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Diagnosis and treatment of paradoxical vocal fold motion in infants

Ashley P. O'Connell Ferster[a,*], Tom Shokri[a], Michele Carr[b]

[a] Department of Surgery, Division of Otolaryngology – Head & Neck Surgery, The Pennsylvania State University, College of Medicine, Hershey, PA, United States
[b] Department of Otolaryngology — Head and Neck Surgery, West Virginia University School of Medicine, Morgantown, WV, United States

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ABSTRACT

Importance: Paradoxical vocal fold motion (PVFM) is a disorder often misdiagnosed in children presenting with shortness of breath and stridor. In infants, little is known about the clinical course and best approach for treatment of PVFM. This retrospective study assesses the approach to treatment and outcomes for infants with PVFM.
Objective: To investigate the clinical course of paradoxical vocal fold motion (PVFM) in infants.
Design: Retrospective review.
Setting: Tertiary academic medical center.
Participants: Patients less than 2 years of age diagnosed with PVFM were identified and included in the study.
Main outcomes and measures: History, physical exam findings, and clinical course of treatment for patients less than 2 years old with PVFM were reviewed. Findings including those on flexible fiberoptic laryngoscopy (FFL) and subjective assessment by parents and clinicians were compiled for review.
Results: Seven infants were diagnosed with PVFM. All patients were full term at birth, and average age at diagnosis was 7 months. All patients initially presented with inspiratory stridor, and two patients had stertor. Two of seven patients also had a history of reactive airway disease and one with laryngomalacia. Five had a history of reflux. Two of seven patients had weight percentiles at diagnosis lower than the 25th percentile, while the remainder were between 37th and 75th percentiles. Initial voice evaluation revealed stridor in all patients, as well as finding of PVFM on FFL. All patients were started on anti-reflux medication. Average time to resolution of PVFM was 5.9 months after treatment.
Conclusions: PVFM can be challenging to diagnose in the infant population. PVFM resolves uneventfully with reflux treatment, however, it is unknown whether reflux treatment is essential or if PVFM would spontaneously resolve. The rarity of infantile PVFM mandates formal evaluation and monitoring by a pediatric otolaryngologist.

1. Introduction

Paradoxical vocal fold motion (PVFM), also known as vocal cord dysfunction (VCD), is the untimely adduction of vocal cords during inspiration resulting in obstruction at the level of the glottis. The width of the rima glottides, the space between vocal folds, is determined largely by vocal fold movement thereby determining air flow through the glottic aperture. During inspiration, the posterior cricoarytenoid muscle contracts resulting in abduction of vocal folds and widening of the rima glottides, while during expiration contracture of the lateral cricoarytenoid results in adduction of vocal folds and narrowing of the rima glottides. In PVFM, there is aberrant adduction of vocal folds during the act of inspiration [1,2]. On visualization of the vocal cords, there is a characteristic, yet uncommon, finding of adduction of the anterior two-thirds with a diamond-shaped gap or “chink” in the posterior aspect of the vocal folds [3,4]. Clinically, this manifests as resulting dyspnea and noisy breathing in the form of stridor.

Due to the similarity in presentation, this entity is often misdiagnosed as reactive airway disease. Therefore, patients are often subjected to extraneous testing and medication administration. Increased awareness has resulted in an increase in the reported incidence of this disorder. Although there have been several proposed classification schemes, to date little is known regarding the pathophysiology of PVFM.

We report seven infants evaluated and diagnosed with PVFM at a tertiary medical center. The goal of our study is to investigate the patterns of presentation and clinical course of infants with paradoxical vocal fold motion (PVFM). Although this disorder has been previously reported in the literature, few studies have described this entity within the pediatric subpopulation, specifically in infants.

* Corresponding author. The Pennsylvania State University, College of Medicine, Department of Surgery, Otolaryngology – Head & Neck Surgery, Hershey, PA 17033-0850, United States.
E-mail address: aoconnell2@pennstatehealth.psu.edu (A.P. O'Connell Ferster).

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2. Materials and methods

Approval to conduct this study was obtained from the Pennsylvania State University Institutional Review Board. Patients with a diagnosis of congenital paradoxical vocal fold motion were identified using ICD-9 and ICD-10 codes. All patients presented to the Pediatric Otolaryngology clinic at Penn State Health within the last 10 years and were evaluated by the senior author (M.C.).

Each patient presented with a history of noisy breathing which prompted further evaluation. Diagnosis of congenital paradoxical vocal fold motion was defined by the child's age at presentation being less than two years old. Paradoxical vocal fold motion was diagnosed based on findings of vocal fold adduction during inspiration and/or expiration on flexible fiberoptic laryngoscopic exam. Flexible fiberoptic laryngoscopy was performed on all patients by the senior author (M.C.). Clinical history and exam findings were reviewed. Voice quality at initial evaluation and follow-up visits was documented based on parent/guardian descriptions, as well as the evaluation in clinic. Findings from procedures and/or imaging studies performed during their diagnostic work-up were also recorded and reviewed via descriptive analysis of the patient population.

3. Results

A total of four male and three female infants were included. The average age at diagnosis was 7.03 months ± 0.64. All seven patients were full term at birth. Past medical history was significant for gastroesophageal reflux in five of seven patients. Feeding issues were documented in two of seven patients. Two of seven patients had a weight less than the 25th percentile; the remainder were between the 37th and 75th percentile.

All patients presented with a history of stridor as reported by parents/guardians. Five of the seven patients presented with complaints of noisy breathing since birth; one patient did not have time of onset documented, and one patient had noisy breathing since 10 months of age. Inspiratory stridor was documented in clinic for six of seven patients. Flexible fiberoptic laryngoscopy revealed paradoxical vocal fold motion in all patients on initial evaluation. Other findings included those consistent with laryngopharyngeal reflux, including post-glottic edema and erythema. Two patients also had short aryepiglottic folds, and two other patients had findings of posterior laryngeal collapse. Vocal fold mobility was intact in all patients. Clinical history and exam findings for each patient are reviewed in Table 1.

4. Discussion

Both the prevalence and incidence of PVFM in the literature is unclear, particularly among infants. Paradoxical vocal cord motion in the pediatric population has been shown to be predominantly in teenagers with a 2:1 female predominance [1–3]. There have been few reports of infants with PVFM, including a case report by Heatley and Swift in 1996 reporting a 4 month old with diagnosed PVFM secondary to gastroesophageal reflux [5]. A second case study also noted PVFM in four infants who were originally, incorrectly diagnosed with bilateral vocal fold paralysis [6]. Although there are limited studies in the pediatric population, none have specifically addressed this clinical entity within the infant subpopulation, and, therefore, little is known of the disease process within this age group.

Several classification schemes have been proposed, but the specific pathophysiology underlying PVFM remains unclear [7]. Christopher and Morris described three categories of PVFM: psychogenic, exertional, and irritant-associated [1]. Historically, PVFM was solely attributed to psychological causes. Although this theory has since been revised, there continues to be a documented association between psychiatric disorders and PVFM. The proportion of comorbid psychiatric disorders in patients with PVFM has been documented as ranging

Table 1
Descriptive data for infants diagnosed with paradoxical vocal fold motion.

Patient ID No.	Age at Diagnosis	Weight Percentile at Diagnosis	Feeding Issues (Y/N)	Past Medical History	Voice Quality (Initial)	In-office FFL Findings	Rx for Reflux Meds (Y/N)	Time to resolution
1	1 week[b]	67.4	Y	Reflux	Inspiratory stridor, hoarse cry	PVFM, Short aryepiglottic folds, mild epiglottic retroflexion, mild VF edema	Y	1.5 months
2	10 months[b]	44.4	Y	Reflux, Pierre Robin, Speech Delay	Inspiratory stridor	PVFM, short aryepiglottic folds, omega epiglottis, mild post-cricoid edema	Y	10 months
3	2 months[b]	22.6	N	Reflux, Neonatal rhinitis, choanal stenosis	Inspiratory stridor, stertor at rest	PVFM, mild post-glottic edema, bilateral choanal stenosis	Y	6 months
4	15 months	75.0	N	Reflux, Asthma	Inspiratory stridor	PVFM, post-glottic edema and erythema	Y	N/A[a]
5	1 month[b]	7.0	N	Reflux, Laryngomalacia, Meconium Aspiration	Inspiratory stridor	PVFM, post-glottic edema, posterior laryngeal collapse	Y	N/A[a]
6	19 months	37.0	N	Developmental delay, Asthma	Quiet inspiratory sound without stridor	PVFM, posterior laryngeal collapse	Y	6 months
7	2 months[b]	53.88	N	Bilateral polydactyly	Inspiratory stridor	PVFM, post-glottic edema and erythema	Y	LTF
Mean (± SD)	7.03 months (0.64)	43.90 (0.24)						5.88 months (3.47)

FFL = flexible fiberoptic laryngoscopy.
PVFM = paradoxical vocal fold motion.
LTF = lost to follow-up.
[a] N/A indicates that PVFM is not resolved and patient is still under treatment.
[b] Indicates noisy breathing present since birth.

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