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ORIGINAL ARTICLE

Outcomes of endoscopic percutaneous suture lateralization for neonatal and infantal bilateral vocal ford paralysis

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HIGHLIGHTS

- Endoscopic percutaneous suture lateralization is an effective and safe treatment for bilateral vocal fold paralysis.
- 89% of our cases were able to avoid tracheostomy without significant impact on the function of swallowing or phonation.
- The nondestructive nature and simplicity of ESPL is valuable.

KEYWORDS
Vocal fold paralysis;
Endoscopic surgical procedure;
Lateralization;
Airway;
Neonatal, infants

Abstract
Objective: Bilateral Vocal Fold Paralysis (BVFP) is a rare but significant resource of respiratory distress in neonates and infants. The objective of this study was to evaluate the efficacy and functional outcomes of Endoscopic Percutaneous Suture Lateralization (EPSL) for the treatment of BVFP in neonates and infants.
Methods: A case series study of nine patients undergoing EPSL for BVFP between January 2019 and June 2021 was conducted. All patients were candidates for tracheostomy prior to EPSL. Demographic features including gender, age at diagnosis and surgery, main symptoms, airway comorbidities, airway support, and etiology were collected preoperatively. Patients were evaluated for breathing, swallowing and phonation postoperatively. Surgical success was defined as the ability to avoid tracheostomy. Functional Endoscopic Evaluation of Swallowing (FEES) was conducted to identify aspiration. Voice evaluation was based on clinical observation.
Results: Nine patients underwent ten EPSL procedures (one in the left vocal fold, and nine in the right vocal fold). Eight patients (8/9) were able to successfully avoid tracheostomy and feed orally without aspiration after the procedure. One patient experienced clinical improvement in respiratory support requirements and underwent laparoscopic nissen and gastrostomy tube placement. At the last follow-up, two patients regained normal voice, two patients had mild dysphonia, and five patients had moderate dysphonia. Five patients showed partial return of the contralateral vocal fold function.

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Conclusion: EPSL is an effective and safe treatment for neonatal and infantal BVFP, which enables patients free from tracheostomy without significant impact on swallowing function or phonation.
Level of evidence: Level 4.

Introduction

Bilateral Vocal Fold Paralysis (BVFP) is a rare condition which can cause significant airway, swallowing and voice impairment in neonates and infants. The primary goal of treatment is to establish a patent airway for ventilation, while maintaining a safe swallow function and acceptable voice. Historically, tracheostomy has been a standard intervention to achieve this goal, particularly in infancy.[1]

Several alternative endoscopic procedures have been developed to avoid adverse effects following tracheostomy, such as arytenoidectomy and posterior cordotomy,[2–5] anterior/posterior cricoid split,[6,7] arytenoid abduction lateropexy[8,9] and suture lateralization.[10] Each procedure has its own advantages and disadvantages. The purpose of this study is to report our experience with Endoscopic Percutaneous Suture Lateralization (EPSL) for neonates and infants with BVFP, and to develop a discussion of its most important aspects and findings.

Methods

Patients

In the present study, we reviewed nine patients with BVFP who underwent EPSL at the Otolaryngology Department of Children's Hospital, Fudan University between January 2019 and June 2021. Demographic information included gender, age at diagnosis, at surgery, airway, main symptoms, comorbidities, airway support and etiology. The type and length of surgery, duration of postoperatively endotracheal intubation, and airway support after extubation were recorded. Outcome measures were airway support, aspiration, feeding status, voice, and vocal fold mobility. This study was approved by our institutional ethical committee (n° (2020) 68).

All patients were diagnosed with BVFP upon awake flexible laryngoscopy, with clinical correlation that BVFP was causing associated respiratory distress or feeding difficulty. All of them were candidates for tracheostomy prior to EPSL and underwent Microlaryngobronchoscopy (MLB) to identify airway comorbidities as well. Since the instability of airway, no patient was evaluated for the swallowing function preoperatively.

Endoscopic percutaneous suture lateralization and post-operative management

All patients underwent suspension direct laryngoscopy while spontaneously breathing under total intravenous anesthesia. A supraglottoplasty was performed if necessary to provide adequate exposure to the glottis and subglottis for EPSL. Fig. 1 shows the brief surgical procedure of EPSL, as reported by Montague in 2018.[10] First, sutures were prepared before surgery. Briefly, a 4–0 Prolene suture (suture #1) was loaded into an 18-gauge needle and secured with a 1-cc stub-tip syringe with the tip of the suture just inside the bevel of the needle. A second 4–0 Prolene suture loop (suture #2) was loaded into an 18-gauge needle, and also secured with a 1-cc stub-tip syringe. The silastic button was fashioned and soaked in betadine on the sterile field. Next, lateralization of the vocal cord was performed. A 4-mm neck incision was made in a relaxed skin tension line at inferior border of thyroid cartilage, 1-cm lateral to midline. After suture #1 passed through the external cervical skin through paramedian cricothyroid membrane to entered the airway under the vocal cord, suture #2 passed through the same position and entered the airway through thyroid cartilage, superior to the vocal cord. After the end of suture #1 passed through the loop of suture #2, the suture loop was then pulled out of the back of the needle, bringing suture #1 with it. While observing the lateralization of vocal cord, suture #1 was tied over the silastic button.

All patients were nasotracheally intubated and sedated in the intensive care unit postoperatively. They received anti-reflux medication with proton inhibitors, analgesics plus intravenous antibiotics, and inhalation of budesonide following extubation. Clinical and swallow evaluation, advancement of oral intake and weaning off ventilatory support were managed on a case-by case basis.

Postoperative evaluation and follow-up

The functional outcomes of the surgery in terms of breathing, swallowing and voice, were evaluated and recorded. Anatomic follow-up was based on awake flexible laryngoscopy (1st month, 3rd month, 6th month and later twice per year, postoperatively). The grade of the vocal fold movement recovery was noted and recorded. Since the assessment of voice in neonates and infants was challenging, the following voice grading system based on clinical observation was used: 1) Normal voice; 2) Mild dysphonia: hoarse voice with some difficulties being heard or understood in loud environment; 3) Moderate dysphonia: weak voice or ventricular band phonation with easy fatigability; 4) Severe dysphonia: breathy voice with difficulty to communicate; 5) Aphonia.[11] Voice was assessed during hospitalization postoperatively and at the last follow-up by the parents. Functional Endoscopic Evaluation of Swallowing

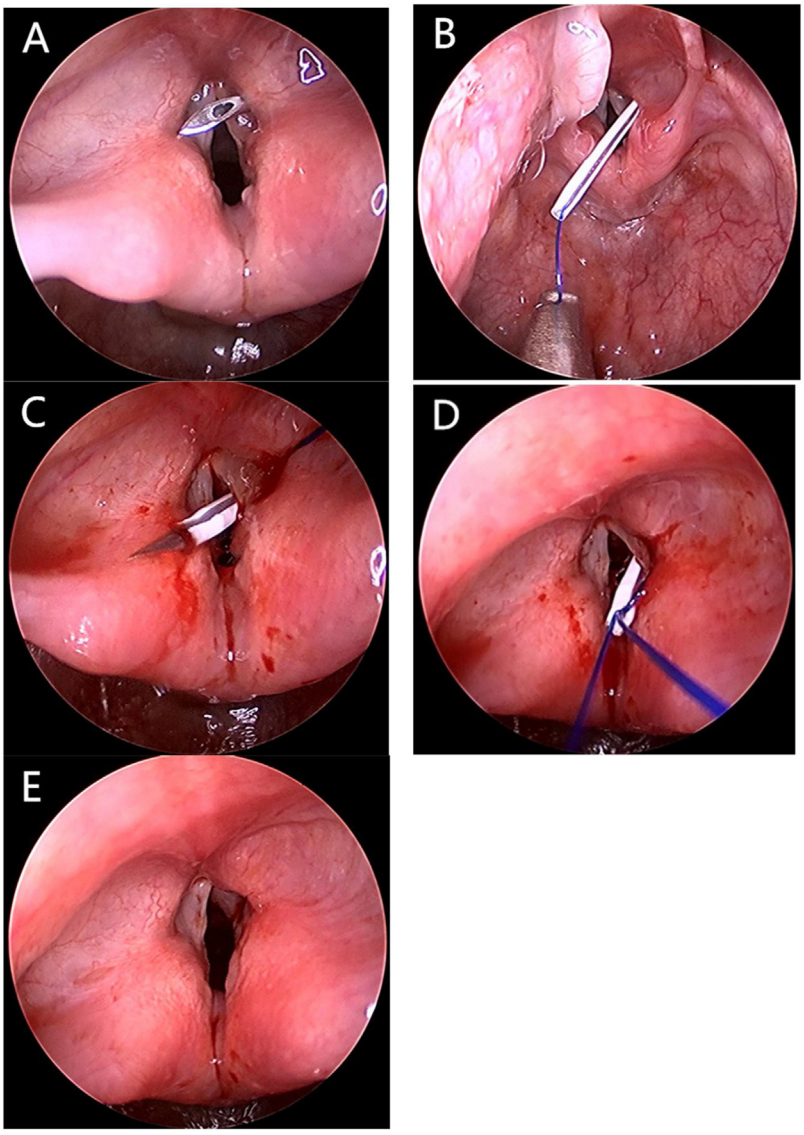

Figure 1 Endoscopic suture lateralization (A). Placement of 18-gauge needle superior to the vocal cord (B). Introduction of suture #1 (C). Placement of the 22-gauge needle inferior to the vocal cord for introduction of suture #2 (D). Passage of suture #1 through the loop of suture #2 (E). Lateralization suture under tension after securing to subcutaneous button in neck.

(FEES) evaluation was performed to identify aspiration (1st month, 3rd month, and 6th month, postoperatively). Naso-gastric tube and anti-reflux therapy was continued through the whole postoperative period until aspiration improved. At each visit, parents were asked to describe any swallowing difficulty and report the number of episodes of pneumonia.

Results

Both the EPSL procedure and the postoperative review on nine patients were performed by our senior author (Chao Chen). The age at diagnosis ranged 2–238 days (median age 16 days). The age at surgery ranged 15–250 days (median age 30 days). The interval between diagnosis and surgery ranged 8–77 days. All patients had severe dyspnea, and three of them had stridor and/or hoarseness. Preoperative airway managements included high-flow nasal cannula (n = 1), continuous positive airway pressure (n = 2) and endotracheal intubation (n = 6). Attempts were made to wean off the ventilatory support on each patient preoperatively, but to no avail. All patients required naso-gastric tube feeding prior to EPSL. Comorbidities were laryngomalacia (n = 2) and micrognathia (n = 3). The etiologies included idiopathic (n = 7) and neurological (n = 2) (Table 1).

Nine patients underwent ten EPSL procedures (one in the left vocal fold, and nine in the right vocal fold), and three of them underwent supraglottoplasty simultaneously. Follow-up was available to all patients (range 13–23 months, median 17 months). The length of surgery varied from 20 to 56 min s. Postoperatively, all patients were nasotracheally intubated for 3–14 days. After extubation, a non-invasive ventilatory equipment was employed on a case-by case basis, including Continuous Positive Airway Pressure (CPAP) (n = 7) and Non-Invasive Positive Pressure Ventilation (NIPPV)

Table 1 Demographic features of 9 patients.

Patient	Gender	Age at diagnosis (days)	Age at surgery (days)	Main symptoms	Comorbidities	Airway support	Etiology
1	M	19	30	Dyspnea	No	HF	Idiopathic
2	F	11	20	Stridor, hoarseness, dyspnea	No	ET	Idiopathic
3	F	238	250	Dyspnea	Micrognathia	CPAP	Idiopathic
4	M	84	92	Dyspnea	Laryngomalacia	ET	Neurological
5	M	75	152	Dyspnea	No	ET	Neurological
6	M	16	36	Stridor, dyspnea	Micrognathia	ET	Idiopathic
7	M	3	15	Dyspnea	No	CPAP	Idiopathic
8	M	2	19	Stridor, dyspnea	Micrognathia, laryngomalacia	ET	Idiopathic
9	M	12	26	Dyspnea	No	ET	Idiopathic

HF, High-Flow nasal cannula; CPAP, Continuous Positive Airway Pressure; ET, Endotracheal Intubation.

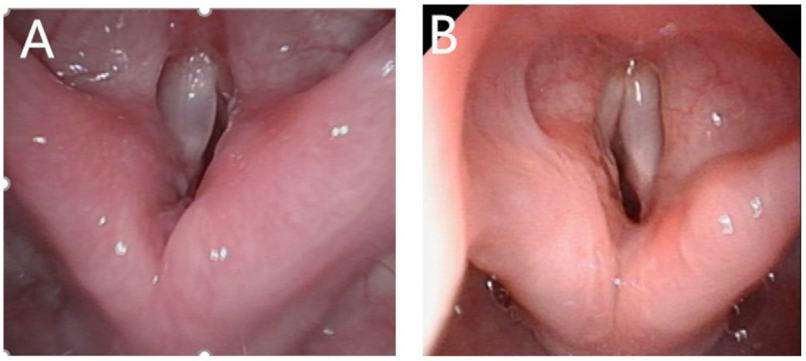

Figure 2 Postoperative flexible laryngoscopy (A). One year after right-sided EPSL (B). One year after left-sided EPSL.

(n = 3). Over a period of 3–7 days, eight of the nine patients (89%) were able to successfully wean off the non-invasive ventilatory equipment. Of these eight patients, five were limited exercise tolerant and three were full exercise tolerant on room air. Subsequent flexible laryngoscopies showed stable lateralization in all the nine patients (Fig. 2). They then had aspiration confirmed with FEES one month postoperatively and were able to tolerate an oral feeding without evidence of aspiration within 6 months. None of the patients presented with pneumonia episodes postoperatively. One patient experienced clinical improvement in ventilatory support (endotracheal tube to high-flow nasal cannula) and underwent laparoscopic nissen and gastrostomy tube placement one month after EPSL.

In terms of voice, two patients regained normal voice, two patients had mild dysphonia, and five patients had moderate dysphonia at the last follow-up. Of the eight patients undergoing right-sided lateralization, four had partial return of the left vocal fold function, but the other four showed no improvement. The one patient undergoing left-sided lateralization reported partial return of the right vocal fold function. Full data mentioned above are shown in Table 2.

No major perioperative or postoperative complications occurred. During the surgery, two complications were noted. One was the injury of the vocal cord and airway bleeding caused by the tip of needle passing through thyroid cartilage, and the other was acute laryngospasm. Briefly, four vocal cords were injured during the surgery, but, fortunately, no hematoma or ulcer evolved postoperatively. Though not serious in most cases, airway bleeding could blur the surgical field and prolong the length of surgery, and precise needle placement was critical for minimize the problem. In the two cases of acute laryngospasm complication, the surgery was suspended, and the patient was re-intubated till vocal cords completely relaxed. They recovered well without negative pressure pulmonary edema.

Three of the nine patients were found in more complicated condition. One patient (Patient 2) experienced re-intubation postoperatively. Another patient (Patient 8) experienced revision right-sided EPSL and cervical abscess. Still another patient (Patient 5) required high-flow nasal cannula and had aparoscopic nissen and gastrostomy tube placement performed postoperatively. Details are as follows.

Table 2 Outcomes of 9 patients.

Patient	Surgery		Duration of ET postop (days)	Post EX	Airway support		Feeding status		Aspiration (FEES)			Voice	VF mobility	FU (months)
	Type	Length (minutes)			Duration (days)	Last FU	Postop	Last FU	1st month postop	3rd month postop	6th month postop	Last FU	Recovery	
1	R-EPSL + SGP	40	3	CPAP	3	RA (FET)	NG	PO	Yes	No	No	normal	LP	23
2	L-EPSL	32	3 + 7	CPAP	3	RA (LET)	NG	PO	Yes	Yes	No	mild Dysphonia	RP	23
3	R- EPSL	31	7	NIPPV	3	RA (LET)	NG	PO	Yes	No	No	mild Dysphonia	LP	20
4	R- EPSL	37	7	CPAP	5	RA (FET)	NG	PO	Yes	No	No	moderate Dysphonia	LP	19
5	R- EPSL	20	7	CPAP	7	HF	NG	GT	Yes	Yes	–	moderate Dysphonia	None	14
6	R-EPSL + SGP	53	7	CPAP	3	RA (FET)	NG	PO	Yes	No	No	normal	LP	17
7	R- EPSL	36	7	NIPPV	5	RA (LET)	NG	PO	Yes	No	No	moderate Dysphonia	None	17
8	R-EPSL + SGP	51 + 56	7 + 7	NIPPV + CPAP	5 + 7	RA (LET)	NG	PO	Yes	Yes	No	moderate Dysphonia	None	17
9	R-EPSL + SGP	26	7	CPAP	3	RA (LET)	NG	PO	Yes	No	No	moderate Dysphonia	None	13

ET, Endotracheal Intubation; FEES, Functional Endoscopic Evaluation of Swallowing evaluation; VF, Vocal Fold; FU, Follow-Up; postop, postoperatively; EX, Extubation; R-EPSL, Right-sided Endoscopic Percutaneous Suture Lateralization; L-EPSL, Left-sided Endoscopic Percutaneous Suture Lateralization; SGP, Supraglottoplasty; CPAP, Continuous Positive Airway Pressure ; NIPPV, Non-Invasive Positive Pressure Ventilation; RA, Room Air; LET; Limited Exercise Tolerant; FET, Full Exercise Tolerant; HF, High-Flow nasal cannula; NG, Nasogastric Feeding; GT, Gastrostomy Tube; PO, Oral Feeding; LP, Left Partial; RP, Right Partial.

Patient 2 was born with hoarse crying, stridor, and respiratory retraction, and was intubated within a few days after birth. The diagnosis of BVFP was confirmed at 11 days of age by flexible laryngoscope. She underwent left-sided EPSL at 20 days of age and was extubated 3 days later. She was in good condition the first 2 days on CPAP until respiratory distress and stridor reoccurred on Day 3. As flexible laryngoscopy revealed obvious edema of glottic mucosa and vocal folds. Then, she was re-intubated for another 7 days. After the second extubation, no dyspnea reoccurred at the last follow-up.

Patient 8 was born with severe ventilatory effort and stridor and was intubated immediately. Flexible laryngoscopy revealed BVFP, micrognathia and laryngomalacia at 2 days of age. He underwent right-sided EPSL and supraglottoplasty simultaneously at 19 days of age. Seven days later, the endotracheal tube was successfully removed, followed by NIPPV. However, the patient's airway and breathing deteriorated on Day 3. Flexible laryngoscopy and MLB indicated that the right-sided suture was so close to the end of vocal fold that the vocal fold was not lateralized enough. He returned to OR and had a second right-sided EPSL performed. During the second right-sided EPSL, with the original suture reserved in situ, another suture was placed more anteriorly to provide competent vocal fold abduction. After this procedure, he showed consistent improvement in his airway, and was discharged home shortly afterwards. He revisited our outpatient department 13 days later because of an acute cervical abscess on the surgical site. He underwent cervical drainage and was treated with oral antibiotics for one week. His condition has remained stable so far.

Patient 5 was born at 32 weeks plus 3 days with generalized cyanosis and without cry at home. Fifteen minutes after birth, he underwent endotracheal intubation and cardiopulmonary resuscitation. Upon recovery of heartbeat and respiration, he was transferred to our hospital, and was diagnosed with ischemic-hypoxic encephalopathy on cranial MRI afterwards. Dysphagia presenting as poor suck and absent swallow reflex was identified secondary to poor muscle tone and coordination. BVFP was identified by flexible laryngoscopy at 2.5 months of age, and EPSL was performed at 5 months of age to improve his respiratory status. After this procedure, he was allowed to extubate endotracheal tube but still needed high-flow nasal cannula. Persistent silent aspiration was confirmed with FEES postoperatively, and he underwent laparoscopic nissen and gastrostomy tube placement instead of nasogastric feeding tube at 6 months of age, before he was transferred to a rehabilitation hospital. At the one-year follow-up, he was gaining weight well, and had improved airway patency.

Discussion

Bilateral vocal fold paralysis in neonates and infants is a delicate challenge for the laryngologist who has to optimize the balance between the three main functions of the larynx (breathing, swallowing and phonation). Given that more than 50% of children with BVFP have spontaneous resolution in the first 12 months of life, traditional management was presented as early tracheostomy to establish an effective airway and waiting for the recovery of vocal fold mobility.[12,13] In a large review, 68.6% of children with BVFP were found to require tracheostomy, and ultimately 64.3% were successfully decannulated.[14] Although it is potentially life-saving, tracheostomy comes at the expense of significant morbidity and mortality.[1] For neonates and infants, it is still devoid of consensus regarding surgical indications, timing for tracheostomy, and perioperative and follow-up management.[15] Tracheostomy also requires intense support from parents and caregivers, who all need to be appropriately trained in caring for the tracheostomy. Such care needs, dependencies, and impact on families need to be addressed before placing a tracheostomy in a child.[16] Thus, in the past 30 years, advances in endoscopic laryngeal surgery have been developed to avoid tracheostomy. Naunheim et al. undertook a cost-effectiveness analysis to evaluate endoscopic management and tracheostomy of BVFP in both short-term and long-term scenario and found that endoscopic management of BVFP appeared to be more cost-effective than tracheostomy. Although it demanded expertise and specialized equipment, an endoscopic strategy demonstrated utility gains and long-term cost advantages over tracheostomy.[17]

Several endoscopic procedures have been developed, including arytenoidectomy and/or posterior cordotomy, anterior/posterior cricoid split, and arytenoid abduction lateropexy and suture lateralization. In the published literature, the rates of decannulation or avoiding tracheostomy of all these procedures were satisfying. As previously reported, in a series of 17 pediatric patients with BVFP who underwent arytenoidectomy alone or combined with posterior cordotomy, 13 patients (76.5%) achieved decannulation or extubation eventually: nine of 12 tracheotomy-dependent patients were decannulated, and four of the rest five patients were extubated postoperativiely.[3] In another study, endoscopic Anterior and Posterior Cricoid Split (APCS) procedures for BVFP was reported in 19 neonates across 4 institutions, with a success rate of 74%.[7] Madani et al. introduced a minimally invasive and quick Endoscopic Arytenoid Abduction Lateropexy (EAAL) using a modified Endolaryngeal Thread Guide Instrument (ETGI) for 4 newborns with BVFP, and reported no dyspnea or swallowing disorder occurred postoperatively.[8] Sztanó et al. reported on three patients treated with EAAL with ETGI that no dyspnea or swallowing disorder occurred over three years of observation.[9] Montague et al. reported that of six neonates with BVFP undergoing EPSL, one patient expired due to unrelated neurodegenerative disease, and the other five surviving patients were stable on room air without tracheostomy.[10] Similarly, in the present study, we performed EPSL in nine patients with BVFP using a technique that did not require any demanding surgical craft or specialized equipment, and all patients were able to avoid tracheostomy afterwards. Clearly, even though the outcomes of EPSL were not significantly superior to other techniques, the nondestructive nature of EPSL gives it advantage over cordotomy/arytenoidectomy operations and the simplicity of EPSL over anterior/posterior cricoid split and EAAL with ETGI.

The most focused concern with regard to endoscopic operations is that they all might produce the changes at the level of the larynx that increase the risk of aspiration and/or dysphonia postoperatively.[7] In the Montague's series,

5/5 surviving patients were able to resume a full oral diet without aspiration eventually.[10] In our series, the patients' swallowing function gradually recovered as well. In the first month postoperatively, all patients suffered obvious aspiration so that nasal gastric feeding tube necessitated. In the third month, aspiration improved in most patients. By the sixth month, all but one patient were fed orally without aspiration. It is therefore concluded that EPSL might interfere with swallowing in a short term but not in a long term, regardless of recovery of vocal fold mobility or not. Our finding is consistent with that of Hsu et al., who suggested that compensatory swallowing function could be achieved even if bilateral vocal fold mobility did not return.[18] In other words, partial vocal fold lateralization was safe on swallowing function. While voice quality was difficult to be measured objectively at this age in our study, the results were acceptable to parents. A longer-term follow up of voice is planned to determine the impact of this procedure on phonation.

One criticism of endoscopic management of BVFP is the need for reoperation postoperatively.[17] As previously reported, ten of the 19 patients (52.6%) required adjunctive procedures following the initial APCS, and these procedures included balloon dilation, supraglottoplasty, and revision APCS[7]; in one patient out of 5 (20%) who underwent EPSL, right-sided suture was removed and left suture lateralization was performed.[10] In our study, however, only one patient (11.1%) underwent revision right-sided suture. We perceive two main factors may impact on the revision rate. One is the airway comorbidities of the patients. More complicated the comorbidities may lead to higher revision rate, and revision rates could be more controllable on well-selected patients. The other is the experience of the surgeons. Surgeons with significant endoscopic experience appeared more likely to have lower revision rates, hence the importance of expertise and specialty training in endoscopic management of BVFP.

Although detailed reports on the surgeon-related causes of revision EPSL are sparse, they have revealed quite similar findings. In a series of adult patients with BVFP, two underwent second operations; one due to dislocation of the silicone block, and the other due to inadequate lateralization.[19] In a mentioned-above study, one patient underwent left-sided EPSL after the removal of the tightened right-sided suture which led to false vocal fold edema.[10] In our study, Patient 8 underwent a second right-sided EPSL because of inadequate lateralization. In the Patient 8, the concomitant micrognathia caused increased difficulty in providing adequate exposure to the glottis and precise needle placement under MLB, resulting in misplacement of suture and eventually revision EPSL. Our study therefore consolidates the understanding that the placement, tightness, and fixation of the suture are crucial for the success of the surgery.

Although EPSL is generally reliable and safe, potential risks or complications may occur, including suture breaking, granulomas on vocal fold, chronic submucosal cordotomy, medialization of vocal fold, and alteration of crico-arytenoid joint dynamics or even ankylosis, etc. Overall, our cases were free from these complications by now. In addition, an important consideration is what to do with the suture over time. The suture was removable, but we did not undertake any removal during this study period. Removal needs to be carefully considered, because reinnervation of the two vocal folds might not occur simultaneously. We believe further investigation in this study is needed, which may involve more study cases and longer follow-up duration.

The limitations of the study are obvious. First, as a small case series study, this research has its inherent limitations, such as absence of a control group, which limits its generalizability. Second, our follow-up periods were short and variable, which limits the conclusions that can be drawn about the long-term outcome of EPSL. In addition, a larger cohort via a prospective, multi-institutional effort with a standardized approach would yield more patients and possibly more statistically significant results.

Conclusion

Our preliminary results suggested that endoscopic percutaneous suture lateralization appears to be an effective and safe treatment for neonatal and infantal BVFP, which enables patients free from tracheostomy without significant impact on swallowing function or phonation.

Conflicts of interest

The authors declare no conflicts of interest.

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