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Type 1 laryngeal cleft: Establishing a functional diagnostic and management algorithm

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Modified barium swallow

Summary

Objectives: To report our experience with all patients diagnosed with type 1 laryngeal cleft over a period of 3 years in our referral practice and to describe a functional diagnostic and management algorithm for children with this disorder.
Methods: A prospective longitudinal study in a tertiary care referral center. Twenty pediatric patients diagnosed with type 1 laryngeal cleft in a 3-year period (5/1/2002–5/1/2005) were included in this study. The incidence, presenting symptoms, diagnostic procedures, medical and surgical interventions performed, and clinical outcomes were evaluated.
Results: The incidence of type 1 laryngeal cleft was 7.6%. Among the 20 patients in this study, aspiration with thin liquids was the most common presenting symptom (18 patients, 90%). Three patients underwent modified barium swallow (MBS) alone, 3 patients underwent functional endoscopic evaluation of swallow (FEES) alone, and 11 patients underwent both MBS and FEES prior to intraoperative endoscopic evaluation. Four patients (20%) were successfully treated with conservative therapy. Sixteen patients (80%) required endoscopic surgical repair after failing a course of conservative measures. The success rate of surgical repair was 94% (15 out of 16 patients).
Conclusions: Type 1 laryngeal cleft can be challenging diagnostically. We propose a functional diagnostic and management algorithm that includes MBS, FEES, suspension laryngoscopy with bimanual interarytenoid palpation, and a trial of conservative

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therapy, as a way to diagnose and manage type 1 laryngeal cleft prior to consideration of surgical repair. If conservative therapy fails, then surgical intervention is indicated.

1. Introduction

Laryngeal cleft is an uncommon congenital malformation where an anomalous connection between the laryngo-trachea and the esophagus is found. The incidence of laryngeal cleft is reported to be less than 0.1% [1] in the general population. It was first described by Richter in 1792 [2], and Pettersson performed the first successful repair in 1955 [3].

Multiple classification systems have been devised to characterize laryngeal cleft [4–6]. The most commonly used classification system today was described by Benjamin and Inglis [6]. In this classification system, laryngeal clefs are divided into four categories (Fig. 1). Type 1 laryngeal clefts are supraglottic interarytenoid clefts, where the clefts are above the level of true vocal cords. Type 2 laryngeal clefts extend below the level of the vocal cords but do not involve the posterior cricoid lamina completely. Type 3 laryngeal clefts extend completely through the cricoid cartilage, with or without further extension into the cervical tracheoesophageal wall. Type 4 laryngeal clefts extend through the majority of the tracheoesophageal wall.

The diagnosis of type 1 laryngeal cleft can be challenging: the presenting symptoms are usually non-specific (e.g. chronic cough and aspiration with feeds), and the laryngeal defect may be difficult to visualize endoscopically. The management of type 1 laryngeal cleft is also controversial, with some studies advocating surgical repair [7] while others advocating medical management as the first-line therapy [8].

The goal of the present study is to attempt to improve the diagnosis and treatment of type 1 laryngeal cleft by identifying features that are commonly seen in these patients. An outline of our multi-dimensional and functional diagnostic and management algorithm, a review of the treatment decision process, and a discussion on endoscopic repair with regards to technique as well as to perioperative course are included.

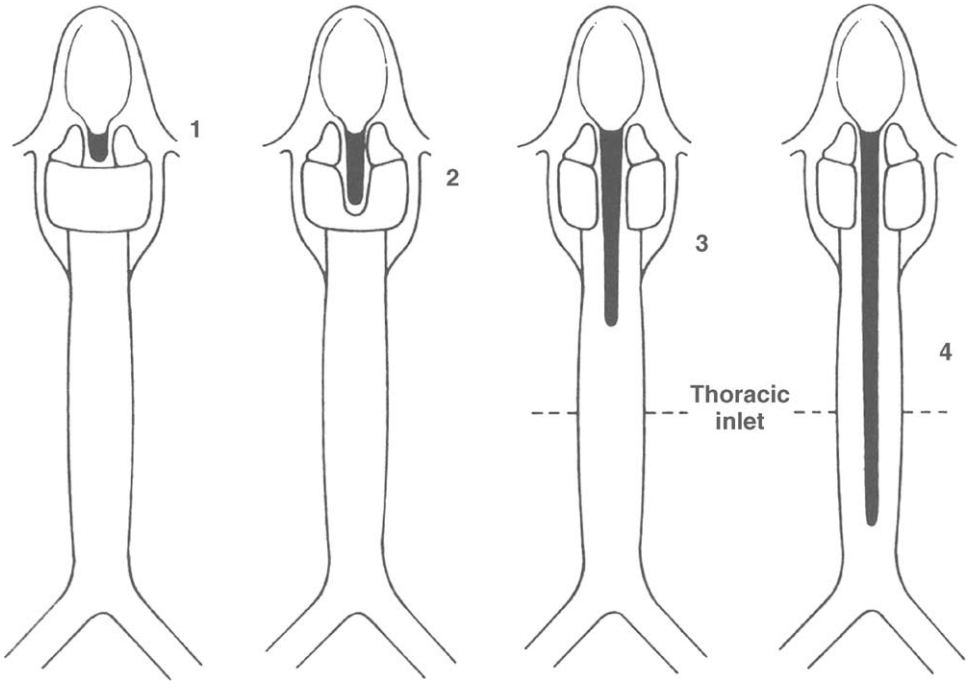

Fig. 1 The Benjamin–Inglis classification system of laryngeal clefts. Type 1 clefts are supraglottic interarytenoid clefts, where the cleft is above the level of true vocal cords. Type 2 clefts extend below the level of the vocal cords but do not involve the posterior cricoid lamina completely. Type 3 clefts extend completely through the cricoid cartilage, with or without further extension into the cervical tracheoesophageal wall. Type 4 clefts extend through the majority of the tracheoesophageal wall ([6], permission granted).

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2. Methods

A prospective longitudinal study of all patients who were diagnosed with type 1 laryngeal cleft by the senior author (C.J.H.) from 05/01/2002 to 05/01/2005 was performed. Demographic information (age and sex) of the patients was collected. The clinical work-up, diagnostic studies performed, treatment modalities implemented, and clinical outcomes were followed and examined.

3. Results

During a 3-year time period (05/01/2002–05/01/2005), a total of 264 pediatric patients were seen in our institution with the presenting symptom of either chronic cough or aspiration (ICD# 786.2 and 933.1). Out of the 264 cases, 20 patients were diagnosed with type 1 laryngeal cleft (7.6%) by a combination of modified barium swallow (MBS), functional endoscopic evaluation of swallow (FEES), and intraoperative endoscopic examination. The age of diagnosis ranged from 4 to 63 months, with the mean of 24.7 months (standard deviation 18.6 months). There were 12 female patients (60%) and 8 male patients (40%). Aspiration with thin liquids was the most common presenting symptom, being seen in 18 patients (90%) (Table 1). Recurrent aspiration pneumonia was seen in 10 patients (50%), chronic cough was seen in 7 patients (35%), stridor was seen in 3 patients (15%), and hypoxia with cyanotic attack was seen in 2 patients (10%). Several patients (75%) had associated abnormalities (Table 2). There were four patients with a history of prematurity at birth (20%), one patient with concomitant tracheoesophageal fistula (5%), four patients with laryngomalacia (20%), one patient with tracheomalacia (5%), one patient with subglottic hemangioma (5%), one patient with duodenal web (5%), and one patient with Nemaline myopathy (5%). Gastroesophageal reflux, as defined by symptom complex, pH probe, or prior endoscopy, was found in 14 patients (70%).

Table 1 Presenting symptoms of patients with type 1 laryngeal cleft

Symptoms	# of patients	% of patients
Aspiration with thin liquids	18	90
Recurrent pneumonia	10	50
Chronic cough	7	35
Stridor	3	15
Hypoxia	2	10

The number of patients and the percentage are shown.

Table 2 Concomitant diagnosis in patients with type 1 laryngeal cleft

Diagnosis	# of patients	% of patients
GERD	14	70
Prematurity	4	20
Laryngomalacia	4	20
TEF	1	5
Subglottic hemangioma	1	5
Duodenal web	1	5
Nemaline myopathy	1	5

The number of patients and the percentage are both shown.

Most patients received MBS and/or FEES to assess the presence, severity, and mechanism of aspiration. Three patients received only MBS (15%), 3 patients received only FEES (15%), and 11 patients received both MBS and FEES (55%) prior to intraoperative endoscopic evaluation (Table 3). There were three patients who did not receive either MBS or FEES due to parental refusal. All 20 patients were taken to the operating room for suspension laryngoscopy with bimanual palpation of the interarytenoid region under magnification using two suction probes to distract the interarytenoid tissue to assess for the presence of laryngeal cleft, as well as rigid bronchoscopy.

After the diagnosis of type 1 laryngeal cleft was made, all patients were treated initially with conservative measures for 1 month (including antireflux therapy, thickened feeds, and maneuvers during feeding to prevent aspiration). Four patients (20%) had successful clinical outcomes with conservative therapy alone, judged by improved cough and resolution of aspiration. Sixteen patients (80%) failed conservative therapy and eventually required surgical repair. Of the 16 patients who underwent surgical repair, 15 patients had successful outcomes as defined by parental report of lack of aspiration with thin liquids and further pulmonary infections after the first 3 months postoperatively. The success rate of surgical repair was 94%. One patient (6%) continued to have episodes of vomiting and recurrent aspiration pneumonia after 3 months postoperatively (see Section 4 for further details).

Table 3 Numbers of type 1 laryngeal cleft patients who received MBS, FEES, or both

Diagnostic study	# of patients	% of patients
MBS only	3	15
FEES only	3	15
MBS and FEES	11	55
None	3	15

The number of patients and the percentage are both shown.

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4. Discussion

Type 1 laryngeal cleft is an uncommon congenital malformation. The 20 cases of type 1 laryngeal clefts described here represent an overall incidence of 7.6% among patients who presented to our pediatric airway center with chronic cough or aspiration. This is comparable to the 6.2% and 7.1% figures reported in two recent large-center case series of type 1 laryngeal cleft [7,8]. The fact that our institution has a specialized airway center may help to explain the high incidence rate reported in this study. Our airway center is staffed by pediatric otolaryngologists, general pediatricians, pediatric pulmonologists, pediatric speech-language pathologists, and pediatric gastroenterologists. By working closely with pediatricians, who are often the first to see patients with chronic cough and aspiration, the awareness of laryngeal cleft is raised.

The presentation of type 1 laryngeal cleft can be non-specific. The most common presenting symptoms in our series were aspiration with feeds (90%), recurrent aspiration pneumonia (50%), and chronic cough (35%). This is consistent with other large-center studies of laryngeal cleft, where symptoms of aspiration [9], recurrent aspiration pneumonia [7], and wheezing/asthma-like complaints (including cough) [8] were found to be most common. The differential diagnosis of patients presenting with chronic cough and aspiration includes neuromuscular disorders effecting deglutition, central nervous system lesions (such as Arnold-Chiari malformation), hydrocephalus, vocal cord paralysis, patulous cricopharyngeal sphincter, esophageal strictures, gastroesophageal reflux, tracheoesophageal fistula (especially H-type), and adenotonsillar hypertrophy.

It has been reported that as many as 50% of patients with laryngeal cleft have associated congenital abnormalities [10]. These congenital anomalies include tracheo-esophageal fistula (TEF), tracheomalacia, cleft lip and palate, anomalous right subclavian artery, pyloric atresia, imperforate anus, pancreatic ectopia, congenital heart defect, and congenital subglottic stenosis. In the present study, seven patients (35%) have associated congenital anomalies, including TEF, laryngomalacia, tracheomalacia, subglottic hemangioma, subglottic stenosis, duodenal web, and Nemaline myopathy (a hereditary neuromuscular disorder characterized by extreme muscle weakness since birth). Only one patient in our study had TEF (5%), which is comparable with the figures reported by Parsons and Watters (5% and 8.3%, respectively). It is interesting to note that none of our patients had either Pallister-Hall syndrome (congenital hypothalamic hamartoblastomas, hypopituitarism, imperforate anus, postaxial polydactyly, and cleft larynx) or Opitz-Frias syndrome (hypertelorism, hypospadia, cleft lip and palate, and cleft larynx), which are commonly associated with laryngeal cleft. We suggest genetic counseling only in cases where there is a family history of genetic abnormalities, or if the patient has other syndromic features.

Most of our patients either underwent MBS and/or FEES prior to intraoperative endoscopic examination. MBS and FEES are both valuable tools for assessing aspiration. MBS has the advantage of being a non-invasive test, but it offers an inferior resolution of the detailed laryngeal anatomy. Functional endoscopic evaluation of swallow, on the other hand, offers direct visualization of the larynx during swallowing, which allows physicians to assess the mobility of the vocal cords, the amount of pooling and penetration of both secretions and liquid/food substances, and the location of aspiration [11,12]. The limitation of FEES stems from the age of the patient at which FEES can yield accurate results. The senior author (C.J.H.) and the senior speech pathologist (J.A.) feel that the optimal ages for attempting FEES in children are 3–12 months and greater than 4 years of age. Children under 1 year of age can undergo FEES with the exception of newborns, where the suck/swallow sequence is too rapid for evaluation; the view of the laryngeal and pharyngeal swallowing mechanisms may also be limited in newborns due to their small anatomic size. In our experience, toddlers can become extremely distressed with the examination and this interferes with the accuracy of swallow assessment; the crying itself can result in episodes of aspiration while trying to swallow simultaneously. Children older than 4 years of age, in general, are better able to participate in the examination, and even attempt swallow treatment maneuvers to improve swallow coordination and safety in cases of aspiration. In the 11 patients who received both MBS and FEES, 7 patients had abnormal findings in both studies (64%), and 4 patients had abnormal findings in FEES with a normal MBS (36%). It is important to note that the results of MBS and FEES can be normal in patients who aspirate intermittently, and thus a normal result in either test does not definitively rule out laryngeal cleft. However, it is our view that MBS and FEES offer complimentary diagnostic information, and should both be performed in the work-up of laryngeal cleft.

Intraoperative endoscopic examination has been held as the gold standard diagnostic modality for type 1 laryngeal cleft [13], and the endoscopic finding which confirms the diagnosis is the absence of the interarytenoid muscle [7]. However, it has been shown histologically that the interarytenoid

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muscle is actually present in patients with type 1 laryngeal cleft, but that the muscle is atrophic [14,15]. This suggests that endoscopic diagnosis of type 1 laryngeal cleft can be subjective, since the extent of interarytenoid muscle atrophy can be variable. This may also help to explain why some patients with type 1 laryngeal cleft respond to conservative therapy alone (these patients probably have a limited amount of interarytenoid muscle atrophy), versus those who require surgical intervention (these patients probably have more extensive interarytenoid muscle atrophy).

We believe that all patients with a clinical history consistent with laryngeal cleft should undergo a diagnostic and management algorithm (Fig. 2) that includes a careful history and physical examination,

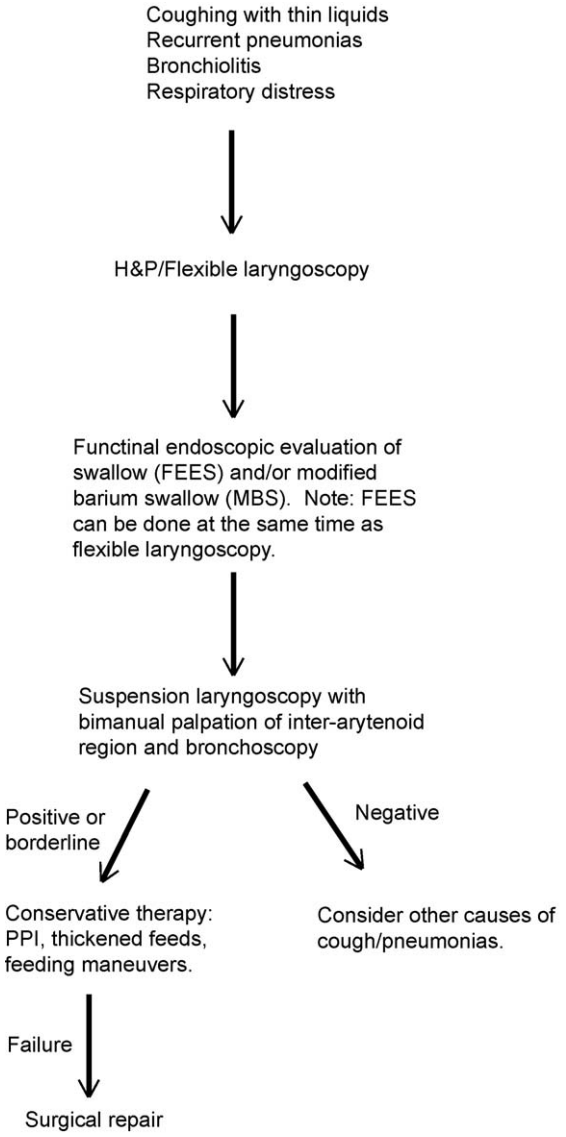

Fig. 2 A proposed algorithm for evaluating patients with type 1 laryngeal clefts.

MBS and/or FEES to evaluate aspiration, and suspension laryngoscopy with bimanual interarytenoid palpation (and rigid bronchoscopy) to evaluate the laryngeal defect. All patients should receive a trial of conservative therapy (including proton pump inhibitor (PPI) for gastroesophageal reflux, maintaining an upright position during feedings, thickened feeds, and maneuvers during feeding to prevent aspiration) prior to consideration of surgical intervention. The rationales for having patients undergo conservative therapy initially are: (1) it helps to differentiate between patients who will benefit from surgical intervention versus those who will not; and (2) in patients who eventually need surgical repair, conservative therapy can optimize their preoperative status by controlling for reflux. In this study, 80% of our patients did not have significant improvement with conservative measures alone, and required surgical intervention. This rate is comparable with the rate of 75% reported by Watters and Russell [7].

Prior to considering surgical repair, it is important to carefully evaluate the results of MBS and FEES to determine whether the mechanism of aspiration warrants the laryngeal cleft repair. For example, one of our patients who were diagnosed with a type 1 laryngeal cleft endoscopically was found to a pattern of aspiration that did not go through the laryngeal cleft defect, but it occurred in a lateral-to-medial direction across the aryepiglottic folds. In this patient, surgical repair of the laryngeal cleft would not have improved the aspiration. This example demonstrates the importance of MBS and FEES in the diagnostic work-up and management planning of type 1 laryngeal cleft.

Multiple surgical techniques have been described for treating laryngeal cleft. The least invasive method is Gelfoam injection into the laryngeal cleft to mimic an intact posterior glottic wall. In a recent study, Kennedy et al. used Gelfoam injection both as a diagnostic and therapeutic method for treating type 1 laryngeal cleft and reported favorable results [16]. However, since Gelfoam is absorbable, the long-term outcomes of these patients remain to be seen. There are multiple surgical approaches to repair type 1 laryngeal cleft. The anterior approach exposes the larynx through a laryngofissure [17], which offers excellent visualization of the laryngeal defect, but may cause laryngeal instability as well as vocal dysphonia if the anterior commissure is damaged or misapproximated. The lateral pharyngotomy approach is advocated by some [5], but there is an increased risk of injuring the recurrent laryngeal nerve. In all of our patients that underwent surgical repair, the endoscopic approach was used. The endoscopic approach is excellent for

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type 1 laryngeal cleft repair [18]. In our institution, we advocate the necessity for spontaneous ventilation, "tubeless" surgery without an endotracheal tube for optimal exposure. This requires excellent cooperation with an experienced anesthetist. If an endotracheal tube were in place, then surgical exposure and suturing would be severely limited. Once anesthesia is induced, the patient is suspended with either an infant or pediatric Lindholm laryngoscope. The interarytenoid space is palpated bimanually with suction probes and injected with 1% lidocaine with 1:100,000 epinephrine. A V-shaped incision is made around the cleft, and dissection is carried out to create an anterior and a posterior mucosal flap bilaterally. These flaps are then reapproximated with 5-0 vicryl sutures on S-12 needles. Of particular importance is the need for appropriate instrumentation to facilitate suturing and tying knots. We use the Kleinsasser needle holder (Karl Storz Endoscopy-America Inc., Culver City, CA) to manipulate the sutures, and the Jako knot pusher (Pilling Surgical Instruments, Limerick, PA) to place knots. After the surgery is completed, an age-appropriate endotracheal tube is then inserted to be used as a laryngeal stent while wound healing takes place. Patients are transported to the pediatric or neonatal intensive care unit for postoperative care. In general, patients are kept intubated for 1–10 days, and are extubated once wound healing takes place. There was one patient who did not require endotracheal tube placement at the end of the surgical repair, and had an uneventful postoperative course.

The rationale for keeping patients intubated postoperatively is to provide a secure airway until wound healing takes place. The duration of intubation, however, has been controversial. There has not been any convincing study which specifically addresses the issue of postoperative intubation duration after laryngeal cleft repair. Some advocate extubation immediately after surgical repair [18], whereas others feel that postoperative intubation for a small amount of time may be necessary [19]. This issue deserves further investigation.

All of the patients that underwent surgical repair in this study did not experience any complications associated with the surgical procedure itself. Some of the possible surgical complications include recurrent laryngeal nerve injury, wound dehiscence, granulation tissue formation, supraglottic stenosis, and deepening of the initial cleft. The main complications experienced by our patients were associated with the postoperative sedation and muscle relaxant administration and appeared to increase in frequency and severity in relation to the length of time the patient was intubated. Three patients (19%) intubated for 1 week experienced serious short-term sequelae: one patient experienced prolonged global hypotonia and required occupational therapy; another patient developed global hypotonia that caused persistent aspiration which finally resolved fully by 3 months postoperatively; a third patient failed to wake up for 48 h after sedation and muscle relaxants were withdrawn, but fortunately this patient spontaneously awoke with no residual deficits and has done well. In our series, the senior author (C.J.H.) began by keeping patients intubated and sedated for 1 week postoperatively but has since adapted a practice of intubating the patients for one night postoperatively without any change in outcome.

One patient in this study continued to have recurrent aspiration pneumonia 3 months postoperatively. However, this patient initially had a history of tracheo-esophageal fistula (TEF), and developed tracheal stenosis after surgical repair of TEF. As a result, she had a long history of recurrent pneumonia prior to the diagnosis of type 1 laryngeal cleft was made (up to 32 episodes in 1 year). After the surgical repair of type 1 laryngeal cleft, even though the patient continued to have occasional aspiration pneumonia, the frequency was greatly reduced (2–3 episodes a year).

5. Conclusions

Type 1 laryngeal cleft is an uncommon congenital malformation of the larynx that requires a high index of clinical suspicion to make the correct diagnosis. We believe that all patients suspected to have a type 1 laryngeal cleft should undergo the functional diagnostic and management algorithm outlined above, which includes a careful medical history and physical examination, MBS, FEES, suspension laryngoscopy with bimanual interarytenoid palpation (and rigid bronchoscopy) as well as a trial of conservative therapy. We advocate that patients should be treated conservatively at first, and only undergo surgical repair if conservative measures fail to yield improvement and if the aspiration pattern is through the laryngeal cleft defect. We prefer the endoscopic approach for type 1 laryngeal cleft repair, which has low morbidity and complication rates. The open technique is reserved for revision surgery or more extensive clefts. Postoperatively, patients are kept intubated for a minimal amount of time before extubation. The duration of postoperative intubation remains controversial and requires further investigation.

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