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Pediatric Paradoxical Vocal-Fold Motion: Presentation and Natural History

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KEY WORDS

paradoxical vocal, vocal dysfunction, exercise-induced stridor

ABBREVIATIONS

PVFM—paradoxical vocal-fold motion
GERD—gastroesophageal reflux disease
FOE—fiber-optic examination
PFT—pulmonary function test
PPI—proton-pump inhibitor

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WHAT'S KNOWN ON THIS SUBJECT: Paradoxical vocal-fold motion (PVFM) is a commonly overlooked diagnosis in children with breathing complaints, especially in those with exercise-induced symptoms. Standardized evaluation and treatment protocols are lacking for children with PVFM.



WHAT THIS STUDY ADDS: PVFM can be treated successfully with a multidisciplinary approach that involves physicians, speech therapists, and behavioral therapists. Clinical presentation may help dictate which treatment modality best addresses the individual needs of a child with PVFM.

abstract

OBJECTIVES: To describe (1) a cohort of children with paradoxical vocal-fold motion (PVFM) who were referred to a multidisciplinary airway center and (2) the outcomes of various treatment modalities including speech therapy, gastroesophageal reflux disease treatment, and psychiatric treatment.

PATIENTS AND METHODS: This was a case series with chart review of children younger than 18 years with PVFM evaluated at a tertiary care pediatric airway center over a 36-month period.

RESULTS: Fifty-nine children with PVFM were evaluated. The cohort had a mean age of 13.64 years (range: 8–18 years) and a female-to-male ratio of 3:1. Speech therapy as an initial treatment resulted in a 63% (24 of 38) success rate after an average of 3.7 treatment sessions. Speech therapy was a more successful treatment than antireflux therapy ($P = .001$). Ten percent (6 of 59) of the children presented with a known psychiatric diagnosis, and 30% (18 of 59) of children in the cohort were ultimately diagnosed with a psychiatric condition. Children with inspiratory stridor at rest had a lower initial success rate with speech therapy (56%), a higher rate of underlying psychiatric disorders (75%), and a high rate of success after psychiatric treatment (100%) that required, on average, 3 sessions over a 2-month period.

CONCLUSIONS: To our knowledge, this is the largest study to date on pediatric PVFM. The majority of children with PVFM improve with speech therapy. Children with PVFM at rest may be better treated with psychiatric therapy than speech therapy. Furthermore, children who present with symptoms at rest may have a higher likelihood of underlying psychiatric disease. *Pediatrics* 2011;128:e1443–e1449

Paradoxical vocal-fold motion (PVFM) is inappropriate adduction of the vocal folds during inspiration. PVFM has been known as paroxysmal vocal-cord dysfunction, episodic laryngospasm, irritable larynx syndrome, and Munchausen stridor.¹⁻⁵ Abu-Hasan et al¹ reported close to a 10% incidence in children with exercise-induced dyspnea. Reisner and Borish² reported a 10% rate in patients referred for inpatient evaluation of refractory asthma.³⁻⁹ PVFM can be diagnosed on the basis of physical examination findings or the combination of clinical history and symptoms. Maschka et al³ proposed the following PVFM classification scheme based on etiology: (1) brainstem compression; (2) upper motor neuron injury; (3) lower motor neuron injury; (4) movement disorder; (5) gastroesophageal reflux disease (GERD); and (6) factitious or malingering disorder.³ Tilles⁴ described several PVFM etiology phenotypes: (1) a primary psychiatric disorder; (2) irritant-induced; (3) exercise-induced; (4) laryngopharyngeal reflux; (5) postnasal drip; and (6) chronic cough. Published classification schemes stress the importance that PVFM is not solely a manifestation of an underlying psychological problem.

Diagnosis of and treatment regimens for PVFM continue to be problems; the authors of 1 adult study reported that the average time between symptom onset and diagnosis is >4 years.⁵ Speech therapy is the mainstay of treatment, and adult success rates are >80%.⁶⁻⁹ Biofeedback, self-hypnosis, and psychotherapy have been successful psychiatric treatment modalities.¹⁰⁻¹² Empiric treatment of laryngopharyngeal reflux or GERD has been recommended, although no prospective studies have been conducted.⁸ Antianxiety medications may be helpful in the acute setting of PVFM, but long-

term anxiolytic therapy has not been found to be effective.¹³ If an underlying psychiatric condition is the cause of PVFM, specific medical therapy can be beneficial.¹⁰ Surgery has a limited role in the management of PVFM. Injection of botulinum toxin (Botox [Allergan, Inc, Irvine, CA]) into the thyroarytenoid muscle has been used in patients for whom speech therapy failed.^{14,15}

The literature has recognized the confusion in diagnosing and treating PVFM.^{3-5,9,16} It is common for many children to undergo unnecessary tests, ineffective medical treatment, and prolonged delays without a proper diagnosis. The goal of this study was to review the cases of patients with PVFM seen at a pediatric airway center with an emphasis on their presenting symptoms, physical examination findings, treatment course, and treatment outcome. A secondary goal was to establish a multidisciplinary algorithm that can help pediatricians, speech-language pathologists, and pediatric subspecialists treat patients with PVFM optimally.

PATIENTS AND METHODS

Institutional review board approval was obtained. Children younger than 18 years were eligible. All patients were seen at the multidisciplinary Massachusetts Eye and Ear Infirmary Pediatric Airway Center. A clinic database was examined for all children diagnosed with PVFM between January 1, 2008, and December 30, 2010. The electronic medical records were then reviewed for demographics, initial symptoms, physical examination findings, treatment modalities, and treatment results. If the medical record was incomplete, then attempts were made to contact the families. Children whom we could not contact or for whom we could not compile complete data were excluded. Successful treatment was defined as the ability to re-

turn to activity or complete symptom resolution.

In our airway-center model, a child is initially assessed by a pediatric otolaryngologist, pulmonologist, and gastroenterologist. Children are evaluated by laryngeal fiber-optic examination (FOE) both at rest and after exercise. Pulmonary function tests (PFTs) are conducted as dictated by clinical history, yet many children have already had PFTs performed. A PVFM diagnosis is made if the clinical history is consistent, and other pulmonary abnormalities are ruled out or if there is inappropriate vocal-fold adduction seen with the FOE. Once a diagnosis of PVFM is made, and if there is no concern for a cardiac or neurologic etiology, the child is sent to speech pathology. The speech pathologist, on an individualized basis, instructs the patient on laryngeal control exercises, laryngeal desensitization strategies, and laryngeal breathing techniques. If it is felt that there may be an underlying psychological component, the child is sent to speech therapy and psychiatry concurrently or to psychiatric treatment alone depending on scheduling availability. Child psychiatrists carry out a detailed evaluation. Patients then undergo psychiatric treatment, which includes biofeedback, hypnosis, and medication management, either alone or in combination. The selection of an initial treatment modality (and progression to additional modalities) is individualized by the psychiatrist. All patients in psychiatric treatment also have a portion of each session devoted to supportive therapy and behavioral management. Empiric medical therapy, most commonly proton-pump inhibitors (PPIs), are only given if there are clinical reflux symptoms or if laryngeal examination has concerning findings. Surgical intervention, the most common of which is botulinum toxin, is reserved for refractory cases.

TABLE 1 Treatment Results

	<i>n</i> (%)
Overall treatment success	45 (76)
Speech therapy for patients with symptoms during exercise	23 (68)
Speech therapy for patients with symptoms at rest	5 (56)
Psychiatric therapy for patients with symptoms at rest	8 (100)

Botulinum toxin is also used as a therapeutic intervention when the consulting psychiatrist feels that the symptoms are so severe that they preclude initiation of psychiatric treatment. The goal is to make the symptoms manageable so that the psychiatric treatment can begin.

Statistical analysis was performed by using SAS 9.2 (SAS Institute, Inc, Cary, NC). Fischer's exact or χ^2 tests were used to evaluate an association between categorical outcomes and categorical variables. $P < .05$ was accepted as the minimal statistical significance level.

RESULTS

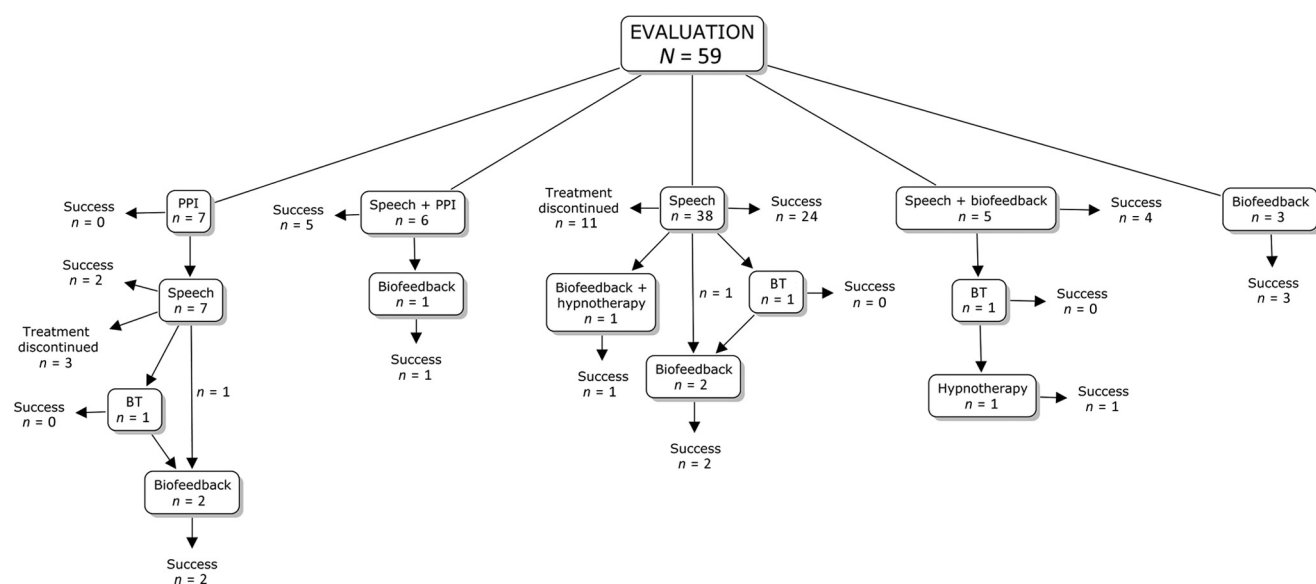
Database review revealed 68 patients diagnosed with PVFM, and complete in-

formation was available for 59 patients. There were 45 (76%) girls and 14 (24%) boys. Mean age at evaluation was 13.64 years (range: 8–18 years). Before airway-clinic presentation, 44.07% ($n = 26$) reported an asthma diagnosis, and 13.56% ($n = 8$) had a diagnosis of GERD. Six patients (10.17%) had been previously diagnosed with depression and/or anxiety. Thirty-six children had PFTs documented, the majority (88.89%) of which yielded normal results. Two patients had PFT results suggestive of asthma, and another was suggestive of PVFM.

Our overall treatment-success rate as defined by symptom resolution and/or return to activity was 76% (45 of 59) (Table 1). Speech therapy was the first-line treatment for most patients (Fig 1). Speech therapy was successful as an initial therapy for 68% ($n = 23$) of the patients who were symptomatic with exercise alone and 56% ($n = 5$) successful for those with any symptoms at rest. Patients attended a mean of 3.7 sessions (range: 1–10). Those who needed >1 session ($n = 20$) were treated for an average of 10.3 weeks.

At the last documented follow-up, these patients had remained asymptomatic and/or continued to participate in their activity an average of 18 months after speech therapy began. Of the 14 children for whom treatment failed, 3 showed some improvement in symptoms but not complete resolution. An additional 3 patients who quit treatment noticed symptom resolution in an average of 27 months after presentation. Eight patients were not able to return to their activity or did not have resolution of symptoms. Mean follow-up time for all 14 patients for whom treatment failed was 17.2 months (range: 2–35).

First-line treatment involving psychiatric treatment, either alone or in combination with speech therapy, was 100% ($n = 8$) successful for children symptomatic at rest. Psychiatric treatment after any speech therapy failure was successful for those with symptoms at rest and for those with symptoms while exercising. PPI therapy alone was not successful for any child. Seven children were sent for pH-probe testing, and 4 demonstrated evidence

**FIGURE 1**

Differences in symptomatology and physical examination findings were identified when patients were analyzed according to treatment group. BT indicates botulinum toxin.

TABLE 2 Initial Treatment Results: Speech Therapy Versus a PPI

Therapy Success ^a	Speech Therapy	Speech Therapy + PPI	PPI
Yes	24	5 ^b	0 ^c
No	14	1	7

^a Success was defined as resolution of symptoms or ability to return to activity.

^b $P = .65$ when comparing speech therapy with speech therapy + a PPI.

^c $P = .001$ when comparing speech therapy alone with a PPI alone.

TABLE 3 Most Common Presenting Symptoms of Children With PVFM

Symptom	<i>n</i> (%) ^a
Shortness of breath with exercise	45 (76.27)
Throat discomfort or tightness	36 (61.02)
Stridor with exercise	23 (38.98)
Shortness of breath at rest	15 (25.42)
Symptoms at rest only	8 (13.56)
Reflux symptoms	14 (23.73)
Chest pain	13 (22.03)
Stridor at rest	12 (20.34)
Wheezing with exercise	6 (10.17)
Hoarseness	3 (5.08)
Wheezing at rest	2 (3.39)

^a Many children had multiple initial symptoms. The denominator value is 59.

of GERD. Three of these 4 patients were started on PPIs without treatment success, but after institution of speech therapy their symptoms resolved. There was no significant difference in success rates between speech therapy alone and the combination of speech therapy with a PPI ($P = .65$). However, speech therapy alone or with a PPI was more successful than a PPI alone ($P = .001$) (Table 2).

Further analysis of the children who were treated by psychiatry yielded interesting results. Twelve of the 14 patients were formally diagnosed with a major psychiatric disorder: anxiety ($n = 11$) or major depressive disorder ($n = 1$). The average time required for airway symptom alleviation with psychiatric care was 2.2 months. The average number of biofeedback sessions was 3.14 (range: 1–6), and the average number of hypnotherapy sessions was 3 (range: 2–4). Five children were also treated with psychotropic medications. Five children required psychiatric care beyond resolution of their PVFM symptoms. Three children, ini-

tially treated successfully with speech therapy, were later referred to psychiatric care for new-onset psychiatric symptoms unrelated to PVFM.

The most common presenting symptoms were shortness of breath with exercise ($n = 45$ [76.27%]), throat discomfort or tightness ($n = 36$ [61.02%]) and stridor with exercise ($n = 23$ [38.98%]) (Table 3). Eight (13.5%) patients had symptoms at rest only. One of these 8 patients had been diagnosed previously with depression, and an additional 6 patients were formally diagnosed with psychiatric disorders after beginning psychiatric care for PVFM. Children who received any psychiatric treatment were more likely to have symptoms only present at rest than patients who did not receive psychiatric treatment ($P = .001$) (Table 4). The more common activities associated with exercise were running, soccer, and swimming. Of the 57 children with documented FOE findings, 14 (24.56%) had inappropriate vocal-fold adduc-

tion on inspiration. Patients who received psychiatric care were more likely to have inappropriate vocal-fold adduction while sitting comfortably than children who did not receive psychiatric care ($P = .001$) (Table 5). No children were found to have neurologic or cardiac etiologies for their symptoms.

DISCUSSION

Our demographic data are consistent with those in previous reports showing that PVFM predominantly affects adolescent girls.^{1,4,17} Many of our patients carried a diagnosis of asthma, yet only 2 had a PFT result consistent with asthma. Asthma and PVFM can occur simultaneously, but the lack of response to asthma medications, a change in a child's typical asthma pattern, or absence of expiratory obstruction on spirometry testing should point to PVFM.^{11,17} PVFM symptoms predominate with exercise; <15% of our patients had symptoms at rest only. Our symptom data are also in agreement with those of previous research in which the most common complaints were dyspnea, throat tightness, and stridor with a predominant inspiratory component.^{6,8} It should be noted, however, that inspiratory stridor is not a

TABLE 4 Symptoms Present at Rest: Children Who Received Psychiatric Treatment Versus Those Who Did Not Receive Psychiatric Treatment

Symptomatic at Rest Only	Psychiatric Therapy, <i>n</i> (%)	Nonpsychiatric Therapy, <i>n</i> (%)
Yes	6 (43)	2 (4) ^a
No	8	43

^a $P = .001$ when comparing patients who received psychiatric treatment and had symptoms at rest with patients who received nonpsychiatric treatment and had symptoms at rest.

TABLE 5 FOE Findings at Rest: Children Who Received Psychiatric Treatment Versus Those Who Did Not Receive Psychiatric Treatment

Exam Findings	Psychiatric Therapy, <i>n</i> (%)	Nonpsychiatric Therapy, <i>n</i> (%)
Adduction on inspiration	5 (42)	1 (2) ^a
Abduction on inspiration	7	44
Total	12 ^b	45

^a $P = .001$ when comparing patients who received psychiatric treatment and had vocal-fold adduction on inspiration compared with those who did not receive psychiatric treatment.

^b Of 14 patients treated by psychiatry, 2 refused the FOE (total $n = 12$ with examination findings).

requirement for the diagnosis, because expiratory and biphasic stridor have been reported in patients with PVFM.⁵

The majority of our patients were not found to have vocal-fold adduction on inspiration during examination. It is difficult to elicit findings in many children with PVFM, because it is often impossible to replicate the specific exercise conditions under which their symptoms begin. Previous series have shown the ability to elicit physical examination findings in <50% of patients.^{18,19} However, it is interesting that we were able to visualize inappropriate vocal-fold adduction in the majority of children who complained of symptoms at rest. Similar to other clinicians and researchers, it has been our approach to not regard FOE as the gold standard for diagnosing PVFM. Although we do evaluate all of our patients with FOE, our experience has indicated that clinical history, symptoms, and thorough evaluation and exclusion of primary pulmonary, neurologic, and cardiac entities are sufficient to make a diagnosis of PVFM. Our overall treatment-success rate was 76%. Speech therapy had a high success rate in patients who presented with symptoms only during exercise. Speech therapy was not as successful for patients who presented with symptoms at rest only. No child improved when placed on PPI therapy alone. Even in 3 children for whom a pH-probe study confirmed GERD, there was no improvement with PPI therapy alone. There was no significant difference in success rates between speech therapy alone and speech therapy with a PPI, which is most likely because of the small sample size; a much larger, prospective efficacy study would need to be conducted to determine if PPIs have a synergistic effect with speech therapy or if PPI therapy is necessary at all. Although GERD is often cited as a

common cause of PVFM in adults, the literature does not support a strong association. Arytenoid edema, interarytenoid edema, and pachydermia have been suggested as physical examination signs of GERD and associated with PVFM, but these physical examination findings have not been correlated with results of pH-probe studies or impedance testing in children.^{5,8} Given our findings, we would not advocate a PPI as a sole treatment for PVFM. We do not start all of our patients with PVFM on empiric PPI therapy, and we recommend only giving it to children with overt GERD symptoms. Furthermore, the children who are put on PPI therapy are also sent to speech therapy as the primary treatment modality.

Our treatment failures deserve special comment. Three children's cases did not meet our definition of successful treatment. Interviews with these children and their families revealed that although they had occasional symptoms, their overall status improved significantly with speech therapy, enough so that they felt that they did not need further treatment. The symptoms of an additional 3 patients did not respond to initial speech therapy, and they did not seek additional treatment. These children had symptoms that resolved spontaneously, on average, 2 years after being initially seen. The remaining 8 patients continued to have symptoms at their last follow-up visit or phone call, and they did not plan on seeking further treatment. There were 2 common reasons why the patients for whom treatment failed discontinued treatment. One explanation was the lack of available local speech therapy or psychiatric resources and the inability to commit significant travel time to attend specialized therapy. A second explanation was that many children simply stopped playing the sport that caused the symptoms. From

these data it is difficult to comment on the natural history of PVFM, because initial treatment strategies have a fairly high success rate.¹⁹ Only prospective studies will provide insight into whether children simply outgrow their symptoms or continue to be at risk through their adult life.

Many case reports have associated PVFM as a laryngeal manifestation of stress or other psychiatric disorder.^{12,13,20} Approximately 10% of the patients in our cohort presented with a known diagnosis of anxiety or depression. An additional 12 children were found to have an underlying psychiatric diagnosis once PVFM treatment began; thus, 30.5% of our patients had an underlying psychiatric condition. Anxiety was particularly prominent, which suggests that there is a significant association between PVFM and anxiety. These children's PVFM symptoms responded well to psychiatric treatment: resolution occurred in <3 months for most patients. Overall, psychiatric treatment was successful both for those patients who had symptoms at rest and those whose symptoms did not improve with speech therapy. Patients referred to psychiatric care were more likely to have symptoms at rest and to have inappropriate vocal-fold adduction at rest. Although the numbers are small, our data would suggest that children with PVFM symptoms at rest are better treated with psychiatric treatments initially. PVFM symptoms at rest may also be a harbinger of psychiatric disease that requires additional treatment, because half of the children in our study who had symptoms at rest and received psychiatric care required prolonged psychiatric care after their PVFM symptoms resolved.

From symptom presentation and outcome data, we formulated a treatment algorithm that may aid those who see children with PVFM (Fig 2). The goal of

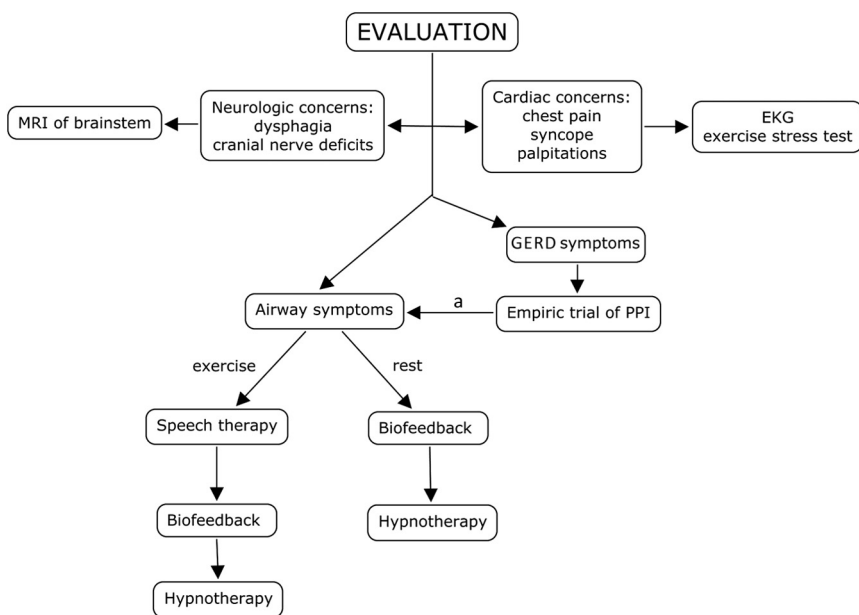


FIGURE 2

Proposed algorithm for children with suspected PVFM. ^a Children with GERD symptoms will most likely need a trial of speech therapy or biofeedback for long-term successful treatment. EKG indicates electrocardiogram.

this algorithm is to help reduce treatment inefficiencies; studies have found that patients with undiagnosed PVFM use health care resources at a higher rate than age-matched controls with moderate-persistent asthma.²¹ One portion of this algorithm that should be stressed is the need to rule out cardiac and neurologic disease. Finally, as we alluded to in “Results,” there is a very small number of children whose symptoms are so debilitating that surgical intervention may be required as a temporizing measure to alleviate significant symptoms that prevent normal activities of daily life. Botulinum toxin may provide temporary relief so that a patient can participate in speech or psychiatric treatment. Botulinum toxin injection is not a long-term cure and should only be used when traditional therapies have failed.

Limitations of our data include their retrospective nature and the fact that all patients were evaluated in a multidisciplinary airway clinic with relatively easy initial access to specialized speech therapists and child psychia-

trists. Our outcome data are limited by the subjective nature of reporting, because there is no objective test to determine the resolution of airway symptoms. We are unable to determine long-term results, because the children were followed until their last documented clinic visit. In most instances, this last visit was marked by successful speech or psychiatric therapy and the provider felt that no further treatment was necessary. This prevents us from commenting on recurrence rates, but our general clinical experience is that once the children learn adequate breathing control, they can successfully use these techniques during symptom recurrence. From our originally identified 68 patients, 9 were lost to follow-up, which left us with a cohort of 59 patients. Six of the patients for whom we considered treatment to have failed had partial or complete resolution of their symptoms; thus, our symptom-resolution rate is likely higher than our treatment-success rate. Furthermore, although our reported population was robust, the populations of certain treatment groups

such as those children who received PPI therapy and those who received psychiatric care were relatively small. To carry out a treatment-efficacy study of speech therapy, speech therapy with concurrent PPI therapy, and PPI therapy alone, 103 subjects per group would be needed to obtain at least 80% power.

PVFM can be a frustrating entity for both patient and physician. Patients are usually frustrated because they can go for a significant time without an accurate diagnosis and appropriate treatment. Providers can be frustrated with the various terminology used to describe this disorder, the fact that the symptoms overlap with asthma, and the lack of clear treatment algorithms.⁹ Because of the relative infrequency of this diagnosis, future insights into evaluation and treatment will only come from collaborative studies at multidisciplinary centers. We hope that our proposed algorithm will be a starting point for more efficient evaluation, treatment, and research of these patients.

CONCLUSIONS

PVFM should be suspected in children who have dyspnea or stridor on exertion and normal PFT results or for whom adequate asthma medication fails. Speech therapy is a successful treatment for most children with PVFM, but a subset of children, mainly those with PVFM at rest, most likely benefit from psychiatric treatment. More prospective research is needed to identify validated screening instruments that may best identify which treatment modality best serves an individual patient. Further research is also necessary to determine if PVFM is a harbinger of more serious psychiatric illness as a child matures.

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