



Pediatric Unilateral Vocal Fold Paralysis: Workup and Management

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Abstract

Purpose of Review This paper will examine recent advances in the assessment and management of pediatric unilateral vocal fold motion impairment (VFMI).

Recent Findings Unilateral VFMI can have a considerable impact on pediatric voice, swallowing, and pulmonary hygiene. In addition, postoperative VFMI in children undergoing congenital cardiac procedures is associated with longer lengths of stay. Laryngeal ultrasound (LUS) is an adjunctive tool that can be used to assess vocal fold mobility in patients who cannot tolerate traditional flexible nasolaryngoscopy (FNL) or for those in whom FNL is non-diagnostic. Laryngeal electromyography (LEMG) can help differentiate neuronal causes of vocal fold paralysis from mechanical cricoarytenoid joint fixation. Recent data suggests that preoperative LEMG may be used to predict voice outcomes after non-selective laryngeal reinnervation (NSLR). NSLR is a promising treatment modality for permanent vocal fold medialization in children.

Summary An emphasis on early diagnostic and prognostic techniques, such as the use of LUS and LEMG, can inform decision-making in unilateral VFMI treatment. NSLR is emerging as the preferred treatment modality for pediatric unilateral VFMI due to neuronal injury.

Keywords Non-selective laryngeal reinnervation · Laryngeal electromyography · Vocal fold motion impairment · Pediatric · Vocal fold paralysis · Vocal cord paralysis · Laryngeal ultrasound

Introduction

Vocal fold motion impairment (VFMI) describes any abnormal or restricted movement of the vocal fold, regardless of etiology. Unilateral VFMI may result in glottic insufficiency leading to dysphonia and increased risk of swallowing dysfunction. In addition to flexible nasolaryngoscopy (FNL) or transoral laryngoscopy, laryngeal ultrasound (LUS) and laryngeal electromyography (LEMG) can be used as adjunctive

procedures to diagnose and characterize VFMI. Non-selective laryngeal reinnervation (NSLR) has also gained popularity among pediatric otolaryngologists for the treatment of unilateral VFMI due to neuronal injury.

Impact

Unilateral VFMI can have a complex impact on voice, swallowing, and pulmonary hygiene, and in turn, negatively impact a child's quality of life.

Voice

VFMI and the resultant glottic incompetence can result in a breathy or weak voice. Older children may also report vocal fatigue. Dysphonia negatively impacts a child's essential ability to communicate, which can elicit negative personality perceptions by their non-dysphonic peers [1]. In a study based on voice recordings alone, dysphonic children were attributed negative physical, social, and personal characteristics by

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adults and peers. Fear of negative opinions by others can cause children to become hesitant to engage in speaking and stunt overall speech development [2].

The advent of various validated pediatric voice outcome measures such as the Pediatric Voice-Related Quality of Life (PVRQOL) and the Pediatric Voice Handicap Index (pVHI) highlights the growing social-emotional impact of a child's self-perception of their voice [3–5]. Previously, young children were hypothesized to be unaware of their dysphonia with earlier voice-related quality of life surveys focused solely on parental perception of their child's voice [1]. However, there has been a shift to incorporate child interviewing to determine the impact of self-perception of voice. This has led to a greater understanding of the emotional and psychological stigma associated with childhood dysphonia. Dysphonic children who are undergoing a rapid phase of physical and social development with negative perceptions of self or by peers can lead to social isolation, frustration, shame, and overall decreased quality of life [1].

Swallowing and Pulmonary Hygiene

Swallowing is a complex coordinated mechanism involving multiple muscles and nerves to pass a bolus from the oral cavity to the esophagus. Along this pathway, the vocal folds provide protection against aspirated material that may intrude into the airway. Although the vocal folds do not solely facilitate safe swallowing, VFMI and glottic incompetence can compromise airway protection, and is linked to aspiration pneumonia, recurrent pulmonary infections, need for hospitalizations, and malnutrition.

In a study of over 400 pediatric patients with VFMI, as many as 50% of participants presented with dysphagia [6]. Unlike the adult population where patients can largely communicate symptoms of choking or coughing with eating, recognizing and obtaining a history of dysphagia and aspiration in children can be challenging. With a non-verbal population, such as neonates or young children who are unable to adequately express themselves, silent aspiration is more common than overt aspiration. Over half of the patients in a retrospective chart review of 28 patients aged 0.4–4.3 years old at a single institution with unilateral VFMI had silent rather than overt aspiration [7]. It is difficult to quantify the incidence of aspiration in the pediatric population as postoperative laryngeal exams are usually not performed unless patients are symptomatic. Underdiagnosis can lead to untreated recurrent respiratory infections and prolonged hospitalizations.

Understanding the implications of dysphagia and aspiration is paramount in children undergoing congenital heart surgery where unilateral VFMI is a common postoperative complication [6, 8]. In a population-based study of neonates undergoing congenital heart surgery, 6.9% ($n=3725$) had VFMI [9]. The circuitous route of the left recurrent laryngeal nerve

around the aortic arch leaves the nerve vulnerable to strain or compression from manipulation of the aortic arch during congenital heart procedures. Management of dysphagia secondary to unilateral VFMI requires coordination of alternative forms of nutrition through enteral feeding or modified diets, which can increase postoperative length of hospitalization [9, 10]. In a retrospective review of 76 neonates undergoing cardiac surgery, 94% of patients without unilateral VFMI were discharged on a regular diet by mouth compared to only 50% of patients with unilateral VFMI who were discharged on regular diet. Additionally, neonates in this study with unilateral VFMI had an average 12-day longer hospital stay than those without unilateral VFMI [10]. Neonates with VFMI after congenital heart surgery also have a greater incidence of readmission for poor weight gain and feeding difficulties [11]. Chronically, dysphagia has been reported to lead to lung injury, oral aversion, and refusal behaviors [12]. This highlights the importance of postoperative nutrition and feeding plan counseling in the neonatal population to prevent prolonged hospitalizations.

Workup

Most causes of unilateral VFMI can be attributed to mechanical joint fixation, neuronal injury, or a neoplastic process. Proper workup to establish the etiology is key to determine the appropriate treatment strategy.

Diagnosis of unilateral VFMI can be made via office laryngoscopy: either FNL or transoral rigid laryngoscopy. Visualization can provide an excellent view of laryngeal structures and dynamic vocal fold movement; however, in-office laryngoscopy can be difficult in young children.

Imaging

Ultrasound is a non-invasive imaging modality that requires little patient cooperation. This procedure is well tolerated with data supporting its reliability as a medium to diagnose VFMI. With pediatric cadaveric and normal functioning larynxes as controls, Wang et al. [13] utilized maximum glottic angle and vocal fold-arytenoid angle to successfully diagnose VFMI via ultrasound. In pediatric patients, laryngeal ultrasound is a comparable alternative to FNL in the evaluation of VFMI with fewer effects on physiological parameters. In a case control study of 46 infants from a cardiovascular intensive care unit, there was considerable agreement between ultrasound and FNL findings in diagnosis of VFMI. Physiologically, laryngeal ultrasound had fewer changes on oxygen saturation, blood pressure, and pulse rate [14•]. The decreased physiological impact of laryngeal ultrasound compared to FNL is beneficial in single ventricle and pulmonary hypertensive patients where rapid changes in these vitals can lead to devastating

health consequences [14•]. Ultrasound image quality is user-dependent and can be challenging in adults with calcified thyroid cartilages or in children with tracheostomy tubes [15].

Non-invasive imaging techniques such as computerized topography (CT) and magnetic resonance imaging (MRI) can be useful in determining the etiology of VFMI when history and physical exams do not provide a clear clinical picture. However, CTs and MRIs cannot perceive mobility and are less useful for visualization of the dynamic glottis. Imaging may also expose children to radiation and possibly sedation.

Laryngeal Electromyography

First introduced in 1957, laryngeal electromyography (LEMG) is a valuable diagnostic tool for laryngeal neuromuscular disorders [16]. In adults, LEMG has been used for prognosis for vocal fold movement recovery after neuronal injury by assessing recruitment, waveform morphology, spontaneous activity, and presence of synkinesis [17]. Unfortunately, true, voluntary, LEMG is usually not possible in children. Thus, pediatric LEMG is typically performed in the operating room, under general anesthesia, with a tubeless spontaneous ventilation technique. A monopolar needle or fine wire electrode is placed in the posterior cricoarytenoid muscles on each side and motor unit potentials measured with laryngeal abduction observed with respiration. The thyroarytenoid (TA) muscles can also be assessed for synkinetic firing with respiration. Insertional and spontaneous activity, such as fibrillation potentials and positive sharp waves, can be appreciated.

Ongkasuwan et al. [18•] and Smith and Houtz [19] both found that children with preoperative LEMG's with evidence of denervation had greater improvements in objective voice measures after NSLR when compared to those with evidence of spontaneous (though unfavorable) reinnervation [18, 19]. While the mechanism is not entirely clear, the authors speculate that in children with evidence of denervation, the motor endplates may be occupied by collateral sprouting from the superior laryngeal nerve or contralateral RLN and thus not be available for the reinnervated ansa cervicalis nerve fibers. This preliminary research suggests that LEMG may be used to predict voice outcomes with NSLR.

In 2019, Caloway et al. [20•] performed intraoperative evoked vagal EMG with hook wire electrodes in the TA muscle on three patients during NSLR [20•]. Two of the three individuals had either normal amplitude and/or normal latency intraoperatively. These two patients had resolution of dysphagia and improvement in dysphonia 6 months postoperatively. The patient with both low amplitude and prolonged latency signals had continued dysphagia, and reported a suboptimal voice result [20•]. The authors posit that evoked vagal EMG may be useful to determine NSLR candidacy in the operating room.

Management

Management of unilateral VFMI is best achieved through a collaborative approach with speech language pathology. In addition, these patients often have comorbid conditions necessitating collaboration with other specialties including pulmonology, gastroenterology, and cardiology.

Non-Surgical Interventions

Because a majority of iatrogenic unilateral VFMI will self-resolve, an observation period of 12–24 months is recommended prior to surgical interventions [6, 21]. A literature review of 717 cases of adult unilateral VFMI revealed 25–87% of patients experienced some to complete recovery within 12 months [22]. In a 2017 systematic review of unilateral VFMI secondary to birth trauma, 63% of this group had spontaneous recovery and fared well with observation alone [23].

In infants, the clinical impact of glottic incompetence is primarily on swallow. Feeding and swallow therapy with an experienced speech language pathologist is essential. As children develop more language skills around age 2–3 years, voice quality and communication become a larger focus. Voice therapy techniques can be employed to help maximize voicing efficiency and address compensatory phonatory mechanisms such as supraglottic phonation. In older children particularly, postmedialization voice therapy may be needed to help the child adjust to the sound of their “new voice” which may seem foreign to them.

Surgical Interventions

Three main surgical interventions can be used to improve glottic closure: injection laryngoplasty, laryngeal framework surgery, and laryngeal reinnervation procedures.

Injection Laryngoplasty

Injection laryngoplasty is quick, outpatient, procedure which allows for temporary vocal fold medialization. The duration of effect can be 1 month to 1 year depending on the material used [24]. Of note, some longer lasting injectable materials such as hyaluronic acid and calcium hydroxylapatite can cause inflammatory reactions leading to edema, vocal fold stiffness, or airway obstruction [25].

In the adult population, some authors suggest that early injection laryngoplasty (within 6 months of injury) may influence vocal fold positioning and aid with synkinetic reinnervation, decreasing the need for further surgical interventions [26, 27]. While, in adults, injection laryngoplasty can be performed at the bedside or in the office, young children typically require general anesthesia. Repeated injection laryngoplasty is not a viable long-term solution in the pediatric population,

especially given the potential neurocognitive effects of repeated anesthetics in children under age three [28].

Laryngeal Framework Surgery

Type 1 thyroplasty, with or without arytenoid adduction (to close the posterior glottis), involves placing an implant lateral to the TA muscle and medial to the thyroid cartilage via an external approach. Ideally, the procedure is performed with the patient awake and phonating to determine the optimal implant size and placement. However, most children cannot tolerate an awake approach limiting the surgeon's ability to "tune" the implant. Laryngeal framework surgeries are static medialization procedures and do not address vocal fold atrophy which can continue over time. In addition, the implant does not grow with the child and may require surgical revision as the child goes through puberty.

Laryngeal Reinnervation Procedures

Laryngeal reinnervation was first described in 1924 and reported in the pediatric population in 2007 [29, 30]. This procedure involves the anastomosis of a nearby functioning nerve with the recurrent laryngeal nerve (RLN). The phrenic, hypoglossal, superior laryngeal, and ansa cervicalis have all been candidates for anastomosis. However, the ansa-RLN non-selective laryngeal reinnervation (NSLR) is currently the most popular approach [31].

NSLR does not result in vocal fold movement; rather, it medializes the vocal fold and restores muscle bulk and tone. In addition, NSLR can reestablish the mucosal wave, height, and arytenoid rotation, allowing closure of the posterior glottis. Barring additional open neck surgeries, NSLR results should be durable over time.

In 2015, Zur et al. [32] found better voice outcomes in patients after NSLR compared to injection laryngoplasty. The same year, a systematic review of 15 studies demonstrated that NSLR resulted in more sustainable, longer term voice results compared to static medialization with type 1 thyroplasty or injection laryngoplasty [33]. Unlike static medialization procedures, results with NSLR are not immediate. Voice change does not occur until roughly 4.5 months postoperatively. In the interim, some patients may benefit from temporizing injection laryngoplasty [34].

Larger single institution series have demonstrated that pediatric NSLR improves voice quality and can be replicated by surgeons of different institutions [18, 19]. Closing the glottic gap with NSLR can also improve dysphagia [35, 36]. Unlike the NSLR adult literature, there is no clear age cut off or time from RLN injury for NSLR [18, 19].

A survey of fellowship trained pediatric otolaryngologists in 2018 revealed practice shifts with 37% of the participants reflecting they would offer NSLR as first-time treatment for

unilateral neuronal VFMI. This was attributed to increased comfort level with the procedure itself [37]. Most recently, Caloway et al. [38] presented a discussion of surgical challenges in a cohort of 21 pediatric patients who underwent NSLR. This study's "lesson's learned" and surgical commentary highlights the growth in expertise and comfort with the NSLR procedure [38].

Conclusion

Unilateral VFMI has a complex impact on quality of life for children, warranting thorough evaluation and tailored treatment options. LUS and LEMG are excellent adjunctive tools that clinicians can add to their armamentarium. NSLR is emerging as the preferred treatment modality for pediatric unilateral VFMI due to neuronal injury.

Declarations

Conflict of Interest The authors declare no competing interests.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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