

# The Clinical Course of Idiopathic Bilateral Vocal Fold Motion Impairment in Adults: Case Series and Review of the Literature

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**Summary: Aim.** Steps for assessment and successful management of bilateral vocal fold motion impairment (VFMI) are (1) recognition of its presence, (2) identifying the etiology and factors restricting vocal fold motion, (3) evaluation of airway patency, and (4) establishing a management plan. No large series documenting the course and outcome of adult idiopathic bilateral VFMI has been published within the past 15 years.

**Methods.** Retrospective chart review of adult patients with idiopathic bilateral VFMI at a tertiary academic center. A diagnosis was established if history, physical examination with laryngoscopy, and initial imaging excluded a cause. Records were reviewed for demographics, clinical characteristics, surgical intervention details, and length of follow-up.

**Results.** Nine adult patients with idiopathic bilateral VFMI were identified. There were five males and four females with a mean age of 59.6 years. The mean follow-up period was 54.4 months (range, 6–111 months). Upon presentation to our laryngology service, three patients were advised observation, three patients were advised to undergo urgent tracheostomy, and three patients were advised to undergo elective surgery for airway management. By the end of the follow-up period, only four patients (4/9, 44.4%) were tracheostomy dependent, one of them was lost to follow-up after tracheostomy tub downsizing for decannulation.

**Conclusions.** To our best knowledge, this is the largest series so far of adult patients with idiopathic bilateral VFMI. Conservative treatment can be considered as an alternative to surgery in select cases.

**Key Words:** Idiopathic–Bilateral–Vocal fold motion impairment–Adult–Paralysis–Immobility.

## INTRODUCTION

Bilateral vocal fold motion impairment (VFMI), hypomobility or immobility, reduces the area of the glottic aperture which consequently increases airway resistance. Glottic level narrowing induces persistent stridor and dyspnea that worsen with upper airway inflammatory conditions and exercise. An accurate diagnosis and appropriate treatment are necessary as this condition may progress to acute respiratory failure. The requirements for assessment and successful management of bilateral VFMI are (1) recognition of its presence, (2) identifying the etiology and the factors restricting vocal fold (VF) motion, such as intra-arytenoid adhesions or cricoarytenoid joint fixation, (3) evaluation of airway patency, and (4) establishing a management plan. Idiopathic bilateral VFMI is a rare unrecognized condition in adults. The diagnosis is made when a thorough history and physical examination in concert with imaging, usually computed tomography (CT) with intravenous contrast along the course of the recurrent laryngeal nerve (RLN), do not reveal an etiology for the motion impairment.

The treatment of patients with bilateral VFMI presents a challenge to the otolaryngologist-head and neck surgeon who needs to balance airway against voice quality. There is no universal agreement either regarding the exact criteria defining the severity of bilateral VFMI or regarding the preferred surgical procedure. Although several techniques for airway augmentation have been proposed to manage patients with bilateral VFMI, tracheostomy remains the most effective management, especially in the urgent acute airway setting.<sup>1</sup> However, tracheostomy is often refused by the patient as a long-term solution.

In adults, trauma is the most common cause of bilateral VFMI, followed by idiopathic cases at a rate of 34.34%.<sup>2</sup> However, there is limited literature on the outcome and natural history of adult patients with idiopathic bilateral VFMI.<sup>2–4</sup> No large series documenting the course and outcome of adult idiopathic bilateral VFMI has been published within the past 15 years. The purpose of this study was to analyze the natural history of and therapeutic approaches to idiopathic bilateral VFMI in adults and to review the current literature.

## MATERIALS AND METHODS

This retrospective study was approved by the institutional review board of this facility. The charts of adult patients from January 2006 to August 2017 evaluated for idiopathic bilateral VFMI were reviewed. Eligible patients were extracted from our institution's electronic medical record with the help of Stanford Translational Research Integrated

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Database Environment. Stanford Translational Research Integrated Database Environment is a research and development project at Stanford University to create a standard-based informatics platform supporting clinical and translational research. Patients aged 18 or older with bilateral vocal fold/cord paresis/hypomobility or paralysis/immobility (ICD 9 codes 478.30–478.34) and “idiopathic” diagnosis were included in the initial database extraction. During the review, only those with documented laryngoscopy findings confirming bilateral VF hypomobility or immobility were included. Idiopathic bilateral VFMI was diagnosed if clinical history, head and neck (H&N) physical examination including flexible or rigid laryngoscopy, and initial imaging from skull base to chest excluded a cause. Patients with known neurological or rheumatologic diseases—whether upon presentation or manifesting during follow-up—were excluded. Patients with a history of head and neck, thoracic, or cranial base malignancy, radiation therapy, or surgery; traumatic mechanisms including prolonged intubation ( $\geq 48$  hours); known mediastinal disease; or with findings indicating an obvious cause for VFMI on workup were excluded. In addition, patients with missing data were also excluded. Medical records were reviewed, and data on selected demographics (age, gender) were extracted. Also, retrieved were data on the presenting symptom, surgical procedures performed, and last follow-up status.

Statistical analyses were performed with the SAS system software package version 9.2 (SAS, Cary, North Carolina). The numeric variables were presented as total numbers, percentages, and mean  $\pm$  standard deviation values.

**RESULTS**

Overall during the study period, based on ICD9 codes used, the charts of 66 adults with the diagnosis of idiopathic bilateral VFMI were reviewed. Only nine patients met our inclusion criteria. None of the patients had undergone laryngeal electromyography (LEMG). There were five (55.6%) males and four (44.4%) females. The mean age was 59.6 years (range, 37–86 years). Table 1 summarizes the demographics and clinical presentation of these patients. The majority of patients presented to the laryngology service with dyspnea and stridor (7/9, 77.8%). Three patients had missing data on the duration of presenting symptoms, and six patients were symptomatic for 1 week to 3 years before being referred for evaluation. Three patients had a history of idiopathic unilateral VF immobility and were found with bilateral VFMI upon presentation to our clinic. Two of them had previously undergone unilateral type 1 thyroplasty before developing bilateral immobility. Two patients presented with no previous laryngology exam. Four patients were referred with known untreated bilateral VFMI.

Length of follow-up for patients with idiopathic bilateral VFMI ranged from 6 to 111 months, with a mean of 54.4 months (Table 2). Upon presentation to our laryngology service, three patients were offered follow-up alone with no surgical or medical intervention, three patients were

**TABLE 1.** Demographics and Presenting Symptoms of Patients with Idiopathic Bilateral Vocal Fold Motion Impairment

	First Laryngology Encounter Location	Age/Gender at Presentation	Main Complaint	Duration of Symptoms	Prior known VFMI Diagnosis	Previous Procedures	VF Mobility on Laryngoscopy (Rt/Lt VF)
1	Outpatient clinic	71/F	SOB and stridor	12 months	Bilateral VFMI	None	Hypomobile/Hypomobile (worse on the Rt)
2	Outpatient clinic	66/F	SOB and stridor	3 months	Bilateral VFMI	None	Hypomobile/Hypomobile
3	Outpatient clinic	53/F	SOB and stridor	Unknown	No	None	Immoble/Immoble
4	inpatient consult	72/F	SOB and stridor	6 months	Bilateral VFMI	None	Hypomobile/Hypomobile (worse on the Lt)
5	Outpatient clinic	70/M	SOB and stridor	3 years	Bilateral VFMI	None	Hypomobile/Hypomobile
6	Emergency room	86/M	SOB and stridor	1 week	No	None	Immoble/Immoble
7	Outpatient clinic	42/M	SOB and stridor	Unknown	Rt VF immobility	Rt type 1 thyroplasty	Immoble/Immoble
8	Outpatient clinic	39/M	Voice breaks	Unknown	Rt VF immobility	None	Immoble/Hypomobile
9	Outpatient clinic	37/M	Worsening dysphonia	10 months	Rt VF immobility	Rt type 1 thyroplasty	Immoble/Hypomobile

F, Female; L, Left; M, Male; RT, right; SOB, shortness of breath; VFMI, vocal fold motion impairment.

**TABLE 2.**  
**Immediate and Follow-up Findings and Interventions of Patients with Idiopathic Bilateral Vocal Fold Motion Impairment**

	Recommended Management at First Encounter	Time to Procedure	Additional Procedures During Follow-up	Time to Decannulation	Duration of Follow-up (Months)	Status at Last Follow-up	VFMI Recovery at Last Follow-up
1	Elective surgery- Pt refusal	—	Urgent tracheostomy, TCF closure	1 year	32	Decannulated	Rt VF hypomobility improved (Partial recovery) 2 y)
2	Elective surgery- Pt refusal	—	Urgent tracheostomy. No further treatment (Pt refusal)	No decannulation	100	With TT	No
3	Elective surgery- Pt refusal	—	Urgent Tracheostomy, CO2 laser-assisted left cordotomy (mobile joints)	Downsized. No decannulation	111	TT down-sized, lost to follow-up	Rt VF hypomobility (Partial recovery)
4	Tracheostomy	2 weeks	No	No decannulation. Pulmonary toilet	66	With TT	No
5	Tracheostomy	8 days	No further treatment (Pt refusal)	No decannulation	62	With TT	No
6	Tracheostomy	3 days	No	6 months	6	Decannulated	Bilateral VF mobility (Full recovery)
7	Follow-up	—	No	—	49	No TT	No
8	Follow-up	—	No	—	47	No TT	No
9	Follow-up	—	No	—	17	No TT	No

Pt, patient; TCF, Tracheocutaneous fistula; TT, tracheostomy tube; URI, Upper respiratory infection; VF, vocal fold.

advised to undergo urgent tracheostomy, and three patients were advised to undergo elective surgery for airway management. All patients agreed with the offered plan and recommendations, except for the elective surgery group. Interestingly, this group of three patients all refused elective intervention but later required urgent tracheostomy. By the end of follow-up, only four patients (4/9, 44.4%) had a tracheostomy tube (TT), one of which was lost to follow-up after TT downsizing for decannulation. By the end of follow-up, three patients were found with partial to complete recovery of VF motion. One patient regained full bilateral VF mobility (patient number 6) within 6 months following diagnosis.

## DISCUSSION

The different terms used to describe VFMI are confusing, used interchangeably, and not standardized. Based on the Rosen et al VFMI nomenclature proposal from 2016, the following terms were described in detail: VF paralysis, VF paresis, VF immobility/hypomobility associated with mechanical impairment of the cricoarytenoid joint and VF immobility/hypomobility related to malignant laryngeal tumors.<sup>5</sup> The terms VF paresis or paralysis refer to the reduced or absent function of the vagus nerve or its distal branch, the RLN.<sup>5</sup> While the general terms of VF immobility and VF hypomobility describe the qualitative physical exam finding of VF motion, they make no assumption of the etiology, and do not imply an idiopathic status.<sup>5</sup> Interestingly, this nomenclature did not include separate group of unilateral/bilateral idiopathic VFMI or VF immobility/hypomobility of unknown origin.

Several studies have reviewed large numbers of patients with bilateral VFMI and analyzed them based on etiology.<sup>6–10</sup> The incidence of bilateral VFMI was reported to be about one-third of all VFMI cases.<sup>11</sup> Feehery's comparative study of the etiologic factors reported before and after 1980, showed a significant decrease in cases of bilateral VFMI secondary to thyroidectomy (66.9% versus 16.6%) and an increase of bilateral VFMI due to trauma (intubation) and extrinsic compression by neoplasms. A significant increase was also found in the idiopathic group (3.4% versus 13.9%).<sup>9</sup> Similar results were found by Rosenthal et al that showed a lower incidence of bilateral VFMI by iatrogenic causes and an increase in idiopathic cases.<sup>10</sup> Idiopathic cases were reported to be as high as 39.34%.<sup>2</sup>

Compared with unilateral VFMI, where normal ventilation can be relatively spared, the classical presentation of patients with bilateral VFMI is a reduction of the glottal area resulting in various degrees of airway compromise. Patients typically have noisy inspiratory breathing with minimal voice change. Patients may present with a breathy, weak voice, and minimal airway symptoms if the vocal folds are in a more abducted position. In the current study, seven out of the nine patients (77.8%) first presented to our laryngology service with shortness of breath and worsening

stridor (7/9, 77.8%). Two patients with known idiopathic right VF immobility, one of them after type 1 thyroplasty, presented with voice change as the main presenting symptom and complaint.

Most of the literature on the evaluation of a patient with VFMI, and specifically idiopathic VFMI, focuses on unilateral immobility/hypomobility.<sup>12–15</sup> Yet, in general, the diagnosis of idiopathic VFMI, unilateral or bilateral, is made after a complete workup, including clinical history and H&N physical examination with flexible laryngoscopy. CT imaging of the neck and chest with intravenous contrast, visualizing the path of the vagus and RLNs, is usually part of the diagnostic workup of immobility/paralysis, though not always with hypomobility/paresis.<sup>14</sup>

The diagnostic evaluation begins with a thorough clinical history, which in many instances can lead directly to the etiology.<sup>16,17</sup> Patients need to be explicitly questioned about trauma, history of intubation, known rheumatologic disease, previous H&N or thoracic malignancy, surgery or radiation, history of infection or recent upper respiratory infection, and the degree and onset of symptoms including stridor, shortness of breath, dysphonia, dysphagia, and aspiration. Physical examination includes a complete H&N exam including videolaryngoscopy, preferably flexible, which is the mainstay investigation in the diagnosis of VFMI.<sup>18</sup> Given the frequency of associated neurological conditions in adult patients with idiopathic VF immobility, a careful neurological examination should be considered for all patients.<sup>12</sup> In cases of suspected posterior glottic stenosis (interarytenoid scar) and/or cricoarytenoid fixation, palpation of the cricoarytenoid joints and the posterior glottis is important. Traditionally, the palpation examination is done in the operating room by direct laryngoscopy and with general anesthesia. Another option is in-office arytenoid palpation with an Abraham cannula while performing flexible laryngoscopy, as described by Krishna and Rosen.<sup>19</sup> None of the patients had a history or physical findings suggestive of posterior glottic stenosis (interarytenoid scar) and/or cricoarytenoid fixation, and palpation was not done.

The role and routine use of CT scan in the evaluation of idiopathic unilateral VF immobility is well established and was found justified and cost-effective.<sup>20–22</sup> The utility of CT scan in bilateral VFMI is not clear. Yet, all patients included in this cohort underwent CT scan as part of their initial workup. Based on the study by Noel et al from 2016, patients that were initially diagnosed with idiopathic unilateral VF immobility following normal CT scan may have an occult cause that later becomes evident.<sup>15</sup> Therefore, repeated imaging was recommended within two years after diagnosis. A retrospective study by White et al supported the utility of serological evaluation in the initial investigation of idiopathic VFMI, including a complete blood count, and testing for Lyme disease, syphilis, and myasthenia gravis in the appropriate clinical setting.<sup>23</sup> If a neurologic condition is suspected, magnetic resonance imaging of the posterior cranial fossa, LEMG, and antibody studies were suggested to help with the diagnosis.

LEMG can play an essential role in the assessment, differential diagnosis, and prognosis of bilateral VFMI, evaluating denervation, reinnervation, and the potential for recovery.<sup>24,25</sup> LEMG may assist with differentiating between mechanical fixation and neurogenic paralysis. According to the statement paper by American Association of Neuromuscular & Electrodiagnostic Medicine, LEMG changed the diagnosis of VF immobility approximately 48% of the time when the original diagnosis was suspected to be injury RLN.<sup>26</sup> Yet, the authors suggest that future prospective studies should examine patient outcomes regarding clinical decisions that were made using LEMG data as compared with decisions made without this information. We have found that LEMG may not readily provide information on etiology or guide treatment recommendations. Therefore, LEMG has not been a routine component of evaluation of patients with VFMI, and none of the patients in our study had undergone LEMG.

During the follow-up period, three patients recovered VF mobility (patients 1, 3, 6). One year is widely used as the interval from onset during which recovery is expected. A recent study by Husain *et al* found that 38 of 55 patients (69%) with idiopathic unilateral VF paralysis recovered vocal function, two-thirds doing so within six months of onset.<sup>27</sup> There is limited literature on the potential recovery of idiopathic bilateral VFMI. Gupta *et al* reported that this group is associated with the best prognosis, with spontaneous recovery of 58.33%, when compared to 56.25% when traumatic and 40% when iatrogenic. However, this study included pediatric patients (24 patients  $\leq 15$  years, 37 patients  $\geq 16$  years) and it is not clear what diagnostic imaging was done in order to establish the diagnosis of idiopathic.<sup>2</sup> Interestingly, three patients in our study were previously diagnosed with unilateral paralysis. Based on our extensive literature review, there are no studies which estimate the risk to the contralateral uninvolved side in idiopathic paralysis.

The primary objective of intervention for bilateral VFMI is to improve patients' ventilation. Various surgical procedures have been developed to address the airway restriction. Nevertheless, an ideal treatment of bilateral VFMI has not been found in spite of advances in surgery and laryngology materials.<sup>1,28</sup> Tracheostomy remains the most commonly used surgical procedure for bilateral VFMI, especially in the urgent acute setting.<sup>29</sup> It provides the largest airway and maintains the structural integrity of the glottic larynx. Despite its effectiveness, tracheostomy is less favored because it presents an open wound that requires long-term care and is associated with psychosocial problems including adjustment reactions, especially if the procedure was done nonelectively.<sup>30–32</sup>

Since the beginning of the 20th century, other approaches to enlarge the stenotic airway have been introduced first with external methods and later with endoscopic techniques including arytenoidectomy, cordotomy, arytenoid abduction, and laterofixation.<sup>1,16,33,34</sup> In addition, new techniques such as laryngeal pacing and reinnervation procedures have also

been studied and applied in some cases of bilateral VFMI.<sup>35,36</sup> In recent years, botulinum toxin (Botox) injection has been suggested for temporary relief of airway obstruction symptoms in adult patients with bilateral VFMI.<sup>37,38</sup> Other treatment approaches currently being investigated include gene therapy and stem cell therapy, however, the application for bilateral VFMI is largely unknown.<sup>39,40</sup>

Recognizing different degrees of airway obstruction and restoring or maintaining airway patency is an integral part of airway management. In the current study, three patients were offered immediate tracheostomy, three patients were offered elective laryngeal surgery to expand the airway, and three were offered watchful waiting. The insight into clinical decision-making process is often limited and difficult to assess. Yet, several factors were taken into consideration before making airway management recommendations. These include patient's general appearance at presentation (stable and with normal vital signs), consult/visit setting (outpatient versus emergency department versus inpatient), symptoms' duration (acute versus non acute), endoscopic findings upon laryngoscopy exam (VF hypomobility versus immobility, VF median versus paramedian position, glottic airway patency), physician's personal experience and confidence level, and patient's preference and understanding of the diagnosis. Glottic opening in millimeters was not routinely reported.

In the present study, all three patients who were offered elective surgery refused to have surgical intervention (patients 1–3). All of them returned later and underwent urgent tracheostomy due to worsening symptoms with upper respiratory tract infection. None of the patients that were offered watchful waiting underwent further surgical intervention (patients 7–9). Based on these findings, appropriate and justified decisions were made based on clinical judgment.

At the end of the follow-up period, four out of nine patients had a TT (patients 2–5). Patient four remained with TT and was not a candidate for surgical intervention or decannulation due to cerebrovascular accident (following the VFMI diagnosis and tracheostomy) with subsequent poor clearance of airway secretions. The TT was kept for pulmonary toilet/hygiene. Two patients were offered further airway procedures but refused and remained with TT (patients 2 and 5). Patient 3, underwent urgent tracheostomy 11 months after she was first seen in clinic and refused elective procedure to improve her airway. Two years later the patient had a CO<sub>2</sub> laser left cordotomy. She subsequently returned to clinic 7 years later with unilateral VF motion. The TT was downsized with an intention of decannulation, but the patient never returned and was lost to follow-up.

The limitations of the current study are its retrospective nature and small study group. Nevertheless, it provides further knowledge on the natural course of idiopathic bilateral VFMI.

## CONCLUSIONS

Bilateral VFMI is a potentially fatal condition. Due to this risk, prompt and accurate diagnosis followed by adequate treatment is mandatory. In this retrospective investigation,

we examined the clinical course of nine adult patients with idiopathic bilateral VFMI. To the best of our knowledge, this is the largest series in the English literature. Our results suggest a very small risk of developing contralateral involvement following idiopathic unilateral VFMI. One third of patients experience eventual and spontaneous recovery of VF motion. Further studies and reports are needed to more fully characterize the natural course and proper management of this important group of patients.

### DISCLOSURES

The authors have no funding, financial relationships, or conflicts of interest to disclose.

### SUPPLEMENTARY MATERIALS

Supplementary material associated with this article can be found, in the online version, at <https://doi.org/10.1016/j.jvoice.2018.11.012>.

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