ORIGINAL ARTICLE ÖZGÜN ARAŞTIRMA

Evaluation of Swallowing Dysfunction in Children with Recurrent Respiratory Symptoms

Tekrarlayan Solunum Sistemi Semptomları Olan Çocuklarda Yutma Disfonksiyonun Değerlendirilmesi

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Abstract

Introduction: This study aims to determine the role of swallowing disorders in the etiology of recurrent lower respiratory tract infections (LTRIs) and persistent respiratory symptoms in children.

Materials and Methods: The records of 32 patients aged 0-18 years who applied to the outpatient clinic of the pediatric pulmonology department between November 2016-June 2019 with LTRIs or persistent respiratory symptoms and diagnosed as swallowing dysfunction were evaluated retrospectively. The swallowing function of the patients were imaged and recorded by videofluoroscopic swallowing study (VFSS), and parents were surveyed for with pediatric eating assessment tool-10.

Results: Median age of the patients was 14 (7.25-32.25) months, 81.3% were male. It was noted that 50% of the patient population had metabolic and/or neurologic disease, and half of the remaining 16 patients had risk factors for swallowing dysfunction, while the other half did not. Aspiration of thin liquids was found in 90.6% of patients, of which 68.8% were silent, 56.3% had oral phase dysfunction and 40.6% had a delayed swallowing reflex. Oral phase dysfunction and delayed swallowing reflex for thin and thick liquids were more frequent in 16 (50%) patients with metabolic and/or neurological disease on VFSS compared to those without metabolic and/or neurological disease ($p<0.05$). There was reduction in the annual number of LTRIs reported following implementation of swallowing therapy ($p=0.01$).

Conclusion: Swallowing dysfunction is an important diagnosis to consider when approaching patients with history of recurrent respiratory symptoms, regardless of their comorbidities and risk factors. Early diagnosis and treatment is crucial to avoid subsequent complications associated with it.

Keywords
Swallowing dysfunction, pediatric, dysphagia, respiratory symptoms

Anahtar kelimeler
Yutma disfonksiyonu, pediatrik, disfaj, solunum sistemi semptomları

Öz

Giriş: Bu çalışmanın amacı çocuklardaki tekrarlayan alt solunum yolu enfeksiyonu (ASYE) ve persistan solunum sistemi semptomlarının etiyolojisinde yutma disfonksiyonun rolünü belirlemektir.

Gereç ve Yöntem: Kasım 2016-Haziran 2019 tarihleri arasında çocuk göğüs hastalıkları polikliniğine ASYE veya persistan solunum sistemi semptomlar ile başvuran ve yutma disfonksiyonu tanısı alan 0-18 yaş arası 32 hastanın kayıtları geriye dönük olarak kaydedildi. Hastaların yutma fonksiyonu videofloroskopik yutma çalışması (VFSS) ve pediyatrik yeme değerlendirme aracı-10 ile değerlendirildi.

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Bulgular: Hastaların medyan yaşı 14 (7.25-32.25) aydı, %81.3'ü erkekti. Hastaların %50'sinin metabolik ve/veya nörolojik hastalığı vardı, %25'inin yutma disfonksiyonu için risk faktörlerine sahipti ve %25'inin herhangi bir risk faktörü veya altta yatan bir hastalığı yoktu. Hastaların %90.6'sında ince sıvıların aspirasyonu bulundu, bunların %68.8'i sessiz, %56.3'ünde oral faz disfonksiyonu ve %40.6'sında gecikmiş yutma refleksi vardı. VFSS'de metabolik ve/veya nörolojik hastalığı olan 16 (%50) hastada metabolik ve/veya nörolojik hastalığı olmayanlara kıyasla ince ve puding kıvamlı sıvılar için oral faz disfonksiyonu ve gecikmiş yutma refleksi daha sıktı ($p<0.05$). Yutma terapisi alan takipli hastalarda yıllık ASYE sayısında azalma saptandı ($p=0.01$).
Sonuç: Yutma disfonksiyonu, tekrarlayan solunum sistemi semptomları öyküsü olan hastaları değerlendirirken eşlik eden hastalıkları ve risk faktörleri ne olursa olsun etiyolojide düşünülmesi gereken önemli bir tanıdır. Erken tanı ve tedavi, yutma disfonksiyonu ile ilişkili komplikasyonları önlemek için çok önemlidir.

Introduction

The mechanics of respiration and swallowing have common neuromuscular structures and work in coordination with each other. This coordination is vital for airway protection and prevention of aspiration. Swallowing is a complex process and is divided into three consecutive phases: oral, pharyngeal and esophageal. It is influenced by multiple neurophysiological, anatomic, environmental and social factors. If any component is affected, it can lead to dysphagia, which is defined as abnormal swallowing due to incoordination, obstruction or weakness affecting swallowing biomechanics and can occur at any stage of swallowing. There are multiple causes of dysphagia including prematurity, gastroesophageal reflux, congenital malformations (cleft lip/palate, Moebius syndrome, Down syndrome, etc.), neurologic and metabolic disease (1). It can also be present in children without underlying risk factors (2,3).

Approximately 1% of children in the general population experience swallowing difficulties (2). This percentage is higher in patients with known comorbidities, such as cerebral palsy, traumatic brain injury, and airway malformations (4).

Children with dysphagia may not manifest with symptoms and may be clinically silent. Others present with vomiting, coughing, wheezing, recurrent respiratory tract infections, and/or choking (1,4,5). Aspiration is defined as food or liquid entering the airway below the level of the vocal cords (1). Silent aspiration is defined as the absence of coughing, despite aspiration before, during, or after swallowing (1). Although coughing is a mechanism that protects the airway against penetration and/or aspiration, most infants do not cough following aspiration (6). Therefore, recurrent pulmonary infections may be indicative of ongoing aspiration.

This study aims to determine the role of swallowing disorders in the etiology of recurrent lower respiratory tract infections (LRTIs) and persistent respiratory symptoms in children referred to the pediatric pulmonology department and whether there is a decrease in the number of LRTIs after treatment.

Materials and Methods

In the study, patients aged 0-18 years with recurrent LRTIs or persistent respiratory symptoms who were admitted to the outpatient clinic of the pediatric pulmonology department, between November 2016 and June 2019 were evaluated retrospectively through electronic medical reports. Patients with known swallowing dysfunction at admission were excluded from the study. Thirty-two patients were included in the study. The demographic characteristics of the patients, their symptoms and signs, whether they had any complaints while feeding, whether they were exposed to cigarette smoke, chest X-ray findings, the number of LRTIs before and after swallowing therapy, and whether they had known persistent diseases were noted (Table 1).

Patients who present to the pediatric pulmonology department with LRTIs or persistent respiratory symptoms and those with suspected swallowing dysfunction based on history, physical examination, and chest X-ray are routinely referred to the swallowing disorders unit. In this center, swallowing functions of the patients are imaged and recorded by videofluoroscopic swallowing study (VFSS). VFSS is a method that evaluates the oral, pharyngeal, and oesophageal phases of swallowing with liquid and viscous barium. Loss or delay in oropharyngeal reflexes, laryngeal penetration, reflux, residuals, and aspiration can be detected by VFSS. As described earlier, aspiration occurs when food or liquid enters the airway below the level of the vocal cords. On the other

hand, penetration is defined as the entry of food or liquid into the larynx without passing the vocal cords (1). The penetration aspiration scale (PAS) is an 8-point scale determined by the depth of the material passing through the airway and whether the material entering the airway can be expelled during videofluoroscopy (5). During their visits, parents are surveyed using the pediatric eating assessment tool-10 (PEAT-10). This PEAT-10 is effective in predicting the aspiration risks of patients and in monitoring the severity of initial symptoms and treatment effectiveness. The family is asked to score between 0 and 4 for 10 questions. A PEAT-10 score higher than 3 is predictive for airway aspiration (7).

The Gazi University Non-Interventional Clinical Research Ethics Committee approved the study (protocol number: 916100558, date: 04.01.02).

Statistical Analysis

The Statistical Package for Social Sciences (SPSS) version 18 was used for statistical analysis. The normal distribution of variables was tested with the Kolmogorov-Smirnov test. Continuous variables with normal distribution were presented as mean (standard deviation); non-normal distribution variables were reported as median (interquartile range). Categorical variables were shown as numbers and percentages. The Mann-Whitney U test and Student's test were used, respectively, to compare two groups of variables not normally distributed and normally distributed. The Pearson chi-square or Fisher's Exact test were used to evaluate categorical data. Correlations were analyzed using Spearman's test and Pearson's correlation analysis. A value of $p<0.05$ was considered as statistically significant.

Results

In the 3 years of the study period, a total of 2,666 patients were admitted to the outpatient clinic of the pediatric pulmonology department due to LRTIs or persistent respiratory symptoms. Seven patients had known swallowing dysfunction, were already followed up at the swallowing disorders unit, and were excluded from the study. Swallowing studies were performed in 73 patients due to complaints during swallowing or LRTI/persistent respiratory symptoms of unknown cause. Swallowing dysfunction was found in 32 of 73 patients (43.8%).

The median age of the 32 patients with swallowing dysfunction was 14 (7.25-32.25) months. The demographic and clinical characteristics of the patients are shown in Table 1. Of the patients, 16 (50%) had metabolic and/or neurologic disease and eight (25%) had risk factors for swallowing dysfunction (prematurity, congenital heart disease, laryngomalacia, and cleft lip and palate). Swallowing dysfunction was detected in eight patients (25%) without any underlying diseases or conditions that might affect swallowing. Of these eight patients, four patients had a chronic cough, three patients had recurrent LRTI and one patient had wheezing. On VFSS, among these eight patients, laryngeal aspiration was found in six children with thin liquids and in one child with both thin and thick liquids. Silent aspiration was detected in four of eight patients (50%). Half of these eight patients were exposed to cigarette smoke.

During admission, 26 patients (81.3%) had oral feeding, five patients (19.2%) had liquid modified feeding, and one patient (3.8%) had non-oral feeding. After VFSS, oral feeding was recommended for two patients (6.2%), liquid modified feeding for 22 patients (68,8%), and non-oral feeding for eight patients (25%). The results of the VFSS of the patients are shown in Table 2. The most common VFSS finding was a silent aspiration in 22 patients (68.8%). The mean of the PEAT-10 score of the patients was 14.4±11, and the median PAS score for thin liquids was 8 (7-8). Aspiration of thick liquids was detected in only six patients (18.8%), and all were silent aspirations.

No statistical difference was found in the PAS (thin-thick liquid) score according to gender of patients, birth weight-week, and underlying disease or risk factors. A significant difference was found between parental smoking and the PEAT-10 score of the patients ($p=0.046$). The PAS score of the patients exposed to cigarette smoke was higher, but the difference was not statistically significant ($p=0.081$). While no correlation was found between the number of LRTIs in one year and the PAS, a positive correlation was found with the PEAT-10 score ($p=0.008$, $r=0.781$). A comparison of the VFSS results of patients with and without metabolic and/or neurologic disease is shown in Table 3. On VFSS, 16 patients with metabolic and/or neurologic disease had oral phase dysfunction and a delayed swallowing reflex was more common in both thin and thick fluids than in those without metabolic

Table 1. Demographic and clinical characteristics of patients with swallowing dysfunction (n=32)	
	N (%)
Gender	
Male	26 (81.3)
Female	6 (18.8)
Age (months)	14 (7.2-32.2)*
Weight	
>(-2) SD**	14 (43.8)
<(-2) SD	18 (56.3)
Gestational age, week	
Term	23 (71.9)
Preterm	9 (28.1)
Symptoms	
Recurrent LRTI#	18 (56.3)
Persistent cough	9 (28.1)
Wheezing	5 (15.6)
Number of LRTI	
Total	5 (3-7)*
Annual (previous year)	3 (2-4)*
Symptoms while feeding	
Cough	14 (43.8)
Vomiting	9 (28.1)
Choking-growling	7 (21.9)
None	2 (6.3)
Chronic disease	
Yes	24 (75)
No	8 (25)
Neurometabolic disease	
Yes	16 (50)
No	16 (50)
Pulmonary infiltration	
Right	16 (50)
Bilateral	12 (37.5)
Left	2 (6.3)
Normal	2 (6.3)
Exposure to cigarette smoke	
Yes	20 (62.5)
No	12 (37.5)

#LRTI: Lower respiratory tract infection. *Median (interquartile range), SD: Standart deviation

Table 2. Evaluation of videofluoroscopic results of patients at admission and after swallowing therapy		
	At admission n=32 (%)	After swallowing therapies n=14 (%)
Penetration aspiration scale (thin liquid)		
None	2 (6.2)	8 (57.1)
Penetration	1 (3.1)	-
Aspiration despite patient's response	7 (21.9)	3 (21.4)
Aspiration absent patient's response	22 (68.8)	3 (21.4)
Penetration aspiration scale (thick liquid)		
None	26 (81.3)	1 (3.1)
Penetration	-	13 (40.6)
Aspiration despite patient's response	-	-
Aspiration absent patient's response	6 (18.7)	-
Oral phase dysfunction (thin liquid)		
Yes	18 (56.25)	1 (7.2)
No	14 (43.75)	13 (92.8)
Delayed swallowing reflex (thin liquid)		
Yes	13 (40.6)	4 (28.6)
No	19 (59.4)	10 (71.4)
Nasal regurgitation (thin liquid)		
Yes	3 (9.4)	1 (7.2)
No	29 (90.6)	13 (92.8)
Aspiration (thin liquid)		
Yes	29 (90.6)	6 (42.8)
No	3 (9.4)	8 (57.1)
Coughing (thin liquid)		
Yes	7 (21.9)	3 (21.4)
No	25 (78.1)	11 (78.6)
Oral phase dysfunction (thick liquid)		
Yes	17 (53.1)	1 (7.2)
No	15 (46.9)	13 (92.8)
Delayed swallowing reflex (thick liquid)		
Yes	10 (31.3)	2 (14.3)
No	22 (68.8)	12 (85.7)

Table 2. Continued		
	At admission n=32 (%)	After swallowing therapies n=14 (%)
Nasal regurgitation (thick liquid)		
Yes	2 (6.25)	-
No	30 (93,75)	14 (100)
Aspiration (thick liquid)		
Yes	6 (18.7)	-
No	26 (81.3)	14 (100)
Coughing (thick liquid)		
No	32 (100)	14 (100)

and/or neurologic disease, and it was statistically significant (p=0.037 and p=0.003 for thin liquids and p=0.004 and p=0.006 for thick liquids, respectively). Of the patients with metabolic and/or neurologic disease, 12 (75%) had growth retardation (weight is below 2 standard deviations according to his/her gender and peers). The weight Z-scores for the age of patients with metabolic and/or neurologic disease was lower than those without metabolic and/or neurologic disease (p=0.037).

In 20 patients (12 of 32 patients lost the follow-up in the pediatric pulmonology department), the median number of LRTIs per year before swallowing therapy was 3 (2-4), while the annual number of LRTIs after therapy was 1 (1-2.75). A reduction in the annual number of LRTIs of patients after swallowing therapy was statistically significant (p=0.010). The annual number of LRTIs was higher in patients with chronic disease or risk factors (p=0.035). There was no statistical difference between the annual number of LRTIs and the type of swallowing dysfunction.

Discussion

Children with swallowing dysfunction may present with recurrent LRTIs and persistent respiratory symptoms (1,3). Swallowing dysfunction should be suspected in children with persistent respiratory symptoms. In our study, swallowing dysfunction was found in eight children with normal neuromotor development without any underlying risk factors. The decrease in the symptoms of our patients with swallowing dysfunction after changing diets shows the importance of swallowing therapy in the management of children with recurrent LRTIs and persistent respiratory symptoms.

In our study, 1.2% of patients with recurrent LRTIs and persistent respiratory symptoms were diagnosed with swallowing dysfunction. Eight of these patients had no chronic disease or risk factor for swallowing dysfunction. Studies have shown varying degrees of swallowing dysfunction in children without risk factors (2,8-10). Silent aspiration is common among infants and young children with swallowing dysfunction (2-4,6). In a retrospective study, swallowing dysfunction was detected in 19 patients (19/517) who had no risk factors for dysphagia and had unexplained respiratory symptoms, and they detected silent aspiration in 57.9% of these patients (3). In our study, silent aspiration was found in half of the patients without risk factors and in 75% of the patients with risk factors.

In our study, patients with swallowing dysfunction presented mostly with recurrent LRTIs. Respiratory tract symptoms in patients with swallowing dysfunction vary in the literature (2-4,9). About half of our patients had a cough, while feeding and silent aspirations were detected in eight of these patients. The cough is one of the mechanisms that protects the respiratory tract (1). Silent aspiration was observed on VFSS even in patients who coughed while feeding, suggesting that coughing during feeding does not guarantee safe swallowing and does not protect airways completely, especially in children with persistent respiratory symptoms. One of the reasons for the high frequency of silent aspiration in children with dysphagia is thought to be incomplete maturation of airway clearance mechanisms (11). The absence of primary airway protection responses in recurrent aspirations may impair lung maturation, especially in 2-year-old children (12,13).

The position of the patient during aspiration determines the segment of the lung where aspiration is most likely to occur. If patients aspirate while in the supine position, the superior segment of the lower lobe of the right lung and the posterior segment of the upper lobe of the right lung are usually involved. Patients who aspirated in the upright position may have involvement of the basal segments of the bilateral lower lobes (14). In our study, most of our patients had infiltration in the right lung segments on chest X-rays. Suspicion of aspiration should be considered in children with recurrent LRTIs, especially with involvement of the right lung.

Table 3. A comparison of videofluoroscopic results of patients with and without neurometabolic disease				
		Neurometabolic disease		p-value
		Yes n=16 (%)	No n=16 (%)	
Thin Liquid	Oral phase dysfunction			
	Yes	12 (75)	6 (37.5)	0.037*
	No	4 (25)	10 (62.5)	-
	Delayed swallowing reflex			
	Yes	11 (68.8)	2 (12.5)	0.003
	No	5 (31.3)	14 (87.5)	-
	Nasal regurgitation			
	Yes	2 (12.5)	1 (6.3)	0.544
	No	14 (87.5)	15 (93.8)	-
	Aspiration			
	Yes	16 (100)	13 (81.3)	0.226
	No	-	3 (18.8)	-
	Coughing			
	Yes	3 (18.8)	4 (25)	1,000
	No	13 (81.3)	12 (75)	-
	Penetration aspiration scale			
	None	-	2 (12.5)	-
	Penetration	-	1 (6.3)	0.276
	Aspiration despite patient's response	3 (18.8)	4 (25)	-
	Aspiration absent patient's response	13 (81.2)	9 (56.3)	-
Thick Liquid	Oral phase dysfunction			
	Yes	13 (81.3)	4 (25)	0.004
	No	3 (18.8)	12 (75)	-
	Delayed swallowing reflex			
	Yes	9 (56.2)	2 (12.5)	0.006
	No	7 (43.8)	14 (87.5)	-
	Nasal regurgitation			
	Yes	2 (12.5)	-	0.484
	No	14 (87.5)	16 (100)	-
	Aspiration			
	Yes	5 (31.3)	1 (6.3)	0.333
	No	11 (68.8)	15 (93.8)	-
	Penetration aspiration scale			
	None	11 (68.8)	15 (93.8)	-
	Penetration	-	-	-
	Aspiration despite patient's response	-	-	0.172
	Aspiration absent patient's response	5 (31.3)	1 (6.3)	

Half of the patients without risk factors were exposed to cigarette smoke. Studies have shown that smoking is one of the etiologies of dysphagia in adults (15,16). Although this has not been observed in studies involving the pediatric population, the higher scores on the PEAT-10 and PAS of the patients exposed to cigarette smoke in our study suggest that smoking exposure may play a role. Although smoking has not yet been shown as an etiology of dysphagia in studies conducted in children, the higher scores on the PEAT-10 and PAS of the patients exposed to smoking in our study suggest that smoking exposure may also be involved in the etiology of swallowing dysfunction in children. Since smoking is also a risk factor for recurrent respiratory tract symptoms, suggestions to avoid smoking in the management of children with swallowing dysfunction may also be beneficial in the management of patients with persistent respiratory tract symptoms (17).

After changing the diet of the patients with swallowing dysfunction, the frequency of admission to the hospital due to LRTIs decreased. This shows the importance of swallowing therapies and nutritional recommendations in the management of patients with recurrent respiratory tract symptoms. Since multiple factors, such as other treatments received by the patients and environmental factors affecting the patients, could not be evaluated in our study, the decrease in the number of LRTIs cannot be attributed solely to changing the diet.

In patients with metabolic and/or neurologic disease, oral phase dysfunction and delay in the swallowing reflex were more common. Oral motor dysfunction is common in children with delayed neuromotor development. In these patients, problems such as sucking, chewing, swallowing, drooling, and persistent tongue thrust are frequently encountered (18,19). Oral motor dysfunction is seen in 90% of children diagnosed with cerebral palsy (20). Especially in patients with neurological disease, untreated nutritional problems can cause growth retardation, which may increase morbidity and mortality (18,21). Similarly, in our study, the weight of patients with metabolic and/or neurologic disease was lower compared to other patients and their age, and respiratory tract infections were more common. Many of these children with metabolic and/or neurologic diseases would benefit from a regular nutritional assessment and management as part of their overall care. In addition, patients without risk factors but with isolated swallowing dysfunction should be followed up for neurological diseases that may develop in the future. Swallowing dysfunction in infancy may be the first sign of associated neurological diseases or syndromes (9).

Management of pediatric dysphagia includes nutritional recommendations, medical and surgical interventions, positioning guidelines, and oral-motor/swallowing exercises. Underlying disease, neuromotor and behavioral development of children, and social and environmental factors affect the treatment recommendations of children (1,4). A multidisciplinary approach to the management of dysphagia in pediatric populations is required, involving physiotherapists, otolaryngologists, pediatric gastroenterologists, and pediatric pulmonologists (1). In addition to respiratory system symptoms, swallowing dysfunction can cause malnutrition, which can negatively affect cognitive and behavioral development in children (1,22). Successful management of swallowing disorders improves nutritional status and significantly reduces morbidity (23). The prognosis of patients with isolated swallowing dysfunction is very good in long-term follow-up (9). In our study, after the appropriate nutritional recommendations for our patients, there was a decrease in the number of lung infections and, accordingly, a decrease in the morbidity of the patients.

The retrospective design of the study and the small number of patients are the limitations of our study. There is a need for multicentre prospective studies with larger patient groups.

Conclusion

Swallowing dysfunction may cause recurrent respiratory tract symptoms in children. Patients with underlying risk factors should be evaluated for swallowing dysfunction. Swallowing dysfunction should be kept in mind in the differential diagnosis of patients with recurrent respiratory symptoms but without any risk factors for dysphagia. Early diagnosis of swallowing dysfunction and initiation of appropriate treatment is important and can reduce the morbidities associated with dysphagia.

Ethics

Ethics Committee Approval: The Gazi University Non-Interventional Clinical Research Ethics

Committee approved the study (protocol number 916100558-604.01.02).

Conflict of Interest: No conflict of interest was declared by the authors.

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