ORIGINAL ARTICLE

The Presentation and Management of Laryngeal Cleft

A 10-Year Experience

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Objective: To review the presentation and associated congenital abnormalities of laryngeal cleft and present guidelines for its evaluation and management.

Design: A 10-year retrospective study (1994-2004) with institutional review board approval.

Setting: Two pediatric tertiary care medical centers.

Patients: Twenty-two pediatric patients (mean age, 21 months) with laryngeal cleft.

Intervention: Surgical repair of laryngeal cleft.

Main Outcome Measures: Sex, age, symptoms, other associated abnormalities, method of evaluation, type of laryngeal cleft, method of surgical repair, treatment outcome, complications, and long-term follow-up.

Results: All 22 patients underwent surgical repair for laryngeal cleft. Airway endoscopy confirmed the following types of laryngeal clefts: type 1 (n=3), type 2 (n=10), and type 3 (n=9). Surgical repair techniques included an open approach with or without interposition graft (n=16) and an endoscopic approach (n=6).

Conclusions: Early diagnosis and proper repair of laryngeal cleft are essential to prevent pulmonary damage and associated morbidity. Each patient should be assessed properly, and the surgical approach should be individualized based on the symptoms, other associated findings on airway endoscopy, and type of cleft.

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LARYNGEAL CLEFT IS A RARE condition. Symptoms range from mild stridor to massive aspiration and respiratory distress, depending on severity of the cleft. Diagnosis requires a high index of suspicion, accurate interpretation of preoperative studies, and thorough endoscopic evaluation. Different surgical approaches have been proposed for the management of laryngeal cleft. The purpose of this study is to present our experience with patients with laryngeal cleft and provide guidelines for its evaluation and management.

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METHODS

Patients treated for laryngeal cleft between 1994 and 2004 at Children's Hospital, Boston, Mass (n=7), and Armand-Trousseau Children's Hospital, Paris, France (n=15) were identified (**Table 1** and **Table 2**). A systematic medical chart review was undertaken to determine the age at the time of presentation, sex, symptoms, significant medical and family history, evaluation findings, initial treatment, need for subsequent treatments, efficacy of each treatment, and complications. We classified the type of laryngeal cleft in our patients based on the Benjamin and Inglis[1] classification system (**Figures 1**, **2**, and **3**A).

RESULTS

Twenty-two patients were diagnosed as having laryngeal cleft (Table 1 and Table 2). Age at the time of diagnosis ranged from 15 days to 12 years (mean age, 21 months). Initial presentations were for airway difficulty (n=6), feeding difficulty (n=2), or a combination of both (n=14). Fifteen patients (68%) presented with another significant medical problem. Fourteen patients (64%) required other surgical interventions.

Table 1. Preoperative Patient Characteristics

Patient No./ Sex/Age	Symptoms at Presentation	Medical History	Significant Surgical History	Swallow Studies Showing Aspiration, No.	Type of Cleft*	Other Significant Findings on Endoscopy
1/F/12 y	Aspiration; recurrent pneumonia	Vater syndrome	Repair TEF	2	1	Left vocal cord paralysis
2/M/6 y	Aspiration; stridor	Asperger syndrome	None	1	1	None
3/F/3 y	Aspiration; recurrent pneumonia	None	Gastrostomy tube placement	2	1	None
4/M/2 y	Aspiration; recurrent pneumonia	IgA deficiency	Gastrostomy tube placement	10	2	None
5/F/3 y	Aspiration; feeding difficulty	Mild bronchopulmonary dysplasia; GER	Nissen fundoplication; gastrostomy tube placement	6	2	None
6/M/1 mo	Respiratory distress	None	Gastrostomy tube placement	3	3	None
7/M/5 y	Stridor	Opitz-Frias syndrome	Tracheotomy	5	2	None
8/M/1 mo	Cyanotic attacks; feeding difficulty	Prematurity; GER; esophageal atresia	Esophagoplasty	3	3 (3 rings)	Tracheobronchomalacia
9/F/14 mo	Stridor; recurrent pneumonia; aspiration	Pallister-Hall syndrome	None	No study obtained	3 (4 rings)	Tracheobronchomalacia
10/F/13 mo	Stridor; recurrent pneumonia; aspiration	None	None	No study obtained	3 (2 rings)	Tracheobronchomalacia
11/M/12 mo	Feeding difficulty	TEF GER	Repair of TEF, tracheotomy; gastrostomy tube placement; Nissen fundoplication	1	2	Tracheobronchomalacia
12/F/10 mo	Respiratory distress	GER	Nissen duplication; gastrostomy tube placement	1	3 (4 rings)	Tracheobronchomalacia
13/M/4 mo	Malnutrition; feeding difficulty; stridor	Esophageal atresia	Repair of esophageal atresia (esophagoplasty)	1	3 (4 rings)	None
14/M/1 mo	Recurrent pneumonia	Opitz-Frias syndrome; GER	Tracheotomy; gastrostomy tube placement Nissen duplication	1	3 (4 rings)	None
15/F/3 mo	Stridor	None	None	1	2	None
16/M/15 d	Cyanotic attacks; feeding difficulty	Cleft lip; GER	Tracheotomy; gastrostomy tube placement; Nissen duplication	No study obtained	3 (6 rings)	Tracheobronchomalacia
17/F/7 mo	Feeding difficulty; recurrent pneumonia	Esophageal atresia	Esophagoplasty gastrostomy tube placement	1	2	Left vocal cord paralysis
18/F/1 mo	Feeding difficulty; stridor; respiratory distress	None	None	No study obtained	2	Tracheobronchomalacia
19/M/7 mo	Respiratory distress	None	Tracheotomy	No study obtained	3 (2 rings)	Tracheobronchomalacia
20/M/2 mo	Feeding difficulty; recurrent pneumonia	None	None	1	2	None
21/F/8 mo	Stridor; recurrent pneumonia; aspiration	Rubella embryofetopathy (deafness, cataract, microphthalmia)	None	No study obtained	2	Laryngomalacia Glossoptosis
22/F/1 mo	Feeding difficulty	Cleft palate	None	1	2	None

Abbreviations: GER, gastroesophageal reflux; TEF, tracheoesophageal fistula.
*Based on the Benjamin and Inglis[1] classification system.

Methods of evaluation included rigid airway endoscopy (n=22), esophagoscopy (n=16), and preoperative modified swallow study (n=16). Airway endoscopy confirmed laryngeal cleft in all patients: type 1 (n=3), type 2 (n=10), and type 3 (n=9). Other significant findings noted at the time of endoscopy included tracheobronchomalacia (n=8), unilateral vocal cord paralysis (n=2), laryngomalacia (n=1), and glossoptosis (n=1).

All patients were managed medically for gastroesophageal reflux (GER) during the perioperative and postoperative periods. Five patients underwent a combination of Nissen fundoplication and gastrostomy tube placement, and 4 patients required gastrostomy tube placement alone. The mean age at the time of laryngeal cleft repair was 21 months. All patients with type 1 laryngeal cleft underwent an initial trial of con-

Table 2. Perioperative and Postoperative Patient Characteristics

Patient/ Sex/Age	Age at the Time of Surgery	Repair Approach	Postoperative Duration of Intubation	Postoperative Swallow Study Findings	Complications	Duration of Follow-up
1/F/12 y	12 y	Endoscopic	1 d	Normal	None	1 y
2/M/6 y	6 y	Endoscopic	1 d	Normal	None	2 y
3/F/3 y	3 y	Endoscopic	1 d	Normal	None	10 mo
4/M/2 y	2 y	Endoscopic	1 d	Normal	None	15 mo
5/F/3 y	3 y	Endoscopic	3 d	First MSS showed aspiration; second study showed no aspiration	Pneumonia	12 mo
6/M/1 mo	4 mo	Endoscopic	4 d	Normal	None	17 mo
7/M/5 y	5 y	Open, with interposition tibial periosteum graft	Preexisting tracheotomy tube in place	Normal	None	5 y
8/M/1 mo	2 m	Open, with interposition tibial periosteum graft	8 d	First MSS showed aspiration; second study showed no aspiration	None	7 y
9/F/14 mo	14 mo	Open, with interposition tibial periosteum graft	6 d	None	None	8 y
10/F/13 mo	13 mo	Open, with interposition tibial periosteum graft	4 d	None	None	8 y
11/M/12 mo	12 mo	Open, with interposition tibial periosteum graft	8 d	None	None	5 y
12/F/10 mo	11 mo	Open, with interposition tibial periosteum graft	12 d	Normal	Secondary surgery for residual communication	2.5 y
13/M/4 mo	7 mo	Open, with interposition tibial periosteum graft	8 d	Normal	Mild aspiration of thin liquid	5 y
14/M/1 mo	2 mo	Open, with interposition tibial periosteum and auricular cartilage graft	Unknown	Normal	None	14 mo
15/F/3 mo	5 mo	Open, with interposition tibial periosteum graft	7 d	Normal	Subcutaneous emphysema postoperatively; thrombosis of the superficial femoral vein	1.5 y
16/M/15 d	1 mo	Open, with interposition tibial periosteum graft	7 d	None	Secondary surgery for loosening of the sutures at the upper part of the graft; tracheotomy due to subglottic edema and tracheomalacia	2 y
17/F/7 mo	18 mo	Open, with interposition auricular cartilage graft	4 d	Normal	Pneumonia	1 y
18/F/1 mo	1 mo	Open, with interposition tibial periosteum graft	8 d	Aspiration with liquids	Mild aspiration of thin liquid	3.5 y
19/M/7 mo	7 mo	Open, with interposition tibial periosteum graft	18 d	None	Tracheotomy due to significant subglottic edema and tracheomalacia	6 y
20/M/2 mo	2 mo	Open, with interposition auricular cartilage graft	6 d	None	None	10 mo
21/F/8 mo	8 mo	Open, with no interposition	4 d	None	None	1 y
22/F/1 mo	1 mo	Open, with no interposition	2 d	None	None	2 y

Abbreviation: MSS, modified swallow study.

servative monitoring with thickened feed. However, they continued to aspirate with thin liquid both clinically and on documented swallow studies.

Open reconstruction approach with or without interposition graft was used in 16 patients: interposition tibial periosteum graft (n=11), interposition auricular cartilage graft (n=2), a combination of these 2 grafts (n=1), and no interposition graft (n=2). Endoscopic approach for repair of laryngeal cleft was used in 6 patients: 3 with type 1 cleft, 2 with type 2, and 1 with type 3.

The duration of postoperative intubation ranged from 1 to 4 days (mean duration, 1.8 days) in the endoscopic group. Patients who underwent an open approach had a mean postoperative intubation period of 7.3 days (range, 2-18 days) in 14 cases, an unknown intubation period in 1 case, and a preexisting tracheotomy in 1 case, decannulated several months after repair of the laryngeal cleft.

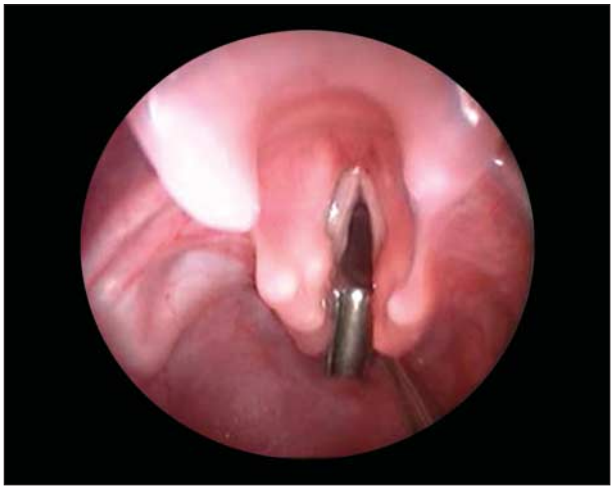

Figure 1. Type 2 laryngeal cleft.

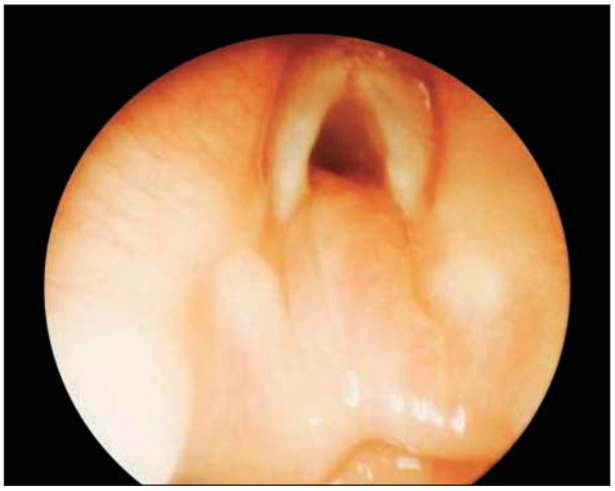

Figure 2. Type 3 laryngeal cleft.

Eight patients (36%) experienced a complication. Overall, complications included pneumonia (n=2), persistence of aspiration with thin liquid (n=2), secondary surgery owing to residual communication or loose sutures (n=2), subcutaneous emphysema and thrombosis of superficial femoral vein (n=1), and need for tracheotomy owing to subglottic edema and tracheomalacia (n=2). The duration of follow-up ranged from 10 months to 8 years (mean follow-up, 36 months) for the 22 patients.

COMMENT

The incidence of laryngeal cleft is approximately 1 in 10 000 to 20 000 live births.[2] It is more common in boys than girls, with a ratio of 5:3.[3,4] The affect of prematurity and hydramnios as contributing factors remains controversial. A possible autosomal dominant pattern of inheritance has also been reported in some families.[3]

Embryologically, the trachea and esophagus share a common lumen until they are separated by the development of the tracheoesophageal septum. Failure of this fusion and incomplete development of the tracheoesophageal septum may lead to congenital abnormalities such as isolated laryngeal cleft, tracheoesophageal fistula, and esophageal atresia, depending on the severity of the abnormality.[5,6]

As Bell et al[7] report, the first patient with laryngeal cleft was described by Richter in 1792. In 1955, Pettersson[8] reported the first surgical repair of a type 1 laryngeal cleft. Several classification systems for laryngeal cleft have been introduced by different investigators: Cohen[9] in 1975, Armitage[10] in 1984, and Evans[11] in 1985. In 1989, Benjamin and Inglis[1] presented a classification system in which 4 types of cleft were described: type 1 is a supraglottic interarytenoid defect that extends inferiorly no further than the level of the true vocal folds; in type 2, the cricoid lamina is partially involved, with extension of the cleft below the level of the true vocal folds; type 3 is a total cricoid cleft that extends completely through the cricoid cartilage with or without further extension into the cervical trachea; and type 4 extends into the posterior wall of the thoracic trachea and may extend as far as the carina.

Any newborn with feeding problems, repeated aspiration, and respiratory distress should have a thorough evaluation. A complete prenatal and birth history must be taken. Congenital infections, maternal drug use, hypoxia, or birth trauma may cause temporary or permanent feeding difficulty. Other conditions such as esophageal stricture, tracheoesophageal fistula, cricopharyngeal spasm, neuromuscular abnormalities, laryngomalacia, GER, and vocal cord paralysis should be included in the differential diagnosis of these patients. It is essential to have a high index of suspicion for other associated abnormalities including gastrointestinal (16%-67%), genitourinary tract (14%-44%), and cardiovascular (16%-33%) that have been reported in patients with laryngeal cleft.[2,4,12] A higher incidence of laryngeal cleft is also reported with Pallister-Hall syndrome and Opitz-Frias syndrome.[12] Roth and colleagues[2] reviewed 85 well-documented cases of all subtypes of cleft and reported an overall mortality rate of 46% due to laryngeal cleft and associated congenital abnormalities.

Stridor, choking, cyanosis, and regurgitation are typical manifestations of laryngeal cleft or laryngotracheoesophageal cleft. Routine chest radiography might show pulmonary infiltrates secondary to aspiration. Modified swallow studies and fiberoptic endoscopic evaluations of swallowing are also helpful in evaluation of laryngeal cleft. However, it is often difficult to differentiate laryngeal incompetence and aspiration due to a laryngeal cleft vs neuromuscular incoordination. It is also important to realize that modified swallow studies and fiberoptic endoscopic evaluations of swallowing only document several cycles of swallowing and may produce normal results if the child is aspirating only intermittently. Flexible laryngoscopy gives a limited view of the posterior glottic space and should not be relied on to make the diagnosis.

Microlaryngoscopy under general anesthesia remains the gold standard in the diagnosis of laryngeal cleft. Laryngeal cleft may be obscured by redundant laryngeal and/or esophageal mucosa prolapsing into the cleft. Therefore, it is of paramount importance to examine the larynx carefully, and the arytenoids must be parted with a probe to make the correct diagnosis. We noted other significant associated findings in 50% of our patients at the time of endoscopy, which is similar to the findings in other

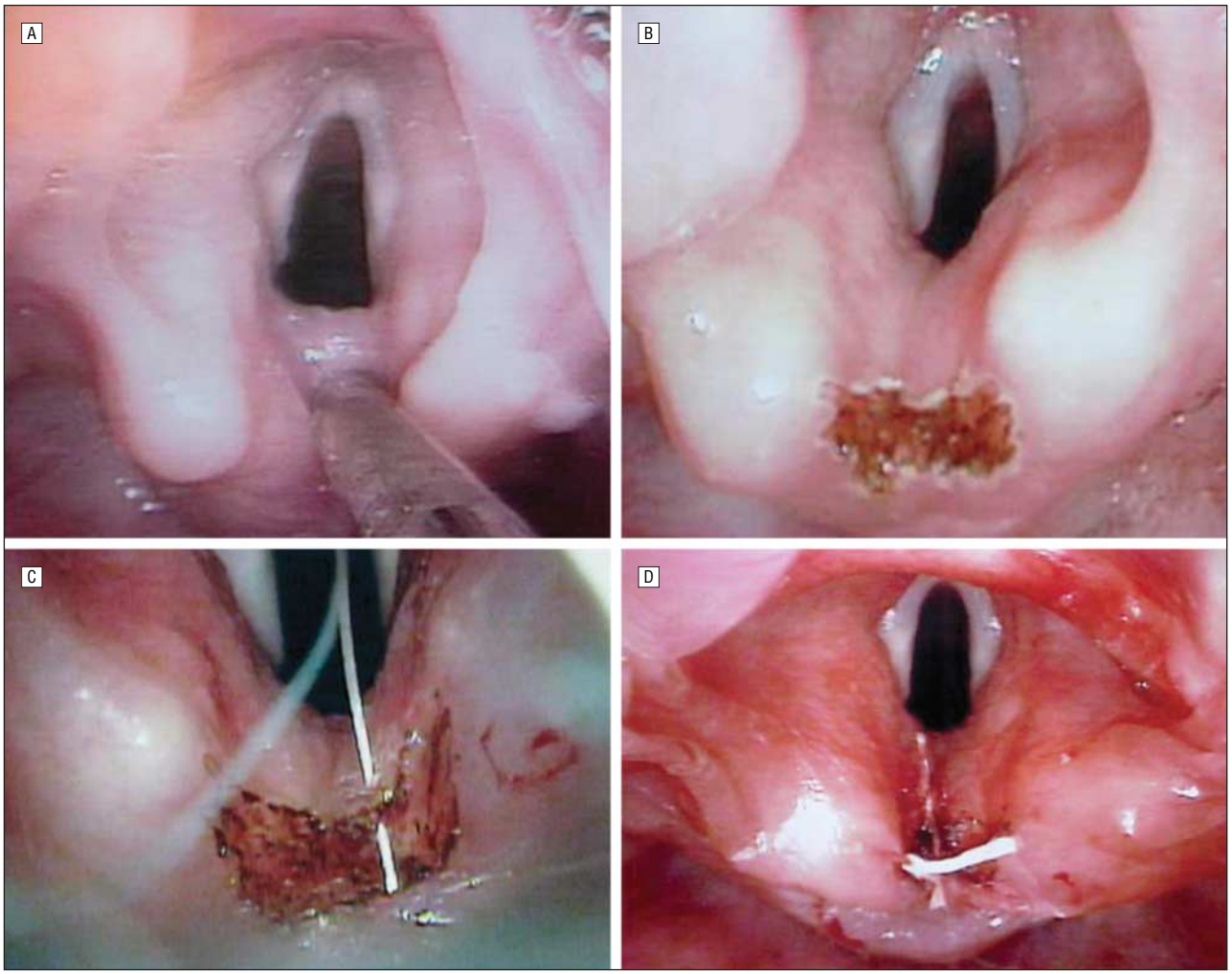

Figure 3. Endoscopic approach to repair a type 1 laryngeal cleft. A, Type 1 laryngeal cleft at presentation; B, after carbon-dioxide laser cut; C, first suture being placed in the deepest part of the cleft; D, closure of the cleft using endoscopic approach.

studies: Parsons et al,[13] 66%; Evans et al,[14] 65%; and Moungthong and Holinger,[15] 64%. One of the most common associated findings is tracheobronchomalacia.[16,17] We recommend performing the airway endoscopy with the patient breathing spontaneously and with no positive airway pressure to avoid masking the signs of airway collapse. The presence of tracheoesophageal fistula has also been reported in 20% to 37% of patients with laryngeal cleft.[6,14] Therefore, it is essential to perform a complete evaluation of tracheobronchial tree and esophagus under general anesthesia to rule out other associated abnormalities in all patients with laryngeal cleft.

The timing and approach for surgical repair depends on the severity of symptoms, associated abnormalities, and the type of cleft. In small clefts, the proper diagnosis might be missed, and the patient could survive into adult life with recurrent respiratory infections.[18,19] In the more extensive form, the condition may be fatal unless properly diagnosed and corrected surgically in a timely fashion. Some authors have advocated observation and expectant management of minor clefts using positioning and thickened food to prevent aspiration.[20,21] We believe early diagnosis and surgical repair of laryngeal cleft is important to reduce the irreversible pulmonary damage and other associated morbidities that may occur as a result of repeated aspiration.

In the past decades different operative approaches have been proposed for the management of laryngeal cleft. In 1967, According to Evans,[11] Jahrsdoerfer and colleagues described the anterior approach, which involves exposure of the cleft by means of thyrotomy, cricoidotomy, and tracheofissure to the first and second rings to provide direct access to the posterior cleft and allow for microsurgical closure. Some authors have advocated avoiding the anterior approach owing to possible risk of laryngeal instability and long-term laryngeal growth disturbances.[11,22] The lateral pharyngotomy approach has also been described as an approach that avoids the laryngofissure.[11] In our experience, and as reported by other investigators,[23] meticulous closure under magnification, proper stenting with a nasotracheal tube, and excellent postoperative care in the intensive care unit will limit the risk of laryngeal instability with the anterior approach. For a more extensive cleft involving the thoracic trachea, a lateral pharyngotomy with a right thoracotomy approach or an anterior approach with a median sternotomy has been used.[5,24]

One of the well-known complications of laryngeal cleft surgery is the risk of loosening and dehiscence of sutures at the site of cleft repair. Anastomotic leaks are reported to occur in approximately 50% of the repairs and generally require reoperation.[4] This can be due to a combination of factors such as laryngeal movement, GER, coughing, and pressure caused by an endotracheal tube or nasogastric tube.[25] Investigators have advocated the use of an interposition graft such as the sternocleidomastoid muscle,[26] costal cartilage,[27] and pleural flap[28] to decrease the risk of dehiscence. In 1998, Garabedian and colleagues[25] reported the use of tibial periosteum as an interposition graft. The theoretical advantages of this graft include a strong resistance to necrosis and its osteogenic properties. It also has the advantages of being thin, rigid, and easy to handle. The disadvantage of the tibial periosteal graft is the possibility of donor site morbidity.

Other complications of surgical repair include laryngeal nerve injury, granulation tissue formation, esophageal stricture, and continued aspiration. It has also been reported that some of the feeding issues may continue for a short period postoperatively, despite successful surgical repair of the cleft, due to neurological disturbance of the swallowing reflex.[23,25]

Six of our patients underwent an endoscopic approach for repair of laryngeal cleft. Endoscopic repair was performed using suspension microlaryngoscopy under general anesthesia with spontaneous breathing. The larynx was visualized with a Lindholm laryngoscope. A carbon dioxide laser at a setting of 3 W at 0.3-second intermittent mode was used to denude the mucosal margin of the cleft. It is of paramount importance to completely remove the mucosa at the apex of the cleft to prevent persistence of the fistula at the lower end of the repair.

Absorbable interrupted sutures (4-0 Vicryl; Ethicon Inc, Somerville, NJ) were used to close the cleft. The first suture is the most important and must be placed at the most inferior extent of the cleft. We generally place 3 to 4 sutures, depending on the extent of the cleft (Figure 3).

The success of the endoscopic approach depends on proper exposure of the cleft at the time of suspension laryngoscopy and magnification under microscope. In our experience, avoiding endotracheal intubation and maintaining general anesthesia under spontaneous breathing allow for much better exposure and increase the chances of success. Based on our experience, we advocate the minimally invasive endoscopic approach for types 1 and 2 clefts. We have repaired 1 case of type 3 in which the cleft extended through the cricoid but did not involve the tracheal rings with a good outcome. Our experience is very limited with the endoscopic repair of a type 3 cleft, and to our knowledge, no large series with long-term follow-up has been conducted. Based on our limited experience, the endoscopic approach for type 3 laryngeal cleft may be considered if there is no involvement of the tracheal rings.

Sixteen of our patients underwent the open approach for repair of the cleft. An interposition graft was used in 14 of these patients, as described by Garabedian and colleagues.[25] The harvesting of the tibial graft is a straightforward process and should be done on the anteromedial surface of the tibia to prevent any consequences on tibial growth.[25] It is also essential to secure the periosteal graft laterally to prevent it from moving and folding on itself.[25] An auricular cartilage graft or temporalis fascia can also be used as an interposition graft with good success. We advocate using the open approach with an interposition graft in patients who have type 2 clefts when adequate endoscopic exposure is limited or in the case of a more extensive cleft.

Regardless of surgical approach, a number of critically important factors should be addressed in the preoperative period. Gastroesophageal reflux must be evaluated and controlled with medical therapy and/or surgical intervention prior to repair of the cleft. We believe that Nissen fundoplication should be considered, especially in children with increased pharyngeal and tracheobronchial secretions and recurrent aspiration pneumonia. All patients should continue with anti-GER medication postoperatively and have a close follow-up to be sure the GER is under control.

The other issue is the degree of tracheomalacia and/or tracheobronchomalacia that is seen in these patients, which may cause further difficulty with airway symptoms and even prevent extubation despite proper surgical repair of the cleft. Even though the natural history of tracheomalacia is one of gradual improvement as the child grows, most of these patients may require intervention during the preoperative and postoperative periods. Treatment options include continuous positive airway pressure, stenting the airway by prolonged intubation, aortopexy, and tracheotomy. We do not advocate prolonged endotracheal stenting postoperatively owing to the risk of laceration and dehiscence of the cleft repair. Tracheotomy is a viable option for some of these patients despite known morbidity and mortality.

Based on our experience and reports by other investigators,[4,7,9,12] it is clear that the management of laryngeal cleft is challenging from both the medical and surgical standpoints. Different surgical options have been proposed, and no option is applicable to all patients. Each patient should be assessed properly, and a surgical approach should be individualized based on the symptoms, other associated findings on airway endoscopy, and the type of the cleft.

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