Pediatric Pulmonology 41:1040–1048 (2006)

Long-Term Follow-Up of Oropharyngeal Dysphagia in Children Without Apparent Risk Factors

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Background. The presence of swallowing dysfunction in children without obvious risk factors remains under appreciated. Early identification and prompt initiation of appropriate treatments are critical for reduction of morbidities associated with dysphagia.

Objective. To describe the clinical presentations, radiologic characteristics, and long-term outcomes in children with oropharyngeal dysphagia presenting as unexplained respiratory problems. We completed a retrospective chart review of all children without known dysphagic risk factors upon presentation to Speech-Language Pathology (December 1991–April 1995) for feeding/swallowing evaluations because of refractory respiratory problems and dysphagic concerns, and who subsequently were diagnosed with dysphagia on Videofluoroscopic Swallow Study (VFSS). In August 2002, follow-up telephone interviews were conducted with caregivers of 14 children.

Results. We identified 19 children (mean age 1.14 years; range 0.9–5.75) with dysphagia presenting as unexplained respiratory problems. On VFSS, delayed pharyngeal swallow onset was the most common abnormal radiologic finding and always preceded penetration or tracheal aspiration. Eleven (57.9%) children aspirated. Aspiration occurred only with liquids and 100% of aspiration events were silent (i.e., no cough). Dysphagia was not a concern in 11 children at a mean age 3.2 years (range 0.7–10) and persisted in three children who were 9 years or older.

Conclusions. Oropharyngeal dysphagia should be considered in the differential diagnosis of young children without known risk factors associated with swallowing dysfunction when they present with unexplained respiratory problems. Although the prognosis for resolution of dysphagic concerns is very good, it may take several years. **Pediatr Pulmonol. 2006; 41:1040–1048.** © 2006 Wiley-Liss, Inc.

Key words: deglutition; deglutition disorders; swallowing; silent aspiration; videofluoroscopy; pediatric; dysphagia.

INTRODUCTION

Infants and young children with feeding and swallowing dysfunction are at increased risk for acute insidious development of aspiration-induced chronic lung disease, malnutrition, neurodevelopmental problems, and stressful interactions with their caregivers.[1,2] Dysfunctional swallowing (oropharyngeal dysphagia) is well recognized in infants and children with pre-maturity, upper aerodigestive tract anomalies, central nervous system impairments, neurodevelopmental delays, and syndromes affecting the craniofacial structures.[3–7] These diagnostic conditions may affect the structural integrity of the oropharynx or the complex coordination of the neuromuscular and airway processes involved in deglutition. Several studies suggest that oropharyngeal dysfunction occurs in children who are without known risk factors[8,9] and also demonstrate that early identification of the problem and prompt initiation of appropriate treatment may reduce the morbidities associated with swallowing dysfunction.[9,10]

This study was undertaken because the presence and nature of swallowing dysfunction in children with unexplained respiratory symptoms at the time of presentation continues to be under appreciated and we have found more variability in the clinical presentation than was previously reported. This is the first investigation to demonstrate this variability in respiratory presentations and detail the specific radiologic abnormalities associated with the swallowing dysfunction in this population. Furthermore, although the prognosis for recovery is quite

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Received 27 November 2005; Revised 5 May 2006; Accepted 6 May 2006.

DOI 10.1002/ppul.20488
Published online in Wiley InterScience (www.interscience.wiley.com).

good, the resolution of dysphagia can occur at older ages than previously reported.[7,8]

METHODS

From December 1991 through April 1995, approximately 517 patients were referred to Speech-Language Pathology for swallowing evaluations. We completed a retrospective chart review and identified 19 children with unexplained respiratory problems and dysphagic concerns, and who were subsequently found to have radiologic evidence of dysphagia (Table 1). Diagnostic exclusion criteria for risk factors associated with dysphagia included pre-maturity (<37 weeks gestational age), known neurologic deficits or upper aerodigestive tract anomalies, or other conditions commonly associated with impairments in deglutition at the time of presentation. Each subject participated in a clinical feeding/swallowing evaluation which followed a standard protocol including: a review of the medical, developmental, and feeding history; an examination of oral-motor structures and their functions; and an observation of a typical meal. A Videofluoroscopic Swallow Study (VFSS) was performed because of symptoms and clinical presentations suggestive of oropharyngeal dysphagia (Table 2).[11,12] Data collected included the caregiver's description of the onset and characteristics of the dysphagic symptoms, identification of primary referral sources, and results of other swallow-related diagnostic procedures.

The VFSS

One investigator (ML-G) and a pediatric radiologist performed each VFSS following modifications of swallow study protocols described elsewhere.[13] Videofluoroscopic data were obtained with a General Electric fluoroscope (RFX 90) and recorded on either a 3/4 or 1/2-inch videotape using a Sony VHS (VO-5800H) or Sony Super VHS (SVO-9600) videocassette recorder. Lateral views were imaged for all children.

Children were seated to simulate feeding positions used at home and observed during the clinical portion of this evaluation. Seating positions varied from a semi-reclined angle (approximately 60 degrees) for infants to an upright-seated position (90 degrees) for children who were able to sit independently. Age, customary feeding routines, and dysphagic concerns (e.g., coughing when drinking milk) guided the preparation of barium contrasts and selection of feeding utensils. One investigator (ML-G) prepared all barium consistencies using a 45% w/w (70% w/v) E-Z-PAQUE® barium sulfate suspension for thin liquid (TL) contrast. The TL base was used to prepare very thin (two parts TL: three parts water) and thick (one tablespoon rice cereal per 60 cc TL) liquid contrasts. The puree consistency was a 2:1 mixture of Stage II baby food (typically applesauce) and Esophotrast®, and solid foods (graham cracker or ''favorite'' soft cookie) were coated with Esophotrast®. The TL contrast was usually presented first; however, the order of presentation was adjusted to simulate each child's typical mealtimes and encourage cooperation. All children were presented with TL contrast. In addition, 13 children were presented with thick liquid, 5 with very TL, and 5 with solid foods. The very TL and solid foods were presented because of specific concerns about these textures. Liquids were presented by bottle, cup, or straw. Purees and solid foods were presented as per the child's usual feeding routine. Each child was fed by a familiar caregiver.

TABLE 1— Characteristics of Children at Presentation and Caregivers at Follow-Up

	Value
Feeding/swallowing evaluation	
Children: male, female	19: 13, 6
Age (years)	
Average (median)	1.14 (0.73)
Range	0.09–5.75
Follow-up interviews	
Caregivers contacted	14
Relationship to patient	
Mother	11 (79%)
Father	2 (14%)
Grandmother	1 (7%)
Children: male, female	14: 11, 3
Age (years)	
Average (median)	9.82 (9.39)
Range	7.72–14.45
Interval between feeding/swallowing evaluation and follow-up interview (years)	
Average (median)	8.7 (8.67)
Range	7.63–10.44

VFSS Analysis

Radiologic abnormalities were categorized according to the phase of swallow impairment (e.g., oral, pharyngeal, or cervical esophageal) and the presence of a delay in pharyngeal swallow onset. The oral phase was reviewed for problems with anterior or posterior bolus containment, and bolus formation or transfer.

We defined a delay in swallow initiation as contrast remaining in the valleculae for more than 2 sec or accumulating in the pyriform sinuses before the onset of the pharyngeal swallow. On-line timers were not used during videofluoroscopy. Delays in swallow initiation were estimated.

In this investigation, the primary pharyngeal phase abnormalities of interest were contrast entering the airway and the elicitation of a cough after contrast entered the trachea. We differentiated between two levels of airway entry—supraglottic penetration and aspiration. Supraglottic penetration (henceforth penetration) refers to the passage of contrast into the supraglottic airway

TABLE 2— Clinical Presentations of 19 Children Referred for Feeding/Swallowing Evaluations and With Oropharyngeal Dysphagia on Videofluoroscopic Swallow Study (VFSS)

Clinical presentations[1]	Descriptions of specific problems	Children no. (%)
Respiratory problems	Coughing or choking during feeds	14 (73.7)
	Wheezing	10 (52.6)
	Bronchitis or respiratory syncytial virus[2]	8 (42.1)
	Asthma or reactive airway disease	8 (42.1)
	Wet vocal quality	7 (36.8)
	Chronic upper respiratory infections	6 (31.6)
	Noisy breathing	5 (26.3)
	Apparent life threatening episode	3 (15.8)
	Stridor	1 (5.3)
	Cyanosis	1 (5.3)
	Oxygen requirement	1 (5.3)
Other feeding/swallowing-related problems	Gastroesophageal reflux, regurgitation, or emesis	10 (52.6)
	Poor weight gain	6 (31.6)
	Failure to thrive	2 (10.5)
	Nasopharyngeal reflux (NPR)	
	With emesis	8 (42.1)
	With oral feeding[3]	2 (10.5)
	Drooling	8 (42.1)
	Otitis media	
	Chronic or recurrent	9 (47.4)
	With pressure equalization tubes	4 (21.1)
	Gagging during meals	4 (21.1)
	Oral hypersensitivity	3 (15.8)
	Long mealtimes (average >40 min)	2 (10.5)
	Feeding refusal	1 (5.3)

[1]Clinical presentations were not mutually exclusive.
[2]Three months before the feeding/swallowing evaluation, one patient with aspiration on VFSS was diagnosed with RSV.
[3]One patient with aspiration on VFSS exhibited NPR with emesis and during feeding.

(i.e., into the laryngeal vestibule, including to the level of the vocal folds) which was then expelled from the airway. The term aspiration describes entry of contrast into the subglottic airway or trachea. Other pharyngeal phase abnormalities included nasopharyngeal reflux (NPR) and post-swallow residue.

The presence of cough in response to aspiration was noted. A cough response was reported as absent when it did not occur within 20 sec of the aspiration event.[3] The term silent aspiration refers to the failure to demonstrate an overt response to aspiration (e.g., coughing or choking).

Caregiver Telephone Follow-Up Interviews

In August 2002, one investigator (ML-G) conducted semi-structured follow-up telephone interviews with caregivers of 14 (74%) children. Interviews included questions about when caregivers were no longer concerned about feeding or swallowing problems; current feeding routines, health, and education status; and caregivers' recollections of what information was or would have been helpful when their children were diagnosed with oropharyngeal dysphagia.

Caregivers of five children could not be located. All caregivers who were contacted agreed to participate in telephone interviews. Approval was obtained from The Joint Committee for Clinical Investigation of The Johns Hopkins Hospital.

Statistical Analysis

A *t*-test was used to compare mean ages of children with penetration and aspiration. The presence of penetration or aspiration between children with and without GTs was compared using a Pearson Chi-Square (χ^2) test. All statistical computations were performed using the Statistical Package for the Social Sciences (SPSS).[14]

RESULTS

Patient Demographics

Nineteen children (mean age 1.14 years) who were referred to SLP for feeding/swallowing evaluations because of questions about swallowing dysfunction presenting as unexplained respiratory problems, and demonstrated radiologic evidence of oropharyngeal dysphagia (Table 1). Of

these children, 12 also presented with other feeding/swallowing-related difficulties (Table 2). Referrals for feeding/swallowing evaluations were made by pulmonologists (68%), gastroenterologists (21%), primary pediatricians (21%), otolaryngologists (21%), and immunologists (5%); occasionally two or more specialty services simultaneously requested evaluations.

Sixteen (84%) children began exhibiting dysphagic problems within the first 3 months of life. Of the remaining three children, one became symptomatic between 3 and 6 months, one between 6 and 12 months, and one when older than 36 months. At the time of their clinical feeding/swallowing evaluations, 17 were feeding orally. Two children presented with respiratory and dysphagic problems during efforts to resume oral feedings after surgical intervention (i.e., nissen fundoplication and gastrostomy tube [GT]) for gastroesophageal reflux (GER) which had been refractory to maximum medical management. Although these two children had GER, they were without risk factors associated with oropharyngeal dysphagia (i.e., diagnostic exclusion criteria) and swallowing difficulties coinciding with the resumption of oral feedings were not expected.

VFSS Findings

Oral phase abnormalities occurred infrequently and were considered mild because they did not interfere with adequacy of nutritional delivery. The number of children with inefficient anterior or posterior bolus containment for TLs, thick liquids, and puree was three, one, and one, respectively. Alterations in bolus formation or transfer occurred in two children with TLs and four with purees.

The most common abnormal radiologic finding was delayed pharyngeal swallow onset. The proportion of children with delayed swallow onset was 100% with very TLs, 94.7% with TLs, 100% with thick liquids, and 61.5% with purees.

The frequency of contrast entering the airway differed across the five consistencies of barium. The number of children with one or more episodes of either penetration or aspiration while swallowing very TLs, TLs, thick liquids, or purees was 4 (80.0%), 16 (84.2%), 10 (76.9%), and 2 (15.4%), respectively. Ten (52.6%) children displayed airway entry with more than one consistency. Airway entry did not occur with solid food boluses.

Aspiration occurred only with liquids, with 57.9% (11 of 19) children aspirating with either thin or thick liquid. One patient aspirated thin and thick liquids. The frequency of silent aspiration was 100%; that is, none of the 11 children with radiologic evidence of aspiration coughed or demonstrated any overt behaviors indicative of contrast entering the airway. Mean ages were comparable for children with penetration (1.46 years) and aspiration (0.90 years) ($t = 0.914$, $P > 0.05$).

NPR occurred in three children with TLs and two with thick liquids. In the plane imaged, all episodes of NPR appeared to be secondary to a delay in swallow onset versus inadequate velopharyngeal closure. One patient had traced post-swallow residue in the valleculae when swallowing thin and thick liquids, and puree. Cervical esophageal function was grossly within normal limits for all children.

Other Evaluations

Chest radiographs (including scout films immediately before the VFSS procedure) and upper gastrointestinal series (UGI's) were completed in 100% and 63% of the children, respectively (Table 3). The mean interval between VFSS and chest X-ray was 2.8 days (median, range 0, 0–19 days) and VFSS and UGI was 107.8 days (median, range 28.5, 1–1,006 days). Other evaluations included bronchoscopies, indirect and direct laryngoscopies, endoscopies, and pH probes. One patient underwent a VFSS at an outside facility.

Management Recommendations

Following discussions among the primary pediatrician, specialists, and primary caregivers, three general categories of management recommendations included changes in the route of nutritional support, modifications in oral feeding routines, and referrals or follow-up with pediatric specialty services. Modifications in oral feeding routines were recommended for the 17 children without feeding tubes. Of the two children with fundoplications and GTs, one was transitioned to a combination of oral and tube feedings and the other remained on gastrostomy feedings only.

Recommendations for 95% of the children included dietary modifications such as thickening liquids. Other oral feeding routine modifications included changes in feeding position (upright versus reclined), the pacing of meals (slowing the rate of feeding), and the overall feeding schedule (smaller and more frequent meals).

Additional evaluation or therapy needs prompted recommendations for referrals or follow-up with pediatric specialty services and multi-disciplinary feeding/swallowing programs. Five children continued to exhibit respiratory problems following the implementation of modifications in oral feeding routines and the initiation of conservative medical management (Table 4). Subsequently, two of these children underwent placement of a percutaneous GT (PEG) and three required GTs plus nissen fundoplications to manage oropharyngeal dysphagia and GER. Placement of GTs was unrelated to findings of penetration versus aspiration on VFSS ($\chi^2 = 0.707$, $P > 0.05$).

TABLE 3— Results of Upper Gastrointestinal Series, Chest X-rays, and Other Relevant Studies

Case	Upper gastrointestinal series (UGI)[1]	Chest X-ray[1]	Other studies
1		Minimal increase in central lung markings	Direct laryngoscopy: Normal; bronchoscopy: no report Endoscopy: esophageal erythema
2		Minimal hypoinflation	Bronchoscopy: no tracheoesophageal fistula, no malacia Upper endoscopy: normal
3	Limited examination: minimal gastroesophageal reflux	Minimal increased interstitial markings + diffuse peribronchial thickening; no focal infiltrates or atelectasis; minimal hyperinflation lungs	
4	Normal	Normal	Indirect laryngoscopy: normal anatomy with excessive pooling of secretions in hypophayrnx (out side hospital [OSH])
5		Diffuse central and peripheral peribronchial infiltrates; atelectatic changes in left lower lobe	Direct laryngoscopy: slightly deep interarytenoid space, no cleft; bronchoscopy: diffuse tracheobronchitis numerous lipid laden macrophages (LLM) pH probe: normal
6		No focal infiltrates or consolidations, minimally diffuse increased density both lung fields	
7	GER + aspiration	Normal	Upper endoscopy: normal
8	Question cricopharyngeal function; nasopharyngeal reflux, laryngeal penetration; significant GER	Normal	MRI of brain: normal; VFSS (OSH): normal
9	Minimal GER	Normal: clear lungs	Bronchoscopy: completed, no report available Endoscopy: positive for duodenitis and esophagitis
10		Mild central peribronchial thickening; no hyperinflation	
11	GER, normal peristalsis, no aspiration	Normal	
12		Mildly pronounced central and interstitial lung markings	Upper endoscopy: normal
13		Hyperinflation of lungs, no infiltrates	pH probe: positive
14	Aspiration, GER, no other esophageal abnormalities	Normal	
15	GER, no aspiration	Normal	Indirect laryngoscopy (OSH): redness
16	Aspiration, no GER	Normal	Recommended MRI of brain—no record of completion
17	Normal anatomy, GER	Increased central lung marking	Bronchoscopy: LLM upper endoscopy: normal
18	Normal	Normal	Direct laryngoscopy: mild laryngomalacia
19	Normal peristaltic activity, GER, spontaneousgastric emptying	Bilateral hypoinflation, no infiltrates or pleural effusion	Upper endoscopy: Normal

[1]Results are reported from the study closest to the date of the VFSS for children with more than one UGI and chest X-ray.

TABLE 4— Children With Aspiration or Penetration on Videofluoroscopic Swallow Study (VFSS) and Feeding Tube Status

Patient	Aspiration or penetration	Placement of tube and nissen fundoplication[1,2,3]	Reasons for tube placement[4]	Age of tube removal (years)[5]
1	A	A (+)	Asthma, GER	3
2	P	A (+)	Chronic cough, wheezing, gastroesophageal reflux, dysphagia	6
3	P	—		
4	A	—		
5	A	B (+)	Aspiration, GER	10
6	A	—		
7	A	—		
8	P	—		
9	P	—		
10	P	—		
11	P	—		
12	P	—		
13	A	—		
14	A	—		
15	A	—		
16	A	A	Chronic aspiration secondary to dysfunctional swallowing, failure to thrive	6
17	P	A (+)	Aspiration pneumonia, GER	8
18	A	A	Aspiration pneumonia and GER	6
19	A	B (+)	Asthma, GER	4

[1]A = After VFSS for failure to adequately respond to conservative management (e.g., feeding routine modifications and pharmacologic therapies).
[2]+ = Fundoplication.
[3]B = Before VFSS for respiratory problems presumed to be associated with GER.
[4]Reasons for feeding tube placement were obtained from operative reports of feeding tube placement.
[5]Ages for removal of feeding tubes were obtained from medical records of patients (Cases 16–19) whose caregivers could not be located for participation in follow-up interviews.

Follow-Up Caregiver Telephone Interviews

Caregivers of 11 children were no longer concerned about swallowing problems when children were a mean age of 3.2 years (range 0.7–10 years). Although none of the children had any risk factors associated with dysphagia during their initial evaluations, four children (Cases 1, 2, 3, and 5) had dysphagic problems until they were 9 years of age or older and three of these children continued to have problems at the time of the telephone interview (Table 5). Only one of these children (Case 1) aspirated during the VFSS.

In response to questions about their recollections of the swallowing evaluation process and what information would have been helpful, four (29%) caregivers reported being frustrated by difficulty convincing pediatricians about the possibility of swallowing difficulties. Three respondents (21%) wanted more realistic predictions of when the swallowing dysfunction would resolve and its potential impact on other family members. Two (14%) wanted to speak with other families facing similar problems and reassurances that their children would not die.

DISCUSSION

We identified the presence of oropharyngeal dysphagia in 19 children who presented with unexplained respiratory problems and were without known risk factors associated with swallowing dysfunction. Our observations have several clinical and research implications. First, dysphagia occurs in children without known risk factors for swallowing dysfunction. Consequently, there should be a high index of suspicion regarding dysphagia with concomitant aspiration in children with refractory respiratory problems. Second, when dysphagia results in aspiration, aspiration is likely to occur with liquids and is frequently silent. Third, even if children are thought to be low risk for dysphagia, dysphagia may persist for several years, not months as previously reported.[7,8,15]

Varying degrees of swallowing dysfunction have been reported in the children not thought to be at risk for such problems.[4,7,8,15] Descriptions of dysphagia in this population have ranged from profound problems characterized by ''very weak or absent pharyngeal contraction'' or ''incomplete pharyngeal palsy,''[7,16] to moderate pharyngeal dyscoordination,[7,8] to suck/swallow/breathing incoordination.[7] In this investigation, the most common radiologic abnormality was delayed swallow onset which preceded all penetration or aspiration events. We identified dysphagia in a cohort of children with swallowing dysfunction more characteristic of swallow/breathing incoordination than the moderate or profound abnormalities previously described.[7,15,16] Furthermore, respiratory presentations may be more variable than previously

TABLE 5— Caregiver Reports During Follow-Up Interviews[1]

Child	Age dysphagia resolved (years)	Current status
1	Problems continue	Nine years ♂ gags when eats too much. Asthma and frequent colds during winter. Respiratory medications. Missed 3–4 school days[1]. Fourth grade, average student.
2	Problems continue	Eleven years ♀ intermittent choking with liquids, usually able to compensate. Picky eater. Borderline slow weight gain. Frequent upper respiratory infections. Medication: lactulose. Missed 4 school days. Sixth grade, below average in math and comprehension.
3	Problems continue	Fourteen years ♂ continued swallowing problems. Aspirates when he drinks liquids. Picky eater. Asthma, recurrent pneumonia, no weight gain. Respiratory medications. Missed 19 school days. Eighth grade, special education, below average in all subject areas.
4	1	Nine years ♂ allergies. No medications. Missed 1 school day. Fourth grade, average student.
5	10	Twelve years ♂ enjoys eating. Frequent colds. Overweight. No medications. Seventh grade, above average student. Feeding tube removed at 10 years of age.
6	1	Eight years ♀ enjoys eating. Asthma. Overweight. Respiratory medications. Missed 3 school days. Fourth grade, above average reading and spelling, below average math.
7	3	Nine years ♀ picky eater without other problems for years. Medication: ranitidine for recent diagnose of gastroesophageal reflux. Home schooled, therefore no missed school days. Fourth grade, average math and above average reading.
8	4.5	Nine years ♂ picky eater w/o problems. Asthma. Poor weight gain. No medications. No missed school days. Fourth grade, above average.
9	2.5	Nine years ♂ enjoys eating. Asthma and colds last longer than other children. Respiratory medications. Misses 3–4 school days past year. Third grade, slightly above average student.
10	1	Eight years ♂ enjoys eating. No medications. Missed 8–9 school days past year, more than usual and attributed to strep throat. Third grade, above average student.
11	1.5	Nine years ♂ enjoys eating. No medications. Missed 1 school day. Fifth grade, average student.
12	4.5	Ten years ♂ enjoys eating. Slow weight gain. Medications for attention deficit hyperactivity disorder. No missed school days. Fifth grade, tutored for math and and reading comprehension, and speech therapy for articulation.
13	5	Eight years ♂ enjoys eating. No medications. Mildly overweight. Missed 8 school days. Third grade, average student with mild cerebral palsy needing physical therapy.
14	0.7	Seven years ♂ enjoys eating. No medications. Missed 3 school days. Second grade, above average student.

[1]School days missed during the academic year 2001–2002.

appreciated. Whereas isolated dysphagia has been identified in children who presented with cyanosis, wheezing, or stridor,[4,7,8] some of our children demonstrated chronic but more ambiguous respiratory symptoms (e.g., ''increased congestion'').

In this investigation, penetration occurred during liquid and puree swallows; however, aspiration occurred only with liquids. The relationship between penetration and aspiration continues to be controversial and a source of confusion. Whereas some investigators and clinicians believe that penetration signals an increased risk of aspiration,[5,17] others judge it to be less of a problem and not confirmatory of aspiration.[8,18] We concur that penetration may occur without aspiration; however, aspiration cannot occur without penetration because materials must pass through the laryngeal aditus before entering the trachea. Furthermore, in infants, aspiration typically occurs later than penetration during VFSS evaluations.[5] In this investigation, age did not distinguish between children with aspiration versus penetration secondary to oropharyngeal dysphagia. The likelihood that penetration will evolve into aspiration is probably determined by multiple factors including the presence of other swallowing abnormalities; the frequency, depth, and clearance of penetration; and characteristics of the particular child, such as tiring towards the end of meals.[5,17–19]

In our study, 100% of aspirators were silent aspirators. These findings are consistent with previous studies, which have reported an 89–100% rate of silent aspiration in young children with aspiration on VFSS.[5,8,20] Silent aspiration occurred even among children who were reported to cough while drinking, suggesting that coughing during meals does not guarantee that children swallow safely, particularly in the setting of other dysphagic problems or unexplained respiratory symptoms. Although reasons for the high prevalence of silent aspiration in dysphagic children remain unclear, potential explanations include the development of a blunted cough in response to recurrent aspiration, disruptions in the post-natal maturation of airway clearance mechanisms, or other processes not yet defined.[21,22] Further research is needed to define the relationship between aspiration and the evolution of cough and other protective airway mechanisms in young children with dysphagia.

Two of our subjects had nissen fundoplications and GTs placed before participating in swallowing evaluations. These children were without known dysphagic risk factors

and feeding tubes were placed because of the complex interactions between asthma and GER with presumed indirect aspiration. When symptoms persisted after surgical intervention, these children were evaluated by VFSS and exhibited oropharyngeal dysphagia with silent aspiration. Although our sample size is small, we recommend a careful review of every child's feeding history before removing feeding tubes even when they were placed during procedures primarily for GER and indirect aspiration, and swallowing evaluations for those with histories suggestive of oropharyngeal dysphagia.

While the long-term prognosis for recovery is very good for children without obvious dysphagic risk factors, dysphagic concerns may persist longer than previously reported. Isolated dysphagia has been reported to resolve within the first 3–4 years of life;[4,7,8,23] however, four of our children (Cases 1, 2, 3, and 5) exhibited swallowing problems beyond 9 years of age. Unfortunately, one of the limitations of this study design is that we do not have airway or neurologic data for all children. The reason for persistent swallowing dysfunction remains unclear for one child (Case1) given the available data. For the remaining three children, dysphagia may have been one of the first indications of other underlying conditions. The potential of a non-specific underlying etiology, particularly in the setting of learning problems and developmental delays, is a possible explanation for persistent problems in two children (Cases 2 and 3). Anatomic abnormalities may account for the third child (Case 5) exhibiting dysphagic problems until he was 10 years of age. After the VFSS, he underwent a direct laryngoscopy, which revealed a ''somewhat deeper'' than normal intra-arytenoid space; however, a laryngeal cleft was not identified. Another plausible explanation for persistent dysphagia is that caregivers of children previously diagnosed with dysphagia are more sensitive to the presence of any feeding or swallowing difficulty. Finally, the reason for dysphagic concerns on follow-up may be attributed to one of the limitations of this investigation. Given the lengthy interval between the initial evaluation and the telephone survey, we were unable to determine whether the reported problems represented new dysphagic concerns versus unresolved difficulties. Regardless of the reasons for dysphagic problems at these older ages, our cases illustrate that swallowing dysfunction in this population may persist longer than previously recognized.

During telephone interviews, nearly one-third of the caregivers recalled being frustrated by their pediatrician's failure to initiate diagnostic procedures for swallowing dysfunction promptly. They reported that pediatricians did not respond to their concerns about feeding difficulties because their children were ''normal'' and hence ''would quickly outgrow'' these problems. Furthermore, they were not prepared for the ongoing and stressful impact of these problems on other family members, particularly siblings.

The findings of this investigation are subjected to all the limitations associated with retrospective studies and interviews based upon recollections. Well-planned prospective studies are needed to determine the prevalence of unexplained pediatric dysphagia and identify clinical pathways which support the best outcomes for these children. One of the unavoidable weaknesses of this study design was the variability in evaluation procedures completed after identification of oropharyngeal dysphagia. Although none of our children exhibited dysphagic risk factors at the time of presentation, systematic and consistent evaluations may have revealed other underlying conditions contributing to the dysphagia or respiratory problems in some of our subjects. Unfortunately, data from your follow-up telephone survey were limited to responses from only 76% of the caregivers. Despite these limitations, our findings are consistent with other studies, which have confirmed the presence of oropharyngeal dysphagia in children without identifiable etiologies at the time of presentation.[4,7,8] Increased awareness of the potential of oropharyngeal dysphagia in this population will enable clinicians to initiate prompt referrals to specialists for appropriate diagnostic procedures. A thorough feeding history should be obtained when children present with unexplained respiratory problems. Children with histories suggestive of oropharyngeal dysphagia should be referred for a swallowing assessment, including a VFSS when appropriate.

Oropharyngeal dysphagia should be considered in the differential diagnosis of young children presenting with unexplained respiratory problems even if they are without apparent dysphagic risk factors. Early evaluation of swallowing dysfunction and prompt initiation of appropriate therapies are critical to lessening the impact of the morbidities associated with dysphagia. Although the long-term prognosis for recovery is very good, dysphagic concerns may persist for several years, which is longer than previously reported.

REFERENCES

1. Tawfik R, Dickson A, Clarke M, Thomas AG. Caregivers' perceptions following gastrostomy in severely disabled children with feeding problems. Dev Med Child Neurol 1997;39:746–751.
2. Loughlin GM. Respiratory consequences of dysfunctional swallowing and aspiration. Dysphagia 1989;3:126–130.
3. Rogers B, Arvedson J, Buck G, Smart M. Characteristics of dysphagia in children with cerebral palsy. Dysphagia 1994;9:69–73.
4. Kohda E, Hisazumi H, Hiramatsu K. Swallowing dysfunction and aspiration in neonates and infants. Acta Otolaryngol Suppl (Stockh) 1994;517:11–16.
5. Newman LA, Keckley C, Petersen MC, Hamner A. Swallowing function and medical diagnoses in infants suspected of Dysphagia. Pediatrics 2001;108:E106.
6. Griggs CA, Jones PM, Lee RE. Videofluoroscopic investigation of feeding disorders of children with multiple handicap. Dev Med Child Neurol 1989;31:303–308.

7. Heuschkel RB, Fletcher K, Hill A, Buonomo C, Bousvaros A, Nurko S. Isolated neonatal swallowing dysfunction: A case series and review of the literature. Dig Dis Sci 2003;48:30–35.
8. Sheikh S, Allen E, Shell R, Hruschak J, Iram D, Castile R, McCoy K. Chronic aspiration without gastroesophageal reflux as a cause of chronic respiratory symptoms in neurologically normal infants. Chest 2001;120:1190–1195.
9. Khoshoo V, Edell D. Previously healthy infants may have increased risk of aspiration during respiratory syncytial viral bronchiolitis. Pediatrics 1999;104:1389–1390.
10. Khoshoo V, Ross G, Kelly B, Edell D, Brown S. Benefits of thickened feeds in previously healthy infants with respiratory syncytial viral bronchiolitis. Pediatr Pulmonol 2001;31:301–302.
11. Arvedson JC, Rogers BT. Pediatric swallowing and feeding disorders. J Med Speech Lang Pathol 1993;1:203–221.
12. Lefton-Greif MA. Diagnosis and treatment of pediatric feeding and swallowing disorders: Role of the speech-language pathologist. In: Tuchman DN, editor. Pediatric feeding and swallowing disorders: Pathophysiology, diagnosis, and treatment. San Diego: Singular Publishing Group; 1994. p 97–113.
13. Logemann JA. Manual for the Videofluorographic Study of Swallowing. 2nd ed. Austin, TX: Pro-Ed; 1993.
14. SPSS (Statistical Package for the Social Sciences). 13th ed. Chicago: SPSS Inc; 2004.
15. Inder TE, Volpe JJ. Recovery of congenital isolated pharyngeal dysfunction: Implications for early management. Pediatr Neurol 1998;19:222–224.
16. Mbonda E, Claus D, Bonnier C, Evrard P, Gadisseux JF, Lyon G. Prolonged dysphagia caused by congenital pharyngeal dysfunction. J Pediatr 1995;126:923–927.
17. Friedman B, Frazier JB. Deep laryngeal penetration as a predictor of aspiration. Dysphagia 2000;15:153–158.
18. Delzell PB, Kraus RA, Gaisie G, Lerner GE. Laryngeal penetration: A predictor of aspiration in infants? Pediatr Radiol 1999;29:762–765.
19. Robbins J, Coyle J, Rosenbek J, Roecker E, Wood J. Differentiation of normal and abnormal airway protection during swallowing using the penetration-aspiration scale. Dysphagia 1999;14:228–232.
20. Arvedson J, Rogers B, Buck G, Smart P, Msall M. Silent aspiration prominent in children with dysphagia. Int J Pediatr Otorhinolaryngol 1994;28:173–181.
21. Loughlin GM, Lefton-Greif MA. Dysfunctional swallowing and respiratory disease in children. Adv Pediatr 1994;41:135–162.
22. Thach BT. Maturation and transformation of reflexes that protect the laryngeal airway from liquid aspiration from fetal to adult life. Am J Med 2001;111:69S–77S.
23. Macaulay JC. Neuromuscular incoordination of swallowing in the newborn. Lancet 1951;1:1208.