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Swallowing dysfunction among patients with laryngeal cleft: More than just aspiration?

Julie E. Strychowsky [a,c,*], Pamela Dodrill [a], Ethan Moritz [a], Jennifer Perez [a], Reza Rahbar [a,b]

[a] Department of Otolaryngology and Communication Enhancement, Boston Children's Hospital, 333 Longwood Ave, 3rd Floor, Boston, MA 02115, USA
[b] Department of Otology and Laryngology, Harvard Medical School, Boston, MA, USA
[c] Department of Otolaryngology-Head and Neck Surgery, University of Western Ontario, 800 Commissioners Rd E, VH B3-444, London, ON, N6A 5W9, Canada

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ABSTRACT

Background: The Modified Barium Swallow (MBS) is the most widely utilized instrumental assessment of swallowing disorders in children; however, the exact role in the evaluation of laryngeal clefts remains controversial.
Methods: This study was an IRB-approved retrospective review on patients diagnosed with laryngeal cleft from 2002 to 2014. The objective was to describe the range of swallowing dysfunction that may be present in patients with laryngeal clefts both pre- and post-intervention (conservative management versus surgery). A speech-language pathologist reviewed MBS studies and medical records to determine Penetration–Aspiration Scale (PAS) and Functional Oral Intake Scale (FOIS) scores.
Results: One hundred seventy-five patients who underwent laryngeal cleft repair during the study period (type 1, $n = 111$; type 2, $n = 54$; type 3, $n = 9$; type 4, $n = 1$) were included. Fifty patients who were managed conservatively (type 1) were also included. Swallowing impairment was demonstrated in all phases of swallowing for all cleft types. Oral phase impairment ranged from 27–67% pre-intervention to 19–75% post-intervention, triggering impairment from 24–42% pre-intervention to 24–75% post-intervention, and pharyngeal phase impairment (laryngeal penetration and aspiration) from 57–100% pre-intervention to 40–100% post-intervention. Laryngeal penetration and aspiration on thin and thick liquids, silent aspiration, PAS, and FOIS scores are reported. Significant improvements in swallowing function ($p < 0.05$) were documented in all of the conservatively and surgically managed sub-groups.
Conclusions: The MBS study is a useful tool for evaluating swallowing function in patients with laryngeal cleft and provides information beyond the lack or presence of aspiration. Understanding impairments in all phases of swallowing may be beneficial for perioperative management.

1. Introduction

Laryngeal clefts are rare congenital anomalies that result from failure of fusion of the tracheoesophageal septum or two lateral growth centers of the posterior-cricoid cartilage during embryological development [1–3]. The annual incidence ranges from 1 in 10,000 to 1 in 20,000 live births, with a male predominance [4]. Most cases are sporadic while others associated with syndromes such as Opitz-Frias or Pallister-Hall, or congenital anomalies such as tracheoesophageal fistulas [5]. Patients with laryngeal clefts may present with airway and/or swallowing impairments, which can lead to recurrent aspiration pneumonia, respiratory distress, and failure to thrive [1–3]. Therefore, appropriate and timely diagnosis is paramount.

Management of laryngeal clefts ranges from conservative monitoring to surgical intervention. Both groups generally require feeding therapy input to address potential swallowing impairments. Feeding therapy techniques may involve skill building activities (e.g. oral-motor therapy) and/or teaching the patient or their care-givers how to use compensatory strategies, such as the use of modified liquids/foods (e.g. thickened liquids, pureed foods), modified feeding equipment (e.g. slower or faster flow bottle nipples, open cups vs straw cup), modified feeding positioning (e.g. upright vs reclined) and modified feeding strategies (e.g. having the feeder actively pace the feed, having the child take single sips from a straw vs sequential sips). It is the role of the Otolaryngologist, with input from the

* Corresponding author. Tel.: +1 519 685-8242, Fax: +1 519 685-8185.
E-mail addresses: julie.strychowsky@lhsc.on.ca (J.E. Strychowsky), pamela.dodrill@childrens.harvard.edu (P. Dodrill), emoritz@wesleyan.edu (E. Moritz), jennifer.perez@childrens.harvard.edu (J. Perez), reza.rahbar@childrens.harvard.edu (R. Rahbar).

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speech-language pathologist, to determine whether a patient would best be managed through feeding therapy alone or if they also require surgical repair of their cleft.

There is a lack of standardized evaluation for the diagnosis of laryngeal cleft [1–3,6–9]. Diagnostic practices include history and physical examination, chest X-ray, swallowing assessment (modified barium swallow [MBS] and/or fiberoptic endoscopic evaluation of swallowing [FEES]), flexible fiberoptic laryngoscopy, and the gold standard of operative endoscopy to gently palpate the interarytenoid area. Other diagnostic adjuncts include measurement of lipid laden macrophage levels obtained by bronchial alveolar lavage [10] and interarytenoid injection [11–13]. The MBS (also known as video fluoroscopic swallow study, VFSS) is the most widely utilized tool for the assessment of swallowing disorders in children [14,15]; however, the exact role in the diagnosis and monitoring of laryngeal clefts remains controversial.

The purpose of this study was to describe the range of swallowing dysfunction that may be present in patients with laryngeal clefts both pre- and post-intervention (conservative management versus surgery).

2. Materials and methods

A retrospective review of a prospective database on all patients diagnosed with laryngeal cleft at Boston Children's Hospital was performed. Institutional review board approval was obtained. All patients who were diagnosed with laryngeal cleft from 2002 to 2014 by the senior author (R.R.) and had MBS studies performed were included. A speech language pathologist (P.D.) reviewed all of the MBS studies to classify types of swallowing impairments and determined Penetration–Aspiration (PA) scale [16] scores and Functional Oral Intake Scale (FOIS) [17] scores for each MBS study. The original reports from the MBS studies were used for data collection and when this data was insufficient, the radiologic examination was reevaluated.

MBS swallowing impairments were classified as follows: oral phase impairment, swallow triggering impairment, and pharyngeal phase impairment (including laryngeal penetration on thin liquids, laryngeal penetration on thickened liquids, aspiration on thin liquids, aspiration on thickened liquids, and silent aspiration). Laryngeal penetration was defined as food or liquid penetrating the laryngeal inlet above the level of the vocal folds, whereas aspiration was defined as food/liquid passing below the vocal folds. For those who underwent surgery, the MBS study that preceded surgical repair was analyzed. In addition, where available, post-operative MBS studies at or around 4 months post-surgery were analyzed for comparison (a four-month timeframe was used to allow for adequate surgical healing and assumed swallowing retraining). For those who did not undergo surgical management, the first MBS available was used as a baseline measure. A subsequent study at or around 4 months post-initial assessment was used for a similar comparison.

PA and FOIS scores were determined based on the findings and recommendations at the time of each MBS study. Rosenbek and colleagues developed the PA scale in 1996 [16]. It is an 8-point validated multidimensional assessment tool for swallowing impairment that relies on the classification of the depth to which material passes into the airway and whether or not it is expelled (Table 1). The FOIS pediatric scale was adapted by Crary and colleagues [17] from an existing adult tool [18]. It is a 7-point ordinal scale that documents the functional intake of food and liquid in patients (Table 2).

Data from patient medical records was extracted by one member of the research team (E.M.) and included: patient demographics, type of cleft, medical comorbidities, symptomatology, feeding history, and number of clinic visits and MBS studies.

Table 1
Laryngeal Penetration–Aspiration (PA) Scale [16].

1	Material does not enter airway
2	Material enters the airway, remains above the vocal folds, and is ejected from the airway
3	Material enters the airway, remains above the vocal folds, and is not ejected from the airway
4	Material enters the airway, contacts the vocal folds, and is ejected from the airway
5	Material enters the airway, contacts the vocal folds, and is not ejected from the airway
6	Material enters the airway, passes below the vocal folds, and is ejected into the larynx or out of the airway
7	Material enters the airway, passes below the vocal folds, and is not ejected from the trachea despite effort
8	Material enters the airway, passes below the vocal folds, and no effort is made to eject

The diagnosis of laryngeal cleft was confirmed by palpation of the interarytenoid area during direct laryngoscopy by the senior author (R.R.). The type of laryngeal cleft was defined by the Benjamin–Inglis classification system, which describes type 1 as an interarytenoid defect to the level of the vocal folds, type 2 as partial extension through the posterior cricoid cartilage, type 3 as extension completely through the posterior cricoid cartilage and possible extension into the cervical trachea, and type 4 as extension into the intrathoracic trachea [19].

Statistical analysis was performed (P.D. and K.K.) using the SPSS Statistics for Windows (Version 19, SPSS Inc., Chicago, IL). Chi-square analysis was used to analyze trends in dichotomous measures (e.g. presence/absence of oral phase impairment, pharyngeal phase impairment, laryngeal penetration, aspiration). Kruskal Wallis and Mann Whitney U analyses were used to compare non-parametric measures (e.g. PA scale and FOIS scores). *t*-Tests and ANOVAs were used to compare parametric measures (e.g. number of MBS studies, ORL visits, pneumonias). A p-value < 0.05 was considered statistically significant.

3. Results

One hundred seventy-five patients underwent laryngeal cleft repair during the study period (type 1, $n = 111$; type 2, $n = 54$; type 3, $n = 9$; type 4, $n = 1$), while 50 patients with laryngeal cleft were managed conservatively with feeding therapy and compensatory strategies (type 1 only). Pre-operative MBS studies were available for 138 surgical patients (79%) (type 1, $n = 98$; type 2, $n = 37$; type 3, $n = 3$; type 4, $n = 0$) and all conservatively managed patients. Patient demographics and comorbidities are detailed in Table 3.

MBS swallowing impairments varied by cleft sub-type and for pre- versus post-surgery (Table 4). All laryngeal cleft sub-types demonstrated some degree of oral phase impairment, triggering impairment, and pharyngeal phase impairment (including laryngeal penetration, aspiration, and silent aspiration). Interestingly, within these groups, some patients did not demonstrate any

Table 2
Functional oral intake scale (FOIS) for infant/toddler [17] Table 14-8.

1	Nothing by mouth
2	Tube dependent, with minimal attempts at liquids/food
3	Tube dependent, with consistent intake of liquids/food
4	Total oral diet, but requiring modified liquids ± compensations*
4.5	Total oral diet, but requiring modified solids ± compensations*
5	Total oral diet, without special preparation (i.e. developmentally appropriate), but with compensations*
6	Total oral diet (developmentally appropriate), with no restrictions

* Compensations = special feeding equipment (e.g. special nipples/cups), special therapy strategies (e.g. pacing), or special positioning (e.g. side-lying for infants, head support for older children).