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Endoscopic repair of laryngotracheoesophageal clefts

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ABSTRACT

Background/Purpose: In Japan, surgical repair of a laryngotracheoesophageal cleft (LTEC) typically consists of the anterior approach, with the lateral approach as an alternative. Endoscopic surgery to repair the tracheoesophageal septum has been reported, and this study reviewed our experience treating several cases of LTEC endoscopically.
Methods: Endoscopic repair of LTEC was performed in 7 patients (3 boys, 4 girls; age range 4 months to 2 years 10 months; mean age 11 months; mean weight at surgery 7.23 kg; weight range 3.85–12.24 kg) between 2009 and 2014. LTEC was type I in 5 patients and types II and IV in 1 patient each. The patient with type IV was first operated on by the lateral approach, and the remaining cleft, which level was type III, was repaired endoscopically. Postoperative outcomes were retrospectively studied.
Results: Endoscopic surgery was successful in all patients. All 6 patients with types I and II LTEC were extubated easily, while in the patient with type IV LTEC, it was difficult to remove the tracheostomy cannula because of tracheomalacia. Postoperatively, tracheostomy cannulation became more stable, and the patient is gradually being weaned off the ventilator. All patients could be fed orally without difficulty postoperatively.
Conclusions: Endoscopic surgery provides a view from the cephalic aspect permitting the surgeon to form a normal larynx with only minimal risk of complications.

Laryngotracheoesophageal cleft (LTEC) is a particularly rare congenital deformity that occurs in the embryonic stage because of a malformation of the trachea and the tracheoesophageal septum [1]. According to Benjamin and Ingles' classification of the disorder [2], type I cleft limited to the supraglottic interarytenoid and type II is a partial cricoid cleft. Type III is a cleft extending to the proximal trachea. Rarer still is types IV, which extend to the distal trachea or main bronchus. (Fig. 1) In Japan, surgical repair typically consists of the anterior approach [3], where a longitudinal incision is made in the trachea to the cleft in order to form a septum. An alternative surgical strategy is the lateral approach [4], which attempts to reconstruct the malformation by lateral separation of the larynx, trachea, and esophagus. Endoscopic surgery to reconstruct the tracheoesophageal septum has been reported in the field of laryngology but is largely unknown among pediatric surgeons. In endoscopic surgery, the cephalic end of the cleft can be formed to closely resemble the shape of a normal larynx as much as possible. It is also possible to simultaneously repair laryngeal stenosis, which can cause extubation difficulties after repair of LTEC. Since the major advantages of endoscopic surgery also make it a meaningful option for pediatric surgeons, the present study describes our experience treating several cases of LTEC.

1. Materials and methods

Endoscopic repair of LTEC was performed in 7 patients between 2009 and 2014. LTEC was classified as type I in 5 patients and types II and IV in 1 patient each. The patients were 3 boys and 4 girls, aged 4 months to 2 years 10 months (mean age: 11 months). Mean body weight at surgery was 7.23 kg (range, 3.85–12.24 kg). (Table 1) Of the patients with type I LTEC, 2 presented with stridor and aspiration, and 2 had stridor and suckling disorder. These 4 patients had not needed intubation or tracheostomy preoperatively. They were receiving tube feeding via a nasogastric tube because of their difficulty to ingest food orally and were referred to us for surgery (Table 1; cases 3, 4, 5, 7). Gastroesophageal reflux was observed in 2 of them, and we started antireflux medication preoperatively (Table 1; cases 3, 7). The remaining patient with type I LTEC had tracheomalacia as the associated anomaly. The only symptom of the patient was stridor, so she was under follow-up observation. Her tracheomalacia worsened after an infection, and then external stenting was performed. However, following surgery, she developed inspiratory obstruction that made extubation difficult, and repair of LTEC was performed (Table 1; case 1). The patient with type II LTEC had difficulty taking food orally because of stridor and aspiration and was, therefore, administered tube feeding via a nasogastric tube. He had gastroesophageal reflux and took antireflux medication. Given the significant nature of the stridor, flexible airway endoscopy was performed, but his breathing deteriorated during the examination. Temporary intubation or tracheostomy was not performed. Therefore, he was referred to us for surgery

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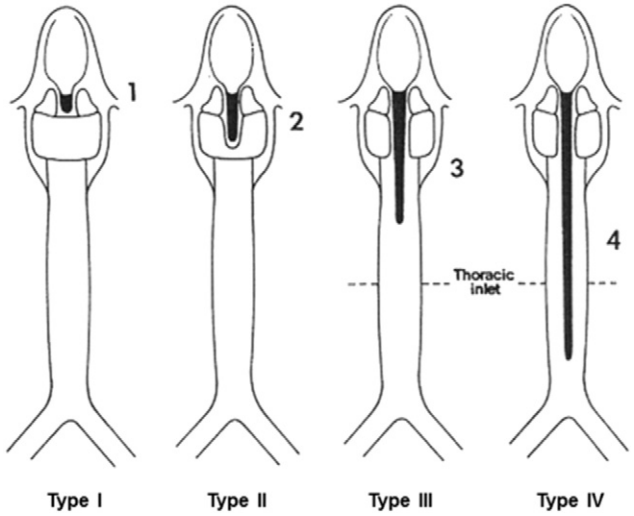

Fig. 1. Benjamin and Ingles' classification.

(Table 1; case 2). As previously reported [5], the patient with type IV LTEC received ligation of the abdominal esophagus and gastrostomy to allow enteral feeding while preventing gastroesophageal reflux. The repair of septum was performed using the lateral approach, after a tracheostomy to secure the airway. At that time, the cleft was left unrepaired from the third tracheal ring upwards to preserve laryngeal function. The remaining cleft, which level was type III on this occasion, was repaired endoscopically (Table 1; case 6). Postoperative outcomes were retrospectively studied in these 7 patients.

2. Surgery

Apart from the patient with type IV LTEC, all of the surgeries were done by oropharyngeal intubation with a 3-0 cuffed orotracheal tube. Using suspension laryngoscopy designed for use in the posterior glottis, we could see the caudal end of the cleft, which was then magnified using an operating microscope. First, the mucosal layer was carefully removed from the caudal end of the cleft commissure to the cranial end using cupped forceps with sharp edges. CO_2 laser, scissors or electric knife could be also used to remove the mucosal layer. The cleft was sutured using interrupted sutures (Fig. 2). After inserting the laryngoscope, the suture was basically performed with a single needle carrier using the curve of the needle. Because of the limited space, unlike laparoscopy and thoracoscopy, it is often difficult to suture using 2 pairs of forceps. The septum was securely sutured using a single layer of 4-0 PDSs suture with a 13-mm needle, with particularly close spacing at the caudal end. We tied the stitch using knot pusher and the knot was inside tracheal lumen. If the cleft was long, the process of removing the mucosal layer and suturing was repeated after reacquiring a view of the cephalic head (Fig. 3). The cephalic head was sutured to beneath the bilateral folds of the cleft. Finally, the entire larynx was observed to confirm that there were no issues with its shape. Any stenoses caused by contraction of the aryepiglottic folds were removed based on a surgical procedure used to treat laryngomalacia [6]. Apart from the single patient with type IV LTEC, extubation was performed on the third day after surgery.

Table 1
Patients and symptoms.

Case	Age (months)	Sex	Body weight (kg)	Type	Symptom
1	34	F	12.24	I	Failed extubation
2	8	M	7.8	II	Stridor, aspiration
3	7	M	6.28	I	Stridor, suckling disorder
4	11	F	8.08	I	Stridor, aspiration
5	15	F	8.31	I	Stridor, aspiration
6	4	M	4	IV	Unstable tracheostomy
7	5	F	3.85	I	Stridor, suckling disorder

3. Results

Six patients had stenosis caused by contraction of the aryepiglottic folds; in these cases, the contracted area was simultaneously incised (Table 2; cases 2–7). In 1 patient, the stenosis was observed at LTEC diagnosis and was so severe that it had to be removed prior to surgery to ensure an adequate field of view (Table 2; case 4). After the repair of LTEC, mild stenoses that pulled the cleft in the ventral direction were found in 4 patients; in these cases, a slight incision was made in the aryepiglottic folds (Table 2; cases 2, 3, 5, 7). In 1 patient, a marked bilateral imbalance caused by severe contraction in the left fold was observed after LTEC repair, and an incision was made only in the left aryepiglottic fold in order to correct the bilateral imbalance (Table 2; case 6).

One patient was found to have laryngomalacia with excess prolapsed mucosa that made extubation difficult after surgery; this mucosa was removed endoscopically [6] (Table 2; case 1). Two patients were found to have laryngomalacia with epiglottal prolapse that made extubation difficult after surgery; in these cases, the epiglottis was endoscopically sutured to the base of the tongue [6] (Table 2; cases 4, 7). One patient developed a granuloma of the larynx after surgery caused by irritation caused by the sutures. The granuloma was endoscopically removed because it was obstructing the patient's breathing (Table 2; case 7). The patient with type IV LTEC had a pinhole remaining in the caudal end of the newly-formed septum, which was closed using the anterior approach [5] (Table 2; case 6).

Extubation was successful in all 6 of the patients with types I and II LTEC. In the patient with type IV LTEC, because of the presence of tracheomalacia which was diagnosed after first LTEC repair using the lateral approach, it was difficult to remove the tube via tracheostomy. After the surgery, however, tracheostomy cannulation became more stable, and he is gradually being weaned off the ventilator. After his respiratory status had improved, the abdominal esophagus band was untied and fundoplication was performed (Table 2; case 6). All patients could be fed orally with no problems after surgery. We have had no recurrence of LTEC after endoscopic repair for 1 year and 10 months to 5 years and 5 months (mean period: 3 years and 5 months) of following-up period.

4. Discussion

Endoscopic repair of LTEC was first reported in 1979 [7]. Since then, the procedure has undergone various modifications by laryngologists [8–10], but it remains largely unknown among pediatric surgeons. In Japan, patients with LTEC are often referred to pediatric surgeons, who typically adopt the anterior approach to construct the septum directly in the cleft by making a longitudinal incision in the trachea [3], or the lateral approach to repair the cleft by lateral separation of the larynx, trachea, and esophagus [4]. In our experience, LTEC is often complicated by laryngeal stenosis triggered by contraction of the aryepiglottic folds. It depends on the degree of severity, but in severe cases, this stenosis can cause technical difficulties during extubation after repair of LTEC. The stenosis resembles the pathology of laryngomalacia involving the aryepiglottic folds, and it can be removed endoscopically [6]. The ability to remove laryngeal stenoses during repair of LTEC is one of the advantages of endoscopic surgery. In the anterior approach, difficulty in forming the cephalic end of the cleft can lead to fusion of the bilateral folds, malalignment of the vocal cords, and hoarseness of the voice after surgery. Even in the lateral approach, forming the cephalic end involves the risk of nerve damage. Endoscopic surgery provides a one-way view from the cephalic aspect that allows the surgeon to form a normal larynx with only minimal risk of the above complications.