The role of fiberoptic endoscopic evaluation of swallowing in the assessment of pediatric dysphagia
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Introduction
Swallowing is a basic, life-sustaining function. It is an innate ability present in the developing fetus [1]. Swallowing is so natural that we do it ~580 times daily even without thinking about it [2].

There are specific physiological components of normal swallow that occur in a series of unfolding stages called phases.

(1) The oral phase: this phase is subdivided into two phases
(a) The oral preparatory phase.
(b) The oral transport phase.
(2) The pharyngeal phase.
(3) The esophageal phase.

Any change or damage that occurs in any of the sensory or motor events that comprise the oral preparatory, oral transport, and pharyngeal stages of swallowing causes difficulty in moving the food from the mouth to the stomach, or what is called dysphagia [3].

Causes of pediatric dysphagia
(1) Structural etiologies (e.g. craniofacial anomalies and laryngomalacia).
(2) Neurologic etiologies (e.g. prematurity and cerebral palsy).
(3) Respiratory compromises (cardiac, pulmonary, or airway anomalies).

Background
Swallowing is a basic, life-sustaining function that involves interplay between two distinct but related phenomena, airway protection and bolus transport. Pediatric dysphagia is one of the most important symptoms to be assessed and managed. The standard fiberoptic endoscopic evaluation of swallowing (FEES) protocol of Langmore (2001) was designed to assess dysphagia on all populations.

Aim
The aim of this work was to clarify the role of FEES in the diagnosis of pediatric dysphagia and the signs related to it.

Study design
This was a retrospective study that was conducted to assess pediatric dysphagia using FEES as a clinical diagnostic tool.

Participants and methods
The study included 64 children (38 male, 26 female). Of them, 32 patients were suffering from difficulty in swallowing of different degrees and 32 were controls (they were not suffering from any difficulty in swallowing). The mean age in months for symptomatic children was 41.47 ± 36.25 and the mean age in months for control cases was 42.08 ± 35.61. The examination was carried out using FEES applying the standard FEES protocol of Langmore (2001).

Results
Application of the standard FEES protocol of Langmore (2001) showed highly related signs of pediatric dysphagia, such as handling of secretions, pharyngeal function in part I and timing of the bolus flow/initiation of the swallow, structural movements during the swallow, and residue after the swallow and between swallows in part II.

Conclusion and recommendation
There are more common signs related to pediatric dysphagia than others and should be considered in any therapeutic program for overcoming dysphagia in children. Laryngomalacia is a structural disorder causing pediatric dysphagia in a considerable number of children. The standard FEES protocol should be applied on a larger number of pediatric populations with different disorders.

Keywords:
fiberoptic endoscopic evaluation of swallowing, pediatric dysphagia, swallowing

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(4) Gastrointestinal causes (mucosal, anatomical, and functional).
(5) Maladaptive feeding behaviors [4].

Dysphagia is not a disease, but rather a symptom that results from an underlying etiology or cause. Swallowing impairments are frequently classified in the clinical setting on the basis of three factors:

(1) The extent of oral, pharyngeal, or esophageal phase deficits.
(2) The underlying pathology or cause.
(3) The severity of the underlying disorder.

In children, the purpose of swallowing assessment and treatment is designed to prevent the development of more costly medical complications, including malnutrition and pneumonia. Identifying strategies to keep the child orally fed and able to maintain safe and adequate oral nutrition and hydration can achieve major cost saving in the healthcare system.

A fiberoptic endoscopic evaluation of swallowing (FEES) was described in 1988 by Langmore and colleagues as a procedure to assess oropharyngeal dysphagia. Initially, it was presented as an alternative to the fluoroscopy procedure, when it was not available. Over the years, endoscopy has become a standard tool for diagnosing and treating dysphagia [5]. Phoniatricians and speech–language pathologists use endoscopy to diagnose dysphagia in children, to assess the nature of the problem, and to guide its dietary and behavioral management.

**Indications of pediatric fiberoptic endoscopic evaluation of swallowing examination [5]**

1. Patient has never eaten orally, or has been maintained on enteral feedings for an extended period and the need to assess readiness for oral feeding.
2. Patient has a known or suspected structural abnormality of the pharynx or the larynx.
3. Patient is a surgical candidate for reconstruction of structural abnormality involving the oral cavity, pharynx, larynx, trachea, or esophagus.
4. Previous abnormal video fluoroscopic swallowing study and the need to assess the airway protection further and formulate strategies to improve safety of the swallowing.
5. Suspected difficulty with secretion management.
6. Patient cannot be transferred to radiology for videofluoroscopy.
7. Patient cannot be positioned adequately for fluoroscopic viewing.

Epidemiologic data on the incidence and prevalence of feeding and swallowing disorders in pediatric populations are limited. However, the prevalence of feeding problems in typically developing children has been estimated to be in the range of 25–40% and significantly higher in children with developmental delays [6]. The incidence of dysphagia is unknown; however, there is a general agreement that the incidence of swallowing dysfunction is increasing [7,8].

Langmore [5] established her protocol for dysphagia assessment through FEES examination. This protocol may help in discovering the signs of dysphagia in children and which of them has the most important role in inducing difficulty in swallowing in those children.

**Aim of the work**

The aim of this study was to clarify the role of FEES in the diagnosis of pediatric dysphagia and the signs related to it.

**Participants and methods**

This retrospective study assessed the signs of dysphagia in children. The study included 64 children (38 male, 26 female) from the Unit of Phoniatrics, Ain Shams University Hospitals. Of them, 32 were suffering from difficulty in swallowing of different degrees and 32 were controls (they were not suffering from any difficulty in swallowing). The mean age in months for symptomatic children was 41.47 ± 36.25 and the mean age in months for controls was 42.08 ± 35.61. All candidates were recruited from Ain Shams University Hospitals from March 2012 until August 2014 after obtaining consent from their parents. All candidates were evaluated according to Ain Shams Phoniatric Unit’s protocol for dysphagia assessment. All participants were subjected to examination using FEES applying the standard FEES protocol of Langmore (2001).

The standard FEES protocol of Langmore (2001) consists of two parts:

1. Part I consists of assessment of the anatomy and the sensorimotor function of the base of the tongue and the pharyngeal and the laryngeal structure. Parameters such as range or amplitude, symmetry, and briskness of movement can be assessed through specific tasks given to the patient, and the following could be observed: velopharyngeal closure, appearance of the hypopharynx and the larynx at rest, handling of secretions, the base of the tongue, the pharyngeal function, the laryngeal function, and sensory testing.
Part II involves the delivery of food and liquid to directly assess the swallowing of these materials. Throughout part II FEES examination, the examiner should comment on the following: the oral preparatory stage, the oral transit time, timing of the bolus flow/initiation of the swallow, adequacy of structural movements during the swallow, penetration/aspiration before and at the onset of the swallow, residue/aspiration after the swallow and between swallows, hypopharyngeal reflux, and sensations.

The assessment was performed by two expert phoniatricians and the results were obtained. Thus, we can compare children with symptoms of dysphagia and control candidates according to the signs of the standard FEES protocol mentioned before.

Score calculation
(1) A score for part I assessment was calculated by adding up values of the following variables: velopharyngeal closure, appearance of the hypopharynx and the larynx at rest, handling of secretions, the base of the tongue, the pharyngeal function, the laryngeal function, and sensory testing. Each variable was given a value of 1 for normal finding and 0 for abnormal finding, yielding a total score of 7.

(2) A score for part II assessment was calculated by adding up values of the following variables: the oral preparatory stage, the oral transit time, timing of the bolus flow/initiation of the swallow, adequacy of structural movements during the swallow, penetration/aspiration before and at the onset of the swallow, residue/aspiration after the swallow and between swallows, hypopharyngeal reflux, and sensations. Each variable was given a value of 1 for normal finding and 0 for abnormal finding, yielding a total score of 10.

(3) Scores were then used for correlations.

Results
Both cases and controls were matched as regards sex \( (P = 0.611) \) and age \( (P = 0.846) \). There was no significant difference between cases and controls as regards sex and age (Table 1).

Results as regards sex
(1) There was no significant difference between the number of male patients among cases (20; 62.5%) and those among controls (18; 56.25%).

(2) There was no significant difference between the number of female patients among cases (12; 37.5%) and those among controls (14; 43.75%).

The correlation of the mean age in months between cases \( (41.19 \pm 41.79) \) and controls \( (49.19 \pm 49.53) \) showed no significant difference.

Table 2 shows a significant difference between cases and controls for all signs of part I.

The results indicated that cases had marked signs, which were prominent and impaired the normal swallowing process:

(1) Velopharyngeal closure was impaired in eight (25%) cases and only in two (6.25%) controls.

Results as regards age

Table 1 Comparison between cases and controls as regards sex and age

| Distribution of candidates | Cases [N (%)] | Controls [N (%)] | \( P \) value \( ^a \) |
|---------------------------|--------------|-----------------|------------------|
| Sex                       |              |                 |                  |
| Male                      | 20 (62.5)    | 18 (56.25)      | 0.611*           |
| Female                    | 12 (37.5)    | 14 (43.75)      |                  |
| Age (months)              |              |                 |                  |
| Median (IQR)              | 25 (11–61.5) | 24 (10–89.5)    | 0.846*           |
| Mean ± SD                 | 41.19 ± 41.79| 49.19 ± 49.53   |                  |
| Range                     | 2–168        | 2–167           |                  |

IQR, interquartile range (first quartile–third quartile). \( ^a \)The value is calculated using the \( F^2 \)-test, \( ^b \)The value is calculated using the Mann–Whitney U-test.

Table 2 Comparison between cases and controls as regards part I of the assessment

| Items or signs of part I | Cases [N (%)] | Controls [N (%)] | \( P \) value \( ^a \) |
|-------------------------|--------------|-----------------|------------------|
| Velopharyngeal closure  |              |                 |                  |
| Poor                    | 8 (25)       | 2 (6.25)        | 0.039            |
| Good                    | 24 (75)      | 30 (93.75)      |                  |
| Appearance of the       |              |                 |                  |
| hypopharynx and the     |              |                 |                  |
| larynx at rest          |              |                 |                  |
| Laryngomalacia          | 16 (50)      | 3 (9.375)       | <0.0001          |
| Normal                  | 16 (50)      | 29 (90.625)     |                  |
| Handling of secretions  |              |                 |                  |
| Bad                     | 29 (90.625)  | 0 (0)           | <0.0001          |
| Good                    | 3 (9.375)    | 32 (100)        |                  |
| Base of the tongue      |              |                 |                  |
| Weak                    | 9 (28.125)   | 0 (0)           | 0.002*           |
| Good                    | 23 (71.875)  | 32 (100)        |                  |
| Pharyngeal function     |              |                 |                  |
| Weak                    | 30 (93.75)   | 0 (0)           | <0.0001          |
| Normal                  | 2 (6.25)     | 32 (100)        |                  |
| Laryngeal function      |              |                 |                  |
| Weak                    | 12 (37.5)    | 0 (0)           | <0.0001          |
| Normal                  | 20 (62.5)    | 32 (100)        |                  |
| Sensory testing         |              |                 |                  |
| Weak                    | 14 (43.75)   | 0 (0)           | <0.0001          |
| Normal                  | 18 (56.25)   | 32 (100)        |                  |

\( ^a \)The values are calculated using the \( \chi^2 \)-test, \( ^{\ast} \)The value is calculated using the Fisher’s exact test.
Appearance of the hypopharynx and the larynx in rest showed signs of laryngomalacia in 16 (50%) cases, whereas laryngomalacia was seen only in three (9.375%) controls.

Handling of secretions was bad in 29 (90.625%) cases, whereas all controls were handling secretions well.

The mobility of the base of the tongue was weak in nine (28.125%) cases, whereas all controls had a good mobility of the base of the tongue.

Pharyngeal function in swallowing was weak in 30 (93.75%) cases, whereas all controls had a normal pharyngeal function.

Laryngeal function was weak in 12 (37.5%) cases, whereas all controls showed normal laryngeal function.

Sensations were weak in 14 (43.75%) cases, whereas all controls showed normal sensations.

Table 3 shows a significant difference between cases and controls for all items of part II, except for the item of hypopharyngeal reflux, which showed no significant difference between cases and controls. The results showed that the cases had marked signs, which may be said to cause the pediatric dysphagia or worsen it:

(1) Oral preparatory stage was prolonged in nine (28.125%) cases, whereas all controls showed a within-normal duration of the oral preparatory stage.

(2) Oral transit time was prolonged in nine (28.125%) cases, whereas all controls showed within-normal oral transit time.

(3) Timing of the bolus flow/initiation of the swallow showed delay in 30 (93.75%) cases, whereas all controls showed within-normal timing of the bolus flow/initiation of the swallow.

(4) Structural movements during the swallow showed inadequacy in 26 (81.25%) cases, whereas all controls showed within-adequate structural movements during the swallow.

(5) Penetration before and at onset of swallow was present in 16 (50%) cases, whereas all controls showed no penetration before, nor at onset of the swallow.

(6) Aspiration before and at the onset of the swallow was present in 11 (34.375%) cases, whereas all controls showed no aspiration before, nor at the onset of the swallow.

(7) Residue after the swallow and between swallows was present in 30 (93.75%) cases, whereas only one (3.125%) control showed residue after the swallow and between swallows.

(8) Aspiration after the swallow and between swallows was present in 11 (34.375%) cases, whereas all controls showed no aspiration after the swallow, nor between swallows.

(9) Hypopharyngeal reflux was present in five (15.625%) cases, whereas one (3.125%) control showed hypopharyngeal reflux, with no significant difference between the two.

(10) Sensations were weak in 14 (43.75%) cases, whereas sensations were within normal in controls.

Table 4 shows that part I and part II assessment scores were significantly highly correlated among cases but not among controls. Age was not correlated with either part of assessment.

Results of cases as regards age

(1) Correlation between part I and part II was significant ($P < 0.001$).
(2) Correlation between part I and age was not significant ($P = 0.082$).

(3) Correlation between part II and age was not significant ($P = 0.278$).

Results of controls as regards age

(1) Correlation between part I and part II was not significant ($P = 0.674$).

(2) Correlation between part I and age was not significant ($P = 0.745$).

(3) Correlation between part II and age was not significant ($P = 0.341$).

Table 4 shows that sex was not correlated with either part of assessment. Thus, sex does not affect the assessment findings, neither in cases nor in controls.

Results of cases as regards sex

(1) Correlation of part I between male and female patients was not significant ($P = 0.886$).

(2) Correlation of part II between male and female patients was not significant ($P = 0.569$).

Results of controls as regards sex

(1) Correlation of part I between male and female patients was not significant ($P = 0.442$).

(2) Correlation of part II between male and female patients was not significant ($P = 0.387$).

Table 4 Correlation between part I and part II assessment scores and age

|          | Part II | Age |
|----------|---------|-----|
|          | Correlation coefficient | $P$ value | Correlation coefficient | $P$ value |
| Cases    |         |     |         |                        |           |
| Part I   | 0.864   | 0.000 | 0.312   | 0.082                  |
| Part II  |         |     | 0.198   | 0.278                  |
| Controls |         |     |         |                        |           |
| Part I   | -0.077  | 0.674 | 0.06    | 0.745                  |
| Part II  |         |     | 0.174   | 0.341                  |

*Pearson correlation coefficient.

Table 5 Association between part I and part II assessment scores and sex

|          | Male (mean ± SD) | Female (mean ± SD) | $P$ value* |
|----------|------------------|---------------------|------------|
| Cases    |                  |                     |            |
| Part I   | 3.35 ± 1.69      | 3.25 ± 2.18         | 0.886      |
| Part II  | 4.75 ± 2.63      | 5.33 ± 2.99         | 0.569      |
| Controls |                  |                     |            |
| Part I   | 6.89 ± 0.32      | 6.79 ± 0.43         | 0.442      |
| Part II  | 8.89 ± 0.47      | 9 ± 0               | 0.387      |

*The values are calculated using the independent sample $t$-test.

Data management and analysis

The collected data were revised, coded, tabulated, and introduced to a PC using Statistical Package for Social Science (2001, SPSS 15.0 for Windows; SPSS Inc., Chicago, Illinois, USA). Data were presented and suitable analysis was carried out according to the type of data obtained for each parameter.

Descriptive statistics

(1) Mean ± SD.

(2) Minimum and maximum values (range) for numerical data.

(3) Frequency and percentage of non-numerical data.

Analytical statistics

(1) The independent-samples: A $t$-test was used to assess the statistical significance of the difference between the two study group means.

(2) The $\chi^2$-test was used to examine the relationship between two qualitative variables.

$P$ value: level of significance.

A $P$ value more than 0.05 was considered nonsignificant (NS).

A $P$ value less than 0.05 was considered significant (S).

A $P$ value less than 0.01 was considered highly significant (HS).

Discussion

In this study we used the FEES protocol [5] on pediatric patients with dysphagia and on controls. In part I of the assessment we found that the most common signs found in most of the pediatric patients with dysphagia were as follows:

(1) Weak pharyngeal function in 93.7% of patients, which indicates weak pharyngeal peristalsis and consequential difficulty in clearing the bolus. It was observed that the absence of pharyngeal wall contraction on gag is an indicative sign of pharyngeal dysphagia [9].

(2) Bad handling of secretions in 90.6% of patients. In a normal individual, oropharyngeal secretions are cleared from the hypopharynx by periodic spontaneous swallows throughout the day. In a dysphagic patient, the accumulation of these secretions is thought to reflect impairment in the efficiency of laryngopharyngeal clearance [10].
Among the factors contributing to a reduction in secretion clearance in a dysphagic patient would be a reduction in the frequency of spontaneous swallows, a weakness in the pharyngeal response during the swallow, or a combination of weakness, and decreased swallowing frequency. It was found that the accumulation of endoscopically visible oropharyngeal secretions located within the laryngeal vestibule was highly predictive of aspiration of food or liquid. As patients who show trouble in clearing oropharyngeal secretions from the laryngeal vestibule for whatever reason will also likely demonstrate the same trouble with food or liquid while swallowing [11].

Thus, we considered that these two signs are the most common ones in pediatric dysphagia.

Weak base of the tongue and laryngeal function did not occur in controls; however, they occurred in pediatric patients but in less number (28.1 and 37.5%, respectively).

The tongue is used not only for moving the bolus about and forming a cohesive mass but also to sequentially press the bolus through the oropharynx and into the upper esophageal segment. Thus, weakness of the base of the tongue results in food sticking in the pharynx and increased incidence of coughing [12,13]. It was reported that, in conjunction with palatal weakness, tongue weakness was strongly correlated with dysphagia [14].

In case of affection of the laryngeal function, failure of the supraglottic and glottis laryngeal valve closure results in leakage through the supraglottis and glottis during swallow and in decreased ability to generate adequate hypopharyngeal pressures to propel the bolus through the pharyngeoesophageal segment and into the esophagus. Moreover, incomplete or absent laryngeal elevation may contribute to incomplete hypopharyngeal clearing [15]. Clinical measures for laryngeal function in case of dysphagia showed compelling results. It was observed that the clinical sign of 'dysphonia' was correlated with dysphagia after stroke [16]. It has been reported as one of the six signs, two or more of which indicate a problem, in the detection of aspiration [17] and more severe outcomes with dysphagia [18]. Most studies investigating the clinical signs of dysphagia cite a rating of vocal quality as useful [19].

This indicates that these two signs also contribute to dysphagia but they are not common signs in pediatric dysphagia.

Poor velopharyngeal closure was seen in 25% of patients and signs of laryngomalacia were seen in 50% of patients. Although few numbers of controls showed these two signs, there was a significant difference between cases and controls. In case of the velopharyngeal incompetence, dysphagia is explained by the inability to create appropriate negative introral pressure during suckling in infants. However, in older children, dysphagia is explained by leakage of the bolus into nasopharynx and diminished ability to generate appropriate oropharyngeal pressure to propel the bolus through the oropharynx.

Although among structural signs, laryngomalacia is the most prominent, we assumed in this study that its presence was as a cause but not a sign of dysphagia. As laryngomalacia is primarily a respiratory disorder, because of airway obstruction, patients had feeding difficulty due to poor coordination of the suck-swallow-breathe sequence. The feeding difficulty in pediatric patients can lead to failure to thrive and even aspiration, which is a sign of dysphagia. A 5-year retrospective study [20] on 50 consecutive infants with severe laryngomalacia who were assessed with FEES found that 44 infants (80%) showed laryngeal penetration and 36 infants (72%) showed laryngeal penetration with aspiration. Those infants underwent supraglottoplasty and postoperative FEES indicated resolution of laryngeal penetration and aspiration in 36 infants (72%) and 31 (86.1%) patients, respectively. Other studies also showed that supraglottoplasty in case of severe laryngomalacia opened the airway in more than 80% of patients, improved swallowing, and decreased aspiration risk in carefully selected patients who otherwise would not be at risk for aspiration [21,22].

In part II of the assessment, we found that the most frequent signs were as follows: the delayed timing of the bolus flow/initiation of the swallow (93.7%), the presence of residue after the swallow and between the swallows (93.7%), and the inadequacy of the structural movements during the swallow (81.2%). Thus, these signs highly contribute to pediatric dysphagia. However, the prolonged oral preparatory stage was seen in 28.1% of the patients and prolonged oral transit time in 28.1% of patients. Thus, the affection in the pharyngeal phase of swallowing is more common than the affection in the oral phase of swallowing in pediatric dysphagia.

Aspiration may occur before, during, and after swallowing. The time sequence may have clinical importance in predicting the etiology. When aspiration occurs before a swallow, a delay in pharyngeal swallow initiation or abnormal tongue movements may play a role [23]. In this study, the rate of patients with delayed timing of the bolus flow/initiation of the swallow was
93.7%, with 34.3% showing penetration before and at the onset of swallow and 34.3% of patients with aspiration before and at the onset of swallow. A delayed swallow is serious, as if it is too long, penetration or aspiration will occur [24]. The bolus spills into the laryngeal vestibule before it has closed off for the swallow. Moreover, one of the justifications for that is the weak sensations, which was detected in 43.7% of the assessed patients. It may also be related to observed alterations in the oral phase [25]. It was found that, in 125 children with dysphagia, 60% had laryngeal penetration and 31% had deep laryngeal penetration (residue left on the true vocal folds) [26]. When deep laryngeal penetration occurred, 85% had associated aspiration. Aspiration that occurs before the swallow or at the onset of swallow is reportedly one of the most frequent times for aspiration to be seen [27]. A study conducted on 469 adult patients showed that aspiration before the swallow was the second most frequent time for aspiration to occur, accounting for about 25% of all aspiration events [28].

Aspiration during a swallow indicates ineffective laryngeal closure or timing in coordination [23]. However, we could not assess aspiration during swallowing using FEES (swallow white-out), but in this study 37.5% of patients had weak laryngeal function and this may be an indicator for aspiration during swallowing.

Aspiration after swallow is from residue in the larynx and the hypopharynx being drawn in the larynx with the next breath [23]. In our study, 93.7% of patients had residue after the swallow and between the swallows, and 34.3% of patients showed aspiration after and between the swallows. The presence of residue may be associated with alterations in the oral phase and delay in initiation of the swallow reflex, in agreement with other studies [29,24]. Aspiration after the swallow is the most frequent time for this event to occur according to a study conducted on 469 adult dysphagic patients; aspiration after the swallow accounted for about 65% of all events of aspiration [28].

However, in this study we found that there is no specific time at which aspiration occurs more frequently in pediatric dysphagia.

The source of this pooling may be from the oral cavity or emerged from the esophagus through a relaxed upper esophageal sphincter (UES). Whether the material came from the esophagus or the stomach would not be known; hence, the term hypopharyngeal reflux is used [30].

A study conducted on 28 patients aged from 1 to 32 months with evidence of gastro-esophageal reflux disorder (GERD), as well as evidence of dysphagia with aspiration or hypopharyngeal pooling by FEES. Each child underwent either medical or surgical intervention to control GERD. A significant improvement in swallow function after GERD treatment was found. GERD may lead to decreased laryngopharyngeal sensitivity, which may contribute to swallowing dysfunction [31]. It was also found that the incidence of oropharyngeal dysphagia is prominent in infants with GERD-like symptoms [32].

We also found in this study that neither the age nor the sex has any influence on the two parts of assessment in both cases and controls. Thus, these findings are only related to dysphagia and neither the age nor the sex of the patients affects our findings.

**Conclusion and recommendation**

We found that there are more common signs related strongly to pediatric dysphagia than others, such as handling of secretions, pharyngeal function, timing of the bolus flow/initiation of the swallow, structural movements during the swallow, and residue after the swallow and between swallows. They need a tailored program to overcome these signs. Laryngomalacia should be suspected as a common cause in pediatric dysphagia. We recommend applying this protocol on a larger group of dysphagic children to reveal the common signs related to each disorder accused of pediatric dysphagia.

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**Conflicts of interest**

There are no conflicts of interest.

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