Paradoxical vocal fold motion (PVFM) in pediatric otolaryngology

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A B S T R A C T

Introduction: Paradoxical vocal fold motion (PVFM) is a condition in which the vocal cords exhibit inappropriate inspiratory adduction, and it has been poorly studied in the pediatric population.

Methods: Pediatric patients diagnosed with PVFM by a pediatric otolaryngologist and doctor of speech pathology from 2008 to 2012 were reviewed. Patients in whom another cause for their respiratory disturbance was eventually identified were excluded. Patient demographics, characteristics, treatment, and outcomes were reviewed.

The study was approved by the Institutional Review Board at our institution.

Results: Thirty patients met criteria for inclusion; one with a Chiari malformation was excluded. 17/29 (59%) were female. Body mass index (BMI) numbers ranged from 16 to 25 with a mean of 21. 9/29 (31%) competed at the highest level of a sport; only 3/29 (10%) did not participate in athletics. Average age of onset was 12.0 years; average diagnosis delay was 1.3 years. Mean follow up was 2.3 years. 24/29 (83%) were previously treated for reflux. 25/29 (86%) completed at least one session of respiratory and laryngeal control therapy with overall average of 2.2 sessions completed. All patients who attended a second therapy session were recorded as having improvement in symptoms.

Conclusions: Pediatric patients with PVFM often participate in high levels of organized sports and the frequency of concurrent asthma and reflux symptoms in this population supports the theory that laryngeal hypersensitivity contributes to the pathophysiology of PVFM. These patients were not found to have any associated psychiatric diagnoses. Pediatric patients with PVFM have an excellent prognosis when treated with speech therapy and for comorbid conditions as indicated.

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1. Introduction

Paradoxical vocal fold motion (PVFM) is a condition in which the true vocal cords exhibit inappropriate adduction during the inspiratory phase of the respiratory cycle resulting in inspiratory stridor, dyspnea, cough, and anxiety. The disorder was first described in 1842 by Robley Dunglison who described laryngeal spasms induced by “hysteria”. Literature from the 1970s further depicted a syndrome frequently mimicking asthma and carrying numerous misnomers including “Munchausen’s stridor” [1] and “factitious asthma” [2]. Rodgers and Stell first employed the more physiologic terminology “paradoxical vocal cord movement” in 1978 [3]. However, it is only in the last decade that terminology, diagnostic criteria, and treatment have begun to move toward a universal standard.

The true incidence and prevalence of PVFM in the general population has not been defined [4,5]. Likely due to limited awareness and challenges associated with diagnosis. While felt to be relatively uncommon, certain subgroups demonstrate significant prevalence. Newman et al. reported that 30% of patients diagnosed with refractory or persistent asthma had some component of PVFM, and 10% of the patients had PVCM as the sole cause of their symptoms [6]. A notable 2:1 predominance exists amongst female patients diagnosed with PVFM and studies suggest that about 30% of patients are under the age of 18 [4].

Symptomatology typically consists of dyspnea, stridor, throat tightness, voice changes, and cough [6,7,8]. Pediatric patients often have normal pulse oximetry, blood gas values, and chest radiographs [9]. Flow volume loops obtained during attacks may show a blunted inspiratory component. Patients frequently are non-responsive to methacholine and histamine challenges [9,10] and refractory to standard asthma treatment regimens. While diagnosis can be made based on history, flexible laryngoscopy with visualization of paradoxical vocal fold motion during inspiration has been considered by most the gold standard. However, the attacks are frequently unwitnessed by physicians, and the symptoms are often misdiagnosed as asthma. Patel et al. in 2004 reported an average time between symptom onset and diagnosis of over four years [5].

Misdiagnoses often result in inappropriately directed treatment which may include bronchodilators, corticosteroids, intubation, or even tracheostomy with associated side effects and iatrogenic

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morbid disorders with PVFM has been well described [5,6,9,11,12] and the treatment of such comorbid conditions is pivotal. However, speech language pathology (SLP) techniques and behavioral intervention have demonstrated widespread success in the treatment of the PVFM population [6,8,10,13] and remain the mainstay of treatment. Despite reports establishing the effectiveness of treatment with SLP approaches, these techniques have not been well described in the literature.

The purpose of this study was to characterize the typical pediatric patient with PVFM, review treatment methods used with specific focus on SLP techniques, and report success rates achieved using a multidisciplinary approach at a tertiary care facility.

2. Methods

Retrospective chart review was conducted of all pediatric patients, age ≤18 years, who had been diagnosed by a pediatric otolaryngologist and doctor of speech pathology over a 5-year period at a single tertiary care center. An initial cohort was identified by reviewing all patients diagnosed at the Cleveland Clinic Head and Neck Institute from August 2008 to August 2012 with ICD codes including shortness of breath (786.05), respiratory distress (786.09), laryngeal spasm (478.75), other forms of asthma (493.8), or paradoxical vocal fold motion (478.5). Patients who were ultimately diagnosed with PVFM by the senior author and a doctor of speech pathology either by clinical history or flexible laryngoscopy were included. Patients were excluded if the practitioner did not specifically state the diagnosis of PVFM or if another cause for their respiratory distress was eventually identified. The final cohort was then analyzed for demographics, coexisting conditions, and time from presentation of symptoms to diagnosis. Prescribed treatment regimens, compliance, and outcomes were also reviewed.

2.1. Speech language pathology techniques:

The therapeutic intervention aimed to relieve the exercise induced dyspnea, and increase exercise tolerance. The techniques used included

a) establishing abdominal breathing control
b) eliminating or decreasing muscle tension in the torso associated with high ventilatory output tasks
c) establishing forceful activation of the inspiratory muscles, while relaxing throat and laryngeal muscles. This is accomplished by a series of graduated breathing exercises using pursed lip inhalation and exhalation, which induces inspiratory resistance, and increases inspiratory muscle strength
d) Respiratory practices were introduced initially in static positions — supine and upright, gradually progressing into dynamic practice, coordinating breathing rhythm with walking, light jogging, and running. Adaptations were made to incorporate breathing control practices in different sports [14–18].

3. Results

Thirty patients with a diagnosis of PVFM were identified. One patient with chiari malformation was excluded. Seventeen (59%) of twenty-nine patients were female. The average BMI of our study group was 21 with a range of 16–25. Nine (31%) of twenty-nine patients participated in an organized sport at the most competitive level, while only three patients (10%) did not participate in a regular organized sport. The average age of symptom onset was 12.0 years and the average time until diagnosis was 1.3 years. The mean follow up duration was 2.3 years.

The majority of our patients (80%) were diagnosed based on clinical history alone, while six (20%) were observed having PVFM at the time of flexible laryngoscopy done immediately after exertion. Twenty-four (82%) patients of twenty-nine were treated for asthma prior to their diagnosis of PVFM. Additionally, twenty-three (79%) were placed on reflux regimen during the course of their treatment.

All patients were advised to undergo therapy with a doctor of speech language pathology. Twenty five (86%) completed at least one session with the mean number of sessions completed being 2.2 per patient. All patients who attended a second therapy session were recorded as having improvement in symptoms.

4. Discussion

The diagnostic and therapeutic challenges posed by PVFM stem from its incompletely understood pathophysiology. Historically, it has been characterized as a psychiatric condition. Newman reported in 1995 that over 70% of patients studied with PVFM had an associated psychiatric diagnosis [6]. One of the first large scale studies by Christopher et al. published in 1983 described the typical patient as a woman between the ages of 20–40 who worked in the medical field and had a history of physical or sexual abuse [19]. Maturo et al. reported in 2011 that 75% of their pediatric population had underlying psychiatric disorders and suggested that children with PVFM may be better treated with psychiatric therapy than speech therapy [12].

However, the picture has evolved as new studies are added to the literature. It has become evident that its cause is more likely multifactorial and generally a functional disorder as opposed to a purely a psychiatric or conversional disorder. In a large percentage of patients there seems to be an association with factors that may predispose the larynx to hypersensitivity like asthma, reflux disease, or chronic post nasal drainage. Powell et al. noted that over one third of the patients in his pediatric population had anatomic changes suggestive of reflux disease. Other studies have reported evidence of laryngeal inflammation in the general PVFM population to be as high as two thirds or more [9]. Similarly, Perkner has described a population that developed PVFM after exposure tonoxious volatile chemicals [20].

Additionally, exercise has been recognized as yet another potential instigator of PVFM and a recent review reported its prevalence as the cited underlying etiology at 18.2% [4]. This same group prospectively evaluated active military subjects with exertional dyspnea and reported 12% with PVFM [4]. Rundell et al. in 2003 noted a prevalence of 5% amongst 370 elite athletes evaluated [21].

Our series supports this trend with the vast majority of patients having associated asthma, reflux symptoms, and often both. Furthermore, the majority of our population participated in organized sports with over 30% being involved at the most competitive level. In contrast, none of our patients were noted to have an associated psychiatric diagnosis.

Successful management of PVFM with speech therapy has been reported as early as 1978. Since that time, numerous studies have demonstrated the efficacy of SLP in controlling symptoms. A recent literature review by Patel et al. in 2015 pointed out that 89% (54/61) of studies reporting on symptoms of PVFM demonstrated improvement or resolution following SLP [8]. Rameau et al. treated pediatric patients within a single multidisciplinary session consisting of an otolaryngologist, speech pathologist, and nurse practitioner and reported a success rate of 86.4% [13]. While most consider speech therapy to be the primary therapy for PVFM [4,5,8,22], descriptions of specific techniques have been few. In our study, all patients had resolution or improvement in symptoms within two sessions with SLP techniques and methodology employed by our doctor of speech pathology.

While the importance of identifying and treating underlying and comorbid disorders associated with PVFM is not to be underemphasized, the need for prompt and accurate diagnosis by either history or flexible endoscopy followed by appropriately targeted therapy is essential to patient safety, satisfaction, and cost utilization. Given the complex and multifactorial etiology of PVFM, it is the opinion of the authors that early intervention incorporating the care of a SLP is both safe and effective and should continue to be first line therapy.
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