Citation for published version:
Rabone, C & Wallace, V 2021, 'A thematic analysis exploring the psychological well-being of adults born with esophageal atresia', *Journal of Psychosomatic Research*, vol. 145, 110474.
https://doi.org/10.1016/j.jpsychores.2021.110474

DOI:
10.1016/j.jpsychores.2021.110474

Publication date:
2021

Document Version
Peer reviewed version

Link to publication

Publisher Rights
CC BY-NC-ND

University of Bath

Alternative formats
If you require this document in an alternative format, please contact:
openaccess@bath.ac.uk

General rights
Copyright and moral rights for the publications made accessible in the public portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognise and abide by the legal requirements associated with these rights.

Take down policy
If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Download date: 07. Nov. 2026

A Thematic Analysis Exploring the Psychological Well-Being of Adults Born with Esophageal Atresia

Caitlin Rabone, MSc[1]

Vuokko Wallace, DClinPsy[1]

[1]Department of Psychology, University of Bath, UK

Author Note

Vuokko Wallace https://orcid.org/0000-0002-9451-6598

We have no known conflict of interest to disclose.

Correspondence concerning this article should be addressed to Vuokko Wallace, Claverton Down, 10 West, University of Bath, Bath BA2 7AY, UK. Email: vw273@bath.ac.uk

Acknowledgements: We would like to thank all the parents taking part in the study, and the TOFS Charity UK for their invaluable help with participant recruitment.

Highlights

- Psychological well-being important in adults born with Esophageal Atresia (EA)
- Online survey methods can be applied to qualitative research in rare diseases
- Dysphagia, surgical scars, and medical trauma as health-related challenges in EA
- Many adults with EA expressed resilience, positively impacting on their PWB
- Multidisciplinary approach, including psychology, needed for adults born with EA

ABSTRACT

Background: Living with a rare and chronic health condition can have a significant impact on psychological well-being and mental health. There is a growing understanding that Esophageal Atresia (EA), a rare birth defect often accompanied by a Trachea-Esophageal Fistula (TEF), is a complex health condition that requires lifelong medical attention beyond paediatric care into adulthood. Given the reciprocal relationship between one's physical and psychological well-being, the aim of this study was to develop a better understanding of the mental health of adults born with EA/TEF.

Methods: An international online qualitative survey was designed and disseminated in collaboration with an EA/TEF patient charity. The qualitative data was analysed using a reflexive and inductive Thematic Analysis to explore the research question "How can being born with EA/TEF affect psychological well-being in adulthood?".

Results: A total of 92 adults born with EA/TEF completed the online survey from 11 different counties. Five themes were generated during the analysis: 'Negative Experience with Healthcare Professionals', 'The Perception of Surgical Scars', 'The Psychosocial Consequences of Dysphagia', 'The Legacy of Medical Trauma', and 'Resilience in the Face of Adversity'.

Conclusion: The results indicated that adults born with EA/TEF might face emotional challenges that can negatively affect their psychological well-being and mental health. It was also found that some adults with EA/TEF demonstrate resilience through positive reappraisal of adverse experiences. The current study suggests that a multidisciplinary approach to the care of adults born with EA/TEF is necessary and directions for future research are discussed.

Esophageal Atresia (EA) is a rare congenital malformation, occurring in approximately 2.4 infants per 10,000 live births (Roberts et al., 2016). It is characterized by a separation of the tube connecting the esophagus to the stomach, and often occurs together with a connection between the esophagus and the trachea known as Trachea-Esophageal Fistula (TEF; Martin & Crabbe, 2016). Other associated complications include a long-gap presentation and a VACTERL syndrome, in which two or more additional birth anomalies are present (e.g., vertebral defects, anal atresia, cardiac malformations, renal anomalies and limb abnormalities; Raam et al., 2010).

Due to advances in modern surgery, the survival rates for babies born with EA/TEF have increased to up to 97% in the Western world and there is a growing population of adults born with the condition (Dellenmark-Blom et al., 2015; Sistonen et al., 2011). As a result, it has become clear that many EA/TEF patients experience long-term health complications including a chronic cough, recurrent chest infections, asthma, dysphagia and gastroesophageal reflux (Roberts et al., 2016; Martin & Crabbe, 2016). EA/TEF is therefore a complex and lifelong condition requiring the attention of healthcare professionals beyond pediatric care (Martin & Crabbe, 2016).

Health psychology is interested in using interdisciplinary perspectives such as the biopsychosocial model to consider and promote physical health and wellness (Johnson & Acabchuk, 2018). One reason for this is because research suggests that there is a reciprocal relationship between the development and course of physical conditions and disease, and psychological well-being (PWB) and mental health difficulties (Luo et al., 2019; Uhlenbusch et al., 2019; The King's Fund, 2012; Rare Disease UK, 2018; Moussavi et al., 2007). Qualitative research for instance indicates that rare and chronic conditions may negatively affect PWB through physical and emotional challenges, such as a dependency on others and the subjective feeling of being different (von der Lippe et al., 2017; Claessens et al., 2005).

Previous research with adults born with EA/TEF has focused on measuring general and health related Quality of Life (QoL), and no studies have directly examined PWB in this population (Diseth & Emblem, 2017; Svoboda et al., 2018). Therefore, given the lack of research and the importance of interdisciplinary care for people with rare and chronic health conditions, the current research aimed to develop a deeper understanding of the mental health of adults born with EA/TEF using qualitative research methods. The study aimed to answer the research question: "How can being born with EA/TEF affect PWB in adulthood?"

Methods

Design

Patient and public involvement is an invaluable and essential aspect of healthcare research in the United Kingdom (Mader et al., 2018). Consequently, the current study was designed in collaboration with an international EA/TEF patient charity, and the project was supervised by a Clinical Psychologist born with EA/TEF. The study received ethical approval from the Department of Psychology at the University of Bath and was carried out in accordance with the BPS Code of Ethics and Conduct (BPS, 2018).

This study employed a cross-sectional qualitative research design and adults born with EA/TEF were recruited through an international online survey (Davies, 2016). A qualitative research strategy was considered most appropriate because the processes of "how" and "why" people may be affected by a phenomenon can be explored by describing and interpreting their lived experiences and perspectives (Willig, 2013). This approach has been used traditionally in health psychology as it can offer rich and meaningful insights into the impact of physical health conditions (Braun & Clarke, 2014; Crowe et al., 2015; Kimura et al., 2015; Moradi et al., 2014; Fawaz & Samaha, 2020).

Participants

A total of 106 participants submitted their survey responses to the study but 14 were excluded because the answers lacked descriptive detail (short or one-word answers such as "N/A"). The final sample size therefore included 92 participants, which for a qualitative survey falls within the upper mid-range (Braun et al., 2020). Survey responses came from 11 different countries, although most came from the UK. A total of 75 participants were female and 17 were male. They varied in age from 18–24-years-old to 55–64-years-old. Furthermore, 54 participants described their EA sub-type as EA/TEF, 23 as Long-gap EA/TEF, and 15 as VACTERL. See Table 1 for the participant demographics.

Table 1.
Participant demographics.

Demographics	**Number of Participants (*n* = 92)**
Age	
18-24	18
25-34	20
35-44	25
45-54	23
55-64	6
Gender	
Female	75
Male	17
Residence	
Asia	1
European Union	5
North America	12
Oceania	10
United Kingdom	63
No response	1
EA Sub-Type	
EA/TEF	54
Long-gap EA/TEF	23
VACTERL	15

Research Strategy

Using the specialized service platform Qualtrics (2019), one online survey was created jointly for two MSc projects investigating the PWB and mental health of adults born with EA/TEF. For this current study, the survey included nine, open-ended questions designed to produce rich data regarding the impact of EA/TEF on the participants' well-being (Turner III, 2010). The qualitative questions were created using the biopsychosocial model to consider how different aspects of people's lives (physical, social, and psychological) may be affected by EA/TEF and how these may interact to influence their experiences of mental health (Engel, 1977). In addition to answering the qualitative questions, the participants were invited to complete two quantitative measures for anxiety and depression. This paper reports only on the findings derived from the qualitative open-ended questions.

Data Collection

A purposive sampling strategy was employed to ensure only those individuals with personal experiences of living with EA/TEF were recruited (Etikan et al., 2017). People were eligible to take part if they were born with EA/TEF, aged 18 or over, and had access to the online survey through an electronic device. For this investigation, data collection stopped once inductive thematic saturation was achieved (Saunders et al., 2018). This ensured the sample size was appropriate and the research question could be answered in depth (Braun & Clarke, 2013). The study collected participant-generated textual data using the Qualtrics software over a period of 3 months in 2019. The final dataset included 21,941 words in total; see Table 2 for a breakdown of the words provided by the participants to each question.

The participants were recruited through a digital advertisement inviting adult members of an international EA/TEF patient charity to take part in the research project. The advertisement with a direct link to the online Qualtrics survey, was posted onto the EA/TEF patient charity's website, closed social media platforms such as private Facebook groups, and

emailed directly to members of the charity. When the participants followed the link to the survey, they were presented with an information sheet regarding the purpose and procedure of the study. The participants were then asked to give informed consent, provide demographic information such as age and gender, complete two quantitative measures, and answer the nine open-ended questions. The participants typed their responses to the survey questions without a word limit and submitted them at the end. Finally, the participants were provided with a debrief form thanking them for their involvement. Participants completed the survey anonymously without direct contact with the researchers. In accordance with the Data Protection Act, the data was held securely, and all electronic files were password protected and encrypted where appropriate (Carey, 2018).

Table 2.

The total, range and mean number of words provided by the participants for each question in the online qualitative survey (n = 92).

Question Number	Qualitative Question	Total Number of Words per Question	Range of Words per Question	Mean Number of Words per Question
1.	What does it mean to you, to be an adult survivor of EA/TEF?	3940	1 – 299	43
2.	How does living with EA/TEF affect your physical well-being and daily activities?	3753	1 – 207	41
3.	What are the emotional challenges of living with EA/TEF?	2496	1 – 121	27
4.	What are the positive emotional experiences of living with EA/TEF?	1611	1 – 209	18
5.	How does living with EA/TEF affect your self-esteem and body image?	2759	1 – 119	30
6.	How does living with EA/TEF affect your relationships with your family and friends?	2239	1 – 150	24
7.	How does living with EA/TEF affect your romantic relationships?	1495	1 – 109	16
8.	Where do you receive emotional support from in regards to living with EA/TEF?	896	1 – 59	10
9.	Is there anything else you would like to share about your personal experiences of living as an adult with EA/TEF and how these impact upon your well-being?	2752	1 – 310	30

Data Analysis

The data was analyzed using reflexive thematic analysis (TA), which is one of the most widely used methods for identifying and interpreting important patterns of shared meaning across a large qualitative dataset (Braun & Clarke, 2006; Braun & Clarke, 2019; Braun & Clarke, 2020). TA is a highly flexible research approach, allowing the data to be analyzed using the researcher's chosen epistemological framework critical realism, whereby it is assumed that language reflects meaning and that one's perception of reality is socially influenced (Braun & Clarke, 2006, 2013). The reflexive TA was conducted systematically using the six-stage process specified by Braun & Clarke (2006) and the themes were developed on a semantic level using an inductive approach (Braun & Clarke, 2013). For further details regarding the analytic process see Table 3.

To establish the trustworthiness of the research findings, steps were taken to increase credibility, transferability, dependability, and confirmability (Guba, 1981; Shenton, 2004). These steps included keeping a reflective diary, attending frequent research supervision sessions to discuss and reflect on interpretations of the data, and following the Standards for Reporting Qualitative Research guidelines (O'Brien et al., 2014).

To ensure reflexivity, the researchers acknowledged and critically evaluated the influence of their own life experiences and assumptions on the analysis (Palaganas et al., 2017). This included reflection through peer supervision on aspects of diversity (Burnham, 2013; Divac & Heaphy, 2005) and the researchers' lived experience of chronic physical health conditions.

Table 3.

Details of the reflexive Thematic Analysis conducted.

Analysis Stage	**Brief Description**	**Process**
1. Familiarisation	Immersion in the data to become familiar with the depth and breadth of the content.	The first author actively read through the data three times, recording their initial thoughts.
2. Generating Initial Codes	The production of initial codes and organization of the data into meaningful groups.	The entire dataset was highlighted and coded manually in relation to the research question using the comments feature in Microsoft Word. Similar data extracts and codes were collated, merged and refined using NVivo.
3. Searching for Themes	The synthesis and analysis of codes to generate initial themes.	The first author printed and cut out the codes on paper to sort them visually into theme-piles. Data extracts and codes relevant to the themes were collated and reviewed using NVivo. Thematic maps were developed to explore relationships within and between the themes.
4. Reviewing Themes	The refinement of candidate themes to ensure they are supported by the codes, data extracts and entire dataset.	Stage 3 of the analytic process was repeated, and some initial themes were combined, separated or discarded. The final themes, subthemes and thematic map were chosen.
5. Defining and Naming Themes	The development of clear theme definitions and names to capture their ‘story’.	Scientific literature was used to develop a detailed analysis of the themes in relation to the research question.
6. Producing the Report	Writing up the analysis to produce a detailed narrative of the data.	Vivid and compelling data extracts were chosen to evidence the themes. A final academic report was produced.

Results and Discussion

As represented in Table 4, five themes were identified in relation to the research question "How can being born with EA/TEF affect psychological well-being (PWB) in adulthood?".

Table 4.

The five themes and subthemes identified in relation to the research question.

Themes	Subthemes	Brief Description
1. Negative Experiences with Healthcare Professionals	*1.1 Lack of Understanding* *1.2 Lack of Support*	Adults born with EA/TEF can experience distress because of a lack of understanding and support from healthcare professionals.
2. The Perception of Surgical Scars	*2.1 Dissatisfaction* *2.2 Acceptance*	Adults born with EA/TEF can experience negative and positive body-image depending on how they perceive their surgical scars.
3. The Psychosocial Consequences of Dysphagia	*3.1 Anxiety with Every Meal* *3.2 Social Withdrawal*	Adults born with EA/TEF can experience anxiety and social withdrawal because of symptoms of dysphagia.
4. The Legacy of Medical Trauma	*4.1 Psychological Distress* *4.2 Parental Anxiety*	Adults born with EA/TEF and their families can experience ongoing medical events and procedures as traumatizing.
5. Resilience in the Face of Adversity	*5.1 Finding the Silver Lining* *5.2 Keeping Perspective*	Adults born with EA/TEF can experience resilience though the positive reappraisal of negative and stressful events.

Theme 1. Negative Experiences with Healthcare Professionals

People living with a chronic condition are often frequent users of healthcare services. Research has shown that the relationship these patients have with medical professionals is important for their overall health and PWB (Naylor et al., 2012; Budych et al., 2012). The current theme explored how a lack of understanding and support from healthcare practitioners can negatively impact the mental health of adults born with EA/TEF.

1.1 Lack of Understanding

The participants described a lack of knowledge, awareness and understanding about EA/TEF amongst healthcare professionals, particularly regarding the long-term nature of the condition. As a result, some expressed feeling dismissed, "angry" and "frustrated":

> [I] Really hope there will be a greater understanding of the condition […] As probably the hardest parts of living with the condition is the lack of understanding by medical professionals and being made to feel that you are making it up. (Participant 84)

1.2 Lack of Support

Due to a lack of understanding amongst the healthcare community, some participants thought they received inadequate medical and emotional support. One participant discussed for instance how having EA/TEF "means that I am alone medically to navigate my health and well-being" (Participant 54). A few participants reported the transition from pediatric to adult care as particularly "stressful".

> I found transitioning to the adult health system very difficult. My health concerns often get written off by GPS who do not understand or think I look too healthy to be so unhealthy. (Participant 38)

Consequently, some of the participants expressed feelings of "fear" and anxiety about their future healthcare: "I also have some concerns as I age as I have had difficulties finding specialists who understand our complications as adults and only want to look at the "normal" things" (Participant 90).

In summary, the participants reported a lack of understanding and support from healthcare professionals and feelings of anger, anxiety, and stress as a result. This is a common experience shared with other people living with rare and chronic conditions (von der Lippe et al., 2017; Zurynski & Elliott, 2013). In a study with 1,350 rare disease patients for example, 88% of respondents agreed that poor professional awareness of their condition can negatively impact upon mental health (Rare Disease UK, 2018).

Theme 2. The Perception of Surgical Scars

Following the initial surgical repair and further medical procedures across the lifespan, many adults born with EA/TEF have visible surgical scars on their thorax, abdomen and dorsal area (Martin & Crabbe, 2016). As represented by the current theme, the way in which these adults perceive their surgical scars can have implications for their PWB.

2.1 Dissatisfaction

Many participants expressed that they disliked their surgical scars and were dissatisfied with their physical appearance. One participant explained: "as I've aged and put

on some weight, my abdomen scars look worse […] so I don't really like how I look" (Participant 24), and another reported "I had my scars realigned and it made them worse, I've always hated my stomach, and hate explaining my scars" (Participant 42). Consequently, some participants reported feeling self-conscious of their body: "I have issues with the scaring mainly on my stomach area, and has caused being self-conscious getting undressed in front of partners especially" (Participant 51).

2.2 Acceptance

In contrast to the previous subtheme, there were some participants who perceived their surgical scars as a positive part of their body: "my scars are my badge. They are me. I had a chance to 'fix' them but when it came to it I couldn't do it. They are part of me and who I am" (Participant 75). Additionally, several participants described their scars as a symbol of "strength", and reported wearing them with confidence and pride.

> I am very proud of my scar […]. I feel like it connects me to my past and to other TOFs. It's a sign that I survived something that not everyone does - it's a visual reminder that life can be scary but that's ok and I'll get through it […] I feel like my scar is a positive part of my body, so when I went to prom I had a prom dress that specifically showed it off. (Participant 100)

In summary, the participants' perceptions of their surgical scars seemed to have an impact on their self-confidence and body image. Similarly, a study exploring QoL in adults born with EA/TEF found that some participants reported surgical scars as a negative consequence of the condition (Deurloo et al., 2005). Previous research has found that living with visible differences in physical appearance such as scars can result in negative self-

perceptions and evaluations (Ngaage & Agius, 2018; Van Loey, 2020), which in turn may be associated with poor mental health outcomes (Williamson et al., 2018). A positive body image, on the other hand, is associated with indicators of good mental health such as optimism, self-compassion, and life-satisfaction (Swani et al., 2018).

Theme 3. The Psychosocial Consequences of Dysphagia

Dysphagia is one of the commonly reported chronic health problems for adults born with EA/TEF. It can be described as a difficulty swallowing or moving food from the mouth to the stomach (Ijsselstijn et al., 2013; San et al., 2018). The symptoms include persistent episodes of coughing or choking during meals and the involuntary regurgitation of food (NHS, 2018). As captured by the current theme, dysphagia can negatively affect the PWB of adults born with EA/TEF.

3.1 Anxiety with Every Meal

The participants were often concerned about choking on food and this was described as a distressing event: “on a few occasions I have nearly had to go to hospital because something became lodged in my oesophagus, which I found very stressful” (Participant 100). Consequently, some participants described living with feelings of anxiety as they perceived every meal as a potential risk for choking: “psychologically, I think it can be daunting as you don’t know when food will get stuck and it can sometimes be a cause of worry” (Participant 86), and “I constantly worry about eating if I don't have a drink nearby. Although I don't have as many symptoms as others might, it does cause me anxiety” (Participant 15).

3.2 Social Withdrawal

When eating in public many participants were self-conscious of dysphagia and they described coughing or choking on food as "embarrassing". One participant expressed for example, "choking and gurgling in public is horrifying for me and everyone watching" (Participant 45). Additionally, many participants reported feeling anxious about eating in front of others and avoiding social events such as going out for meals with their friends and family.

> Now I am having to think more of what I will eat when I'm in public and with friends in case it gets stuck or causes problems. If this happens it can ruin my night and I would need to leave. (Participant 21)

> I often avoid going out for meals or eating in crowded places due to worrying about how long it takes me to eat and having any issues in public. I also often feel as though friends and family may judge how slow I am at eating and I often become very anxious when eating in front of people. (Participant 81)

In summary, many adults born with EA/TEF described feelings of anxiety because of their symptoms of dysphagia, leading to the avoidance of social activities. This has also been reflected in qualitative studies that have examined the lived experience of dysphagia in stroke survivors and patients with head and neck cancer (Moloney & Walshe, 2018; Ganzer et al., 2015). Furthermore, previous research has found a positive association between impaired swallowing function and symptoms of anxiety (Verdonschot et al., 2013; Verdonschot et al., 2017), and that avoidance of social activities can lead to increased feelings of isolation and depression (McQuestion et al., 2011).

Theme 4. The Legacy of Medical Trauma

In addition to the initial surgical repair, many young people and adults born with EA/TEF will need regular health interventions, including hospitalizations and invasive medical procedures (Martin & Crabbe, 2016; Leibovitch et al., 2018). As captured by the current theme, some patients and their families can experience such medical events as distressing and traumatic.

4.1 Psychological Distress

Some participants recalled medical experiences from their childhood that were characterized by feelings of fear, humiliation, and helplessness, and these were described as having an impact on their mental health. Additionally, some participants felt guilty about the distress experienced by their family.

> The emotional effect [of EA/TEF] has been enormous. The trauma relates more to medical events from my childhood […] As a child I had some extremely frightening and humiliating experiences that have left me quite traumatised and this has led to the psychological and social difficulties I have now. Anything that has happened to me medically since I was 18 pales into insignificance compared to the events in my childhood. (Participant 25)

> For a long time I felt guilty for the way I was born, and for the hurt my family went through. I felt like it was my fault and that I didn't meet the expectations of the first grandchild/child in the family. (Participant 59)

4.2 Parental Anxiety

Some participants described their parents as traumatized by the paediatric medical events associated with EA/TEF. One participant explained, “My mum doesn't talk about it, it's like she's blocked it, my dad talks with me about it but mum I feel she went through a very hard time having a sick child” (Participant 71). Another participant reported, “I feel I was able to deal with my problems well but got more upset seeing my mum getting upset when I could not swallow or eat properly, I think she needed more support than I did” (Participant 29). Additionally, some adults born with EA/TEF described their parents as being overprotective or hypervigilant and anxious about their health, “I do believe she [mother] loved me very much but [she] was always anxious I would die.” (Participant 27)

> I think my parents might be a bit more protective of me than they would otherwise be - I'm an only child and they experienced the worst of my TOF because of my surgery when I was a baby. Mum sometimes gets worried about me eating certain foods that I know I'm fine with. (Participant 100)

In summary, some of the participants described themselves and their parents as traumatized by medical events associated with EA/TEF and as a result they experienced feelings of distress, anxiety, and guilt. Similarly, previous research has shown that the diagnosis and treatment of a life-threatening or chronic pediatric condition can cause long-term psychological distress, depression and anxiety in patients and their families (Pinquart, 2018; Ari et al., 2018; Woolf et al., 2016; Bronner et al., 2010). Importantly, parental mental health is linked to less effective parenting practices, insecure child attachment and increased distress in their children (Skreden et al., 2010; Ari et al., 2018).

Theme 5. Resilience in the Face of Adversity

Due to the nature of a chronic condition, adults born with EA/TEF are likely to experience ongoing challenges (Gumuchian et al., 2018). Consequently, some might struggle to cope adaptively with the stress of living with EA/TEF, resulting in poor PWB (Meng & D'Arcy, 2016). However, many participants showed resilience in the face of adversity. Specifically, some participants used positive reappraisal as an adaptive coping response to re-evaluate and find positive meaning from their stressful experiences (Troy & Mauss, 2011; Garland et al., 2011; Nowlan et al., 2015).

5.1 Finding the Silver Lining

Upon reflection, many participants were able to find a benefit or 'silver lining' from their experiences. The participants explained that through living with a chronic health condition, they had gained personal strength and the ability to overcome difficulties: "I feel it has made me a stronger person. Despite a tough start in life I have achieved many things, travelled the world and am able to handle set backs well" (Participant 60). Furthermore, the participants described an increased understanding and empathy for others, leading some into a career in health or social care. One participant explained, "I'm resilient. I'm caring. I have great empathy for others. I seem to have intuition and can tell when others are not ok. Thus I work as a welfare worker which is very rewarding" (Participant 68). Some participants also reported appreciating their health more and looking after their physical well-being: "I know I need to look after myself if I want to stay healthy so I take responsibility for my wellbeing" (Participant 29). When such 'silver linings' were identified, the participants reported feeling "proud" for overcoming their difficulties and EA/TEF was viewed as a positive and essential part of their identity. One participant expressed, "EA/TEF is part of me, there is no separation - it is an integral part in making me who I am today" (Participant 31).

5.2 Keeping Perspective

The participants described being able to positively re-evaluate their stressful experiences by keeping them in perspective. In contrast to the first theme 'Negative Experiences with Healthcare Professionals', many participants recognized that without the help of modern medicine and surgical interventions, they would not be alive:

> I watched the recent surgeons programme and things like that do help me to pause and think yes, I am very lucky to benefit from science, research, the NHS, surgeons etc and feel grateful to have made it this far. (Participant 42)

Some participants also compared themselves to other adults born with EA/TEF and accepted that their physical health problems could be worse.

> I feel grateful and lucky to be here. I have a sense of pride in my condition but I am aware that I don't have as many issues as other TOF survivors and that's where my feeling of gratefulness comes from. (Participant 95)

By keeping perspective, the participants expressed feelings of happiness, luck, and appreciation for their life, as well as gratitude for medical professionals. When asked what being an adult with EA/TEF means to them, one participant shared the following:

> Extremely lucky, I feel as if I've had someone watch over me from the moment I was born! I feel inspired to do the best for myself as I've been given a chance to live and survive with my condition. (Participant 40)

In summary, the adults born with EA/TEF demonstrated resilience by finding positive meaning and growth from their stressful experiences. Previous research has shown that higher levels of resilience might be associated with improved mental health outcomes (Hu et al., 2015; McGowan et al., 2018; Cal et al., 2015; Färber & Rosendahl, 2018).

Conclusions

To our knowledge, this is the first study to investigate the PWB of adults born with EA/TEF. The aim of the study was to develop a better understanding of the mental health of this population, and this was achieved through a qualitative online survey design. The data was analyzed using a reflexive TA, on an inductive and semantic level, and five themes were generated during the analysis. The participants reported feelings of anxiety and psychological distress because of their relationship with healthcare professionals, symptoms of dysphagia and traumatic paediatric medical experiences. Additionally, their perceptions of surgical scars appeared to have an impact on their body image and self-confidence. These findings suggest that adults born with EA/TEF might face emotional challenges that can negatively affect their PWB and mental health (Rare Disease UK, 2018; Williamson et al., 2018; Verdonschot et al., 2017; Pinquart, 2018). Some participants also reported finding opportunities for personal growth and positive meaning from their experiences of adversity, which indicates that some adults born with EA/TEF are resilient and likely to have good PWB (Hu et al., 2015; McGowan et al., 2018).

Strengths and Limitations

The findings of the current study provide new, meaningful, and rich insights into the PWB of adults born with EA/TEF. The use of an online survey is also a relatively novel and innovative method in qualitative research (Braun et al., 2020), providing a variety of advantages and benefits for this study. These included the ability to recruit a large, diverse,

and international sample which is often a challenge when working with a rare disease population (Griggs et al., 2009). The online survey design was also ideally suited to the sensitive nature of the research question, and it allowed the researchers to give voice to people that might not have been able to participate "face-to-face".

However, the research design had some limitations. Firstly, the sample may not be representative of the whole population of adults born with EA/TEF because the participants were self-selected volunteers, and the majority were female (Davies, 2016). Additionally, the fixed nature of the online survey meant the qualitative questions could not evolve or be revised once data collection had commenced. It could also be argued that the large number of qualitative questions increased the risk of participant burden or disengagement; however, the flexibility offered to the participants through the online survey design (e.g., when they completed the study and for how long) may have mitigated this (Braun et al., 2020).

Implications

The results of the study suggest that a multidisciplinary approach to the care of adults born with EA/TEF might be necessary. Specifically, medical professionals such as GPs may need to consider the mental health needs of adult patients born with EA/TEF and help them to receive appropriate psychological support. Evidence-based psychological interventions could be offered to adults born with EA/TEF to 1) help them manage and reduce anxiety surrounding symptoms of dysphagia, 2) promote a positive body image and the acceptance of surgical scars, and 3) teach adaptive coping strategies in response to stressful experiences such as traumatizing medical events. The benefit and design or adaptation of psychological interventions for adults born with EA/TEF is an important area for future research.

References

Ari, A. B., Peri, T., Margalit, D., Galili-Weisstub, E., Udassin, R., & Benarroch, F. (2018). Surgical procedures and pediatric medical traumatic stress (PMTS) syndrome: Assessment and future directions. *Journal of pediatric surgery*, *53*(8), 1526-1531. https://doi.org/10.1016/j.jpedsurg.2017.10.043

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. Qualitative research in psychology, 3(2), 77-101.

Braun, V., & Clarke, V. (2013). *Successful qualitative research: A practical guide for beginners*. Sage.

Braun, V., & Clarke, V. (2014). What can "thematic analysis" offer health and wellbeing researchers?. *International journal of qualitative studies on health and well-being*, *9*. https://doi.org/10.3402/qhw.v9.26152

Braun, V., & Clarke, V. (2019). Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health*, *11*(4), 589-597. https://doi.org/10.1080/2159676X.2019.1628806

Braun, V., & Clarke, V. (2020). Can I use TA? Should I use TA? Should I not use TA? Comparing reflexive thematic analysis and other pattern- based qualitative analytic approaches. *Counselling and Psychotherapy Research*. https://doi.org/10.1002/capr.12360

Braun, V., Clarke, V., Boulton, E., Davey, L., & McEvoy, C. (2020). The online survey as a qualitative research tool. *International Journal of Social Research Methodology*, 1-14. https://doi.org/10.1080/13645579.2020.1805550

British Psychological Society. (2018). Code of ethics and conduct. BPS.

Bronner, M. B., Peek, N., Knoester, H., Bos, A. P., Last, B. F., & Grootenhuis, M. A. (2010).

Course and predictors of posttraumatic stress disorder in parents after pediatric intensive care treatment of their child. *Journal of pediatric psychology*, *35*(9), 966-974. https://doi.org/10.1093/jpepsy/jsq004

Budych, K., Helms, T. M., & Schultz, C. (2012). How do patients with rare diseases experience the medical encounter? Exploring role behavior and its impact on patient–physician interaction. *Health policy*, *105*(2-3), 154-164. https://doi.org/10.1016/j.healthpol.2012.02.018

Burnham, J. (2013). Developments in Social GGRRAAACCEEESSS: visible-invisible, voiced-unvoiced. In I. Krause (Ed.), Cultural Reflexivity. London: Karnac.

Cal, S. F., de Sá, L. R., Glustak, M. E., & Santiago, M. B. (2015). Resilience in chronic diseases: A systematic review. *Cogent Psychology*, *2*(1), 1024928. https://doi.org/10.1080/23311908.2015.1024928

Carey, P. (2018). *Data protection: a practical guide to UK and EU law*. Oxford University Press, Inc..

Claessens, P., Moons, P., de Casterlé, B. D., Cannaerts, N., Budts, W., & Gewillig, M. (2005). What does it mean to live with a congenital heart disease? A qualitative study on the lived experiences of adult patients. *European Journal of Cardiovascular Nursing*, *4*(1), 3-10. https://doi.org/10.1016/j.ejcnurse.2004.12.003

Crowe, M., Inder, M., & Porter, R. (2015). Conducting qualitative research in mental health: Thematic and content analyses. *Australian & New Zealand Journal of Psychiatry*, *49*(7), 616-623. https://doi.org/10.1177/0004867415582053

Davies, W. (2016). Insights into rare diseases from social media surveys. *Orphanet journal of rare diseases*, *11*(1), 1-5. https://doi.org/10.1186/s13023-016-0532-x

Dellenmark-Blom, M., Chaplin, J. E., Gatzinsky, V., Jönsson, L., & Abrahamson, K. (2015).

Health-related quality of life among children, young people and adults with esophageal atresia: a review of the literature and recommendations for future research. *Quality of Life Research*, *24*(10), 2433-2445. https://doi.org/10.1007/s11136-015-0975-x

Deurloo, J. A., Ekkelkamp, S., Hartman, E. E., Sprangers, M. A., & Aronson, D. C. (2005). Quality of life in adult survivors of correction of esophageal atresia. *Archives of surgery*, *140*(10), 976-980. https://doi.org/10.1001/archsurg.140.10.976

Diseth, T. H., & Emblem, R. (2017, October). Long-term psychosocial consequences of surgical congenital malformations. In *Seminars in pediatric surgery* (Vol. 26, No. 5, pp. 286-294). WB Saunders. https://doi.org/10.1053/j.sempedsurg.2017.09.009

Divac, A., & Heaphy, G. (2005). Space for GRRAACCES: training for cultural competence in supervision. Journal of Family Therapy, 27: 280–284. https://doi.org/10.1111/j.1467-6427.2005.00318.x

Etikan, I., Musa, S. A., & Alkassim, R. S. (2016). Comparison of convenience sampling and purposive sampling. *American journal of theoretical and applied statistics*, *5*(1), 1-4. https://doi.org/10.11648/j.ajtas.20160501.11

Engel, G. L. (1977). The need for a new medical model: a challenge for biomedicine. *Science*, *196*(4286), 129-136. https://doi.org/10.1126/science.847460

Färber, F., & Rosendahl, J. (2018). The association between resilience and mental health in the somatically ill: a systematic review and meta-analysis. *Deutsches Ärzteblatt International*, *115*(38), 621. https://doi.org/10.3238/arztebl.2018.0621

Fawaz, M., & Samaha, A. (2020). <? covid19?> The psychosocial effects of being quarantined following exposure to COVID-19: A qualitative study of Lebanese health care workers. *International Journal of Social Psychiatry*, *66*(6), 560-565. https://doi.org/10.1177%2F0020764020932202

Ganzer, H., Touger-Decker, R., Byham-Gray, L., Murphy, B. A., & Epstein, J. B. (2015). The eating experience after treatment for head and neck cancer: a review of the literature. *Oral oncology*, *51*(7), 634-642. https://doi.org/10.1016/j.oraloncology.2015.04.014

Garland, E. L., Gaylord, S. A., & Fredrickson, B. L. (2011). Positive reappraisal mediates the stress-reductive effects of mindfulness: An upward spiral process. *Mindfulness*, *2*(1), 59-67. https://doi.org/10.1007/s12671-011-0043-8

Griggs, R. C., Batshaw, M., Dunkle, M., Gopal-Srivastava, R., Kaye, E., Krischer, J., ... & Merkel, P. A. (2009). Clinical research for rare disease: opportunities, challenges, and solutions. *Molecular genetics and metabolism*, *96*(1), 20-26. https://doi.org/10.1016/j.ymgme.2008.10.003

Guba, E. G. (1981). Criteria for assessing the trustworthiness of naturalistic inquiries. *Ectj*, *29*(2), 75-91. https://doi.org/10.1007/BF02766777

Gumuchian, S. T., Peláez, S., Delisle, V. C., Carrier, M. E., Jewett, L. R., El-Baalbaki, G., ... & Thombs, B. D. (2018). Understanding coping strategies among people living with scleroderma: a focus group study. *Disability and rehabilitation*, *40*(25), 3012-3021. https://doi.org/10.1080/09638288.2017.1365954

Hu, T., Zhang, D., & Wang, J. (2015). A meta-analysis of the trait resilience and mental health. *Personality and Individual Differences*, *76*, 18-27. https://doi.org/10.1016/j.paid.2014.11.039

Ijsselstijn, H., van Beelen, N. W. G., & Wijnen, R. M. H. (2013). Esophageal atresia: long-term morbidities in adolescence and adulthood. *Diseases of the Esophagus*, *26*(4), 417-421. https://doi.org/10.1111/dote.12059

Johnson, B. T., & Acabchuk, R. L. (2018). What are the keys to a longer, happier life?

Answers from five decades of health psychology research. *Social Science & Medicine, 196,* 218-226. https://doi.org/10.1016/j.socscimed.2017.11.001

Kimura, N. R., Maffioletti, V. L., Santos, R. L., Baptista, M. A. T., & Dourado, M. C. (2015). Psychosocial impact of early onset dementia among caregivers. *Trends in psychiatry and psychotherapy*, *37*(4), 213-219. https://doi.org/10.1590/2237-6089-2015-0038

Leibovitch, L., Zohar, I., Maayan-Mazger, A., Mazkereth, R., Strauss, T., & Bilik, R. (2018). Infants Born with Esophageal Atresia with or without Tracheo-Esophageal Fistula: Short-and Long-Term Outcomes. *The Israel Medical Association journal: IMAJ*, *20*(3), 161-166.

Luo, M. S., Chui, E. W. T., & Li, L. W. (2019). The Longitudinal Associations between Physical Health and Mental Health among Older Adults. *Aging & mental health*, 1-9. https://doi.org/10.1080/13607863.2019.1655706

Mader, L. B., Harris, T., Kläger, S., Wilkinson, I. B., & Hiemstra, T. F. (2018). Inverting the patient involvement paradigm: defining patient led research. *Research involvement and engagement*, *4*(1), 1-7. https://doi.org/10.1186/s40900-018-0104-4

Martin, V., & Crabbe, D. (2016). *The TEF Book: Oesophageal atresia (OA), trachea-oesophageal fistula (TEF) and VACTERL from infancy to adulthood.* TOFS.

McGowan, J. A., Brown, J., Lampe, F. C., Lipman, M., Smith, C., & Rodger, A. (2018). Resilience and physical and mental well-being in adults with and without HIV. *AIDS and Behavior*, *22*(5), 1688-1698. https://doi.org/10.1007/s10461-017-1980-6

McQuestion, M., Fitch, M., & Howell, D. (2011). The changed meaning of food: Physical, social and emotional loss for patients having received radiation treatment for head and neck cancer. *European Journal of Oncology Nursing*, *15*(2), 145-151. https://doi.org/10.1016/j.ejon.2010.07.006

Meng, X., & D'Arcy, C. (2016). Coping strategies and distress reduction in psychological

well-being? A structural equation modelling analysis using a national population sample. *Epidemiology and psychiatric sciences*, *25*(4), 370-383. https://doi.org/10.1017/S2045796015000505

Moloney, J., & Walshe, M. (2018). "I had no idea what a complicated business eating is…": a qualitative study of the impact of dysphagia during stroke recovery. *Disability and rehabilitation*, *40*(13), 1524-1531. https://doi.org/10.1080/09638288.2017.1300948

Moradi, M., Parker, M., Sneddon, A., Lopez, V., & Ellwood, D. (2014). Impact of endometriosis on women's lives: a qualitative study. *BMC women's health*, *14*(1), 1-12. https://doi.org/10.1186/1472-6874-14-123

Moussavi, S., Chatterji, S., Verdes, E., Tandon, A., Patel, V., & Ustun, B. (2007). Depression, chronic diseases, and decrements in health: results from the World Health Surveys. *The Lancet*, *370*(9590), 851-858. https://doi.org/10.1016/S0140-6736(07)61415-9

National Health Service (NHS). (2018). Dysphagia (swallowing problems). https://www.nhs.uk/conditions/swallowing-problems-dysphagia/#

Ngaage, M., & Agius, M. (2018). The psychology of scars: A mini-review. *Psychiatr. Danub*, *30*, 633-638.

Naylor, C., Parsonage, M., McDaid, D., Knapp, M., Fossey, M., & Galea, A. (2012). Long-term conditions and mental health: the cost of co-morbidities.

Nowlan, J. S., Wuthrich, V. M., & Rapee, R. M. (2015). Increasing older adults' use of positive reappraisal coping for chronic physical illness. *Clinical Gerontologist*, *38*(5), 428-437. https://doi.org/10.1080/07317115.2015.1067274

O'Brien, B. C., Harris, I. B., Beckman, T. J., Reed, D. A., & Cook, D. A. (2014). Standards for reporting qualitative research: a synthesis of recommendations. *Academic Medicine*, *89*(9), 1245-1251. https://doi.org/10.1097/ACM.0000000000000388

Palaganas, E. C., Sanchez, M. C., Molintas, M., Visitacion, P., & Caricativo, R. D. (2017). Reflexivity in qualitative research: A journey of learning. The Qualitative Report, 22(2), 426-438.

Pinquart, M. (2018). Posttraumatic stress symptoms and disorders in children and adolescents with chronic physical illnesses: A meta-analysis. *Journal of Child & Adolescent Trauma*, 1-10. https://doi.org/10.1007/s40653-018-0222-z

Qualtrics. (2018). Provo, UT, USA: Qualtrics. https:///www.qualtrics.com

Raam, M. S., Pineda-Alvarez, D. E., Hadley, D. W., & Solomon, B. D. (2011). Long-term outcomes of adults with features of VACTERL association. *European journal of medical genetics*, *54*(1), 34-41. https://doi.org/10.1016/j.ejmg.2010.09.007

Rare Disease UK. (2018). *Living with a Rare Condition: The Effect on Mental Health [PDF]*. Rare Disease UK. https://www.raredisease.org.uk/our-work/living-with-a-rare-condition-the-effect-on-mental-health-2018/

Roberts, K., Karpelowsky, J., Fitzgerald, D. A., & Soundappan, S. S. (2016). Outcomes of oesophageal atresia and tracheo- oesophageal fistula repair. *Journal of paediatrics and child health*, *52*(7), 694-698. https://doi.org/10.1111/jpc.13211

San, S. W., Ahmad, A., & Mahadevan, B. (2018). The Swallowing Ability and Psychosocial Domains of Patients with Dysphagia among Head and Neck Cancer Population. *Jurnal Sains Kesihatan Malaysia (Malaysian Journal of Health Sciences)*, *17*(1). https://doi.org/10.17576/JSKM-2019-1701-01

Saunders, B., Sim, J., Kingstone, T., Baker, S., Waterfield, J., Bartlam, B., ... & Jinks, C. (2018). Saturation in qualitative research: exploring its conceptualization and operationalization. *Quality & quantity*, *52*(4), 1893-1907. https://doi.org10.1007/s11135-017-0574-8

Shenton, A. K. (2004). Strategies for ensuring trustworthiness in qualitative research

projects. *Education for information*, *22*(2), 63-75. https://doi.org/10.3233/EFI-2004-22201

Sistonen, S. J., Pakarinen, M. P., & Rintala, R. J. (2011). Long-term results of esophageal atresia: Helsinki experience and review of literature. *Pediatric surgery international*, *27*(11), 1141. https://doi.org/10.1007/s00383-011-2980-7

Skreden, M., Skari, H., Malt, U. F., Haugen, G., Pripp, A. H., Faugli, A., & Emblem, R. (2010). Long- term parental psychological distress among parents of children with a malformation—A prospective longitudinal study. *American Journal of Medical Genetics Part A*, *152*(9), 2193-2202. https://doi.org/10.1002/ajmg.a.33605

Svoboda, E., Fruithof, J., Widenmann-Grolig, A., Slater, G., Armand, F., Warner, B., ... & Hannon, E. (2018). A patient led, international study of long term outcomes of esophageal atresia: EAT 1. *Journal of pediatric surgery*, *53*(4), 610-615. https://doi.org/10.1016/j.jpedsurg.2017.05.033

Swami, V., Weis, L., Barron, D., & Furnham, A. (2018). Positive body image is positively associated with hedonic (emotional) and eudaimonic (psychological and social) well-being in British adults. *The Journal of social psychology*, *158*(5), 541-552. https://doi.org/10.1080/00224545.2017.1392278

The King's Fund. (2012). *Long-Term Conditions and Mental Health: The Cost of Comorbidities [PDF]*. The King's Fund. https://www.kingsfund.org.uk/publications/long-term-conditions-and-mental-health

Troy, A. S., & Mauss, I. B. (2011). Resilience in the face of stress: Emotion regulation as a protective factor. *Resilience and mental health: Challenges across the lifespan*, *1*(2), 30-44. https://doi.org/10.1007/s12671-011-0043-8

Turner III, D. W. (2010). Qualitative interview design: A practical guide for novice

investigators. *The qualitative report*, *15*(3), 754-760.

Uhlenbusch, N., Löwe, B., Härter, M., Schramm, C., Weiler-Normann, C., & Depping, M. K. (2019). Depression and anxiety in patients with different rare chronic diseases: A cross-sectional study. *PloS one*, *14*(2), e0211343. https://doi.org/10.1371/journal.pone.0211343

Van Loey, N. E. (2020). Psychological Impact of Living with Scars Following Burn Injury. In *Textbook on Scar Management* (pp. 429-434). Springer, Cham.

Verdonschot, R. J., Baijens, L. W., Serroyen, J. L., Leue, C., & Kremer, B. (2013). Symptoms of anxiety and depression assessed with the Hospital Anxiety and Depression Scale in patients with oropharyngeal dysphagia. *Journal of psychosomatic research*, *75*(5), 451-455. https://doi.org/10.1016/j.jpsychores.2013.08.021

Verdonschot, R. J., Baijens, L. W., Vanbelle, S., van de Kolk, I., Kremer, B., & Leue, C. (2017). Affective symptoms in patients with oropharyngeal dysphagia: a systematic review. *Journal of psychosomatic research*, *97*, 102-110. https://doi.org/10.1016/j.jpsychores.2017.04.006

von der Lippe, C., Diesen, P. S., & Feragen, K. B. (2017). Living with a rare disorder: a systematic review of the qualitative literature. *Molecular genetics & genomic medicine*, *5*(6), 758-773. https://doi.org/10.1002/mgg3.315

Willig, C. (2013). *Introducing qualitative research in psychology*. McGraw-hill education (UK).

Woolf, C., Muscara, F., Anderson, V. A., & McCarthy, M. C. (2016). Early traumatic stress responses in parents following a serious illness in their child: A systematic review. *Journal of clinical psychology in medical settings*, *23*(1), 53-66. https://doi.org/10.1007/s10880-015-9430-y

Williamson, H., Antonelli, P., Bringsén, Å., Davies, G., Dèttore, D., Harcourt, D., ... &

Yalçın, A. D. (2018). Supporting patients with body dissatisfaction: A survey of the experiences and training needs of European multi-disciplinary healthcare professionals. *NursingPlus Open*, *4*, 8-13. https://doi.org/10.1016/j.npls.2018.05.001

Zurynski, Y. A., & Elliott, E. J. (2013). Challenges of transition to adult health services for patients with rare diseases. *Med J Aust*, *198*(11), 575-576. https://.org/doi:10.5694/mja13.10424