Dysphagia in patients undergoing esophageal atresia surgery: Assessment using a functional scale

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ABSTRACT

Introduction. Dysphagia is defined as difficulty swallowing. Up to 84% of patients undergoing esophageal atresia surgery have dysphagia beyond the neonatal period.

Materials and methods. A retrospective study of patients undergoing esophageal atresia surgery from 2005 to 2021 was carried out. The Functional Oral Intake Scale (FOIS) was used to assess dysphagia in 4 age groups (<1 year old, 1-4 years old, 5-11 years old, and >11 years old). FOIS scores <7 or symptoms of choking, impaction, or food aversion were regarded as dysphagia.

Results. 63 patients were analyzed. 74% (47/63) had dysphagia during follow-up. Prevalence was 50% in patients <1 year old (FOIS mean 4.32), 77% in patients aged 1-4 (FOIS mean 5.61), 45% in patients aged 5-11 (FOIS mean 5.87), and 38% in patients >11 years old (FOIS mean 6.8). The most frequent causes of dysphagia were stenosis, which occurred in 38% of the patients (n=24), and gastroesophageal reflux (n=18), which was present in 28% of the patients. Both conditions were associated with significantly lower mean FOIS scores ($p<0.05$) in the patients under 11 years of age. Differences ($p<0.05$) were found in the dysphagia-associated perinatal factors in the various age groups, with longer ventilation assistance times, parenteral nutrition, and hospital stays.

Conclusions. Dysphagia is an extremely frequent symptom at any given age in patients undergoing esophageal atresia surgery. A standardized, cross-disciplinary follow-up is key to improve quality of life.

KEY WORDS: Esophageal atresia; Oropharyngeal dysphagia; Esophageal dysphagia.

DISFAGIA EN PACIENTES INTERVENIDOS DE ATRESIA DE ESÓFAGO: VALORACIÓN CON UNA ESCALA FUNCIONAL

RESUMEN

Introducción. La disfagia se define como dificultad en el proceso de alimentación. Hasta un 84% de pacientes intervenidos de atresia de esófago tienen disfagia más allá del periodo neonatal.

Material y métodos. Estudio retrospectivo de serie de casos intervenidos por atresia de esófago 2005-2021. Se utilizó la escala FOIS (Functional Oral Intake Scale) para cuantificar la disfagia en 4 grupos de edad (menores de 1 año, 1-4 años, 5-11 años y mayores de 11 años). Se consideró disfagia cualquier valor de FOIS <7 o síntomas de atragantamiento, impactación o aversión alimentaria.

Resultados. Se obtuvieron datos de 63 pacientes. El 74% (47/63) presentó disfagia durante el seguimiento. La prevalencia fue del 50% <1 año (media FOIS 4.32), 77% 1-4 años (media FOIS 5.61), 45% 5-11 años (media FOIS 5.87) y 38% >11 años (media FOIS 6.8). Las causas más frecuentes de disfagia fueron la estenosis, que presentó un 38% de los pacientes (n=24) y el reflujo gastroesofágico (n=18), que presentó a su vez un 28% de los pacientes. Ambas condiciones se asociaron con unos valores medios de FOIS significativamente menores ($p<0,05$) en los pacientes menores de 11 años. Se encontraron diferencias ($p<0,05$) en factores perinatales asociados a disfagia en los distintos periodos de edad, a destacar mayor tiempo medio de: asistencia ventilatoria, nutrición parenteral e ingreso hospitalario.

Conclusiones. La disfagia es un síntoma extremadamente frecuente a cualquier edad en los pacientes intervenidos de atresia de esófago. Un seguimiento estandarizado y multidisciplinar es esencial para mejorar la calidad de vida de estos pacientes.

PALABRAS CLAVE: Atresia de esófago; Disfagia orofaríngea; Disfagia esofágica.

INTRODUCTION

Dysphagia is defined as difficulty swallowing[(1)]. Normal swallowing includes various stages –oral (preparatory and propulsive), pharyngeal, and esophageal. Should any of these stages be impaired, the bolus has difficulty in

DOI: 10.54847/cp.2023.04.10

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This work was presented as an oral communication at the 9th Ibero-American Congress of Pediatric Surgery held in Oporto in 2022.

Date of submission: April 2022 *Date of acceptance:* May 2022

Table 1. Functional Oral Intake Scale (FOIS).

Functionality levels for oral intake	*Characteristics*
1	No oral intake.
2	Tube dependent with minimal/inconsistent oral intake.
3	Tube supplements with consistent solid/liquid oral intake.
4	Total oral intake of a single consistency.
5	Total oral intake of multiple consistencies requiring special preparation or compensations.
6	Total oral intake of multiple consistencies with no special preparation or compensations, but must avoid specific foods or liquid items.
7	Total oral intake with no restrictions.

advancing towards the stomach, thus causing alterations in the efficacy and safety of swallowing.

The incidence of dysphagia in the pediatric age is growing as a result of the increase in the survival of pre-term newborns and children with chronic conditions. Collecting objective clinical data proves uneasy as feeding disorders can be caused by multiple factors. Therefore, a cross-disciplinary team with deep knowledge of swallowing's complexity, the pathologies that may have an impact on it, and the various techniques available for dysphagia's clinical and instrumental diagnosis is essential[2].

Dysphagia is reported as a symptom by patients with esophageal atresia (EA). However, its real incidence is unknown due to the fact most patients learn to adapt to their unique anatomy and physiology, and therefore do not complain. In the literature, prevalence has been reported to be around 50% in patients over 10 years old[3], ranging from 21% to 84% in the studies carried out in other ages[4,5]. Up to 3 out of 4 patients report changes with respect to the general population in terms of feeding habits –need for greater water intake, diet changes, or being the last to finish their meals[6].

There are various scales available for the assessment of dysphagia[7-10], but none of them has been explicitly validated for EA. Gatzinsky et al. prospectively assessed dysphagia in adult patients undergoing EA surgery[11] using the Numerical Dysphagia Score described by Dakkak[12] and modified by Watson[13]. Another scale used in various articles[14,15] to evaluate dysphagia in the pediatric age is the FOIS scale[9] (Table 1), which describes the functional level of oral solid and liquid intake, considering modifications in both and the need for complementary feeding devices –nasogastric probe/gastrostomy.

The final objective of this work was to determine and characterize the presence of dysphagia in patients undergoing esophageal atresia surgery in our department from 2005 to 2021.

MATERIALS AND METHODS

A retrospective study of patients undergoing esophageal atresia surgery in our institution from 2005 to 2021 was carried out.

Each patient's variables were retrospectively studied and grouped in various age periods: < 1 year, 1-4 years, 5-11 years, and > 11 years.

Multiple variables were collected, including demographic data and personal history regarding the perinatal period, surgery, and the postoperative period.

Atresia types were categorized according to Gross classification[16].

In terms of dysphagia clinical signs, best and worst FOIS score (Table 1) over that period, presence of food aversion, presence of choking, recurrent respiratory signs, impaction signs, and reflux signs were recorded.

Regarding the diagnostic assessment of dysphagia's ultimate cause:

- Stenosis was defined as the presence of a change in caliber at an esophagogram or a high digestive endoscopy associated with clinical symptoms.
- Gastroesophageal reflux (GERD) was defined as the presence of a pH-metry with such diagnosis (reflux index > 10%), the passage of contents from the stomach to the esophagus in the gastrointestinal transit, and/or biopsy compatible with gastroesophageal reflux disease.
- Eosinophilic esophagitis was determined based on compatible biopsies. Biopsies of the three esophageal thirds were taken, with diagnosis being established based on the presence of > 15 eosinophils per high power field.

Mastication was considered to be impaired based on the notes from the dysphagia-specialized speech therapist on the medical records.

Airway abnormalities were considered to be so when diagnosed through fibrolaryngoscopy.

Swallowing incoordination was defined as the presence of such disorder at videofluoroscopy.

Table 2. Findings according to FOIS score and clinical signs.

Dysphagia	*Total patients (N= 63)*	*< 1 year*	*1-4 years*	*5-11 years*	*> 11 years*
FOIS < 7	33 (52%)	24 (38%)	29 (50%)	11 (31%)	1 (7%)
Clinical symptoms	46 (73%)	30 (48%)	42 (73%)	16 (45%)	5 (38%)
FOIS < 7 + clinical symptoms	47 (74%)	31(50%)	44 (77%)	16 (45%)	5 (38%)
FOIS mean in patients with dysphagia		4.32	5.61	5.87	6.8

Regarding the therapeutic approach for the treatment of dysphagia, feeding adaptation, esophageal dilatations, and surgical procedures were considered.

In the context of this study, FOIS scores < 7 or the presence of choking, impaction, or food aversion in the various age periods were regarded as dysphagia. As part of the dysphagia characterization work, it was classified as oropharyngeal dysphagia and esophageal dysphagia.

The statistical analysis was carried out using IBM®'s SPSS Statistics 25 software. A descriptive statistical representation was performed by defining the presence of dysphagia and subsequently categorizing it in subgroups –oropharyngeal, esophageal, or both– according to its ultimate cause and in the four age groups. An analytical statistical representation was later conducted by comparing the mean times of the perinatal factors in the four age groups and in the stenosis and gastroesophageal reflux subgroups.

The variables' distribution normality was assessed using the Kolmogorov-Smirnov test. For normal distribution quantitative variables, mean and standard deviation were calculated. For non-normal distribution quantitative variables, median and interquartile range were determined.

For qualitative variables, frequencies and percentages were achieved.

Clinical parameters were compared by means of an independent-sample t-test for normal distribution variables, and ANOVA for non-dichotomous variables. For non-normal distribution variables, Mann-Whitney's U test was used for dichotomous variables, and Kruskal-Wallis' test was employed for non-dichotomous variables.

To assess the association among qualitative variables, the Chi-squared test was used. To compare continuous quantitative data, Student's t-test was employed.

Statistical significance was established at $p<0.05$.

This study was carried out with the approval of Aragón's Research Ethics Committee (CEICA).

RESULTS

63 patients met the study's inclusion criteria.

Data was classified by age groups, with available data diminishing with longer follow-up times. 62 patients were < 1 year old, 57 patients were 1-4 years old, 35 patients were 5-11 years old, and 13 patients were > 11 years old.

Regarding the characteristics of the population meeting inclusion criteria, 40 patients were male and 23 were female. Mean patient age was 7.9 years (range: 0-16).

In terms of EA type, 93% of cases (n= 59) were Gross classification type C, 3 were type A, and 1 was type D. 2 cases were classified as Long Gap in the medical records.

7 cases were associated with VACTERL syndrome.

Mean gestational age was 36.9 weeks (range: 29-41), and mean weight at birth was 2,528 grams (range: 1,250-4,610).

Regarding the variables associated with the postoperative period, mean ventilation assistance time was 7 days (range: 0-37), time to enteral tolerance was 12.9 days (range: 2-124), time to parenteral nutrition was 20 days (range: 6-115), and time to nasogastric probe removal was 27 days (range: 2-118). Mean hospital stay was 41 days (range: 10-166).

Dysphagia was diagnosed in 74% of the patients (47/63) when including all follow-up periods. Table 2 features the number and percentage of patients with dysphagia in the various age periods.

In all patients diagnosed with dysphagia, the latter was classified as esophageal dysphagia and oropharyngeal dysphagia.

Stenosis, gastroesophageal reflux, and/or eosinophilic esophagitis were considered as objective causes of esophageal dysphagia, whereas mastication disorders, swallowing incoordination, and/or airway abnormalities were regarded as causes of oropharyngeal dysphagia.

In the classification process, two additional scenarios were found –patients with both oropharyngeal and esophageal dysphagia, and patients with clinical manifestations of dysphagia or FOIS disorders but with no objective cause of dysphagia.

Following dysphagia classification, the differences in the objective cause of dysphagia were analyzed according to patient age.

- In patients under 1 year of age, the most frequent causes were stenosis (19%) (12/62), GERD (14%) (9/62), and airway abnormalities (8%) (5/62).
- In patients aged 1-4 years old, the most frequent causes were stenosis (28%) (16/57), mastication disorders (15%) (9/57), and GERD (14%) (8/57).

Table 3. Differences in perinatal factors according to age groups.

Perinatal factors	*Dysphagia < 1 year*	*No dysphagia < 1 year*	*p value*
Ventilation assistance time	9.13 ± 1.44 days	5.9 ± 0.49 days	p < 0.05
Time to pleural drainage removal	11.46 ± 1.53 days	13.30 ± 2.69 days	p > 0.05
Time to enteral nutrition	13.74 ± 3.78 days	12.26 ± 2.28 days	p > 0.05
Time to nasogastric probe removal	33.32 ± 5.66 days	21.34 ± 3.08 days	p > 0.05
Parenteral nutrition time	25.36 ± 5.02 days	14.86 ± 1.71 days	p > 0.05
Hospital stay	56.80 ± 7.5 days	25.67 ± 2.47 days	p < 0.05
Perinatal factors	*Dysphagia 1-4 years*	*No dysphagia 1-4 years*	*p value*
Ventilation assistance time	8.18 ± 1.01 days	6.38 ± 1.19 days	p < 0.05
Time to pleural drainage removal	14.89 ± 2.25 days	7.46 ± 0.63 days	p < 0.05
Time to enteral nutrition	15.18 ± 3.07 days	8.46 ± 0.66 days	p < 0.05
Time to nasogastric probe removal	29.75 ± 4.05 days	24.83 ± 7.28 days	p > 0.05
Parenteral nutrition time	23.69 ± 3.64 days	11.41 ± 0.94 days	p < 0.05
Hospital stay	47.38 ± 5.65 days	28.07 ± 6.43 days	p < 0.05
Perinatal factors	*Dysphagia 5-11 years*	*No dysphagia 5-11 years*	*p value*
Ventilation assistance time	11.06 ± 2.31 days	6.63 ± 0.97 days	p > 0.05
Time to pleural drainage removal	13.06 ± 2.08 days	10.37 ± 2.52 days	p > 0.05
Time to enteral nutrition	11.66 ± 1.32 days	10.15 ± 1.72 days	p > 0.05
Time to nasogastric probe removal	29.33 ± 7.50 days	25.41 ± 5.80 days	p > 0.05
Parenteral nutrition time	29.68 ± 6.81 days	13.38 ± 1.99 days	p < 0.05
Hospital stay	57.12 ± 9.65 days	28.42 ± 5.12 days	p < 0.05

- In the 5-11-year-old group, the most frequent causes were stenosis (28%) (10/35), (GERD) 14% (5/35), and mastication disorders (8%) (3/35).
- In patients over 11 years of age, GERD was the only objective cause of dysphagia (7%) (1/13).

According to the presence or absence of dysphagia in the various age groups, differences among perinatal factor means were analyzed, with the following statistically significant differences (Table 3):

- Longer ventilation assistance time and hospital stay in dysphagia patients < 1 year old.
- Longer ventilation assistance time, time to pleural drainage removal, time to enteral nutrition, parenteral nutrition time, and mean hospital stay in dysphagia patients aged 1-4.
- Longer parenteral nutrition time and hospital stay in dysphagia patients aged 5-11.

No differences in mean perinatal factor times in the > 11-year-old group were found.

Of the total patients, 30 (47%) had esophageal dysphagia, 24 (38%) had oropharyngeal dysphagia, 12 (19%) had both, and 5 (7%) had symptoms of dysphagia, but with no objective cause.

Regarding esophageal dysphagia (n=30), 24 patients had stenosis, 18 had GERD, 3 had eosinophilic esophagitis, and 12 had both stenosis and GERD.

In terms of stenosis, it was diagnosed in 24 patients (38%). It was more frequent in the 1-4-year-old (16/57) and 5-11-year-old (10/35) groups. Stenosis patients had statistically significant differences ($p<0.05$) in certain perinatal factors –longer time to pleural drainage removal, longer parenteral nutrition time, and longer time to enteral nutrition (Table 4).

The presence of stenosis was associated with significantly lower FOIS scores ($p<0.05$) in the < 1-year-old (4.95 vs. 6.07), 1-4-year-old (5.41 vs. 6.30), and 5-11-year-old (5.93 vs. 6.94) age groups. 87% of cases (21/24) required dilatation, with a mean of 4.5 dilatations (range: 1-26). 12 cases (19%) had stenosis as the only cause of dysphagia, while other causes were accountable for it in the remaining patients.

GERD was diagnosed in 18 patients (28%). Regarding distribution by age groups, it was more frequent in the < 1-year-old (9/62) and the 1-4-year-old (8/57) groups. Gastroesophageal reflux patients had statistically significant differences ($p<0.05$) in certain perinatal factors –lon-

Table 4. Differences in perinatal factors according to the presence of stenosis.

Perinatal factors	*Stenosis*	*No stenosis*	*p value*
Ventilation assistance time	8.20 ± 1.09 days	7.02 ± 1.03 days	p > 0.05
Time to pleural drainage removal	16.27 ± 3.38 days	9.97 ± 1.32 days	p < 0.05
Time to enteral nutrition	18.60 ± 5.47 days	9.66 ± 1.01 days	p < 0.05
Time to nasogastric probe removal	28.81 ± 4.9 days	26.36 ± 4.25 days	p > 0.05
Parenteral nutrition time	27.29 ± 5.61 days	15.29 ± 2.24 days	p < 0.05
Hospital stay	48.50 ± 7.45 days	36.64 ± 5.24 days	p > 0.05

Table 5. Differences in perinatal factors according to the presence of gastroesophageal reflux.

Perinatal factors	*GERD*	*No GERD*	*p value*
Ventilation assistance time	12.11 ± 2.13 days	5.46 ± 0.34 days	p < 0.05
Time to pleural drainage removal	16.25 ± 3.05 days	10.87 ± 1.85 days	p > 0.05
Time to enteral nutrition	19.83 ± 6.42 days	10.06 ± 1.49 days	p < 0.05
Time to nasogastric probe removal	44.94 ± 8.01 days	19.25 ± 2.29 days	p < 0.05
Parenteral nutrition time	35.38 ± 7.69 days	13.52 ± 1.08 days	p < 0.05
Hospital stay	74.27 ± 8.19 days	27.63 ± 3.63 days	p < 0.05

ger time to extubation, longer parenteral nutrition time and time to enteral nutrition, longer time to nasogastric probe removal, and longer hospital stay (Table 5).

The presence of GERD was associated with significantly lower FOIS scores ($p<0.05$) in the < 1-year-old (3.41 vs. 6.54), 1-4-year-old (4.94 vs. 6.36), and 5-11- year-old (6 vs. 6.76) age groups.

83% of the patients (15/18) received medical treatment, and 66% of the patients (12/18) required surgery. In 6 cases (9%), GERD was the only cause of dysphagia, while other causes were accountable for it in the remaining patients.

Of the total patients with oropharyngeal dysphagia, and throughout the follow-up period (n=24), 11 patients (17%) had mastication disorders according to medical records, 5 patients (7%) had airway abnormalities diagnosed at nasal fibrobronchoscopy, and 7 patients (11%) had swallowing incoordination diagnosed at videofluoroscopy.

DISCUSSION

Dysphagia is an extremely frequent symptom in patients undergoing EA surgery. It occurs throughout development with heterogenous clinical manifestations. In a study published by Legrand et al.[17], dysphagia was estimated to have a prevalence of 60%, lower than in our series, where total prevalence was 74%.

The etiologic diagnosis of the ultimate cause of dysphagia can be complex. Various studies[18,19] have reported such difficulty, and the recent increase in dysphagia publications[20-22] shows there is growing concern regarding this symptom.

In 2016, Gottrand et al. published a highly informative review article objectively describing the nutritional and digestive issues of patients undergoing EA surgery[23]. In our study, stenosis and gastroesophageal reflux emerged as the most frequent causes of dysphagia, but not the only. The percentage of stenosis in other series (18-60%)[24,25] is similar to ours (38%). In addition, the percentage of gastroesophageal reflux reported in the literature (20-63%) [17,26] is higher than in our patients (28%). Arslan et al. published a series of articles[27,28] on mastication disorders as a cause of dysphagia in patients undergoing EA surgery. In our study, up to 17% of the patients had such symptom, and we believe it may be a less known, more difficult to assess cause of dysphagia, which does not make it less significant. Regarding the nutritional repercussion of feeding difficulty, up to 30% of patients undergoing EA surgery have been reported to have growth failur[29]. In our study, no anthropometric data was recorded, since the objective was not to quantify the final repercussions of dysphagia. However, extending our study in this respect could prove interesting.

The FOIS scale is regularly used in the clinical care of other pathologies, but it was first published by Coppens et al.[15] in 2016 to assess dysphagia specifically in EA patients. In this study, it also emerged as a useful, reproducible tool to identify and assess the presence of dysphagia with significant clinical repercussion.

Yalcin et al.[19] propose videofluoroscopy as a useful diagnostic tool to identify and classify dysphagia. In their study, 32 patients undergoing esophageal atresia surgery were assessed through videofluoroscopy, with a lower percentage of oropharyngeal dysphagia (37%) than in our series, where 12 videofluoroscopies were carried out, with 58% of them revealing oropharyngeal dysphagia –however, it should be noted that, in our study, videofluoroscopy was not systematically conducted in all patients. In this respect, we believe it would be interesting to learn about the experience other institutions have with it.

Complementary to the descriptive study of dysphagia, a mean difference analysis in terms of perinatal factors was carried out, with differences in variables being found between patients with and without dysphagia in the various age groups. To our knowledge, there are no studies specifically correlating the presence of dysphagia in the long-term with perinatal factors.

Regarding our study limitations, this study has a retrospective nature. This means we cannot be as objective as if it were a prospective one, since it is based on medical records, and anamnesis aimed at searching for the presence of dysphagia is not always carried out. The variable prevalence of esophageal atresia and the losses or variability in the follow-up periods also mean there is less data available and it is more irregular. When retrospectively analyzing data, only correlations –and not causal statements based on our sample– can be made.

We are also well aware of the limitation implied by the fact the various types of atresia were not compared with each other. Indeed, most of our study patients had Gross type C atresia, and we had an insufficient number of patients with other types for them to be statistically comparable.

Nevertheless, we believe this is a novel study in the field of pediatric surgery that could help standardize a study protocol and establish the therapeutic actions required to treat dysphagia in patients undergoing EA surgery.

ACKNOWLEDGEMENTS

To the Medical Archive and Documentation Center of our institution for letting us review the medical records.

To the patients, parents, and guardians for giving their consent to the use of their data and therefore allowing us to better identify their needs.

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