Endoscopic percutaneous suture lateralization for neonatal bilateral vocal fold immobility☆


A U C S F-B e n i o f f C h i l d r e n ’ s H o s p i t a l , D i v i s i o n o f P e d i a t r i c O t o l a r y n g o l o g y , D e p a r t m e n t o f O t o l a r y n g o l o g y - H e a d a n d N e c k S u r g e r y , U n i v e r s i t y o f C a l i f o r n i a , S a n F r a n c i s c o , U n i t e d S t a t e s
b S e a t t l e C h i l d r e n ’ s H o s p i t a l , D i v i s i o n o f P e d i a t r i c O t o l a r y n g o l o g y , D e p a r t m e n t o f O t o l a r y n g o l o g y - H e a d a n d N e c k S u r g e r y , U n i v e r s i t y o f W a s h i n g t o n , U n i t e d S t a t e s

A R T I C L E I N F O

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Airway
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A B S T R A C T

Objective: Bilateral vocal-fold immobility (BFVI) is a rare but significant cause of severe respiratory distress in neonates. The primary aim of treatment is to provide an adequate airway while minimizing adverse effects such as aspiration and dysphonia. Our objective here is to describe the outcomes of a series of neonates undergoing percutaneous endoscopic suture lateralization for BFVI using a novel technique.

Methods: In this retrospective case series, we present 6 neonates (mean age: 18 days) with BFVI from three tertiary academic medical centers. The etiologies included 4 idiopathic, 1 unspecified neurodegenerative disorder, and 1 acquired from cardiac surgery. All had stridor and respiratory distress with hypoxemia requiring respiratory support at diagnosis. Endoscopic vocal-fold lateralization was performed under spontaneous-breathing suspension laryngoscopy using a novel technique of percutaneous needle-directed placement of 4–0 prolene suture without use of specialized equipment.

Results: All patients had clinical improvement in stridor and respiratory support requirements and avoided tracheostomy. One patient had persistent aspiration after lateralization that resolved after suture removal. One patient required bilateral lateralization procedures. One patient expired of epilepsy due to neurodegenerative disease unrelated to airway pathology. At last follow-up (mean 12.6 months), 5/5 remaining patients were on room air without tracheostomy and feeding orally without aspiration; 4/5 had partial or complete return of vocal-fold function.

Conclusion: Endoscopic percutaneous suture lateralization may be a safe and effective non-destructive primary treatment modality for neonatal BFVI. All neonates undergoing this procedure avoided tracheotomy.

1. Introduction

Bilateral vocal-fold immobility (BFVI) is a rare but significant cause of severe respiratory distress in neonates often requiring urgent intervention. Etiologies include birth trauma, neurological disorders such as Arnold-Chiari malformation, hydrocephalus, cerebral palsy, hypoxia, and cardiac surgery. The etiology in most cases is unknown [1]. The primary aim of treatment is to provide an adequate airway for ventilation while minimizing adverse effects such as aspiration and dysphonia. Current management options vary widely on a spectrum that includes non-invasive positive pressure ventilation (NIPPV), endoscopic surgery such as cricoid split [2–4], cordotomy [5], and tracheotomy. In some series, up to 90% of patients with BFVI underwent tracheostomy [6,7]. The considerable rate of spontaneous recovery, which is greater than 50% in both idiopathic and acquired cases, makes more invasive or destructive treatments less desirable [7,8]. Here, we present the first multi-institutional series of neonates undergoing reversible endoscopic percutaneous suture lateralization for BFVI.

Suture lateralization has been a treatment modality for BFVI for several decades in adults since it was introduced by Ejnell and Tisell in 1993 [9]. The Lichtenberger endo-extralaryngeal needle carrier has become a popular tool to perform lateralization [10]. However, this technique is not feasible in neonates due to the size and angle of the insertion tool obstructing visualization. A recent small case series described a modified version of the Lichtenberger needle insertion tool that was used for endoscopic suture lateralization in neonates at a single institution; however, this specialized instrument is not widely available [11].

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In this study, we describe outcomes in 6 neonates with BVFI who underwent a novel technique for endoscopic suture lateralization. Our technique does not require any specialized modified instruments, but can be performed with equipment and materials that are readily available as part of routine pediatric laryngoscopy and bronchoscopy.

2. Materials and methods

We reviewed the charts of 6 patients with BVFI from two tertiary academic medical centers who all underwent endoscopic suture lateralization. Demographic information obtained included patient age at diagnosis, comorbidities, etiology and length of follow-up. Perioperative information obtained included amount and type of respiratory support, medical evaluation including MRI, aspiration and feeding status. Outcome measures were respiratory support and airway status, vocal fold function, and feeding and aspiration on follow-up. Approval for this retrospective study was obtained from the Committee on Human Research of the University of California, San Francisco and the Institutional Review Board of Seattle Children's Hospital.

Included patients all had BVFI diagnosed upon awake flexible laryngoscopy, with clinical correlation that BVFI was causing associated respiratory distress or feeding difficulty. Preoperative counseling was performed describing the primary benefits of the surgery as improvement in respiratory distress and feeding difficulty with a goal for avoidance of tracheostomy. The risks described included, but were not limited to, airway edema leading to worsening respiratory distress, worsening aspiration, stitch abscess, and granuloma formation.

All children underwent suspension direct laryngoscopy while spontaneously breathing under total intravenous anesthesia using propofol. Intermittent endotracheal intubation was performed with an uncuffed endotracheal tube as needed. 0.5 mg/kg dexamethasone and prophylactic antibiotic for skin coverage were administered at the onset of the case.

We prepare the following equipment: 1) Lindholm laryngoscope with suspension arm; 2) Microlaryngeal instruments including graspers, laryngeal distending forceps, a long-handled knife, scissors, or pick; 3) Minor plastics tray; 4) 4–0 prolene suture x 3; 5) 22 gauge and 19 gauge needles without filters; 6) 1 cc syringe with stub-tip x 3; 7) 1-mm-thick silastic sheet fashioned into a 5 × 5 mm button with two buttonholes; 8) 5–0 fast absorbing suture, histoacryl for skin closure; 9) 4-mm 0-degree Hopkins rod telescope with camera and light cord; and 10) operating microscope.

First, sutures are prepared prior to laryngoscopy. A 4–0 Prolene suture (suture #1) is loaded into a 22 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) is loaded into a 19-gauge needle and also secured with a 1-cc stub-tip syringe. Preloading suture through the needle tip, and securing it loosely with the syringe was found to be critical, as placement of the suture through the hub end of the syringe intraoperatively was challenging. The silastic button is fashioned and soaked in betadine on the sterile field.

Next, the patient is placed into suspension laryngoscopy with a Lindholm laryngoscope and vocal folds palpated to confirm passive mobility and absence of firm fixation. Weight-based topical lidocaine is applied to the larynx. Best visualization is obtained with a 4 mm 0° telescope, but one may use a microscope if single surgeon is performing the operation. Next, a 4-mm neck incision is made in a relaxed skin tension line at inferior border of thyroid cartilage, 1-cm lateral to midline. Skin is elevated to expose the surface of the strap muscles sufficient to accommodate the subcutaneous 5-mm silastic button. Laryngeal distending forceps are placed to allow adequate visualization of the subglottis and true vocal folds (TVFs).

Precise needle placement is critical for surgical success and minimization of airway bleeding. Needles were placed percutaneously, and the airway not entered until the mucosa was clearly seen to be tented by the needle in the proper location. The 22-gauge needle is placed through the incision, approximately 7 mm lateral to midline, at inferior edge of thyroid cartilage and directed through the paramedian cricothyroid membrane to enter the airway under the TVF just anterior to the vocal process. Once the tip of needle is in airway, suture #1 is advanced into the airway and retrieved with endoscopic laryngeal graspers, and the 22-gauge needle is withdrawn. Next, the 19 gauge needle is passed through the same incision aiming more superiorly so that the thyroid cartilage is firmly engaged by the needle tip. This needle passes through the thyroid cartilage and should enter the airway in the ventricle, just superior to the TVF and just anterior to the vocal process. Once the tip of the 19-gauge needle is seen in the ventricle, the suture loop is passed into the airway. With the needle still in the airway, the end of suture #1 is passed through the loop of suture #2 and retrieved out of the laryngoscope under tension. The suture loop is then pulled out the back of the 19-gauge needle, bringing suture #1 with it. Suture #2 is removed together with the 19-gauge needle. At this point, suture #1 is around the vocal ligament with both ends coming out the incision. These two ends are passed through the holes in the silastic button. While observing the airway, suture #1 is tied over the button, while watching the TVF lateralize. At this point, the button and suture knot are within the subcutaneous soft tissue of the neck. The patient was taken out of suspension and the neck wound irrigated and closed over the button and suture, which are completely buried.

Management of post-operative endotracheal intubation was considered individually. All patients received 0.5 mg/kg decadron every 8 h for 24 h and acid suppression through postoperative period. Clinical and radiographic swallow evaluation, advancement of oral intake and weaning off respiratory support were managed on a case-by-case basis.

3. Results

We present 6 neonates (median age at diagnosis, 2 days, range 2–81 days) with BVFI from three tertiary academic children's hospitals who underwent endoscopic percutaneous suture lateralization (Fig. 1, Supplemental Video 1). The etiologies included 4 idiopathic, 1 unspecifiable neurodegenerative disorder, and 1 acquired after cardiac surgery (Table 1). All presented with stridor that was worse on agitation with O2 desaturation. Preoperative airway management varied from intermittent nasal cannula to endotracheal intubation. Most required nasogastric tube feeding prior to surgery. All underwent preoperative MRI, with only one (patient 6) with abnormal findings.

Supplementary video related to this article can be found at http://dx.doi.org/10.1016/j.ijporl.2018.02.032.

All patients experienced clinical improvement in stridor and respiratory support requirements and avoided tracheostomy (Table 2). Of the 6 cases, three had a straightforward course where symptoms of stridor and supplemental oxygen dependence improved immediately after initial suture lateralization with durable benefit and no long-term change in swallow function. One of these 3 had transient clinical concern for new aspiration immediately post-op confirmed with modified barium swallow study. He was allowed to breastfeed without nasogastric tube placement and had clinical resolution of aspiration without complication. Three of the 6 cases were more complicated. Two of them required further procedures, and one patient expired of underlying neurologic disease but still successfully avoided tracheostomy. Details of the complicated cases are as follows.

Patient 2 was diagnosed at 3 months of age due to ongoing stridor, failure to thrive, respiratory distress with oxygen requirement, severe reflux and aspiration. His past medical history was significant for double aortic arch coarctation for which he underwent repair, as well as subsequent plication of the right hemidiaphragm for an iatrogenic right phrenic paralysis. Aspiration was confirmed with videofluoroscopy, and he underwent laparoscopic nissen and gastrostomy tube placement prior to diagnosis of BVFI and suture lateralization. On initial diagnosis of BVFI, he had slight movement of the right TVF and no movement of

![Image](dx.doi.org/10.1016/j.ijporl.2018.02.032)

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the left TVF, requiring endotracheal intubation. After he underwent left suture lateralization, he had immediate improvement in his airway and was weaned from high flow oxygen support. Postoperatively he had persistent aspiration on VFSS. He experienced gradual return of function of the left vocal fold. At 10 months of age, he had the suture removed due to persistent aspiration. After removal, his glottic closure and swallow function improved, and he resumed a full oral diet.

Patient 4 presented immediately after birth with stridor and desaturation with agitation. Flexible fiberoptic laryngoscopy confirmed BVFI. Workup with MRI was unrevealing and there were no co-morbidities. She underwent right suture lateralization on her 6th day of life. Stridor and desaturations improved, she tolerated an oral diet, and was discharged home shortly after. She was readmitted two weeks later for recurrent stridor, failure to thrive, and desaturation with agitation. She returned to the OR on day of life 25 and the suture was not visible in airway. The right sided suture was tightened. Due to persistent symptoms, she returned to OR at 1 month of age and was found to have subglottic stenosis as well as right false vocal fold edema. The right-sided suture was removed and left suture lateralization was performed. After this procedure she had more consistent improvement in her airway symptoms. Postoperative video swallow study revealed no aspiration. At two month follow up, she was gaining weight well, experienced partial return of function, and had greatly improved stridor and airway patency. At two year follow up she demonstrated interval complete return of function bilaterally. She was noted to have some dysphonia, so the left-sided suture was removed with improvement in

Table 1
Patient demographics.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age at diagnosis (days)</th>
<th>Age at surgery (days)</th>
<th>Comorbidities</th>
<th>Gender</th>
<th>Presenting symptoms</th>
<th>Etiology</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>18</td>
<td>29</td>
<td>no</td>
<td>M</td>
<td>S/D</td>
<td>Idiopathic</td>
</tr>
<tr>
<td>2</td>
<td>81</td>
<td>108</td>
<td>Coarctation of aorta, vascular ring</td>
<td>M</td>
<td>S/D, aspiration, intubation</td>
<td>Iatrogenic</td>
</tr>
<tr>
<td>3</td>
<td>2</td>
<td>25</td>
<td>no</td>
<td>F</td>
<td>S/D, NIPPV</td>
<td>Idiopathic</td>
</tr>
<tr>
<td>4</td>
<td>2</td>
<td>6</td>
<td>no</td>
<td>F</td>
<td>S/D</td>
<td>Idiopathic</td>
</tr>
<tr>
<td>5</td>
<td>2</td>
<td>24</td>
<td>no</td>
<td>F</td>
<td>S/D</td>
<td>Idiopathic</td>
</tr>
<tr>
<td>6</td>
<td>2</td>
<td>2</td>
<td>Epilepsy, Micrognathia, clubfeet, hip dysplasia</td>
<td>F</td>
<td>S/D, NIPPV</td>
<td>Neurologic</td>
</tr>
</tbody>
</table>

Abbreviations: NIPPV, non-invasive positive-pressure ventilation; S/D, stridor, desaturations with agitation.
voice. She is doing well postoperatively.

Patient 6 expired from her underlying neurodegenerative disease unrelated to her airway pathology. At time of BFVI diagnosis she had an unknown neurologic condition with abnormal sulcations but no Chiari malformation found on MRI. She was aspirating secondary to poor muscle tone and coordination. Suture lateralization was performed to improve her respiratory status and she was able to be discharged home without tracheostomy. She was ultimately found to have a poor neurologic prognosis based on muscle biopsy results and after family discussion was ultimately made comfort care. She died at home at 4 months of age.

At last follow-up, all surviving patients were stable on room air without tracheostomy and feeding orally without aspiration. Four of 5 had partial or complete return of function of at least one vocal fold.

4. Discussion

Bilateral abductor paresis and paralysis of the vocal folds in neonates is a challenging condition to manage. The aim of treatment of BVFI is to improve airway patency while minimizing long-term impact on swallowing function and phonation. The high rate of spontaneous resolution makes irreversible and morbid procedures undesirable. Historically, the most common treatment for BVFI has been tracheostomy; in a large review, 68.6% of children with BVFI were found to require tracheostomy. Though ultimately 64.3% were successfully decannulated, the morbidity and cost associated with pediatric tracheostomy make this a less attractive option [12]. Therefore, multiple less-invasive options have been described, including non-invasive positive pressure, arytenoidectomy and posterior cordotomy, anterior/posterior cricoid split, and suture lateralization.

Non-invasive supportive means such as continuous positive pressure ventilation as an alternative treatment carries a separate set of complications. Chronic NIPPV is associated with deleterious effects on development of the face and lungs. The force exerted by positive pressure on immobile airways has been implicated in a variety of disorders, most commonly tracheomalacia and tracheobronchomalacia [13]. Furthermore, options for home delivery of NIPPV are limited.

Arytenoidectomy and posterior cordotomy have been performed both separately and in conjunction for adults and children for greater than twenty years. In a series of 17 pediatric patients with BVFI over 13 years who underwent arytenoidectomy, there was a significant incidence of dysphonia (5/17) and a smaller group with persistent aspiration with pneumatopathy (2/17) [14]. Posterior cordotomy represents a more minimally invasive endoscopic approach. By freeing the vocal ligament and vocalis muscle from the vocal process, tissue retraction occurs and there is subsequent enlargement of the airway [5]. While this may have lesser impact on voice and swallowing, it does require an irreversible alteration of the larynx. It can be repeated if necessary and performed on the contralateral side to add adduction benefit. In a review of 11 patients over an 11 year span, the majority who underwent cordotomy either avoided tracheostomy or were ultimately decannulated [5].

In 2017, simultaneous anterior and posterior cricoid split procedures for BVFI was reported in 19 neonates across 4 institutions, with a success rate of 74% defined as avoidance of tracheostomy or successful decannulation [2]. One patient developed new aspiration. Five out of 14 successful and all five unsuccessful patients required further procedures. The largest disadvantage of anterior posterior cricoid split surgery is its irreversible alteration of the laryngeal skeleton. The long-term sequelae to phonation are unknown.

Arytenoidopexy with or without arytenoidectomy have been described extensively in the literature (reviewed in [15]), consisting of either external or endoscopic approaches to place a suture into the vocal process of the arytenoid and secure it against the thyroid cartilage to stably lateralize the vocal fold. This has primarily been described as a secondary management modality for BVFP in older infants and toddlers to facilitate decannulation (successful in 79% of cases), and has a significant rate of long-term dysphonia (38%). Endoscopic arytenoidopexy has not been widely described as a means of primary management of BVFP in the neonatal period to avoid tracheostomy.

In a study most similar to ours, a version of suture lateralization described as endoscopic arytenoid lateralopexy using a modified adult endolaryngeal thread guide instrument on 4 patients [11]. The angle of the thread guide was made less acute so that it would fit into a smaller airway diameter and permit concomitant endoscopy for visualization. All four children were intubated for 3–7 days and then began feeding by mouth between postoperative day 4–10. Only 1 of 4 in that study required reintubation for laryngeal edema, which was treated with antibiotics and steroids with avoidance of tracheostomy. All children were fed via NG tube during the postoperative intubation period then oral diet after extubation, though formal swallow evaluation with video fluoroscopy or functional endoscopic evaluation was not reported. No sutures were removed or revised in this study.

In our study, we performed endoscopic suture lateralization in 6 neonates with BVFI with a variety of etiologies using a technique that did not require any specialized modified equipment. Our procedure is distinct from the more widely described arytenoidopexy, as the suture placed does not directly engage the cartilage of the vocal process of the arytenoid, but rather exerts a lateralization force by sitting on top of the vocal fold mucosa just anterior to the vocal process and pulling the

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### Table 2

Patient outcomes.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Airway support</th>
<th>Feeding Status</th>
<th>Aspiration</th>
<th>Vocal-fold Mobility</th>
<th>FU (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Preop</td>
<td>Last FU</td>
<td>Preop</td>
<td>Last FU</td>
<td>Preop</td>
<td>Postop</td>
</tr>
<tr>
<td>1</td>
<td>NC</td>
<td>RA</td>
<td>NG</td>
<td>PO</td>
<td>PO</td>
</tr>
<tr>
<td>2</td>
<td>ET</td>
<td>RA</td>
<td>GT</td>
<td>GT</td>
<td>PO</td>
</tr>
<tr>
<td>3</td>
<td>CP</td>
<td>RA</td>
<td>NG</td>
<td>NG</td>
<td>PO</td>
</tr>
<tr>
<td>4</td>
<td>NC</td>
<td>RA</td>
<td>PO</td>
<td>PO</td>
<td>PO</td>
</tr>
<tr>
<td>5</td>
<td>NC</td>
<td>RA</td>
<td>PO</td>
<td>NG</td>
<td>PO</td>
</tr>
<tr>
<td>6</td>
<td>HF</td>
<td>RA</td>
<td>NG</td>
<td>NG</td>
<td>GT</td>
</tr>
</tbody>
</table>

Abbreviations: FU, follow-up; NC, O2 by nasal cannula; ET, endotracheal intubation; CP, continuous positive airway pressure; HF, high-flow nasal cannula; RA, room air; NG, nasogastric feeding; GT, gastrostomy tube; PO, oral feeding.
vocal ligament laterally. While we anticipated that this might lead to less robust lateralization of the vocal fold, due to our percutaneous, rather than endolaryngeal, introduction of the needles and suture, and small neonatal airway, we wished to minimize risk of direct trauma, inflammatory reaction, or granulation relating to the arytenoid proper, and risk of cricoarytenoid ankylosis.

In our series, all neonates were able to avoid tracheotomy and maintain long-term airway patency, suggesting that our technique of lateralization without direct arytenoidopexy was sufficiently robust. Complications included need for further procedures and transient aspiration. Though one baby expired due to unrelated neurologic morbidity, the 5 remaining were all feeding exclusively by mouth without clinical signs of aspiration at most recent follow up. Limitations of the study include a heterogeneous and small retrospective cohort without comparator group, which limits generalizability. Impact on phonation was not consistently or formally measured, but the prior study of neonatal suture lateralization using a very similar technique did report satisfactory vocal outcomes [11]. Longer-term follow up of our cohort is planned, in order to determine impact of this procedure on phonation.

An important consideration is what to do with the suture over time. It is felt that some submucosal cordotomy likely occurs and the vocal fold mucosalizes over the suture. We performed suture removal in three cases. In all instances, the suture was no longer present within the airway at time of removal. In one case, the initial suture placed was ineffective for airway patency, so it was removed and a contralateral suture placed with successful airway improvement. The other two were removed on account of long-term symptoms; one was removed for persistent aspiration, the other for dysphonia. In both cases, symptoms improved after suture removal without any worsening of airway and breathing. This suggests that the suture, while no longer within the airway, was still providing some lateral tension on the vocal fold, that was released upon removal, allowing improving of active vocal-fold abduction. We are unable to ascertain whether there is some component of permanent lateralization caused by the suture even after its removal; it is likely that some degree of “auto-cordotomy” occurs, and possible that there is some permanent lateralization due to alteration of crico-arytenoid joint dynamics or even ankylosis. At the time of suture removal, palpation of the cricoarytenoid joint did not reveal any gross fixation, but we were unable to determine whether some subtle changes in joint mobility had occurred. Overall, however, our experience with these cases demonstrates at least some reversability of the suture lateralization: we propose, then, that indications for suture removal may include lack of efficacy, persistent aspiration, or dysphonia.

5. Conclusion

Endoscopic percutaneous suture lateralization may be a safe primary treatment modality for neonatal BVFI. Compared to other described techniques, ours requires no specialized equipment and results in no known permanent alterations of the laryngeal framework or vocal-fold anatomy. Neonates undergoing this procedure avoided tracheotomy.

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Conflicts of interest

The authors have no relevant conflicts of interest.

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