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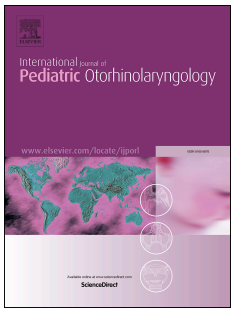

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ABSTRACT

OBJECTIVES: Bilateral vocal cord paralysis often causes severe dyspnea requiring an early airway intervention in neonates. Endoscopic arytenoid abduction lateropexy (EAAL) with suture is a quick, reversible, minimally-invasive vocal cord lateralizing technique to enlarge the glottis. The arytenoid cartilage is directly lateralized to a normal abducted position. It can be performed even in early childhood with the recently-introduced pediatric endoscopic thread guide instrument. The long-term results and the stability of the lateralization were evaluated.

METHODS: Three newborns had inspiratory stridor immediately after birth. Laryngo-tracheoscopy revealed bilateral vocal cord paralysis. Unilateral, left-sided endoscopic arytenoid abduction lateropexy was performed with supraglottic jet ventilation. The follow-up period was >3 years.

RESULTS: After extubation on the 4-7th postoperative day no dyspnea or swallowing disorder occurred. Laryngo-tracheoscopy, clinical growth charts and voice analysis showed satisfactory functional results.

CONCLUSIONS: The endoscopic arytenoid abduction lateropexy might be a favorable solution for neonatal bilateral vocal cord paralysis. In one step, airway patency can be achieved without irreversible damage to the glottic structures. Normal swallowing function was preserved. The results are durable, and neither medialization nor dyspnea re-appeared during observation.

Level of Evidence: 4

KEYWORDS: bilateral vocal fold paralysis, congenital dyspnea, vocal fold immobility, endolaryngeal thread guide instrument, endoscopic arytenoid abduction lateropexy, laterofixation

INTRODUCTION

The treatment of bilateral vocal cord paralysis (BVCP) is a challenge in children and especially in neonates. Associated stridor and respiratory distress generally require urgent interventions [1-3]. Miyamoto et al in a series of 22 patients reported a 68% tracheotomy rate [4]. Earlier reports favored tracheotomy more heavily. Certainly, among the options for surgical management of BVCP, tracheotomy is the most studied. Rates of spontaneous recovery of vocal cord function range from 33 to 65%, and tracheotomy has the theoretical advantage of restoring an airway without altering laryngeal anatomy [5-6].

Endoscopic arytenoid abduction lateropexy (EAAL) was objectively proved to be an effective, reversible 'simple suture procedure' to expand the glottis and avoid tracheotomy in neonates with BVCP [7-8]. The lateralization and fixation of the arytenoid cartilage to its normal abducted position, even in early childhood, is possible with the recently-introduced pediatric endolaryngeal thread guide instrument (ETGI; Mega Kft, Szeged, Hungary) [9]. As this is an age of rapid development in children, the long-term results and the stability of lateralization in the growing larynx is a concern. With the primary outcome measure being a durable resolution of dyspnea and with secondary outcomes focusing on adequate swallowing and voicing functions, herein, we report on 3 children, each with >3 years follow up after EAAL.

MATERIALS AND METHODS

Patients

In 2011-2018 congenital bilateral vocal cord paralysis (BVCP) was diagnosed in eight newborns. They had been admitted to the perinatal intensive care unit (PICU) immediately after birth due to severe stridor and inspiratory dyspnea. In this study the first three cases were chosen in which the postoperative follow-up was longer than three years. Unfortunately, one boy (over 40 months of follow up) was transferred out of the region and could not return for evaluation; according to parent's opinion he was 'symptomless'. The remaining four children had considerably less follow-up.

Unilateral endoscopic arytenoid lateropexy was performed on the 5th, 5th and 19th day of life, respectively.

Surgical technique (Fig 1)

Endoscopic arytenoid abduction lateropexy was performed as described in our earlier publications [7,9]. All operations were performed under general anesthesia via total intravenous anesthesia and high frequency supraglottic jet ventilation.

After disinfection of the laryngeal mucosa with Betadine, the ETGI was passed through the laryngoscope to the glottic level (7). The mobile arytenoid cartilage was tilted backward and upward with the end of the instrument (**Fig 2**). The built-in, curved blade was then pushed through, under the vocal process, and out to the surface of the neck. A nonabsorbable suture thread (0-Prolene; Ethicon, Somerville, NJ) was laced through the hole at the tip of the blade by an assistant surgeon. The doubled-over thread was pulled back with the blade, into the laryngeal cavity. After a repeated tilting of the arytenoid cartilage, the blade was pushed out with the thread above the vocal process to the outer surface of the neck. The assistant surgeon then cut the double-folded thread to remove it from the blade tip. The blade was then pulled back into the laryngeal cavity, and the ETGI could be removed. A small skin incision was then created to withdraw the ends of the thread by a Jansen hook to the surface of the sternohyoid muscle. The corresponding ends were knotted above it.

At the end of the surgery, the infants remained intubated for 5-7 days with uncuffed tracheal tubes. Parenteral antibiotic (amoxicillin/clavulanic acid, 25 mg/5 mg/kg/12 hours) was administered for 4 days, and methylprednisolone (4 mg/kg) was administered for 7 days. Nasogastric feeding was used in all patients while intubated. The postoperative management took place in the PICU in each case.

Follow-up

The functional outcomes of the surgery in terms of breathing, voice, swallowing, were evaluated and recorded. Anatomic follow up evaluations included regular endoscopic examinations (1^{st} postoperative week, 2^{nd} month, 6^{th} month and later twice per year) under general anesthesia using a rigid 0° and 30° endoscope. The grade of the vocal cord movement recovery was noted. Growth in height and weight were recorded over time. At each visit, parents were asked to describe any swallowing difficulty. The voice samples were recorded with a high sensitivity (40Hz-16kHz) condenser head microphone (Audio-Technica ATM75) at a sampling frequency of 96 kHz, 24 bit (Tascam US 122MkII external soundcard) and

analyzed by Praat® 5.3.2.9. software [www.praat.org]. The following acoustic parameters were recorded in this study: mean pitch, jitter, shimmer, and harmonics-to-noise ratio. Follow-up intervals were 38, 39 and 81 months, respectively.

RESULTS

No major perioperative or postoperative complications occurred. The babies could be extubated safely on the 5th, 7th and 7th postoperative day, respectively. In the follow-up period, the primary outcome measure—durable resolution of dyspnea—was observed in all three children. In terms of the secondary outcome measures that assessed voicing and swallowing, all patients were able to tolerate a normal *per os* diet. Growth and development were appropriate. The parents were satisfied with the postoperative outcome. Based on their observations, the voice of the patient was normal in 2 cases and slightly impaired in 1 case. Gurgling and cooing were similar to their siblings. Appropriate voice analysis was possible at the end of the observation period. The late postoperative results are shown in **Table 1**. Regular endoscopies showed stable lateralization (**Fig 3**). Partial regeneration of vocal cord movement was observed in 2 of the 3 cases: bilateral in patient #1 (right side abduction and adduction, left side adduction) and unilateral in #2 (right side abduction and adduction). Patient #3 showed no regeneration, and his postoperative voice was the worst. Lateralization sutures were not removed in any of the cases.

DISCUSSION

A ‘watch and wait policy’ is often recommended after establishing the diagnosis of bilateral vocal cord palsy in early childhood [10]. However, the associated airway limitations significantly impair normal growth and development of the child. Children initially managed with non-invasive positive pressure ventilation are confined to the intensive care unit and may be unable to feed *per os*. In cases of severe dyspnea, and when non-invasive positive pressure ventilation fails, an urgent surgical intervention is necessary. In a meta-analysis of vocal cord paralysis cases, Jabbour et al catalogued the clinical course and outcomes of 102 pediatric BVCP cases. The authors determined that tracheostomy was the most frequently performed surgical intervention, despite its many well-known risks. Tracheotomy was deemed

unavoidable in 69% of cases [11,12]. Alternatives to tracheotomy in the vulnerable, confined neonatal airway are much more limited compared to those in adults. Open or endoscopic cartilage augmentation of the posterior cricoid is technically challenging in the neonate, and the efficacy of partial/total arytenoidectomy and transverse cordotomy is questionable [13-15].

The optimal surgical intervention for congenital BVCP would provide an immediately adequate airway without ruining the anatomical structures bounding the glottis, and the operation would be reversible. Our preliminary results showed that EAAL can be relatively easily and quickly performed with low surgical stress even in the first days of life. With the use of supraglottic jet ventilation, pediatric laryngoscopes and the miniaturized, pediatric endoscopic thread guide instrument (ETGI), precise maneuvering and positioning of the lateralization suture in the narrow laryngeal space of the newborns was possible [7].

During a minimum of three years of observation (in patient #3 81 months), the lateralization was stable, and no medialization occurred. The patients' growth and development were normal according to growth charts. The larynges were also growing [16-17], and with laryngeal growth, we expected that dyspnea due to medialization could have happened. Were medialization to occur, the old sutures could have been easily removed and replaced with new ones in a correct position via repeat EAAL. Regular endoscopic assessments were performed, but, fortunately, no such intervention was required.

In the literature, the rates of vocal cord movement recovery range widely, from 8% to 82% [4,11,18]. Our patients had idiopathic BVCP with no history of birth trauma, cardiac surgery or neurologic disorders. Our patients did not have 'dead' larynges. In one patient no movement recovery was found, but in two cases vocal cords showed recovery—specifically, one patient had bilateral almost complete recovery, and one had unilateral adduction recovery.

Thus, the primary and secondary outcomes of this study were satisfied. Surgeons called on to improve the glottic airway in children with BVCP should value the non-destructive nature of EAAL over cordotomy/arytenoidectomy operations and the technical simplicity of EAAL over cartilage expansion of the posterior cricoid. In our series, all parents were satisfied with the postoperative quality of life. As phoniatry tests proved the patients' voices were socially acceptable, they had no problem with speech development. Finally, as the unilateral lateralization suture did not cause any swallowing or phonation impairment, it was not removed after partial functional recovery. This was in accord with the parents' decision.

CONCLUSION

Endoscopic arytenoid abduction lateropexy (EAAL) represents a favorable solution for neonatal bilateral vocal cord paralysis (BVCP). This surgical intervention may represent a long-term solution, even in fast-growing laryngeal structures.

FIGURES

Figure 1.: Endoscopic arytenoid abduction lateropexy performed on 5th day of life (patient #2, m)

A: Intraoperative picture: Bilateral vocal cord paralysis

B: Intraoperative picture: left-sided arytenoid lateropexy

C: Late postoperative endoscopy (>3 years): left-sided lateropexy, proper airway

Figure 2.: Endoscopic arytenoid abduction lateropexy performed by ETGI

A: Endoscopic Thread Guide Instrument with stem-pipe and blade designed for infants

B: Intraoperative picture: The mobile arytenoid cartilage was tilted backward and upward with the end of the instrument

C: Schematic picture of EAAL

Figure 3. Late postoperative endoscopic pictures

A: Patient #1 (36th postoperative month)

B: Patient #2 (38th postoperative month)

C: Patient #3 (77th postoperative month)

Table 1. Late postoperative results of EAAL

REFERENCES

1. Benjamin JR, Goldberg RN, Malcolm WF. Neonatal vocal cord paralysis. Neo Reviews 2009:10(10);494-501.

2. Takamatsu I. Bilateral vocal cord paralysis in children. Nihon Jibiinkoka Gakkai Kaiho 1996;99(1):91-102.

3. Gentile RD, Miller RH, Woodson GE. Vocal cord paralysis in children 1 year of age and younger. Ann Otol Rhinol Laryngol 1986;95:622-625.

4. Miyamoto RC, Parikh SR, Gellad W, Licameli GR. Bilateral congenital vocal cord paralysis: a 16-year institutional review. Otolaryngol Head Neck Surg. 2005;133(2):241-245.

5. Funk RT, Jabbour J, Robey T. International Journal of Pediatric Otorhinolaryngology Factors associated with tracheotomy and decannulation in pediatric bilateral vocal fold immobility. *Int J Pediatr Otorhinolaryngol*. 2015;79(6):895-899.

6. Lesnik M, Thierry B, Blanchard M, Glynn F, Leboulanger N. Idiopathic Bilateral Vocal Cord Paralysis in Infants : Case Series and Literature Review. 2015;(July):1724-1728.

7. Rovó L, Madani S, Sztanó B, Majoros V, Smehák G, Szakács L, Jóri J. A new thread guide instrument for endoscopic arytenoid lateropexy. Laryngoscope 2010;120(10):2002-2007.

8. Szakács L, Sztanó B, Matievics V, Bere Zs, Bach Á, Castellanos PF, Rovó L. A comparison between transoral glottis-widening techniques for bilateral vocal fold immobility. Laryngoscope 2015 ;125(11):2522-2529.

9. Madani S, Bach Á, Matievics V, Erdélyi E, Sztanó B, Szegesdi I, Castellanos PF, Rovó L. A new solution for neonatal bilateral vocal cord paralysis: Endoscopic arytenoid abduction lateropexy. Laryngoscope. 2017 Jul;127(7):1608-1614.

10. Chen EY, Inglis AF Jr. Bilateral vocal cord paralysis in children. Otolaryngol Clin North Am 2008;41(5):889-901.11. Jabbour J, Martin T, Beste D, Robey T. Pediatric vocal fold

immobility: natural history and the need for long-term follow-up. JAMA Otolaryngol Head Neck Surg. 2014 May;140(5):428-33

12. Jabbour J, North LM, Bougie D, Robey T. Vocal Fold Immobility due to Birth Trauma: A Systematic Review and Pooled Analysis. Otolaryngol Head Neck Surg. 2017 Dec;157(6):948-954.

13. Aubry K, Leboulanger N, Harris R, Genty E, Denoyelle F, Garabedian EN. Laser arytenoidectomy in the management of bilateral vocal cord paralysis in children. Int J Pediatr Otorhinolaryngol 2010;74(5):451-755.

14. Inglis AF, Perkins J a, Manning SC, Mouzakes J. Endoscopic posterior cricoid split and rib grafting in 10 children. *Laryngoscope*. 2003;113(11):2004-2009

15. Yawn RJ, Daniero JJ, Gelbard A, Wootten CT. Novel application of the Sonopet for endoscopic posterior split and cartilage graft laryngoplasty. *Laryngoscope*. 2016;126(4):941-944.

16. Litman RS[1], Weissend EE, Shibata D, Westesson PL. Developmental changes of laryngeal dimensions in unparalyzed, sedated children. Anesthesiology. 2003 Jan;98(1):41-5.

17. Wani TM, Bissonnette B, Malik MR, Hayes D, Ramesh AS, Al Sohaibani M, Tobias JD. Age-Based Analysis of Pediatric Upper Airway Dimensions Using Computed Tomography Imaging. Pediatric Pulmonology 2016; 51:267–271.

18. Takamatsu I. Bilateral vocal cord paralysis in children. Nihon Jibiinkoka Gakkai Kaiho. 1996 Jan;99(1):91-102

Patient /sex	EAAL (days after birth)	Follow-up (months)	Growing percentiles (%) Length-for-age/weight-for-age	Jitter (%)	Shimmer (%)	HNR (dB)	Pitch (Hz)
#1/f	5th	38	50/25	1,1	5,6	9,9	280,3
#2/m	5th	39	25/10	1,5	7,3	16,8	386,4
#3/m	18th	81	10/3	0,9	2,5	14,7	335,3

Table 1. Late postoperative results of EAAL