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Clinical characteristics and videofluoroscopic swallowing study findings in children with swallowing disorders

Charakterystyka kliniczna i wyniki wideofluoroskopowej oceny aktu połykania u dzieci z zaburzeniami połykania

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Key words

swallowing disorders, videofluoroscopic swallowing study, children

Słowa kluczowe

zaburzenia połykania, wideofluoroskopowa ocena aktu połykania, dzieci

Summary

Introduction. Swallowing disorders are a relevant but often unrecognized and underestimated problem in children. Sometimes they lead to aspiration pneumonia.

Aim. The aim of this study was to show the usefulness of the videofluoroscopic swallowing studies' (VFSS) findings in children with various diseases and symptoms of swallowing disorders based on experiences of the Department of Gastroenterology, Hepatology and Nutrition Disorders in Poland.

Material and methods. A total of 36 children were enrolled in this retrospective study. All patients presented swallowing disorders, therefore they were referred to VFSS. Indications for VFSS were defined by a physician and speech-language pathologist. The outcomes were reviewed by a radiologist and speech-language pathologist. Type of feeding, compensation or rehabilitation was recommended by speech-language pathologist. The swallowing problems, VFSS findings and recommendation after examination were analyzed.

Results. The reason for VFSS referral was "the safety of swallowing" (17 children) or the assessment the function of swallowing (15 children). For the rest of patients the reason for VFSS was the simultaneous evaluation of the safety and function of swallowing (4 children). 22 children (61%) presented with respiratory symptoms as a cause of swallowing disorders and the necessity for VFSS. Silent aspiration was observed in 15 of patients, aspiration with cough in 2, pharyngeal residue in 6, residue with penetration in 6 of all patients. The VFSS outcomes indicated the necessity to modify oral feeding in 19 children (53%). In 12 patients (33%) oral feeding was discontinued. Rehabilitation without oral feeding was ordered in 13 patient (36%), general swallowing rehabilitation with the oral use of foods in 11 children (30%). Compensation using different food consistency was used in 9 patient (25%), compensation by proper positioning and modified feeding technique was adopted in 16 children (44%). Feeding disorders therapy was ordered in 8 children (22%).

Conclusions. This method is helpful for defining and diagnosis the problem with swallowing. VFSS allows to choose a proper therapy and to determine the way of feeding accordingly to different pathophysiologic mechanisms of swallowing disorders in children.

Streszczenie

Wstęp. Zaburzenia połykania są istotnym, ale często niedocenianym i nierozpoznanym problemem u dzieci. Czasem prowadzą do zachłystowego zapalenia płuc.

Cel pracy. Celem pracy jest przedstawienie metod terapeutycznych i efektów wideofluoroskopowej oceny aktu połykania (VFSS) u dzieci z różnymi chorobami i objawami zaburzeń połykania leczonych w Klinice Gastroenterologii, Hepatologii i Zaburzeń Odżywiania w Polsce.

Materiał i metody. Do badania retrospektywnego zakwalifikowano 36 dzieci. Wszyscy pacjenci prezentowali zaburzenia połykania, w związku z czym zostali skierowani na badanie VFSS. Wskazania do VFSS zostały określone przez lekarza i logopedę. Wyniki zostały zweryfikowane przez radiologa i logopedę. Sposoby karmienia, kompensacji lub rehabilitacji zostały zalecone przez logopedę. Przeanalizowano problemy z połykaniem, wyniki VFSS oraz zalecenia po badaniu.

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Wyniki. Powodem wykonania VFSS było określenie „bezpieczeństwa połykania” (17 dzieci) lub ocena funkcjonalności połykania (15 dzieci). U pozostałych pacjentów powodem VFSS była jednoczesna ocena bezpieczeństwa i funkcjonalności połykania (4 dzieci). U 22 dzieci (61%) wystąpiły objawy z układu oddechowego wskazujące na konieczność wykonania VFSS. Ciche aspiracje zostały zauważone u 15 pacjentów, aspiracje z kaszlem u 2, zalegania w gardle u 6, zalegania wraz z penetracją u 6 spośród wszystkich pacjentów. Wyniki VFSS wskazały na konieczność zmodyfikowania karmienia doustnego u 19 dzieci (53%). U 12 pacjentów (33%) odstawiono karmienie doustne. Rehabilitacja bez karmienia doustnego została zalecona u 13 pacjentów (36%), ogólna rehabilitacja połykania ze stosowaniem karmienia doustnego u 11 dzieci (30%). Kompensacja z wykorzystaniem różnych konsystencji pokarmu została wykorzystana u 9 pacjentów (25%), kompensacja przez pozycjonowanie i zmodyfikowanie techniki karmienia została podjęta u 16 dzieci (44%). Terapię zaburzeń karmienia zalecono u 8 dzieci (22%).

Wnioski. Metoda jest przydatna w zakresie identyfikowania i diagnozowania problemu z połykaniem. VFSS pozwala na dokonanie wyboru właściwego leczenia i określenie sposobu karmienia odpowiedniego względem różnych patofizjologicznych mechanizmów zaburzeń połykania u dzieci.

INTRODUCTION

Swallowing is a phased process. It is composed of 3 to 5 phases, depending on the definition of the oral phase and its division into 2 or 3 stages, which are as following: a food intake process, bolus forming stage and initiation of pharyngeal phase (1, 2). According to the location of each phase, it is easier to distinguish 3 stages: the oral phase, the pharyngeal phase and esophageal phase. The oral phase is volitional, while the pharyngeal and esophageal ones are achieved according to the reflex actions. The crucial part of the oral phase is the food intake, its processing through crumbling it and melting it with saliva (depending on food consistency we can distinguish biting off, biting, crushing and chewing) and moving the bolus to the back of the oral cavity in order to initiate the pharyngeal phase. The oro-motor phase is carried directly due to the effective functioning of lips, tongue, jaw and cheeks, and indirectly through the face and neck muscles. The pharyngeal phase begins with the activation of the reflex actions, which protects the closing of the respiratory tract during this phase, and moving the bolus towards the pharyngo-esophageal sphincter (UES). In order to do so, the soft palate gets tensed, and together with the adjoining throat's walls form a palato-pharyngeal closure, which prevents the bolus from getting to the nasal cavity. Another reflex action simultaneously protects the entrance to the respiratory tract through the lifting of the larynx and pushing it forward, together with the closure of the epiglottis and the vocal cords. Larynx displacement is possible due to the prior move of the hyoid bone, this pulls the larynx and it is a trigger for UES opening. The passage of food through the UES to the esophagus is the beginning of the esophageal phase. This phase consists of the bolus passage to the stomach due to the esophageal peristalsis. Bolus passing through the gastro-esophageal sphincter is thought to be the last stage of the swallowing process. The motor control during the pharyngeal phase is conducted by the bottom muscles of the oral cavity, pharyngeal muscles, and muscles of the neck and larynx. The esophageal phase is controlled by UES muscles, esophageal muscles and LES muscles.

The neurological control of swallowing process involves different parts of nervous system – from the brain stem which is responsible for inborn swallowing reflexes, to the central uppermost level of the central nervous system, such as motor and pre-motor cortex, which takes over the control over food intake and swallowing upon child's maturation. Simultaneously, the motor habits are imprinted as well as the will-dependent motor skills (3).

The function of swallowing may be compared to the effective system of generated pressures and penstocks, which control the transit of food bolus from the front of the oral cavity to the stomach. The tightness and tension of mouth, esophago-pharyngeal closure, and gastro-esophageal sphincter are the components of the penstocks system helping in controlling the food transit. The functioning of the tongue, cheeks, clenching of pharyngeal muscles and peristalsis of esophagus create a system of generated pressures, which contributes to food transfer (sometimes against the gravity).

The swallowing disorders (dysphagia) may appear at any stage of the process and they may apply to either one or more phases of the swallowing process. The problems may include bolus formation, oral transit, initiation of the pharyngeal phase, transit to the esophagus, opening of the upper esophageal sphincter, transit through the lower esophageal sphincter. Particular concern relates to the timing and coordination deficits that may result in aspiration (4).

Oral phase disorders may lead to some problems with keeping food within the oral cavity and cleaning it off food. They also can cause inability to effective bite crumbling and its transition from the front part of the oral cavity to the back part, which often results in low feeding efficiency. During the pharyngeal phase, disorders may manifest through the food aspiration into the low respiratory tract (fig. 1), its penetration into the nasal cavity (fig. 2), and residue in the pharynx (fig. 3 and 4). When assessing the esophageal phase, the attention is usually paid to detect potential esophageal narrowings, tracheo-esophageal fistulas and reverse passage of food through UES to pharynx.



Fig. 1. Penetration (A) Passing in Aspiration (B).

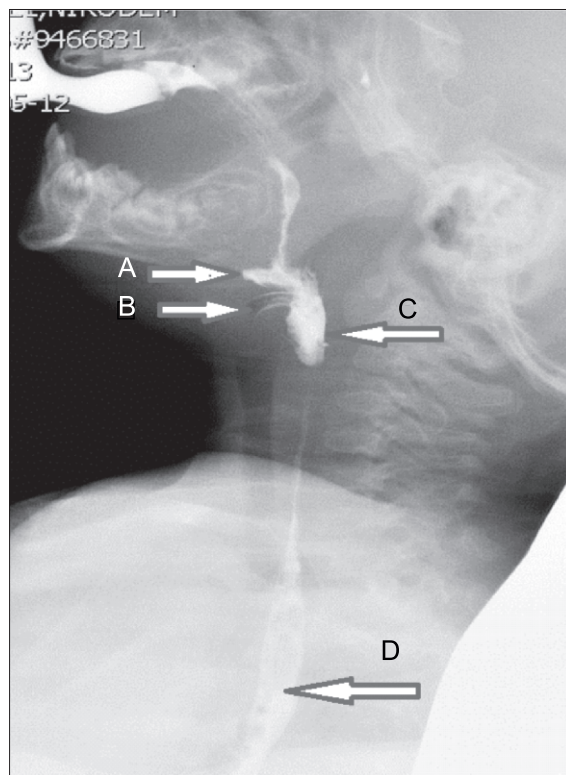


Fig. 3. Material in Valleculae (A), Penetration (B), Residue in Piriform Sinuses (C), Bolus in Esophagus (D).

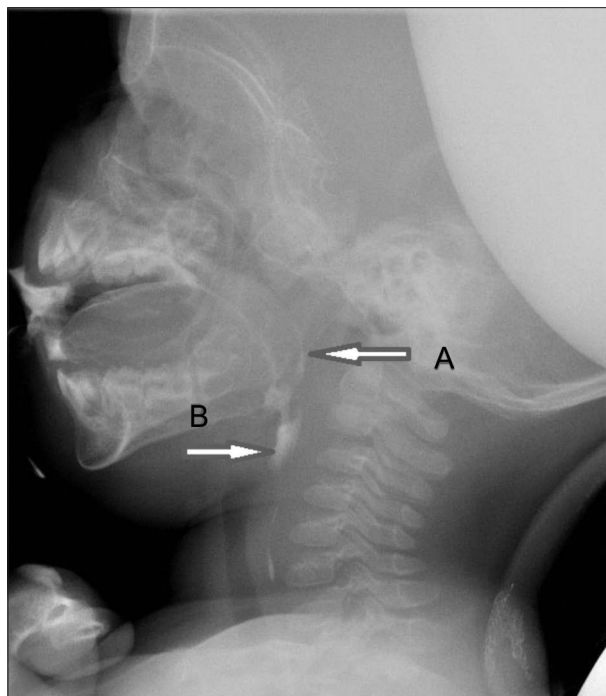


Fig. 2. Penetration in Nasopharynx (A) and Residue in Piriform Sinuses (B).

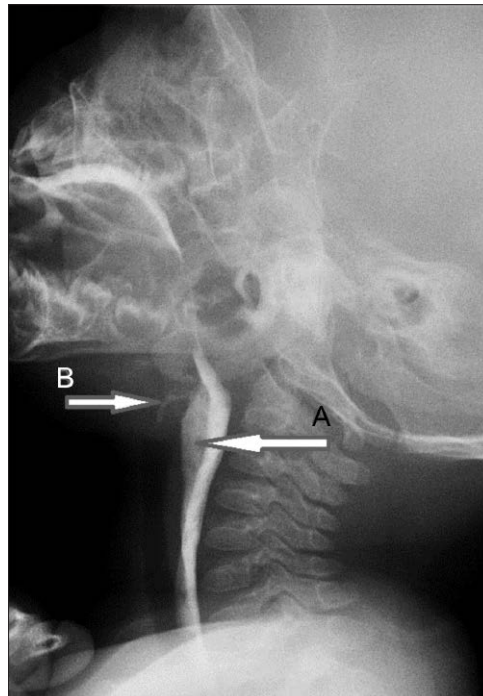


Fig. 4. Residue in Piriform Sinuses (A) with Penetration (B).

Videofluoroscopic evaluation of swallowing process can be used to evaluate each of the phases. During the oral phase, we check the functioning of the particular elements in oral cavity, transfer of the bolus to the back parts of oral cavity and effectiveness of oral cleaning. During the pharyngeal phase, we can evaluate the rapidity of the reflex action's activation due to the food

transit to the pharynx, coordination of successive activation of the reflex actions, the efficiency of closing and opening the particular elements of the pharynx, the effectiveness of respiratory tract protection against food aspiration and pharyngeal cleansing at the end of this phase. The evaluation of esophageal phase consists of evaluating the food passage through the esophagus (5).

The test consists of giving a contrast medium melted food to a patient, what enables assess swallowing act while using observation and recording on videotape the images appearing on a fluoroscopic screen. Since, a child has to take the food orally and swallow it voluntarily, a cooperation with patient is required in order to perform this examination. A speech therapist and radiologist are engaged to the examination. However, a presence and help of parents is often necessary. A food used in the VFSS should resemble the one which parents feed their child with. Apart from food intake and swallowing act, feeding technique is evaluated.

AIM

The aim of this study was to show the usefulness of the videofluoroscopic swallowing studies' (VFSS) findings in children with various diseases and symptoms of swallowing disorders based on experiences of the Department of Gastroenterology, Hepatology and Nutrition Disorders in Poland.

MATERIAL AND METHODS

Subjects

We enrolled 36 children with swallowing disorders treated in Department of Gastroenterology, Hepatology and Nutrition Disorders to the study, who underwent VFSS between April 2012 and October 2013. If a patient underwent VFSS more than once during the study period, only the first VFSS findings was analyzed.

Methods

Clinical characteristic including medical conditions associated with swallowing disorders, and the occurrence of symptoms from respiratory system were evaluated. All patients presented with swallowing disorders and despite of this, suffered from neurological, cardio-respiratory, anatomic-functional or gastrological problems. Indications for VFSS were defined by a physician and speech-language pathologist. Outcomes were reviewed by a radiologist and speech-language pathologist. A type of feeding was defined before and after VFSS. A type of compensation or rehabilitation was recommended by speech-language pathologist.

The problems of patients with dysphagia were divided into 4 groups: neurologic disorders, cardio-respiratory problems, anatomical and functioning disorders, and gastrological problems. The group of neurologic patients was not homogenous with regard to the symptoms intensity. It comprised of infantile cerebral palsy, drug-refractory epilepsy, ischemic and anoxic encephalopathy, psycho-motor retardation, and problems with appropriate muscle strength and tension. Among the patients with gastrological disorders, there were undernourished children, patients presenting with excessive vomiting, feeding disorders, and gastro-esophageal reflux disease. The cardio-respiratory disorders comprised heart defects, broncho-alveolar dysplasia, asthma, pulmonary hypertension, and respiratory failure. Esophagus atrophy, burn of oral cavity and esophagus,

larynx atresia, cleft palate or cleft lip belonged to anatomical and functioning disorders. Nine patients (25%) were included into more than one group because of concomitant diseases.

Another classification was based on patient's previous respiratory symptoms indicating potential risk of food aspiration or swallowing-breathing coordination disorders. Respiratory disorders, which might lead to swallowing problems, included pneumonia, bronchitis, recurrent upper respiratory tract infections, wheezing and whistling either during feeding or just after feeding process (6, 7). In this cases, the aim of the study was to determine the safety of swallowing (8). The indication for VFSS to evaluate swallowing process in these patients was an insufficient food intake in relation to daily requirements, frequently accompanied by malnutrition, as well as anatomical and functioning disorders influencing safety and efficiency of swallowing.

VFSSs were conducted as described by Arvedson with some modifications. The day before examination, speech language pathologist was observing child's natural feeding process; either while eating by him/herself or being fed by its parents/caregivers. Afterwards, a speech language pathologist was feeding a child by herself in order to determine the level of its hypersensitivity to food stimulus, as well as to observe child's reaction to a new situation connected with stranger who feed him. Moreover, the type of food, its consistency and feeding techniques were determined to work out an examination plan. Before VFSS the parent formal consent was obtained. During the examination, a child was sitting on its parent's laps and was fed by a speech language pathologist, however, in several cases, parents fed their child themselves.

Apart from its diagnostic aspect, this study also serves for the evaluation and verification of planned therapy methods for child's management (9, 10). Two methods were used: the method of compensation using different food consistency (11-14) and compensation by proper positioning and modified feeding technique. In some cases, post-examination orders concerned functioning rehabilitation called "Dry" Rehabilitation (rehabilitation without oral feeding) (15, 16) or swallowing rehabilitation with the oral use of foods – FR (17), as well as starting regular feeding disorders therapy (18).

In order to evaluate the level of swallowing disorders the Penetration-Aspiration Scale was used (tab. 1) (19). Lack of swallowing disorders was defined as lack of aspirations and penetrations with no pharyngeal food storage (score 1) (20, 21). Penetrations without food storage (score 2 or 4) were considered to be child's specific, and not to cause direct danger to safety swallowing at the moment. Penetrations accompanied by food storage (score 3 or 5) were classified to a category of potential danger leading to aspiration due to muscle fatigue (elongated feeding time in comparison to short feeding time during examination) and to influence swallowing functioning. This was also a reason

for selecting a separate category which included food storage only as a potential risk of aspiration and impaired swallowing functioning. Aspirations (score 6-8) were considered as dangerous in each case (22) and they were an indication to take up specific actions, such as compensative operations, rehabilitations and feeding technique changing. Silent aspirations were defined as aspiration without cough and choking.

Table 1. Penetration-Aspiration Scale by Rosenbek et al. (19).

Score	Neider penetration nor aspiration	
	1	Material does not enter airway
	Penetration	
	2	Material enters airway, but remains above vocal folds; Ejected from airway; no stasis
	3	Material remains above vocal folds; visible stasis remains
	4	Material contacts vocal folds, but is ejected; no stasis
	5	Material contacts vocal folds, and is not ejected; visible stasis remains
	Aspiration	
	6	Material passes glottis, but is ejected from airway; No visible subglottic stasis
	7	Material passes glottis, but is not ejected from airway; visible Subglottic stasis despite patient's response
8	Material passes glottis, and is not ejected; visible subglottic stasis; Absent patient response	

RESULTS

The results of clinical characteristics of patients (tab. 2) demonstrate, that the largest group included children with neurological disorders (23 children). Patients with gastrological problems and anatomical and functioning disorders constitute of 11 and 9 children respectively. The smallest group comprised patients with cardio-respiratory disorders. The number of children referred to VFSS in order to determine feeding safety (17 children) was comparable to the number of children referred to the examination to determine their swallowing process functioning (15 children). Simultaneous evaluation of the safety and functioning of the swallowing process was an indication for VFSS in 4 children (11%).

The respiratory symptoms indicating the swallowing disorders were observed in 22 of patients (tab. 3). The rest of the children had not presented with such

symptoms before. It is remarkable, that the largest group of swallowing problems were silent aspirations (15 children), among which, respiratory symptoms directly leading to aspiration, have not been noticed in all cases (3 children). The smallest group comprised of children with aspirations accompanied by coughing and choking (2 children). Pharyngeal food residue was registered in 12 patients (33%), half of which proceeded with penetrations, which increased an aspiration risk during child's feeding. Automatically cleansed penetrations were recorded in 4 patients. Three patients (8%) presented with no disorders.

Table 3. Respiratory symptoms and VFSS findings.

Symptoms / Findings	Respiratory symptoms n = 22 (61%)	Without respiratory symptoms n = 14 (39%)
Silence Aspiration n = 15 (42%)	12 (33%)	3 (8%)
Aspiration with Cough n = 2 (5%)	1 (3%)	1 (3%)
Residue n = 6 (17%)	4 (11%)	2 (5%)
Penetration n = 4 (11%)	1 (3%)	3 (8%)
Penetration and Residue n = 6 (17%)	4 (11%)	2 (5%)
Without n = 3 (8%)	0	3 (8%)

Therapeutic approach based on VFSS findings (tab. 4) in the largest number of cases led to change in feeding technique (16 children). 13 children (36%) received a recommendation of swallowing therapy without the use of food, and 11 patients during the therapy could benefit from the use of food as a factor supporting the therapeutic process. In the case of one quarter of patients the degree of compaction of food was recommended, which was conducted to the improvement of safety and efficiency of feeding. Feeding disorders therapy was started in 8 patients. More than one type of therapeutic support (compensation and rehabilitation) was used in 18 patients (half of all children).

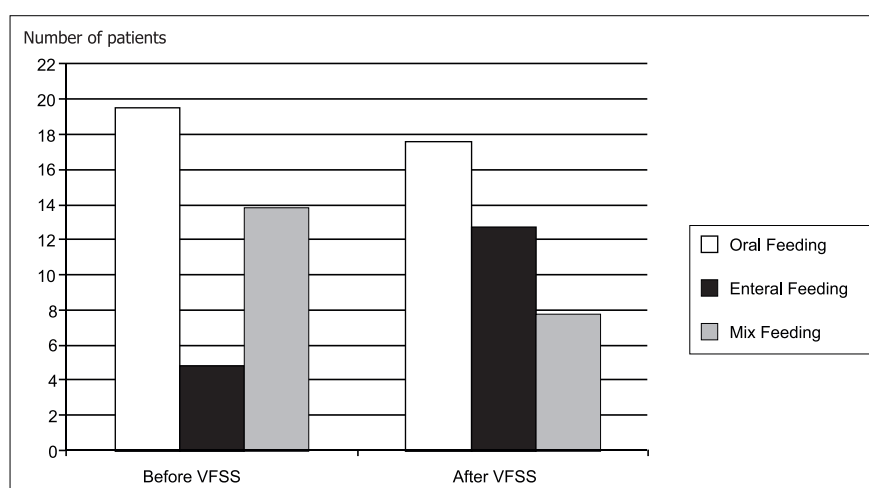
The results of the analysis of feeding technique preceding VFSS (fig. 5) have demonstrated, that the number of children fed exclusively orally has decreased (from 19 to 17), while the number of patients with exclusive enteral nutrition increased (from 4 to 12), and the number of patients fed both orally and through enteral nutrition changed from 13 to 7. The major changes con-

Table 2. Reasons for referral for VFSS and associated medical condition.

Medical Conditions / Reasons	Anatomical or Functional Problems n = 9 (25%)	Cardio-Respiratory Problems n = 5 (14%)	Neurological Problems n = 23 (64%)	Gastrological Problems n = 11 (30%)
Safety of Swallowing n = 17 (47%)	4	1	14	2
Functional of Swallowing n = 15 (42%)	5	3	5	8
Safety and Functional n = 4 (11%)	0	1	4	1

Table 4. Way of therapy and VFSS findings.

Way of Therapy Findings	Correction by Techniques and Positioning n = 16 (44%)	Compensation by Consistency n = 9 (25%)	"Dry" Rehabilitation n = 13 (36%)	Rehabilitation with Feeding n = 11 (30%)	Feeding Disorders Therapy n = 8 (22%)
Silence Aspiration	5	3	9	3	0
Aspiration with Cough	2	1	0	0	1
Residue	4	2	2	4	0
Penetration	2	1	0	1	4
Penetration and Residue	3	2	2	2	1
Without Swallowing Abnormality	0	0	0	1	2

**Fig. 5.** Way of feeding before and after VFSS.

cerning feeding techniques were the discontinuation of the oral feeding in children at the great risk of aspiration and low possibility of rapid effects of swallowing rehabilitation and when compensation with the change of feeding technique did not give expected effects visible in VFSS. Among 12 children with discontinuation of the oral feeding, 8 presented with silent aspirations, 2 with pharyngeal residue and 2 with simultaneous pharyngeal residue and penetrations. In 4 cases (11%) patients had exclusive enteral nutrition before VFSS, and the results of the examination have demonstrated that it was feasible to introduce oral feeding. In 5 cases (14%), the results of VFSS showed the need for enteral nutrition introduction in patients with exclusive oral feeding. In general, the way of food supply has been changed in 21 children (58%).

DISCUSSION AND CONCLUSIONS

The analysis was based on the group of patients referred to the Department of Gastroenterology, Hepatology and Feeding Disorders, Children's Memorial Health Institute in Warsaw. The results of this analysis should be regarded rather as a presentation of diagnostic and therapeutic methods for treatment of feeding disorders with the use of VFSS, than a characteristics of patients with swallowing disorders. This is a limitation of this study. Nevertheless, our results are consistent with the outcomes of the study by Wair

et al., who has demonstrated that the vast majority of children presenting with aspiration symptoms, are patients with silent aspirations (81%), and most of them are children with neurological dysfunctions (23).

Silent aspirations are particularly difficult to diagnose. Apart from clinical symptoms, such as moist voice, buzzing, stridor, coughing, choking, recurrent pneumonia and/or bronchitis, frequent episodes of laryngitis and upper respiratory tract infections, diagnostic methods are being extended of acoustic analysis of sounds uttered while breathing (24) or saturation decrease (25). However, none of these methods provide such strong evidence of silent aspirations, as VFSS does. Another examination comparable to VFSS with respect to diagnostics accuracy is Fiberoptic Endoscopic Evaluation of Swallowing (FEES), which is an invasive procedure not always feasible in infants. Besides, while FEES enables finding aspirations after swallowing, its accuracy decreases in case of liquids (26). Moreover, FEES serves only to evaluate the process before and after swallowing, not a swallowing functioning itself (27).

Our analysis demonstrates that the aspirations not always manifest with respiratory symptoms, and that other factors of swallowing process evaluation may help in swallowing safety and functioning assessment. Food residue with or without penetrations was second most frequent swallowing disorder found during VFSS. In this case, we used the ability of VFSS to assess pharyngeal residue and

cleaning which helps in evaluation of discomfort level and aspiration risk during feeding process. Such conclusions are very important for the therapeutic proceedings.

The diagnostic value of VFSS is to evaluate the feeding technique and compensation methods. The results of the analysis demonstrate the variety of therapeutic approaches, which is the result of specific functioning of each child. Although being aware that neurologic disorders can cause malnutrition and silent aspirations which can lead to pneumonia (28), we still need to know how to prevent consequences of dysphagia or, at least, reduce the problems following this disorder. How to found the right therapeutic method accordingly to specified patient? Some studies showed that in patients with similar basic problems can be recorded different pattern of swallowing disorders. Comparison of the two groups of neurological patients with vocal cord paralysis, either of central or peripheral nature, has shown that despite of pharyngeal dysphagia, patients with peripheral paralysis had additionally more frequently oral phase dysfunctions, while the latter ones more frequently presented with poorer pharyngeal movability (29). On the other hand, study which has used VFSS to compare character of dysphagia in children with spastic cerebral palsy, dyskinetic cerebral palsy and neuromuscular disorders, has demonstrated that in cerebral palsy, swallowing incoordination which required thickened food use is a major problem, while patients with neuromuscular disorders more frequently suffered from muscle fatigue, and thus, food condensed is not recommended for them due to high risk of pharyngeal food storage (30). The examples mentioned above demonstrate the variety of swallowing disorders. Therefore, without proper diagnostics and therapeutic tools, such as VFSS, it would be difficult to establish the most appropriate management for the individual patient.

Long-term results concerning treatment of dysphagia seem to be very promising. Five years retrospective evaluation of 90 children has shown swallowing

improvement in 74 patients. It was strongly correlated with both neurologic and nutritional condition (17). On the other hand, the analysis of children without neurological disorders, but with recurrent respiratory infections caused by aspirations, has demonstrated that only 4 out of 13 patients after first VFSS had their oral food discontinued. However, after 3-9 months, following second VFSS, all children well tolerated oral feeding (31). Such specifications seem to be particularly important regarding the fact that this analysis has demonstrated that after VFSS, the number of children on exclusive enteral nutrition has increased. There was also a group of patients who had additional enteral nutrition due to their inability to cover orally their whole daily caloric requirement. This confirms that patients with dysphagia should pass swallowing therapy and VFSS can help in determining how to proceed.

The limitation of VFSS is its requirement of patient's cooperation. Despite parent's assistance during examination, a procedure itself and sensory hypersensitivity make acquisition of reliable results difficult. In these cases, the tools of indirect evaluation are usually recommended, but their accuracy is not 100% (32, 33).

VFSS is frequently considered as a "golden standard" for evaluation of the swallowing disorders. Therefore, verifications are needed in order to standardize the protocol of examination. It is possible to achieve in adult patients however it seems that in children, the individualized procedures are far more frequent. It is mostly caused by the fact that pediatric dysphagia is connected with not sufficient patient's cooperation, lack of standards and more frequent behavioral disorders than in adults. Still, the ability to evaluate swallowing disorders and therapeutic use of VFSS seem to be very useful.

VFSS is helpful for defining and diagnosis the problem with swallowing. VFSS allows to choose a proper therapy and to determine the way of feeding accordingly to different pathophysiologic mechanisms of swallowing disorders in children.

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received/otrzymano: 20.12.2013
 accepted/zaakceptowano: 06.02.2014