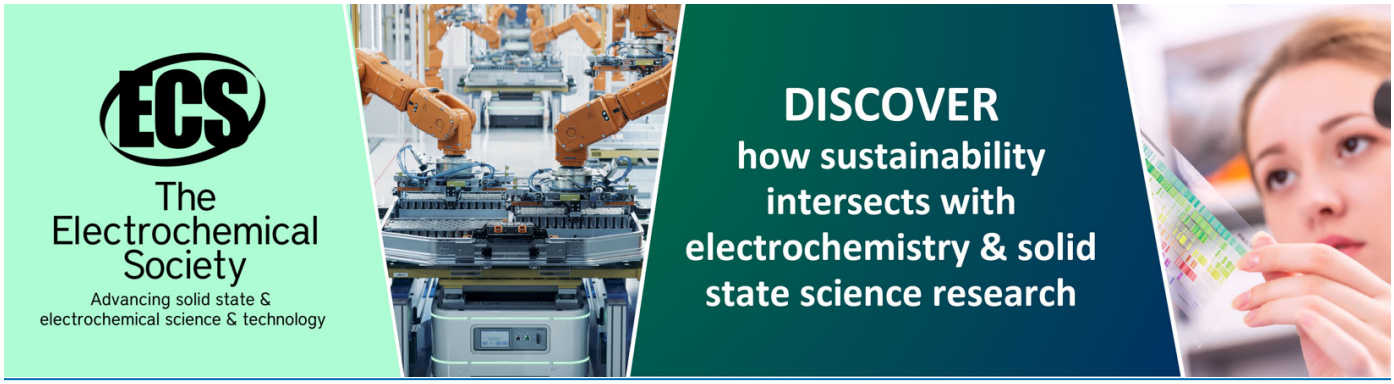
An overview of flexible endoscopic swallowing examination in the pediatric patients with suspected dysphagia



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An overview of flexible endoscopic swallowing examination in the pediatric patients with suspected dysphagia

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Abstract. Children's eating and swallowing ability is dynamic and is closely related to the growth processes. Anatomical structure during growth can impact the maturity of swallowing ability. Disruption of the swallowing process may cause dysphagia. This study is a descriptive cross-sectional design involving 54 subjects with consecutive sampling to assess the difficulties with swallowing and feeding. This study used the flexible endoscopic evaluation of swallowing (FEES) and assessed the characteristics of the subjects including age, gestation length age, caregivers, symptoms, complications, and medical disorders. This study showed the prevalence of dysphagia was 63% in children with suspected dysphagia. Dysphagia symptoms in children <6 months included apnea during bottle/breast feeding (7/34). In children >6 months, postural impairment (10/34), drooling (6/34), and coughing while eating (8/34) were the symptoms of dysphasia. Underlying diseases included structural anomaly (5/34), cardiopulmonary-larynx disorder (24/34), and neurological disorders (23/43). Complications included gastroesophageal reflux disease (GERD) (12/34), failure to thrive (10/34), and aspiration pneumonia (3/34). In FESS examination, standing secretion (22/34) and impaired tongue movement (20/34) indicated dysphagia. Residue was more common occur in gastric rice consistency (44.7%), penetration in thin liquid (44.2 %) and aspiration in thick liquid (34.8%).

1. Introduction

In children, the ability to eat and swallow is dynamic process. Anatomical structure is a crucial component of the swallowing ability, and the ongoing growth and development affect the maturation of the swallowing function. Neurological, physical, cognitive, oromotor status, independence, food preferences, and skills in children are constantly changing. Therefore, improved feeding and swallowing in children is expected to occur in accordance with the growth and maturation of the anatomical structures that affect swallowing [1-3].

The incidence of feeding difficulties and dysphagia in children has increased in the past 20 years. Presumably, this is due to an increase in the survival rates of the children born prematurely, children

with low birth weight, and children with complex medical conditions. For example, the percentage of children born prematurely has increased 20% since 1990, whereas the number of babies with a birth weight of 2500 grams or less has increased in the last 50 years. Another factor is the increased survival rate of children with cerebral palsy and developmental abnormalities. Twenty percent of the children born at 24–26 weeks of gestation suffer from cerebral palsy, whereas only 4% of children born at 32 weeks experience cerebral palsy [4,5].

Feeding difficulties are estimated to occur in 25–45% of the growing children and in more than 80% of children with developmental disabilities. Feeding difficulties and dysphagia in children may be caused by behavioral problems, progression, neurological disorders, respiratory disorders, or anatomical abnormalities. In addition, Sitton et al. [6] reported that the causes include neurological abnormalities, genetic disorders, heart abnormalities, vocal cord dysfunction, premature birth, tonsillary hypertrophy, and micrognathia. Prasse et al. [7] reported dysphagia in children with neurological abnormalities, cerebral palsy, premature birth, tracheostomy or dependent ventilators, craniofacial disorders, and failure to grow [4-7].

Dysphagia may occur in the oral, pharyngeal, or esophageal phase of the swallowing process. Specific clinical evaluations are performed according to the symptoms, and the appropriate diagnostic tests can ascertain the cause and extent of the swallowing disorder. Such tests include general physical examination (nose, oral cavity, hypopharyngeal, and larynx) and oromotor examination. The flexible endoscopic evaluation of swallowing (FEES), a swallowing examination using a flexed fiber-optic endoscopy, has been specifically developed to assess the swallowing difficulties [8,9].

Dysphagia can occur in children with certain diseases including neurological disorders, gastroesophageal reflux disease (GERD), laryngomalacia, cardiac abnormalities, and craniofacial disorders. In a study of 33 children with dysphagia by Ko et al. [10], 13% had cerebral palsy, 15% had a central nervous system disorder, 15% had epilepsy, 6% had a congenital heart disorder, 6% had bronchopulmonary dysplasia, 5% had gastroesophageal reflux, 9% had a developmental delay, and 6% had failure to grow. Pediatric patients with neurological disorders and GERD have decreased sensation in the laryngopharyngeal area and the food entering the hypopharynx is sometimes not followed by airway closure, which can lead to aspiration or silent aspiration. Pediatric patients with neurological disorders tend to have poor esophageal motility, leading to the occurrence of GERD. Patients with laryngomalacia tend to experience GERD because it decreases the vagus nerve function, causing decreased laryngeal tone and esophageal motility [10-12].

Dysphagia is not always due to impaired swallowing ability; however, it may occur due to problems with airway protection and aspiration. Silent aspiration that occurs continuously will cause food to enter the lungs. If this continues long-term, it will lead to an infection known as aspiration pneumonia [13-15].

To date, no studies are available on dysphagia in children in Indonesia. Similarly, research on the swallowing process in the pediatric patients with dysphagia using FEES has not been previously performed. Thus, this study aimed to obtain the baseline data to determine the “big picture” of dysphagia and aspiration problems in the pediatric patients in Indonesia. Presumably, this data will be useful to the clinicians and families for the comprehensive management of dysphagia in children.

2. Methods

This was a descriptive cross-sectional study to determine the FEES descriptions in 54 pediatric patients with suspected dysphagia treated at the Integrated Dysphagia Clinic of ENT Department, Cipto Mangunkusumo Hospital, Jakarta. Patients were consecutively recruited between October and November 2016. Patients that met the study criteria and had no conditions in accordance with the rejection criteria were included in the study.

Patients underwent the FEES examination at the Integrated Dysphagia Clinic of ENT Department. The intent and purpose of the study, the examination stage, and the examination technique were explained to the patients and their parents. The parent/guardian of the patient provided their informed

consent. The Health Research Ethics Committee, Faculty of Medicine, Universitas Indonesia-Cipto Mangunkusumo Hospital approved this study protocol.

The parent/guardian completed a dysphagia questionnaire to determine the patient's history of swallowing difficulty. Patients follow the vital checks and postural control assessments using oximetry during the examination. Then the patient was positioned in an upright sitting position. Preswallowing was assessed by evaluating the oral cavity, tongue movement, buccal masseter condition, oral hygiene, palate, and temporomandibular joints. A flexible nasopharyngolaryngoscopy fiber optic scope was inserted through the cavum nasi to assess the velopharyngeal competence, base of the tongue, vallecula, pyriform sinus, posterior pharyngeal wall, post-cricoid, laryngeal structure, vocal fold movement during phonation and inspiration, accumulation of saliva, penetration or salivary aspiration, and accompanying cough reflex.

The consistency of the food provided for the test was adjusted according to the oromotor development of the patients based on their age, that is, for <6 months, only water and milk were provided; for 6–7 months, milk porridge followed by milk and then water; for 7–9 months, milk porridge followed by filtered rice, milk, and then water; for 9–12 months, milk porridge, filtered rice, gastric rice, milk, and then water; and for >12 months, milk porridge, filtered rice, gastric rice, steamed rice with chopped rough side dishes, milk, and then water. During the FEES examination, the presence of preswallowing leakage, residue, penetration, and aspiration were assessed. The patients who could not swallow or manipulate the consistency of the food, were given a safe consistency and stop at that consistency.

The results of the examination were recorded and statistically analyzed using SPSS 20 (Statistical Package for Social Science) software. Univariate analysis was performed to analyze the subject characteristic data and frequency distributions. The prevalence rates were also calculated. *P*-values < 0.05 were considered as statistically significant.

3. Results

FEES examination confirmed that 37% of the study population had feeding difficulties, 7.4% had mechanical dysphagia, and 55.6% had neurogenic dysphagia (Table 1).

Table 1. Distribution of characteristics related to the diagnosis of swallowing (dysphagia) or feeding difficulties (n = 54).

Samples Characteristic	Quantity (%)
Feeding difficulties	20 (37)
Oral mechanical phase dysphagia	3 (5.6)
Oral neurological phase dysphagia	7 (13)
Pharynx mechanical phase dysphagia	1 (1.8)
Pharynx neurogical phase dysphagia	9 (16.7)
Oral and pharynx mechanical phase dysphagia	0 (0)
Oral and pharynx neurogical phase dysphagia	14 (25.9)

Demographic characteristics are presented in Table 2. Of the 34 pediatric patients with dysphagia, 19 (56%) were male. Patients were divided into four age groups: the group ≤6 months revealed 11 patients with dysphagia; the group 7–12 months had 4 patients with dysphagia; the group 13–36 months revealed 9 patients with dysphagia; and the group >36 months had 10 patients with dysphagia. The median age was 23 months, with the youngest aged 1 month and the oldest aged 13 years.

Table 2. Rates of swallowing (dysphagia) and feeding difficulties by sex, age, caregiver, maternal pregnancy, nutritional status, and medical diagnosis.

Subject Characteristic		Dysphagia (n = 34)	Feeding Difficulties (n = 20)
Sex	Male	19	13
	Female	15	7
Age	≤6 months	11	3
	7–12 months	4	5
	13–36 months	9	6
	>36 months	10	6
Caregiver	Parent (Mother)	27	18
	Guardian	7	2
Pregnancy	Premature	8	6
Duration	Not premature	26	14
Nutritional Status	Poor nutrition	9	8
	NGT/PEG	9	1
	Oral	0	7
	Good nutrition	25	12
	NGT/PEG	14	8
	Oral	11	4
Medical Diagnosis	Neurologic	23	14
	Heart, Lung, Larynx	24	9
	Structural	25	7
	Involvement of 1 organ system	8	10
	Neurologic	5	5
	Heart, Lung, Larynx	2	3
	Structural	1	2
	Involvement of 2 organs system	13	10
	Neurologic—Heart, Lung, Larynx	2	5
	Neurologic—Structural	4	4
	Heart, Lung, Larynx - Structural	7	1
	Involvement of 3 organs system	13	0
	Neurologic - Heart, Lung, Larynx - Structural	12	0
	Heart, Lung, Larynx–Structural - Digestion	1	0
Complications	Failed to grow	4	4
	Aspiration pneumonia	1	0
	PRGE	5	4
	Failure to grow and aspiration pneumonia	0	0
	Failure to grow and GER	5	2
	Aspiration pneumonia and GER	1	0
	Failure to grow, aspiration pneumonia, and PRGE	1	0
	None	17	10

Regarding the primary caregiver, a majority of the patients with dysphagia were cared for by their mother (27 of 34), whereas the remaining were cared for by a grandparent, extended family member, housemaid, baby sitter, guardian, or nurse. For maternal pregnancy, 8 of 34 patients had full-term birth

(not premature) dysphagia. Only 9 of 34 patients with dysphagia had poor nutritional status. Of the 17 patients with poor nutrition, 9 patients were diagnosed with dysphagia and all used NGT/PEG. Of the 8 patients with feeding difficulties, 1 used NGT and 7 used oral intake. There were 37 patients with sufficient nutrition. Of the 25 patients with dysphagia, 14 used NGT and 11 used oral intake. Of the 12 patients with feeding difficulties, 8 used NGT and 4 used oral intake. Of the 34 patients with dysphagia, 23 revealed neurological abnormalities, 24 had a heart/lung/larynx abnormalities, and 25 indicated structural abnormalities. Patients with the structural abnormalities had a 1.9-fold greater risk of dysphagia than the patients without structural abnormalities. Of the 34 patients with dysphagia, 8 had abnormalities in 1 organ system, 13 had abnormalities in 2 organs systems, and 13 had abnormalities in 3 organs system. All patients with abnormalities in three organ systems revealed dysphagia. Of the 54 patients with complications, 6 revealed failure to grow, 3 had aspiration pneumonia, and 18 had PRGE. All patients with aspiration pneumonia also revealed dysphagia.

Table 3. Frequency of symptoms related to dysphagia and feeding difficulties.

	Dysphagia (*n* = 34)	**Feeding difficulties (n = 20)**	**Prevalence Rate (IK 95%)**
<6 months			
Apnea while nursing	8	3	0.727 (0.506–1,044)
Do not want to breastfeeding	5	0	1.500 (0.945–2.381)
General Symptoms			
Posture is disturbed	10	9	0.947 (0.523–1.716)
Meal >30 min	3	6	0.549 (0.208–1.449)
Only eat certain foods	4	3	1.701 (0.520–2.209)
Oral Phase Symptoms			
Excessive drool	6	4	1.157 (0.621–2.158)
Food/saliva accumulated in the cheeks	3	5	0.640 (0.249–1,647)
Food out of the nose	3	1	1.456 (0.756–2.804)
Pharynx Phase Symptoms			
Choking/choking while eating	1	2	0.596 (0.117–3.038)
Trying hard to swallow	2	0	**1.944 (1.409–2.683)**
Cough while eating	8	6	1.095 (0.602–1,994)
Apnea while eating	1	0	**1.895 (1.391–2,581)**
There was a lot of mucus in the throat	5	2	1.429 (0.792–2.576)
Hoarseness	1	0	**1.895 (1.391–2.581)**
Vomiting while eating	2	2	0.917 (0.328–2.563)

As listed in Table 3, the most common complaints at <6 months of age were apnea during feeding, with about 8 of the 11 patients experiencing dysphagia. The most common symptom of discomfort was posture, which was experienced by 19 patients, 10 of who had dysphagia. Oral phase complaints included excessive drooling, with 6 of 10 patients experiencing dysphagia. Coughing during feeding was the most common pharyngeal phase complaint, with 8 of 14 patients experiencing dysphagia. Patients with the symptoms of trying to swallow had a 1.9-fold greater incidence of dysphagia than those without the symptoms. Patients with apnea symptoms at meals and hoarseness were 1.8 times more likely to develop dysphagia than those without these symptoms.

Table 4. Initial FEES findings in patients with dysphagia and feeding difficulties.

Initial Examination of FEES			
	Dysphagia (n = 34)	**Feeding difficulties (n = 20)**	**Prevalence Ratio (IK 95%)**
The movement of the temporomandibular joint is disrupted	3	1	1.210 (0.660–2.217)
Weak Lipseal	10	4	1.190 (0.785–1.806)
The movement of the tongue is disturbed	20	2	**2.078 (1.373–3.145)**
Buccal tone is disrupted	4	1	1.307 (0.799–2.136)
Closure of the asymmetric velopharynx	11	2	**1.508 (1.056–2.514)**
Parese plika vocalist,	2	0	**1.625 (1.311–2.015)**
Epiglottic retroflexion is impaired	13	5	1.238 (0.832–1.843)
Standing secretion	22	3	**2.127 (1.347–3.358)**
Silent aspiration	3	1	**2.000 (1.467–2.727)**

As listed in Table 4, of the 34 patients with dysphagia, 3 experienced a disrupted temporo-mandibular joint movement, 10 had weak lip seals, 20 reported disrupted buccal tone, 11 had impaired velopharyngeal closure, 2 revealed vocal fold paresis, 13 were with disturbed epiglottis retroflexion, and 22 had standing secretion. Patients with impaired tongue movement had a 2-fold greater risk of dysphagia, whereas patients with asymmetric velopharyngeal closure had a 1.5-fold greater risk than those with symmetrical velopharyngeal closure. Patients with vocal cord paresis had a 1.6-fold greater risk of dysphagia, and patients with standing secretion had a 2.1-fold greater risk than those without standing secretion.

At the FEES examination, preswallowing leakage was observed at a similar rate for all food consistencies, residue occurred more frequently for foods with coarse consistency, and there was more frequent penetration for water consistency and aspiration for milk and water consistencies. For all food consistencies, FEES parameter disturbances were more common in the patients with neurological disorders.

4. Discussion

In this population, feeding difficulties and dysphagia occurred in 37% and 63% of children, respectively. Burklow et al. reported that 33–88% of children with developmental disorders have dysphagia [4]. Sensory processing disorder is experienced by 5–15% of children in the general population and 63% of children with autism [4,12].

Interestingly, only 23.5% of dysphagia patients in this study were born prematurely. This is not in accordance with the literature that states that premature birth is one of the main factors leading to the eating and swallowing disorders in children. Another study stated that 37–40% of the children with eating and swallowing disorders were born premature. The discrepancy between the research results and the literature may be due to the inclusion of patients over the age of 6 months. At 6 months of age, maternal pregnancy is one of the factors affecting difficulty in swallowing in children because it affects the suction-swallow-breathe cycle. Premature babies cannot coordinate between sucking, swallowing, and breathing. This coordination is obtained at 34 weeks in premature babies [4,16].

In addition, 26.5% of patients with dysphagia and 40% patients with feeding difficulties had a poor nutritional status. Wolf et al. [13] reported that poor nutrition is one of the complications of eating and swallowing difficulties. Poor nutrition status will lead to failure of growth and delayed development. This discrepancy occurred because the nutritional needs of the children diagnosed with difficulty in eating and swallowing were met via NGT or nutritional fulfillment.

This study found 1 patient of poor nutrition with feeding difficulties using NGT, it is aimed to pursue nutritional requirement and can be tested food texture according to age. In 8 patients of sufficient nutrition with feeding difficulties using NGT, it is because those patients have never performed FEES examination. Therefore it is necessary to examine FEES in children with suspected dysphagia so that children do not need to be given continuous intake per NGT and should be trained to swallow food according their age.

The results found 33 patients with heart, lung, and larynx abnormalities, including 29 patients with laryngomalacia, 10 patients with cardiac abnormalities, and 6 patients with lung abnormalities. Infants with heart, pulmonary, and laryngeal abnormalities often have difficulty initiating and maintaining coordination between the suction-breath during ingestion [17].

The most frequent pharyngeal dysphagia symptom was coughing during feeding, which occurred in 14 patients (35%), followed by mucus in 7 patients (17.5%), and choking on feeding in 3 patients (7.5%). Weir et al. [18] reported similar rates, namely 46% coughing, 32% mucus in the throat, and 2% choking. The difference may be due to the sample age. In this study, 11 of 14 samples were aged ≤6 months with apnea symptoms during breastfeeding. Differences in the pharyngeal dysphagia symptoms were observed at age ≤6 months for coughing symptoms while eating and age >6 months with apnea symptoms during breastfeeding [18].

In FEES, 22 of 34 patients with dysphagia had standing secretion. Standing secretion is the first identifiable FEES parameter in the hypopharynx, which occurs due to hyposensitivity of the hypopharynx so that the pharyngeal peristalsis is not optimal. Residues occurred more frequently for foods with rough consistency, penetration occurred more frequently for water, and aspiration occurred more frequently for milk and water. Barata et al. [19] reported that a thick liquid consistency and solid foods were more likely to result in residue, but were less risky for penetration or aspiration and nasal regurgitation. Bingie et al. [20] also found that the incidence of penetration and aspiration was reduced for foods with a denser consistency [19,20]. These findings may be useful for parents in determining food types that children with swallowing difficulties needed.

5. Conclusion

In this study, the prevalence of dysphagia in children in Indonesia with swallowing difficulties was estimated to be 63%. Dysphagia may occur at age ≤6 months (11/34), 7–12 months (4/34), 13–36 months (9/34), and >36 months (10/34). Dysphagia occurs in males (19/34) and females (15/34). The initial FEES examination revealed that standing secretion (22/34), disturbed tongue movement (20/34), and silent aspiration (41.2%) were the most common symptoms in children with dysphagia.

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