COVID-19 pandemic experiences of parents caring for children with oesophageal atresia/tracheo-oesophageal fistula

Alexandra Stewart, Christina H Smith, Simon Eaton, Paolo De Coppi, Jo Wray

ABSTRACT

Purpose The COVID-19 pandemic has resulted in a global health crisis of unparalleled magnitude. The direct risk to the health of children is low. However, disease-containment measures have society-wide impacts. This study explored the pandemic experiences of parents of children with oesophageal atresia/tracheo-oesophageal fistula (OA/TOF) in the UK.

Design A phenomenological approach underpinned use of an asynchronous online forum method, in collaboration with a patient support group. Data were evaluated using thematic analysis.

Results The online forum ran between 7 November and 18 December 2020 with 109 participants. Pandemic experiences were divided into themes relating to healthcare and disease containment. Participants described positive experiences with remote healthcare but identified limitations. Delays and cancellations led to escalation of care to an emergency level, slower developmental progress and feelings of being abandoned by services. Inpatient care was perceived as safe but caring alone was emotionally and practically challenging. Disease containment themes revealed anxiety regarding health risks, ‘collateral’ damage to well-being because of isolation, and an impact on finances and employment. Parents described a transition from worry about direct health risks to concern about the impact of isolation on socialisation and development. A process of risk–benefit analysis led some to transition to a more ‘normal life’, while others continued to isolate. Benefits to their child’s health from isolation were reported.

Conclusions Parents’ experiences of caring for a child with OA/TOF during the pandemic were varied. Rapid adoption of telehealth has demonstrated the enormous potential of remote healthcare delivery but requires refinement to meet the needs of the individual. Future pandemic planning should aim to retain community healthcare services to avoid escalation of care to an emergency, manage chronic and developmental concerns, and support parental well-being. Accurate and consistent disease-specific information is highly valued by parents. Third sector organisations are ideally positioned to support this.

What is known about the subject?

► Direct health risk to children from COVID-19 is low but there is a high risk of ‘collateral’ damage from strategies required to contain the virus.
► Patient support groups can be powerful allies in providing accurate and consistent messages, that is particularly useful to those with rare diseases.
► Social media can facilitate rapid and effective data collection in a rare disease cohort.

What this study adds?

► A parent perspective of the impact of pandemic-related reduction in healthcare provision and use of telehealth, highlighting areas of need for pandemic and post-pandemic service delivery.
► An insight into parents’ experiences and decision-making surrounding disease-containment measures which highlights the variation, even within a single, rare disease.
► Isolation has resulted in exceptionally low exposure to usual childhood infections resulting in improved health for children with oesophageal atresia/tracheo-oesophageal fistula.

Although it is now suggested that children are approximately 50% less likely to be infected than adults, account for 1%–5% of cases worldwide and rarely experience severe disease, early data indicated that ‘high-risk’ groups for severe disease existed. As a result, children deemed ‘extremely clinically vulnerable’ were advised to ‘shield’, avoiding all contact with others to minimise their risk of being infected.

One group of vulnerable children are those born with oesophageal atresia/tracheo-oesophageal fistula (OA/TOF). A rare, congenital abnormality, OA/TOF occurs in approximately 1/3500 live births in the UK that results in a blind-ending oesophageal...
pouch and/or an anomalous connection between the trachea and oesophagus. While survival rates following surgical repair are excellent, many children experience long-term health challenges: swallowing dysfunction and feeding difficulties in approximately 80%, gastro-oesophageal reflux and oesophagitis in up to 70%, recurrent respiratory infections and chronic cough in 40%–52%. Approximately 50% of children have other congenital abnormalities, most commonly cardiac abnormalities. Hospital readmission with respiratory or gastrointestinal issues in the preschool years is common.

Despite vulnerability to respiratory infection, children with OA/TOF have not experienced severe COVID-19 symptoms. However, disease containment continues to involve restrictions to social contact, education and non-essential business and impacts society beyond the immediate risk to health. Our aim was to describe parental lived experiences of caring for a child with OA/TOF during the COVID-19 pandemic with the following specific objectives:

1. To describe experiences of accessing healthcare and medical advice.
2. To describe parental experiences of disease-containment measures and their impact.
3. To learn from their experiences and make recommendations for delivery of care for this rare disease.

METHOD
A phenomenological approach underpinned use of an online forum to explore parental experiences of accessing healthcare and the impact of disease-containment measures in the UK during the COVID-19 pandemic.

Data were collected using a previously described online forum method and detailed in the online supplemental material.

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.

Patient and public involvement
Patient and public involvement (PPI), through collaboration with TOFS and use of a PPI steering group (including four parents), has been integral to study design, recruitment, data analysis and dissemination. Details are provided throughout the Methods section.

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 to learn from their experiences and make recommendations for delivery of care for this rare disease.

Table 1 Demographic data

<table>
<thead>
<tr>
<th>Relationship to the child</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Mother</td>
<td>58 (89%)</td>
</tr>
<tr>
<td>Father</td>
<td>3 (5%)</td>
</tr>
<tr>
<td>Adult with OA/TOF</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>Did not respond</td>
<td>3 (5%)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Ethnicity</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>White</td>
<td>61 (94%)</td>
</tr>
<tr>
<td>Asian or British Asian</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>Did not respond</td>
<td>3 (5%)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Geographical location</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>England</td>
<td>55 (82%)</td>
</tr>
<tr>
<td>Scotland</td>
<td>10 (15%)</td>
</tr>
<tr>
<td>Wales</td>
<td>2 (3%)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Age of the child</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Under 2 years of age</td>
<td>25 (38%)</td>
</tr>
<tr>
<td>2–4 years of age</td>
<td>33 (34%)</td>
</tr>
<tr>
<td>5–11 years of age</td>
<td>12 (18%)</td>
</tr>
<tr>
<td>Over 12 years of age</td>
<td>3 (5%)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Type of OA/TOF</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>OA and TOF repaired within a week of birth</td>
<td>55 (85%)</td>
</tr>
<tr>
<td>OA and TOF repaired more than a week after birth</td>
<td>5 (7%)</td>
</tr>
<tr>
<td>OA only</td>
<td>3 (5%)</td>
</tr>
<tr>
<td>TOF only</td>
<td>2 (3%)</td>
</tr>
</tbody>
</table>

OA/TOF, oesophageal atresia/tracheo-oesophageal fistula.
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.

Data collection
Questions for the online forum, provided in box 1, were co-developed with the PPI group and checked by TOFS, ensuring pertinent issues were explored in a sensitive manner. Questions were posted individually by the moderator and participants responded by posting a ‘comment’. Data saturation was assumed once no further comments were being made. A new question was then posted. The moderator answered participant questions, prompted for clarification and invited further responses if required. Participants were able to respond to others’ comments. Participants were also able to respond privately to the moderator rather than posting to the whole group. To diversify participation, parents not using Facebook, or not wishing to share information on the forum, could participate via email.

Data analysis
All responses were anonymised by the moderator and sent to the research team as a Word document. Thematic analysis was conducted. Data were independently coded by three members of the research team, followed by group discussion to agree themes. Two thematic maps were generated. Tables of codes with supporting quotes and the maps were reviewed by the PPI group and two other members of the research team providing data triangulation from different professional and personal perspectives.

RESULTS
The online forum ran from 7 November to 18 December 2020. There were 109 members, of whom 65 completed the demographic survey (table 1) and responded to at least one question. An additional six participants responded by email.

Experiences of accessing healthcare
Participants’ experiences of accessing healthcare are summarised in figure 1. Further illustrative quotes are shown in table 2. Themes were grouped into remote healthcare, delays and cancellations, and hospital care.

Remote healthcare
Access to healthcare changed, with a shift to telehealth (telephone or video) appointments reported by most participants.

Figure 1 Thematic map healthcare. a&e, Accident and Emergency; IPC, infection prevention and control.
communication with healthcare professionals, having telephone or email contact that was not previously available. Where parents felt connected, remote healthcare was positively received. The value of specialist nurses in achieving good communication was evident.

We’ve had huge support from our CNS team who has been amazing throughout the entire journey.

However, limitations resulting in uncertainty were also described. Some parents raised concerns that their child’s health or development was being compromised. Parents reported feeling disconnected from their healthcare team, due to communication or organisational challenges and the limitations of telehealth appointments. A few expressed concern at how their child would cope with face-to-face appointments after a period of not attending in person.

I’m worried that my daughter will become withdrawn, nervous, anxious for future appointments.

### Delays and cancellations

Delays and cancellations to inpatient and outpatient care were widely reported. Most cancelled appointments had been rebooked.

Participants described concern at anticipated and realised difficulty accessing timely and appropriate care, with stark accounts of feeling abandoned by...
healthcare services. This was most evident for community services, with speech and language therapy services most frequently cited.

Parents felt that delays impacted directly on their child’s health, including escalation of care to an emergency and slower developmental progress.

Hospital care
Hospital avoidance due to concerns about infection risk was reported but all accounts of hospital treatment were positive. Parents felt safe with infection prevention and control measures.

Caring alone, due to one parent policies, caused the greatest challenge. Participants described distress making decisions regarding care, including surgery alone, the absent parent being omitted from care and the challenge of processing information while simultaneously looking after the child.

One parent highlighted the impact of mask-wearing on bonding. Practical challenges, such as not having a parent kitchen and car-parking, were also reported. Overall, access to hospital care was reported more positively than community care.

Experiences of disease containment and their impacts
Themes relating to disease containment are outlined in figure 2, with further illustrative quotes provided in table 3.

Fear of risk to the child and cutting contact
Fears for their child’s health were expressed almost universally. Several parents described an overwhelming fear that their child was going to die.

I couldn’t shift the feeling again that we that we were going to lose him.

This led to most participants cutting all contact with others outside their household; some following the UK government’s advice to ‘shield’, others without this advice. Whether and when children were advised to shield varied. Some received instruction at the start of the pandemic, some after a couple of months and some not at all. Where shielding advice was not immediate, many participants sought information from healthcare teams and the TOFS support group. Participants identified the TOFS website and online peer support as particularly helpful.

The TOFS Facebook group is the only place where I have seen useful information about our TOFS.

Shielding experiences were varied. Some felt gratitude for the family time. For others, balancing work with children at home or the social isolation resulted in high levels of stress.
Transitional to new worries

Many parents transitioned from worrying about their child’s health to worrying about socialisation and development. They described balancing the health benefits of isolation with the risks to well-being.

Participants highlighted the burden of assessing the risks/benefits of school or childcare attendance. Good communication with the school/nursery and trust in the infection control procedures facilitated attendance. Some reported that infection control measures prevented in-person staff training, disrupting transition to school/nursery. One child was unable to access longer hours in nursery as the parent was unable to go on-site to gastrostomy-feed her child. Two parents highlighted to school staff their child’s chronic OA/TOF-related cough to differentiate it from an infectious COVID-19-related cough.

…wanted to explain that her TOF cough was normal for her…It wasn’t ideal having to try to explain at a distance at the door, but I didn’t want her new teacher to be alarmed (or other kids in the class) when she coughed.

Moving out of isolation and ongoing fear and isolation

Fear led to complete isolation for almost all participants initially, but as the pandemic progressed experiences diverged. For some, increased knowledge provided reassurance. This, coupled with increasing concern about the social and emotional effects of isolation, prompted transition to a more ‘normal life’, although within society-wide restrictions. For others, increased information and knowledge was not reassuring, resulting in continued isolation and anxiety, including continued home-learning.

Health and development benefits

Strikingly, many parents reported benefits of social isolation, highlighting reduced illness and hospitalisations, improved weight gain and improvements to general development.
Impact on parental mental health and work–life balance

Parents made direct reference to the impact that isolation as a result of disease-containment measures, difficulty accessing healthcare and anxiety about the health risk to their child had on their own mental health. A small number required professional support for anxiety or post-traumatic stress disorder.

Combining childcare with home working, managing with limited space and an increased burden of domestic tasks were stress-inducing. Financial hardship was reported. The ability to work was affected by the need to look after children, avoidance of social contact to keep their child safe and parental mental health difficulties.

DISCUSSION

The online forum allowed for timely gathering of parental insights into the impact of the COVID-19 pandemic on children with OA/TOF. The method engaged a large number of parents from a rare disease cohort, avoided face-to-face contact and minimised burden by allowing for asynchronous participation.

Access to healthcare during the pandemic has been shaped by infection prevention and control measures, limiting face-to-face contact and prioritisation of ‘essential’ services. This study highlights the significant impact these changes have had.

Telehealth was widely and rapidly adopted. No parents reported having accessed telehealth prior to the pandemic. Benefits, including access to specialist services from geographically distant locations and reduced costs and time to patients reported in this study, have long been recognised. Recent technological advances in mobile communication, software and high-speed internet have increased feasibility of telehealth and effective use in paediatric surgical conditions has been demonstrated. While many parents reported receiving good care remotely, wanting this to continue post-pandemic, some felt disconnected from their healthcare team and that care was suboptimal. These views are echoed by clinicians. Our study design prevented linking demographic data to individual responses. However, diagnostic complexity and the age of the child varied within the group. Younger children with OA/TOF tend to have more challenging health needs with postoperative morbidity greater for some. We propose satisfaction with telehealth was greater for those at a stable point in their care than those with specific concerns. Dissatisfaction was not reported in other pre-pandemic studies evaluating use of telehealth with paediatric surgical patients. This may be due to the speed with which telehealth was rolled out, with blanket, rather than targeted, use.

It is likely that use of telehealth will continue post-pandemic. Implementation of telehealth long term will be reliant on development of pathways enabling a balance of face-to-face and remote appointments with an analysis of the impact on care. Bird and colleagues have developed a comprehensive co-design framework to develop virtual clinics for the management of chronic illness in children. Use of such a framework would ensure feelings of disconnect identified in this study are minimised. Service providers must now actively engage with the rapidly evolving technology, such as use of remote stethoscopes, understand barriers and support access to technology, and develop appointment-specific telehealth guidance to optimise use of telehealth.

Parents described the impact that delays and cancellations had to their child’s health and developmental progress, with access to community care particularly problematic. A number of parents highlighted the distress caused by feelings of being abandoned by healthcare services. Redeployment had a significant impact
on services identified as ‘non-essential’. Our findings mirror those of research with other rare diseases. Continuation of ‘non-essential’ services, such as community speech and language therapy, during periods of high resource would reduce the need for escalation of care to emergency levels, promote development and support parental well-being.

The wider harms caused by society-wide ‘lockdown’ have been well summarised and are reflected in our findings; fear and anxiety, displaced non-COVID care, social isolation, stress and loss of income were all reported. Similar themes emerged from research exploring the experiences of parents caring for children with cancer. Our findings highlight the diversity of experiences, typified by divergent management of anxiety that enabled some to transition to a more normal life, while others continued to isolate. Although individual differences in risk evaluation are inevitable, clinicians should acknowledge the burden of decision-making and seek to support by providing clear communication of the best available evidence to mitigate unnecessary isolation.

Parents highlighted the challenge of obtaining information about risks to their child. Not all children were identified as ‘extremely clinically vulnerable’ and advice was not always received. Not all children were seen by disease-specialists, or were being screened for discharge. Many parents described the positive impact that isolation had on their child’s health and growth. Exposure to common viruses and other infections is usually an inaccessible part of childhood and supports development of a well-functioning immune system but can necessitate hospital treatment for vulnerable children. The long-term impact of not being exposed is unknown. However, clinicians should be aware of the potential challenge that some parents will face in supporting normal childhood activity with the knowledge that avoidance may improve health post-pandemic.

Chronic cough is common in children with OA/TOF. Interestingly, difficulty differentiating infective coughing from the child’s usual cough was not commonly reported and was not hindering school attendance. TOFS provide excellent resources for families to educate about chronic ‘TOF cough’, limiting misunderstanding and empowering parents to advocate appropriately.

Recommendations, based on parental lived experience data, for OA/TOF service providers are presented in Table 4. While this study focused solely on children with OA/TOF, it is likely that many of these recommendations would be appropriate for children with other complex healthcare needs.

Limitations
Despite efforts to facilitate wider involvement, participants were overwhelmingly female white parents of preschool children, likely reflecting those most commonly using Facebook and engaging with a support group. We acknowledge that although a wide range of experiences were described, they may not be reflective of the whole OA/TOF community.

Description of group rather than individual demographics supported anonymity but prevented subanalysis by OA/TOF type or age. Future research should identify whether such factors impact on satisfaction with telehealth and the assessment of risk.

CONCLUSION
Parents’ experiences of caring for a child with OA/TOF during the pandemic were varied. Rapid adoption of telehealth has demonstrated the enormous potential of remote healthcare delivery but requires refinement to meet the needs of the individual. Future pandemic planning should aim to retain community healthcare services to avoid escalation of care to an emergency, manage chronic and developmental concerns, and support parental well-being. Accurate and consistent, disease-specific information is highly valued by parents. Third sector organisations are ideally positioned to support this.

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Data availability statement  No data are available. Although data have been anonymised, we have not made data available to protect the identity of those involved in the research, due to the detail provided by participants.

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