Dynamic Imaging Grade of Swallowing Toxicity in Children with Esophageal Atresia

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Eur | Pediatr Surg

Abstract

Introduction The Dynamic Imaging Grade of Swallowing Toxicity (DIGEST) scale was developed to evaluate the safety, efficiency, and overall pharyngeal swallowing performance in patients with dysphagia (DIGESTs, DIGESTe, and DIGESTt, respectively). Although various types of swallowing dysfunction are encountered in children with esophageal atresia (EA), oropharyngeal dysphagia poses risk for aspiration. Therefore, a retrospective study was performed to evaluate the safety and efficacy of swallowing by using DIGEST score in children with EA.

Patients and Methods Thirty-nine EA patients were included. The demographic features, respiratory problems, results, and outcomes of surgical treatment were evaluated from medical records. The videofluoroscopic swallowing evaluation investigated for both airway protection and bolus residuals at the level of vallecula, posterior pharyngeal wall, and pyriform sinus at liquid and pudding consistencies. The penetration and aspiration scale (PAS) was used to define penetration and aspiration severity, and DIGEST was used to evaluate DIGESTs, DIGESTe, and DIGESTt.

Results The median age of the patients were 13 months (7–39 months), and male-to-female ratio was 25:14. Sixty-seven percent of patients were type-C EA and 61% of them has associated anomalies; 38% of patients had aspiration (PAS = 6–8) in liquids and 10% in pudding consistency. Life-threatening/profound swallowing dysfunction in DIGESTE (DIGEST = 4) was seen in 13% (n = 5) of patients; 40% of EA patients showed severe problems in DIGESTt.

Conclusion DIGEST is a valid and reliable tool to define the efficacy and safety of swallowing in children with EA.

Keywords

- esophageal atresia
- tracheoesophageal fistula
- swallowing
- dysphagia
- children

Introduction

Esophageal atresia (EA) is the most common congenital anomaly of esophagus with an incidence of 1:3,500 live births.¹ Patients with EA show wide range of swallowing problems and dysphagia is a common long-term morbidity

in all age groups.² Not only esophageal phase of deglutition but also oral and pharyngeal phases are impaired in 35% of cases.^{2,3} Oropharyngeal impairment and aspiration are considered as a significant risk factor for respiratory problems in children with EA.^{3,4} Previous studies have already shown that EA patients have bolus residuals at different anatomical

received July 5, 2023 accepted after revision September 21, 2023 accepted manuscript online September 25, 2023 © 2023. Thieme. All rights reserved. Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany DOI https://doi.org/ 10.1055/a-2181-2293. ISSN 0939-7248.

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locations with different amount of food and consistencies. During the swallowing, the pharyngeal retentions and bolus residuals are significant risk factors for aspiration and considered as one of the causes of respiratory morbidity. Therefore, videofluoroscopic swallowing evaluation (VFSE) have been commonly used for evaluating the pharyngeal phase of deglutition. Bolus residual scale and normalized residual ratio scales have been used to evaluate the pharyngeal impairment in patients with EA. Both scales enable to define the severity of retention in different anatomical locations with observational and quantitative methods. However, none of those studies defines the safety and efficacy of pharyngeal dysphagia in terms of grades.

Dynamic Imaging Grade of Swallowing Toxicity (DIGEST) is an analysis method developed to grade the severity of the pharyngeal dysphagia based on the VFSE findings. 9 It aims to grade the pharyngeal residue and penetration/aspiration in terms of swallowing safety and efficacy. It has been used as simple but robust tool for many clinical situations causing pharyngeal dysphagia such as head and neck cancers. 10 The safety problems in DIGEST suggests a respiratory problem, whereas efficacy may refer a possible nutritional impairment. 10 Thus, it does not only grade the severity of pharyngeal impairment but also correlates with the clinical problems related with pharyngeal swallowing dysfunction. To the best of our knowledge, it has not been used in pediatric population especially in EA patients. Therefore, we performed a retrospective study to evaluate the DIGEST grades of children with EA.

Patients and Methods

The study was approved by local ethical committee (GO/2023-20).

Patients operated for EA and underwent VFSE for swallowing problems were included in the study. The demographic features, associated anomalies, respiratory problems, results, and outcomes of surgical treatment were evaluated from medical records. Patients without VFSE and with no full oral feedings were excluded.

Videofluoroscopic Swallowing Evaluation

VFSE is known to be the basic method for the investigation of the deglutitive functions. Oral, pharyngeal, and esophageal phases of deglutition are evaluated with different consistencies of food in this procedure.³

Two authors (N.D. and S.S.A.) re-evaluated the video records of the VFSE investigation of the included cases. The penetration and aspiration scale (PAS) and DIGEST scale evaluations were evaluated as given below and recorded.

VFSE examinations were performed with liquid (1–3–5–10–20 mL of barium), pudding (3–5–10 mL of barium with pudding), and solid (5–10 mL of barium with biscuit) barium tests and used the 5 mL volume results for the analysis, since this amount provides more effective evaluation of swallowing physiology. Penetration and aspiration scores were evaluated in VFSE by using the PAS, and scores 6 or grater were considered as aspiration (►Table 1).

Table 1 Penetration and aspiration score

1	No penetration and aspiration	No contrast in the airway
2	Penetration	Contrast at the supraglottic level, no contrast residue
3		Contrast at the supraglottic level, visible contrast residue
4		Contrast at the level of glottis, no contrast residue
5		Contrast at the level of glottis, visible contrast residue
6	Aspiration	Contrast at the subglottic level, no contrast residue
7		Contrast at the subglottic level, visible contrast residue despite the response of the patient
8		Contrast at the subglottic level, visible contrast residue with no response of the patient

DIGEST

DIGEST is a validated method evaluating the grade of severity of pharyngeal swallowing impairment based on the degree and the patterns of penetration/aspiration and pharyngeal residues from VFSE findings. In the first step, a swallowing therapist rates all pharyngeal bolus clearance in a standardized VFSE protocol. Then, DIGEST criteria were applied to derive DIGEST grades as reported by Hutcheson et al (Fig. 1). ¹⁰ Pharyngeal dysphagia severity per DIGEST grade 1 = mild, 2 = moderate, 3 = severe, and 4 = life threatening/profound. DIGEST was also evaluated for safety (DIGESTs), efficiency (DIGESTe), and overall pharyngeal swallowing performance (DIGESTt).

Statistical Analysis

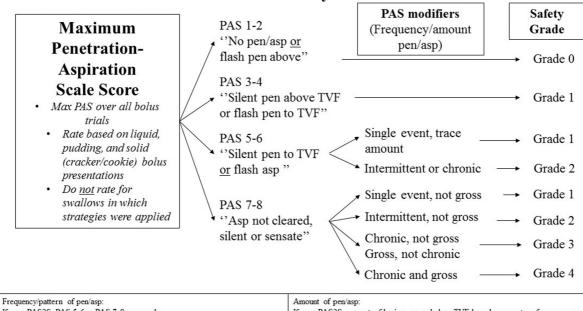
The IBM-SPSS for Windows version 20 software (IBM Corp., Armonk, New York, United States) was used for statistical analyses. The descriptive statistics were calculated as number and percent for qualitative data. Nonparametric tests were used for intragroup comparison of ordinal variables. Pearson's correlation was used to define a relation between PAS scores and DIGEST parameters. A *p*-value of less than 0.05 was considered to be statistically significant.

Results

A total of 39 patients with EA were included in the study. The median age of the patients were 13 months (7–39 months), and male-to-female ratio was 25:14. **Table 2** shows the demographic features, types of EA, clinical findings, surgical methods, and outcomes. The most common Gross type was type-C (67%) and 61% of the patients had associated anomalies.

PAS scores and DIGEST grades are summarized in **Table 3**. Thirty-eight percent of patients had aspiration (PAS = 6-8) in liquids and 10% in pudding consistency. Lifethreatening/profound swallowing dysfunction in DIGESTe (DIGEST = 4) was seen in 13% (n=5) of patients, whereas 40% of EA patients showed severe problems in DIGESTt.

DIGEST Safety Grade

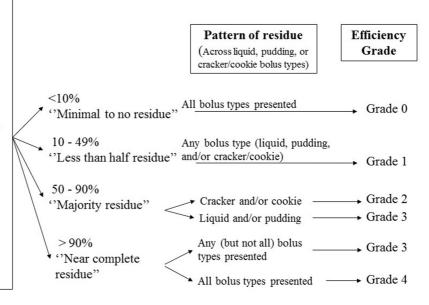


If max PAS2S, PAS 5-6 or PAS 7-8 occured;
Single event
Intermittent (on multiple but <50% of trials on a single consistency
Chronic (majority [>50%] of thin liquid trials and /or on >1
Consistency
If max PAS2S, amount of barium on or below TVF based on worst performance on any single bolus;
Trace (resembles faint coating, droplets or trickle of barium on/below TVF
Neither trace nor gross
Gross (<25% bolus volüme)

Maximum % of pharyngeal residue

- Max % (proportion) of bolus in pharynx over all bolus trials
 - Rate based on liquid, pudding, and solid (cracker/cookie) bolus presentations
- Rate based on %
 pharyngeal residue
 after initial swallow
 attempt of each bolus
- Do <u>not</u> rate based on oral residue
- Do <u>not</u> rate for swallows in which strategies were applied

Efficiency Grade



	S0	S1	S2	S3	S4
E0	0	1	2	3	3
E1	1	1	2	3	3
E2	1	2	2	3	3
E3	2	2	3	3	4
E4	3	3	3	4	4

Fig. 1 Bolus scoring criteria for DIGEST. DIGEST, Dynamic Imaging Grade of Swallowing Toxicity; PAS, penetration and aspiration scale.

Table 2 Demographic features, clinical findings, results, and outcomes of surgical treatment

Parameters (n = 39)	n (%)			
Median age (mo)	13 (7–39.5)			
Gender (male:female)	25:14			
Type of EA				
A	10 (26%)			
С	26 (67%)			
E	3 (8%)			
Surgical treatment				
Primary anastomosis	26 (67%)			
Delayed primary anastomosis	8 (21%)			
Esophageal replacement	5 (13%)			
Respiratory symptoms	30 (77%)			
No respiratory problem	9 (23%)			
RP	10 (26%)			
Bronchiectasis/atelectasis	5 (13%)			
RP + bronchiectasis	2 (5%)			
RP + diagnostic bronchoscopy	4 (10%)			
RP + diagnostic bronchoscopy + bronchiectasis	8 (21%)			
GERD	23 (60%)			
No GERD	16 (41%)			
Medical treatment	17 (44%)			
Surgical treatment	6 (15%)			
Complications	29 (81%)			
No	7 (19%)			
AL	1 (3%)			
AS	5 (14%)			
Median number of dilatations	1 (0-4)			
RTEF	2 (6%)			
AL + AS	12 (33%)			
AL + RTEF	2 (6%)			
AS + RTEF	1 (3%)			

Abbreviations: AL, anastomotic leak; AS, anastomotic stricture; EA, esophageal atresia; GERD, gastroesophageal reflux disease; RP, recurrent pneumonia; RTEF, recurrent tracheoesophageal fistula.

When patients with no penetration and no aspiration (PAS = 1) were compared with patients with penetration (PAS = 2–5) and aspiration (PAS = 6–8), DIGESTs, DIGESTe, and DIGESTt grades were significantly higher in patients with penetration and aspiration (p < 0.05). Only DIGESTe and DIGESTt grades were higher in patients with aspiration compared with penetration in both liquid and solid consistencies (p < 0.05). DIGESTs were similar in patients with aspiration and penetration in liquid swallows (p > 0.05).

PAS scores were correlated with DIGEST grades for safety, efficacy, and total scores. **Table 4** demonstrates the correlation of PAS scores with all DIGEST grades in different

Table 3 PAS scores and DIGEST grades in patients

Parameters	n (%)		
Median PAS liquid	1 (1–8)		
Normal (PAS = $1-2$)	24 (62%)		
Penetration (PAS $=$ 3 $-$ 5)	0		
Aspiration (PAS $= 6-8$)	15 (38%)		
Median PAS solid	1 (1-8)		
Normal (PAS = $1-2$)	35 (90%)		
Penetration (PAS = $3-5$)	0		
Aspiration (PAS $= 6-8$)	4 (10%)		
Residuals liquids			
Vallecula	2 (5%)		
Posterior pharyngeal wall	0		
Piriform sinus	1 (3%)		
Residuals solids			
Vallecula	5 (13%)		
Posterior pharyngeal wall	1 (3%)		
Piriform sinus	1 (3%)		
DIGESTe			
Normal = 0	22 (56%)		
Minimum = 1	0		
Mild = 2	3 (8%)		
Severe = 3	9 (23%)		
Life threatening/profound $=$ 4	5 (13%)		
DIGESTs			
Normal = 0	30 (77%)		
Minimum = 1	7 (18%)		
Mild = 2	2 (5%)		
Severe = 3	0		
Life threatening/profound = 4	0		
DIGESTt	<u>.</u>		
Normal = 0	21 (54%)		
Minimum = 1	1 (3%)		
Mild = 2	3 (8%)		
Severe = 3	14 (40%)		
Life threatening/profound = 4	0		

Abbreviations: DIGEST, Dynamic Imaging Grade of Swallowing Toxicity; DIGESTe, efficiency DIGEST; DIGESTs, safety DIGEST; DIGESTt, overall pharyngeal swallowing performance DIGEST; PAS, penetration and aspiration scale.

consistencies. Since PAS evaluation is integral part of the DIGEST scoring, a significant strong correlation between PAS and DIGESTt scores were found as expected (p=0.001, r=0.954). When DIGEST scores were grouped as DIGEST negative (DIGEST [-]=0) and positive (DIGEST [+]=1-4), there was no statistical difference between DIGEST (+) and DIGEST (-) cases for respiratory problems, gastroesophageal reflux, and surgical complications (p > 0.05).

Table 4 The correlation between PAS scores and DIGEST grades in different consistencies

	p-Values, correlation coefficient (r)				
	DIGESTs	DIGESTe	DIGESTt		
PAS liquids	0.05, 0.440	0.001, 0.954	0.01, 0.956		
PAS solids	0.05, 0.439	0.001, 0.555	0.008, 0.418		

Abbreviations: DIGEST, Dynamic Imaging Grade of Swallowing Toxicity; DIGESTe, efficiency DIGEST; DIGESTs, safety DIGEST; DIGESTt, overall pharyngeal swallowing performance DIGEST; PAS, penetration and aspiration scale.

Discussion

Pharyngeal dysphagia is a well-documented postoperative complication in patients with EA.^{2,3} In EA, structural anomalies may cause disruption in anatomical relation between esophagus and epiglottis and may lead to insufficient airway closure and aspiration.¹¹ In 35% of EA cases, pharyngeal swallowing problems ranging from mild to severe dysphagia have been reported.3 However, severity of pharyngeal dysphagia and its consequences on safe swallowing have not been evaluated. DIGEST is a validated method to define the severity and grade of swallowing dysfunction. The safety problems in DIGEST (DIGESTs) suggests an unsafe swallowing that may cause respiratory problem, whereas efficacy (DIGESTe) may refer ineffective swallowing resulting with nutritional impairment.¹⁰ In this study, we first documented that 40% of patients had severe DIGEST grades. Although the absence of life-threatening results in the safety evaluation was a very good result, efficacy evaluation showed profound problem in 13% of the patients. Therefore, we suggest that problems with effective swallowing are more common than safe swallowing in our cohort of patients. Moreover, we could not find any statistical difference in DIGEST (+) and (-) patients for respiratory problems.

PAS score is also reliable tool to define penetration and aspiration in VFSE. Patients with penetration are at risk for aspiration and recurrent respiratory infection. In EA patients, 20% of cases penetration and aspiration and patients with delayed primary repair had higher PAS scores compared with early repair.8 In addition to structural anomalies, tensioned anastomosis and delayed repair associated with higher penetration and aspiration scores in EA patients. In this study, we also found that 38% of cases had aspiration in liquids and 10% had aspiration in solid consistencies. Also, PAS is strongly (r=0.954-0.056) correlated with high grades of DIGEST efficacy and total especially in liquids in EA cases. Since PAS evaluation is integral part of DIGEST scoring, significant correlation between these two scores is expected.

DIGEST utilizes two scores including safety profiles and efficacy profiles of patients from VFSE derivate images. 10 It is commonly used to grade the pharyngeal swallowing dysfunction in head and neck cancers and other dysphagia causes in adults. 10 This is the first study that DIGEST was used to grade pediatric pharyngeal dysphagia. DIGEST scores

are obtained from VFSE parameters. VFSE is a diagnostic method with proven validity and reliability in children. Therefore, we suggest that it is applicable for pediatric population and defines the severity of pharyngeal problems in terms of efficacy and safety. Since pediatric population is more vulnerable for aspiration, safety is very important for safe swallowing. Also, efficacy information is significant for effective deglutition and nutrition for a developing child. Therefore, we suggest that DIGEST seems a reliable tool superior to other observational and quantitative method in EA patients.

Our study has some limitations. First, we have small number of patients with varying types of treatment. Second, since the symptoms of the patients occur due to various underlying causes, it was not possible to correlate the symptoms with VFSE and DIGEST findings. Also, nutritional assessment of the patients will be more informative to interpret the efficacy parameters. Finally, tensioned anastomosis and other structural anomalies such as motility disorders, tracheomalacia should be evaluated together with DIGEST grades. However, the possible structures are multifactorial, a single structural anomaly could not attribute to impaired DIGEST scores. Despite these limitations, this is the first study showing the use of DIGEST grades in pediatric population and EA patients. Moreover, unlike to other diagnostic evaluations, DIGEST provides more reliable and detailed information about the severity and safety of pharyngeal swallowing.

Conflict of Interest

None declared.

Note

The study was presented in the 24th EUPSA Congress in June 7–10, İzmir, Turkey.

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