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Case Report

Recurrent laryngeal nerve reinnervation for management of aspiration in a subset of children☆

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ABSTRACT

Pediatric aspiration is a multifactorial process that is often complex to manage. Recurrent laryngeal nerve (RLN) injury can cause glottic insufficiency and aspiration. We describe three cases of unilateral vocal fold paralysis resulting in aspiration and the successful use of the RLN reinnervation for its treatment.

The theory for utilizing the reinnervation procedure is that when glottic closure improves and a less breathy vocalization occurs, then the larynx is better equipped to protect the lower airway and avoid aspiration. Our cases demonstrate stronger voice and improved swallow function, with normalization of modified barium swallow evaluation, at approximately 6-months post reinnervation.

1. Introduction

Pediatric aspiration is a multifactorial process that is often complex to manage. In a subset of children, it is due to a unilateral vocal fold paralysis, and the recurrent laryngeal nerve reinnervation (RLN) surgery may be indicated. It can be useful in a child with recurrent aspiration pneumonias and persistent aspiration in whom conservative measures (feeding therapy, dietary modifications and enteric feeding) have been unsuccessful.

This is a report of three pediatric patients who successfully underwent reinnervation surgery for the primary indication of aspiration with dysphonia. Chronic denervation was confirmed with laryngeal electromyography. Details for all case can be found in Tables 1 and 2.

2. Case reports

2.1. Case #1

A 5-year-old full-term male underwent two atrial-septal defect (ASD) repairs, a Ross procedure - bicuspid aortic valve repair, coarctation of aorta repair, and a patent ductus arteriosus (PDA) ligation. He presented to Children's Hospital of Philadelphia at 3 years of age with chronic dysphonia, persistent aspiration of thin liquids on modified barium swallow (MBS) and a left vocal fold immobility. He previously underwent an injection laryngoplasty at his home institution. The injection did not benefit his aspiration nor the voice, and he was referred for consideration of a reinnervation. At presentation, he was on a honey-thick diet, and a stroboscopy revealed a glottic gap.

In May 2015, he underwent a laryngeal electromyography (EMG) and was found to have chronic left thyroarytenoid (TA) muscle denervation. Prolaryn gel (Merz, Raleigh NC) (0.3mL) was injected to temporarily plump his left vocal fold, and he underwent a reinnervation of the recurrent laryngeal nerve utilizing a sternothyroid branch of the ansa cervicalis. The plan was to repeat a swallow study when his voice starts getting stronger.

A swallow study done 6 months postoperatively showed no aspiration, and he was advanced to a non-restricted diet. One month later he was seen by his local academic pediatric otolaryngologist and a laryngoscopy revealed complete glottic closure. Analysis of home-based acoustic recordings revealed steady improvement of voice quality and function and at 22 months post-reinnervation he was judged to have a normal conversational speech with no episodes of aspiration or aspiration pneumonia (Table 2). Table 3 shows the normative values for the acoustic parameters [3–6].

2.2. Case #2

This is a 10-year-old female with a history of 27-week gestation, prolonged QT syndrome, PDA ligation at birth, bronchopulmonary dysplasia, gastroesophageal reflux, and recurrent pneumonias which started at 8 years of age. All illnesses required hospitalization with antibiotic therapy, and a modified barium swallow study revealed silent

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Table 1
Demographics, medical and surgical history.

Case	Gender	Age (yrs.)	Gestational Age	Medical & Surgical History	Age at Injury	MBS Pre-Reinnervation	Preoperative Laryngoscopic Findings
1	Male	5	Full Term	ASD repair x2; Ross Procedure; PDA ligation	Birth	Silent aspiration of thin liquids	Left VFI
2	Female	10	27 weeks	PDA ligation, pacemaker for Prolonged QTc syndrome	Birth	Silent aspiration of thin liquids No penetration or aspiration with nectar thick liquids and puree	Left VFI
3	Female	6	30 weeks	22Q deletion syndrome, Interrupted AoA with VSD repair; Right Hemidiaphragmatic Paralysis	Birth	Frank aspiration during thin liquid intake and nectar thick liquids	Left VFI, synkinetic motion of the left vocal fold and left arytenoid

ASD = Atrial-Septal Defect; PDA = Patent Ductus Arteriosus; VSD = Ventricular Septal Defect; AoA = Aortic Arch MBS = Modified Barium Swallow (also known as Video Swallow Study); VFI = Vocal Fold Immobility.

Table 2
Perceptual, acoustic and swallow function pre-op and post-ANSA.

Perceptual rating includes Grade-Roughness-Breathiness-Aesthenia-Strain (GRBAS) and parental proxy questionnaire Pediatric Voice Handicap Index (pVHI). Acoustic measures include Maximum Phonation Time (MPT) on /a/, Physiological Fundamental Frequency range (vocal range) in semitones (ST), perturbation (jitter%, shimmer%) on sustained vowel, and Yanagihara Hoarseness rating from spectrogram. Swallow function includes Modified Barium Swallow (MBS) results.

	Case#1	Case#2	Case#3
GRBAS (0 = normal and 3 = severely abnormal for each parameter); maximum sum score = 15	Pre-RLN: 2-2-2-2-0 (sum 8) Post-RLN (1 mos) = 1-1-1-0-0 (sum 3) Post-RLN (6 mos): 0-0-1-0-0 (sum 1) Post-RLN (22 mos) = 0-0-1-0-0 (sum 1)	Pre-RLN: 2-2-3-2-2 (sum 11) Post-RLN (1 mos): 0-1-0-1-0 (sum 2) Post-RLN (6 mos): 1-0-1-2-0 (sum 4) Post-RLN (10 mos):1-1-1-1-0 (sum 4)	Pre-RLN: 3-3-1-2-3 (sum 12) Post-RLN (4 mos): 1-1-1-1-1 (sum 5) Post-RLN (16 mos): 1-0-2-1-1 (sum 5)
Pediatric Voice Handicap Index	Pre-RLN = 9 Post-RLN (22 mos) = 12	Pre-RLN = 1 Post-RLN (1 mos) = 8 Post-RLN (4 mos) = 6 Post-RLN (10 mos) = 5	Pre-RLN = 59 Post-RLN (4 mos) = 64 Post-RLN (16 mos) = 53
MPT in seconds (secs)	Pre-RLN = 3 secs	Pre-RLN = 6 secs Post-RLN (1 mos) = 5 secs Post-RLN (6 mos) = 5 secs Post-RLN (10 mos) = 9 secs	Pre-RLN = 2 secs Post-RLN (4 mos) = 7 secs
Vocal range in semitones (ST)	Pre-RLN = 20 ST Post-RLN (6 mos) = 29 ST Post-RLN (22 mos) = 25 ST	Pre-RLN = 19 ST Post-RLN (1 mos) = 29 ST Post-RLN (6 mos) = 26 ST Post-RLN (10 mos) = 25 ST	Pre-RLN = 24 ST Post-RLN (4 mos) = 29 ST
Perturbation	Pre-RLN: could not assess Post-RLN (6 mos): Jitter 3.549%, Shimmer 5.428%	Pre-RLN: Jitter 2.755%, Shimmer 8.668% Post-RLN (1 mos): Jitter 3.881%, Shimmer 9.074% Post-RLN (6 mos): Jitter 1.907%, Shimmer 5.935% Post-RLN (10 mos) = Jitter 2.023%, Shimmer 5.662%	Pre-RLN: Jitter 6.579%, Shimmer 12.029% Post-RLN (4 mos): Jitter 2.859%, Shimmer 4.506%
Yanagihara spectrogram (0 = normal, 4 = severely abnormal)	Pre-RLN = 3 Post-RLN (1 mos) = 3 Post-RLN (6 mos) = 0 Post-RLN (22 mos) = 1	Pre-RLN = 3 Post-RLN (1 mos) = 3 Post-RLN (6 mos) = 3 Post-RLN (10 mos) = 3	Pre-RLN = 4 Post-RLN (4 mos) = 2 Post-RLN (16 mos) = 3
Postoperative MBS	Normal at 6 months post-RLN	Normal 4 months post-RLN - Advanced to unrestricted diet at 5 months post-RLN	Normal MBS at 16 months post-RLN
Reinnervation technique	Sternothyroid branch to Ansa Cervicalis +0.3mL Prolaryn Gel. No laryngeal cleft identified.	Omohyoid branch to Ansa Cervicalis +0.2mL Prolaryn Gel. No laryngeal cleft identified	Omohyoid branch to Ansa Cervicalis +0.05mL Prolaryn Plus into small interarytenoid notch. No vocal fold injection

GRBAS = Grade, Roughness, Breathiness, Aesthenia, Severity; MPT = maximum phonation time; ST = Semitones; MBS = Modified Barium Swallow; RLN = recurrent laryngeal nerve reinnervation.

Table 3
Normal values for six acoustic parameters [3–6].

	Normal Values
GRBAS	0-0-0-0-0 = Σ 0
Physiological Frequency Range (PFR) (highest to lowest frequency, in semitones)	26-34 ST
Perturbation on /a/(jitter%, Shimmer%)	Jitter 1.24% Shimmer 3.35%
Yanagihara Spectrogram rating	0
Maximum Phonation Time	10.0 sec (sd 2.5 sec)
Pediatric Voice Handicap Index	Σ ≤ 2

GRBAS = Grade, Roughness, Breathiness, Aesthenia, Severity; ST = semitone range; sec = seconds; sd = standard deviation.

aspiration of thin liquids with mild-moderate pharyngeal dysphagia characterized by inadequate laryngeal closure patterns. No penetration or aspiration was seen with nectar thick liquids and puree. The etiology of the dysphagia was judged to be due to reduced pharyngeal/laryngeal sensation in the setting of reflux and the presence of a chronic left vocal fold immobility. She was started on nectar thickened liquids, reflux medication and referred to otolaryngology. She had difficulty tolerating the Thick-It® (Kent Precisions Foods Group, MO) and the limited diet.

On clinical assessment, she was noted to have a breathy phonation and a left vocal fold immobility with a visible glottic gap. Due to the silent aspiration and inability to tolerate the thickened feeds, an injection laryngoplasty was recommended with consideration for a recurrent laryngeal nerve reinnervation to address the breathy voice and