Laser Assisted Double-Layer Endoscopic Repair of Laryngeal Clefts: Our Experience in 11 Cases

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Abstract Laryngeal cleft is a rare congenital malformation of the respiratory tract leading to a high level of morbidity and mortality, recently being diagnosed with increased frequency. Management throughout the years included medical and surgical. The open surgical technique is more commonly used although it has higher risk and need longer post-operative care. Recently surgical endoscopic repair was introduced using different techniques. To evaluate the clinical features of infants and children presenting with laryngeal clefts, and review endoscopic management modality especially the technique and results of repair using double-layer (2 layers) technique. Outlining our experience with the surgical technique, complications arising from surgery as well as surgical outcome by evaluating the decannulation rate and cessation of the assisted feeding by the end of treatment. Retrospective case series study. Review of infant and children seen in our clinic with the diagnosis of laryngeal cleft from January 2012 till June 2014., and treated by CO₂ laser assisted double-layer endoscopic closure. The presenting symptoms, patient demographics, diagnostic procedure, cleft type, surgical outcome and complications all were evaluated. We revised the case notes of 34 patients with a workable diagnosis of Laryngeal cleft. A total of 11 patients met our criteria and were included in the study. Clefts typing was according to modified Benjamin–Ingles

classification, type 1 (n = 9) and type 2 (n = 2). All clefts were closed endoscopically by CO₂ laser incision of the mucosa and two-layer endoscopic closure of the cleft without postoperative intubation or tracheotomy. They accepted oral feeding within 5 postoperative days (range 3–11 days). 5 out of 6 patients (83.3%) successfully stopped nasogastric feeding and 1 out of 2 patients successfully weaned from nasal cannula. One patient deceased few months after surgery due to other medical problems. Two patients needed redo after burst of the upper cleft stitches immediately after repair by iatrogenic cause during suctioning. The repair was stable in all patients during the second look 6 weeks after surgery with no recurrence or fistula formed. There were no clinically significant observed complications with this technique. After a mean follow up of 24 months, all children have a good voice, have no sign of residual aspiration, and less hospital admissions. Laryngeal cleft should be suspected in children presenting with recurrent pneumonia and difficulties during feeding. Endoscopic repair is a successful and safe technique for treating laryngeal clefts, and has short post-operative recovery without postoperative intubation or tracheotomy. Using the double-layer technique appeared to be promising but needs more comparative studies in the future.

Keywords Laryngeal cleft · Laryngotracheoesophageal clefts · Endoscopic · CO₂ laser

Introduction

Laryngotracheal cleft is a rare congenital malformation of the respiratory tract. First description was made by Richter [1]. The incidence of posterior laryngeal cleft in children

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examined by direct laryngo-bronchoscopy (DLB) has recently been reported between 6.2 and 7.6% [2, 3]. The significant increase in the incidence of this rare anomaly can be explained by increased awareness and higher suspicion among the referring physicians as well as the new evolution in the diagnostic tools [4]. The first successful surgical repair was made by Pettersson [5] while, the first endoscopic closure of laryngeal cleft described by Yamashita et al. [6]. In spite of only few institutions have extensive experience in treating this condition; the therapeutic surgical techniques have significantly improved over the last few years.

Laryngotracheal clefts have been described based on the distal extent of the cleft, which adds several dimensions to the selection of the exact treatment modality. The most widely used system is that of Benjamin and Inglis [7] which includes four cleft types. In 2006, Sandu and Monnier modified this classification into four types with subdivisions for types III and IV. Type IIIa has posterior cleft extending down to the inferior border of the cricoid plate and type IIIb extends into the cervical trachea. Type IVa, laryngotracheal cleft involves intrathoracic trachea up to the carina; while type IVb, extends to one of the main bronchi [8].

For low grades, type I and type II clefts; the endoscopic closure is considered the primary procedure with a high surgical success rate. The endoscopic management has recently been suggested for treating higher grade clefts [4, 8]. Two endoscopic techniques are currently used, the microflaps approximation with or without laser assistance [9] and laser assisted double-layer microflaps endoscopic closure [8]. The second technique was described by Sandu and Monnier [8] with only four cases. In Leishman et al. [4] presented this technique in 17 cases with successful results.

In this article, we present our experience with eleven cases of type I and II laryngotracheal clefts treated exclusively by CO₂ laser assisted double-layer endoscopic repair without postoperative intubation or tracheotomy. We outline the surgical technique, complications arising from surgery as well as surgical outcome by evaluating the decannulation rate and cessation of the assisted feeding by the end of treatment.

Methods

Patients

This is a retrospective review of 11 pediatric patients who underwent CO₂ laser assisted double-layer endoscopic laryngeal cleft repair at King Abdulaziz Medical City (KAMC) in Riyadh-Saudi Arabia from January 2012 till June 2014. Only children who were treated by this

technique were included. Children who underwent conservative treatment or primary open surgical management of laryngeal cleft were excluded even if the endoscopic tools were used as secondary management. Charts were reviewed for age, gender, history of prematurity, significant medical comorbidities, descriptions of laryngeal cleft types and timing of interventions, feeding status and airway condition at last available follow up visit. The diagnosis was made based on; presenting symptoms, modified barium swallow (MBS) and direct laryngo-bronchoscopy (DLB) results. Modified Benjamin–Inglis classification of laryngo-tracheo-esophageal clefts was used to standardize all descriptions of laryngeal cleft types. All patients had trail of conservative treatment but failed.

Statistical Analysis

All data were analyzed by SPSS version 17, using mean $\pm$ SD for quantitative variables and frequencies and percent for qualitative variables. McNemar's test was used for statistical analysis of qualitative data before and after endoscopic repair of laryngeal cleft. Differences were considered significant when the *p* value was <0.05 and highly significant when the *p* value was <0.01 .

Surgical Technique

All patients were surgically treated under general anesthesia with spontaneous respiration in suspension micro-laryngoscopy. We introduced a Parson (Storz) laryngoscope into the cleft to spread apart its lateral borders and gain access to its lower end. Then, with the CO₂ laser (Coherent Ultrapulse) in the mode (250 microspot, 150 mJ/cm², 10 Hz), a sharp incision of the mucosa dividing the lateral borders of the cleft into double layer microflaps was made from the lower end up to the level of the cuneiform cartilages. With the same CO₂ laser parameters, a very precise; 3–5-mm deep incision of the mucosa could be created to obtain an adequate separation of the tracheal and pharyngoesophageal mucosal layers. Especially at the lower end of the cleft, this allowed optimal placement of the stitches to avoid the risk of a recurrent fistula. Closure of the cleft was done in two layers. First, the tracheolaryngeal closure was performed in the distal to proximal direction by placing inverted stitches through the mucosa of the posterior membranous trachea with the knots tied on the pharyngoesophageal side of the cleft repair.

Sutures were tied to obtain meticulous mucosal contact with the cut surfaces facing inward. The second layer of suturing was placed in a similar fashion in the distal to the proximal direction with the knots tied on the mucosal surface of the pharyngoesophageal repair. The upper limit

of the cleft repair was at the level of the cuneiform cartilages in all cases. Injection of gelfoam and fibrin glue (TISSEEL) helped in proper separation of the two flaps and reduced the incidence of recurrent fistula (Fig. 1).

Positive inspiratory and expiratory pressure was supplied by a biphasic positive airway pressure (BPAP) system installed immediately during the wakeup phase after the anesthesia. Patients transferred to the intensive care unit for surveillance. A small and soft nasogastric tube was routinely placed at the end of the procedure. Antibiotics were given routinely, whereas corticosteroids were only be used if a slight laryngeal obstruction resulting from postoperative edema observed. Oral feeding resumed within 5 postoperative days. MBS and DLB to check stability of the repair and absence of recurrent fistula were routinely done within six weeks of repair (Fig. 2).

Results

We revised the case notes of 34 patients with a workable diagnosis of Laryngeal cleft. Twenty-three patients were excluded because they underwent conservative or open surgery as a primary treatment modality. The demographics of the remaining 11 patients were as follows (Table 1): 6 (54.5%) boys and 5 (45.5%) girls; the mean age at presentation was 14 months ranged from 3 months to 3 years. The main symptoms at referral were recurrent pneumonia in 10 patients (90.9%), difficulties during feeding in 9 patients (81.8%) and stridor in 3 patients (27.2%).

Gastroesophageal reflux disease (GERD) was confirmed in 10 patients (90.9%). The associated other medical comorbid conditions were found in 5 patients as cardiac anomalies (45.5%); 3 patients (27.2%) had trisomy 21 and 3 other patients (27.2%) with neurological disorders.

Regarding pre-op feeding assistance were found in 9 patients (81.8%) where 6 patients depended on nasogastric tube for feeding and 3 patients on gastrostomy tube. Regarding the ventilation support; 8 out of 11 patients (72.7%) did not need any ventilatory support prior to surgery. The extension of clefts in our patients according to modified Benjamin–Ingilis classification was type I in 9 patients and type II in two patients.

The mean follow-up was 11 months range from 6 to 18 months. We considered the management is successful when the patient is completely asymptomatic with no frequent episodes of chest infection and normally breathing and feeding without any assistance. We achieved that in 8 patients (72.7%) (p value <0.01).

Regarding the post-op results feeding assistance, 5 out of 6 patients (83.3%) successfully stopped nasogastric feeding and 1 out of 2 patients successfully weaned from nasal cannula after the dramatic improvement happened to his lung status. Patient on tracheostomy remained on tracheostomy tube planning to decannulate in near future. One patient deceased few months after surgery due to other medical problems (Table 2).

In our series, 6 patients of type I (66.6%) and the two patients of type II (100%) underwent postoperative swallow studies, which revealed no aspiration in 6 weeks post

Fig. 1 Surgical technique steps: **a** CO₂ laser incision and the inverted stitches holding the laryngeal flap, **b** pharyngeal flap stitches in place and gelfoam insertion, **c** gelfoam in place between laryngeal flap and pharyngeal flap and **d** tissue glue reinforces healing and stability of the repair

Fig. 2 Post operative, **a** DLB view and **b** check for repair stability

Table 1 Pre operative patient demographic

Age/sex	Cleft type	Symptom	Comorbidity	Surgical history	Feeding	Ventilation
22 m/M	1	Aspiration Rec. pneumonia	BA GERD	None	NGT	RA
10 m/F	1	Stridor Rec. pneumonia	Hydrocephalus Seizure GERD	VP shunt Fundoplication Gastrostomy	GT	RA
3 year/F	1	Aspiration Rec. pneumonia	Trisomy 21 BA GERD AVSD	None	Oral	RA
1 year/F	1	Stridor Rec. pneumonia	Seizure Encephalopathy BA GERD	Fundoplication Gastrostomy	GT	RA
15 m/F	1	Aspiration Rec. pneumonia	Trisomy 21 ASD VSD PDA GERD	PDA ligation	NGT	Nasal cannula
5 m/F	1	Rec. pneumonia	Trisomy 21 GERD		NGT	RA
6 m/M	1	Aspiration Rec. pneumonia	PDA Hypotonia		NGT	RA
3 year/M	1	Rec. pneumonia	CHD GERD	Tracheostomy	Oral	Trach
3 m/M	2	Stridor Cyanosis	Preterm GERD Pulmo-stenosis		NGT	RA
8 m/M	2	Rec. pneumonia	GERD Bronchopulmonary dysplasia Cong. CMV	Fundoplication Gastrostomy	GT	Nasal cannula
6 m/M	1	Rec. pneumonia	GERD	None	NGT	RA

BA Bronchial asthma, GERD Gastroesophageal reflux disease, AVSD Aterioventricular septal defect, PDA Patent ductus arteriosus, CHD congenital heart disease, NGT Nasogastric tube, GT Gastrostomy tube, RA Room air)

op. However, when accounting for the need for hospitalization, we note an overall decrease in the number of hospital admissions by (60.5%) and two patients with long admission were successfully discharged home (Table 3). Two patients needed redo after burst of the upper cleft

stitches immediately after repair by iatrogenic cause during suctioning. The repair was stable in all patients (100%) during the second look 6 weeks after surgery with no recurrence or fistula formed. There were no clinically significant observed complications with this technique.

Table 2 Post operative patients demographics

No.	Surgery	Complication	Reoperation untill complete closure	Feeding	Ventilation	Follow up
1	Endoscopic	–	–	Oral	RA	17 month
2	Endoscopic	–	–	GT	RA	18 month
3	Endoscopic	Small gap	1	Oral	RA	14 month
4	Endoscopic	–	–	GT	RA	13 month
5	Endoscopic	–	–	Oral	RA	12 month
6	Endoscopic	–	–	Oral	RA	12 month
7	Endoscopic	–	–	NGT	RA	12 month
8	Endoscopic	Short epiglottic fold Intra arytenoid fibrous band	1	Oral	Trach	12 month
9	Endoscopic	–	–	Oral	RA	13 month
10	Endoscopic	–	–	GT	Nasal cannula	6 months
11	Endoscopic	–	–	Oral	RA	6 months

Table 3 Numbers of hospital admissions Pre and Post operative

No.	Pre surgery	Post surgery
1	4	2
2	Long admission	3
3	4	1
4	5	3
5	9	4
6	4	2
7	3	0
8	5	2
9	Long admission	1
10	Long admission	Deceased
11	4	1

Discussion

Laryngeal cleft is a rare congenital abnormality leading to a high level of morbidity and mortality. This is often due to the presence of associated malformations and syndromes [4]. Because it is rare condition, no single institution has sufficient experience to propose a general algorithm for treatment. There is controversy with regard to the proper method for evaluation, diagnosis, and management of laryngeal cleft. These controversies includes: lack of standardized criteria for the evaluation and diagnosis, role of MBS and fiberoptic endoscopic evaluation of swallowing (FEES), universally accepted grading system, the role of feeding therapy and its efficacy and the timing and type of surgical intervention [10].

The management of type I and type II clefts remains controversial. Some studies advocate surgical repair, whereas others advocate medical management. Parsons et al. [3] reported 41 patients with type I laryngeal cleft that

were managed conservatively, while, Evans [11] reported the need for surgical intervention in 50% (13/26) of type I laryngeal cleft patients. Watters and Russel [12] reported that 75% of patients (9/12) had persistence of symptoms that failed to resolve by conservative therapy and required surgical intervention. Chien et al. [2] reported the need for surgical intervention in 80% (16/20) of their patients. It is important to realize that not all patients with type I and type II laryngeal clefts are the same. One can easily monitor a patient with intermittent cough and minimal pulmonary manifestation. However, a patient who has recurrent pneumonia and worsening of pulmonary status despite feeding therapy and thickened feeds should be considered for surgical repair. Our surgical indication is based on a combination of factors including: clinically apparent aspiration with feeding, severity of pulmonary status, imaging studies, presence or absence of other significant comorbid conditions predisposing to aspiration, findings on upper aerodigestive endoscopy, and poor response to medical management and feeding therapy.

Many reports appeared regarding the endoscopic repair of laryngeal cleft after the first report of Yamashita et al. [6]. Rahbar et al. [9] reported a series of 6 patients with type I to type III clefts who underwent endoscopic closure with no complications. Kubba et al. [13] also, presented 35 patients with type I to III clefts which were closed endoscopically. During recent years different techniques were suggested to improve the success rate and decrease post-operative complications. Laser assisted double-layer technique was first described in 2006 by Sandu and Monnier with only four cases with good results, followed by Leishman et al. [4] successfully treated 17 patients up to type IIIa laryngeal clefts with technique. Success rate of endoscopic closure for type I and type II has been generally considered between 81 and 94% [14]. In our case series the repair was stable in all patients 100% during the second

look 6 weeks after surgery but the complete resolution of symptoms achieved in 73%.

Conclusion

Laryngeal cleft should be suspected in all children present with persistent areodigestive tract symptoms especially when present with recurrent pneumonia or feeding difficulties. Surgical treatment in refractory cases allows cessation of feeding assistance and decrease number of hospital admissions. Endoscopic laser assisted double layer repair is a stable, successful and safe technique for repairing laryngeal clefts and could be used in the future in more advanced types of laryngeal clefts.

Compliance with Ethical Standards

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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