

Parents' experiences of feeding children born with oesophageal atresia/tracheo-oesophageal fistula.

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Abstract

Background: Feeding difficulties are widely acknowledged following oesophageal atresia/tracheo-oesophageal fistula repair. However, little is understood about the nature and severity of these difficulties. This study explored feeding in children with oesophageal atresia/tracheo-oesophageal fistula from the parent perspective.

Methods: In collaboration with a patient support group, data were collected using a research-specific online discussion forum. Thematic analysis was used to code the data and identify themes.

Results: One hundred and twenty-seven parents registered for the online forum, of whom 83 (65%) provided demographic data. Seventy-four (89%) of responders were mothers, 75 (90%) were of white ethnicity, 65 (78%) were from the UK. Six key themes were identified: feeding is a traumatic experience, feeding my child is scary, feeding is isolating and filled with uncertainty, feeding outside of the home is difficult, feeding associated emotions, developing coping strategies. Parents described features of medical, nutritional, feeding skill and psychosocial dysfunction across all stages of eating/drinking development. They described how their child's feeding difficulties had an impacted their own well-being. An interactional model of feeding difficulties in OA/TOF is proposed.

Conclusions: Exploring parent experiences provides rich data from which to expand understanding of the complex nature of feeding difficulties in OA/TOF. Feeding should be viewed as a dyadic process, occurring within a

family system. Intervention for feeding difficulties should be family-centred, addressing parental anxiety, trauma and uncertainty, as well as the child's underlying medical/surgical needs to optimise outcome. Further study of clinical correlates with parental experience is required.

Key words: esophageal atresia, tracheo-esophageal fistula, feeding difficulties, deglutition, nutrition, parent.

Level of evidence: IV (non-experimental, qualitative)

Abbreviations:

CPR Cardiopulmonary resuscitation

OA Oesophageal Atresia

PTSD Post-traumatic stress disorder

QOL Quality of life

TOF Tracheo-oesophageal fistula

1. Introduction

Following surgical repair, long-term eating and drinking difficulties are one of the most significant problems affecting those born with oesophageal atresia (OA) and tracheo-oesophageal fistula (TOF)[1-3], placing a considerable burden on patients, parents and health services [4] . Besides delayed introduction of oral feeding, establishing eating and drinking in children with OA/TOF is complicated by other factors including: oesophageal dysmotility, oesophageal stricture, upper airway abnormality, and tracheomalacia, which can result in bolus obstruction, gastro-oesophageal reflux, aspiration and negative mealtime experiences [5-8] .

The literature describing the type and nature of the feeding difficulties experienced by children with OA/TOF is sparse. The common symptoms are reported: coughing or choking when eating, difficulty with specific textures of food, needing water to move food through the oesophagus and prolonged mealtimes, but details of the underlying causes, severity and prevalence are lacking [1, 3]. A recent national study reported that 42% of children born with OA/TOF met the criteria for a paediatric feeding disorder [9]. Feeding difficulties have been identified as a significant contributor to reduced quality of life [10, 11] and faltering growth in those with OA/TOF [2].

It is important to acknowledge that feeding, eating and drinking in children is a dyadic interaction, occurring between parent/caregiver and child and believed to be important to bonding and nurturing. Parents are integral to ensuring the child receives the appropriate amount and type of food/drink, in

an appropriately structured manner, to grow and develop [12]. Yet, for many parents feeding is stressful. A mismatch between the parental expectation of a feed/mealtime with the child's ability or desire to eat creates a feeding difficulty and anxiety which can negatively impact parent-child interaction [12]. . Thus, it is vital that both parent and child factors are considered when assessing feeding function and difficulties.

Recent research has suggested that parents of children with OA/TOF experience increased anxiety compared to the general population [3, 13, 14]. However, our understanding of how feeding their child may contribute to this anxiety is very limited. This study aimed to explore parents' lived experience of feeding in children born with OA/TOF with the following specific objectives:

- To describe parents' experiences of feeding, eating and drinking through childhood.
- To explore how the process of feeding impacts on parental well-being.
- To describe parental experiences of support for managing feeding, eating and drinking.

2. METHOD

A phenomenological approach underpinned use of an online forum to explore parents' experiences of feeding, eating and drinking in children born with OA/TOF. Phenomenological research informs, or even re-orientates, our understanding of a phenomenon through analysis of an individual's lived experience [15]. Using systematic analysis of narrative data, commonalities are identified to develop a generalizable description.

Data were collected using a previously described online forum method [16, 17].

In collaboration with TOFS, the UK support group for OA/TOF, a research-specific, private Facebook group was launched. An experienced member of the TOFS Facebook group, independent of the research team, moderated the forum. This online forum was part of a larger study that was granted ethical approval (NRES 20/LO/0098).

2.1 Participants

Convenience sampling was used to recruit parents of children aged 0-18 years with OA/TOF living in the UK. The TOFS support group advertised participation to their members by email and on their Facebook group.

Interested parents were asked to apply to join the research Facebook group, with access granted by the moderator after participants consented to participation by agreeing the group "rules", acknowledging responses would be anonymised and passed to the research team. Participants provided

demographic data via a link to a separate Survey Monkey questionnaire.

These data were used to describe group characteristics but were not linked to individual responses.

2.2 Data collection

Twelve questions, provided in table 1, were co-developed with a public involvement group consisting of four parents of children with OA/TOF, one dietitian and one speech and language therapist and reviewed by the medical advisory group at TOFS, ensuring pertinent issues were explored in a sensitive manner. Questions were posted individually by the moderator. A new question was posted once there were no further comments to the existing question. The moderator answered participant questions and prompted for clarification or explanation if required. Participants were also able to respond privately to the moderator. Parents not using Facebook, or not wishing to share information on the forum, could participate via email.

2.2.1 Data analysis

All responses were anonymised by the moderator. Thematic analysis was conducted [18]. Data familiarisation was undertaken by AS, JW, CS and RG to enable discussion and reflection and then coded line-by-line by AS. Coding was conducted primarily at a semantic level to identify explicitly stated experiences of parents. Codes were then organised into themes, summarising the parents' experiences, and discussed with the whole research team. Tables of codes with supporting quotes were reviewed by

the PPI group providing reflections on the data from different personal perspectives.

3. Results

3.1 Participants

One hundred and twenty-seven individuals joined the research-specific Facebook group, 83 (65%) of whom provided demographic data, presented in table 2.

3.2 Themes

The results of the analysis are summarised in figure 1. Parents described anatomical and physiological abnormalities that led to a wide variety of clinical presentations. Six key themes, relating to parents' experiences of feeding, eating and drinking were identified and are outlined in the purple boxes. The data generated an overall impression of feeding being burdensome and anxiety-provoking for participants. Parent experiences covered all stages of feeding development, signifying that feeding difficulties are not related to a single time point but, due to the developmental nature of eating and drinking in early childhood, can be pervasive over years. The themes and illustrative quotes are presented in table 3. The quotes are referenced throughout the narrative summary.

3.2.1 Feeding my child is scary

Many parents expressed fear when feeding their child. Feeding was described as “*terrifying*”, “*petrifying*”, “*worrying*” and “*nerve-wracking*”. For some this fear was associated with initial feeding attempts, which then lessened as feeding progressed successfully (Q1).

Others described a fear that was constant, seemingly not diminishing even with positive experiences (Q2).

Parents described being repeatedly exposed to this fear-inducing situation as it occurs at every mealtime.

“...every feed I worry and prepare for the worst. I am on edge, I hold my breath...I don't take my eyes off him for a second”

Parents described multiple events where their child would experience food sticking, gagging, vomiting or going blue. They expressed fear at the unpredictable nature of each feed.. Feeding was seen as an event that could end in needing to go to hospital to remove food that had become stuck or in the child having acute, blue episodes.

A few parents reported preparing for the worst when their child first started weaning onto pureed foods.

“It was nerve wracking. I used to have my phone in one hand ready to dial 999 [emergency services] and a spoon in the other!”

Transitioning between feeding milestones, for example, smooth pureed foods, lumpy and chewable foods or eating away from the family, was identified as particularly challenging, causing increased fear. These events occur throughout early childhood and parents described anticipatory fear, as well as fear during the actual event (Q5).

Several parents described being “brave” when feeding their child, particularly around offering different types of food. Parents demonstrated good awareness of their anxiety and the potential impact on their child’s attitude towards eating, with some describing hiding their fear (Q6),

3.2.2 Feeding is a traumatic experience

Some parents felt that feeding was not just anxiety-inducing at the time, but that their experiences led to longer lasting trauma. Some recalled a single traumatic feeding event, while others described repeated events that led to the development of trauma responses (Q7, Q8),

“the scariest blue-spell happened some 10 days upon being home...it was and still is the most horrifying moment of my entire life...this is one thing that stayed with me always and I still can feel the horror of it...from that moment on every feeding was a nightmare for me that I tried to cover up as hard as I could...but the PTSD [post traumatic stress response] with me stays...”

Descriptions of traumatic events included first feeding attempts and during weaning onto solids foods. Blue episodes, or those perceived as life-threatening, were vividly described as having a long-lasting impact for parents, although were not the only cause of trauma responses.

3.2.3 Feeding is isolating and filled with uncertainty

Many parents reported feeling uncertainty around their child’s feeding.

Parents felt unsure about what is “normal” and what is not. There was

uncertainty about what foods to give, when to try new foods and the speed at

which to progress. Parents described this uncertainty as being stressful (Q9).

. Many parents felt that they were told to “have a go” when weaning their child. They felt dissatisfied that they were left to try different foods and find their way through the process.

Some parents described needing to “fight” to get professional help. There were reports of not being believed that something was wrong and delays accessing appropriate professionals (Q10).

Parents also described lacking confidence in some professionals’ understanding of their child’s difficulties. Accessing specialists with knowledge of OA/TOF-specific feeding was particularly difficult (Q11).

Parents also felt isolated due to –limited support from family and friends, as they did not understand the complexity of the child’s difficulties. This led to some feeling that they were the only one able to feed their child, to the level of excluding their partner (Q12, Q13).

Some parents expressed difficulty about family and friends not understanding the complexity of the situation, again adding to feelings of isolation (Q14).

Parents described not feeling able to leave their child, or having very limited number of people who could support childcare, due to feeling that others did not understand risks or not feeling that it was not fair to give someone else the responsibility of feeding their child (Q15).

“Only a few have been involved with our little boy, my mum and 2 trusted friends are completely trusted with him eating/drinking. Not only for our peace of mind but there’s many that are not confident in managing meals.”

3.2.4 Feeding difficulties make eating outside of the home difficult

Numerous parents reported that eating outside of the home was difficult, often due to concern about managing if food got stuck. Parents described it being easier to manage at home (Q17). Other parents reported taking “easy” foods, such as pureed foods, with them or carefully checking the menu for suitable foods to avoid food getting stuck. For some the reluctance to eat out extended to attending family gatherings or meals with friends, as well as eating in a restaurant.

Numerous parents reported negative reactions from strangers, including panic, staring or negative remarks when food got stuck when eating out (Q18).

They described needing to develop resilience to the negative reactions. For some this enabled them to continue eating out, for others it caused them to avoid these situations.

Eating outside of the home was also difficult when others were taking responsibility for feeding the child. Parents described situations where others “pushed” feeding, by giving more challenging foods that the parent would not usually offer. For some this was positive, enabling them to gain confidence in their child’s skills. For others this caused distress, distrust and conflict with family members or caregivers (Q19).

Many parents described carefully preparing for eating outside of the home for example by training others and providing meticulous written guidance, particularly for childcare settings. Others described how family members had voluntarily done CPR/choking management training in preparation for feeding their child (Q20).

“I had to provide a lot of information and speak to her keyworker at length about what to look out for and what to do or not do.”

3.2.5 Feeding associated emotions

Some parents identified that although feeding their child was scary, it was also exciting. A number expressed the immense pride that they felt when their child managed a new food or did better than expected (Q21).

Mealtime fear and anxiety were juxtaposed with joy when their child fed well. Despite the challenges, there was an awareness that supporting their child to feed had been an enriching experience (Q22). Alongside feelings of failure and guilt when feeding was less successful, there was appreciation when feeding went well or better than expected that other parents would likely take for granted.

3.2.6 Developing coping strategies

Many parents described developing ways to cope and that, with time, the anxiety eases. Improvements related to being less worried about doing something wrong, gradually gaining confidence through taking small steps,

having positive experiences, and having increased knowledge about what to do when feeding becomes difficult.

Parents identified mechanisms that supported coping and resilience. Peer support and that of the TOFS support group were often cited as being very positive, alongside knowledgeable professionals and the help of family and friends (Q23, Q24, Q25, Q26).

Many parents described the strategies that they had developed to support their child's feeding, such as eating slowly, taking small bites, chewing well, drinking after a mouthful of food, and making foods more liquid. Learning and adapting to their child's feeding difficulties over time enabled parents to cope with feeding-related anxiety.

3.3 Proposed model of the feeding dyad in OA/TOF

Figure 2 presents a proposed model of the feeding, eating and drinking dyad in children born with OA/TOF. The lived experiences described in the data are encapsulated in the concept of feeding-related quality of life (QOL), i.e. the enjoyment of mealtimes, participating in social activity involving eating/drinking and feeling confident in their own and others' ability to feed their child. The derived themes highlight factors, relating to both the child and the parent, which impact on feeding-related QOL. The recognition of feeding as a dyadic process, and thus the inter-relation of these factors is supported, although further validation of the strength and direction of these relationships is required.

4. DISCUSSION

This research provides a unique insight into the reality of feeding, eating and drinking in children born with OA/TOF from the parent perspective. The research highlights the considerable anxiety and uncertainty parents experience when feeding their child, particularly at times of transition through feeding milestones. Social isolation because of their child's feeding difficulties is evident. The need to consider these parent factors in the assessment and management of OA/TOF-related feeding difficulties is supported.

4.1 Fear and anxiety

Establishing oral feeding was clearly very stressful. The data strikingly highlights that parents face anxiety at every mealtime or feed, an unavoidable event that occurs multiple times a day.

Recent work has demonstrated increased levels of anxiety generally in parents of children with OA/TOF [13, 14, 19, 20], but there has been little exploration of anxiety specific to eating and drinking. Wallace and colleagues used feeding difficulty as a co-variate in their analysis of parental anxiety in OA/TOF. They found that requiring speech and language therapy input for feeding difficulties (used as a proxy for having feeding difficulties), was the only child-related factor associated with higher levels of anxiety in parents [13]. Bevilacqua and colleagues identified that parental anxiety at mealtimes was higher in those with more frequent choking episodes [3].

Evidence from the general population and those with other chronic health conditions shows that mothers of children with feeding difficulties have elevated anxiety and stress [21, 22], related to inadequate weight gain and not knowing how the child would develop in the future [23]. Our study, however, described the actual process and unpredictability of feeding as anxiety-inducing. This presentation differs from coughing associated with oropharyngeal dysfunction or gagging due to sensory impairment that is often seen in children with neurological, cardiac or gastroenterological conditions [24], suggesting that the anxiety experienced by parents of children with and without OA/TOF may differ but may be a significant contributor to raised parental anxiety. Further research, using validated assessments of feeding ability and anxiety, is required to assess the strength of this potential association.

The direction of any relationship between OA-related feeding difficulties and parental anxiety also remains unclear. Does having a child with more significant feeding difficulties cause increased anxiety, or are the feeding difficulties exacerbated by parents who are more anxious? Parents with higher levels of anxiety or depression may find it more difficult to read their child's feeding cues, provide appropriate mealtime structure and cope when presented with medical conditions that impair feeding [21]. Research undertaken with ex-preterm toddlers found mothers with higher anxiety or depression were more controlling and intrusive in their mealtime interactions, hindering their child's independent feeding and this was associated with higher toddler distress, avoidance of eating and negative mealtime behaviours [25]. A recent study found 42% of children with OA/TOF

presented with a feeding disorder, displaying oppositional and aversive behaviours causing stressful mealtimes [9]. It is evident that assessment of the dyadic feeding relationship could provide valuable insight into the nature of such feeding difficulties, which would guide intervention and improve outcome. Further evaluation of the extent of the impact on parental well-being and whether anxiety, depression or stress is a driver for feeding difficulties in this population is required.

4.2 Trauma responses

Some parents described feeding as a traumatic experience, referencing post-traumatic stress responses. Previous research identified 59% of parents of children with OA/TOF scored above the cut-off for post-traumatic stress disorder (PTSD) [20]. Post traumatic stress disorder was not associated with severity of the neonatal course or OA/TOF sequelae at the time of assessment. Feeding was not used as a co-variate in the analysis. This study indicates that the experiences of establishing oral feeding may contribute significantly to the development of PTSD, warranting further exploration. Multi-disciplinary teams should be aware of the potential for severe anxiety and post-traumatic responses, screen for such difficulties, particularly in parents whose children experience blue episodes, and ensure appropriate onward referral.

4.3 Isolation and uncertainty

An increasing body of evidence indicates that parents of children with OA/TOF have reduced QOL [13, 14, 19]. Alongside the potential for feeding-

related anxiety and trauma to impact QOL, our study identified that parents experience social isolation due to a lack of confidence in others being able to feed their child, others not understanding the extent of the difficulties, avoiding social situations involving eating and not being able to rely on friends and family for support due to their lack of confidence in feeding the child. Social isolation resulting from caring for a child with feeding difficulties has been previously reported [23, 26, 27] and having a good support system has been associated with better QOL [28]. In a clinical situation it can be easy to view feeding as the responsibility of one parent, often the mother. Identifying the feeding process as a family system is key to effectively managing difficulties [29]. Intervention at a family level, for example by facilitating information sharing, can help manage potential conflict and give confidence to the parent that others can care for their child, reducing feelings of isolation [30].

Uncertainty as to where to get appropriate support was also evident. Parents described variable access to appropriate professional support, with reports of needing to “fight” for support and feeling like they were left to establish feeding alone. Service delivery for complex, rare conditions, such as OA/TOF, has been under scrutiny in recent years with calls for centralised, specialist centres where the child and family can access appropriate multi-disciplinary support to optimise outcome, including from patient support groups/charities [6, 31, 32] . As evidenced in non-OA/TOF populations, feeding outcome, including parental well-being, can be improved with appropriate access to specialist intervention and better parental understanding of the problem [33].

4.4 Nature of feeding/swallowing difficulties

The complexity and need for specialist support were highlighted by the parent descriptions of different causes of swallow impairment (oesophageal dysmotility/strictures, oro-pharyngeal dysfunction, recurrent TOF, gastro-oesophageal reflux, tracheomalacia) and symptoms of impairment (blue episodes, coughing, gagging, vomiting, aspiration, bolus obstruction, longer mealtimes, slower feeding development and faltering growth). Such risk factors are widely acknowledged and recent studies have attempted to characterise OA/TOF related feeding difficulties [2, 3, 9, 34, 35], yet their exact nature and prevalence have been poorly defined [36]. Agreed terminology for describing feeding difficulties in clinical practice and research would aid understanding of their nature prevalence and strengthen evidence synthesis. Until recently there was no universally accepted definition of a feeding difficulty. In 2019, the term “pediatric feeding disorder”, defined as “impaired oral intake that is not age appropriate, and is associated with medical, nutritional, feeding skill, and/or psychosocial dysfunction” (p. 125), was proposed and diagnostic criteria outlined [37]. Pham and colleagues recently used this term to highlight the frequency of feeding-related psychosocial dysfunction in the OA/TOF population [9], but did not describe other areas of dysfunction. Our findings and proposed model indicate that OA/TOF feeding disorders are associated with interacting medical, nutritional, feeding skill *and* psychosocial “dysfunction”. We suggest OA/TOF requires assessment of all four aspects of feeding disorders in order that the root causes are understood, and the interaction of child and parent factors are considered in treatment [37]. Use of a consistent framework for multi-

disciplinary, holistic assessment of swallow function, feeding difficulties and QOL would be valuable in clinical care and future research.

4.5 Limitations

This study used an asynchronous, online data collection method which facilitated participation from a larger number of parents than is typically delivered by qualitative research. There was excellent geographical representation from the UK and 20% of responses were from outside the UK, indicating findings have international significance. While rich data, containing information that went beyond a surface description, were obtained, it is acknowledged that other qualitative methods, such as interviews, may have afforded greater depth of understanding.

Participants were overwhelmingly of white ethnicity and mothers. This is likely due to the use of the patient support group for recruitment and Facebook for data collection. The participant demographics are likely to be representative of individuals most likely to access the patient support group and be able and comfortable sharing their experiences on Facebook. As improved coping has been associated with communication with parents of children with the same problems [33], those not accessing the support group may not develop coping strategies as effectively and be at greater risk of poor outcome. Alternatively, those not accessing peer support may be coping well and not feel it is required. We acknowledge this selection bias and that our findings may not be reflective of the experiences of those with different cultural expectations of parenting and feeding, those with limited literacy or English language skills or those who do not access patient support

groups. Alternative qualitative methods, such as interviews which could employ purposive sampling to ensure greater diversity of participants, would add valuable insights.

It should also be noted that the online forum ran in November-December 2020, at a time when the COVID pandemic restricted access to professional support and may have increased feelings of isolation.

4.6 Conclusions

This study describes the daily reality faced by parents of children with OA/TOF in establishing eating and drinking throughout early childhood and beyond. The impact of the feeding/swallowing difficulties reaches beyond the child's health and development to parental well-being and quality of life. The importance of recognising feeding as a dyadic process is evident. Thus, a family-centred approach, addressing parental anxiety, trauma, uncertainty, and isolation is key to improving feeding outcomes in children with OA/TOF. Further research exploring how clinical factors, such as surgical course, explain the phenomena described herein and quantifying the strength of the association between feeding and parental well-being will further characterise OA/TOF feeding difficulties and guide multi-disciplinary care pathways and intervention approaches.

Figure legends

Figure 1. Thematic map of parents' experience of feeding in OA/TOF

Figure 2. Proposed model of feeding dyad in OA/TOF

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References

- [1] Ax SO, Abrahamsson K, Gatzinsky V, et al. Parent-Reported Feeding Difficulties among Children Born with Esophageal Atresia: Prevalence and Early Risk Factors. *Eur J Pediatr Surg.* 2021;31:69-75.
<https://doi.org/10.1055/s-0040-1716880>
- [2] Traini I, Menzies J, Hughes J, et al. Oesophageal atresia: The growth gap. *World J Gastroenterol.* 2020;26:1262-72.
<https://doi.org/10.3748/wjg.v26.i12.1262>
- [3] Bevilacqua F, Ragni B, Conforti A, et al. Fixed the gap, solved the problem? Eating skills in esophageal atresia patients at 3 years. *Dis Esophagus.* 2020;33. <https://doi.org/10.1093/dote/doz102>
- [4] Svoboda E, Fruithof J, Widenmann-Grolig A, et al. A patient led, international study of long term outcomes of esophageal atresia: EAT 1. *J Pediatr Surg.* 2017. <https://doi.org/10.1016/j.jpedsurg.2017.05.033>

- [5] Faure C, Righini Grunder F. Dysmotility in Esophageal Atresia: Pathophysiology, Characterization, and Treatment. *Front Pediatr*. 2017;5:130. <https://doi.org/10.3389/fped.2017.00130>
- [6] Koumbourlis AA-O, Belessis YA-O, Cataletto MA-O, et al. Care recommendations for the respiratory complications of esophageal atresia-tracheoesophageal fistula. 2020.
- [7] Baxter KJ, Baxter LM, Landry AM, et al. Structural airway abnormalities contribute to dysphagia in children with esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg*. 2018. <https://doi.org/10.1016/j.jpedsurg.2017.12.025>
- [8] Mahoney L, Rosen R. Feeding Difficulties in Children with Esophageal Atresia. *Paediatr Respir Rev*. 2016;19:21-7. <https://doi.org/10.1016/j.prrv.2015.06.002>
- [9] Pham A, Ecochard-Dugelay E, Bonnard A, et al. Feeding disorders in children with oesophageal atresia: a cross-sectional study. *Arch Dis Child*. 2022;107:52-8. <https://doi.org/10.1136/archdischild-2020-320609>
- [10] Dellenmark-Blom M, Dingemann J, Witt S, et al. The Esophageal-Atresia-Quality-of-life Questionnaires: Feasibility, Validity and Reliability in Sweden and Germany. *Journal of Pediatric Gastroenterology & Nutrition*. 2018;67:469-77.
- [11] Dellenmark-Blom M, Quitmann J, Dingemann J, et al. Clinical Factors Affecting Condition-Specific Quality-of-Life Domains in Pediatric Patients after Repair of Esophageal Atresia: The Swedish-German EA-QOL Study. *Eur J Pediatr Surg*. 2020;30:96-103. <https://doi.org/10.1055/s-0039-1693729>

- [12] Berlin KS, Davies WH, Lobato DJ, et al. A Biopsychosocial Model of Normative and Problematic Pediatric Feeding. *Children's Health Care*. 2009;38:263-82. <https://doi.org/10.1080/02739610903235984>
- [13] Wallace V, Honkalampi K, Sheils E. Anxiety and depression in parents of children born with esophageal atresia: An international online survey study. *J Pediatr Nurs*. 2021;60:77-82. <https://doi.org/10.1016/j.pedn.2021.02.016>
- [14] Witt S, Dellenmark-Blom M, Dingemann J, et al. Quality of Life in Parents of Children Born with Esophageal Atresia. *Eur J Pediatr Surg*. 2019;29:371-7. <https://doi.org/10.1055/s-0038-1660867>
- [15] Neubauer BE, Witkop CT, Varpio L. How phenomenology can help us learn from the experiences of others. *Perspect Med Educ*. 2019;8:90-7. <https://doi.org/10.1007/s40037-019-0509-2>
- [16] Wray J, Brown K, Tregay J, et al. Parents' Experiences of Caring for Their Child at the Time of Discharge After Cardiac Surgery and During the Postdischarge Period: Qualitative Study Using an Online Forum. *Journal of Medical Internet Research*. 2018;20. <https://doi.org/10.2196/jmir.9104>
- [17] Stewart A, Smith CH, Eaton S, et al. COVID-19 pandemic experiences of parents caring for children with oesophageal atresia/tracheo-oesophageal fistula. *BMJ Paediatr Open*. 2021;5:e001077. <https://doi.org/10.1136/bmjpo-2021-001077>
- [18] Braun V, Clarke V. *Thematic analysis: A practical guide*. London: SAGE; 2022.
- [19] Tan Tanny SP, Trajanovska M, Muscara F, et al. Quality of Life Outcomes in Primary Caregivers of Children with Esophageal Atresia. *J Pediatr*. 2021. <https://doi.org/10.1016/j.jpeds.2021.07.055>

- [20] Le Gouez M, Alvarez L, Rousseau V, et al. Posttraumatic Stress Reactions in Parents of Children Esophageal Atresia. PLoS One. 2016;11:e0150760. <https://doi.org/10.1371/journal.pone.0150760>
- [21] Jones CJ, Bryant-Waugh R. The relationship between child-feeding problems and maternal mental health: a selective review. Advances in Eating Disorders. 2013;1:119-33. <https://doi.org/10.1080/21662630.2013.742972>
- [22] Silverman AH, Erato G, Goday P. The relationship between chronic paediatric feeding disorders and caregiver stress. J Child Health Care. 2021;25:69-80. <https://doi.org/10.1177/1367493520905381>
- [23] Lamm K, Hallstrom IK, Landgren K. Parents' experiences of living with a child with Paediatric Feeding Disorder: An interview study in Sweden. Scandanavian Journal of Caring Sciences. 2022;00:1-10.
- [24] Rommel N, De Meyer A-M, Feenstra L, et al. The complexity of feeding problems in 700 infants and young children presenting to a tertiary care institution. Journal of pediatric gastroenterology and nutrition. 2003;37:75-84.
- [25] Salvatori P, Andrei F, Neri E, et al. Pattern of mother-child feeding interactions in preterm and term dyads at 18 and 24 months. Front Psychol. 2015;6:1245. <https://doi.org/10.3389/fpsyg.2015.01245>
- [26] Garro A, Thurman SK, Kerwin ME, et al. Parent/caregiver stress during pediatric hospitalization for chronic feeding problems. J Pediatr Nurs. 2005;20:268-75. <https://doi.org/10.1016/j.pedn.2005.02.015>
- [27] Estrem HH, Thoyre SM, Knafel KA, et al. "It's a Long-Term Process": Description of Daily Family Life When a Child Has a Feeding Disorder. J Pediatr Health Care. 2018;32:340-7. <https://doi.org/10.1016/j.pedhc.2017.12.002>

- [28] Pedersen SD, Parsons HG, Dewey D. Stress levels experienced by the parents of everally fed children. *Child: care, health and development*. 2004;30:507-13.
- [29] Parrish M. Family adaptation to a child's feeding and swallowing disorder: a social work perspective. *Seminar in Speech and Language* 1997;18:71-8.
- [30] Hewetson R, Singh S. The lived experience of mothers of children with chronic feeding and/or swallowing difficulties. *Dysphagia*. 2009;24:322-32.
- [31] Dingemann C, Eaton S, Aksnes G, et al. ERNICA Consensus Conference on the Management of Patients with Esophageal Atresia and Tracheoesophageal Fistula: Follow-up and Framework. *Eur J Pediatr Surg*. 2020;30:475-82. <https://doi.org/10.1055/s-0039-3400284>
- [32] Slater G, Faulkner J. Towards a holistic model for the treatment of oesophageal atresia. TOFS2021.
- [33] Garro A. Coping patterns in mothers/caregivers of children with chronic feeding problems. *J Pediatr Health Care*. 2004;18:138-44. <https://doi.org/10.1016/j.pedhc.2003.08.001>
- [34] Menzies J, Hughes J, Leach S, et al. Prevalence of Malnutrition and Feeding Difficulties in Children With Esophageal Atresia. *J Pediatr Gastroenterol Nutr*. 2017;64:e100-e5. <https://doi.org/10.1097/MPG.0000000000001436>
- [35] Baird R, Levesque D, Birnbaum R, et al. A pilot investigation of feeding problems in children with esophageal atresia. *Dis Esophagus*. 2015;28:224-8. <https://doi.org/10.1111/dote.12178>

- [36] Krishnan U, Mousa H, Dall'Oglio L, et al. ESPGHAN-NASPGHAN guidelines for the evaluation and treatment of gastrointestinal and nutritional complications in children with esophageal atresia-tracheoesophageal fistula. *Journal of pediatric gastroenterology and nutrition*. 2016;63:550-70.
- [37] Goday PS, Huh SY, Silverman A, et al. Pediatric Feeding Disorder: Consensus Definition and Conceptual Framework. *J Pediatr Gastroenterol Nutr*. 2019;68:124-9. <https://doi.org/10.1097/MPG.0000000000002188>

Table 1: Online forum questions

What were your early experiences of feeding your child? How did you feel in those early days?
For those of you with children who are old enough, what were your experiences of starting weaning/pureed foods? How did offering your child food make you feel?
For those of you with children who are old enough, what were your experiences of moving on to chewable foods? How did moving on to chewable foods make you feel?
Thinking about your child's feeding, what has gone the best and what has been the most challenging aspect for you as a parent?
For those of you with older children, what were your experiences of supporting your child's eating/drinking when they started nursery/early years childcare and/or school?
How have you adapted mealtimes to support your child? How do you promote a positive relationship with food?
What are your experiences of other family members' or friends' involvement with your child's eating/drinking?
What are your experiences of eating out with your child? Do you feel confident eating out with your child (pre-COVID!)?
What has been your experience of supporting your child to feed themselves? How have you helped your child gain independence with eating/drinking?
What was your experience of seeking support or advice for feeding or swallowing related concerns, pre-COVID? Who provided you with the most useful information?
What has been your experience of explaining your child's swallowing to them and others?
If you could give another parent of a child with OA/TOF one piece of advice about feeding, what would it be?

Table 2: Demographic details of participants

		Number (% of responders)
Relationship to child	Mother	74 (89)
	Father	4 (5)
	Individual with OA/TOF*	1 (1)
	Missing	4 (5)
Ethnicity	White	75 (90)
	Asian or Asian British	2 (2)
	Mixed Ethnicity	2 (2)
	Missing	4 (5)
Geographical location	England	53 (64)
	Scotland	10 (12)
	Wales	2 (2)
	Northern Ireland	0 (0)
	Outside of the UK	18 (22)
Respondent age	18-24	1 (1)
	25-34	31 (37)
	35-44	44 (52)
	45-54	7 (8)
	55-64	1 (1)
Age of child	Under 12 months	13 (16)
	12-23 months	16 (19)
	2-4 years	28 (34)
	5-11 years	16 (19)
	12 and over years	6 (7)
	Did not respond	4 (5)

Type of OA/TOF	OA and TOF repaired at birth	70 (84)
	OA and TOF delayed repair	7 (8)
	TOF only (repaired)	4 (5)
	OA only (repaired)	2 (2)
	OA unrepaired	1 (1)

*Unable to verify if any responses were attributed to this individual.

Table 3: Theme summary and illustrative quotes

Theme	Subtheme	Quote reference	Illustrative quote
Feeding my child is scary	Initial fear	Q1	<i>"I was petrified the first few times I fed him"</i>
	Constant fear	Q2	<i>"I'm always scared. Always, even when he goes through a long time of doing well."</i>
		Q3	<i>"...every feed I worry and prepare for the worst. I am on edge, I hold my breath...I don't take my eyes off him for a second"</i>
	Anticipatory fear	Q4	<i>"It was nerve wracking. I used to have my phone in one hand ready to dial 999 [emergency services] and a spoon in the other!"</i>
		Q5	<i>[speaking about moving between textures] "We started weaning 2 months ago...but I feel like I'm not moving on from smooth/runny purees because I'm too scared to try anything else."</i>
	Hiding fear	Q6	<i>"However nervous I was I always tried hard not to show it so that the experience was positive for my daughter."</i>
Feeding is a traumatic experience	Single event	Q7	<i>"I think my first experience scarred me a little. Those earlier memories still haunt me and set me up to feel anxious about feeding..."</i>
	Multiple events	Q8	<i>"We had many many blue episodes including the need for resuscitation as a result of drinking milk/early weaning foods. This has without a doubt left me with a degree of PTSD from everything we've seen him go through."</i>
Feeding is isolating and filled with uncertainty	What is normal?	Q9	<i>"you're constantly asking yourself if your moving too fast or too slow or worrying about what could happen. It's stressful"</i>

	Lack of professional support	Q10	<i>"I felt completely on my own and isolated. Very little support or advice...very much on my own fighting to do the best I could every day"</i>
		Q11	<i>"Most listen and pretend they understand but it's quite clear they have no idea what I'm talking about or little understanding to be able to provide a useful answer".</i>
	Lack of family support	Q12	<i>"even my mother who was our rock when me and my siblings were growing up...she was too lost during such episodes with feeding issues so I relied exclusively on myself."</i>
		Q13	<i>"No one would feed him except for myself and his dad....it was very tough mentally and emotionally."</i>
		Q14	<i>"I don't think anyone really understands the condition and appreciates its challenges unless they have spent a period of time with us and experienced a choking episode."</i>
		Q15	<i>"I didn't really let anyone else look after her or feed her as I didn't feel it was safe and not fair to put that responsibility on them."</i>
		Q16	<i>"Only a few have been involved with our little boy, my mum sons and 2 trusted friends are completely trusted with him eating/drinking. Not only for our peace of mind but there's many that are not confident in managing meals."</i>
Feeding difficulties make eating outside of the home difficult	Avoiding difficult situations	Q17	<i>"I don't feel very comfortable eating out with my 4 year old son as he sometimes gets food stuck and it is easier to manage the situation at home"</i>
	Negative reactions of others	Q18	<i>"I had several people tell me that I shouldn't bring my child out if she has croup/isn't well...if she struggled with</i>

			<i>some food and coughed....I would...throw long medical words at them to shut them up but it was still unpleasant and stressful".</i>
	Conflicting feeding practices	Q19	<i>"They [nursery] were a little gung ho at times, I remember having to remind them no toast. But in hindsight, I think we were being more cautious than we needed to be, and actually nursery did get her eating!"</i>
	Careful preparation	Q20	<i>"I had to provide a lot of information and speak to her keyworker at length about what to look out for and what to do or not do."</i>
Feeding associated emotions		Q21	<i>"Apprehensive and frightened but if she managed it and/or enjoyed it then I felt huge pride."</i>
		Q22	<i>"I never get tired of watching him eat. Little big steps....I'm surprised by what he can manage... I'm also surprised when he can't manage something that seems ok."</i>
Developing coping strategies	Peer support	Q23	<i>"We found the TOF group on Facebook invaluable for information."</i>
		Q24	<i>"I have a group of friends with babies around the same age and they are just wonderful while out and about."</i>
	Knowledgeable professionals	Q25	<i>"Surgical team and professionals very supportive and encouraging"</i>
		Q26	<i>"We have had input from our local SALT team which has been really helpful".</i>

Figure 1

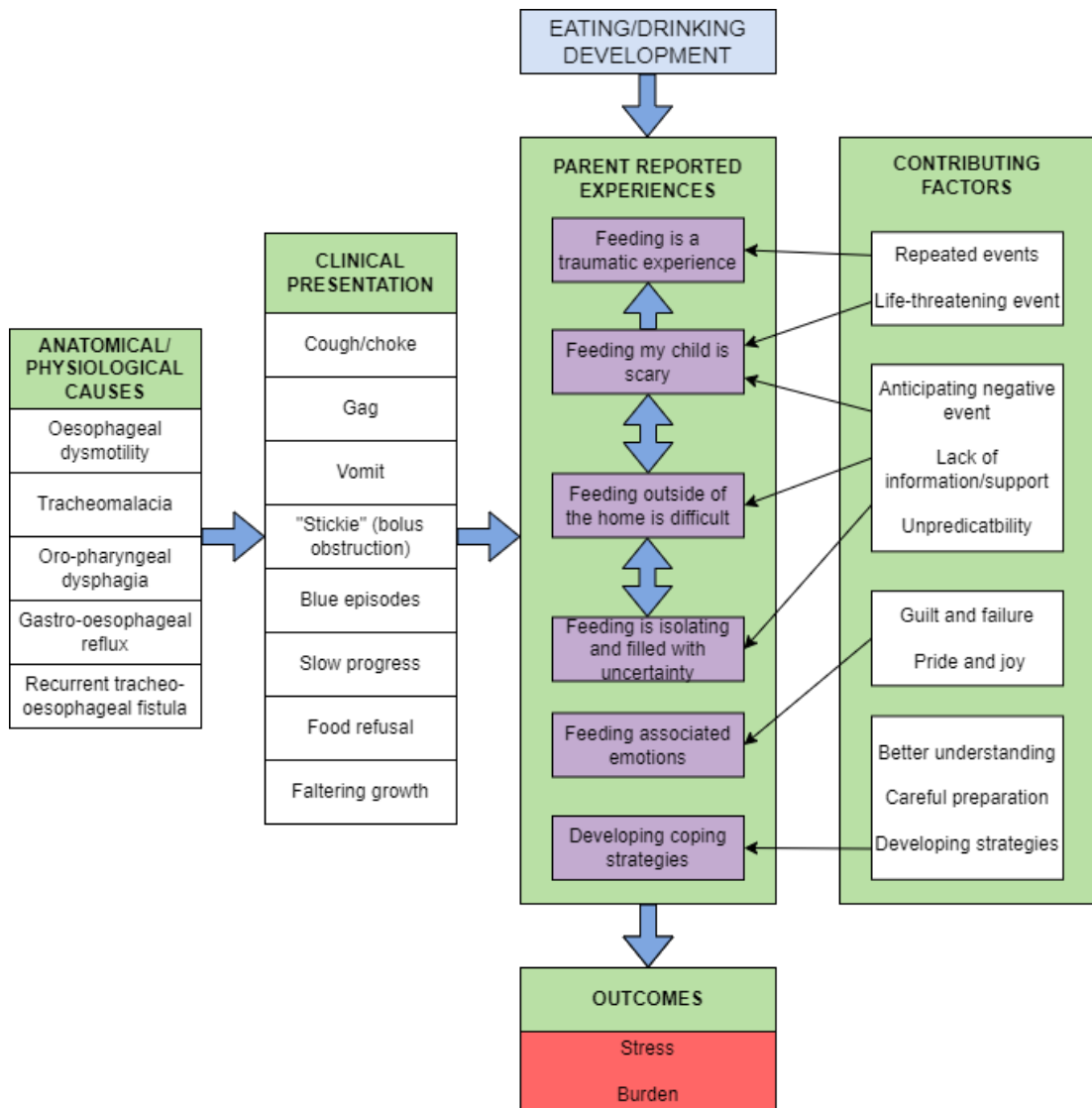


Figure 2

