

RESEARCH ARTICLE

Anxiety and post-traumatic stress disorder in parents of children born with esophageal atresia

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ABSTRACT

Neonates with esophageal atresia (EA) require admission to the neonatal intensive care unit and undergo surgery early after birth. These parents encounter potential stressors that can contribute to mental health manifestations including symptoms of post-traumatic stress disorder (PTSD), anxiety and depression. The primary objective of this study was to examine the occurrence of PTSD symptoms and anxiety among parents of EA children.

We conducted an ambi-directional cohort study that included parents of EA children followed-up in our unit, between 2021 and 2023. Two separate questionnaires were completed by the parents during the child's visit consisting of socio-demographic details, the Perinatal post-traumatic stress disorder questionnaire (PPQ), and the State-Trait Anxiety Inventory (STAI).

During the study period, 20 parents completed 28 questionnaires. The mean PPQ score was 3.72, ranging from 0 to 12. Six parents had a PPQ score of 6 or higher: 2 with a score of 6 and one each with a score of 9, 10, 11 and 12. The mean State STAI was 46 ± 7 , 14% of which had a score of >55 indicating high anxiety levels at the time of visit. Similarly, the mean Trait STAI was 46 ± 5 , with only one parent (4%) scoring above 55.

It is important to identify potential symptoms of PTSD and anxiety in parents of EA children early on, allowing for timely referral for counselling and treatment. This approach benefits both the parental mental well-being and the overall adjustment and coping of the entire family.

HIGHLIGHTS BOX

What is known? EA is a chronic disease that can impact the psychological well-being of children born with this disease. EA parents are known to develop PTSD and anxiety. **What does article add to our knowledge?** Family-centered care of children born with EA is becoming more important. We need to include mental health awareness of parents in the long-term follow-up of EA children. **How does this study impact our current management guidelines?** We need to recognize and address the effects of EA on the parents of these children. Emotional well-being and follow-up should be included in the follow-up protocol of EA patients with inclusion of the appropriate members of the multidisciplinary team.

KEY WORDS

Parental mental well-being; psycho-social follow-up; esophageal atresia.

INTRODUCTION

Esophageal atresia (EA) is a congenital gastro-intestinal abnormality that can be detected during prenatal screening or shortly after birth (1). Neonates with EA typically require admission to the neonatal intensive care unit (NICU) and undergo surgery within the first few days of life. Parents of neonates diagnosed with EA, similar to those whose neonates

necessitate early NICU admission, encounter diverse potential stressors that can contribute to subsequent mental health manifestations (2). Furthermore, the care required for a chronically ill child, such as those born with EA, has been demonstrated to demand specialized attention from parents potentially elevating their susceptibility to the development of mental health symptoms (1). Despite extensive literature on the surgical outcomes of EA, little is known about the psychological impact on the parents of neonates born with EA (1, 3).

Mental health symptoms that are known to develop in these parents include parental anxiety, depression and/or post-traumatic stress disorder (PTSD) (1). Validated questionnaires have been used in previous studies identifying PTSD and anxiety risks for parents of children born with EA and those two tools have been chosen for this specific study (1). PTSD is a condition that may occur after experiencing a traumatic event, encompassing four characteristic symptom clusters: intrusion, avoidance, negative mood alterations, and changes in arousal and reactivity (3, 4). The perinatal post-traumatic stress disorder questionnaire (PPQ) is a validated instrument specifically designed for parents of high-risk neonates (1, 5). Originally developed to explore the relationship between specific perinatal stressors (such as gestational age and postnatal complications) and the development of PTSD in mothers, the PPQ has been adapted for the use in various neonatal conditions, including EA (1).

Higher levels of both anxiety and depression have been reported in parents of children with chronic diseases (6). Anxiety is defined as feelings of tension and worried thoughts that can lead to physical symptoms such as sweating, increase in blood pressure and palpitations (7). It is not the same as fear but the two are often used interchangeably (7). Parental stress and anxiety have been shown to lead to maladaptive parenting practices which can in turn lead to and predict development of mental health disorders in the children themselves (6). Studies of families with children born with EA have found that child-related factors such as the age of the child and the severity of the illness is directly related to the mental health of the parents (6). The State-Trait Anxiety Inventory (STAI) has been used for parents of children born with EA (1). It is a well-established measurement tool that can be used to evaluate the presence of

both transient anxieties caused by a specific situation (State anxiety) and persistent anxiety experienced continuously (Trait anxiety) (8).

The primary objective of this study was to examine the occurrence of PTSD symptoms and anxiety among parents whose children were born with EA in our health care facility.

MATERIALS AND METHODS

An ambi-directional cohort study was conducted at an academic pediatric surgical department. Parents of children born with EA, that were followed-up in our unit, between 2021 and 2023, were invited to participate. Participation was voluntary and parents provided written informed consent. Parents were recruited on a consecutive, convenience basis as their child presented to the pediatric surgical clinic for follow-up or to our unit for hospitalization.

Questionnaires were completed by the parents themselves during their child's follow-up visits. Two separate questionnaires were completed: the "First visit parental psycho-social questionnaire" and the "Follow-up visits parental psycho-social questionnaire" which were scheduled to be completed at least 6 months apart.

First visit parental psycho-social questionnaire

This questionnaire consisted of 3 different sections and was only completed once by the parents, during the child's first visit that occurred during the study period. The first section enquired about the socio-demographic information and personal challenges of the parents. Additionally, the child's age and the reason for the visit (admission to the surgical ward vs. regular out-patient department visit (OPD)) was recorded. The second part was the "Perinatal post-traumatic stress disorder questionnaire" (PPQ) (1, 5). This section relied on parental recall, with symptoms considered present if they persisted for more than one month. The PPQ comprises 14 items, each with a binary response (Yes/No), and a positive response was assigned a score of one. A PPQ score of ≥ 6 is indicative of the presence of PTSD symptoms. The third part was the STAI to evaluate parental anxiety (1, 8, 9). This inventory comprises a 40-item self-report assessment employing a 4-point Likert scale for scoring. Higher scores

indicate elevated levels of parental anxiety, with scores exceeding 55 on either the State or the Trait STAI, indicating severe anxiety.

Follow-up visit parental psycho-social questionnaire

For subsequent visits during the study period, parents were requested to complete a condensed version of the first questionnaire that only included basic socio-demographic information of the parents and the STAI (State and Trait). Details such as the child's age and the purpose of the visit (admission to the surgical ward vs. regular OPD visit) were also documented. Parents were allowed to complete this questionnaire multiple times, provided that the visits were spaced at least 6 months apart.

Clinical data of the children born with EA

Clinical data of the children born with EA were also collected from hospital records. These included gestational age at birth, type of EA, and details regarding their surgical procedures (specifically whether they underwent esophageal replacement and/or had a gastrostomy at any time and the presence of major peri-operative complications).

Statistical analysis

Descriptive analysis was performed using means, standard deviations, medians, and interquartile range for continuous data, as appropriate. Categorical data were described using number and percentage. For the bivariate analysis, Student T-test were used as appropriate. A p-value of <0.05 was considered statistically significant.

Ethical approval was provided prior to the onset of the study (HREC reference number S20/10/260).

RESULTS

During the study period (2021-2023), a total of 22 children with EA were followed-up in our unit. Twenty-four parents were approached for recruitment. Four parents were excluded: two parents attended only one follow-up visit soon after initial discharge and were subsequently

lost to follow-up while two parents declined participation. Twenty parents (18 mothers and 2 fathers) of 18 children completed a total of 28 questionnaires (18 First visit and 10 Follow-up questionnaires). Just less than half ($n = 13$, 46%) of the visits, during which the parents completed a questionnaire, was documented as regular visits to the pediatric surgery OPD. The remaining 15 (54%) questionnaires were completed during an admission of their child (scheduled or emergency) to the surgical ward.

Six (33%) of the children in this cohort were born prematurely. The mean age of the children at the time of parental questionnaire completion was 63 ± 59 months, of which five (28%) were 6 months or younger. Sixteen (89%) children presented with a typical EA with a distal tracheo-esophageal fistula (TEF) and two (11%) with an isolated EA. Major complications were experienced by four (22%) children post-EA repair, including two significant anastomotic leaks and two with recurrent TEF's. Five (28%) of the children received an esophageal replacement and four (22%) a gastrostomy at some point during their treatment.

Socio-demographic information of the parents

The mean age of parents at time of questionnaire completion was 36 ± 10 years. Questionnaires were mostly completed by mothers ($n = 18$, 90%) with the majority ($n = 15$, 75%) of the parents reporting no difficulties in their own personal lives at time of questionnaire completion. The remaining five (25%) parents reported experiencing at least one of the following personal difficulties: health related problems, problems at work, relationship complications, financial difficulties, other problems, or combinations thereof. Parental social demographic data are described in **Table 1**.

Perinatal post-traumatic stress disorder questionnaire (PPQ)

The mean PPQ score was 3.72, ranging from 0 to a maximum score of 12. Six parents (33%) had a PPQ score of six or higher: two parents with a PPQ score of 6 and one each with a PPQ score of 9, 10, 11 and 12. There was no evident correlation between PPQ scores and the children's clinical EA course. One child of a parent with a high PPQ score (PPQ of ≥ 6) had a

straightforward peri-operative course during the neonatal period with no complications. Two children had a colonic interposition as esophageal replacements. Two of the remaining three children had minor post-operative complications (complications that did not require any surgical intervention) and one had a major anastomotic breakdown requiring redo-surgery. There was no significant association between parents with PPQ scores above or below 6 regarding gestational age at birth, type of EA or the need for additional surgical procedure (e.g., esophageal replacement or gastrostomy) as described in **Table 2**. A significant association was found between PPQ scores and the age of the child at time of questionnaire completion as well as the type of visit. We found a significant difference when we compared parents whose children had major peri-operative complications (defined as those who required surgical intervention) in the neonatal period with those who had minor or no complications (requiring no surgical intervention as part of the treatment). Furthermore, the majority (n = 3, 75%) of parents with children that experienced major complications, had PPQ scores >6.

The State-Trait Anxiety Inventory (STAI)

A total of 20 parents, comprising 18 mothers and 2 fathers, completed 28 STAI's. The mean State STAI (reflecting their current feelings) was 46 ± 7 . Four (14%) cases scored above 55 indicating high anxiety levels at the time of visit. The highest State STAI score documented was 59 and was scored by a mother of a premature born infant with an isolated EA and major complications post-operatively requiring a colonic interposition.

The mean score for the Trait STAI (assessing whether parents were generally anxious) was 46 ± 5 , with only one parent (4%) scoring above 55 during an emergency admission of the child for a gastroscopy and removal of a foreign body.

There were no statistically significant differences between the State STAI and Trait STAI scores ($p = 0.26$). The age of children at time of visit, the type of visit, the type of EA, a history of additional surgeries required and major or minor post-operative complications revealed no significant differences in either the State or Trait STAI scores achieved by our parents. The only statistically significant difference was found when we compared the State STAI of parents

with children born prematurely to those who were born term. The rest of the results are described in **Tables 3** and **4**.

All parents found to be at risk for developing PTSD (PPQ scores of ≥ 6) as well as those with scores indicating anxiety were referred for further work-up and counselling.

DISCUSSION

Our study findings indicate that one-third of parents with children born with EA were at risk for developing PTSD, while 14% had scores indicating high levels of anxiety at the time of questionnaire completion. The age of the child at the time of hospital visit, major complications during the peri-operative period as well as type of visit were associated with parental PTSD risk whilst prematurity was significantly associated with increased STAI scores.

Parents of neonates that are admitted to a NICU are exposed to various stressors, contributing to the development of mental health symptoms (2). A systematic review by Malouf *et al.* showed a higher prevalence of PTSD and anxiety in parents with neonates in NICU compared to parents of healthy neonates (2). De Mier *et al.* determined that the severity of neonatal complications, gestational age, and length of hospitalization were significant contributing factors for the development of PTSD in mothers of high-risk neonates (5). Based on these factors, they developed the PPQ as an early PTSD recognition tool for parents (5, 10). The PPQ score encompasses possible symptoms of PTSD as defined by the DSM-IV criteria and include questions about: symptoms of intrusion, avoidance, and changes in arousal (3, 5). Other studies have validated the PPQ's efficacy in identifying PTSD risk in mothers of high-risk neonates (10). In comparison to their studies, we found that major peri-operative complications during the neonatal period were a contributing factor for high PPQ scores in our cohort.

While studies have explored the psychological impact of congenital gastro-intestinal malformations (CGIM) requiring neonatal surgical intervention on parents, specific reports of

similar impact on parents of EA patients are scarce (11). It is acknowledged that parents of neonates with CGIM are at higher risk for developing PTSD later in life (11). Roorda *et al.* found that this group of parents were not only vulnerable to psychological distress, due to the birth itself, but also due to contributing factors such as the diagnosis of CGIM, intervention, and potential long-term complications associated with these malformations, as was also seen in our group of parents (11). Although EA was included in the group of malformations studied, they did not specifically report on parents with children born with EA. The prevalence of PTSD in their group of parents was 16.5% with mothers having a higher risk of developing PTSD when compared to a reference group (23% vs. 5%). This is slightly lower than the 33% of parents in our cohort with, all the at-risk parents being mothers.

Le Gouez *et al.* investigated the associations between the development of PTSD symptoms, and neonatal disease severity, severe complications in EA, as well as the quality of life and global health status of EA patients (1). They found no association between the presence of PTSD symptoms and neonatal disease severity or the presence of severe complications at 2 years of age (1). We found high PPQ scores in 75% of the parents whose children had major peri-operative complications during the neonatal period and a significant difference in the risk for the development of PTSD when compared with those with minor or no complications. Le Gouez *et al.* reported 59% of their participants had a PPQ score of six or more, which was significantly higher than the 33% in our cohort (1). They found that PTSD was only weakly associated with the age of children in their cohort, which again contrasts with our findings where the age of a child >6 months was associated with a high risk of PTSD (PPQ > 6) (1). These differences in outcomes emphasize the need for larger, more in-depth studies specifically focusing on the correlation between risk factors and the development of PTSD in parents of children born with EA.

The State-Trait Anxiety Inventory (STAI) is a valuable tool to distinguish between State and Trait anxiety in parents of children with EA, allowing differentiation between those who

experience anxiety constantly and those with anxiety in specific situations (e.g., during an admission vs. a routine follow-up visit) (8). In our study more parents had higher State STAI than Trait STAI scores indicating anxiety related to the timing of the questionnaire. Although we were unable to demonstrate a statistically significant correlation between high anxiety scores and the purpose of parent's visit in our cohort, we acknowledge the limitation of our small sample size.

The first study examining both PTSD and anxiety in parents of children born with EA, was conducted in 2016 by Le Gouez *et al.* (1). Both our study and the one conducted by Le Gouez *et al.* identified 4 cases with participants having State-STAI scores of above 55. In our study, we followed a similar model as Le Gouez *et al.*, with the addition of allowing for multiple completions of the STAI questionnaire for our study parents. The addition of multiple STAI questionnaire completions can possibly be seen as a form of counselling as we observed a decrease in scores in follow-up visits of parents who initially had scores above 55 in our cohort. This however highlights the need for larger, multicenter studies specifically looking at these different aspects possibly identifying these questionnaires as methods of counselling of parents. Such studies will help shed more light on the psychological aspects and provide valuable insights for better management and support for parents of children born with EA globally.

The small sample size in our study is a limitation that should be acknowledged, as it may restrict the generalizability and applicability of the results. Additional psycho-social determinants, specific to lower-middle income countries were not fully addressed (food insecurity, work insecurity, partner violence, financial worries *etc.*) and should be included in future studies. Only a few parents filled in repeat questionnaires. It may be clinically useful to perform longitudinal assessment to determine an improvement or worsening of at-risk symptoms in these parents with the changing medical state of their children. We also acknowledge that questionnaires identifying parents at risk of developing depression should be included in future studies for a more comprehensive examination of the mental health of

parents of EA patients.

By addressing the limitation of a small sample size, future research in this domain would enhance our understanding of the psychological aspects and experiences of parents facing the challenges of EA, as well as identifying further potential risk factors. This knowledge could contribute to the development of effective support systems and interventions tailored to meet the specific needs of these parents, ultimately improving the overall well-being of families affected by EA.

CONCLUSIONS

Our study highlights the importance of investigating and identifying potential symptoms of distress, specifically PTSD and anxiety, in the parents of children born with EA at an early stage, allowing for timely referral for counselling and treatment, if necessary.

By addressing the emotional needs of parents and incorporating them into the care process, comprehensive and holistic support for families affected by EA can be provided. This approach not only benefits the parents' mental well-being but also positively impacts the overall adjustment and coping of the entire family in managing the challenges of this disease.

COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interests

The Authors have declared no conflict of interests.

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Author contributions

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Ethical approval

Human studies and subjects

The protocol for this article conforms to the provisions of the Declaration of Helsinki (1995)

and has been approved Ethics approval by the Stellenbosch University Health Research Ethics Committee (S20/10/260).

Animal studies

N/A.

Data sharing and data accessibility

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Publication ethics

Plagiarism

Authors declare no potentially overlapping publications with the content of this manuscript and all original studies are cited as appropriate.

Data falsification and fabrication

All the data correspond to the real.

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Table 1. Parental socio-demographic information.

	n = 20
	n (%)
Relationship status	
Married	6 (30)
In a relationship	6 (30)
Widowed	1 (5)
Single	6 (30)
Didn't complete this question	1 (5)
Number of children in the household	
1	7 (35)
2	4 (20)
3	7 (35)
4	2 (10)
Highest level of parental education	
Didn't finish high school	8 (40)
Finished high school	6 (30)
College diploma	1 (5)
Unknown	5 (25)
Ever treated by a psychologist or psychiatrist?	2 (10)

Table 2. Comparison of PPQ scores.

	n = 18	Median PPQ (IQR 25 - 75)	p-value
Premature neonates	6	0.5 (0-10)	0.5
Term neonates	12	2.5 (0.5-6)	
Age at visit ≤6 months	5	0 (0-1)	0.02
Age at visit >6 months	13	4 (1-9.5)	
OPD visits	5	6 (3-10.5)	0.04
Hospital admission	13	1 (0-4.5)	
Isolated EA	2	0	0.1
EA with a distal TEF	16	2.5 (0.5-7.5)	
No additional surgeries	11	2 (0-6)	0.22
Esophageal replacement and/or gastrostomy	7	1 (0-11)	
Major complications post-EA repair	4	10 (4.5-11.5)	0.01
Minor or no complications	12 *	2 (0-5)	

PPQ: Perinatal Post-traumatic Stress Disorder Questionnaire; IQR: Interquartile Range; OPD: Out-patient Department; EA: Esophageal Atresia;

TEF: Tracheo-esophageal Fistula. * Two children had no neonatal repair done and are not included for comparison in this section.

Table 3. Comparison of State STAI scores.

	n = 28	Mean State STAI ± SD	p-value
Premature neonates	9	50 ± 7	0.04
Term neonates	19	44 ± 7	
Age at visit ≤6 months	5	48 ± 6	0.3
Age at visit >6 months	23	46 ± 7	
OPD visits	13	47 ± 7	0.2

Hospital admission	15	45 ± 7	
Isolated EA	4	49 ± 8	0.2
EA with a distal TEF	24	46 ± 7	
No additional surgeries	17	45 ± 7	0.3
Esophageal replacement and/or gastrostomy	11	47 ± 7	
Major complications post-OA repair	8	48 ± 8	0.2
Minor or no complications	18 *	45 ± 6	

STAI: State-Trait Anxiety Inventory; SD: Standard Deviations; OPD: Out-patient Department; EA: Esophageal Atresia; TEF: Tracheoesophageal

Fistula. * Two children had no neonatal repair done and are not included for comparison in this section.

Table 4. Comparison of Trait STAI scores.

	n = 28	Mean Trait STAI ± SD	p-value
Premature neonates	9	48 ± 2	0.06
Term neonates	19	45 ± 6	
Age at visit ≤6 months	5	46 ± 4	0.5
Age at visit >6 months	23	46 ± 5	
OPD visits	13	47 ± 5	0.2
Hospital admission	15	45 ± 5	
Isolated EA	4	49 ± 3	0.1
EA with a distal TEF	24	46 ± 5	
No additional surgeries	17	46 ± 6	0.2
Esophageal replacement and/or gastrostomy	11	47 ± 4	
Major complications post-OA repair	8	47 ± 5	0.3
Minor or no complications	18 *	46 ± 5	

STAI: State-Trait Anxiety Inventory; SD: Standard Deviations; OPD: Out-patient Department, EA: Esophageal Atresia, TEF: Tracheoesophageal

Fistula. * Two children had no neonatal repair done and are not included for comparison in this section.

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