Indian J Otolaryngol Head Neck Surg
(July–Sept 2022) 74(3):296–304; https://doi.org/10.1007/s12070-021-02479-y

ORIGINAL ARTICLE

Endoscopic Repair of Laryngeal Clefts: 8 Years' Experience

Ria Emmanuel[1] · Eswaran V Raman[1] · Deepa Shivnani[1]

Received: 6 October 2020 / Accepted: 19 February 2021 / Published online: 6 March 2021

Abstract To emphasize the need for high clinical suspicion in the diagnosis of Laryngeal cleft in paediatric population, to catalogue the pattern of presentation, time to treatment and the evolution of surgical techniques for Laryngeal cleft repair at our center. A retrospective review of laryngeal cleft cases which presented over a period of 8 years (May 2012–May 2020), from a tertiary care center, was done. Data includes—patient demographics, preliminary investigations, diagnostic methods, type of cleft, surgical steps and post-operative follow up. Extensive literature search was done and we could not find similar studies from South East Asia and the Indian subcontinents. Of the 10 patients 7 were managed surgically and 3 conservatively. There was an equal distribution of type 1 (n = 5) and 2 (n = 5) clefts. 80% cases were males and 9 out of 10 patients had associated congenital anomalies. 80% cases had symptom resolution (75% were managed surgically and 25% managed medically). Surgical intervention should be based on the extent of anatomical defect and the functional impairment caused by cleft such as respiratory problems, persistence of feeding issues despite maximal medical management and feeding therapy. Early surgical management of type I and II clefts have satisfactory outcomes.

Keywords Endoscopic repair · Laryngeal cleft · Management · Type 1 · Type 2

Abbreviations

LC	Laryngeal cleft
FEES	Fiber-optic endoscopic evaluation of swallowing
TE	Tracheo esophageal
TEF	Tracheaesophageal fistula
MBS	Modified barium swallow
DL	Direct laryngoscopy
GA	General anesthesia
OGT	Orogastric tube
LRTI	Lower respiratory tract infection
yrs	Years
IA	Inter arytenoid
pre op	Preoperative
post op	Post operative
CXR	Chest X ray
ETT	Endotracheal tube
PDS	Polydioxanone suture
POD	Postoperative day
TORS	Trans oral robotic surgery
fig	Figure
PICU	Paediatric intensive care unit

✉ Ria Emmanuel
riaishere@gmail.com

Eswaran V Raman
evraman26@gmail.com

Deepa Shivnani
deepa.shivnani14@gmail.com

[1] Department of Otorhinolaryngology and Head and Neck Surgery, Children's Airway and Swallowing Centre, Manipal Hospitals, 98, HAL Airport Road, Bengaluru, India

Introduction

Laryngeal cleft is an under diagnosed pathology with a very low incidence of 1 in 10,000 to 20,000. It's a rare condition which is often not looked for, with non-specific symptoms. LCs are abnormal posterior communications between the laryngotracheal complex and oesophagus. It

was first reported by Ritcher in 1792. Symptomatic laryngeal clefts account for 0.3% to 0.5% of all congenital anomalies of larynx [1–3]. However there has been a recent increase in incidence of LC from 1990 onwards, probably due to better understanding of the disease and high levels of suspicion. The incidence has gone up from 0.1 to 0.47% in initial reports to 6.2–7.6% in recent years [4–6]. Some studies show that type 1 LC is the most commonly diagnosed, but as these patients may be asymptomatic there could be a potential undiagnosed population of LC too [6].

The most common classification system used is the Benjamin Inglis classification [7]—which divides clefts into 4 types.

- Type 1—Supraglottic inter arytenoid cleft located above vocal cord level
- Type 2—Cleft extends below the level of vocal cords into the upper cricoid cartilage
- Type 3—Cleft extends through the cricoid cartilage possibly in to the cervical trachea
- Type 4—Cleft extends into the thoracic trachea and extends towards the carina.

Moungthong and Holinger have added to the Benjamin/Inglis classification a distinct group of occult, submucous clefts of the cricoid cartilage. (Type 0) [8]. This was initially described by Tucker and Madadalozzo [2].

Benjamin Inglis classification was modified or subdivided by Monnier and Sandu [7, 9] for the purpose of endoscopic repair of clefts into

- Type 3a—Complete cleft of the cricoid plate
- Type 3b—Cleft extending down to the level of the sternal notch but not further down into the intrathoracic portion of the trachea
- Type 4a—Cleft extending into the intrathoracic trachea to the carina and
- Type 4b—Intrathoracic extension of the cleft involving one main bronchus.

When cleft approaches but does not reach the level of true vocal cord it is diagnosed as a 'deep interarytenoid groove' (IA height above the cricoid cartilage < 3 mm but remains above true cord) [10].

Most LCs occur sporadically, though autosomal dominant inheritance patterns have been observed. Well recognised maternal risk factors are—substance abuse, premature delivery and polyhydramnios [6].

Clefts develop due to lack of separation of the laryngo tracheal axis and the esophagus. A posterior laryngeal cleft is formed due to failure of development of the IA muscle (submucous cleft) with or without absence of IA mucosa (type 1 LC). Incomplete formation of cricoid cartilage forms a type 2 cleft and incomplete formation of TE septum distal to cricoid leads to type 3 or 4 clefts. The IA muscle and cricoid cartilage are derivatives of the 6th branchial arch and the above anomalies occur between 5 and 6th week of embryonic period [2, 11].

Type 1 and 2 clefts have a heterogenous presentation and the management is controversial [12]. They range in severity from being asymptomatic throughout life (type 1) to being incompatible with life (type 4) [1]. The high level of morbidity of LC is usually due to its association with syndromes like Opitz-Frias, Pallister Hall, VACTERL and CHARGE syndromes. As these cases are often associated with other congenital anomalies (60%) it is while managing those issues that the clefts often get picked up [2].

To our knowledge there are no similar studies from South Asia. We present a series of 10 cases of LCs which were managed at our center and all the 10 children were of Indian origin. Each child had a unique presentation and the duration of diagnosis to surgery also varied. This would be the first of its kind, describing the clinical presentation and management of LC cases, from this subcontinent.

Materials and Methods

After obtaining the institutional review board approval, the details of patients, all those who were diagnosed as laryngeal clefts by the senior author, were collected by reviewing the medical records (maintaining patient confidentiality) from May 2012 to May 2020. The medical records were carefully evaluated for the patient demographics, presentation, time to treatment, method of diagnosis and treatment, follow up and outcome.

The protocol followed for suspected LC cases managed at our centre is given below.

Every patient underwent detailed history taking and clinical examination. The preliminary investigations included CXR, Fiber-optic laryngoscopy and swallow assessment. The treatment options were discussed with the family and further treatment plan was made by a multidisciplinary team involving pediatric otorhinolaryngologist, paediatrician, pediatric airway anesthesiologist and speech and language pathologist. All the children except the ones where the cleft was an incidental finding, were put on a trial of conservative therapy prior to the surgery. Conservative therapy included anti-reflux treatment (Lansoprazole) and feeding therapy (adjustments in liquid viscosity, solid consistency, feeding equipment strategies and positioning). All the kids with comorbid conditions were started on or were already on medical/surgical treatment for the same. Written and informed consent was taken from parents of the children who underwent surgical procedures.

General anesthesia was given by the tubeless technique of anesthesia which gives the full view of laryngeal inlet. 2 of our patients were already on tracheostomy at the time of

surgery. In the non-tracheostomized patients, the ETT was passed either nasally or orally and the tip of the endotracheal tube was placed just above the glottis. Induction was achieved through inhalation of sevoflurane in oxygen or intravenous administration of propofol to induce unconsciousness while maintaining spontaneous respiration. During the procedure propofol was used to maintain the anesthetic plane. The vocal cords were also anesthetized using topical lignocaine (4%).

Patient was positioned on the table (sniffing position, with 35 degree neck flexion and 15 degree head extension) and a silicone teeth protector inserted. An age appropriate Benjamin-Lindholm scope and a 0° Hopkins rod lens 4 mm telescope was used for visualization. The laryngoscope was fixed to the laryngoscope holder and chest support and placed on a support table, such that the inter arytenoid region is in the center of the field. The clefts were classified according to the Benjamin Inglis classification. Diagnosis was based on the simple but most important step of palpating the interarytenoid region using an 'L' shaped probe/nerve hook (Fig. 1).

The mucosal margin of the cleft was splayed using the Lindholm vocal cord retractor also called laryngeal spreader (Fig. 2). The surgery was done using a microscope and the endoscope. Robot assistance was used in one case. C02 laser was delivered either using a micromanipulator or laser fiber. While using the endoscope, we used a 3 handed technique, during which the scope was held by an assistant, so the primary surgeon could use both hands for surgery. The inter-arytenoid mucosa was denuded either using CO2 laser (4–6 watts, 1.0 mm spot size, accupulse mode) or cold steel instruments. The edges were approximated using single or double layer suturing. Suture materials used were 5–0 or 6–0 vicryl and 5–0 PDS. The repaired (Fig. 3) site was reinforced using fibrin glue in certain cases. The surgical procedure also included aryepiglottoplasty, wherever found necessary.

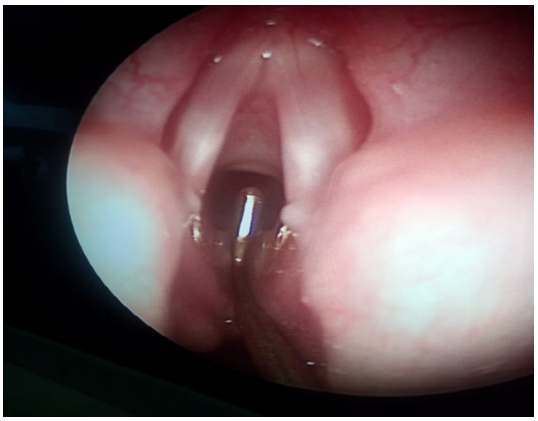

Fig. 1 Cleft identification using "L" shaped probe

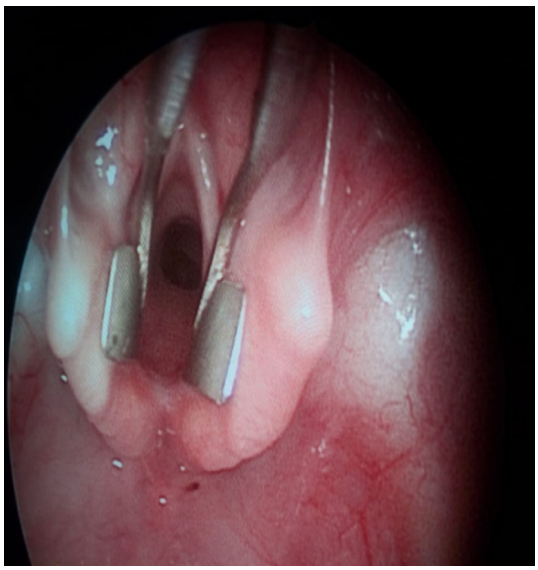

Fig. 2 Cleft exposed using laryngeal spreader

Post operatively all the patients were monitored in the PICU and were put on short course of injectable corticosteroids and nebulisations. Trial of semisolids and thickened liquids were given between 3rd and 5th post op days. All of them were called for a swallow assessment after 4–5 weeks post procedure and swallow therapy was modified accordingly.

Results and Analysis

A total of 10 LC cases were included in the series. 8/10 (80%) patients were males and there was an equal distribution of type 1 (n = 5) and 2 (n = 5) clefts. The age at diagnosis ranged from 1 to 27 months. The mean age, at

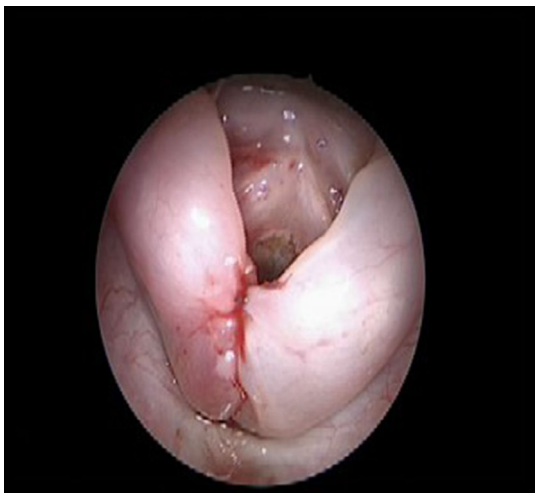

Fig. 3 Post cleft repair

diagnosis was 14.1 months and at surgery was 18.3 months (6 months to 29 months). The mean time span between diagnosis to surgery was 3.5 months (0.75 months to 6 months). The most common presenting symptom was feeding difficulty especially to liquids. 90% cases had associated congenital anomalies, most common of which was laryngomalacia (n = 5) followed by TEF (n = 2) and anorectal malformation (n = 1). Patient selection for surgery was based on severity of symptoms, outcome of swallow studies and failure of trial of conservative management, mean duration of trial was 3.27 months (0.75 to 6 months). Suture repair of cleft was done in 7 cases, a second surgery was required in 2 of these cases. The suturing was done using single layer in 6 cases and double layer in 1 case. Fibrin glue was used in reinforcing the sutured area in 3 cases.4 out of the 7 operated cases underwent bilateral aryepiglottoplasty along with the cleft repair and all except one patient was extubated immediately after surgery. 2 out of the 3 non operated cases were type 1 clefts which were managed conservatively, were in the cleft was an incidental finding whereas the 3rd case, a type 2 cleft was planned for surgery, but the child succumbed to central hypoxia. Two of our cases were already tracheostomized. The mean duration of follow up was 3.9 months (1 to 8 months). Clinical improvement was mostly based on parental judgement and weight gain with or without postoperative FEES/MBS study. Of all the patients who had clinical improvement (80%), 75% (n = 6) were surgically managed and 25% (n = 2) conservatively managed (Tables 1, 2).

Discussion

Although a rare disease, LCs are frequently reported since 1990 as a result of enhanced endoscopic techniques and advanced neonatal care facilities [6]. The low incidence prior to this period could be due the following reasons: minor clefts may be asymptomatic, the endoscopic diagnosis is often difficult and the lesion may be easily missed and severe clefts may lead to deaths, before a diagnosis can be made [2].

The severity of symptoms of LC correlates with type of cleft and the associated anomalies. Patients with high grade clefts (LC 3,4) may present at birth which requires urgent intervention, while diagnosis of low grade clefts (LC 1,2) is delayed due to non-specific symptoms [13]. In a series by Andrieu-Guitrancourt and colleagues, Type 0 clefts were diagnosed after the age of 6 months, type 1 before the age of 6 months, and type 2 before age of 2 months [14]. In our study the 2 cases which were diagnosed before 2 months were LC type2 and there was an equal distribution of type 1 (n = 5) and 2 (n = 3) in cases diagnosed after 6 months. In our series the mean age at diagnosis was 14.1 months which is lower than the mean age in the series by Rhabar and colleagues (21 months) and the series by Parsons and Herr (2 years 10 months) [2].

LCs are more common in boys than girls with a male to female ratio of 5:3 [11], and in our study the ratio was 8:2.

LCs are associated with congenital abnormalities in 58% to 68% of cases and are most commonly associated with gastrointestinal anomalies [3, 15]. Thirty percent (3/10) of our cases had associated digestive problems TEF (n = 2), esophageal and duodenal atresia (n = 1) which goes with the estimated prevalence 16–67% of digestive problems in LCs patients. Some other common associations are genitourinary (14–44%), cardiac (16–33%), craniofacial 5–15%, tracheal and pulmonary 2–9% [2]. In a series by Myer and colleagues 21% of clefts were diagnosed by accident, which is similar to our finding [16]. In 20% of our series, cleft was an incidental finding.

LC should be considered in the differential diagnosis of children who present with swallow issues (50%), breathing difficulty (43%) and recurrent pneumonia or respiratory distress at birth (37%) [2].

Most common investigations used for the work up in suspected patients include CXR, FEES and MBS. CXR provides evidence of parenchymal changes associated with aspiration. Penetration of fluid bolus into the interarytenoid area is easily picked up in FEES study. It gives a direct view of the larynx as well as the changes which occur after applying swallow maneuvers. It is superior to MBS as it doesn't subject patients to radiation. Barium swallow, though it helps us to evaluate the global swallowing coordination—oral, pharyngeal, oesophageal, gastric phases, sometimes immediate passage of contrast into trachea and may lead to false diagnosis of TEF. However, advantages of MBS include the lack of white out and it doesn't require patient co-operation. However FEES and MBS can be normal in patients with intermittent aspiration [11]. Though flexible endoscopy may help us identify a cleft, definitive diagnosis can be made only by evaluation of the patient under GA. Careful palpation of the IA area using a hook is of paramount importance, as LC may be obscured by redundant, laryngeal/esophageal mucosa protruding into it. Once cleft is diagnosed a thorough systemic evaluation should be done to identify associated anomalies [10, 17].

While doing a flexible bronchoscopy mucosal edema and cobblestoning of the mucosa are commonly found in these patients. Tracheal/bronchial secretions can be measured for Lipid laden macrophage index and neutrophil percentage which are markers for inflammatory response in the tracheobronchial tree [5].

Controversy exists in the management of type 1 and 2 clefts and the age to consider surgical repair, however there are studies that show satisfactory outcomes in the early

Table 1 Pre operative patient details

Sl no	Age at Dx in months	Sex	Symptoms at presentation	Medical history	Surgical history at time of presentation	Swallow studies and its outcome	Flexible bronchoscopy findings	Duration of trial of conservative Mx (in months)
1	24	Female	Difficulty in swallowing liquids, nasal regurgitation, recurrent chest infections, delay in speech	Anomalous left coronary artery from pulmonary artery in ECHO	Nil	MBS—aspiration present	Suspected laryngeal cleft	3
2	25	Male	Noisy breathing, feeding difficulties	Tongue tie, Laryngomalacia	Nil	FEES—aspiration to liquids	Laryngomalacia Suspected laryngeal cleft	6
3	13	Male	Bluish discolouration during feeding, nasal regurgitation	TEF, high arched palate, anorectal malformation with imperforate anus, hypospadiasis, chordee, congenital hypothyroidism, cardiac anomaly with bicuspid aortic valve	Colostomy, Gastrostomy	FEES—aspiration to liquids, MBS—no e/o TEF,GERD—grade 3	Suspected laryngeal cleft	6
4	5	Male	Choking while feeding	Tachypnoea and NICU stay for 20 days after birth, pneumothorax. BOOP	ICD insertion, Pleurodesis	MBS—aspiration present	Laryngomalacia-swollen AE folds, suspected laryngeal cleft	0.75
5	8	Female	Cough and vomiting while feeding, noisy breathing	Failure to thrive, GERD with feeding difficulty, PFO with L-R shunt Severe pneumonia, Vit D deficiency	Nil	MILK SCAN-no significant reflux	Floppy epiglottis blocking laryngeal inlet, omega shaped epiglottis, suspected IA cleft	3
6	24	Male	Noisy breathing with suprasternal and intercostal retractions	H/o Road traffic accident requiring ICU stay and intubation for 10 days	Nil	Nil	Subglottic stenosis Grade 3	Nil
7	11	Male	Recurrent chest infections, cough while drinking liquids	Repeated hospital admissions for wheeze/LRTI/Pneumonia	Nil	FEES—aspiration to liquids, MBS—no e/o TEF/GERD	Laryngomalacia Suspected laryngeal cleft	2
8	24	Male	Referred for Mx of SGS	Esophageal atresia, duodenal atresia, SGS—grade3 hypopharyngeal stenosis, hypotonia, global developmental delay	Cervical esophagectomy with trans anastomotic tube, Gastrostomy, Tracheostomy, Balloon dilatation of subglottic stenosis 5 times	Nil	Suspected laryngeal cleft	6

Table 1 continued

Sl no	Age at Dx in months	Sex	Symptoms at presentation	Medical history	Surgical history at time of presentation	Swallow studies and its outcome	Flexible bronchoscopy findings	Duration of trial of conservative Mx (in months)
9	2	Male	Referred for Mx of Laryngeal cleft	pre term baby, had inpsiratory stridor since birth, laryngotracheobronchomalacia,on mechanincal ventillation, on GERD treatment	Tracheostomy done on day 9 of life, on mechanical ventilatory support	Nil	Laryngo tracheobronchomalacia@Laryngeal cleft	1
10	1	Male	Intermittent stridor, repeated desaturations on oral intake	TEF—TYPE C, ostium secondum atrial septal defect, single umbilical artery polydactyly with syndactyly	TEF and prox esophageal atresia repair, Tracheostomy, Gastrostomy	Subjective analysis—desaturation on feeding	Suspected laryngeal cleft, redundant IA mucosa	5

surgical intervention of such cases. This has been advocated in view of prevention of irreversible pulmonary damage and associated comorbidities. Trial of conservative therapy–feeding therapy and medical therapy is a must before a definitive surgery. Medical management should consist of treatment of GERD, food allergies, eosinophilic esophagitis, reactive airway disease. The mean duration between diagnosis to surgery, if less than 3 months, was defined as "early surgical intervention" by Day et al. This period was less than 3 months (2.8 months) in our study too [15, 18].

Over the years the surgical techniques for LC have evolved quite a lot and there is difference in opinion on deciding the indication for proceeding to surgical repair, assessing patient candidacy for surgery, endoscopic versus open approach, single versus double layer closure, age at surgery, duration of trial of conservative treatment prior to surgery etc. It was concluded that there was no impact of age on the safety or efficacy of surgical intervention in a study by Cole and colleagues [19]. 65% of members who joined the multi national-multi institute study on LCs did not use minimum weight as criterion for endoscopic repair of LCs [10].

In our series of surgically treated LCs (n = 7) one case was Trans oral Robotic assisted and in the rest of the 6 cases we used microscope and endoscope. TORS has many benefits over traditional endoscopic approach as it provides three dimensional visualization, increased range of instrument movement and filtration of tremor. But paediatric-sized robotic instruments and equipment are not readily available yet, the initial operation times and learning curve are longer and involve high cost [20].

Endoscopic closure of LC was first reported by Yamashita [11]. Endoscopic repair is the preferred option for type I and some minor type 2 clefts. For major type 2, type 3 a with complete vertical division of the cricoid plate, and type 3 b with extension of the cleft down to the level of the sternal notch, open surgery with anterior laryngotracheal fissure and two-layer posterior repair is usually recommended with or without graft interposition. In type 4 clefts with intrathoracic extension, a lateral approach through the tracheoesophageal groove from the intra to the extra thoracic portion of the airway is the preferred method [9].

In a series of 35 patients with LC (1–3) Kubbe et al. [12] performed endoscopic repair in 15/35 cases, this remains one of the largest series in the category and it didn't show any significant difference in the functional outcome between the two groups. Initially endoscopic repair was limited to type 1 and type II which was also extended to type 3b [13–15]. Gardedian et al. had a 100% successful rate using a double layered approach for 4 type 3b clefts [16].

Table 2 Summary of patients (surgical details included)

Sl no	Age at Sx in months	Time span BTN Dx and Sx in months	Tracheostomised at time of SX	Type of cleft	Cleft repair approach	Method of denudation of mucosa	Technique of suturing	Other findings/ procedures	Post op swallow study and its outcome	Complications	Duration of follow up in months	Outcome
1	29	2	No	2	Robot assisted enodscopic approach	Diathermy	2 Layers	Nil	Nil	Sepsis	1 M	Diseased
2	31	6	No	2	Endoscopic	CO_2 laser	Not sutured	Tongue tie release, B/L aeplasty	Salivogram-normal	Nil	6 M	Recovered
3	18	5	No	I	Microscopic	CO_2 laser	Single layer, fig of 8	Nil	FEES—normal	Nil	3 M	Gastrostomy closed
4	5.75	0.75	No	I	Endoscopic assisted	CO_2 laser	Single layer, fig of 8	Laryngomalacia	Subjective assesment—no aspiration	Nil	3 M	Recovered
5	13	2	No	2	Endoscope assisted	CO_2 laser	Single layer	B/L aeplasty	Subjective assesment—no aspiration	Nil	6 M	Recovered
6	26	4	Yes	1	Endoscope assisted	CO_2 laser	Single layer	SGS grade 3, balloon dilatation	Nil	Nil	8 M	Condition stable
7	6	5 M	Yes	2	Endoscope assisted	Cold steel instruments	Single layer, fig of 8	Nil	contrast study (through PEG) for reflux—nil reflux	Nil	3 M	Weigth gain +
8	8.75	0.75	No	1	Not repaired	Not done	Not repaired	Short AE folds, B/L AE plasty done	Subjective assesment—no aspiration	Nil	4 M	Recovered, on oral feeds
9	25	Nil	No	1	Not repaired	Not done	Not repaired	SGS grade 3 balloon dilatation	Subjective assesment—no aspitation	Nil	3 M	Recovered
10	2	Nil	Yes	2	Not repaired	Not done	Not done	Nil	Nil	Central hypoxia	2 M	Diseased

Most of the studies on clefts employ a double layer closure, but Douglas and colleagues advocate single layer closure with absorbable suture [11]. In 80% of our cases we tried the single layer closure, only 2 cases had to be sutured at a later stage. Waltzmans and Bent described anterior and posterior flaps over opposite arytenoid with an "S" shaped incision in the IA cleft, whereby the flaps were rotated to over the demucosalisied part of contralateral arytenoid, creating a double mucosal layer in IA area without overlapping sutures [21].

Most of the airway surgeons used absorbable sutures for cleft repair (PDS or VICRYL ranging from 4–0 to 7–0), while Kristine and colleagues in their series used non absorbable sutures for early surgical intervention in type 1 cleft [18]. Chiang et al. found good results using P2 needles for the surgical management of type 1 and 2 clefts. The upper limit of cleft repair was just below the cuneiform cartilage in a study by Crispin and colleagues [23]. In the multi-institutional study by Jeffrey et al., 40% of members routinely sutured the corniculate cartilage and 60% did not suture the corniculate cartilage [10]. In our series the upper limit of repair was just below the cuneiform cartilage.

Interarytenoid injection augmentation for type 1 LC have been tried in various studies and is found to be a predictor of success rate of suture augmentation [22], though none of our cases underwent this trial. This was mainly due to concerns about cost and repeated exposure to general anaesthesia.

The sutured area was reinforced using collagen glue in 3 of our patients but we didn't find a difference in wound strength. Surgical failure secondary to poor wound healing can be due to Factor 13 deficiency and concurrent GERD which is not adequately treated [5, 23].

The presence of LC enhance the supraglottic collapse and closure of LC itself can contribute to supraglottic constriction and create a situation similar to laryngomalacia. 80% of cases (LC type 1 and 2) included in study by Chiang et al. underwent concurrent supraglottoplasty along with cleft repair. In our LC series 5/7 patients underwent bilateral AE plasty using curved laryngeal scissors along with LC repair [13].

Estimated success rates of endoscopic LC repair was between 71 and 94% as per previous studies, which was also reflected in our series, 80% patients had resolution of the symptoms (75% in the surgical group). The success rates of medical management varies between 20 and 100%, 2 of our patients who were conservatively managed (type 1 LC), completely recovered [11]. Complete resolution rates in a recent meta analysis were slightly different from the above, the rates were as follows—conservative management 52.3%, injection augmentation – 69.2% and endoscopic surgery – 65.4% [6].

A recent study shows that patients with LCs can have dysfunction in all phases of swallowing (some degree of oral phase impairment-triggering and pharyngeal phase impairment such as laryngeal penetration, aspiration and silent aspiration). So the patients may still have to continue diet modification post surgical repair [24].

We found in our literature review that the terms failure, relapse, revision, fistula formation etc. have been used interchangeably, in cases requiring multiple stage surgeries. In our series 2 patients required a second stage surgery. Success of treatment was assessed if there is subjective improvement according to parental report in follow up visits with or without MBS findings. Successful decannulation, elimination of aspiration pneumonia, disappearance of clinical signs of aspiration and tolerance of oral feeding have been used to define success [13, 19].

Ours was a retrospective record based study from a tertiary care centre. Most of the patients were referred from other centres and we did not witness the initial presentation, and some of them were already on medical treatment. The sequence of occurrence of symptoms and duration of conservative treatment, in our study, was based on information obtained from parents and available medical records. Hence the accuracy of clinical history is doubtful.

Conclusion

LC in the pediatric population is a diagnosis of suspicion. The timing between presentation, diagnosis and initial treatment varies significantly among the patients. Trial of conservative management and treatment of associated anomalies should go hand in hand. The surgical intervention should be based on the anatomical defect and the functional impairment caused by cleft such as respiratory problems, persistence of feeding issues despite maximal medical management and feeding therapy. The treatment options should be discussed by a multidisciplinary team and it should be individualized for each case as there is no single option applicable to all. Such studies help clinicians to identify individual variations and clinical heterogeneity. Further research is required in understanding the natural history of LC as most of them present with other congenital anomalies.

Author Contributions All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by RE, EVR and DS. The first draft of the manuscript was written by RE and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

References

1. Chitkara AE, Tadros M, Kim HJ, Harley EH (2003) Complete laryngotracheoesophageal cleft: complicated management issues. Laryngoscope 113:1314–1320
2. Pezzettigotta SM, Leboulanger N, Roger G, Denoyelle F, Garabédian EN (2008) Laryngeal cleft. Otolaryngol Clin North Am 41(5):913–ix. https://doi.org/10.1016/j.otc.2008.04.010
3. Roth B, Rose KG, Benz-Bohm G, Gunther H (1983) Laryngotracheoesophageal cleft: clinical features, diagnosis and therapy. Eur J Pediatr 140:41–46
4. Chien W, Ashland J, Haver K, Hardy SC, Curren P, Hartnick CJ (2006) Type 1 laryngeal cleft: establishing a functional diagnostic and management algorithm. Int J Pediatr Otorhinolaryngol 70(12):2073–2079
5. Rahbar R, Chen JL, Rosen RL et al (2009) Endoscopic repair of laryngeal cleft type I and type II: When and why? Laryngoscope 119(9):1797–1802
6. Loh R, Phua M, Shaw L (2019) Diagnosis and management of type 1 laryngeal cleft: systematic review Aust J Otolaryngol 2:5
7. Benjamin B, Inglis A (1989) Minor congenital laryngeal clefts: diagnosis and classification. Ann Otol Rhinol Laryngol 98(6):417–420
8. Moungthong G, Holinger LD (1997) Laryngotracheoesophageal clefts. Ann Otol Rhinol Laryngol 106(12):1002–1011
9. Sandu K, Monnier P (2006) Endoscopic laryngotracheal cleft repair without tracheotomy or intubation. Laryngoscope 116(4):630–634. https://doi.org/10.1097/01.mlg.0000200794.78614.87
10. Yeung JC, Balakrishnan K, Cheng ATL et al (2017) International pediatric otolaryngology group: consensus guidelines on the diagnosis and management of type I laryngeal clefts. Int J Pediatr Otorhinolaryngol 101:51–56
11. Johnston DR, Watters K, Ferrari LR, Rahbar R (2014) Laryngeal cleft: evaluation and management. Int J Pediatr Otorhinolaryngol 78(6):905–911
12. Saxena RC, Scott AR (2015) The varying presentations of type I laryngeal cleft: illustrative cases in a set of triplets. Int J Pediatr Otorhinolaryngol 79(4):620–622
13. Chiang T, McConnell B, Ruiz AG, DeBoer EM, Prager JD (2014) Surgical management of type I and II laryngeal cleft in the pediatric population. Int J Pediatr Otorhinolaryngol 78(12):2244–2249
14. Andrieu-Guitrancourt J, Borde J, Mitrofanof P, Ensel J, Dehesdin D (1982) Association of posterior laryngeal cleft and atresia of the oesophagus. Two cases treated successfully (author's transl). Chir Pediatr 23(2):75–80
15. Rahbar R, Rouillon I, Roger G et al (2006) The presentation and management of laryngeal cleft: a 10-year experience. Arch Otolaryngol Head Neck Surg 132(12):1335–1341
16. Myer III CM, Cotton RT, Holmes DK et al (1990) Laryngeal and laryngotracheoesophageal clefts: role of early surgical repair. Ann Otol Rhinol Laryngol 99(2pt):98–104
17. Bowe SN, Hartnick CJ (2017) Management of type I and type II laryngeal clefts: controversies and evidence. Curr Opin Otolaryngol Head Neck Surg 25(6):506–513
18. Day KE, Smith NJ, Kulbersh BD (2016) Early surgical intervention in type I laryngeal cleft. Int J Pediatr Otorhinolaryngol 90:236–240
19. Cole E, Dreyzin A, Shaffer AD, Tobey ABJ, Chi DH, Tarchichi T (2018) Outcomes and swallowing evaluations after injection laryngoplasty for type I laryngeal cleft: Does age matter? Int J Pediatr Otorhinolaryngol 115:10–18
20. Leonardis RL, Duvvuri U, Mehta D (2014) Transoral robotic-assisted laryngeal cleft repair in the pediatric patient. Laryngoscope 124(9):2167–2169
21. Waltzman M, Bent III J (2001) Repair of type 1-A and 1-B posterior laryngeal clefts. Oper Tech Otolaryngol Head Neck Surg 12(4);252–255
22. Miller AL, Hersh CJ, Johnson KE, Hartnick CJ (2019) Short-term swallowing outcomes following type 1 laryngeal cleft injection. Int J Pediatr Otorhinolaryngol 116:159–163
23. Leishman C, Monnier P, Jaquet Y (2014) Endoscopic repair of laryngotracheoesophageal clefts: experience in 17 cases. Int J Pediatr Otorhinolaryngol 78(2):227–231
24. Strychowsky JE, Dodrill P, Moritz E, Perez J, Rahbar R (2016) Swallowing dysfunction among patients with laryngeal cleft: More than just aspiration? Int J Pediatr Otorhinolaryngol 82:38–42