Development of a Pediatrics Dysphagia Screening Questionnaire

Original Article

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ABSTRACT

Purpose: This study aimed to develop a non-invasive marker to screen for Pediatric Oropharyngeal Dysphagia among susceptible groups," The Pediatric Dysphagia Screening Questionnaire (PDSQ), "and compare its efficacy to clinical and instrumental assessments.

Patients and Methods: We applied the 12-item "yes/no" Pediatric Dysphagia Screening Questionnaire (PDSQ) to a study set of 60 children from Oropharyngeal Dysphagia susceptible groups, such as children with neurodevelopmental disorders, Down syndrome, underweight of unknown etiology, and submucous cleft. We also administered the questionnaire to 60 healthy controls with the same age range and sex distribution. In addition, we validated the questionnaire responses to clinical feeding-swallowing observation outcomes supported by clinical consensus in Cases and Controls. Finally, we performed a Fiberoptic Endoscopic Evaluation of Swallowing (FEES) study on the dysphagia-susceptible group.

Results: There was a statistically significant difference in the questionnaire scores between the cases and controls. In our Dysphagia susceptible group of patients, the PDSQ showed a cutoff score of 3 predicted Oropharyngeal Dysphagia diagnosed with clinical feeding observation with 97.9% sensitivity and 100% specificity, a cutoff score of 7 predicted penetration with 83.3% sensitivity and 80.6% specificity, and a cutoff score of 9 predicted aspiration with 84.2% sensitivity and 85.4% specificity.

Conclusion: The PDSQ showed acceptable Sensitivity and Specificity, indicating its validity as a screening tool for pediatric Oropharyngeal Dysphagia.

Key Words: Aspiration, dysphagia, pediatrics, penetration, screening.

Received: 1 August 2023, **Accepted:** 29 August 2023

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ISSN: 2090-0740, 2023

INTRODUCTION

Dysphagia is a difficulty in swallowing that involves impairment, specifically in the safety and efficiency of swallowing[1]. Swallowing safety involves the transfer of food or liquid from the oral cavity to the stomach without entering the airway (penetration/aspiration). Swallowing efficiency is the ability to move a bolus of food or liquid through the oropharynx without leaving any residue in the digestive tract. When residue is present in the pharynx after swallowing, there is a danger of post-swallowing aspiration during the inspiratory phase of respiration[2].

The diagnosis of Dysphagia also expands to include mechanical disorders of oral food intake, which comprise behavioral, sensory, and motor disorders in preparation for swallowing, for example, disorders of cognitive awareness, visual and olfactory recognition of food, and physiological responses to the smell and presence of food[3]. The execution of a safe, effective, and efficient swallowing throughout human growth from infancy to late childhood depends strongly on intricate sensory development, efficient motor coordination of the swallowing muscles, and maturation of feeding skills to ensure airway protection and full bolus clearance from the oropharyngeal segment[4]. This dependency makes children with developmental delays particularly vulnerable to the disorder and all possible comorbidities. Approximately 1% of children in the general population experience swallowing difficulties[5]; the incidence rate is much higher in some clinical disorders, such as cerebral palsy, traumatic brain injury, and airway malformations. Ancel, Livinec & Larroque, 2006 noted an increase in swallowing dysfunction when premature infants are born early compared with later-born premature infants[6]. Moreover, the incidence of swallowing dysfunction is increasing, possibly due to improved survival rates of children with a history of prematurity (birth at <37 weeks gestation), low birth weight, and complex medical conditions[7]. Pediatric Dysphagia is a severe problem that may lead to slow weight

 DOI: 10.21608/ejentas.2023.226365.1661

gain, disrupted nutrition, and cause acute choking that could lead to life-threatening aspiration-based infections[8].

In oropharyngeal Dysphagia, the oral preparatory, voluntary, and pharyngeal phases of swallowing are affected. If the esophageal phase is disturbed, we categorize Dysphagia as esophageal Dysphagia. All stages of swallowing physiology may influence each other; therefore, diagnosing Dysphagia requires a comprehensive evaluation of the aerodigestive tract from the oral cavity to the stomach[9]. Such a thorough clinical assessment is time-consuming and requires the dedication of resources and personnel. There are a variety of instrumental tools that assist clinicians, but they are also labor-demanding. This situation has brought about the need for screening tools to support clinicians in rationalizing their decision-making and efficiently managing caseloads.

The gold standard for diagnosing Dysphagia is the fiberoptic endoscopic evaluation of swallowing (FEES) and modified barium swallowing studies[10,11]. Various screening tools are available to assist clinicians in determining the need for these diagnostic tools. Most available screening tools offer assistance to adult cohorts of patients. They vary from the questionnaire to the drinking task. A few earlier studies have attempted to develop Pediatric Dysphagia screening tools. The PEDI-EAT was developed by Thoyre, Pados & Park in 2014[12]; The Behavioral Pediatrics Feeding Assessment Scale in Young Children with Autism by Allen, Smith, & Duku in 2015[13]; The Children's Eating Behavior Inventory: Reliability and Validity Results in 1991 Archer, Rosenbaum & Streiner;[14] and Development of the Children's Eating Behavior Questionnaire by Wardle, Guthrie & Sanderson in 2001[15]. Earlier studies did not validate these tools against either a dysphagia-specific instrumental assessment or a clinical evaluation of the disorder. Some are clinically population-specific, while others are lengthy in content and would not be suitable for rapid clinical screening. In 2018, Arslan, Demir, Karaduman, & Belafsky published the PEDI-EAT-10 questionnaire for pediatric dysphagia screening; they validated the tool against clinical videofluoroscopy in a subject population of children with Cerebral palsy[16]. The earlier questionnaire was rich in questions exploring aspects of the physiology reflective of the pharyngeal phase of swallowing and swallowing outcome-related components with limited attention to the oral stage, which is also commonly disordered in these patients.

The authors believe clinicians need more choices to address the requirements of a diverse array of pediatric patients. An ideal screening tool would include assessing the pediatric population's oral and pharyngeal aspects of swallowing. The distinction of affected phases could only occur after the screening process flags a potentially diseased patient and during standardized assessments. The validation of an ideal tool will be against instrumental evaluations. The time lapse between screening and validation should be minimal. Finally, the tool application must be easy and rapid.

Assessing children's feeding problems with a standardized questionnaire is a useful clinical option because feeding problems coexist and can be undetectable with a broad spectrum of pediatric diseases. This study aimed to detect Oropharyngeal Dysphagia in children using a questionnaire to allow early intervention, rehabilitation, and prevention of complications, which may be life-threatening. To establish this tool in this preliminary phase, we elected to make the study inclusive of variable cohorts of high-incidence patients to make the tool applicable as a broad-spectrum option. Future work will follow the path of PEDI-EAT-10 and focus on more homogenous diagnostic entities.

PATIENTS AND METHODS:

This study received approval from the local institutional research ethics board.

Participants

This study initially included a dysphagia susceptible group of 60 children referred to the phoniatrics units of Cairo University and Fayoum University. We originally intended to recruit children aged 2 to 18; however, a post-recruitment demographic analysis showed that the subjects were 24 females and 36 males aged 2 to 14. The referring clinicians diagnosed all study group subjects with a primary disorder identified by the researchers as at an additional risk of dysfunctional swallowing. However, the primary clinicians referred those patients to the phoniatrics clinic for Language, Speech, Voice, or resonance assessments; the only exception was underweight children – pediatricians referred those patients for a swallowing assessment. All participants were asymptomatic regarding their physiological swallowing function to meet the inclusion criteria. The diagnostic categories were clinically significant underweight children with no apparent cause; children with neuromuscular conditions, such as cerebral palsy, myopathy, and Down syndrome; children with anatomical abnormalities of the oropharynx, larynx, trachea, esophagus, and congenital anomalies associated with orofacial deficits (e.g., Pierre Robin Syndrome); and cases with a submucous cleft. Although some clinicians believe submucous cleft patients rarely present with Velopharyngeal Incompetence or Dysphagia, earlier work in 1990 by Moss, Jones, and Pigott recommended including a submucous cleft in the differential diagnosis of pediatric Dysphagia[17]. Supported by this, the research team included this disorder in the study population. Intentionally, the study excluded children presenting with a previously confirmed diagnosis of Dysphagia or specifically seeking medical advice for a complaint of difficulty in eating and

swallowing. Additionally, underweight children with a specific etiology, such as marasmus, were excluded.

We also recruited a healthy post hoc control group of 24 females and 36 males aged 2-14 from the Fayoum University Pediatric outpatient clinic. In addition, we interviewed the participants' caregivers to screen for possible swallowing abnormalities or any of the conditions included in the dysphagia-susceptible cohort; healthy subjects were either coming to the clinic for regular vaccinations or siblings accompanying a case seeking medical advice.

Protocol

The initial step after parental consent was general history taking from parents/caretakers to monitor eligibility for inclusion; following this was a thorough feeding history guided by Arvedson, Brodsky, & Reigstad, 2002[18]. The next step was administering the Pediatric Dysphagia Screening Questionnaire (PDSQ). Finally, we followed the previous two stages with a clinical feeding assessment in both groups. However, due to its invasive nature, only the subjects in the Dysphagia susceptible group underwent Fiberoptic Endoscopic Evaluation of Swallowing (FEES). We implemented all the protocol steps on the same day for each participant. The clinician's history and delivery of the questionnaire items differed from the clinicians performing the clinical feeding assessment and FEES.

Similarly, the clinician performing FEES differed from those who conducted the clinical feeding examination. The study design blinded all the clinicians when completing their tests on the questionnaire results. Two clinicians at each site implemented all evaluations and agreed with the consensus on every diagnostic criterion in clinical feeding observation. The administered food for clinical feeding evaluation was broad regarding variability across participants according to what food the caretaker gave at home, varying from milk in severely affected children to meat and bread in milder cases. In the FEES examination, we tailored bolus administration according to the level of risk and clinical progress across the study. We started with smaller milk volumes and gradually increased them according to child tolerance; for semisolids, we started with half a teaspoon of yogurt and increased progressively according to safety and efficiency. In cooperative tolerating children, we provided a biscuit to represent harder consistencies. The post hoc blinded intra-rater reliability for FEES rating penetration and/or aspiration was ICC= 1.0.

The screening questionnaire PDSQ:

It consists of 12 screening questions guided by a comprehensive review of earlier literature and adult screening tools: the EAT-10, swallowing disturbance questionnaire, self-report symptom inventory, and Sydney Swallow Questionnaire.[19,20,21,22] We selected the questions by expert consensus from a list of 10 items collected from every research team member; the team members were four phoniatrics consultants and a specialist, all with expertise in pediatric swallowing disorders. In the beginning of the study, PDSQ consisted of 20 items then pilot study was conducted at 20 children. The most yield questions were selected, collected and some of them were merged to be summarized in only 12 items.

All the questions were yes or no (Table 1). Yes was marked as 1 and no as 0. Thus, the total score ranged from 0 to 12. Statistical analysis differentiated the numerical boundary that discriminated cases based on the presence or absence of Dysphagia.

Table 1: Yes or no Pediatric Dysphagia Screening Questionnaire PDSQ

Questions	Yes	No
1- Is it difficult to bite or chew?	1	0
2- Does the consistency of food affect greatly feeding and swallowing act?	1	0
3- Is it difficult to sip from a cup?	1	0
4- Is it difficult to use a straw?	1	0
5- Is there excessive and persistent drooling of saliva?	1	0
6- Is there frequent cough during or after swallowing?	1	0
7- Does feeding and swallowing take extra time (>30 min.)?	1	0
8- Is there a preferable posture facilitating feeding and swallowing?	1	0
9- Does eating/feeding cause embarrassment outwards?	1	0
10- Is it hard to anyone else to feed the child?	1	0
11- Is there loss of weight or failure to gain weight?	1	0
12- Is there frequent chest infection?	1	0
Total score	Out of 12	

Clinical evaluation of feeding and swallowing:

We examined the oral structures and functions before introducing food or liquids to the child. Next, we observed the parent-child interactions around feeding. Finally, we classified children showing abnormalities, guided by Arvedson *et al*., 2002[18]. The components of this evaluation are improper chewing, food stuck to the palate, the presence of abnormal suckling movement during feeding, an improper labial seal, anterior spillage, vomiting after feeding and nasal regurgitation of food or fluid.

Fiberoptic Endoscopic Evaluation of Swallowing (FEES)

This instrumental assessment was used to visualize the tongue base, nasopharynx, hypopharynx, and larynx. In addition, we evaluated the structure and function of the upper aerodigestive tract at rest and work and classified cases showing penetration and/or aspiration as cases of Dysphagia[23]. We Included FEES in our assessment for all participants in the dysphagia susceptible group, supported by the possibility of occult Oropharyngeal Dysphagia in a child that is not visible during clinical evaluation of feeding and swallowing. Neurologically-based Dysphagia in the Pediatric population is commonly associated with silent aspiration[24].

ANALYSIS

The collected data were organized, tabulated, and statistically analyzed using the Statistical Package of Social Science (SPSS) software statistical computer package version 22 (SPSS Inc., U.S.A.). First, we calculated the mean and standard deviation (S.D.) for quantitative data. We performed an independent t-test to determine the significance of the differences in questionnaire scores between cases and controls. We used a one-way ANOVA and Tukey post hoc analysis of the mean scores of the questionnaire in patients classified as oral-motor and oropharyngeal by clinical observation. Qualitative data were presented as numbers and percentages, and the chi-square (χ^2) test was used. Third, we used the receiver operating characteristic (R.O.C.) curve to determine the discrimination value of the total questionnaire score for Dysphagia, penetration, and aspiration and to define optimal cutoff points for sensitivity and specificity. Finally, we used Cohen's kappa (κ) to determine the concordance between FEES and clinical observations in detecting Dysphagia. For interpretation of the results of the tests of significance, $P \leq 0.05$ was significant.

RESULTS:

The Dysphagia susceptible group consisted of 60 children with different disorders associated with the possible presence of Dysphagia: 65% had neurological lesions, 11.7% had non-specifically diagnosed underweight, 10% had a submucous cleft, 8.3% had Down syndrome, and 5% had congenital anomalies with an oral-facial deficit (Table 2). The Mean questionnaire score in this group of patients was 6.6, significantly higher than the mean of 0.3 scores in the control group (Table 3).

Table 2: Percentage of each disorder among the Dysphagia susceptible cohort

Type of disorder	No.	%
Neurological disorders (Cerebral palsy & Duchenne Muscular Dystrophy	39	65.0%
Underweight of unidentified cause	7	11.7%
Submucous cleft	6	10%
Down's syndrome	5	8.3%
Congenital anomalies with oral facial Dysmorphia (Pierre Robin Syndrome)	3	5.0%

Table 3: Mean and Standard deviation (SD) questionnaire scores in Dysphagia susceptible participants and Healthy controls

	Dysphagia susceptible participants	Healthy controls	*P value*
PDSQ Score	6.6(4)	0.3(0.1)	$< 0.001^*$

Independent Samples t-test ($^*P < 0.05$)

Clinical observations of feeding and swallowing showed that 49 participants in the Dysphagia susceptible group had signs suggesting the presence of Dysphagia. The most common were improper chewing (76.6%) and food stuck to the palate (60%). In addition, there was an abnormal suckling movement during feeding, an improper labial seal in 55% of the cases, and anterior spillage in 48.3%. The least common signs among the Dysphagia susceptible group were vomiting after feeding and nasal regurgitation of food or fluid (1.7%). (Fig. 1). All the control group participants showed no abnormalities in their clinical feeding observations.

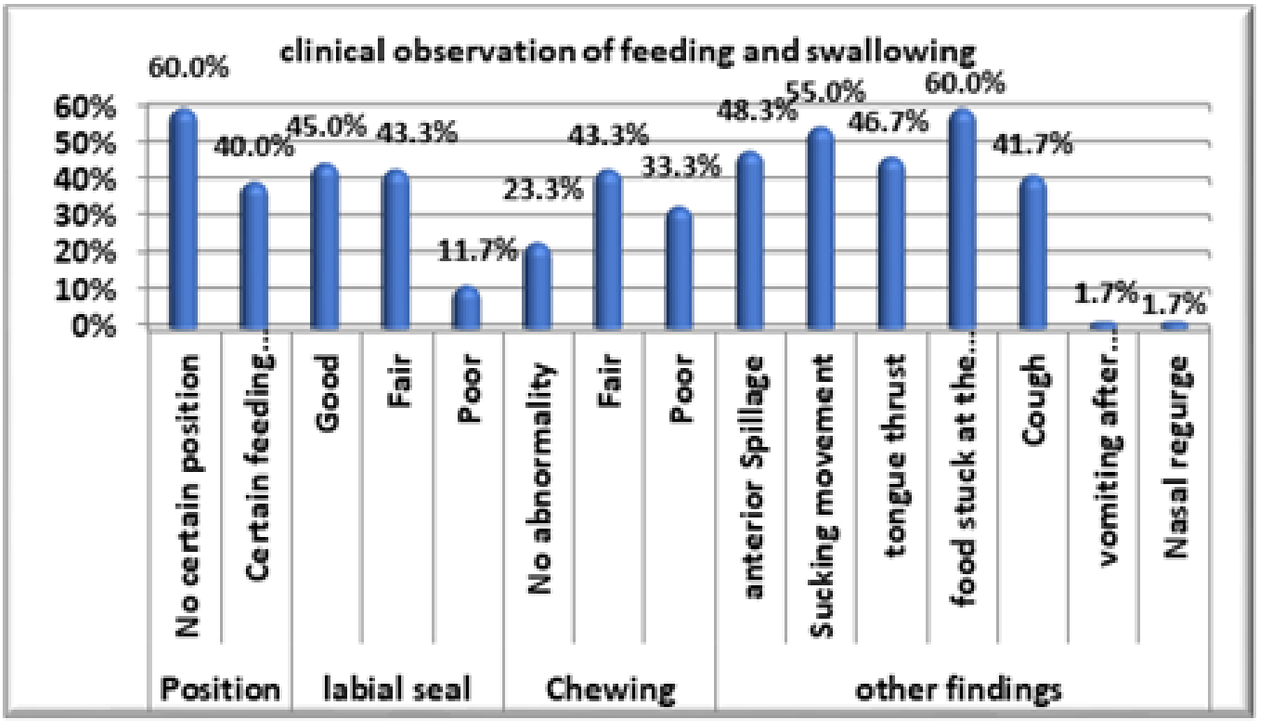

Fig. 1: Clinical observation of feeding and swallowing in the Dysphagia susceptible cohort

Fiber Optic Endoscopic Evaluation of Swallowing (FEES) shows that 27 participants had either penetration or aspiration, or both signs were present simultaneously. Other clinical observations during the examination showed that the Velo Pharyngeal Valve was competent in all dysphagia-susceptible cases except 1. Premature spillage was present in seven children, residue in the vallecula in six, delayed triggering in 15, and residue in the pyriform in 16. Four patients had GERD, one had laryngomalacia, and one had esophageal pooling. (Fig. 2)

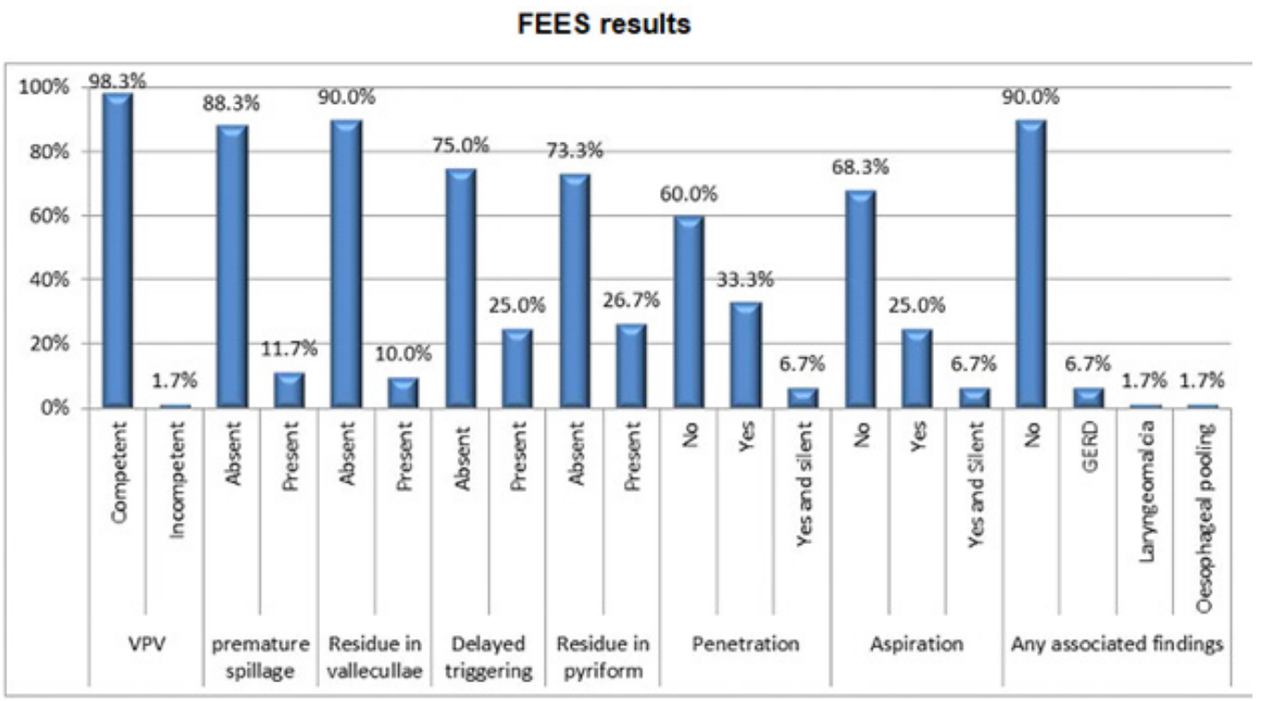

Fig. 2: FEES results in the Dysphagia susceptible cohort

Clinical observations and FEES in the Dysphagia susceptible cohort showed that 27/60 (45%) participants had Dysphagia and 11/60 (18.3%) had no dysphagia, with a total agreement of 38/60 (63.3%), Cohen's kappa (κ) = 0.310, and *P=0.001* (Table 4). Thus, all cases Diagnosed with Dysphagia in FEES manifest as clinical feeding observation, but not all instances of Dysphagia detected by clinical examination are diagnosed with FEES.

Table 4: Comparing Clinical observation and FEES results the Dysphagia susceptible cohort

		FEES		Cohen's kappa (κ)	*P-value*
		Dysphagia	No dysphagia		
Clinical observation	Dysphagia	27	22	0.310	*0.001 (S)*
	No dysphagia	0	11		

*statistical significance difference; *p-value <0.05*, Cohen's kappa (κ)

Table 5 shows the quantitative response "yes" to each pediatric Dysphagia Screening Questionnaire (PDSQ) item in susceptible cases and healthy controls. The percentages varied from 35% to 76.7% in the dysphagia-susceptible cohort. The highest rates were in swallowing abilities with different consistencies and weight loss or failure to gain at 76.7%, taking a long time to finish a meal followed by 68.3%, and difficulty biting and chewing at 65%. Among the 12 items, difficulty in sipping from a cup (35%) was uncommon. It is worth noting that the total yes score in the Dysphagia susceptible participants set was 404 compared to 19 in the healthy controls.

Table 5: Yes scored for each Question item in the Dysphagia susceptible participants and healthy controls

Yes scored for each Question item	Dysphagia Susceptible participants		Healthy Controls
Questions	N.	%	N.
1. Is it difficult to bite or chew?	39	65.0%	0
2. Does the consistency of food affect greatly feeding and swallowing act?	46	76.7%	0
3. Is it difficult to sip from a cup?	21	35.0%	0
4. Is it difficult to use a straw?	37	61.7%	1
5. Is there excessive and persistent drooling of saliva?	35	58.3%	1
6. Is there frequent coughing before, during or after swallowing?	29	48.3%	0
7. Does feeding and swallowing take extra time?	41	68.3%	4
8. Is there a certain preferable posture that can facilitate feeding and swallowing?	25	41.7%	0
9. Does eating/feeding cause embarrassment outwards?	28	46.7%	0
10. Is it hard to anyone else to feed the child?	26	43.3%	0
11. Is there loss of weight or failure to gain it?	46	76.7%	10
12. Is there frequent chest infections?	31	51.7%	0
Total Yes scored in each group	404		19

Comparing questionnaire answers in the dysphagia-susceptible cohort of patients classified as with or without Dysphagia using clinical observation showed a significant difference between Yes and No responses related to the presence or absence of Dysphagia. In every questionnaire item, the *p-value* was *<0.05*, and the higher percentage of yes was among the positive dysphagia patients. (Table 6). Additionally, questionnaire scores are significantly higher in patients with Dysphagia diagnosed with clinical observation, with a mean of 7.8 ± 3.2 compared to 1.1 ± 0.9 with no Dysphagia diagnosis using the same assessment technique. (Fig. 3).

Table 6: Comparison between questionnaire responses among Dysphagic and Non-Dysphagic subgroups diagnosed by clinical observation in the Dysphagia susceptible cohort

Questionnaire items		Clinical observation				*P-value*
		Dysphagic (n=49)		Non-Dysphagic (n=11)		
1- Is it difficult to bite or chew?	Yes	39	79.6%	0	0.0%	*<0.0001**
	No	10	20.4%	11	100.0%	
2- Does the consistency of food affect greatly feeding and swallowing act?	Yes	46	93.9%	0	0.0%	*<0.0001**
	No	3	6.1%	11	100.0%	
3- Is it difficult to sip from a cup?	Yes	21	42.9%	0	0.0%	*0.006**
	No	28	57.1%	11	100.0%	
4- Is it difficult to use a straw?	Yes	35	71.4%	2	18.2%	*0.002**
	No	14	28.6%	9	81.8%	
5- Is there excessive and persistent drooling of saliva?	Yes	33	67.3%	2	18.2%	*0.005**
	No	16	32.7%	9	81.8%	
6- Is there frequent coughing before, during or after swallowing?	Yes	29	59.2%	0	0.0%	*<0.0001**
	No	20	40.8%	11	100.0%	
7- Does feeding and swallowing take extra time?	Yes	38	77.6%	3	27.3%	*0.003**
	No	11	22.4%	8	72.7%	
8- Is there a certain preferable posture that can facilitate feeding and swallowing?	Yes	25	51.0%	0	0.0%	*0.002**
	No	24	49.0%	11	100.0%	
9- Does eating/feeding cause embarrassment outwards?	Yes	28	57.1%	0	0.0%	*0.001**
	No	21	42.9%	11	100.0%	
10- Is it hard to anyone else to feed the child?	Yes	26	53.1%	0	0.0%	*0.001**
	No	23	46.9%	11	100.0%	
11- Is there loss of weight or failure to gain it?	Yes	41	83.7%	5	45.5%	*0.014**
	No	8	16.3%	6	54.5%	
12- Is there frequent chest infections?	Yes	30	61.2%	1	9.1%	*0.002**
	No	19	38.8%	10	90.9%	
Total questionnaire score Mean ± SD		7.8 ± 3.2		1.1 ± 0.9		*0.0001*

Chi-Squared test Qualitative, Independent-t test Quantitative, *Statistically significant Difference; *p-value<0.05*

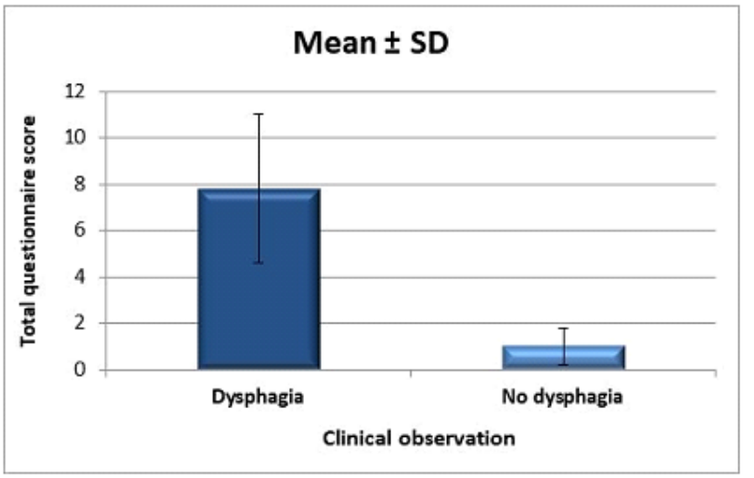

Fig. 3: Comparison between questionnaire responses among Dysphagic and Non-dysphagic subgroups diagnosed by clinical observation in the Dysphagia susceptible group

Table 7 shows that within the Dysphagia susceptible cohort, a significant difference between Yes and No responses in 11/12 questions related to the presence of Dysphagia diagnosed by FEES (penetration &/or Aspiration). Only " Is there a loss of weight or failure to gain it?" shows a *p-value >0.05* as the majority of +ve Dysphagia and –ve Dysphagia patients responded yes to that question (Table 7). In addition, the mean questionnaire score is significantly higher in patients with Dysphagia diagnosed with FEES, with a mean of 9.4 ± 2.6 compared to 4.2 ± 3.4 with no Dysphagia (Fig.4).

Table 7: Comparison between questionnaire responses among Dysphagic and Non-Dysphagic subgroups diagnosed by FEES (Penetration&/or Aspiration) in the Dysphagia susceptible cohort

Questionnaire items		FEES				*P-value*
		Dysphagic (n=27)		Non-Dysphagic (n=33)		
1- Is it difficult to bite or chew?	Yes	24	88.9%	15	45.5%	*<0.0001**
	No	3	11.1%	18	54.5%	
2- Does the consistency of food affect greatly feeding and swallowing?	Yes	26	96.3%	20	60.6%	*0.002**
	No	1	3.7%	13	39.4%	
3- Is it difficult to sip from a cup?	Yes	16	59.3%	5	15.2%	*<0.0001**
	No	11	40.7%	28	84.8%	
4- Is it difficult to use a straw?	Yes	22	81.5%	15	45.5%	*0.007**
	No	5	18.5%	18	54.5%	
5- Is there excessive and persistent drooling of saliva?	Yes	20	74.1%	15	45.5%	*0.036**
	No	7	25.9%	18	54.5%	
6- Is there frequent coughing before, during or after swallowing?	Yes	20	74.1%	9	27.3%	*0.001**
	No	7	25.9%	24	72.7%	
7- Does feeding and swallowing take extra time?	Yes	23	85.2%	18	54.5%	*0.013**
	No	4	14.8%	15	45.5%	
8- Is there a certain preferable posture that can facilitate feeding and swallowing?	Yes	20	74.1%	5	15.2%	*<0.0001**
	No	7	25.9%	28	14.8%	
9- Does eating/feeding cause embarrassment outwards?	Yes	20	74.1%	8	24.2%	*<0.0001**
	No	7	25.9%	25	75.8%	
10- Is it hard to anyone else to feed the child?	Yes	20	74.1%	6	18.2%	*<0.0001**
	No	7	25.9%	27	81.8%	
11- Is there loss of weight or failure to gain it?	Yes	21	77.8%	25	75.8%	*1*
	No	6	22.2%	8	24.2%	
12- Is there frequent chest infections?	Yes	22	81.5%	9	27.3%	*<0.0001**
	No	5	18.5%	24	72.7%	
Total questionnaire score	Mean ± SD	9.4 ± 2.6		4.27± 3.4		*<0.0001*

Chi-Squared test Qualitative, Independent t-test Quantitative, * Statistically Significant Difference; *p-value <0.05*

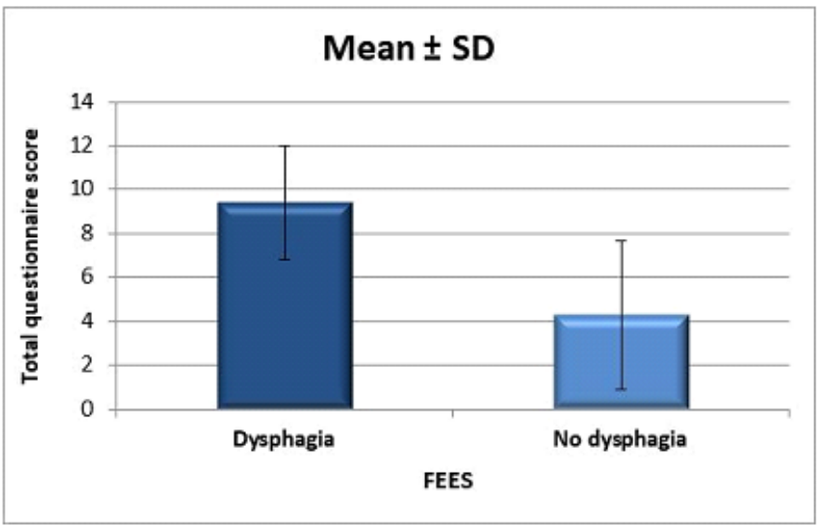

Fig. 4: Comparison between questionnaire responses among Dysphagic and Non-Dysphagic subgroups diagnosed by FEES

There was a significant difference in Yes and No responses to 11/12 questions with the presence or absence of penetration in the FEES examination within the dysphagia-susceptible cohort. However, only " Is there a loss of weight or failure to gain it" showed a *p-value >0.05*, as the majority of patients with positive Dysphagia and Dysphagia responded yes to that question (Table 8). Additionally, the mean questionnaire score is significantly higher in patients with penetration, with a mean of 9.5 ± 2.7 compared to 4.6 ± 3.5 with no penetration (Fig. 5).

Table 8: Comparison of PDSQ response in presence or absence of Penetration in FEES in the Dysphagia susceptible cohort

Questionnaire items		Penetration				*P-value*
		Present (n=24)		Absent (n=36)		
		N	%	N	%	
1. Is it difficult to bite or chew?	Yes	22	91.7%	17	47.2%	*<0.0001**
	No	2	8.2%	19	52.8%	
2. Does the consistency of food affect greatly feeding and swallowing?	Yes	23	95.8%	23	63.9%	*0.004**
	No	1	4.2%	13	36.1%	
3. Is it difficult to sip from a cup?	Yes	14	58.3%	7	19.4%	*0.002**
	No	10	41.7%	29	80.6%	
4. Is it difficult to use a straw?	Yes	20	83.3%	17	47.2%	*0.005**
	No	4	16.7%	19	52.8%	
5. Is there excessive and persistent drooling of saliva?	Yes	19	79.2%	16	44.4%	*0.008**
	No	5	20.8%	20	55.6%	
6. Is there frequent coughing before, during or after swallowing?	Yes	19	79.2%	10	27.8%	*<0.0001**
	No	5	20.8%	26	72.2%	
7. Does feeding and swallowing take extra time?	Yes	20	83.3%	21	58.3%	*0.041**
	No	4	16.7%	15	41.7%	
8. Is there a certain preferable posture that can facilitate feeding and swallowing?	Yes	19	79.2%	6	16.7%	*<0.0001**
	No	5	20.8%	30	83.3%	
9. Does eating/feeding cause embarrassment outwards?	Yes	17	70.8%	11	30.6%	*0.002**
	No	7	29.2%	25	69.4%	

10. Is it hard to anyone else to feed the child?	Yes	18	75.0%	8	22.2%	*<0.0001**
	No	6	25.0%	28	77.8%	
11. Is there loss of weight or failure to gain it?	Yes	18	75.0%	28	77.8%	*0.803 NS*
	No	6	25.0%	8	22.2%	
12. Is there frequent chest infections?	Yes	21	87.5%	10	27.8%	*<0.0001**
	No	3	12.5%	26	72.2%	
Total questionnaire score	Mean ± SD	9.5 ± 2.7		4.6 ± 3.5		*<0.0001**

Chi-Squared test Qualitative, Independent t-test Quantitative, * Statistically Significant Difference; *p-value <0.05*

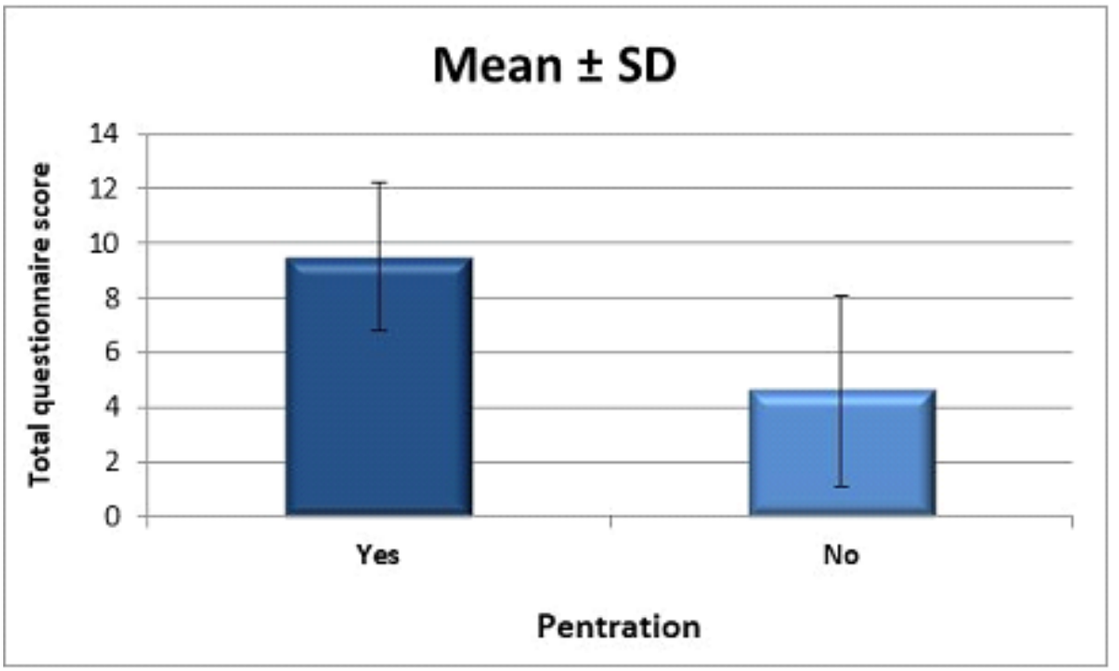

Fig. 5: Comparison between questionnaire responses in presence or absence of Penetration in FEES in the Dysphagia susceptible cohort

Within the Dysphagia susceptible cohort, there was a significant difference in Yes and No responses to 11/12 questions regarding the presence or absence of aspiration in the FEES examination. Only " Is there a loss of weight or failure to gain it" showed a *p-value >0.05*, as the majority of patients with positive Dysphagia and Dysphagia responded yes to that question (Table 9). Additionally, the mean questionnaire score is significantly higher in patients with aspiration, with a mean of 10.2 ± 2.3 compared to 4.9 ± 3.4 with no aspiration (Fig. 6).

Table 9: Comparison of PDSQ response in presence and absence of Aspiration in FEES in the Dysphagia susceptible cohort

Questionnaire items		Aspiration				*P-value*
		Present (n=19)		Absent (n=41)		
		N	%	N	%	
1- Is difficult to bite or chew?	Yes	19	100%	20	48.8%	*<0.0001**
	No	0	0.0%	21	51.2%	
2- Does the consistency of food affect greatly feeding and swallowing?	Yes	19	100.0%	27	65.9%	*0.003**
	No	0	0.0%	14	34.1%	
3- Is it difficult to sip from a cup?	Yes	14	73.7%	7	17.1%	*<0.0001**
	No	5	26.3%	34	82.9%	
4- Is it difficult to use a straw?	Yes	17	89.5%	20	48.8%	*0.003**
	No	2	10.5%	21	51.2%	

5-	Is there excessive and persistent drooling of saliva?	Yes	17	89.5%	18	43.9%	*0.001**
		No	2	10.5%	23	56.1%	
6-	Is there frequent coughing before, during or after swallowing?	Yes	16	84.2%	13	31.7	*<0.0001**
		No	3	15.8%	28	68.3%	
7-	Does feeding and swallowing take extra time?	Yes	17	89.5%	24	58.5%	*0.017**
		No	2	10.5%	17	41.5%	
8-	Is there a certain preferable posture that can facilitate feeding and swallowing?	Yes	16	84.2%	9	22.0%	*<0.0001**
		No	3	15.8%	32	78.0%	
9-	Does eating/feeding cause embarrassment outwards?	Yes	14	73.7%	14	34.1%	*0.004**
		No	5	26.3%	27	65.9%	
10-	Is it hard to anyone else to feed the child?	Yes	14	73.7%	12	29.3%	*0.001**
		No	5	26.3%	29	70.7%	
11-	Is there loss of weight or failure to gain it?	Yes	14	73.7%	32	78.0%	*0.749 NS*
		No	5	26.3%	9	22.0%	
12-	Is there frequent chest infections?	Yes	18	94.7%	13	31.7%	*<0.0001**
		No	1	5.3	28	68.3%	
Total questionnaire score		Mean ± SD	10.2 ± 2.3		4.9 ± 3.4		*<0.0001**

Chi-Squared test Qualitative, Independent t-test Quantitative, * Statistically Significant Difference; *p-value <0.05*

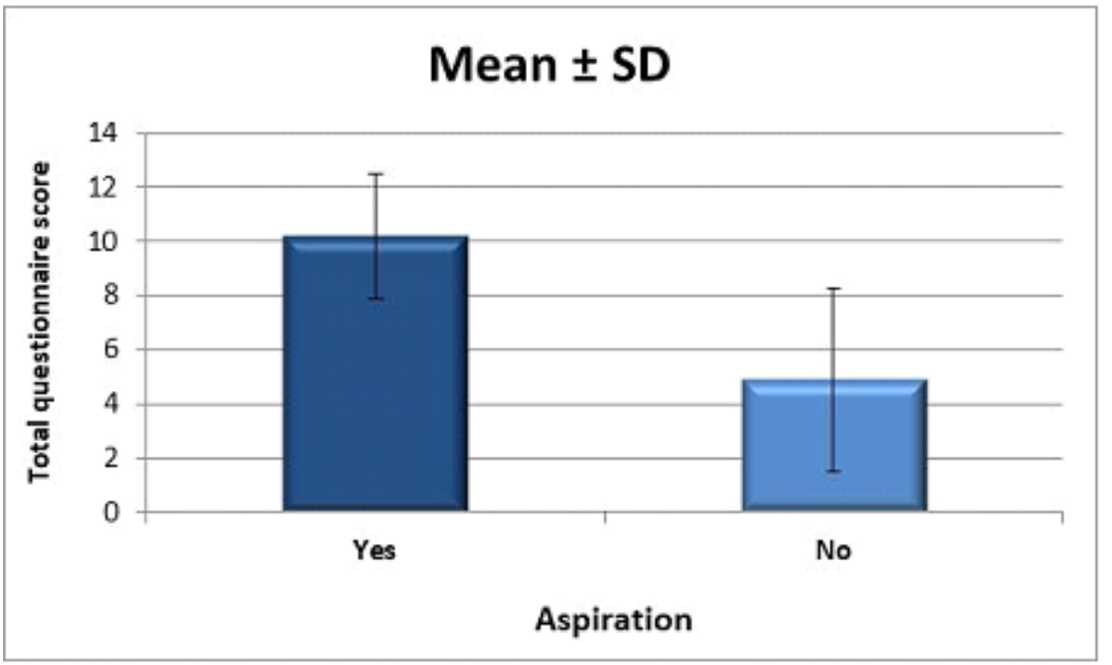

Fig. 6: Comparison between questionnaire responses in presence and absence of Aspiration in FEES in the Dysphagia susceptible cohort

We performed a Receiver Operating Characteristics (R.O.C.) curve analysis to determine the discrimination value of the total questionnaire scores within the Dysphagia susceptible cohort for Dysphagia diagnosed with clinical observation, penetration and/or aspiration, penetration only, and aspiration only as diagnosed by FEES. We plotted the cutoff points for different levels of sensitivity and specificity. (Figures 7, 8, 9, & 10). A cutoff point of 2.5 Showed adequate sensitivity (97.9%) and specificity (100%) to detect Dysphagia diagnosed by clinical observation (Table 10). 6.5 is the cutoff to detect penetration and/or aspiration in FEES, with a sensitivity of (85.2%) and specificity (of 81.8%) (Table 11). In addition, a 6.5 cutoff revealed to have a sensitivity (83.3%) and specificity (80.6%) to detect Penetration in FEES (Table 12), and 8.5 is the most acceptable cutoff point to predict aspiration with a sensitivity of 84.2 and specificity 85.4 (Table 13).

Table 10: Sensitivity and Specificity of the Questionnaire in predicting Dysphagia in comparison with clinical evaluation in the Dysphagia susceptible cohort

Cut off points	Sensitivity %	Specificity %
0.50	100	33.3
1.50	97.9	58.3
2.50	97.9	100.0
3.50	91.7	100.0
4.50	85.4	100.0
6.50	60.4	100.0
7.50	56.3	100.0

Table 11: Sensitivity and Specificity of the Questionnaire in predicting Dysphagia in comparison with the presence of Penetration &/or Aspiration in FEES in the Dysphagia susceptible cohort

Cut off points	Sensitivity %	Specificity %
2.50	100	39.3
3.50	96.3	45.5
4.50	92.6	51.5
5.50	88.9	69.5
6.50	85.2	81.8
8.50	66.7	87.9
9.50	59.3	90.9

Table 12: Sensitivity and Specificity of the Questionnaire in predicting Dysphagia in comparison with the presence of Penetration in FEES in the Dysphagia susceptible cohort

Cut off points	Sensitivity %	Specificity %
2.50	100	36.1
3.50	95.8	41.7
5.50	87.5	62.9
6.50	83.3	80.6
8.50	70.8	85.1
10.5	45.8	91.7

Table 13: Sensitivity and Specificity of the Questionnaire in predicting Dysphagia in comparison with the presence of Aspiration in FEES in the Dysphagia susceptible cohort

Cut off points	Sensitivity %	Specificity %
3.50	100	39.0
4.50	94.7	43.9
5.50	89.5	58.5
7.50	89.5	75.6
8.50	84.2	85.4
9.50	78.9	91.2
10.5	57.9	92.7

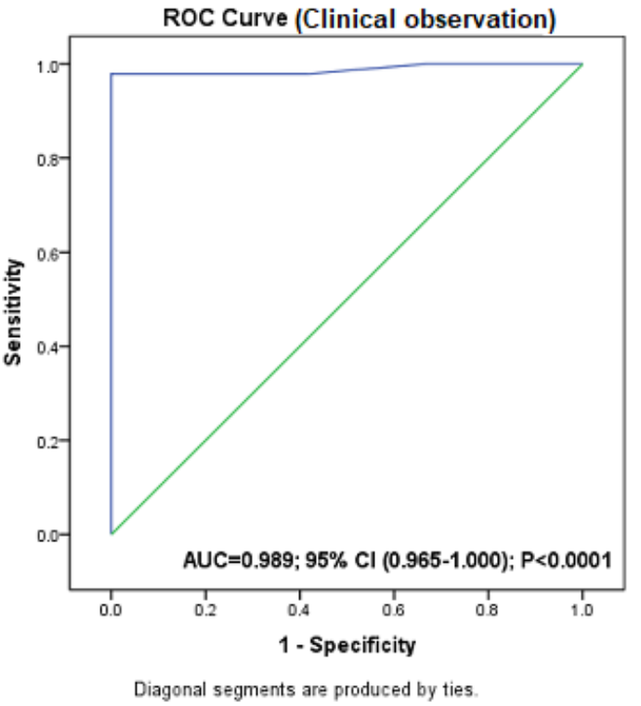

Fig. 7: Receiver Operating Characteristics curve (clinical observation)

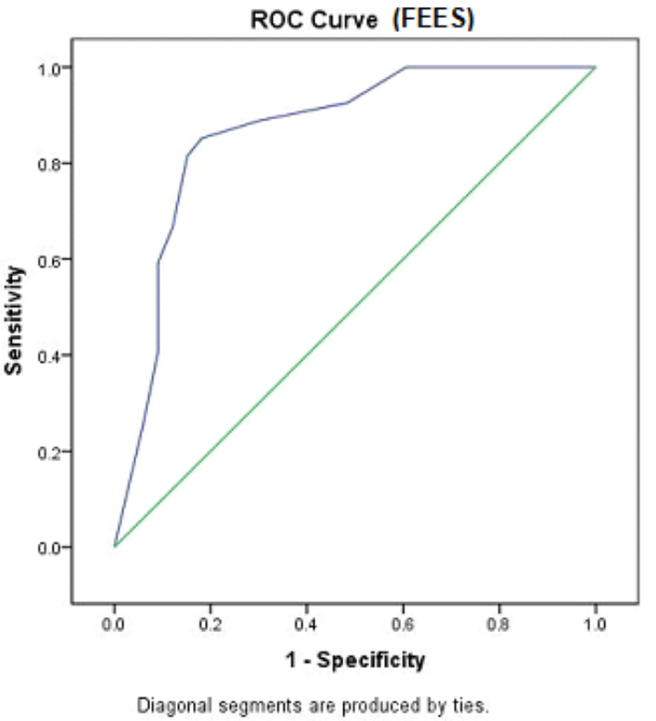

Fig. 8: Receiver Operating Characteristics curve (FEES)

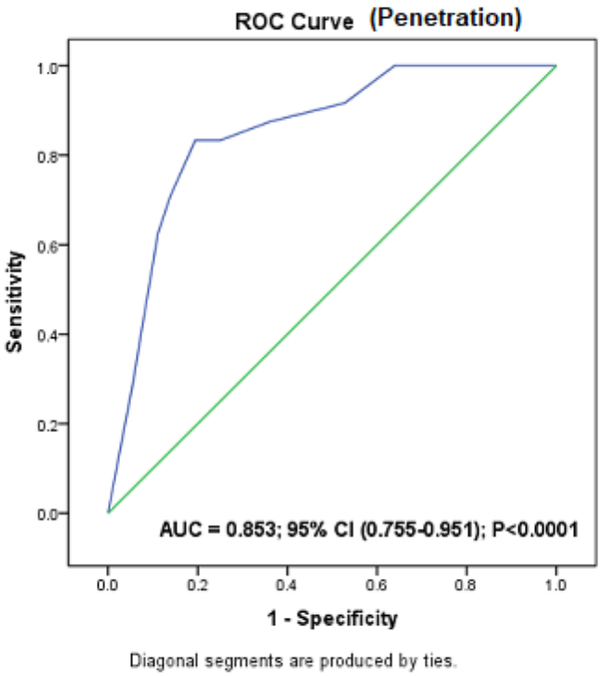

Fig. 9: Receiver Operating Characteristics curve (Penetration)

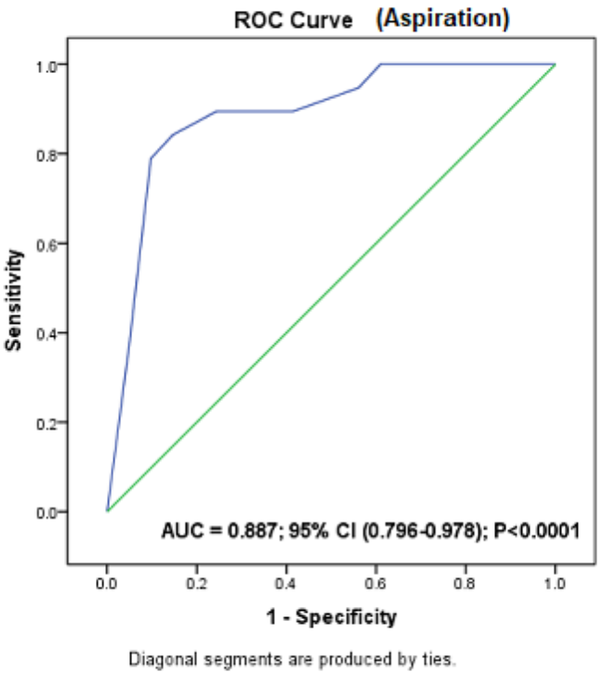

Fig. 10: Receiver Operating Characteristics curve (Aspiration)

DISCUSSION

The primary objective of this study was to design a Pediatric Dysphagia Screening Questionnaire. The rationale for pursuing this goal is to assist clinicians in prioritizing the need for instrumental and complete clinical evaluations. The analysis showed that this tool could potentially contribute to clinical practice. We partially modeled our questionnaire using the EAT-10 tool and validated the PDSQ against the FEES and clinical feeding observations. Forty-nine patients from the dysphagia-susceptible cohort of subjects not specifically seeking medical advice for a complaint of difficulty in eating and swallowing showed abnormalities in clinical observation; only twenty-seven of which had penetration and/or aspiration detectable by FEES. Twenty-two subjects had a functional pharyngeal swallow, even though they had a clinically impaired oral voluntary motor phase. This condition still impairs oral intake, warrants further evaluation, and may require clinical intervention. Specifically, this subset of twenty-two had abnormal clinical observations in feeding positions, abnormal labial seal, anterior spillage, abnormal suckling, tongue thrust, or food sticking to the palate with no coughing or choking during or after swallowing. This finding underpins the idea that a screening questionnaire for pediatric Dysphagia must include questions to probe physiological performance in both the oral motor and pharyngeal phases of swallowing.

We performed a post hoc analysis of the mean scores of the questionnaire in patients classified as oral-motor or oropharyngeal by clinical observation (Table 14). The mean questionnaire score for patients with isolated disordered oral motor performance was 5.8, a statistically lower score compared to patients with pathology classified as oropharyngeal 9.9. The clinical discrimination between the two categories depends on the presence or absence of coughing and throat clearing during or after swallowing in Oropharyngeal Dysphagia, along with any previously mentioned observations in the oral voluntary motor phase. Patients with only oral motor symptoms would have a less severe pathology than those with pharyngeal affection. Therefore, designing questionnaire items targeting oral motor pathology and validating the screening tool across clinical observations is warranted to improve the sensitivity of the screening tool in the pediatric population.

Table 14: Dysphagia by clinical observation in the susceptible cohort of Patients

	Non-Dysphagic (n=11)		Dysphagic (n=49) Suspected Oral-Motor Dysphagia (n=24)		Suspected Oropharyngeal Dysphagia (n=24)		Suspected Esophageal Dysphagia		*P-value*
	Mean	SD	Mean	SD	Mean	SD	(n=1)	SD	
Total Questionnaire score	1	0.9	5.8	2.5	9.9	2.7	7		*<0.0001*c*
			*<0.0001*a*		*<0.0001*b*		------		
					*<0.0001*d*				

One-way ANOVA followed by Tukey post hoc test

*a: High statistically significant difference; *p-value <0.0001* between Non-Dysphagic and Suspected Oral-Motor Dysphagia

*b: High statistically significant difference; *p-value <0.0001* between Non-Dysphagic and Suspected Oropharyngeal Dysphagia

*c: High statistically significant difference; *p-value <0.0001* between Non-Dysphagic and all Types of Dysphagia

*d: High statistically significant difference; *p-value <0.0001* between mean scores of Oropharyngeal Dysphagia and Oral-Motor Dysphagia

The two questions with the highest Yes response in dysphagic patients were: Does food consistency greatly affect feeding and swallowing? Is it difficult to bite or chew? In those two questions, patients diagnosed with FEES scored 88.9 and 96.3, respectively; the corresponding scores in patients diagnosed with Clinical observation were 79.6 and 93.9. This trend further supports the ability of FEES to detect more severe cases, highlighting that clinical observation is more sensitive in the pediatric population and debating its suitability as a validation tool for the PDSQ.

The question with the lowest rating of Yes among dysphagic patients diagnosed by either FEES (penetration and/or aspiration) or clinical feeding observation was: Is it difficult to skip from a cup? with scores of 59.3 and 42.9 percent, respectively). Caretaker comfort in presenting food with the cup could affect this parameter rather than the efficiency of the feeding process.

The only questionnaire item showing a lack of significant difference between patients with and

without Dysphagia within the dysphagia-susceptible cohort was question 11: Is there a loss of weight or failure to gain it? We make this observation when inspecting data from FEES using benchmarks for aspiration, penetration, or both. Interestingly, question eleven scored 10/19 out of instances of a yes score in the healthy controls. Notably, this is the first question in the E.A.T. 10 and PEDI- E.A.T. 10 questionnaires. The lack of significance in our data may reflect a cultural/ psychological perception of slow progress in weight gain rated by the child's parent or caretaker. However, in the broader cohort of patients diagnosed by clinical observation, the question had a significantly higher Yes rating among dysphagic patients.

Our Data contrasted the PEDI- EAT-10 cutoff of 4 /40 (one-tenth of the scale) when we used FEES as a validation tool; PDSQ showed good sensitivity and specificity for detecting penetration at 6.6, aspirations at 8.5, and notably scored from a possible 12 point range. In 2018 Arslan, Kılınç, Yaşaroğlu, Demir, & Karaduman[25] applied their tool to cases of neurological, pediatric Dysphagia undergoing videofluoroscopy. They found that the mean score of the PEDI EAT 10 in children with aspiration was 22.32 and in children with penetration 12.35, thus assigning a rating of 13/40 as the cutoff for discrimination in this specific population. The later study results are more on par with our scores and contradict the previously mentioned cutoff of 4/40 in a very similar patient population, possibly because of a difference in the temporal gap between the questionnaire application and the validation tool in both studies. Arslan *et al*., 2018[25] mentioned that the researchers administered the questionnaire on two separate occasions without identifying whether it occurred on the same day as the Videofluoroscopy. Arslan *et al*., 2018[16] confirmed that screen and instrumental assessments co-occurred on the same day.

The PDSQ achieved Par sensitivity and specificity to PEDIEAT 10 at a cutoff of 3 when validated against clinical feeding observations. This cutoff differs from the earlier tool used in the published studies. The PEDI- E.A.T. 10 targets children with neurologically based swallowing pathophysiology. The PDSQ is for a broader group of patients, including those with anatomical or developmental oral motor deficits. The questionnaire thoroughly explores this element with questions specifically targeting volitional motor performance. Arslan *et al*. (2018)[25] confirmed that oral phase dysfunction was present in 46.1 percent of 254 patients with cerebral palsy in their study population; however, there was no specification if any of those patients had or did not have pharyngeal phase deficiencies. The PDSQ has a simple rating scale of Yes and No questions, a different approach than the graded 0-4 response scale used in the PEDI-EAT 10. Videofluoroscopy validated the PEDI- E.A.T. 10, while the PDSQ was validated using FEES and clinical feeding observations.

CONCLUSION

All items of the PDSQ had a statistically significant higher Yes rating in dysphagic patients compared to the non-dysphagia group within the susceptible cohort; this warrants and supports the inclusion of all questions in the tool. The results highlight the impact of developmental and anatomical oral motor deficits on swallowing physiology. The broadness of the study population and simplicity of using the tool advocate its value as a possible screening alternative for use in children with disorders associated with Dysphagia as a comorbidity. The contrast between cutoff scores of patients only affected in the voluntary oral phase versus those with oropharyngeal impairment may be clinically helpful in suggesting the choice of one instrumental assessment over the other. Clinicians must interpret this cautiously; a study with Videofluoroscopy cross-validating the PDSQ in patients must occur before making this conclusion. The value of these findings would increase if the study had a more extensive cohort of patients. The validity of the PDSQ will increase if the scores of healthy participants are collected and compared with the results from dysphagic patients. Future application of this tool in dysphagic patients before and after therapeutic interventions would also increase its clinical value.

CONFLICT OF INTEREST

There are no conflicts of interest.

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