at primary school who, motivated by curiosity, would invade their personal space, and prod the bits of their body that might signify difference:

"Some kids would squeeze my pad or poke and press my scars"

Other participants described being subject to rumours and misinformation about their body, becoming the focus of other people's gossip:

"It spread around school was that my bum was fake and was made out of plastic, and I had people come up and want to touch my bum because they thought it was fake"

And

"[In response to the rumour] I remember this girl messaging me on Facebook asking me, were you born with a penis?"

Both of these examples illustrate how some participants can struggle with other people assuming control of the stories about their bodies. It appears that participants can experience both physical and psychological intrusions as other people comment, question, and command the narrative around their bodies, perhaps driven by curiosity or fascination with any cloaca related difference. There is a sense that, for those with cloaca, the body can be treated as if it exists in the public domain and can subsequently become subject to public interest and scrutiny.

Some participants described similar experiences of grappling with a lack of control and agency over their body in a medical setting. For some, aspects of their medical care, particularly surgeries and decisions that were made when they were very young - often due to unforeseen complications and difficulties with incorrect diagnoses - were hard to reconcile as they got older and became aware of them:

"I was really angry when I discovered they had removed my fallopian tubes"

Participants described how their bodies and their intimate areas have been the subject of medical examinations and interventions from a young age.

"It would be sore, and it would be in your private areas, and you know they would get a rubber glove and they would shove their finger up your bum"

This sense of being poked and prodded in a medical context, participants note, is a necessity of their condition. However, some participants reflect on how the complexity and rareness of cloaca can attract a significant amount of medical interest and curiosity:

"If you have a rare condition, it's always like a lot of doctors you don't know who are there and most of the time you are not really asked if you are comfortable with a room full of doctors"

Similarly to above, these experiences can contribute to a sense of the body existing in the public, as opposed to the personal domain. The body can therefore become something to be looked at and worked out, a spectacle of medical interest and novelty. Some participants reflected that this lack of ownership over their bodies has made it difficult for them to find and use their voice in medical settings. They were either unaware of their right to speak up or reluctant to be perceived as the *"difficult"* patient.

"I had this pap smear like two months ago and I had this medical student, a male medical student the same age as me, in there watching the pap smear and I was so uncomfortable but if I knew from that young age, that you can ask for what you need, and it's okay, and you don't have to feel embarrassed, like you don't have to say yes to everything"

Furthermore, one participant reflected on how these experiences in a medical context have impacted on her ability to have an awareness of her boundaries in personal settings. This participant described a difficult experience of non-consensual sex, and she reflected on how the sense of lack of ownership over her body contributed to this:

"I never felt that my body was about me, it was about other people and I think that with that first experience being non-consensual, it was also because I was so used to doctors always have a right over my body"

Another participant, whilst similarly reflecting on these challenges of cloaca shared a positive experience with a medical professional.

"She [the doctor] came the day before on her free day to meet me because we hadn't met each other said she wouldn't want anybody to look down there if they were a stranger either"

The validation of the strangeness of having to both discuss and grant access to intimate areas of the body with an unfamiliar person appeared to help the participant feel safer with the doctor and less exposed by the experience.

Theme 2: Impact of medical complexity

2a. "Invisible burden work"

Due to the complexity of the condition, cloaca requires ongoing multi-disciplinary involvement of professionals and the development of multi-faceted symptom management approaches. The first years of life are often marked by complicated surgical procedures, and interventions continue as the individual ages to ensure adequate bowel, urinary, and gynaecological function.

Each individual's intervention and symptom management journey can be unique, as the constellation of symptoms can vary across individuals. Moreover, as an individual ages, complications can emerge, particularly during puberty when gynaecological issues might be discovered. Unforeseen complications or unintended side effects of treatment approaches can arise, requiring further examinations or changes in symptom management techniques. An example of this unpredictable, trial-and-error approach is illustrated in the quote below:

"Off and on throughout this journey I've had issues with my bowel management routine, with it taking too long and not working very well or causing me accidents,

erm and stuff like that, and so I was doing well for a little bit and then it always seems like I'm always off and on, like I'm doing really well and then oh the solution has stopped working, okay what else can we try and that's been going on for a couple years now."

The experience of managing cloaca appears to reflect the complexity of the condition itself, necessitating involvement with various medical professionals, sometimes across different parts of the healthcare system, and the engagement in changeable interventions and treatment routines.

The participant who was a caregiver reflected on her experience of being thrust into a complex health system when her daughter was born:

It was just there was so many specialists, so many levels of care…. so, it was very overwhelming. And I just was sort of thrust into this health system that I didn't understand

This participant continued to reflect on the amount of work involved in managing a condition like cloaca, becoming close to a full-time job:

"So, I approached it like a job, so I'm like, OK, we need a printer I need a photocopier. So, we got one of those photocopier printers. I'm having a desk…so anytime during nap time, I've got to find the right support"

This participant named all of the work involved in managing cloaca care as *"invisible burden work",* signifying an important, yet mostly overlooked, component of living with a complex condition such as cloaca. This participant identified particular challenges, alongside the medical, such as fighting to secure the appropriate funding and support for her daughter to attend nursery, highlighting how cloaca often fell through the gaps of existing provision in the country she lives in:

"I was like, well, how am I going to go back to work because I'm going to have to find a childcare that could change an ostomy. And so I called like everywhere and

they're like, oh, well, you need, like, a specialised childcare, you know. But the only specialised childcares are for, like, people who are like, fragile or have cognitive delays. With cloaca, you're medically complex, but you don't have mobility issues… you're not fragile but you're complex"

As an individual grows older, this responsibility for the provision of cloaca care transitions from paediatric to adult services and, alongside this, there is an accompanying shift of responsibility from caregiver to the young person with cloaca, as services no longer prioritise the involvement of families. The caregiver participant described their worries about this forthcoming shift for her daughter:

"It's so much and we worry about that because it's so much work. It's so much work that's not living. What will she do when I'm dead?"

This shift from paediatric to adult services, with the responsibility for the *"invisible burden work"* shifting into the hands of the young person with cloaca was also felt to be significant by participants with cloaca:

"There really wasn't a lot of help after you became an 'adult' in regard to care. So, my transition from paediatric care to adult care was terrible, there's no other way to put it. Everything is so different and then the number one thing that changes is that you are advocating for yourself"

Several participants shared this experience of transition to adult services being difficult, as they take over responsibility for managing their own care in a context of services that require a lot of coordination:

"I feel like I have taken over my mother's role and I feel everything is all disjointed"

Participants spoke about how their experiences of this *“invisible burden work”* includes advocating for themselves, in an adult medical world where knowledge and understanding of their condition is limited.

“The biggest challenge as an adult is the medical professionals like every time, I go to a doctor I have to explain to them what my medical condition is, and I think that’s quite hard”

One participant voiced how this can feel lonely and isolating, as if the sense of being known by people and feeling listened to that she had experienced in the paediatric world had been lost in the adult setting:

“When I was younger, I was taught to tell a doctors if something didn’t feel right with my body. Now if I do that as an adult, it’s like, who’s listening to hear that?”

Some participants reflected on this sense of “*invisible burden work”* in the context of their experiences with pregnancy. One participant, wanting to seek out more information about their future prospects of pregnancy, described their attempts to do this:

“I have been trying over the past few months to set up appointments with doctors and professionals to be able to get a better understanding of it but it’s really hard because erm there aren’t many experts in this field”

Another participant described her proactive efforts to secure safe and appropriate medical care throughout her pregnancy, including appropriate medication:

“I was going through IVF and these enemas I didn’t think were safe, so it was me, I looked up research articles and stuff; it was pretty much a case of me finding my own medication erm which is quite concerning”

Moreover, she described using personal contacts, to secure a face-to-face meeting with her consultant to ensure she was listened to, and that her needs were met, following an initial lack of reassurance and understanding about her condition:

"My partner's aunty who was a midwife, she's really high up phoned the head midwife because she thought that is ridiculous, she doesn't have a birth plan, she emailed a professor who emailed her back in the morning and also I got in contact with my surgeon who did the original surgery to send the consultant the notes"

In the meeting with her consultant, the participant described advocating for herself, having equipped herself with a significant amount of research and how she exhausted herself in order to ensure appropriate care:

"So I brought my folder with my notes, I said I want progesterone, I want to know there's going to be a c-section. I feel incredibly proud of myself but that took a lot of time and a lot of exhaustion when I was at risk of having a miscarriage"

As an individual grows up and moves from child to adult services, the "invisible burden work" often shifts from caregiver to the individual. The work involves medical advocacy and the navigation of systemic gaps to ensure that care needs are met and demonstrates the resilience required in living with and managing cloaca.

2b. Relationships with medical professionals

All participants described the complexity and uncertainty involved in growing up and living with cloaca. Due to the rare nature of cloaca, participants often had the experience of learning about the condition at the same time as the medical professionals treating them:

“What the heck is going on, what is this! They had never seen it before”

The condition can manifest in ways that are inter-related and unpredictable. Participants describe dealing with infections, unexplained symptoms and a continuous *“trial and error”* approach to symptom management

“It was more like here’s plan a,b,c,d,e,f,g… there’s just no other way to put it than to prepare for everything”

It is interesting, therefore, to consider how participants’ experience of cloaca has been shaped and framed by their interactions with medical professionals who are similarly confronted with the complexity and rarity of the condition.

All participants describe how their experiences as a young adult, in the context of adult services, have been complicated by this complexity. One participant highlighted how regularly they are referred to and labelled as “complex”:

“I think I made a joke of it the other day that I just constantly hear off medical professionals, ‘you’re so complex’”

She reflected on how this can have the effect of either being treated as a ‘case’ with curiosity and novelty, or feeling rejected as they resist taking on her care as they are intimidated by the complexity.

“Then they'll think I don't want to deal with her or my god I want to have this person as my patient because I'm so interested in the complexity and their anatomy, you know?”

However, most of the time, she reflects, it is the former and subsequently she can find herself waiting for and struggling to obtain support

"Well, they sent me to this other hospital, and they said, you're too complex, we're sending you back... and I was like, waiting months and months for these referrals. So, it can get annoying in that way where it's like they try to throw me to other people because they don't want to deal with me. That's frustrating"

Participants describe how, especially as they move from the paediatric to the adult setting, the complexity and rarity of cloaca means that medical professionals with experience and knowledge of cloaca seem to be harder to find. Therefore, they are often confronted with the task of educating the professionals responsible for providing their care. Subsequently, some participants describe a process where the idea of the infallible and all-knowing doctor breaks down and, as a result, they can lose trust in doctors:

"It's like realising the person who is supposed to have all the answers has never even heard of your condition"

For some participants, this can be unsettling and can contribute to a sense of feeling alone and unsupported:

"I guess what I didn't realise as a kid was how much doctors didn't know, but now, I like have the fuller picture that I'm the only one in charge of myself"

However, for other participants, this presents an opportunity for a renegotiation, and a rebalancing of power that can have positive outcomes across different areas of their life, with doctors becoming partners in care as opposed to all-knowing authorities:

"I'd always be more in hospital than in any other environments, like with my friends etc and even now that's the case, so if it's in the dominant setting, where I'm

learning those skills, like that's going to be most helpful and if it's where there's the greatest power dynamic between patient and doctor, like that's going to be where it's going to be most helpful"

Another way in which the complexity and rarity of cloaca can impact upon participants' relationships with medical professionals is a sense of being dismissed or even made to feel stupid. Some participants have encountered medical professionals reacting defensively, speculating that this may be due to the professionals' discomfort with their own lack of knowledge or experience with such a complex and rare condition.

"I think for me it's recognising that they [medical professionals] actually do get frustrated, but I think some of them took it out on me"

Unfortunately, this has sometimes led to a lack of empathy and understanding towards the patient. For example, one participant described a challenging experience with a smear test:

"*The whole time I was cramping and apologising to him that he's not able to do the examination and it was really awkward and he was angry at me that he could not do the examination and I was almost crying because of the experience"*

Once again, it appears there is a tendency for cloaca to take centre stage and the person with the condition can feel obscured by the complexity, curiosity and novelty. This focus on the complexity can have a detrimental impact on patient, doctor and the relationship between them, as one participant summarised:

"We need to stop thinking about it as a condition where it's like 'the most difficult paediatric surgery that exists of all the paediatric surgery' – no - these are people"

It is important to highlight that not all participants describe wholly negative experiences. In fact, some participants reflected on the gratitude and appreciation they have for their care, acknowledging the life-saving care they received in the context of medical complexity:

“Now looking back, I’m incredibly grateful because there were so many things that… well a lot of complex stuff happened, but I was OK”

Others spoke about how their unique constellation of symptoms required a variety of management approaches, some of which were recognized as essential and transformative in changing their experience of the condition and its limitations:

“[The cecostomy] gave me so much quality of life”

Moreover, it seems that positive relationships and experiences were most achievable when participants had been able to maintain contact with medical professionals that have been part of their care team since childhood which suggests a sense of being known is reassuring and containing:

“I’m very lucky that I’m still in contact with some of them, and my gynaecologist is still the same from when I was born”

Although, even without continuity of care, some participants used words such as *“understanding”* and *“kindness”* when describing positive interactions with medical professionals. In the context of complexity, it appears that the opportunity to slow things down, with time to connect with a medical professional is greatly valued:

“I was like wow I always thought my doctors were good but she was brilliant… we had a really good talk and yeah she just had the patience I needed”.

2c. Medical anxiety and trauma

For participants living with cloaca, the complexity of the condition means they have been, from a very young age, subject to frequent and numerous medical procedures, examinations and interventions. One participant, in describing her medical journey, summarised her experiencing by saying:

"When I was younger, I was mostly on my back"

This is a powerful quote which illustrates the substantial amount of time that she has spent in hospitals and other medical settings, which is a common experience for someone living with cloaca. Consequently, all participants shared a range of emotional reactions from their experiences, spanning the spectrum from medical anxiety to what could arguably be understood as medical trauma.

Some participants reflected on memories of time spent in hospital as young children, with little understanding of what they were enduring:

"I remember being held down for IVs by like five different people"; "I remember waking up from the hospital procedure, they'd put me under anaesthetic and waking up and my mum wasn't there and shouting for my mum"

Outside of the hospital environment, other participants reflected on challenging psychological aspects of their symptom management routines. There is a sense of uncertainty and anxious anticipation, with participants describing a state of vigilance as they worry about unexpected challenges such as infections or accidents. This could be a similar process to what is seen in conditions like health anxiety, where vigilance towards threat, and difficulties to disengage from threat are key maintaining processes:

"I never knew how bad the pain would be or how long the meds would take to work because it would be anywhere to 6 hours to never"; "I feel like there's a lot of uncertainty, especially if you're dealing with medical issues, like what if I have an accident, what if I pee myself, what if I get another infection"

One participant shared the anticipatory anxiety and the subsequent difficulties with sleeping she can experience in the time period leading up to an examination or intervention that requires her to have anaesthesia:

"When I found out I had to go under anaesthesia again, I wouldn't be able to sleep for the entire week leading up to the appointment because like going to sleep in my brain was equated with going under anaesthesia which has all of its unknowns like what if I wake up in pain, and all of that stuff"

Another participant described how this can cumulate in a generalised type of medical anxiety that manifests itself whenever they have an appointment with a medical professional:

"But now like I have really bad medical anxiety like even a doctor's appointment that I know is just like a talking appointment like my anxiety spikes so high"

Another participant shared this observation and gave an insight from an online group they are part of, where others with cloaca also share their experiences. This highlights the widespread prevalence of medical anxiety within the community:

"I'm in several adult groups and stuff and there's regularly posts about, 'oh I'm having a doctor's appointment, keep me in your thoughts, anxiety is high right now"

Some participants shared how their experiences of medical interventions have left them with strong sensory associations that, when encountered, act as reminders of the interventions and evoke a continued sense of anxiety and distress:

"I don't think I'll ever forget the smell of the oxygen mask or what anaesthesia smells like. Sometimes even, erm, cause we usually stay in the same areas when we go for appointments and stuff, so even just the drive from the hotel to the hospital, it triggers my anxiety a little bit"; "I remember after my surgery, I would hear the sound of an automatic hand sanitiser, like I was in the airport, and I heard that sound and immediately I was on edge"

The experiences, as described in the quotes, function in a similar way to a trauma memory and suggests that the participant is likely to have experienced a significant amount of distress in these contexts.

One participant described going into *"freeze mode",* to help them cope with the unease and distress that they may experience when undergoing frequent medical interventions:

"When I'm in the hospital with a doctor I go in freeze mode and I'm like the perfect patient and so I'm not able to say that I don't want him to examine me, I just go along with it"

If seen through the lens of medical trauma, this can be understood as an adaptive response to a significant threat. Moreover, it is interesting to consider how this sense of pressure to mask and endure pain and discomfort is shared among a number of participants:

"I'm just lying there and taking it and it's because it's all I've known, and I've learned that if you panic and freak out it's just going to make it worse for you and

it's going to make it worse for everyone around you so just be calm you know. So, then the issue is that these people think I'm fine, but like, no, mentally, not fine"

This strategy, whether automatic or intentional, can help the participant to endure procedures in the short-term. However, it can make it challenging for a medical professional to recognise and respond to their needs sensitively and appropriately. Freezing or masking pain and discomfort can both manifest as a symptom of feeling helpless and vulnerable in these situations and can also reinforce a patient's sense of helplessness and vulnerability.

Theme 3: Growing up with cloaca

3a. Navigating disclosure

This sub-theme captures a common experience among participants: how they navigate disclosure, and how this interacts with their adolescence. Initially, the majority of participants keep cloaca a closely guarded secret for several reasons: to defend against anticipated judgment and shame, and to assert boundaries and control over their bodies, and the narrative of their bodies, in the context of a condition which makes this challenging. However, it appears that as participants move through adolescence, the impact of anticipated judgement and shame can lessen and they can become more open about their experiences, finding empowerment through connection with a supportive community.

Many participants told how they worked to keep cloaca a well-kept secret as they were growing up. Some described anticipation of negative judgement from others, if the truth about their condition was to be shared or known:

"If people find out about this then they will laugh at you and you're not going to have friends and people aren't going to like you, that kind of thing"

For some participants, this assumption was based on observing others react negatively to certain symptoms they experienced, such as flatulence.

"Eventually I was like, I can't go to gymnastics anymore because it was too much – people would laugh, and they would make fun of me"

Others spoke about a resistance to cloaca obscuring or defining their identity. They wanted to stand apart from their condition, not wanting their actions and emotions to the attributed or overshadowed by their condition, and so chose not to share information as a way to achieve this:

"For other people to not know too, that was just always something pretty fun in my head, so no one is judging me or treating me differently. If I decide to cry, it's because we lost a sports game, I'm not crying because my stomach hurts, I'm crying because we lost"

Moreover, there is a sense of wanting to be recognised for their own abilities and achievements, without the knowledge of cloaca influencing others' perceptions and being looked at through a lens of pity or sympathy:

"I was like okay I don't want anyone else to really know, I want to make it on my own merits"

Resistance to sharing information about cloaca appeared to be complicated by its complexity, as some participants described not necessarily understanding the entirety of their condition, nor having the language to describe it. Their sense was of being overwhelmed by it themselves, which prevented them from sharing it with anyone else:

“I was very very very hesitant and it’s because it’s such a story, right, you have to sit down like this for quite some time, to really understand it all”

Moreover, limiting disclosure also seemed to be a strategy some participants used to assert boundaries, regain control, and carve out personal space in response to a condition that can feel invasive, especially throughout adolescent years:

“Because she [mum] knew too much when I was younger and saw too much of me. I like to keep some things to myself, and I think that's probably why”

The caregiver participant described how her daughter who, at the time of interview was in the middle of adolescence, had stopped sharing information and having conversations with her about aspects of her condition:

“She does tend to not want to talk about anything, she doesn’t want to talk about the next surgery that's coming up the vaginoscopy or like any sexual activity she will not talk about that… And she keeps saying, mommy, you keep talking about my poo and my pee and that is really weird”

In fact, this participant sees her daughter keeping everything in and wonders about the impact of this:

“She’s also developed headaches every day... I don’t know if it’s with the stress or because she keeps everything all in”

Limiting disclosure and ‘keeping it all in’ was also described by those with Cloaca, and this could be a lonely experience, a ‘turning inwards’:

“It was something I hid and dealt with myself independently, so I think I internalised a lot and dealt with it privately”

There is a sense here of it being an emotional burden, but also a constant source of stress to closely guard information about their condition and this part of themselves:

"*I kept it so well hidden; it was such a weight on my shoulders that other people didn't find out"*

However, although limiting disclosure might have been an attempt to assert boundaries and regain control, one participant reflected that, in actuality, the constant pressure to keep everything hidden actually contributed to her feeling more out of control:

"Growing up, if there's a secret and you're trying to hide something you constantly feel out of control cause you're just trying to hide it, hide this big secret and it feels out of your control"

As participants moved through adolescence, the majority shared a sense of liberation from this attempt at tight information control:

"Everyone around here knows now and they just don't care, it's just my norm, which is like a weight off my shoulders just being open"

In fact, connection to the wider cloaca community, either through online forums or groups on social media, or meeting others in person at events such as residential camps for children with a range of conditions similar to cloaca, appeared to be a significant contributing factor to this shift. They moved to a different position of empowered openness, as participants described a sense of reclaiming their narrative:

"I remember asking other campers like, what do you say when people ask you when people ask you about your scars and what do you say in simple terms? One of them

turned around and said well I just tell them I got bit by a unicorn or, that it was a shark attack… I was like wow, you can just tell people whatever you want them to know, as much as you want to"

Another participant shared how a difficult medical experience, where she became aware of her mortality, helped her reach out and connect with the wider community and realise the impact of sharing her story and experiences:

"It was a big drama, and, in the end, I needed emergency surgery, and obviously I pulled through, but it made me really scared for my life and I thought… oh okay I must not be the only one, I want to reach out and see how other people are doing. From then, when I started sharing my story in an online Facebook community, I saw how it had a lot of power and impact."

With this sharing and openness, there is an associated letting go of embarrassment, shame and stigma that they realise have been heavy weights, carried through their years of adolescence:

"I think since that moment I've been so embarrassingly open in so many ways, through advocacy, I think that I will say anything to anyone, but it doesn't really matter to me anymore, because I think that I can't tell people to not carry shame and stigma about it if I still carry shame and stigma about it. So I've used that to help me let go of the crap I was carrying"

One participant spoke about how they have moved from being open in the cloaca community space to other areas of their life and how this has been positively received:

"I've never had someone turn around and go 'oh that's really weird and gross' which I think was also a huge lesson because people's understanding, and kindness can really surprise you"

It is important to highlight, however, that although there is a shared sense of participants becoming open in disability, advocacy and cloaca community spaces, not all participants necessarily share a keenness to open themselves up to the same extent in more public spaces, even as they have moved through adolescence into young adulthood.

3b. What's been lost, what's been gained: who am I now?

This sub-theme captures another common experience among participants, as they move through adolescence and grow up with cloaca. Participants reflect on the aspects of their lives that have been compromised by or lost to the condition,

Participants described how the experience of cloaca, whether through managing symptoms or spending time in hospitals for medical appointments, examinations, and procedures, leads to a sense of loss and separation from the normalcy of adolescence:

"It was things like coming home from school or missing out on school trips…I would never go to like friends, sleepovers, and things like that"

One participant highlights several contributing factors, including the disruptive unpredictability of symptom experiences, the time-consuming nature of required flushes that interfere with daily activities like homework, and the inability to participate in typical adolescent experiences such as sleepovers and school trips. They emphasise how these aspects deepened their sense of isolation from their peers.

"There was unpredictability of when it would happen and also that it would pull me away from my peers. I think the other thing that would frustrate me was that everyone else would have a very normal life, or at least from the outside, and if I wanted that I'd have to go home every single night and do a flush. It would take up my time and it would be so annoying, and it would stop me from doing my homework and then if I wanted to go on sleepovers, like it would stop me from doing things. I didn't want to go away on school camps because of it, all of that made me feel more separate from people"

This loss can, understandably, be felt intensely, with one participant describing a feeling that the demands of cloaca on their time and energy have unfairly deprived them of opportunities or experienced they witness their peers having:

"*You do feel that things have been taken away from you, it has been robbed"*

This experience of loss can also be felt in relation to aspects of life such as fertility and childbirth, which can be impacted by cloaca. This could be the difficulty or impossibility of conceiving or giving birth naturally:

"I would suppose the biggest thing on my mind is pregnancy erm I know that I can't have a natural birth, I will have to have a caesarean"

Or, for some people, this could include the loss of fertility:

"I realised that you can't do a lot when you're older because of our condition... so, you have to cancel plans, how do you explain that? So you're in a romantic relationship and then – uhoh, you don't have a vaginal canal, or you remember crap, I don't have half a uterus, I can't bear children"

This participant described how, due to the impact of cloaca, she is now facing the loss of part of her body and the reproductive system:

"Now, full disclosure, we are planning a hysterectomy because of everything that's happened these last few years"

Due to the significant loss experienced by participants, there is also a substantial process of negotiation as participants discover themselves alongside cloaca and develop an identity separate from it. All participants reflected on how this negotiation of life and identity unfolds within the context of cloaca.

One participant spoke about how their experience of cloaca interacted with their experience of religion. As they became increasingly aware of both religion and their condition, they reflected on an internal struggle with their place in the world and their belief system in the context of a Catholic school:

"So, science saved me, so if God is real, maybe I should be alive, as the reason I am alive is because of science and so I used to have this horrible mindset about God and religion through Catholic school because of that"

However, there was also reflection on how the experience of cloaca has positively contributed to the development of their identity as a young adult. For some participants, there is a perception of how the 'good' can exist alongside the 'bad':

"It is hard, but it can be made good in a lot of ways"

For example, one participant reflected on how she used to spend long periods of time on the toilet during flushes with a book, or spending time in hospital doing her schoolwork and how this has contributed to the development of her positive character traits, such as intelligence and resourcefulness:

"I remember when I used to have my enemas, that's when I would do my reading and I was reading by 3"

Another participant spoke about how difficult medical experiences due to cloaca have inadvertently shaped her belief system and future trajectory, after experiencing the warmth and kindness of a hospital chaplain whilst in hospital and the welcoming, non-judgemental nature of a church community:

"I'm studying theology and I want to become a hospital chaplain…I think I like church particularly because of my disability because a parish priest visited me in the hospital"; "they [church holiday camp] would ask me what I would need to be able to participate and that gave me a lot of self-confidence"

This is echoed by another participant who attributed her experience of cloaca as key in shaping her career aspirations as a nurse:

"I don't think I would have chosen to be a nurse. I think cloaca and my experiences with it have really influenced my desire to be a nurse and my passion for nursing, especially to specialise in paediatrics and colorectal"

Moreover, whilst the experience of cloaca can be challenging, all participants shared a perception of it shaping their identity in significantly positive ways.

Some participants attribute the connection with the wider cloaca community and the opportunity to advocate and speak up about their experiences as key in helping them open their perspective up to the strengths they have developed along the way:

"Going to the conference was really good and I feel proud to have done what I've done and went through what I've went through, and I think it has given me a lot. I

think it’s you know made me who I am today you know, I see myself as a strong person and I can cope with things”

In fact, one participant highlighted how the strength and resilience they have developed through managing the experiences associated with cloaca have enabled them to approach other, more universally human challenges, more successfully:

“It’s been a walk-through hell; I’m not going to lie. It’s been rough and really hard; I’m not going to lie but it’s given me a lot of strength and resilience. I don’t think I would be as outspoken, as confident, as I am now if I didn’t have cloaca. I mean, even if I didn’t have cloaca, I would still have the normal teenager, or things that girls or women go through, those normal stressors like body image and stuff like that. I think that if I didn’t have cloaca, I would still be dealing with things, but I wouldn’t, it would be harder for me to overcome those challenges”

When it comes to the future however, whilst there is strength and hope, there is also a sense of tentativeness. For example, one participant spoke about how their medical challenges meant that they were closed off to the possibility of a future for a significant amount of time and how, more recently, they have started to allow themselves to imagine what a future might look like:

“I think it was only last year that I opened up my mind up to the possibility that ‘you know you might actually live a long time”

There is also a sense of uncertainty, as participants continue to grapple with a complex condition with moving and shifting pieces:

“I’ve spoken about with a lot of cloaca girls is that, you know, there’s a few of us that have urinary continence, and the ones of us that do can wake up one day and not have it anymore, so that scares me a little bit”

Discussion

The findings in this study have captured the reflections of individuals with cloaca who have journeyed through adolescence, to young adulthood. They provide a unique perspective on how certain challenges have been navigated, what factors helped in overcoming them, and the ongoing nature of other difficulties. These challenges relate to a developing sense of self in relation to sexual and psychological wellbeing, independence and autonomy and how cloaca is integrated into identity. Moreover, the nature of cloaca itself, in terms of its medical complexity and rarity, appears to be an underlying feature underscoring the experience of the condition and its impact for both individuals with the condition and medical professionals treating it.

The next section situates the main themes from the study within the existing literature. I will then go on to reflect on the strengths and weaknesses of the study, and potential future research and clinical implications.

Sense of the bodily self

This theme from the analysis highlights how cloaca and experiences of treatment and symptom management can impact individuals' relationship with their physical body, including their sense of ownership and the development of a cohesive and confident sexual identity.

Experiences of examination, scrutiny, and fascination in both medical and social contexts can exacerbate these challenges, sometimes leading to a struggle with ownership over the body, as if it exits in the public as opposed to the personal domain. These factors can pose challenges to psychological well-being and overall identity development.

In the social context, some participants describe experiences of others physically interacting with their bodies or creating stories about them and influencing the narrative about their bodies. One aspect of this experience relates to the development of identity in the context of difference that is associated with illness or disability. The existing literature demonstrates how others’ reactions to illness or disability and the associated stigma can significantly affect how individuals incorporate their condition into their identity (O’Donnell & Habenicht, 2022). This is particularly relevant during adolescence and young adulthood, a time when identity development can be heavily influenced by how others perceive physical characteristics (Valeras, 2010).

However, the findings highlight another dimension of this experience: specifically, how others' interest in and command over the narrative about their body may lead to its objectification, rather than being seen as part of personal identity. This objectification can contribute to the porous boundaries that individuals with cloaca might experience regarding their own bodies. This is particularly significant in the medical context, where the complexity and rarity of the condition attract interest and curiosity. Not only can individuals be subject to numerous examinations and interventions, but individuals are also often labelled as ‘complex’ and are referred to as such; their bodies are framed by and interacted with in the context of this complexity. Subsequently, personal identity can become obscured by the complexity of the medical body, which can lead to feelings of disconnection from their own body and struggles with a sense of ownership and agency.

Whilst it is difficult to make any definitive claims considering the qualitative nature of the current study, the findings suggest a number of mitigating factors that are important in supporting the integration of the body with the self, helping individuals to reclaim agency and maintain personal boundaries. These primarily focus on the quality of relationships with medical professionals, and the nature of social contexts which are explored in more detail in the clinical implications section.

Furthermore, the findings demonstrate that individuals growing up with cloaca can face challenges in connecting to their sexual body, developing a sense of a sexual self and forming their sexual identity. This finding reflects a growing evidence-base which highlights how chronic physical health conditions and their management can impair the development of the sexual self (Engelen et al., 2020). For individuals with cloaca, several mechanisms appear to be at play.

Firstly, symptom management procedures and symptoms themselves, such as bloating and constipation, can be uncomfortable or painful which can make it difficult for individuals with cloaca to relate to a subjective sense of attractiveness, confidence and sexual appeal. This is reflected in existing literature that captures perceptions of ostomised women regarding sexual activity and emphasises how challenges related to personal care and physical discomfort can understandably hinder sexual activity (Kimura et al., 2013). Moreover, this can be compounded by factors which impact on body confidence or body image; individuals with cloaca can often have significant scar tissue on their body and intimate areas from surgeries or stomas and have described the impact of this on their confidence, their tendency to avoid sexual contact, or need to seek reassurance from sexual partners. Again, this links to research on other conditions where individuals might also be faced with these challenges to positive body image (Manworren et al., 1996; Suris et al., 2004).

It is also interesting to consider perceptions in society. Existing literature suggests that the difficulties with developing a sexual identity for those with a chronic condition can be influenced by the way in which society may view people with health conditions or disabilities as non-sexual beings with a lack of interest in sexual activities (Neufeld et al., 2003). In fact, it has been said that the social stigma can be more of a tangible hindrance than the condition itself (Esmail et al., 2010; Shah, 2017). Therefore, whilst only touched on in the findings by a few participants, it is interesting to consider how dominant narratives in society may unconsciously influence the way health professionals, teachers or family members perceive or interact with individuals with cloaca and how this might impact on an individual's sense of themselves as a sexual person.

Other mechanisms at play that impact the development of the sexual identity for individuals with cloaca are not necessarily captured in the existing literature. These relate to an inherent aspect of cloaca; the fact that sexual organs and intimate areas can be the direct focus of examination, intervention and symptom management. This study therefore builds on the existing literature by demonstrating how the development of a sexual identity and connection with the sexual body can be challenged by the medicalisation of the genitalia and intimate areas. Individuals with cloaca can first become familiar with their intimate areas within a medical context, where these body parts are discussed, examined, and interacted with. As sexual development progresses and individuals begin to form a new relationship with these areas it appears that it can be challenging to integrate or create space for sexual associations. Subsequently, genitalia and intimate areas can be perceived more as reminders of medical challenges rather than sources of pleasure, making it difficult to separate them from painful, intrusive or simply clinical medical contexts.

In general, positive aspects of sexuality remain under studied among young people globally (Kagesten & Reeuwijk, 2021), especially for those with enduring health conditions. However, existing literature on the components of positive sexual identity development generally agrees that it encompasses sexual self-esteem, sexual pleasure and sexual self-efficacy (Anderson, 2013). One especially powerful finding of the current study highlights how the two concepts within this theme of the sense of bodily self, body ownership and sexual identity, may interact together; how growing up with cloaca can pose a challenge to an individual's sense of personal agency within a sexual context, and the potential implications of this. For example, challenges in developing a sexual sense of self, combined with challenges to body ownership, can lead to difficulties in asserting personal boundaries in non-medical contexts. This was reflected in a participant's first sexual experience being without any sensation and ultimately non-consensual; she noted that her difficulty in knowing and exercising her own boundaries was influenced by the learned perspective that her body was not her own. This both aligns with and extends existing literature such as Arbeit et al. (2014) who describes a process whereby an individual's sexual selfhood may influence their personal agency, which in turn determines their ability to negotiate with others which, in a sexual context, can relate to obtaining or giving consent.

However, the findings highlight that despite the potential challenges, a positive connection with the sexual self was more achievable and perceived as safe within the context of long-term relationships as individuals transitioned from adolescence to young adulthood. This aligns with existing research that highlights the positive influence of supportive partners when it comes to sexual activity (Kimura et al., 2013). It appears that emotional safety and trust within a relationship are important, alongside a sense of acceptance, reassurance and encouragement that a partner may provide. A safe, intimate relationship provides an opportunity for individuals to experience and connect with their sexual self, facilitated through their partner's perspective.

Medical Complexity

The findings highlight that the complexity of cloaca is mirrored in its management, requiring involvement with various medical professionals, sometimes across different parts of the healthcare system. The intervention and symptom management experience appears to be unique to an individual, as the constellation of symptoms can vary across individuals. As the person ages and transitions from paediatric to adult services, the responsibility for the management of care often shifts from caregivers to the individuals who are then faced with the task of coordination of and advocation for their care and alongside management of symptoms.

This experience has been captured succinctly by a participant in the current study as "invisible burden work". This concept of the self-management of care being a burden is prevalent within the literature, particularly when it comes to chronic conditions such as diabetes (Gonzalez et al., 2016). If overwhelming, the experience of one's burden has been found to be linked to worse clinical outcomes including a poorer overall health-related quality of life (Ridgeway et al., 2014). The evidence base outlines how a range of emotional, social and health resources can be drawn on to help individuals manage complex care demands, which includes social support from immediate family members, or from a wider social network (Ridgeway et al., 2014). This links into the current findings which highlight the significance of support that can be accessed from the international community of individuals with cloaca. It appears that this offers an invaluable sense of normalisation and comfort, as individuals share an understanding of similar demands on their time or necessary adjustment to routines.

Wider research indicates that supporting individuals to take on more autonomy and become psychologically empowered in managing their condition leads to better physical health and quality of life (McDougall, Evans, & Baldwin, 2010). However, this must be balanced against the current findings which suggest that the transition to adult services and self-management can sometimes be overwhelming and discouraging in the context of cloaca. In fact, an overreliance on individual autonomy and responsibility in the healthcare context, without adequate support may lead to a sense of 'care left undone' as described by Greaney and Flaherty (2020). Due to its complexity and rarity, a significant part of self-management appears to involve care coordination, navigating services and systems, and self-advocacy; challenges that are less pronounced in other conditions within the existing evidence base and that can significantly add to an individual's perception of burden.

Therefore, it is useful to consider what the existing evidence suggests about supporting and intervening to lessen the experience of burden. A model proposed by Shippee et al. (2020) suggests that burden arises from an imbalance between an individual's daily demands and responsibilities (not solely health-related) and their capacity to address these demands. Capacity includes health status, financial status, social support, and attitudes and beliefs about themselves and their condition (Shippee et al., 2020). Alongside this, the literature also highlights how the relational quality between the individual and medical professional can also mitigate feelings of burden and support patients to develop confidence in self-management (Eton et al., 2017). This highlights the importance of considering individuals with cloaca within their social and psychological context and provides opportunities for further intervention and support that account for both the patient and their relationship with medical professionals.

Another important component of medical complexity highlighted in the findings is the experience of medical anxiety and distress resulting from surgical procedures, medical treatments, and time spent in hospitals or medical appointments, which can significantly impact psychological wellbeing. This aligns with an existing evidence base regarding the possibility of experiencing psychological distress and posttraumatic symptoms following paediatric surgery (Turgoose et al., 2021). Referred to as paediatric medical traumatic stress (PMTS), it can be defined as "a set of psychological and physiological responses of children and their families to pain, injury, serious illness, medical procedures, and invasive or frightening treatment experiences" (National Child Traumatic Stress Network, 2018) and can include reactions such as hyperarousal, avoidance of anxiety-inducing stimuli and negative impacts on cognition and mood (Stanzel & Sierau, 2021). It is important to highlight that medical events such as surgery or interventions are not considered to be inherently traumatic; rather, their impact largely depends on an individual's interpretation and sense-making. Moreover, in the immediate aftermath, acute stress symptoms and transient emotional distress are to be expected and, in fact, the research suggests can help an individual to adapt (Kazak et al., 2006).

The findings of the current study highlight that individuals with cloaca can be at risk of enduring psychological distress in relation to their experiences of medical intervention and treatment. It is apparent that memories of past medical procedures, along with symptoms like avoidance or increased anxiety, may persist into young adulthood. These symptoms could indicate ongoing experiences of medical post-traumatic stress.

According to the existing evidence base, there are a number of factors which have been found to increase the risk for lingering psychological distress following a medical event that can impact on recovery and functioning (Ben-Ari et al., 2018). For example, the length of hospital stays, the number of procedures, high levels of anxiety during hospitalization, elevated or unmanaged pain, the intrusiveness of interventions, the experience of a lack of control, and perceiving hospitalization as frightening and life-threatening are al considered to be significant. It is interesting to consider how the findings of the current study relate to these risk increasing factors and how they also might provide opportunities for intervention and mitigation.

The findings highlight that medical intervention, including initial reconstructive surgery and ongoing symptom management, can be complex, physically intrusive, and changeable. Interventions and examinations may be experienced as intrusive, particularly those involving dilation. Additionally, for those with associated anomalies or the experience of complications and changing symptoms, the number of procedures is likely to be higher, which increases their vulnerability to long-lasting psychological effects. The findings also underscore the high levels of anxiety experienced by individuals with cloaca, manifesting as generalized medical anxiety and in relation to specific medical procedures, such as anaesthesia, which has been defined in the literature as a medical phobia (Ben-Ari et al., 2023).

Furthermore, the study highlights the importance of understanding the coping strategies adopted by participants. Many describe adopting a detached state, trying hard to appear composed and compliant during medical procedures to manage unease and distress. While these coping mechanisms may provide short-term relief, they can also perpetuate feelings of helplessness and vulnerability in medical settings. This is compounded by difficulty in developing a sense of ownership over the body, as highlighted in earlier findings, contributing to an experience of a lack of control, which is another risk factor for lingering psychological distress. This complexity can hinder sensitive recognition and response by medical professionals, especially during the transition from paediatric to adult care, where awareness of an individual’s past medical experiences may not necessarily be known or held in mind.

In terms of risk factors, the literature also highlights the importance of family support and the impact of parental anxiety (Ben-Ari et al., 2018). However, this was not something specifically explored in the current study but could provide rich ground for further exploration in the context of cloaca.

The existing literature does indicate that there is a growing awareness of the possibility and prevalence of medical traumatic stress, alongside efforts to enhance preventive and treatment initiatives. However, systemic obstacles like resource limitations, inadequate training, and institutional support remain prevalent barriers to improving outcomes (Cuneo et al., 2023). Furthermore, it is important to note that this study did not include the perspectives and experiences of medical professionals in relation to medical anxiety and possible experiences of medical trauma. Capturing these perspectives in future research will be crucial for gaining a deeper understanding of this aspect of the cloaca experience.

The findings of this study also point to another interesting consideration: how the complexity of the condition may impact patient perception of medical professionals and how this in turn can impact the nature and quality of interactions between medical professionals and individuals. As an individual grows up with cloaca, their understanding of cloaca can be shaped by interactions with professionals who may simultaneously be learning about the condition and grappling with its complexity and rarity. This can especially be the case following transition from paediatric to adult services, where there might be even less expertise in the system. This aligns with existing literature that highlights the challenge of transition; it is an established finding that adult providers are generally viewed to be less knowledgeable and caring by patients and families than their paediatric counterparts (Hilliard et al., 2014).

For individuals with cloaca, it can feel as if the task of educating new health providers falls solely on their shoulders, contributing to the sense of self-management being daunting “invisible burden work” as opposed to a positive opportunity to develop control and autonomy, as discussed above. These types of experiences can, understandably, undermine perceptions of doctors as infallible authorities which, aligning with the existing evidence base, can lead to stress, uncertainty and medical anxiety in the place of feelings of trust and being cared for (Forsberg et al., 2024).

As an attempt to understand experiences of being dismissed, misunderstood or treated negatively by health professionals, some participants in the current study contextualised their experiences with the idea that medical professionals may too be reacting to the complexity and rarity of cloaca and any negative reaction may possibly stem from discomfort with uncertainty or their own lack of knowledge. This is interesting to consider in the context of existing literature which highlights the challenges for medical professionals in a system where, due to the rise of evidence-based medicine and advances in medical care, there is paradoxically more awareness of the unknown than ever before, and they are therefore tasked with the challenge of managing and adapting to uncertainty, an unextractable component of complexity. This can be compounded by a medical culture which typically values the medical expert role and promotes a binary distinction between knowing and not knowing (Chaukos et al., 2023; Lanham et al., 2020). Considering this, existing literature has started to highlight how relationships between providers and patients, in the context of uncertainty and complexity, could be improved through attention to time and space for conversation and tools to promote communication in this context (Oerlemans et al., 2023).

Moreover, it is interesting to note that the current findings also illuminate how a levelling of the traditional patient-doctor power dynamic is positively received by participants. These instances of collaborative partnerships with medical professionals in the findings reflect the societal shift from paternalistic communication and practice, towards the more idealised patient-centred care. Tiffermans (2020) introduces the term 'engaged patient' to reflect current practice, highlighting that patients are taking a more active role in advocating for their health, and there is growing acknowledgment of this dynamic within patient-doctor interactions. For example, this is evidenced by patients increasingly expressing their preferences and asking more probing or in-depth questions of clinicians (Tiffermans, 2020). However, this is interesting to consider in relation to cloaca, where it becomes crucial to strike a delicate balance: supporting the development of autonomy while preventing individuals from feeling overwhelmed and isolated by the responsibilities and complexities of their own care and unsupported by the medical system, as underscored by the findings. Drawing on the expertise of participants within the study, positive interactions that appeared to foster collaboration without leaving the patient unsupported seemed to take the form of “I’m not sure about this… let’s find out together”. Within these types of communication efforts, the individual with cloaca is invited to be an active participant in the discovery process, which is likely to positively impact a sense of agency. Additionally, by framing the management of uncertainty and complexity as a joint effort appears to help alleviate anxiety and foster trust and support.

Growing up with Cloaca

The findings highlight how, as individuals navigate adolescence, they engage in a process of making sense of their experience with cloaca, in relation to their identity and sense of self. It is interesting to note how the experience of cloaca interacts with the development of identity, one of the key developmental tasks in adolescence (Branje, 2022). The wider literature suggests that having a strong sense of personal identity, which includes addressing questions such as “who am I; what defines me; where am I heading with my life?” can be protective against negative outcomes and a key resource for transitioning to adulthood (Luyckx et al., 2011).

Firstly, the findings demonstrate how cloaca is integrated into social identity, involving the processes of withholding and disclosing information about their condition. This includes understanding the functions these processes serve, the factors involved, and the journey from withholding to disclosure. Initially, it appears that withholding information from others can be a way of protecting oneself from anticipated rejection, or perceptions of being seen as vulnerable or different. This aligns with the existing evidence base for other congenital and chronic conditions such as cystic fibrosis and spina bifida, which highlights that withholding disclosure can be driven by a need to manage image and reputation as individuals are finding their place among peers and seeking for belonging and acceptance (Kaushanksy et al., 2016).

Moreover, the findings highlight that for a condition like cloaca, where interventions and symptom management can be experienced as intrusive, the tendency to withhold information extends to caregivers during adolescence. Individuals attempted to assert their independence and privacy by concealing more information from their parents. This process aligns with expectations in both normative adolescent development and healthy family relationships (Caughlin, 2003; Ebersole and Hernandez, 2016). The broader evidence base from other chronic illnesses, including congenital conditions, illustrates a contradiction that exist between the normative strive for independence and autonomy and the health-driven dependence that can be reminiscent of childhood (Wicks et al., 2019). It is interesting to consider this in the context of cloaca, where caregivers can have significant responsibilities and physical involvement in personal care and symptom management, in relation to areas of the body associated with privacy. The findings suggest that, for some individuals, the drive to conceal information and establish a separation may be more pronounced. This can lead to a desire to manage their condition and associated worries independently, which could increase resistance to accepting support or being recognised by others as benefitting from it.

A significant mitigating factor in the findings that facilitated a move from withholding information to disclosure, and from resistance to acceptance and integration, was connection with the wider cloaca community. Discovering others with the same condition and similar experiences provided individuals with comfort and normalisation, as well as the language to understand and discuss their condition and experiences with others. This phenomenon is widely documented in the literature on chronic conditions and their impact on self-concepts; through shared experiences and understanding, openness and honesty tend to increase (Hajdaveric et al., 2024).

In fact, connecting with others and having a sense of community is linked to increased strength and hope, helping people integrate their identity with their condition and feel more whole (Hajdaveric et al., 2024). This was evident in the current study, where participants reported that connecting with others in the cloaca community helped them to liberate themselves from shame and develop a clearer, more confident identity. Moreover, the findings revealed narratives about identity development that align with the broader literature, acknowledging the nuanced impact of chronic conditions that go beyond simplistic, binary positive or negative categorisations. For example, whilst the findings depicted narratives that contained expressions of loss and missing out, these were complemented by narratives of strength and resilience, as individuals acknowledged their skilful management of their condition. This aligns closely with research that emphasises the positive aspects, or illness gains, in the context of chronic conditions (Wicks et al., 2019).

Strengths and Limitations

Given the international spread of participants, experiences of growing up with cloaca are likely to have been influenced by a number of contextual differences. For example, it is possible that participants could have different experiences due to different healthcare provision. Although, considering the unique and rare nature of cloaca, the medical community does have an international reach and presence, and there are concerted attempts within the medical community to work towards largely consistent provisions of care. Therefore, whilst the international spread of the data has inherent limitations, the qualitative and exploratory nature of the study means that the findings are not intended to be generalisable. In fact, it is also a strength of the study that a wide range of experience has been captured, rather than focusing on experience within the context of one country. Despite geographical differences, commonalities in experiences suggest central aspects inherent to living with cloaca. The study has highlighted the international connectivity, both among the medical and patient community, as a significant feature of this condition.

Moreover, while the study aimed to recruit both young people and caregivers, only one caregiver participant was recruited. This limited the richness that multiple caregiver perspectives could have added to the findings. Nevertheless, the insights from the single caregiver were valuable, providing a helpful point of reference between the young people's reflections on growing up with the condition and the experiences of the caregiver’s daughter, who was experiencing adolescence at the time of the study. Again, the commonalities of themes that emerged between the young people and the caregiver’s perspective pointed to inherent aspects of growing up and living with cloaca and helped add a level of validation to the data set.

Similarly, whilst the initial aims of the study aimed to focus on the experiences of young people, the mean age of participants was 25 years old, with one participant being 38 years old. Due to the rarity of this condition and to time constraints in data collection, a decision was made to widen the age range to ensure adequate recruitment success. The wider spread of ages could be seen as a limitation of the study. However, all participants were born after the introduction of surgical techniques such as the PSARP/PSARVUP and the TUM which would have significantly improved outcomes, in comparison to people born before certain surgical and medical advances in treatment. Moreover, the framing of the interview focused on people’s experiences of growing up with cloaca, encouraging reflections on the experience as a child, teenager and young person.

An important strength of the study to highlight is the support the researcher had to discuss and reflect on the themes that were arising in the research within their supervisory team. However, it is also important to highlight that the researcher and supervisory team are not medical professionals and had no prior personal or professional experience with cloaca or other types of ARM before this study. Understanding cloaca and the necessary interventions requires comprehension of complex medical information. Therefore, it was seen to be important for the researcher and supervisory team to have had an adequate medical understanding to limit the possibility of misinterpretation and misrepresentation. To mitigate this, they were supported throughout the research process by a consultant paediatric surgeon with expertise in this area. The researcher also engaged in independent study and in conversations with a wide range of medical professionals to support an adequate level of medical comprehension. However, the non-medical background of the researcher is equally a strength of the study, as it reduced the likelihood of bias that might arise from preconceived notions about cloaca. This can be advantageous in ensuring the research findings are not implicated by pressure or influence, however implicit, that might stem from a medical perspective or agenda. However, the position and expertise of the researcher is a multifaceted area of consideration and is discussed in further detail below in the methodological reflections.

Methodological reflections

This study employed constructivist grounded theory as its underpinning methodology. It is important to reflect on how this approach has influenced the research process and overall findings, considering its contributions, suitability and potential limitations.

Central to CGT is the position of the researcher. As opposed to attempted objectivity, it is seen to be a strength of CGT that the researchers are located as being within the research process, as opposed to hovering above or outside of it. According to Charmaz, this positioning allows for the emergent process of meaning-making, where interpretations of data are shaped by the researcher's worldview, experiences, and personal characteristics (Charmaz, 2006).

As a trainee clinical psychologist without prior personal or professional experience with cloaca or ARM, I have wondered how my positionality might have influenced the interview experience. For example, it is possible that my identity as a psychologist within the interview, alongside supportive supervision by clinical psychologists outside of the interview, facilitated an open and empathetic environment. I also wonder whether not having prior professional experience with cloaca enabled participants to discuss healthcare experiences more openly, especially those experiences that might have been less positive. However, on the other hand, I wondered if this could have also felt daunting, or could have replicated the dynamic of having to continuously educate the ‘professional’.

Throughout memo writing, I reflected on the warmth and relational quality of the interviews. Being a female researcher of similar age to many of the participants likely contributed to a sense of rapport and might have influenced my inclination towards certain themes in the data, particularly those related to sex and the development of the sexual self. In my reflective memos, I attempted to acknowledge and explore how my identity and own experiences as a woman might influence data interpretation. Rather than aiming to disregard these influences, I worked on embracing them as part of the meaning-making process in CGT. This reflexivity allowed me to approach the data with curiosity and openness, recognizing the interplay between the participants' narratives and my reactions and interpretations.

Another key component of CGT is the construction of a theoretical model that is grounded in the data. Whilst the findings of this study work towards a model for understanding the psychological experience of growing up with cloaca, it remains open and loosely defined, rather than fitting into a coherent diagram that could, arguably, be more easily applied to achieve one of the study’s aims: to elucidate the patient and care experience and inform and support care for individuals with cloaca. Therefore, it could be argued that a different method could have been more effective or appropriate to meet the aims of the study. However, it is important to consider the definition of theory used here. CGT, from an interpretivist perspective, emphasizes how theory illuminates understanding rather than providing a strict explanation (Charmaz, 2006). Following this approach, through constant comparison of and iterative engagement with the data set, the focus has been on exploring patterns and connections, allowing for multiple interpretations and maintaining indeterminacy. This aligns with the epistemological stance of both the researcher and the methodology, rather than proposing a more defined and potentially restrictive model of experience which would not fit with the complexity of the condition and the unique way it can manifest or be experienced by each individual. While alternative methods could have been just as valid and might offer different insights, that does not diminish the value and applicability of the CGT approach used in this study. By embracing CGT, this research provides a nuanced understanding of growing up with cloaca that honours the inherent complexity associated with the condition.

Future research

This study presents a first exploration into the experiences and psychosocial impacts of cloaca, offering a starting point for further research. Reflecting on both the findings and limitations, there are several opportunities for future research that emerge.

Whilst the study focused on the experiences of individuals with cloaca, the need for the complementary medical professional's perspective was apparent throughout. Therefore, future qualitative research could aim to capture the experiences and perspectives of surgical professionals in paediatric services who are directly involved in treating cloaca, as well as practitioners in adult services whose encountering of cloaca patients may not be part of their expected or regular practice. It is essential to investigate how medical professionals experience and manage uncertainty and complexity associated with conditions like cloaca, considering systemic factors that influence patient care. Additionally, exploring the dynamics of interactions between patients with cloaca and medical professionals within the healthcare system by focusing on relational exchanges between patient and medical professional could provide deeper insights into the challenges and nuances of support and care for cloaca.

Future research could also focus on exploring caregiver experience in more detail, through the focused recruitment of caregivers. Similarly, there would be value in focusing on participant experience within a younger age bracket. Moreover, future research could also delve deeper into specific experiences to enhance the richness of initial findings in this study, which appear particularly significant. Examples include exploring sexual experiences or the transition from paediatric to adult services. By making concerted efforts in recruitment, such as accessing and engaging with existing online cloaca communities, researchers could capture experiences from a larger number of participants. Employing a quantitative methodology could then test the identified themes or an exploration of relationships on a larger scale, aiming for generalisability of findings.

Clinical Implications

Undoubtedly, significant advances in medical and surgical treatment and expertise have enabled lifesaving and life-enhancing care. This is particularly true for cloaca, a condition that often requires some of the most complex medical and surgical interventions, where advances in medical and surgical techniques have markedly improved the physical functionality of individuals. Historically, scientific medicine has often been viewed through the lens of a metaphor: the body as a machine and the physician as a mechanic who repairs malfunctioning or broken components (Greenhalgh, 2001). This perspective, however, does not fully account for the psychological experience of the individual, encompassing the condition, symptoms, and necessary interventions. It is crucial to consider how these factors are experienced and the potential psychological impacts they may have in both the short and long term, particularly as individuals transition from paediatric to adult services. Therefore, this study highlights the importance of understanding these psychological implications.

This section outlines key considerations for clinical practice and the organisation and delivery of services to support psychological well-being and improve psychological outcomes in the long term. It is important to highlight the limitations of these recommendations, given that the researcher's professional experience sits outside of the medical field and clinical services for individuals with cloaca and their families. As a result, the recommendation may include potential inaccuracies, points of repetition or overlap in terms of what is already being provided. Nonetheless, and most importantly, these considerations are grounded in the findings.

1. Given the vulnerability of individuals to experiencing medical anxiety or trauma reactions following experiences of surgical procedures, medical treatments and time spent in hospitals or medical appointments, several approaches could be considered. For example, training for healthcare

professionals to increase awareness of medical anxiety and trauma responses is important, particularly in recognising risk factors such as previous trauma or parental distress. Screening tools could be embedded during assessments and follow-ups, including check-ins after an appropriate amount of time following surgeries. Additionally, normalising feelings of distress or fear in individuals and their families can support the understanding of experiences and development of beliefs about the experiences.

2. Further to Svetanoff et al (2023), the current study adds support for the approaches suggested by them to address an individual's relationship with their body, their intimate areas and the development of their sexual self. For example, opening space and ensuring protected time for individuals to explore their sexual goals and concerns and their relationship with their sexual body. Permission to discuss these topics could be given by asking questions about how they feel about their genitals, how they feel about touching their genitals or engaging in forms of sexual activity with a partner. It may be that psychological colleagues could provide support to think about these conversations, whether that be through having conversations together with the medical professional or providing consultation-type support to medical professionals.

3. The concept of "invisible burden work," as highlighted in the study, refers to the unseen complexities involved in managing cloaca, especially following the transition from paediatric to adult services. It is important to consider strategies that support individuals and caregivers to mitigate potentially feeling overwhelmed and lessen a sense of burden.

   As suggested in the study, looking to existing research can develop our understanding of the potential for burden experienced in self-management of enduring health conditions such as cloaca and help address broader influencing factors. This understanding can guide psychological and social interventions. For example, understanding the interaction between existing demands and responsibilities and capacity can focus attention on the consideration of factors such as the individual's relational and social environment, psychological capacity, attitudes, beliefs, social demands, financial status, and social support (as outlined in Shippee et al., 2020). By integrating a consideration of these factors into consultations and assessments, particularly when considering supporting the transition to adult settings, healthcare providers can better highlight the need for potential psychological and social interventions.

Moreover, another approach could be to align medical care with an individual's priorities and goals. Participant experience suggests that using models like the "six F-words," originally developed for child disability but now applied more broadly to child health can be helpful. These six F-words—Functioning, Family, Fitness, Fun, Friends, and Future (Rosenbaum & Gorter, 2011)—can provide a framework for prioritising care and focusing on holistic well-being, which may help reduce feelings of overwhelm. It could be important to explore how this model can be adapted for use in adult care settings to ensure continued support with a holistic perspective.

4. It is notable that, within the findings of the current study, the interactions between the medical professional and the individual with cloaca and the context of the interactions is a recurring aspect of experience that appears to be important. For example, the interactions are taking place often within a context of complexity and uncertainty; the combination of complexity and the rarity of cloaca can often mean that even for those medical professionals with skill and expertise are not necessarily well-practiced or confident, as they might not come across the condition as regularly as other conditions.

   The current study highlights the experience of interactions with medical professionals, from the perspectives of individuals with cloaca. Therefore it is important to highlight that this is one-sided and further research is needed, particularly in relation to the experiences of medical professionals, before more meaningful clinical implications can be suggested. However, considering how to improve communication and support in these contexts is a valuable starting point. The study suggests that positive interactions often involve collaboration and openness. Framing the management of uncertainty and complexity more as a shared effort can help build trust and enhance the individual's sense of agency. Additionally, supporting medical professionals in reflecting on their own experiences and feelings related to complexity and uncertainty could also be beneficial and another starting point to consider.

5. The importance of connection with the wider cloaca community was consistently demonstrated to be a transformative aspect of how the individual related to themselves and to their condition in the study, with participants crediting the community as a rich source of support, normalisation and empowerment that had a positive and protective impact on their psychological wellbeing. Therefore, this could be held in mind by both medical and psychological professionals that provide care to individuals with cloaca; introducing the idea and supporting efforts to support connections wherever possible, among both individuals with the condition and their caregivers.

6. The study also highlights that identity formation is an ongoing process and can be acutely and intimately impacted by enduring health conditions such as cloaca. Therefore, ensuring there are opportunities for young people to reflect on their experiences and explore their identities is crucial to support the identity formation process and can provide invaluable opportunity for stories of resilience, strength and hope to emerge. It is important to consider how this may be achieved within services; whether this may provide guidance for psychological interventions and support that is offered, or whether there are opportunities to integrate this within routine medical care.

7. The clinical implications discussed above have been drawn from the expertise of experiences of individuals and caregivers living with and navigating life with cloaca. Therefore, it is worth considering how their expertise could be more routinely incorporated into care planning and provision, similar to the role of "experts by experience" in mental health

services. However, challenges such as the rarity of the condition and the geographical spread of individuals may pose barriers to this involvement. It is also possible that some form of this engagement already exists within the cloaca community. Therefore, it is important to explore how existing networks and resources within the community can be utilised and expanded upon to enhance care planning and support.

Delivery and organisation of services

Alongside the specific clinical improvements as highlighted above, it is important to consider how care for individuals with cloaca can effectively integrate psychological support. Firstly, while not all patients will need one-on-one psychological therapy at every stage, there should be ongoing opportunities to access psychological support throughout their entire journey. This would involve an established referral route that individuals and their families can access at various points throughout their life and care experience. Moreover, increasing accessibility also requires the normalisation of psychological support as a routine part of care, rather than a distant or secondary option. This could be achieved by integrating psychological professionals into the care team, ensuring their presence and visibility, even if they are not directly involved in the treatment at that time. For example, this could involve the physical presence of psychological colleagues in care planning meetings, being referred to in discussions, or simply having their contact details and photos displayed in the hospital to make them more visible and familiar to patients and families.

Moreover, psychological support can take many forms beyond individual therapy. It might include systemic or indirect support, such as providing training on psychosocial challenges or offering consultation to support a medical professional open a conversation about a young person’s sexual wellbeing in the context of a medical appointment. Psychologists could also work with medical teams to create and implement specific assessment tools and communication strategies. Additionally, given the complexity of working with cloaca, it is equally important to support the well-being of medical professionals. Therefore, psychology colleagues could offer psychological support for healthcare teams, such as opportunities for reflection on the emotional impact of the work.

The findings also provide insights into how the organisation and delivery of clinical services could be enhanced to improve key outcomes, such as patient experience. Given the complexity of cloaca, establishing a centralised, specialised treatment centre dedicated to rare conditions like cloaca could offer substantial benefits. This approach mirrors the care provided for other congenital conditions, such as cystic fibrosis, where specialised care is paramount. A centralised model would allow professionals to pool their knowledge and offer mutual support, reducing the pressure on any one individual to hold all of the necessary expertise. Additionally, it could extend valuable support to general adult healthcare providers through consultation with the centralised system.

Although, centralisation raises concerns about the impact of travel, which could be especially challenging for individuals and families with limited financial resources or social support. While this may be less problematic in smaller countries like the UK, the impact could be more significant in larger countries with vast geographical areas, necessitating further consideration. Though this research does not provide definitive solutions, it highlights the importance of considering how centralisation might affect patient experience, particularly in terms of perceived treatment burden, and whether it would help both patients and healthcare professionals navigate the complexities of cloaca. Understanding these factors, along with strategies to address potential drawbacks, is essential for shaping the future of cloaca care. Ultimately, the findings underscore the need for continued exploration to enhance patient experience and improve care provision.

Conclusion

The complexity of cloaca, compounded by its rarity, creates a unique combination that inevitably poses significant challenges. There is no doubt that the significant advances in medical and surgical treatment and expertise have enabled lifesaving and life-enhancing care, without which individuals with cloaca would likely die or face debilitating complications. However, psychological understanding can often lag behind advances in medical care, perhaps taking longer to develop and to be integrated into medical care. Within this gap, unintended side-effects of complex medical treatments fall that, if not understood, can complicate the idea of a successful outcome (Van den Eynde et al., 2021). An example of this is highlighted in regard to hand replacement surgery, an undeniable feat of medical innovation. While the first ever hand transplant was surgically successful, the recipient struggled to accept and integrate it psychologically and subsequently requested the removal of the hand (Kanitakis et al., 2003). An understanding of the psychological implications has led to the establishment of psychological support throughout the pre- and post-surgical processes (Kumnig et al., 2014). This also speaks to the importance of ensuring a holistic consideration of outcomes when designing and providing services in care. A prevailing focus on the biomedical can often mean that the impact of conditions and subsequent treatment on a patient’s experience and self-perception are overlooked (Hajdarevic et al., 2024). In fact, patient experience at every stage is important to contextualise clinical evidence and outcome, providing key information about the needs of a patient which takes into account the impact of the condition itself, as well as the unintended effects of treatment (Rand et al., 2019).

In conclusion therefore, this study has focused on the experience of younger adults with cloaca and has provided context to an important outcome: the patient and care experience. It is clear that individuals growing up with cloaca can face significant challenges as they move through adolescence to young adulthood, with various threats to their psychological wellbeing and the development of a cohesive sense of self. Subsequently, this study has worked towards a model for understanding of how the experience of the condition itself, including the experience of treatment, might influence psychological wellbeing and self-discovery and definition, alongside providing an understanding of factors that are important in mitigating these challenges. It is hoped that by increasing understanding of this outcome, the patient and care experience can sit equally alongside and positively influence other routine outcomes such as process, resource and clinical results.

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List of Abbreviations

ARM – Anorectal malformation

CGT – Constructivist Grounded Theory

GOSH – Great Ormond Street Hospital

PMTS - Paediatric medical traumatic stress

SF-36 - The Short Form 36 Health Survey Questionnaire

TUM – Total urogenital mobilization (surgical technique)

VACTERL – Vertebral defects, anal atresia, cardiac defects, tracheo-esophageal fistula, renal anomalies, and limb abnormalities.

PSARVUP - Posterior sagittal anorectal vaginal urethral plasty (a type of reconstructive surgery for cloaca)

QoL – Quality of life

RP – Rectoperineal fistula (type of ARM)

RV - Rectovestibular fistula (type of ARM)

Appendix A

Participant Information Sheet

Clinical Psychology, Leeds Institute of Health Sciences, School of Medicine

UNIVERSITY OF LEEDS

Participant Information Sheet
The experience of young people with cloaca.

You are invited to take part in a research project. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Please ask if anything is unclear, or if you would like more information.

What is the purpose of this project?

This project aims to explore the experience of young people 16-40 with cloaca. We would like to better understand the experiences of young people with cloaca and how it might impact on the lives of young people and their families. We hope to do this by hearing directly from young people and their parent or caregiver. It is hoped that by doing this, we can help service providers improve the support they provide.

Why have I been asked?

You have been invited to take part in the study as you are either a young person 16-40 or a parent/caregiver of a young person with cloaca.

Do I have to take part?

Participation is entirely voluntary. It is up to you to decide whether or not to take part. If you do decide to take part, you will be asked to sign a consent form. You can still decide to withdraw at any point without it having any negative consequences. You do not have to give a reason.

What will happen if I decide to take part?

You will take part in a one-to-one interview with the researcher, Emily Samuels. The interview is likely to ask you questions about your experience of cloaca, whether that be your own experience or as a parent or caregiver of a young person with cloaca. There will be a mix of some closed questions that will ask about specific details of your experience and some open questions which will invite you to share any feelings or experiences in relation to the condition and different areas of your life. You do not have to answer anything that makes you feel uncomfortable. The interview will last no more than 60 minutes, and with your permission, it will be audio recorded.

What happens to the information I share?

All the information that you share will be kept confidential and all possible steps will be taken to anonymise the research data to limit the chance that you can be identified in the results. However, whilst no names or identifying information will be used, some of the words you use in the interview may be used in the final research report. You have the opportunity to withdraw information and identify any information you do not want to be used as a quote at any point during the interview and up to two working days after the end of the interview. If you decide to withdraw within this time period, responses already provided will be deleted.

Appendix B

Participant Consent Form

Clinical Psychology, Leeds Institute of Health Sciences, School of Medicine

UNIVERSITY OF LEEDS

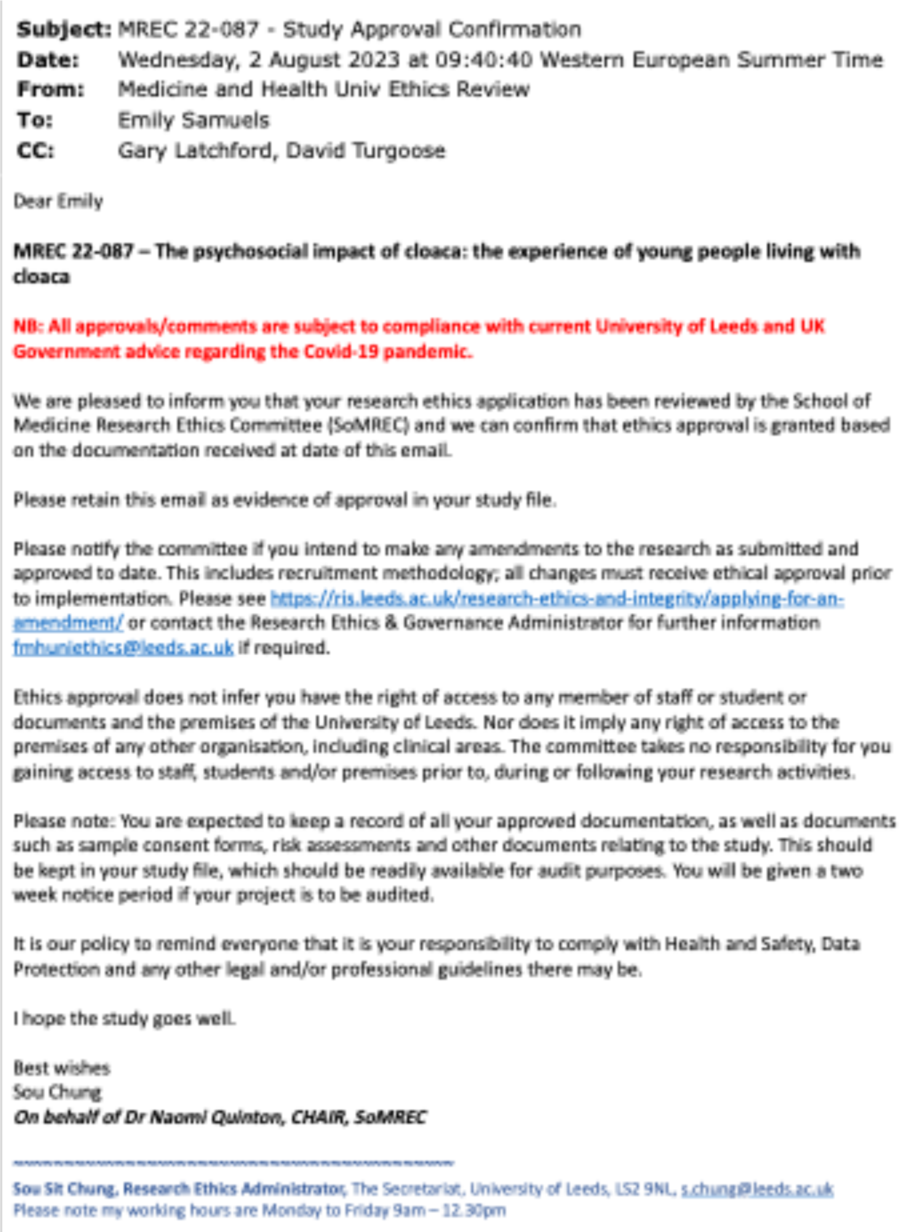
Consent to take part in **'The experience of young people with cloaca'.**	Add your initials next to the statement if you agree
I confirm that I have read and understand the information sheet dated *[insert date]* explaining the above research project and I have had the opportunity to ask questions about the project.	
I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without there being any negative consequences. I can withdraw up to two working days after the interview has taken place. If I decide to withdraw within this time period, any responses I provide will be deleted. In addition, should I not wish to answer any particular question or questions, I am free to decline. I can contact the researcher, Emily Samuels at umes@leeds.ac.uk	
I understand that members of the research team have access to my anonymised responses. I understand that my name will not be linked with the research materials, and that my responses will be kept strictly confidential. Anonymised extracts from my interview may be quoted in the final research report.	
Despite all possible steps to anonymise my responses, I understand that there is a possibility I may be identifiable by others that know me. I understand that I can identify any specific pieces of information that I am not happy to be used in the final research report up to two working days after the interview has taken place.	
I understand that my interview will be recorded, and the recording will be stored securely.	
I understand that the data collected from me will be anonymised and then stored securely for 3 years after the end of the study.	
I understand that if I inform the researcher that myself or someone else is at risk of harm, they may have to report this to the relevant support agencies - they will discuss this with me first but may be required to report with or without my permission	
I agree to take part in the above research project and will inform the researcher should my contact details change.	

Name of participant	
Participant's signature	
Date	
Name of lead researcher	Emily Samuels
Signature	
Date*	

MREC 22-087 – Version 2.0 – 19/07/2023

Appendix C

Confirmation of Ethical Approval



Subject: MREC 22-087 - Study Approval Confirmation
Date: Wednesday, 2 August 2023 at 09:40:40 Western European Summer Time
From: Medicine and Health Univ Ethics Review
To: Emily Samuels
CC: Gary Latchford, David Turgoose

Dear Emily

MREC 22-087 – The psychosocial impact of cloaca: the experience of young people living with cloaca

NB: All approvals/comments are subject to compliance with current University of Leeds and UK Government advice regarding the Covid-19 pandemic.

We are pleased to inform you that your research ethics application has been reviewed by the School of Medicine Research Ethics Committee (SoMREC) and we can confirm that ethics approval is granted based on the documentation received at date of this email.

Please retain this email as evidence of approval in your study file.

Please notify the committee if you intend to make any amendments to the research as submitted and approved to date. This includes recruitment methodology; all changes must receive ethical approval prior to implementation. Please see https://ris.leeds.ac.uk/research-ethics-and-integrity/applying-for-an-amendment/ or contact the Research Ethics & Governance Administrator for further information fmhuniethics@leeds.ac.uk if required.

Ethics approval does not infer you have the right of access to any member of staff or student or documents and the premises of the University of Leeds. Nor does it imply any right of access to the premises of any other organisation, including clinical areas. The committee takes no responsibility for you gaining access to staff, students and/or premises prior to, during or following your research activities.

Please note: You are expected to keep a record of all your approved documentation, as well as documents such as sample consent forms, risk assessments and other documents relating to the study. This should be kept in your study file, which should be readily available for audit purposes. You will be given a two week notice period if your project is to be audited.

It is our policy to remind everyone that it is your responsibility to comply with Health and Safety, Data Protection and any other legal and/or professional guidelines there may be.

I hope the study goes well.

Best wishes
Sou Chung
On behalf of Dr Naomi Quinton, CHAIR, SoMREC

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Sou Sit Chung, Research Ethics Administrator, The Secretariat, University of Leeds, LS2 9NL, s.chung@leeds.ac.uk
Please note my working hours are Monday to Friday 9am – 12.30pm
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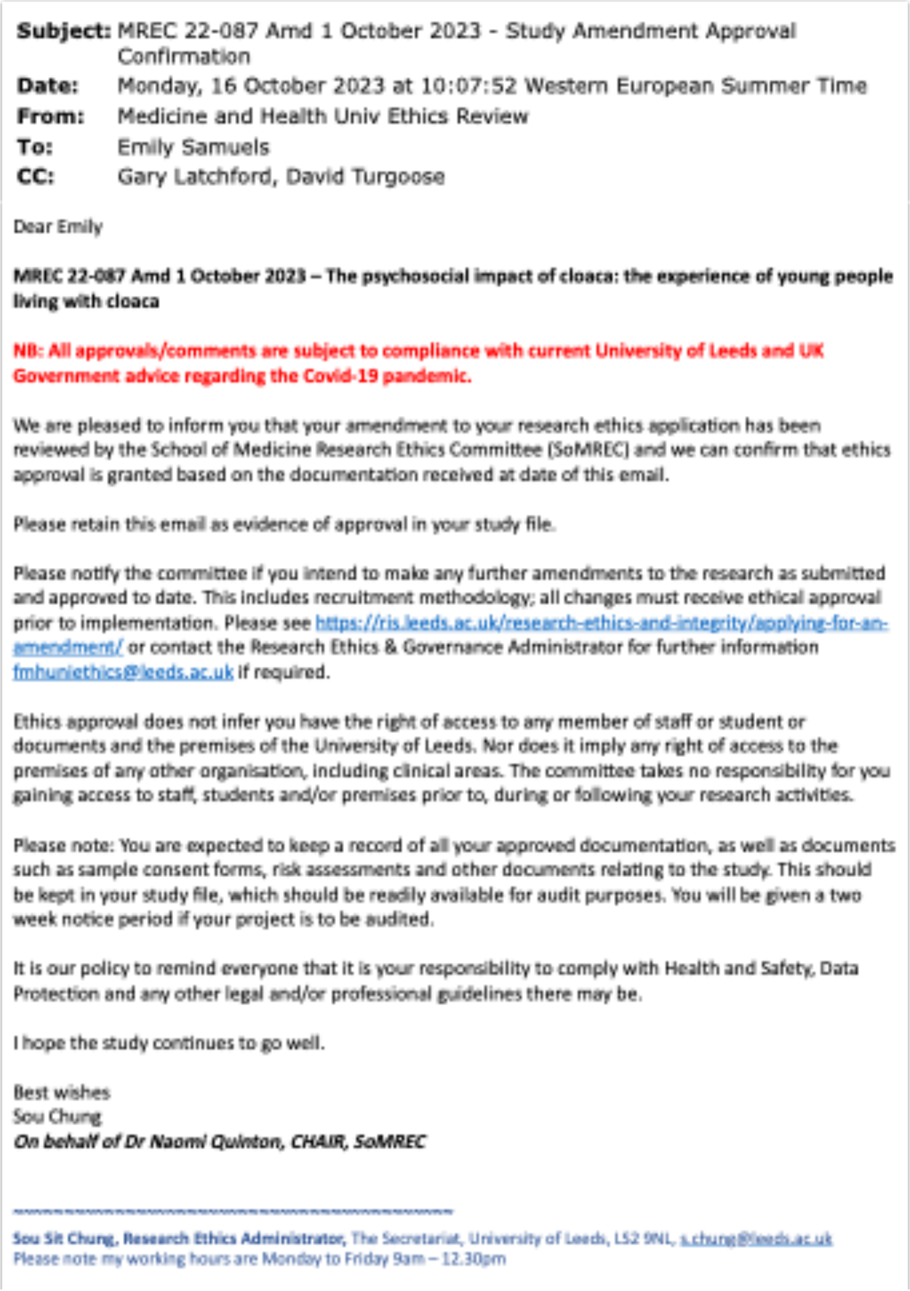
Appendix D

Confirmation of Ethical Approval - Amendment



Subject: MREC 22-087 Amd 1 October 2023 - Study Amendment Approval Confirmation
Date: Monday, 16 October 2023 at 10:07:52 Western European Summer Time
From: Medicine and Health Univ Ethics Review
To: Emily Samuels
CC: Gary Latchford, David Turgoose

Dear Emily

MREC 22-087 Amd 1 October 2023 – The psychosocial impact of cloaca: the experience of young people living with cloaca

NB: All approvals/comments are subject to compliance with current University of Leeds and UK Government advice regarding the Covid-19 pandemic.

We are pleased to inform you that your amendment to your research ethics application has been reviewed by the School of Medicine Research Ethics Committee (SoMREC) and we can confirm that ethics approval is granted based on the documentation received at date of this email.

Please retain this email as evidence of approval in your study file.

Please notify the committee if you intend to make any further amendments to the research as submitted and approved to date. This includes recruitment methodology; all changes must receive ethical approval prior to implementation. Please see https://ris.leeds.ac.uk/research-ethics-and-integrity/applying-for-an-amendment/ or contact the Research Ethics & Governance Administrator for further information fmhuniethics@leeds.ac.uk if required.

Ethics approval does not infer you have the right of access to any member of staff or student or documents and the premises of the University of Leeds. Nor does it imply any right of access to the premises of any other organisation, including clinical areas. The committee takes no responsibility for you gaining access to staff, students and/or premises prior to, during or following your research activities.

Please note: You are expected to keep a record of all your approved documentation, as well as documents such as sample consent forms, risk assessments and other documents relating to the study. This should be kept in your study file, which should be readily available for audit purposes. You will be given a two week notice period if your project is to be audited.

It is our policy to remind everyone that it is your responsibility to comply with Health and Safety, Data Protection and any other legal and/or professional guidelines there may be.

I hope the study continues to go well.

Best wishes
Sou Chung
On behalf of Dr Naomi Quinton, CHAIR, SoMREC

Sou Sit Chung, Research Ethics Administrator, The Secretariat, University of Leeds, LS2 9NL, s.chung@leeds.ac.uk
Please note my working hours are Monday to Friday 9am – 12.30pm

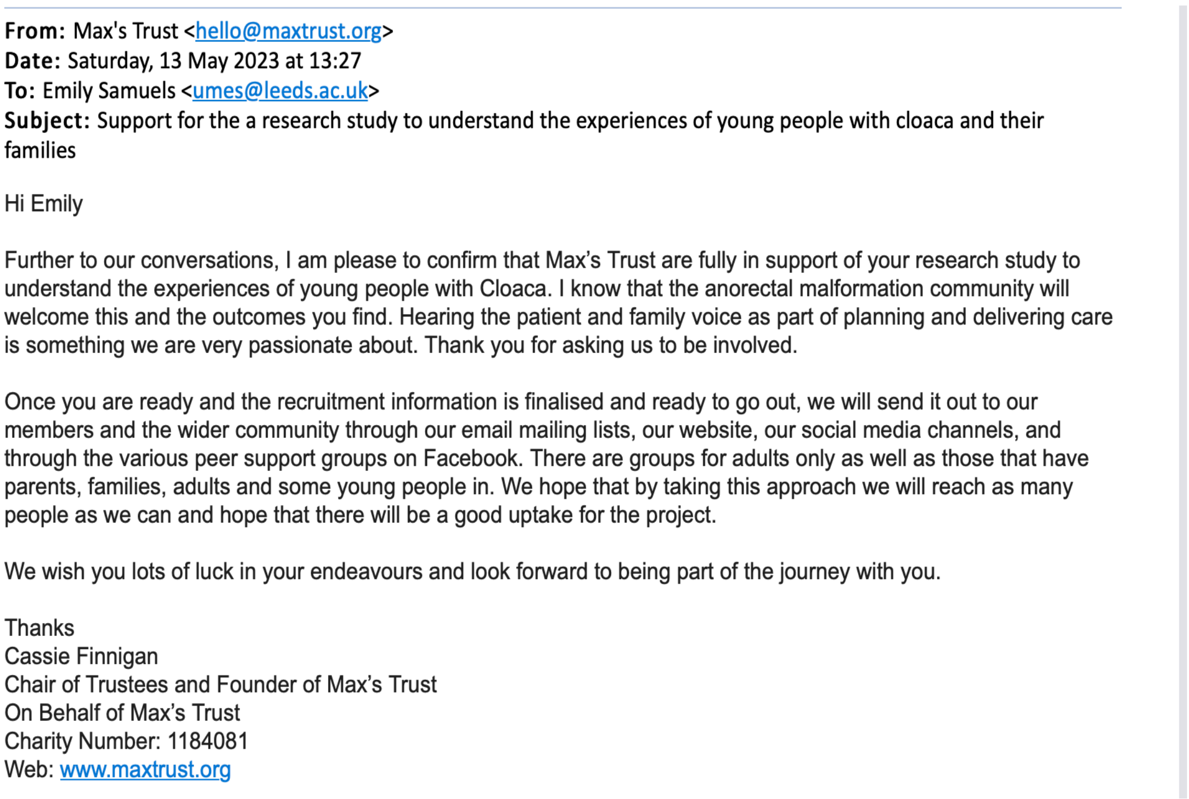
Appendix E

Excerpt from transcript to show example of coding

S2: Yeah, because it becomes such a big thing. So, I would be reading these messages and I'd be like I can't even prove it… I can't even prove that I wasn't born with a penis. Cause if I say I was born with cloaca, they'll google Cloaca and it sounds horrific on Google as well... so, like I can't win.	Feeling the pressure to prove she wasn't born with a penis/debunk the rumour Awareness of the condition being hard to understand and explain
S1: mmm	
S2: So, it's just like I don't know what to say. I was just like, no, I wasn't, no, I wasn't, no I wasn't. I remember just being like denying it, but they're gonna, you know, it's just why would this grown woman say that…it has to be true. It was crazy. It was so weird. Why she did that, I don't know.	Questioning the motivations/motives of this woman Trying to deny the rumour Experiencing others' believing the rumour
S1: it sounds like it felt like you felt forced to share more than you wanted to at that point, is that right?	
S2: Yeah, because I said, I kept everything through school in my teens a secret. I was only open about being born with and cloaca probably about 21 after I'd had an ostomy bag. Through school and my teens, I wouldn't tell anyone. Like I said, my close, close friends knew I had stomach (emphasis) problems and so they could be like 'ohh [name] is off school because she's got stomach problems', but I never went into detail like 'ohh I'm constipated or you know, I have to have enemas' and things like that, I never told anyone like that because it was embarrassing for me.	Keeping cloaca a secret throughout her teens Disclosing the minimum amount of information Keeping it vague
S1: Mmmm	
S2: I just kept everything a secret. So yeah, I was so scared of people finding the truth, which is stupid when I think of it now, because I am so open about it. But as a teenager, especially I suppose back then as well, I think things are less taboo now. I think people are more open about disabilities and illness, and ostomy bags, and especially in the media now, so it's not as taboo, whereas back then people weren't really open about things like that, so in my head I thought, Oh my God, I don't want people to find out anything about me… how am I supposed to explain that I wasn't born with a penis, but I was born with all these other things. But you don't even know what they are because they're so complex, teenagers are not going to understand what the heck cloaca is. Also, teenagers hearing the word anus will just find it funny. Do you know what I mean? I just felt I can't explain myself to these teenagers. They didn't understand that I was panicking and upset, but like most rumours, as long as I didn't react to it, it kind of died away and people forgot.	Reflecting on being scared for people to find out the truth Revaluating how she feels about keeping her condition a secret Perceiving an increased openness in society about disability, illness and ostomy bags (Previously) not wanting anyone to find anything out about her Worrying that people would find it funny Others not understanding her response/distress to the rumour

Appendix E

Confirmation of support from Max's Trust



From: Max's Trust <hello@maxtrust.org>
Date: Saturday, 13 May 2023 at 13:27
To: Emily Samuels <umes@leeds.ac.uk>
Subject: Support for the a research study to understand the experiences of young people with cloaca and their families

Hi Emily

Further to our conversations, I am please to confirm that Max's Trust are fully in support of your research study to understand the experiences of young people with Cloaca. I know that the anorectal malformation community will welcome this and the outcomes you find. Hearing the patient and family voice as part of planning and delivering care is something we are very passionate about. Thank you for asking us to be involved.

Once you are ready and the recruitment information is finalised and ready to go out, we will send it out to our members and the wider community through our email mailing lists, our website, our social media channels, and through the various peer support groups on Facebook. There are groups for adults only as well as those that have parents, families, adults and some young people in. We hope that by taking this approach we will reach as many people as we can and hope that there will be a good uptake for the project.

We wish you lots of luck in your endeavours and look forward to being part of the journey with you.

Thanks
Cassie Finnigan
Chair of Trustees and Founder of Max's Trust
On Behalf of Max's Trust
Charity Number: 1184081
Web: www.maxtrust.org