Endoscopic Surgical Treatment of Laryngotracheal Clefts

Indications and Limitations

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Objective: To present the indications, techniques, results, and limitations of endoscopic surgical treatment of laryngotracheal cleft.


Setting: Department of Otolaryngology–Head and Neck Surgery, Armand Trousseau Children’s Hospital, Paris, France.

Patients: Eleven patients who underwent endoscopic cleft closure as a primary (n=8) or secondary (n=3) procedure among 22 patients treated for laryngotracheal clefts. We report patients’ demographics, symptoms leading to the diagnosis, endoscopic evaluation method, medical examination results, and surgical techniques.

Intervention: Endoscopic closure of the cleft under spontaneous ventilation via 2-layer interrupted sutures after excision of the mucosal edge using a carbon dioxide laser in 10 patients and a thulium laser in 1.

Main Outcome Measures: Analysis of postoperative complications, revision surgery, need for intensive care unit admission, closure of the cleft, and long-term symptom results.

Results: Successful closure of the 11 clefts (with revision surgery in 3 patients) without the need for intubation or intensive care unit admission enabled the elimination of aspiration in 10 patients and significant improvement in 1 patient with bilateral vocal cord paralysis. There were no significant postoperative complications.

Conclusions: Endoscopic closure of laryngotracheal clefts is a reliable technique that significantly reduces perioperative and postoperative morbidity. The results of this technique are entirely satisfactory, and we suggest that it is suitable as a primary procedure for the treatment of type I, II, and III clefts extending to the cervical trachea, including in neonates.


Original Article

SINCE ITS FIRST DESCRIPTION BY Pettersson1 in 1955, cervical surgery, with or without a combined thoracic approach, was the standard for the treatment of laryngotracheal clefts.2 Among different kinds of procedures, we also proposed in 1998 an open approach via a laryngofissure that included interposition of a tibial periosteal graft between the esophageal and laryngotracheal mucous membranes.3 However, the advent of minimally invasive endoscopic surgery in the 1980s4-7 modified the treatment of type I and II clefts and gave rise to the possibility of endoscopic treatment of type III clefts. Through 11 clinical cases involving primary or secondary endoscopic procedures, we examine the advantages and limitations of the endoscopic approach.

METHODS

Twenty-three patients with laryngotracheal clefts underwent surgery between 1994 and 2008 in the Department of Otolaryngology–Head and Neck Surgery at the Armand Trousseau Children’s Hospital. We describe 11 of these patients treated endoscopically via primary or secondary procedures between May 1, 2005, and February 28, 2009.

We conducted a retrospective case note review to determine the following factors: sex, age at diagnosis, initial symptoms, associated malformations, associated surgical procedures, results of the endoscopic examination (type of cleft and associated laryngotracheobronchial malformations), age at surgery, number of procedures, follow-up period, and post-surgical complications.

In our practice, endoscopic examination is the only examination necessary for diagnosis. For each case, the larynx is exposed using a l-
ryngal spreader and is palpated using a probe to confirm the diagnosis and measure the length of the cleft. Exposure and palpation are greatly facilitated by local application of a lidocaine hydrochloride spray, the dilution of which is adjusted for the child’s age and weight. The type of cleft is classified according to the method of Benjamin and Inglis.8

The procedure consisted of 2 stages. First, the mucosal margins and apex of the cleft were excised using a carbon dioxide laser linked with a micromanipulator (AcuSpot; Lumenis UK Ltd, London, England) in continuous superpulse mode at power of 3 to 5 W in 10 patients and a thulium laser in 1 patient. Second, the mucosa was sutured along the pharyngoesophageal and laryngotracheal wall using interrupted monofilament 5/0 and 7/0 sutures (Monosof; Covidien, Mansfield, Massachusetts) and an 8-mm half-curved needle. We used a knot pusher (Pilling, Fort Washington, Pennsylvania) and an adapted Storz needle holder.

At the end of the procedure, a nasogastric tube was inserted to commence feeding on day 1, and antireflux treatment with proton-pump inhibitors was begun. The child was awakened and monitored in the recovery room during the first night before returning to the department ward. Oral feeding was begun between days 7 and 14 after an endoscopic examination. Endoscopic examinations were then performed after 1 month, 3 months, 6 months, and 1 year, and annually thereafter.

### RESULTS

Of the 11 patients with laryngotracheal clefts treated endoscopically, 8 were treated with a primary procedure and 3 with a secondary procedure after previous surgery using an external approach. We report the 2 groups separately.

### PRIMARY PROCEDURE

For these 8 patients, age at diagnosis ranged from 3 days to 5 years, with a mean age of 14 months (Table 1). The initial symptoms consisted of aspiration (n=6), recurrent pneumonia (n=4), and aphonia (n=1). Untypically, there were no patients with stridor. The endoscopy revealed 4 cases of type III cleft extending to the trachea (first and second tracheal rings) and 2 cases each of type I and II clefts.

Patient age on the day of surgery ranged from 14 days to 5 years, with a mean age of 12.5 months. Complete

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### Table 1. Patients Who Underwent Primary Endoscopic Treatment

<table>
<thead>
<tr>
<th>Patient Sex/ Age at Referral</th>
<th>Initial Symptoms</th>
<th>Associated Malformations or Conditions</th>
<th>Associated Surgical Procedures</th>
<th>Endoscopy: Type and Associated Malformations</th>
<th>Age/Weight, kg, at Endoscopic Surgery</th>
<th>Procedures for Complete Closure, No.</th>
<th>Follow-up, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>M/3 d</td>
<td>Aphonia</td>
<td>None</td>
<td>None</td>
<td>Type III (1 partial ring)</td>
<td>20 d/3.2</td>
<td>1</td>
<td>27</td>
</tr>
<tr>
<td>F/9 d</td>
<td>Aspiration</td>
<td>None</td>
<td>None</td>
<td>Type III (2 full rings); tracheal right bronchial stump</td>
<td>14 d/2.6</td>
<td>1</td>
<td>29</td>
</tr>
<tr>
<td>F/1 mo</td>
<td>Aspiration/recurrent pneumonia</td>
<td>Pyelocalyceal dilatation</td>
<td>None</td>
<td>Type III (2 full rings)</td>
<td>1 mo/3.4</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>M/24 mo</td>
<td>Aspiration/recurrent pneumonia/ bronchial obstruction</td>
<td>Optiz syndrome</td>
<td>Testicular ectopia</td>
<td>Type III (1 partial ring)</td>
<td>24 mo/11</td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td>M/2 mo</td>
<td>Aspiration/recurrent pneumonia</td>
<td>Trisomy 21; type III esophageal atresia</td>
<td>Correction of esophageal atresia at day 1</td>
<td>Type II</td>
<td>2 mo/3.5</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>M/5 mo</td>
<td>Bronchial obstruction</td>
<td>Type IV esophageal atresia; mild tracheomalacia</td>
<td>Ligation of the tracheal fistula; esophagostomy on day 0; esophageal anastomosis at 4 mo; Nissen gastrostomy</td>
<td>Type II; right laryngeal palsy</td>
<td>5 mo/4.5</td>
<td>1</td>
<td>31</td>
</tr>
<tr>
<td>M/5 y</td>
<td>Aspiration</td>
<td>Microcephalic encephalopathy (ATR-X genetic anomaly)</td>
<td>None</td>
<td>Type I</td>
<td>5 y/15</td>
<td>1</td>
<td>14</td>
</tr>
<tr>
<td>M/10 mo</td>
<td>Aspiration/recurrent pneumonia</td>
<td>None</td>
<td>None</td>
<td>Type I</td>
<td>28 mo/11</td>
<td>1</td>
<td>48</td>
</tr>
</tbody>
</table>

Abbreviation: ATR-X, X-linked thalassemia mental retardation syndrome.
closure of the cleft and resolution of aspiration was achieved with a single procedure in 6 patients. In 2 patients with type III cleft, it was necessary to perform an additional procedure to extend the closure above the glottis to eliminate a few infrequent aspirations involving liquids, not associated with pneumonia. No complications were associated with the surgery. With mean follow-up of 18.3 months (range, 2-48 months), we did not observe any secondary dehiscence.

SECONDARY PROCEDURE

For the 3 patients treated with a secondary procedure after the primary open approach, the time between the last procedure using a cervical approach and the endoscopic procedure was 2 to 9 years. Symptoms that led to a subsequent supplemental procedure consisted of aspiration (n=3) and recurrent pneumonia (n=2) (Table 2). Endoscopy revealed 1 residual type I cleft and 2 residual type II clefts. The patients’ ages on the day of endoscopic surgery were 2, 4, and 9 years.

Complete closure of the cleft was achieved in all 3 patients (1 procedure in 2 patients and 3 procedures in 1 patient), with resolution of all initial symptoms in 2 patients. However, the third patient, with bilateral vocal cord paralysis in abduction, continued to aspirate but with a significant reduction in the number and severity of episodes. This child died 1 year later of digestive complications not directly related to the laryngeal cleft.

There were no complications of surgery. With mean follow-up of 17 months (range, 12-24 months), we did not observe any secondary dehiscence. No patients required postoperative intubation or intensive care unit admission. All of the patients who underwent primary and secondary procedures were treated for reflux as follows: feeding exclusively via a nasogastric tube plus gastrointestinal antireflux drug treatment (n=3). Nissen gastrostomy (n=3), or thickened diet plus gastrointestinal antireflux drug treatment (n=5). During the postoperative phase, patients who did not undergo Nissen gastrostomy were treated with proton-pump inhibitors for 1 year.

For first-line closures, feeding was resumed on day 1 for patients with type I cleft and on days 8 to 10 for those with type II and III clefts. For secondary closures, feeding was resumed on day 14. The clefts were associated with a syndrome in 40% of the patients and with other malformations in 30%, which is consistent with other series reported in the literature.

<table>
<thead>
<tr>
<th>Patient Sex/ Age at Referral</th>
<th>Initial Symptoms</th>
<th>Associated Malformations or Conditions and Surgical Procedures</th>
<th>Cleft and Cervical Surgery</th>
<th>Type of Cleft and Vocal Cord Mobility</th>
<th>Age, y/ Weight, kg, at Endoscopic Surgery</th>
<th>Procedures for Complete Closure, No.</th>
<th>Follow-up, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>F/1 y</td>
<td>Aspiration/ recurrent pneumonia</td>
<td>Nissen gastrostomy</td>
<td>Type III (4 rings); cervical approach (3 times) at 1 mo, 1 y, and 2 y</td>
<td>Type I</td>
<td>4/9.7</td>
<td>3</td>
<td>15</td>
</tr>
<tr>
<td>F/1 mo</td>
<td>Aspiration/ stridor</td>
<td>Tracheal duplication; dextrocardia (chromosome 22q11 microdeletion)</td>
<td>Type III; cervical approach at 1 mo</td>
<td>Type II</td>
<td>2/9.6</td>
<td>1</td>
<td>24</td>
</tr>
<tr>
<td>F/9 y</td>
<td>Aspiration/ recurrent pneumonia</td>
<td>Opitz syndrome; type III esophageal atresia; Roux-en-Y anastomosis and gastrostomy at birth; tracheomalacia and tracheostomy; obstructed Aboulker stent; Wharton and Stenson ligation</td>
<td>Type II cleft; cervical approach at 1 mo</td>
<td>Type II; bilateral laryngeal palsy in abduction</td>
<td>9/25</td>
<td>1</td>
<td>12 (Death due to digestive complications)</td>
</tr>
</tbody>
</table>

Endoscopic treatment of clefts was reported for the first time by Yamashita in 1979. Taking advantage of the technological advances in endoscopy equipment during the 1980s, and to reduce the perioperative and postoperative morbidity associated with open management, several researchers proposed this technique for the primary treatment of type I and II laryngeal clefts or as a supplemental procedure in the event of upper dehiscence after a primary external procedure.

Most researchers acknowledge that this procedure should be performed under spontaneous ventilation to provide an optimal operating field. Coordination with the anesthesia team is essential to the success of the surgery. None of the present patients needed endotracheal intubation during or after the procedure.

This technique depends on good endoscopic exposure, and a potential limitation of this exposure is limited surgical access, such as in newborns with low weight and patients with Pierre Robin sequence or craniofacial abnormalities. Assessment of access at initial endoscopy is essential.

However, opinions differ regarding the excision technique for the mucosal margins of clefts. Similar to Rah-
bar et al,9 we excise the mucosal margins and apex of the cleft using a carbon dioxide laser in superpulse mode. We believe that the value of the laser lies in its ability to induce a reactionary fibrosis that makes the closure more stable. Using the carbon dioxide laser in ultrapulse mode, Sandu and Monnier12 excised the mucosa of the lateral edge of the cleft superficially from bottom to top and then more deeply to separate the laryngotracheal wall from the pharyngoesophageal wall. Koltai et al10 excised the cleft mucosa from one arytenoid to the other, passing through the base of the cleft, and formed 2 mucosal flaps using microscissors. Waltzman and Bent13 formed an anterior mucosal flap and a posterior mucosal flap on either side of the cleft. These flaps were then crossed, overlapped, and sutured along the medial line, shifting their free edges.

The mucosa is then sutured with 1 or 2 layers, from bottom to top, using interrupted polyglactin 910 (Vicryl) or polyamide 6 (Ethilon) (both from Ethicon Inc, Johnson & Johnson Co, Somerville, New Jersey) 4/0 to 6/0 sutures and a half-curved needle. We prefer to close the laryngeal cleft with the standard 2 layers. However, it is not always achievable and, in this series, 2 type III cases were closed with a single layer, but we did not observe any difference in outcome.

Successful instrumentation of the distal part of type III clefts is a potential limitation of the procedure. Studies with more cases of endoscopic type III repairs are necessary to determine whether there is any real effect on efficacy. We believe that more specifically designed instrumentation will facilitate access to the apex and allow the endoscopic management of type III clefts extending further than the second tracheal ring.

Three patients required extension of the closure to eliminate a few infrequent aspirations. The revision rate is comparable with that of the open procedure.9,11-13 Nevertheless, of 4 patients who underwent type III endoscopy, 2 required a revision procedure. No cases were closed leading to dyspnea or dysphagia.

Other endoscopic techniques have been described, but their indications remain to be evaluated and seem to be limited to type I clefts: (1) injection of collagen14 or absorbable gelatin (Gelfoam; Pfizer, New York, New York)15 (however, the resorption of these materials requires long-term studies before any conclusions can be reached regarding their efficacy) and (2) injection of polidimethylsiloxane gel (Bioplastique; Enteric Medical Technologies, Inc, Foster City, California)16 (however, its long-term tolerance remains to be demonstrated).

At the end of the procedure, we insert a nasogastric tube for feeding, and the child is awakened immediately and is monitored under spontaneous ventilation in the recovery room during the first night. Sandu and Monnier12 described the use of a bilevel positive airway pressure machine in the immediate postoperative period, without suture leakage. Only Chien et al17 recommended routine intubation for 1 to 10 days.

Feeding is then resumed, depending on the type of cleft, between days 1 and 14. We believe that feeding can be begun on day 1 for type I and II clefts and on day 7 for type III clefts. We did not conduct systematic formal swallow studies after surgery but rather based surveillance on the clinical assessment of aspiration.

In the context of a second-line surgical procedure, we start feeding on day 14. We recommend a soft diet for 10 days to preserve the integrity of the sutures. We also believe that the major advantage of this minimally invasive technique is the lower morbidity perioperatively and postoperatively. Indeed, there is negligible risk of nerve damage; complications involving the stability and growth of the larynx and trachea; ischemia associated with intubation or tracheotomy; infections, hematomas, and so on associated with external surgery; and general complications associated with prolonged sedation and endotracheal intubation in the intensive care unit. Moreover, operating time is substantially reduced, thereby minimizing anesthesia-related complications.

This surgery seems to be easily reproducible and does not affect the options associated with an external approach in case of failure. It should be performed as early as possible to protect the respiratory tree from aspiration. In our experience, early surgery has allowed us to maintain excellent ventilatory function to allow spontaneous ventilation in some patients for up to 2 hours.

The initially reported results for the treatment of type I and II clefts through primary or secondary procedures were satisfactory. Koltai et al10 reported an 81% success rate for 6 primary and 5 secondary type I cases. Evans7 reported 81% for 11 type I and 2 type II cases, Chien et al17 reported 94% for 16 type I cases, and Ketcham et al18 reported 68.8% for 16 type I cases. The results of the present series of type I and II clefts (83% closures for single procedures) seem to be consistent with those of other studies.

This surgery was recently proposed in the treatment of type III clefts. A review of the literature3-12 revealed 1 case of endoscopic treatment of a cleft extending to the trachea and 4 cases of total cricoid clefts.

Although needing more than 1 endoscopic procedure in 2 patients, our 4 cases of endoscopic surgical treatment of clefts extending to the trachea support the efficacy of the single case previously reported in the literature. However, the rate of wound breakdown for type III clefts is approximately the same as that for the open approach.

Three of the present procedures were performed on neonates younger than 1 month. At mean follow-up of 16.5 months (range, 4-29 months), these clefts seem to be closed, with excellent clinical results in terms of resolution of aspiration.

We did not experience respiratory difficulties related to tracheomalacia in 10 patients, including 1 with mild tracheomalacia with associated esophageal atresia, but in 1 patient with Opitz syndrome, severe tracheomalacia necessitated a tracheostomy preoperatively. Tracheomalacia does not seem to be correlated with the extent of the cleft, at least for type II to IV.

In conclusion, the results of this technique seem to be established for the treatment of type I and II clefts and are very promising for type III clefts. However, further studies involving more patients are required to definitively establish efficacy. However, we believe that endoscopic repair is a suitable primary procedure in the treatment of type I, II, and III clefts extending to the cervical trachea.
Submitted for Publication: June 11, 2009; final revision received August 26, 2009; accepted September 29, 2009.

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Study supervision: Garabedian, Pezzettigotta, and Roger.

Financial Disclosure: None reported.

Previous Presentations: This work was presented at the American Society of Pediatric Otolaryngology Annual Meeting; May 23, 2009; Seattle, Washington.

REFERENCES


