Presenting Signs and Symptoms do not Predict Aspiration Risk in Children

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Objectives To determine if any presenting symptoms are associated with aspiration risk, and to evaluate the reliability of clinical feeding evaluation (CFE) in diagnosing aspiration compared with videofluoroscopic swallow study (VFSS).

Study design We retrospectively reviewed records of children under 2 years of age who had evaluation for oropharyngeal dysphagia by CFE and VFSS at Boston Children's Hospital and compared presenting symptoms, symptom timing, and CFE and VFSS results. We investigated the relationship between symptom presence and aspiration using the Fisher exact test and stepwise logistic regression with adjustment for comorbidities. CFE and VFSS results were compared using the McNemar test. Intervals from CFE to VFSS were compared using the Student *t* test.

Results A total of 412 subjects with mean (±SD) age 8.9 ± 6.9 months were evaluated. No symptom, including timing relative to meals, predicted aspiration on VFSS. This lack of association between symptoms and VFSS results persisted even in the adjusted multivariate model. The sensitivity of CFE for predicting aspiration by VFSS was 44%. Patients with a reassuring CFE waited 28.2 ± 8.5 days longer for confirmatory VFSS compared with those with a concerning CFE ($P < .05$).

Conclusions Presenting symptoms are varied in patients with aspiration and cannot be relied upon to determine which patients have aspiration on VFSS. The CFE does not have the sensitivity to consistently diagnose aspiration so a VFSS should be performed in persistently symptomatic patients. *(J Pediatr 2018;201:141-6).*

Infants and children are typically referred for swallow evaluation if they have signs or symptoms suspicious for aspiration.[1-3] These symptoms typically include coughing, choking, eyes turning red, difficulty feeding, or changes in color with feeding.[4] Little is known about the actual correlation between presenting symptoms and the risk of finding aspiration either by clinical feeding evaluation (CFE) or videofluoroscopic swallow study (VFSS).[4,5] The CFE typically consists of assessing feeding with 1 or more textures (eg, thin, nectar, honey thick, or purees) using 1 or more methods of feeding (eg, bottle, cup, spoon) by a speech-language pathologist (SLP) specializing in the treatment of pediatric dysphagia and feeding disorders.[3,6] A VFSS typically involves similar trials though the feeding is assessed using fluoroscopy of the oropharynx, larynx, and upper esophagus to determine if there is evidence of aspiration.[7-9] There is limited data on the sensitivity of CFE compared with the VFSS to assess for aspiration risk, and prior studies have only included small numbers of patients.[4,5,10-13]

Objective assessment of swallow function is critical in children with chronic respiratory symptoms because some of the classic signs of aspiration such as aspiration pneumonia are rare, occurring in less than 10% of children.[14-16] Determining the best method to assess for aspiration risk is not known, and each method has pros and cons. The VFSS can assess if there is direct aspiration or laryngeal penetration because the airways are visualized, but involves radiation exposure.[17-19] Although the CFE does not involve radiation risk, it can only identify signs and symptoms during feeding. This is not ideal because more than 80% of pediatric aspiration is silent and, therefore, occurs without overt clinical signs.[11,20-22] Choosing the most sensitive test is critical because inadequately treated aspiration can lead to a variety of poor outcomes including pulmonary injury, failure to thrive, and oral aversion.[1,23-25] The aim of this study was to describe the range of symptoms in children presenting for both CFE and VFSS and determine if any presenting symptoms, and the timing of those symptoms relative to meals, could predict aspiration risk in the pediatric population. An additional aim was to determine the reliability of the CFE in making the diagnosis of aspiration compared with VFSS in children.

Methods

We retrospectively reviewed the records of all children under 2 years of age who had both a CFE and VFSS for the evaluation of oropharyngeal dysphagia at Boston Children's Hospital in 2015. Records were reviewed for patient characteristics, comorbidities, and swallow study characteristics including radiation dose. VFSS

CFE	Clinical feeding evaluation
SLP	Speech-language pathologist
VFSS	Videofluoroscopic swallow study

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results were considered abnormal if there was evidence of aspiration or laryngeal penetration seen for any texture. Laryngeal penetration was considered abnormal based on our clinical experience that these patients have similar outcomes to patients with frank aspiration.[14,26] All CFEs were performed by speech language pathologists specializing in pediatric dysphagia, and all VFSS were performed by SLPs in conjunction with pediatric radiologists. The CFE and VFSS examinations were performed in standard fashion, starting with evaluation of thin liquids followed by increasing the thickness of liquids delivered in stepwise fashion (from thin to nectar to honey to puree) if there is concern for aspiration/penetration, as previously described.[6-9]

The primary aims were to determine if presenting symptoms could determine which patients would be at greatest risk for having an abnormal VFSS and whether the CFE could reliably predict aspiration or laryngeal penetration such that radiation exposure might be avoided. Presence of symptoms was obtained from the medical record based on parental and SLP report and included gastrointestinal symptoms and pulmonary symptoms in addition to how symptoms were related to meals (during, after, or both). We first described the prevalence of presenting symptoms in this cohort and then used the Fisher exact test to determine if there was any association between each individual symptom and the result of each subject's CFE and VFSS. A stepwise logistic regression model was used to determine symptoms independently associated with CFE and VFSS, after adjustment for age at VFSS, male sex, and all comorbidities (neurologic, cardiac, metabolic, immunologic, pulmonary, gastrointestinal, prematurity). The Firth penalized maximum likelihood estimation was used to reduce bias because of sparse table cells.[27] In addition, a multiple logistic regression model containing all presenting symptoms, adjusted for age at VFSS, male sex, and comorbidities (neurologic, cardiac, metabolic, immunologic, pulmonary, gastrointestinal, prematurity), using the Firth penalized maximum likelihood estimation, was used to obtain Wald χ^2 results and *P* values to put all symptoms in a single model to determine the relative strength of each effect.

We next compared the dichotomous assessment (normal vs abnormal) of CFE with VFSS as the gold standard to report test characteristics, including sensitivity, specificity, positive predicted value, and negative predicted value with 95% CI, and used the McNemar test to assess the concordance between these 2 modalities. Lastly, we used the Student *t* test to compare the time in days from initial CFE with initial VFSS for subjects who were ultimately found to have aspiration to determine the delay in aspiration diagnosis as a result of having a normal CFE. Data are presented as mean ± SE and % (n) unless indicated otherwise. Data were analyzed using SPSS (SPSS Inc, Chicago, Illinois) and multivariate analysis was conducted with SAS (SAS Institute, Cary, North Carolina). The study was approved by the Institutional Review Board at Boston Children's Hospital.

Results

We evaluated 412 total subjects with a mean age of 8.9 ± 6.9 months, all of whom had VFSS performed; 160 of these had both CFE and VFSS performed. Within the entire cohort, 38% (n = 156) of the VFSS showed aspiration, 33% (n = 137) showed penetration alone, and 27% (n = 107) did not show evidence of aspiration or penetration; 3% (n = 12) of subjects were unable to complete their VFSS. Subject characteristics, symptoms present at the time of referral, and subject comorbidities are shown in **Table I**. Notably, subjects were symptomatic for a mean (±SD) of 5.3 ± 5.0 months prior to their first formal swallow evaluation. Overall, 29% of the subjects had a neurologic comorbidity and 32% of the subjects were premature with a mean gestational age of 31.9 ± 0.3 months. A flow diagram of the patient population, including the overall rates of swallow testing by VFSS and CFE and the results of these evaluations, is shown in the **Figure**.

A total of 234 patients had radiation exposure reported in the VFSS results, with a mean exposure of 1.98 ± 1.30 mGy. There were significantly lower exposure values in subjects with normal VFSS (1.54 ± 0.12 mGy) compared with those with an abnormal VFSS (2.20 ± 0.11 mGy, $P < .0001$).

The association between individual symptoms and VFSS results are shown in **Table II**. No single symptom predicted

Table I. Subject characteristics and presenting symptoms

Patient characteristics	All subjects (n = 412)	Abnormal VFSS (n = 293)	Normal VFSS (n = 107)
Male	59% (243)	60% (177)	55% (59)
Age at VFSS	9.0 ± 0.4	8.8 ± 0.4	8.8 ± 0.7
Duration of symptoms prior to VFSS	5.3 ± 0.6	5.5 ± 0.7	4.3 ± 0.9
Abnormal VFSS result	71% (293)	100% (293)	0% (0)
Aspiration	38% (156)	38% (156)	0% (0)
Silent aspiration	81% (127/156)	81% (127/156)	0% (0)
Penetration	33% (137)	33% (137)	0% (0)
Comorbidities			
Neurologic	29% (120)	31% (90)	27% (25)
Cardiac	11% (46)	11% (33)	9% (10)
Metabolic	13% (53)	13% (37)	13% (14)
Immunologic	1% (3)	1% (2)	1% (1)
Pulmonary	14% (58)	16% (46)	11% (12)
Gastrointestinal	21% (81)	17% (50)	26% (28)
Prematurity	32% (130)	34% (100)	27% (29)
GI symptoms			
Choking/gagging	37% (153)	38% (112)	36% (38)
Regurgitation	29% (121)	28% (81)	33% (35)
Vomiting	27% (112)	25% (72)	33% (35)
Poor feeding	23% (94)	22% (63)	26% (28)
Slow feeding	6% (24)	6% (16)	8% (8)
Pulmonary symptoms			
Coughing	58% (239)	59% (173)	52% (56)
Noisy breathing	25% (104)	28% (81)	20% (21)
Congestion	21% (87)	20% (58)	24% (26)
Spells	17% (68)	18% (53)	14% (15)
Respiratory distress	12% (50)	13% (38)	11% (12)
Recurrent pneumonia	11% (44)	12% (34)	7% (7)
Oxygen requirement	5% (19)	6% (16)	3% (3)
Relationship to meals			
Only during	53% (217)	55% (162)	46% (49)
Only after	8% (34)	9% (25)	8% (9)
During and after	21% (87)	20% (58)	24% (26)
No relationship to meals	18% (74)	16% (48)	22% (23)

Baseline characteristics, VFSS results, and comorbidities are shown above. There were varied presenting symptoms for the cohort overall and those with abnormal and normal VFSS results are shown. Data are expressed as percentage (n) and mean ± SE. The total number of patients includes 12 patients who could not complete the VFSS. Therefore the abnormal VFSS column plus the normal VFSS column do not always add to the total number of patients.

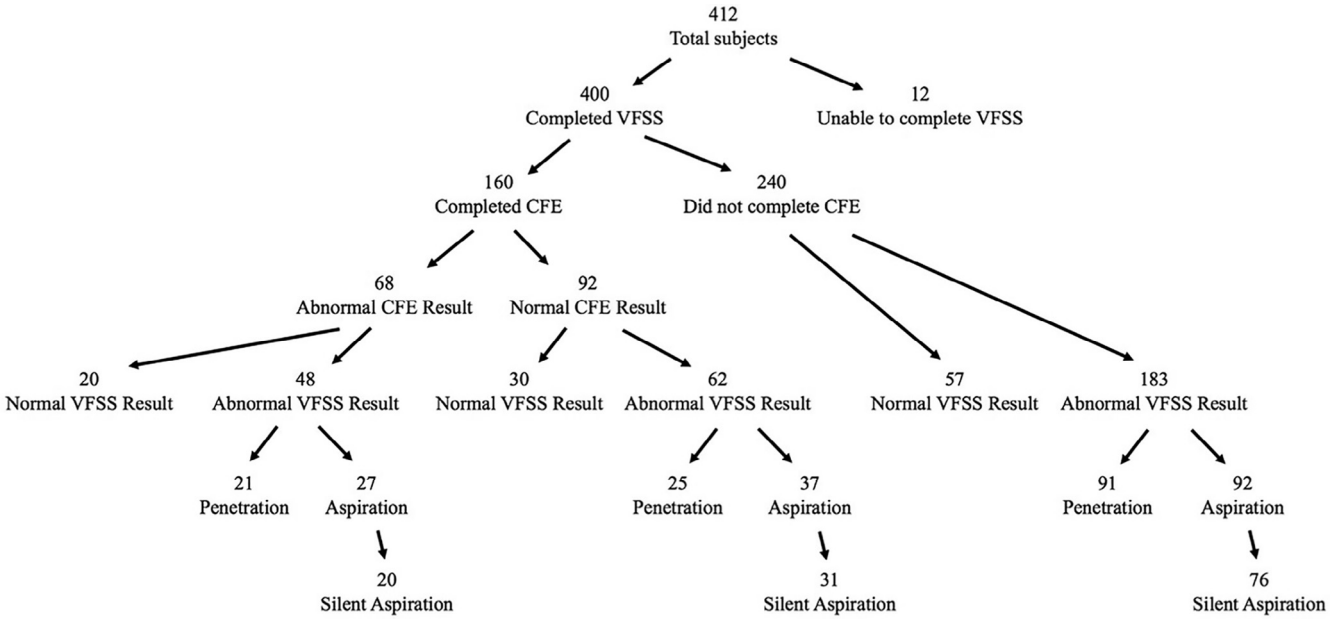

Figure. Study population. The flow diagram above shows the numbers of subjects in each group, including the overall rates of swallow testing by VFSS and CFE and the results of these evaluations.

risk of aspiration (all $P \geq .05$) by VFSS; even timing of symptoms relative to meals did not predict aspiration risk. In contrast, symptoms of coughing (OR 2.67, 95% CI 1.4-5.09, $P = .003$), choking/gagging (OR 2.412, 95% CI 1.248-4.666, $P = .012$), noisy breathing (OR 2.55, 95% CI 1.135-5.730, $P = .025$), and the presence of symptoms during meals (OR 3.36, 95% CI 1.37-8.26, $P = .008$) were significantly associated with CFE results suggesting that presenting symptoms may bias results of the CFE which did not then correlate with the VFSS.

No presenting symptoms were associated with an abnormal VFSS result even when combined in a multivariate model with adjustment for comorbidities as shown in **Table III**. A best-fitting model by stepwise regression identified only

Table II. Association between presenting symptoms and VFSS

Symptom characteristics	VFSS result: OR for abnormal VFSS	95% CI	*P* value
Gastrointestinal symptoms			
Choking/gagging (n = 150)	1.12	0.71-1.78	.64
Reflux (n = 116)	0.79	0.49-1.27	.32
Vomiting (n = 107)	0.67	0.41-1.09	.13
Poor feeding (n = 91)	0.77	0.46-1.29	.35
Slow feeding (n = 24)	0.72	0.30-1.72	.49
Pulmonary symptoms			
Coughing (n = 229)	1.31	0.84-2.05	.25
Noisy breathing (n = 102)	1.57	0.91-2.69	.12
Congestion (n = 84)	0.77	0.45-1.30	.33
Spells (n = 68)	1.35	0.73-2.52	.37
Respiratory distress (n = 50)	1.18	0.59-2.35	.73
Recurrent pneumonia (n = 41)	1.88	0.81-4.37	.19
Oxygen requirement (n = 19)	2.00	0.57-7.01	.43
Relationship to meals			
During meals (n = 295)	1.38	0.84-2.27	.24
After meals (n = 119)	0.80	0.50-1.29	.39
During and after meals (n = 84)	0.78	0.46-1.32	.41

OR for each presenting symptom and abnormal VFSS results are shown above. No single clinical symptom predicted risk of aspiration by VFSS. However, symptoms of choking/gagging, coughing, noisy breathing, and symptoms during meals were significantly correlated with CFE results but not with the gold standard VFSS results.

Table III. Association between presenting symptoms and VFSS after adjustment for comorbidities

Symptoms	VFSS result: Normal (n = 107)	Abnormal (n = 293)	$P_{unadjusted}$	$P_{adjusted}$
Choking/gagging	38 (35.5%)	112 (38.2%)	.64	.44
Reflux	35 (32.7%)	81 (27.7%)	.32	.43
Vomiting	35 (32.7%)	72 (24.6%)	.13	.27
Poor feeding	28 (26.2%)	63 (21.5%)	.35	.44
Slow feeding	8 (7.5%)	16 (5.5%)	.49	.34
Coughing	56 (52.3%)	173 (59.0%)	.25	.16
Noisy breathing	21 (19.6%)	81 (27.7%)	.12	.15
Congestion	26 (24.3%)	58 (19.8%)	.33	.45
Spells	15 (14.0%)	53 (18.1%)	.37	.36
Respiratory distress	12 (11.2%)	38 (13.0%)	.73	.90
Recurrent pneumonia	7 (6.5%)	34 (11.6%)	.19	.24
Oxygen requirement	3 (2.8%)	16 (5.5%)	.43	.54
During meals	75 (70.1%)	220 (75.1%)	.24	.28
After meals	36 (33.6%)	83 (28.3%)	.39	.38
During and after meals	26 (24.5%)	58 (20.2%)	.41	.43

No symptoms were associated with VFSS results even when assessed in a multivariate model with adjustment for comorbidities. Data are expressed as n (percentage). $P_{unadjusted}$ by the Fisher exact test. $P_{adjusted}$ by a logistic regression model containing a single symptom, adjusted for age at VFSS, male sex, and comorbidities (neurologic, cardiac, metabolic, immunologic, pulmonary, gastrointestinal, prematurity), using the Firth penalized maximum likelihood estimation to reduce bias because of sparse table cells.

gastrointestinal comorbidity as independently associated with abnormal VFSS (OR 0.56, 95% CI 0.33-0.96, $P = .03$). A similar model identified neurologic comorbidity, vomiting and slow feeding as independently associated with aspiration on VFSS (OR 2.1, 95% CI 1.34-3.31, $P = .001$ for neurologic comorbidity, OR 0.59, 95% CI 0.36-0.97, $P = .04$ for vomiting, and OR 0.28, 95% CI .09-0.85, $P = .02$ for slow feeding).

No presenting symptoms were associated with abnormal VFSS result even when combined in a multiple logistic model using Wald χ^2 analysis (all $P > .08$).

There was poor agreement between the 2 assessments of swallow function by the McNemar test ($P < .0001$). Even when we only included those patients with aspiration (excluding patients with penetration only) on VFSS, there was still poor agreement ($P = .03$). Follow-up CFEs compared with follow-up VFSS testing were also found to show poor concordance ($P = .0004$). Using the VFSS as a gold standard, the CFE had a sensitivity 44% (34%-53%), specificity 60% (45%-74%), positive predictive value 71% (62%-78%), and negative predictive value 33% (27%-39%) for predicting aspiration. Similar results were found even when we only compared CFE for those patients with aspiration (without penetration) on VFSS, with sensitivity 42% (30%-55%), specificity 60% (45%-74%), positive predictive value 58% (46%-68%), and negative predictive value 45% (37%-53%).

The time from the initial CFE to VFSS was 55.9 ± 8.5 days on average in patients with a CFE that did not raise concerns for aspiration. In contrast, in patients in whom the CFE was concerning for aspiration, there was a mean 27.7 ± 7.6-day time lag between CFE and the VFSS, which was significantly shorter than in patients with a CFE that did not raise concerns. Therefore, patients with a reassuring CFE waited 28.2 ± 8.5 days longer for confirmatory VFSS compared with those with a concerning CFE ($P < .05$).

Discussion

In the present study, we evaluated presenting symptoms for children under 2 years of age who had their first CFE and VFSS, and compared the agreement between these 2 modes of swallow evaluation, and determined the ability of each presenting symptom to predict VFSS and CFE results. We found that there was no single symptom that could reliably predict which patients would have evidence of aspiration on VFSS, and this translates to decreased sensitivity of the CFE compared with the VFSS.

Relatively few studies have examined the presentation and epidemiology of swallowing dysfunction in young children; this remains both an understudied and underappreciated area in clinical pediatrics, and the prevalence of oropharyngeal dysphagia appears to be increasing due to increased survival of premature infants and other children with chronic medical problems.[2,28,29] We focused our study on infants and children under age 2 years because this group has the highest rate of oropharyngeal dysphagia of any pediatric age group. In addition, their symptoms are often nonspecific, making diagnosis and management difficult for the general or specialist provider.[1,30] Pediatricians may treat patients with swallowing dysfunction with medications for reflux, perhaps because of frequent symptom overlap, which might do more harm than good.[31,32] Our group recently showed that oropharyngeal dysphagia and aspiration are associated with brief resolved unexplained events (formerly known as apparent life threatening events), suggesting that this disorder may play a significant role in common pediatric events.[12] In most cases, oropharyngeal dysphagia with aspiration can be treated with thickening of feeds in the great majority of pediatric patients, and continued oral feeding with thickened liquids has superior outcomes than feeding with enteral tubes.[33] However, to treat oropharyngeal dysphagia appropriately, it first must be diagnosed correctly.

Weir et al have consistently shown the high prevalence of silent aspiration in the pediatric population and suggested the need to consider VFSS to appropriately diagnose oropharyngeal dysphagia, but this practice has not become standard of care in many pediatric centers.[4,11] Svystun et al recently reviewed their cohort of 128 otherwise healthy children with aspiration and described the range of presentations in addition to varied diagnostic and management strategies in this patient population with similar age range and rates of VFSS abnormalities.[5] They reported choking, coughing, and respiratory distress in more than 60% of their patients and also notably found vomiting as a presenting symptom in 26% of their cohort. In contrast to our results, however, they did find an association between recurrent pneumonia and abnormal VFSS.[5] Several groups have examined characteristics of children with swallowing dysfunction and consistently identified neurologic impairment as a major risk factor as we have shown in the current study.[30,34]

Many of what have traditionally been thought of as typical presenting symptoms of aspiration, such as recurrent pneumonia, were actually relatively infrequent in this cohort, and symptoms that are not typically considered, such as vomiting, were more common. In addition, almost one-third of patients had symptoms either only after meals or both during and after meals, which may explain why oropharyngeal dysphagia with aspiration is so frequently misdiagnosed as gastroesophageal reflux disease. These varied symptoms suggest that providers should be mindful of the myriad presentations of oropharyngeal dysphagia in this age group.

Our results also show that one-third of children with a normal CFE were actually found to have aspiration on VFSS. Because of the high prevalence of silent aspiration in children, patients with persistent symptoms, even with a reassuring CFE, may still need a VFSS to diagnose aspiration. A high index of suspicion is needed if symptoms persist to order a VFSS to avoid a delay in diagnosis. A systematic review showed poor agreement between the CFE and VFSS but even the largest study included only 91 subjects.[35] Other research groups have attempted to modify the approach to the CFE to improve its accuracy but even the addition of maneuvers such as cervical auscultation have not been shown to improve its sensitivity to the extent that the CFE could be used in isolation.[36]

In addition to merely assessing for the presence or absence of aspiration, there are multiple other components to the clinical feeding evaluation that should be considered. The CFE assesses the oral phase of swallowing to evaluate oral structures and document current oral motor skill level and developmental feeding skills. The CFE also assesses the patient's ability to receive, contain, and propel the bolus to determine appropriate diet level.[6] Therefore, the CFE remains a valuable tool that contributes significantly to the management of these patients.

There are a number of practical implications from our findings. First, specific presenting symptoms cannot be relied on in our pre-test consideration of whether a child will be found to have aspiration on their swallow study. None of the common symptoms predicted aspiration and, conversely, none of the symptoms ruled-out aspiration. In addition, providers should not be fully reassured by a normal CFE in the case of troublesome or ongoing symptoms. Another important finding was the relatively low radiation exposure involved in obtaining VFSS, especially in children who go on to have a normal VFSS result. Our results suggest that the exposure is significantly less than an upper gastrointestinal series.[17] Pediatric radiologists continue to work with SLPs to minimize radiation in performing these studies and other groups have already shown that radiation exposure can be decreased in the performance of VFSS.[17,18,37] Lastly, the children in our study were symptomatic for a mean of more than 5 months prior to having formal swallow evaluation performed, suggesting there might be room for earlier diagnosis of oropharyngeal dysphagia, which might prevent morbidity. A multidisciplinary approach for these patients is essential to optimize their care, limit unnecessary testing, and maximize their future pulmonary and feeding outcomes.[3,23,38]

There are a number of limitations to the present study that should be considered. First, the retrospective nature of the study makes it difficult to know if there might be other historical characteristics or presenting features that might reliably discriminate between different presentations of oropharyngeal dysphagia. In addition, we only had records for those patients who ultimately went on to receive a videofluoroscopic swallow study and, therefore, might be missing some proportion of patients who only had a clinical evaluation because of the mild degree of symptoms or in whom their symptoms could be modified by changes in feeding techniques such as pacing without needing a VFSS. Lastly, the children in the study all received their care at a tertiary children's hospital, which might bias the study group toward a more medically complex population. However, the majority of the subjects included did not have any comorbidities and we did control for these in our multivariate model.

Presenting symptoms are varied in patients with aspiration and these symptoms cannot be relied upon to determine which patients have oropharyngeal dysphagia and aspiration. The evaluation of oropharyngeal dysphagia in young children should always include an objective assessment of VFSS as clinical feeding evaluations are inadequate to assess swallow function and lead to delays in making a diagnosis of aspiration. ■

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