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Endoscopic repair of laryngotracheoesophageal clefts: Experience in 17 cases

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ABSTRACT

Objective: To review the presentation and evaluation of laryngotracheoesophageal clefts as well as their treatment modalities, especially endoscopic closure.
Study design: retrospective case series.
Methods: All patients treated for laryngotracheoesophageal clefts in our clinic during the last 15 years were included. Analysis of preoperative data, surgical success and functional outcome was performed.
Results: A total of 18 patients were included in our study. Cleft distribution was: type I (n=1), type II (n=3), type IIIa (n=5), type IIIb (n=8) and type IVa (n=1). All clefts were closed endoscopically by CO_2 laser repair except for two patients who benfited from open surgery (one type I, one type IIIb). 7 of our 18 patients (39%) experienced a complication necessitating reoperation. Surgical treatment of LTEC allowed cessation of feeding tube assistance and artificial ventilation in 47% and 42% of patients respectively.
Conclusion: Surgical treatement of laryngotracheoesophageal clefts remains a complex procedure with a high rate of morbidity for high grade clefts. Post-surgical difficulties in feeding and breathing are associated with concomitant congenital anomalies. Endoscopic repair is a successful technique for treating up to grade IIIa laryngealc lefts. Further investigation is needed to assess the best approach for treating longer clefts.

1. Introduction

Laryngotracheoesophageal cleft (LTEC) is a rare congenital malformation of the respiratory tract which was first described by Richter in 1792. The first successful surgical repair was described by Pettersson in 1955 [1]. Many pathophysiological descriptions have been proposed concerning the development of LTECs but no definitive explanation has arisen [2].

The incidence of congenital laryngeal abnormalities is estimated at 0.2–1.5% with LTEC being found in approximately 1/10 000–20 000 [3–5] live births though most authors agree that this is an underestimation. The incidence of posterior laryngeal cleft in children examined by microlaryngoscopy has recently been observed between 6.2 and 7.6% [6,7]. This significant difference has been explained by a higher referral rate for suspicion of laryngeal cleft combined with better diagnostic techniques, notably by systematic palpation of the posterior larynx with a probe during microlaryngoscopy. A slightly higher incidence has been noted in boys than in girls.

Common symptoms due to laryngotracheal cleft arise from feeding difficulties and respiratory problems. These symptoms include cough, wheezing, stridor, hoarse cry, dyspnea, cyanotic spells during feeding, and recurrent pulmonary infections. Generally the severity of symptoms is proportionate to the length of the cleft [8,9]. While some grade I clefts are practically asymptomatic, a more extensive cleft will rapidly bring the patient to consult. Type III and type IV clefts are associated with a high mortality rate, often precipitated by the fact that multiple congenital anomalies are present. Despite different methods to surgically repair these high grade clefts, the post-operative mortality rate has remained high between 10 and 75% [10].

Development of different surgical techniques for LTECs has significantly improved the morbidity and mortality of this disease over the years. Low grade type I and type II clefts for which endoscopic closure is now considered a recommended primary procedure have a high surgical success rate. Controversy remains, however, concerning the repair of high grade type III and type IV clefts and the post-operative complication rate remains elevated. Endoscopic surgery has only recently been suggested for treating these high grade clefts which remain technically difficult to repair.

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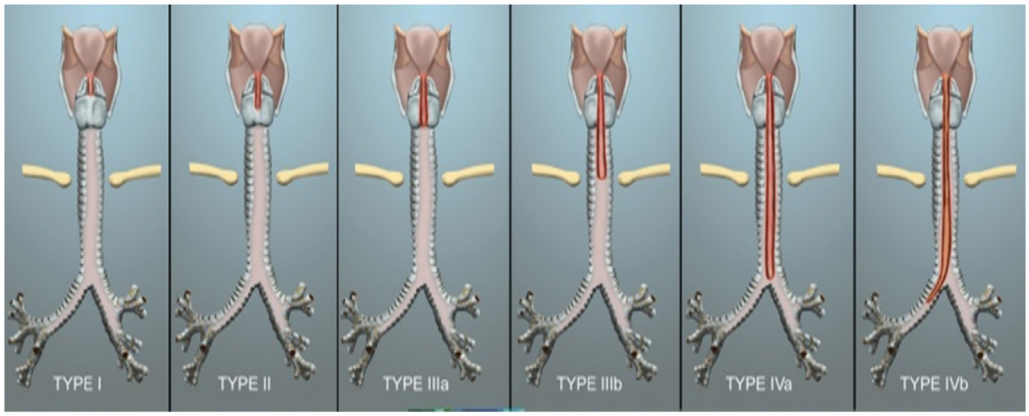

Fig. 1. Modified Benjamin-Inglis classification of laryngo-tracheo-esophageal clefts (LTECs). Type 0: submucosal cleft; Type I: interarytenoid cleft; Type II: partial cricoid cleft; Type IIIa: total cricoid cleft; Type IIIb: LTEC extending into the extrathoracic portion of the trachea; Type IVa: LTEC extending to the carina; Type IVb: LTEC extending into one main bronchus.

This could lead to a high rate of complications and residual tracheoesophageal fistula (TEF). It is essential therefore to have a surgical team with an extensive experience in endoscopic surgery and especially an experienced pediatric anesthesiologist in order to bring together the proper conditions for a successful endoscopic repair, without need for a tracheostomy.

We present our experience with laryngeal cleft repair and especially endoscopic closure of high grade clefts by CO_2 laser and double layer suturing. We outline our surgical technique, complications arising from surgery as well as surgical outcome by evaluating decannulation and cessation of feeding tube by the end of treatment.

2. Methods

We performed a retrospective case study of all patients in our database treated in our ENT clinic between 1997 and 2012. Data concerning demographics, past surgery, treatment modalities, surgical outcome, complications and long-term follow-up were collected. Our primary outcome for successful surgery was proof of complete closure during post-operative endoscopic examination. We also evaluated the improvement in feeding and breathing by determining which patients no longer needed feeding tubes, non-invasive ventilation or cannulas by the end of their treatment.

To classify cleft extension, we used a modified Benjamin's classification (Fig. 1) which distinguishes between partial and total cricoid clefts, extrathoracique and intrathoracique clefts, as well as bronchus extension [11].

2.1. Diagnostic endoscopy

In order to determine the staging of the cleft, all our patients first underwent direct laryngoscopy and bronchoscopy exams under general anesthesia with spontaneous ventilation. Low grade laryngeal clefts can sometimes be difficult to diagnose endoscopically due to overlapping mucosa giving the impression of an intact posterior wall. Palpation using a laryngeal spreader and probe during the exam was therefore used systematically in order to not misdiagnose these patients.

2.2. Surgical technique

Surgery was performed under general anesthesia with spontaneous respiration for all endoscopic closures. A Parsons laryngoscope was used to expose the cleft up to its caudal extremity. Using an ultrapulse CO_2 laser, we incised along both mucosal borders of the cleft in a V form from the caudal edge up to the cuneiform cartilage and thus separated the tracheal and pharyngoesophageal layers. We then proceeded to close alternately both laryngotracheal and pharyngoesophageal walls using separate Vicryl 5.0 sutures. The knots are placed alternately on both walls allowing a double layer closure. The upper limit of the cleft repair is situated just below the cuneiform cartilages. We generally used a collagen glue to reinforce the suture line after repair. Up to 20 stitches may be necessary in order to complete endoscopic closure. Reoxygenation of the patient was performed intermittently when necessary by introducing a ventilation tube in the airway. Almost all of our patients were transferred directly during the post-operative period to the pediatric ICU under bilevel-positive airway pressure if their respiratory status was permitting. Intubation was avoided as long as possible in order to prevent suture breakdown due to pressure lesions and inflammatory reaction caused by the tube. Positive pressure ventilation was used when necessary as a non-invasive approach to airway control helping to overcome post-operative tracheo-bronchomalacia, especially prevalent in type III and type IV LTECs. A small soft nasogastric tube was routinely placed at the end of the operation if the patient was not already equipped with a gastrostomy tube.

3. Results and analysis

A total of 19 patients treated between 1997 and November 2012 were included in our study (Tables 1 and 2). There was a slightly higher ratio of girls than boys, respectively, 53% and 47%. The main symptoms at referral were multiple bronchoinhalations, mucus hypersecretion, stidor, and difficulties during feeding. The most common associated pathologies were: gastroesophageal reflux ($n = 9$), atrial septal defect ($n = 7$), hypospadias ($n = 4$) and esophageal atresia ($n = 3$). Five patients presented with an associated syndrome including 3 Opitz, 1 VATER and 1 Pallister–Hall. Three of our patients had undergone previous unsuccessful surgery for their laryngeal cleft before being referred to our clinic.

Cleft distribution was: type I ($n = 1$), type II ($n = 4$), type IIIa ($n = 5$), type IIIb ($n = 8$) and type IVa ($n = 1$). The main other clinical findings during endoscopy were tracheomalacia ($n = 6$) and congenital subglottic stenosis ($n = 2$). Two patients were noted to have limited vocal cord abduction.

Significant associated abnormalities were discovered during the pre-surgical endoscopic examination in 50% of our cases. This remains on par with other studies where estimations range between 50% and 66% (7–50%, 3–65%, and 17–66%).