The evaluation of deglutition with videofluoroscopy after repair of esophageal atresia and/or tracheoesophageal fistula

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ABSTRACT

Aim: A retrospective study was performed to evaluate the functional disorders of deglutition with videofluoroscopy (VFS), in children operated for esophageal atresia (EA) and/or tracheoesophageal fistula (TEF).
Methods: Patients with the repair of EA-TEF were evaluated in respect to the type of malformation, operative procedure, postoperative complications, deglutitive and respiratory symptoms, and gastroesophageal reflux disease (GERD). The dysphagia score, VFS findings of oral, pharyngeal and esophageal phases, and penetration–aspiration scale (PAS) score were recorded in the evaluation of the deglutitive functions.
Results: Thirty-two cases with a median age of 48 months (2–120 months), and male to female ratio of 14:18 were included in the study. Most of the cases had Gross type C anomaly (n = 26, 81.3%), and the others were type A (n = 3), D (n = 2) and E (n = 1). The incidence of associated anomalies was 71.8%. The patients underwent primary (n = 26, 81.3%) or delayed (n = 6, 18.7%) anastomosis. Postoperative complications including anastomotic stricture (n = 12), leak (n = 2) and recurrent fistula (n = 2) were managed by dilatation, conservative approach and repair of the fistula, respectively. Recurrent pneumonia (n = 13), cough with liquid intake (n = 10) and food impaction (n = 7) were recorded in the history. Management of GERD included medical (n = 11) and surgical (n = 7) treatment. The median dysphagia score was 3.5 (min: 0–max: 27). The oral phase of VFS was normal in most of the cases (n = 29, 90.6%). Only three had mild or moderate impairment, and none had severe. The pharyngeal phase showed no impairment in 23 of the cases (71.8%), and severe impairment was observed only in 3 of all, for the parameters of hyolaryngeal elevation and airway closure. Opposite to the first two phases of the deglutition, the esophageal phase was normal in only 2 of the cases (6.3%). Among the other 30 cases with impairment, only two had mild, and the rest had moderate to severe problems. Esophageal backflow, motility and residue were the most severely impaired parameters of this phase. The PAS evaluation revealed no penetration and aspiration in 26 of the cases (81.3%), while 1 had penetration and five had aspiration.
Conclusion: The patients with repaired EA-TEF may reveal deglutitive and respiratory symptoms in follow-up, necessitating certain investigations. The deglutition is functionally evaluated with VFS. While the disorders of oral and pharyngeal phases are less frequent and prominent, the esophageal phase reveals disorders with higher incidence and severity.

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Esophageal dysfunction is a common problem in children with repaired esophageal atresia (EA) with or without tracheoesophageal fistula (TEF), and considered as a long-term sequel of these cases [1]. Esophageal motility disorders in EA are multifactorial. Impaired esophageal motility in EA survivors is attributed to primary abnormal esophageal innervation and vagal nerve damage during esophageal repair [2]. Dysphagia, regurgitation, aspiration and chronic respiratory tract infections are considered as clinical findings of esophageal dysmotility. Even in the absence of symptoms, the evaluation of esophageal motility reveals dysfunction in majority of the

cases [3,4]. Dysphagia is a common clinical problem in EA-TEF survivors in adulthood, but the prevalence of dysphagia in childhood is not clarified yet, because of lack of objective criteria. The dysphagia symptom score (DS) is thought to be supportive for a standardized definition of dysphagia [5].

Esophageal manometry, videomanometry and multichannel intraluminal impedance with pH monitoring are the diagnostic tools used to demonstrate the esophageal motility [3,4,6]. Videomanometric evaluation of patients with repaired EA revealed dyscoordination of pharyngeal contraction and upper esophageal sphincter relaxation, which caused airway aspiration [4]. Videofluoroscopy (VFS) is a dynamic method investigating the oral, pharyngeal and esophageal phases of deglutitive function in children [7]. Although these studies suggest deglutitive dysfunction in children with EA-TEF, the prevalence of dysphagia and impairment in deglutition need further clarification. This

retrospective study was performed to evaluate the dysphagia symptom and deglutitive functions in children operated for EA-TEF, with dysphagia score (DS) and VFS.

1. Patients and methods

Patients operated for EA-TEF between 2003 and 2014 were investigated in respect to the type of malformation, associated anomalies, operative procedure, postoperative complications, deglutitive and respiratory symptoms, and gastroesophageal reflux disease (GERD) retrospectively. Type A patients who underwent esophageal replacement were not included in the study. Symptom of dysphagia was evaluated by the scoring system first introduced by Dakkak et al and modified by Watson et al (Table 1) [8,9]. Dysphagia for liquids and solid substances was revealed by parents as 'absent' when the patient never experienced swallowing difficulty, 'improved' when had difficulty in the past but not now, and 'ongoing' when the patient has still difficulty. Dysphagia score (DS) included the information about the presence of any dysphagia for each liquid or solid substance. The score for each item (always = 1 point, sometimes = 1/2 point, never = 0 point) was multiplied by the adjacent line number, and the sum of all nine lines revealed the total DS. A score from 0 (no dysphagia) to 45 (severe dysphagia) was assigned for each patient.

1.1. Videofluoroscopic evaluation

Videofluoroscopy is known to be the basic method for the investigation of the deglutitive functions. We performed VFS for all patients with repaired EA and/or TEF 6 months after the operation, and earlier if they had symptoms of dysphagia and/or aspiration. The VFS evaluation for the patients operated in another center was performed on admission, even they were at an older age. Oral, pharyngeal and esophageal phases of deglutition are evaluated with different consistencies of food in this procedure [10]. We performed 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. The parameters for the three phases of deglutition were analyzed and scored with a 0–3 point scale (Table 2). The score of function (0: no impairment, 1: mild, 2: moderate, 3: severe impairment), residue (no residue, 1: minimal, 2: moderate, 3: severe residue), delay in swallowing response (0: no delay, 1: 0–2 sec, 2: 2–5 sec, 3: >5 sec delay), airway closure (0: no delay, 1: minimal penetration, 2: supraglottic penetration, 3: subglottic aspiration), and aspiration (0: absent, 1: present) were recorded in accordance with the correspondent definitions. The penetration–aspiration scale (PAS) was also used in the evaluation of VFS findings, and the score of 1–2 defined 'no penetration and aspiration', 3–6 'penetration', and 7–8 'aspiration' (Table 3) [11].

Table 1
Dysphagia score system.

FOOD	SCORE		
	Always (1 point)	Sometimes (1/2 point)	Never (0 point)
1. Water			
2. Milk/thin soup			
3. Yogurt/pureed fruit			
4. Jam/jelly			
5. Scrambled egg/mashed potato			
6. Fish/boiled vegetables			
7. Bread			
8. Fresh fruit			
9. Meat			

Table 2
Videofluoroscopy scale.

VFS findings	Score
Oral phase (0–3)	
Lip closure	
Tongue elevation	
Tongue retraction	
Oral residue	
Pharyngeal phase (0–3)	
Delay in swallowing response	
Touch of tongue root to pharynx	
Velopharyngeal closure	
Hyolaryngeal elevation	
Vallecular residue	
Pharyngeal residue	
Closure of airway	
Pyriform sinus residue	
Aspiration	
Silent aspiration	
Esophageal phase (0–3)	
Opening of UES	
UES residue	
Esophageal backflow	
Motility problem	
Esophageal residue	
LES dysfunction	

VFS: videofluoroscopy UES: upper esophageal sphincter LES: lower esophageal sphincter.
Score of function: 0: no impairment, 1: mild, 2: moderate, 3: severe impairment.
Score of residue: 0: no residue, 1: minimal, 2: moderate, 3: severe residue.
Score of delay in swallowing response: 0: no delay, 1: 0–2 sec (mild risk), 2: 2–5 sec (moderate risk), 3: >5 sec (severe risk) delay.
Score of airway closure: 0: no delay, 1: minimal penetration, 2: supraglottic penetration, 3: subglottic aspiration.
Score of aspiration: 0: absent, 1: present.

1.2. Ethics and statistical analysis

The study was approved by Ethical Committee of Hacettepe University (HU 16969557-732, 2013), and informed consent was obtained from the parents. The descriptive analysis of the data was performed with SPSS 15.0.

2. Results

Thirty-two cases with a median age of 48 months (2–120 months), and male to female ratio of 14:18 were included in the study. Most of the cases had Gross type C anomaly (n = 26, 81.3%), and the others were type A (n = 3), D (n = 2) and E (n = 1). The incidence of associated anomalies was 71.8%, the cardiopulmonary (n = 18) and gastrointestinal (n = 6) anomalies were the most common ones. The patients underwent primary (n = 26, 81.3%) or delayed (n = 6) anastomosis. Seventeen of the cases were operated in our center, and the other 15 admitted after being operated in another one. The postoperative complications including anastomotic stricture (n = 12), leak (n = 2) and recurrent fistula (n = 2) were managed by dilatation, conservative approach and repair of the fistula, respectively. Oral feeding could be started within 1st week (n = 16, 50%), 1st–4th week (n = 6), and

Table 3
Penetration and aspiration scale.

1	**No penetration and**	No contrast in the airway
2	**aspiration**	Contrast at the supraglottic level, no contrast residue
3	**Penetration**	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		Contrast at the subglottic level, no contrast residue
7	**Aspiration**	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