

Original Investigation

Usefulness of Upper Airway Endoscopy in the Evaluation of Pediatric Pulmonary Aspiration

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IMPORTANCE There is no consensus on the evaluation of pediatric patients with aspiration.

OBJECTIVES To determine the yield of direct laryngoscopy and bronchoscopy (DLB) using general anesthesia in pediatric patients who aspirate and to identify clinical predictors of aspiration-related airway lesions.

DESIGN, SETTING, AND PARTICIPANTS Retrospective review at a tertiary referral children's hospital. A medical record review was performed on all patients with a documented diagnosis of pulmonary aspiration who underwent DLB using general anesthesia during a 5-year period (January 2010 to December 2014).

INTERVENTION Direct laryngoscopy and bronchoscopy using general anesthesia.

MAIN OUTCOMES AND MEASURES Data were collected and analyzed, including age, sex, history of intubation, flexible laryngoscopy results, DLB findings, recurrent pneumonia, and associated diagnoses.

RESULTS Five hundred thirty-two patients met the inclusion criteria. Their mean (SD) age was 2.2 (3.6) years (age range, 0.1-25.0 years), with more than half younger than 1 year. Sixty-two percent (328 of 532) of the participants were male. Flexible laryngoscopy examination alone identified 93 patients with an airway lesion. Direct laryngoscopy and bronchoscopy identified 173 additional diagnoses and had a greater diagnostic yield for airway lesions (45.1% [240 of 532]) than flexible laryngoscopy examination alone ($P < .001$). Patients with an aspiration-related airway lesion were older (mean [SD] age, 2.7 [3.8] vs 2.2 [3.8] years; $P = .02$) and more likely to have another aerodigestive disorder than were patients without an airway lesion (21.7% vs 11.6%; $P = .004$). Older age (adjusted risk ratio [95% CI], 1.37 [1.08-1.73]; $P = .01$), recurrent pneumonia (1.40 [1.11-1.76]; $P = .004$), and history of intubation (1.35 [1.07-1.70]; $P = .01$) were significantly associated with the presence of an aspiration-related airway lesion in the multivariable model. Patients with an aspiration-related airway lesion were less likely to have neurologic disease than were patients without an airway lesion (0.50 [0.34-0.73]; $P < .001$). In all, 66.3% of patients (110 of 166) eventually underwent surgical repair of an identified aspiration-related airway lesion.

CONCLUSIONS AND RELEVANCE In children with chronic aspiration who warrant further evaluation, flexible laryngoscopy alone is not sufficient. There is a high incidence of aspiration-related airway lesions identified on DLB and not seen on flexible laryngoscopy, with 66.3% (110 of 166) of those lesions eventually requiring surgical intervention. Patients 1 year or older with a history of recurrent pneumonia or intubation are more likely to have an aspiration-related airway lesion.

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The management of aspiration in children can be challenging for both physicians and caregivers. Patients often manifest nonspecific symptoms, including chronic cough and upper airway or chest congestion.¹ In more severe cases, patients can initially be seen with recurrent pneumonia. The diagnosis of pulmonary aspiration requires a high index of suspicion by physicians to select appropriate testing, with modified barium swallow study or fiberoptic endoscopic evaluation of swallowing being the primary diagnostic modalities.²⁻⁴ In certain cases, especially in patients who grossly aspirate, the diagnosis of aspiration is made clinically with limited oral testing.

Once the diagnosis of pediatric pulmonary aspiration is confirmed, there is no consensus regarding the evaluation of these patients. At Boston Children's Hospital, patients undergo a multidisciplinary evaluation by an otolaryngologist, pulmonologist, gastroenterologist, and speech pathologist. From an otolaryngology perspective, all patients with complicated or persistent aspiration are evaluated with flexible laryngoscopy and direct laryngoscopy and bronchoscopy (DLB). If indicated by the appropriate service, esophagogastroscope and pulmonary bronchoscopy are coordinated with the DLB using the same general anesthetic.

Because DLB requires general anesthesia, caregivers often question the usefulness of DLB over flexible laryngoscopy alone. The objective of this study was to review our airway findings in patients who undergo a complete upper airway evaluation to determine the added value of DLB in the evaluation of these patients. We also aimed to identify clinical predictors of aspiration-related airway lesions.

Methods

The Boston Children's Hospital Institutional Review Board approved this study, and its guidelines were followed. A retrospective medical record review was performed on all patients with a documented diagnosis of pulmonary aspiration who underwent DLB using general anesthesia at our institution during a 5-year period (January 2010 to December 2014). Patients with no follow-up data were excluded from this study. Data were collected and analyzed, including demographics, comorbidities, history of intubation, airway lesions, and management. Patients were subdivided into groups for analyses. Those with aspiration-related airway lesions, including laryngeal cleft, vocal fold immobility, glottic web, and H-type tracheoesophageal fistula, were compared with those with no airway lesion.

To compare the characteristics of patients with an aspiration-related airway lesion with those of patients with no airway lesion, we used χ^2 test for categorical variables and Wilcoxon rank sum test for continuous variables. Multivariable analysis was also performed to examine clinical predictors of an aspiration-related airway lesion. The adjusted risk ratios (95% CIs) were estimated from the multivariable binomial regression model with a log-link function. The multivariable model was built using a backward selection procedure, with $P < .05$ as the retention criterion. All analyses were con-

Key Points

Question: Should direct laryngoscopy and bronchoscopy (DLB) be performed in the evaluation of children with chronic aspiration?

Findings: In this retrospective study, DLB had a greater diagnostic yield for airway lesions compared with flexible laryngoscopy alone. Children with aspiration-related airway lesions were more likely to be older (≥ 1 year) and have another aerodigestive disorder and a history of intubation and recurrent pneumonia than those without an airway lesion.

Meaning: Direct laryngoscopy and bronchoscopy should be performed in the evaluation of children with chronic aspiration who require further investigation.

ducted with a statistical significance threshold of $P < .05$ and performed using a software program (SAS, version 9.3; SAS Institute Inc).

Results

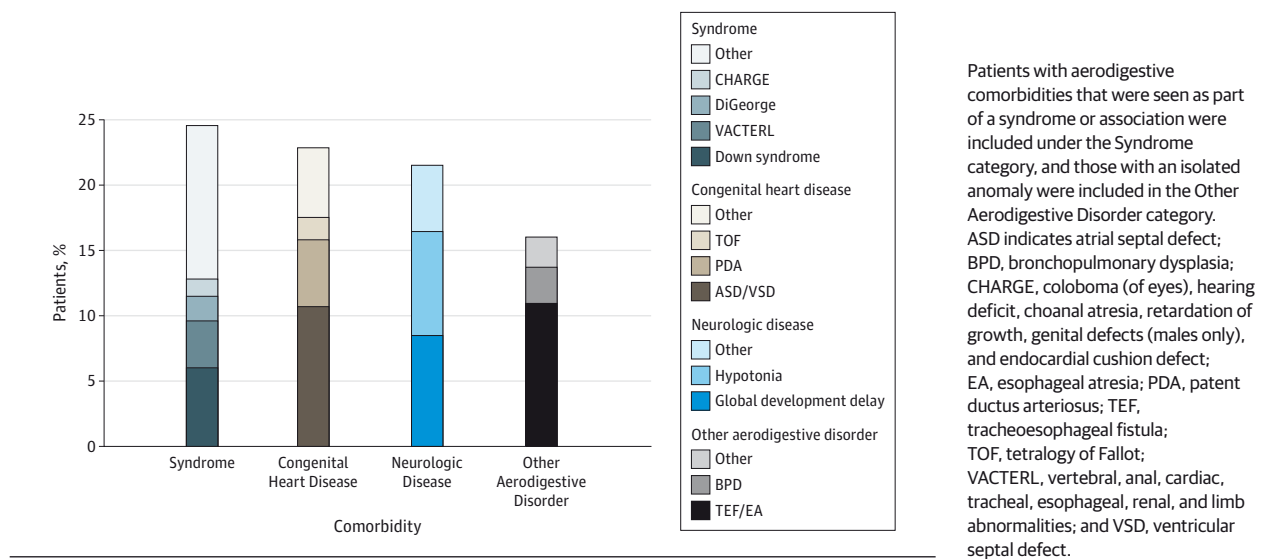
Baseline Characteristics

Five hundred thirty-two patients met the inclusion criteria. Sixty-two percent (328 of 532) of the participants were male. Their mean (SD) age was 2.2 (3.6) years (age range, 0.1-25.0 years), with 56.6% (301 of 532) younger than 1 year. Forty-five percent (240 of 532) had a history of intubation, and 38.7% (206 of 532) had recurrent pneumonia. Comorbidities in 85 patients are shown in the **Figure** (note that some patients had >1 comorbidity). Sixteen percent (85 of 532) had another aerodigestive comorbidity (ie, esophageal atresia, bronchopulmonary dysplasia, cleft palate, or choanal atresia). In this cohort of patients, 91.8% (78 of 85) were diagnosed as having aspiration based on modified barium swallow study alone.

DLB and Flexible Laryngoscopy Findings

Flexible laryngoscopy examination alone identified 93 patients with an airway lesion. Laryngomalacia ($n = 57$) was the most common lesion noted on flexible laryngoscopy, followed by vocal fold immobility ($n = 34$) and glottic web ($n = 2$). Direct laryngoscopy and bronchoscopy identified 173 additional diagnoses and had a greater diagnostic yield for airway lesions (240 of 532 [45.1%]) than flexible laryngoscopy examination alone ($P < .001$ by McNemar test). Almost half of the patients had an airway abnormality. Among 532 patients, 292 (54.9%) had normal findings. Direct laryngoscopy and bronchoscopy identified 136 patients (25.6%) with laryngeal cleft, 40 patients (7.5%) with laryngomalacia, 25 patients (4.7%) with vocal fold immobility, 16 patients (3.0%) with tracheomalacia, 12 patients (2.3%) with subglottic stenosis, 6 patients (1.1%) with laryngotracheomalacia, 3 patients (0.6%) with H-type tracheoesophageal fistula, and 2 patients (0.4%) with glottic web. Among 136 patients with laryngeal cleft, 115 cases (84.6%) were type 1 clefts, 18 cases (13.2%) were type 2 clefts, and 3 cases (2.2%) were type 3 clefts.

Figure. Comorbidities in the Study Population



Patients with aerodigestive comorbidities that were seen as part of a syndrome or association were included under the Syndrome category, and those with an isolated anomaly were included in the Other Aerodigestive Disorder category. ASD indicates atrial septal defect; BPD, bronchopulmonary dysplasia; CHARGE, coloboma (of eyes), hearing deficit, choanal atresia, retardation of growth, genital defects (males only), and endocardial cushion defect; EA, esophageal atresia; PDA, patent ductus arteriosus; TEF, tracheoesophageal fistula; TOF, tetralogy of Fallot; VACTERL, vertebral, anal, cardiac, tracheal, esophageal, renal, and limb abnormalities; and VSD, ventricular septal defect.

Table 1. Characteristics of Patients With an Aspiration-Related Airway Lesion vs Patients Without an Airway Lesion

Variable	No. (%)		P Value ^a
	Aspiration-Related Airway Lesion (n = 166)	No Airway Lesion (n = 292)	
Sex			
Female	60 (36.1)	114 (39.0)	.54
Male	106 (63.9)	178 (61.0)	
Age group, y			
<1	74 (44.6)	168 (57.5)	.02
1-2	43 (25.9)	67 (22.9)	
≥3	49 (29.5)	57 (19.5)	
Prematurity	50 (30.1)	81 (27.7)	.59
Congenital heart disease	43 (25.9)	61 (20.9)	.22
Syndrome	35 (21.1)	69 (23.6)	.53
Other aerodigestive disorder	36 (21.7)	34 (11.6)	.004
Neurologic disease	23 (13.9)	78 (26.7)	.001
History of intubation	86 (51.8)	120 (41.1)	.03
Recurrent pneumonia	76 (45.8)	101 (34.6)	.02
Reactive airway disease	36 (21.7)	75 (25.7)	.34

^a The P values are based on χ^2 test.

Aspiration-Related Airway Lesions

Patients with an incidental finding of subglottic stenosis, laryngomalacia, laryngotracheomalacia, or tracheomalacia were excluded from further analyses. Patients with an aspiration-related airway lesion were older than patients without an airway lesion (mean [SD], 2.7 [3.8] vs 2.2 [3.8] years; $P = .02$) (Table 1). The presence of another aerodigestive disorder was also more common in patients with an aspiration-related airway lesion than in those without (21.7% [36 of 166] vs 11.6% [34 of 292], $P = .004$). A history of intubation (51.8% [86 of 166] vs 41.1% [120 of 292]) and recurrent pneumonia (45.8% [76 of 166] vs 34.6% [101 of 292]) were also more common in patients with an aspiration-related airway lesion compared with patients with no airway lesion ($P = .03$ and $P = .02$, respec-

tively). Patients with an aspiration-related airway lesion were less likely to have neurologic disease than patients without an airway lesion (13.9% [23 of 166] vs 26.7% [78 of 292], $P = .001$).

In the multivariable model, older age (≥ 1 year), recurrent pneumonia, and a history of intubation remained significantly associated with the presence of an aspiration-related airway lesion (Table 2). Patients with an aspiration-related airway lesion were less likely to have neurologic disease than patients without an airway lesion. Among 166 patients, 110 (66.3%) with an aspiration-related airway lesion eventually underwent surgical repair of the identified airway lesion. Surgical procedures were performed in 110 patients with the following conditions: 96 (87.3%) with laryngeal cleft (76 type 1 clefts, 17 type 2 clefts, and 3 type 3 clefts), 11 (10.0%) with vocal

Table 2. Multivariable Analysis of Factors Associated With the Presence of an Aspiration-Related Airway Lesion

Variable	Adjusted Risk Ratio (95% CI) ^a	P Value
Age ≥1 y vs <1 y	1.37 (1.08-1.73)	.01
Recurrent pneumonia	1.40 (1.11-1.76)	.004
History of intubation	1.35 (1.07-1.70)	.01
Neurologic disease	0.50 (0.34-0.73)	<.001

^a The adjusted risk ratios (95% CIs) are based on the multivariable binomial regression model that includes all the variables listed.

fold immobility, 2 (1.8%) with glottic web, and 1 (0.9%) with H-type tracheoesophageal fistula.

Discussion

Pediatric aspiration can manifest with a spectrum of symptoms and signs, ranging from silent aspiration to chronic lung disease.⁵ In patients with suspected aspiration, an objective test (eg, modified barium swallow or fiberoptic endoscopic evaluation of swallowing) is performed to assist with diagnosis. Flexible laryngoscopy is also a valuable adjunct that can be performed at the bedside in most pediatric patients.

The major disadvantage to an evaluation with flexible laryngoscopy alone is that its usefulness is limited to the level of the glottis. Laryngeal cleft and H-type tracheoesophageal fistula are clinically significant aspiration-related airway lesions that cannot be detected with flexible laryngoscopy alone. Barium swallow study could be helpful in identifying H-type tracheoesophageal fistula but would not detect laryngeal cleft. In our study group, DLB identified 136 patients with laryngeal cleft and 3 patients with H-type tracheoesophageal fistula. Of these 139 patients, 97 (69.8%) eventually underwent surgical repair of their airway lesion.

In addition to evaluating the diagnostic yield of DLB, another aim of this study was to identify clinical predictors of aspiration-related airway lesions. As shown in the Figure, 24.4% (130 of 532) of patients were syndromic, and 22.7% (121 of 532) of patients had congenital heart disease. Neither of these factors was associated with the presence of an airway lesion. On the other hand, the presence of another aerodigestive disorder increased the risk of identifying an aspiration-related airway lesion. This finding is similar to prior work that identified an association between laryngeal cleft and tracheoesophageal fistula.⁶⁻⁸

Histories of recurrent pneumonia and intubation were also significantly associated with the presence of an aspiration-related airway lesion. Therefore, recurrent pneumonia is an important point on which to query caregivers about because it increases the likelihood of identifying an aspiration-related airway lesion, and serves as an indication for surgical repair of the identified lesion. A history of intubation is also important to elicit from the patient history because it can be associated with laryngeal abnormalities, such as vocal fold immobility and glottic web, and may also reveal a history of respiratory distress secondary to aspiration pneumonia.

Patients with neurologic disease were significantly less likely to have an aspiration-related airway lesion. In children

with global developmental delay, cerebral palsy, and hypotonia, it has been shown that dysphagia is most likely to occur during the oral and pharyngeal phases of swallowing, which explains the lack of significant airway findings in these patients.⁹⁻¹¹ In our study, 24 of 117 patients (20.5%) with a neurologic comorbidity had an aspiration-related airway lesion. Therefore, although the yield is lower in these patients, DLB may have a role in the evaluation of select patients.

It should be noted that, at our institution, the initial DLB performed for patients with aspiration is for diagnostic rather than therapeutic purposes. In the absence of an emergent airway finding, patients are awoken from anesthesia after diagnostic DLB, and the airway findings are discussed with the caregiver before any additional surgical intervention.

Not all patients with an aspiration-related airway lesion required further surgical intervention. The decision and technique to repair an airway lesion depend on the type of lesion, the patient's overall medical status, and surgeon preference. Sixty-six percent (110 of 166) of our patients eventually underwent surgical repair of their airway lesion. Seventy-one percent (96 of 136) of patients with laryngeal cleft eventually underwent endoscopic repair of their lesion. A review of the past 15 years of laryngeal cleft studies (not including studies from our institution) revealed that the rate of endoscopic repair of type 1 laryngeal clefts ranges from 10% to 80% in the literature.¹²⁻¹⁵ Most studies consider a thickened liquid diet to be conservative therapy and a nonoral diet as a treatment failure. However, in the study¹² reporting that 90% of patients were managed conservatively, 14 of 28 patients required nasogastric tube feeding (mean duration, 8 months) (n = 9), gastrostomy tube placement (n = 3), or gastrojejunostomy (n = 2). When that study is eliminated from the literature review, the rate of endoscopic repair rises to 64% to 80%,¹³⁻¹⁵ which is similar to our rate of endoscopic laryngeal cleft repair.

There are several notable limitations to this study. As with any retrospective study, we are limited to data that have already been collected for purposes other than this study. This investigation is also a single-center experience, and we have a large referral base for patients with suspected airway lesions. In addition, the institution has a large tracheoesophageal fistula population, which is also associated with laryngeal cleft. Therefore, the incidence of lesions may be overestimated in our study and not representative of the general population. Because an administrative database was used to identify patients, diagnoses may have been miscoded, inadvertently excluding patients from the study.

Conclusions

Direct laryngoscopy and bronchoscopy is a valuable adjunct in the evaluation of pediatric patients with aspiration. Its diagnostic yield is significantly higher than that of flexible laryngoscopy alone. Therefore, DLB should be considered in select pediatric patients with chronic aspiration, particularly in those 1 year or older, those with recurrent pneumonia, or those with a history of intubation.

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Study concept and design: Adil and Watters.

Acquisition, analysis, or interpretation of data: All authors.

Drafting of the manuscript: Adil, Gergin, Kawai, Watters.

Critical revision of the manuscript for important intellectual content: Adil, Rahbar, Watters.

Statistical analysis: Kawai.

Administrative, technical, or material support: Watters.

Study supervision: Watters.

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