




Bilateral Vocal Fold Motion Impairment Associated With Diffuse Idiopathic Skeletal Hyperostosis

Diego E. Razura, BS^{1,2} , Elizabeth A. Shuman, MD² ,
Michael M. Johns, III, MD², and Karla O'Dell, MD² 

OTO Open
2024, Vol. 8(3):e70003
© 2024 The Author(s). OTO Open
published by Wiley Periodicals LLC
on behalf of American Academy of
Otolaryngology-Head and Neck
Surgery Foundation.
DOI: 10.1002/oto2.70003
<http://oto-open.org>

WILEY

Abstract

Objective. To describe the clinical courses and interventions of symptomatic patients with bilateral vocal fold motion impairment (BVFMI) attributed to diffuse idiopathic skeletal hyperostosis (DISH).

Study Design. Retrospective cohort study.

Setting. Single Institution Academic Health Center.

Methods. Retrospective chart review of patients ≥ 18 years old evaluated and treated for symptomatic BVFMI secondary to DISH between February 2021 and March 2023. A literature review was conducted.

Results. A total of 4 cases were identified. All patients were male and had symptomatic BVFMI attributed to cervical spine DISH, as seen on imaging. Symptoms ranged from life-threatening dyspnea to breathy dysphonia in addition to dysphagia. Each patient was offered surgery for DISH. Two patients underwent osteophyte removal at the C5-C6 level with improved vocal fold (VF) mobility, breathing, and voice quality. Two patients elected serial observation as voice, swallow, and airway symptoms were manageable. The literature review showed a male-dominant (100%) presentation with an average of 70 years of age. Hypertension (45%) and diabetes mellitus (36%) were the most common comorbidities. Most patients were treated surgically (55%).

Conclusion. Both surgical and conservative interventions may be considered for symptomatic relief and improvement in VF mobility on a patient-to-patient basis. Further study is warranted to investigate the etiology and treatment outcomes in these cases.

Keywords

anterior cervical osteophytes, bilateral vocal fold immobility, diffuse idiopathic skeletal hyperostosis, dysphonia, dyspnea

Received May 16, 2024; accepted August 10, 2024.

Diffuse idiopathic skeletal hyperostosis (DISH), or Forestier disease, is a primarily asymptomatic condition characterized by ossification of the anterior vertebral ligaments. This condition is commonly seen in males and is more prevalent among individuals of advanced age, with an incidence reaching up to 42%.¹ The diagnosis for DISH, originally established by Resnick and Niwayama in 1973, continues to encompass 3 main criteria: (a) the presence of “flowing” calcifications and ossifications along the anterolateral vertebral bodies, (b) relative preservation of disc height in the ossified regions without degenerative disc disease, and (c) no apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or bony fusion.² DISH is often an incidental radiographic finding, but when symptomatic the presentations can have severe complications such as dysphagia, myelopathy, aspiration pneumonia, esophageal obstruction, stridor, dysphonia, and thoracic outlet syndrome.³

In a few reported and rare instances, DISH can present with bilateral vocal fold motion impairment (BVFMI). When both vocal folds (VFs) are involved, the consequences can range from mild discomfort to severe, life-threatening airway obstruction necessitating emergent surgical management. The exact etiology of BVFMI secondary to DISH remains unclear.⁴ The proposed mechanisms suggest that DISH direct compression on the cricoarytenoid joint, which can limit joint motion, or recurrent laryngeal nerve impingement may be responsible.^{5,6} Due to the rarity of BVFMI secondary to DISH, there is no current established management protocol based on patient outcomes, but when

¹Quillen College of Medicine, East Tennessee State University, Johnson City, Tennessee, USA

²Caruso Department of Otolaryngology-Head and Neck Surgery, Keck School of Medicine, University of Southern California, Los Angeles, California, USA

This article was presented at AAO-HNSF 2023 Annual Meeting & OTO Experience; September 30 to October 4, 2023; Nashville, TN.

Corresponding Author:

Karla O'Dell, MD, USC Department of Otolaryngology-Head and Neck Surgery, Keck School of Medicine of USC, 1537 Norfolk Street, Suite 5800, Los Angeles, CA 90033, USA.
Email: Karla.ODell@med.usc.edu

emergent treatment for airway obstruction occurs, tracheostomy is often necessary.⁷

In addition, DISH's pathomechanism is unclear with recent studies suggesting a potential link with metabolic disorders as a contributor.⁸ With the increased prevalence of metabolic disorders in the United States, it can be assumed that the incidence of BVFMI secondary to DISH will also increase. To our knowledge, there are only a few published reports of BVFMI secondary to DISH composed of single case reports. By presenting our management of 4 unique cases, we aim to elucidate the often-delayed diagnosis and unrecognized etiology of BVFMI secondary to DISH and increase awareness of this presentation to prevent adverse outcomes due to delayed treatment.

Methods

The Institutional Review Board (HS-16-00003) of the University of Southern California approved the study. Two laryngologists (M.M.J. III, K.O.) selected 4 patients whose radiographic and laryngoscope findings were consistent with BVFMI secondary to DISH. Patients were evaluated and treated for their symptomatic DISH-associated BVFMI over a year. Clinical notes, diagnostic studies, operative reports, flexible nasolaryngoscopy interpretations, and postoperative data were collected. Factors including demographic data, symptom severity, comorbidities, level and size of osteophytes, interventions, complications, and voice and breathing outcomes were considered.

Databases and Search Strategy

A literature review spanning the last 20 years ending on December 12, 2023, was conducted using the online search databases PubMed, CINAHL, SportDiscus, Web of Science, Cochrane Methodology Register, Cochrane CDSR, Cochrane CENTRAL, and Google Scholar. The general search strategy included Medical Subject Headings (MeSH) terms and title/abstract (tiab) free text phrases that combined condition-specific terms (“Hyperostosis, Diffuse Idiopathic Skeletal” [MeSH] OR Diffuse Idiopathic Skeletal Hyperostosis [tiab] OR Diffuse idiopathic hyperostosis[tiab] OR Cervical Osteophyte [tiab] OR (“Osteophyte”[Mesh] AND cervical[tiab])) with symptomatic presentation terms (((“Vocal Cord Paralysis”[Mesh] AND bilateral[tiab]) OR bilateral vocal fold paralysis[tiab] OR bilateral vocal cord paralysis[tiab] OR Bilateral Vocal Fold Motion Impairment[tiab] OR BVFMI[tiab])). For database-specific search strategy, see Supplemental File S1, available online. Cochrane Methodology Register, Cochrane CDSR, and Cochrane Central were searched but did not yield any results. To further refine the search, additional criteria were implemented: articles had to be written in English and have confirmed cases of BVFMI secondary to DISH. Duplicates were removed. A review of abstracts

and full articles was conducted by 2 authors (D.E.R., E.A.S.). A review of the references cited in the articles was performed to locate further reports. However, this search yielded no new articles beyond those already found in the initial search. Demographic, medical history, symptom, VF motion, level and size of osteophyte, intervention, and outcome data were extracted using a standardized spreadsheet document.

Results

At a single institution, a retrospective chart review from January 2020 to June 2023 yielded 4 patients who were found to have BVFMI attributed to cervical spine DISH seen on imaging. All patients were male (100%), and the average age was 69 years.

In this cohort, the history of hypertension (3, 75%) and diabetes (2, 50%) were the most common medical comorbidities. Most patients presented with dysphagia (4, 100%), dyspnea (3, 75%), and dysphonia (3, 75%). Only 1 patient (25%) presented with weight loss. Diagnostic imaging and management evaluation was done by cervical X-ray (4, 100%), computed tomography (CT) scan of the neck (4, 100%) (**Figures 1 and 2**), modified barium swallow study (MBSS) (3, 75%) (Supplemental File S2, available online), flexible nasolaryngoscopy with stroboscopy (4, 100%) (**Figure 2**), fiberoptic evaluation of swallowing (1, 25%), and magnetic resonance imaging (1, 25%). On examination of VF motion, all patients (100%) had bilateral impairment with laterality variations between patients. Cervical vertebra locations C4, C5, and C6 were involved in all patients. Each patient was offered a surgical option. Ultimately, interventions ranged from conservative (2, 50%) to surgical management (2, 50%). For the 2 (50%) surgically treated, an anterior cervical osteophylectomy was performed with exposure at the C5-C6 level. In the surgically managed patients (2, 50%), improvements in symptoms, breathing, and VF motion were observed. Two (50%) patients elected serial observation as voice, swallow, and airway symptoms were manageable. No harm or adverse effects were encountered aside from temporary dysphagia for the patients who underwent surgery. Complete presentations of individual patients can be found in **Table 1**.

We identified a total of 7 English language articles. Articles were excluded if pre-established inclusion criteria were not met. The literature review search strategy can be found in **Figure 3**. From the 7 articles and our 4 patient cases, all patient cases were male (100%), and the average age was 70. The median was 69 years of age, with a range of 56 to 82. Of reported comorbidities, hypertension (5, 45%) and diabetes mellitus (4, 36%) were the most common. Comorbidities were unspecified in 3 cases (27%). The most common initial symptoms were dysphagia (7, 64%), dysphonia (7, 64%), dyspnea (7, 64%), stridor (5, 45%), and weight loss (4, 36%). Cervical vertebra C4 (10, 91%), C5 (9, 82%), and C6 (7, 64%) were the most involved in DISH pathology. Three patients

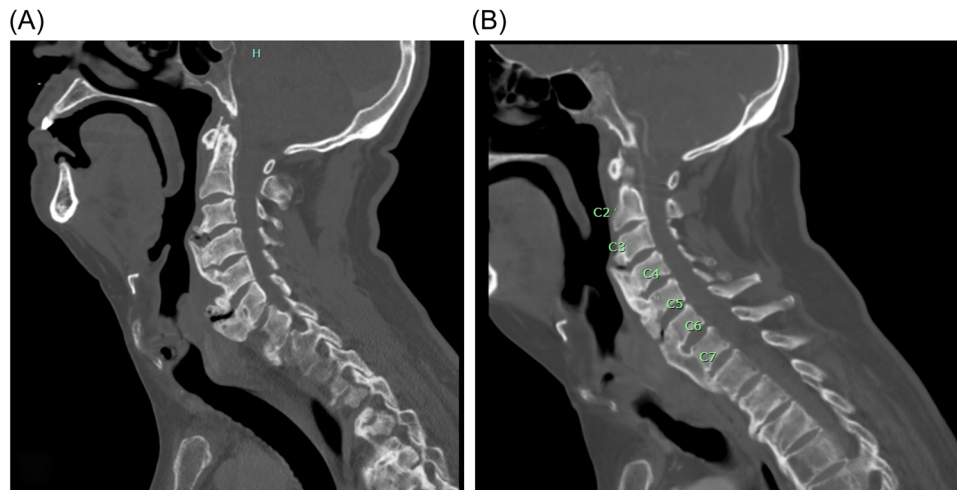


Figure 1. Patient 4 sagittal CT scan of DISH. (A) Preosteophyte removal demonstrates cervical spine hyperostosis over C4-T1. (B) Postosteophyte removal of C5-C6. CT, computed tomography; DISH, diffuse idiopathic skeletal hyperostosis.

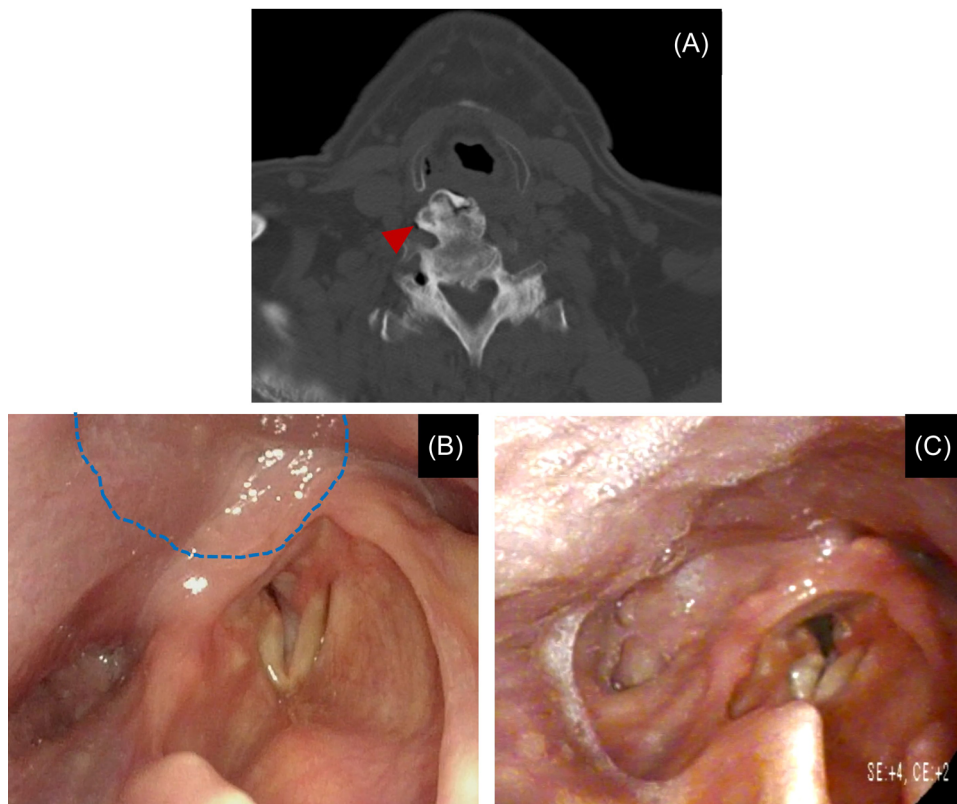


Figure 2. Patient 2: (A) Axial CT scan shows an osteophyte mass (red arrow). (B) Preremoval nasolaryngoscopy view of the VFs with DISH (blue outline). (C) Postremoval nasolaryngoscopy view of the VFs. CT, computed tomography; DISH, diffuse idiopathic skeletal hyperostosis; VF, vocal fold.

(27%) were treated nonsurgically or conservatively. Six (55%) patients were treated with surgical osteophy-tectomy. Two (18%) patients received pharmacological treatment. Young et al reported the use of racemic epinephrine, dexamethasone, and lorazepam without improvement, and Jain et al used unspecified antibiotics and injectable and nebulized steroids.^{9,10} Two (18%) had a tracheostomy performed for airway management. A

summary of our patients and the literature review can be found in **Tables 1** and **2**, respectively.

Discussion

Our presentation of 4 unique cases highlights the multi-system approach needed to diagnose and manage an unforeseen etiology of BVFMI secondary to DISH.

Table 1. Data From 4 Cases of Bilateral VF Motion Impairment Due to Diffuse Idiopathic Skeletal Hyperostosis

Case	Age/sex	Medical history	Symptoms (initial presentation)	VF motion (initial presentation)	Osteophyte location	Treatment	Outcome (last visit)
Patient 1	75/M	OSA	Dysphagia (solids and liquids), dysphonia	RVF: immobile LVF: hypomobile w/edema Bilateral: aperiodic vibratory pattern, paresis	C2-C6	Conservative	No breathing problems or eating problems, functional voice
Patient 2	82/M	Bladder cancer, HTN	Dysphonia, stridor, dyspnea	RVF: restricted motion LVF: hypomobile	C4-C7	Surgery removal of osteophyte at C5-C6	No breathing problems or eating problems, improved voice with restricted RVF abduction
Patient 3	61/M	Colorectal cancer, CVA, prostate enlargement, mechanical ventilation, HTN, DM, CKDIII	Dysphagia, dyspnea, cough	RVF: immobile LVF: immobile Bilateral: complete glottal closure	C4-C6	Tracheotomy; refused osteophyte removal surgery	Decannulated and presented again with acute respiratory distress requiring another tracheotomy
Patient 4	56/M	Ankylosing spondylitis, HTN, DM, HLD, ½ pack a day former smoking history	Dysphagia (liquids > solids), dyspnea, odynophagia, dysphonia, weight loss	RVF: hypomobile LVF: immobile Bilateral: sufficient glottal closure	C4-T1	Surgery removal of osteophyte at C5-C6	Mild dysphagia and dysphonia, improved glottal opening

Abbreviations: C#, cervical vertebra number; CKDIII, stage III chronic kidney disease; CVA, cerebrovascular accident; DM, diabetes mellitus; HLD, hyperlipidemia; HTN, hypertension; LVF, left vocal fold; M, male; OSA, obstructive sleep apnea; RVF, right vocal fold; VF, vocal fold(s).

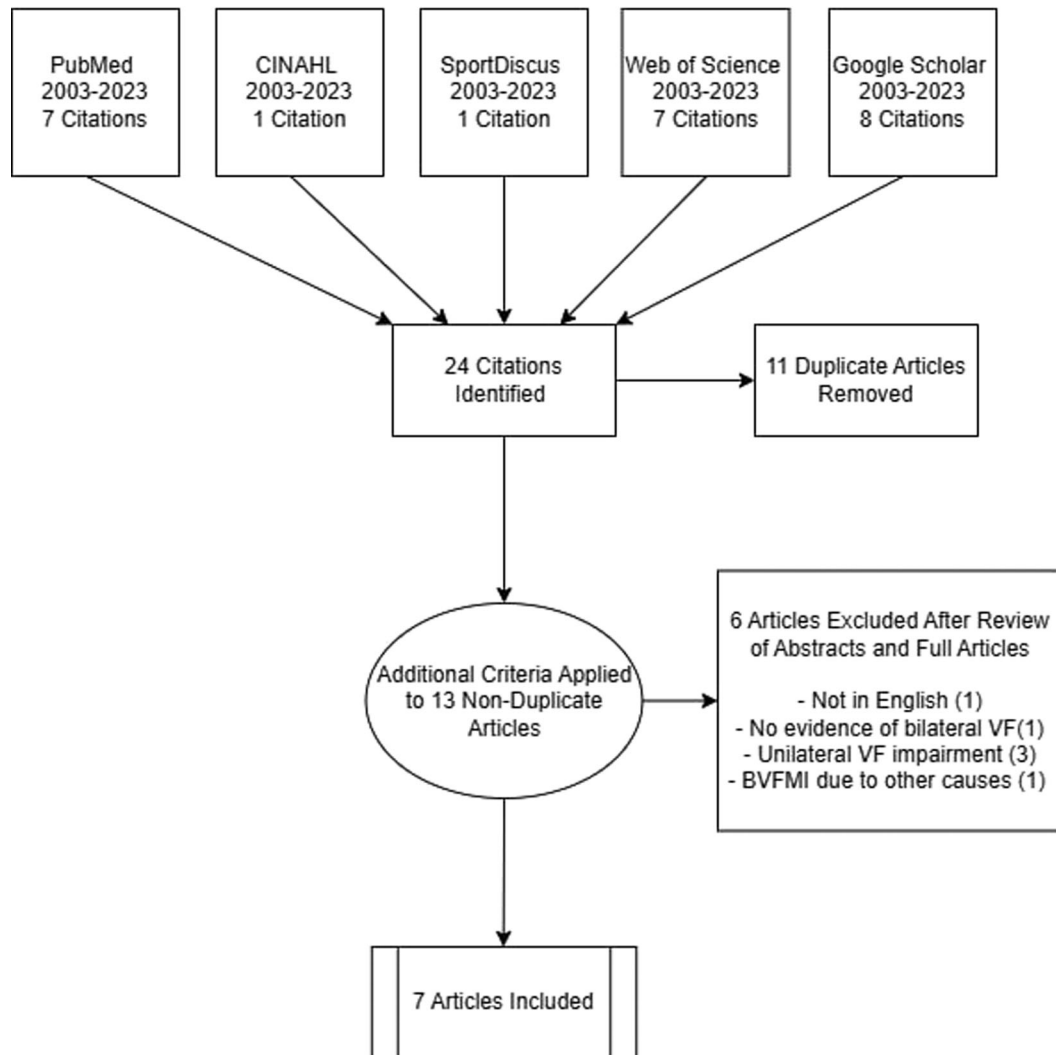


Figure 3. Flow diagram demonstrating the literature review search strategy. The diagram shows the process for the inclusion and exclusion of articles. BVFMI, Ibilateral vocal fold motion impairment; VF, vocal fold.

BVFMI is generally considered one of the most challenging problems laryngologists face. Although a rare presentation,¹⁴ the limited understanding of the involvement of DISH with BVFMI poses an additional significant challenge. A multidisciplinary approach involving laryngologists, spine surgeons, and speech-language pathologists was consistently required to provide comprehensive care for these patients. DISH can present itself in multiple ways and should be taken on a case-by-case basis regarding the decision of when to incorporate surgical intervention for the removal of osteophytes.

Our patient case series, although rare, shares common attributes with more typical presentations of DISH such as male sex, older age, hypertension, and diabetes mellitus as demonstrated in our literature review. The exact etiology of DISH is still unknown, but evidence demonstrates a strong involvement of metabolic disorders.⁸ With the expected increase of metabolic disorders in the United States, further exploration, and guidance on managing this condition should be pursued to improve patient care.

Diagnostic tests, including flexible laryngoscopy with stroboscopy, CT scan, MBSS, and X-rays, were crucial in evaluating the extent of disease progression and determining the appropriate management strategy. For 3 cases, the patients presented with acute worsening dyspnea that warranted urgent treatment. Patients 2 and 4 needed acute surgical intervention due to worsening respiratory distress. Before presenting to our care, patient 3 had a tracheotomy performed due to worsening dyspnea. It is possible that if the inciting etiology had been identified earlier, he could have avoided a tracheotomy. The patient refused surgery once the diagnosis was made and requested decannulation. He later presented again and required an additional tracheotomy. Only patient 1 was managed conservatively with swallow and breathing therapy.

The question of when to proceed with surgery or manage conservatively is up for debate. It is understood that patients with severe symptoms such as airway obstruction and severe dysphagia should proceed with osteophyte removal surgery.^{15,16} For all 4 cases, DISH or

Table 2. Data From 7 Cases From Literature Review of Bilateral Vocal Fold Motion Impairment Due to Diffuse Idiopathic Skeletal Hyperostosis

References	Age/sex	Medical history	Symptoms (initial presentation)	VF motion (initial presentation)	Osteophyte location	Treatment	Outcome (last visit)
Young et al ⁹	61/M	Schizophrenia, traumatic brain injury	Stridor, dyspnea, periodic, apnea	Bilateral: paralysis with retained ability to phonate	C2-C7	Medication (no improvement), tracheostomy	Discharged to an inpatient hospice
Sebaaly et al ⁵	76/M	Not specified	Dysphagia, dyspnea, dysphonia, weight loss	Bilateral: paralysis	Not specified	Surgery removal of unspecified osteophytes	Normal swallow on barium enema, normal VF movement, and no recurrence of ossification
Goico-Alburquerque et al ¹¹	80/M	HTN, DM, 10-year smoking history	Dysphagia (solids and liquids), odynophagia, dyspnea, sore throat, cough, weight loss	LVF: fixed RVF: minimal abduction	C2-C4	Tracheostomy, gastrostomy tube	Discharged to a long-term acute facility
Allensworth et al ¹²	61/M	DM	Dysphagia, dysphonia, stridor, dyspnea, weight loss	Bilateral: immobile VFs in a paramedian position	Skull base-C7	Surgery removal of osteophytes at C4-C7	No recurrence of breathing or swallowing symptoms. Later development of myelopathy and spinal stenosis in the cervical spine requiring posterior decompression using laminectomy and fusion of C4-T1 1 year after anterior osteophylectomy
Jain et al ¹⁰	69/M	CVA, HTN	Dysphonia, stridor	LVF: immobile in a median position, fixed arytenoid RVF: restricted movements, arytenoid mobile	C4-C5	Conservative, medication (antibiotics and steroids)	Stridor decreased with medication, discharged in a stable condition
Certo et al ¹³	68/M	Not specified	Dysphagia, dyspnea	Bilateral: paralysis	C4-C5	Surgery removal of osteophyte at C4-C5	Progressive restoration of normal VF motility, no recurrence of ossification
Aydin et al ⁶	81/M	No significant medical history	Dysphonia, stridor, neck stiffness	Bilateral: immobility of the paramedian located VFs	C3-C7	Surgery: unilateral arythenoidectomy, cordopexy	Not specified

Abbreviations: C#, cervical vertebra number; CVA, cerebrovascular accident; DM, diabetes mellitus; HTN, hypertension; LVF, left vocal fold; M, male; OSA, obstructive sleep apnea; RVF, right vocal fold; VF, vocal fold(s).

osteophyte mass effect was not initially considered as a differential diagnosis. Patients had several visits for shortness of breath and dysphagia before eventually presenting acutely with worsening symptoms. Timely diagnosis can potentially prevent life-threatening presentations and the need for tracheotomy. Despite the lack of severe symptoms, patients who undergo surgery can demonstrate immediate improvement in symptom severity and quality of life.

BVFM is an uncommon etiology that significantly increases the risk of needing a tracheotomy when severe and untreated. BVFM presentations are commonly iatrogenic and derived from causes such as neck surgery or tracheal intubations.¹⁷ Noniatrogenic causes are rare, but can be caused by neurotoxicity, stroke, and brainstem compression.⁹ Therefore, it is important to also consider DISH as a differential for noniatrogenic causes of BVFM in an adult to avoid delay of surgical intervention. The complications for surgical osteophyte removal are rare but include possible vocal fold paralysis, vertebral disc prolapse, fistula formation, hematoma, infection, dysphagia and aspiration, and Horner's syndrome.¹⁸ With the anterior cervical approach for osteophyctomy, it is important to note that the presence of temporary dysphagia is universally associated with this procedure.¹⁹⁻²¹ The presence of early onset dysphagia should be regarded as postoperative recovery rather than due to DISH recurrence. Further outcome studies are needed to elucidate the algorithmic management of BVFM secondary to DISH.

The main limitation of this study includes the small sample size which precludes broader generalization and conclusions. Despite being the largest series report of BVFM secondary to DISH in this search, the findings' generalizability requires further case reports. Nevertheless, all patients who underwent surgery had near immediate improvement in symptom severity, VF motion, and substantial enhancements in overall well-being, highlighting the effectiveness of surgery as a critical treatment option. We anticipate that increased awareness and the projected increase in prevalence will lead to more comprehensive reporting.

Conclusion

BVFM is a rare manifestation of cervical spine DISH, thought due to neurologic and/or mechanical impingement of the laryngeal framework. Both surgical and conservative (eg, voice therapy) intervention may be considered for symptomatic relief and improvement in vocal fold mobility on a patient-to-patient basis. Patients who underwent surgery had near immediate resolution of symptoms and improved well-being. Further study is warranted to investigate the etiology and treatment outcomes in these cases.

Author Contributions

Diego E. Razura, data acquisition and analysis, drafting of manuscript, critical revisions; **Elizabeth A. Shuman**, revision of literature review articles and abstracts, drafting of manuscript,

critical revisions; **Michael M. Johns III**, conception and design of study, drafting of manuscript, critical revisions; **Karla O'Dell**, conception and design of study, drafting of manuscript, critical revisions.

Disclosures


Competing interests: None.


Funding source: None.


Supplemental Material

Additional supporting information is available in the online version of the article.

ORCID iD

Diego E. Razura  <http://orcid.org/0000-0002-0133-3582>

Elizabeth A. Shuman  <http://orcid.org/0009-0009-0872-869X>

Karla O'Dell  <http://orcid.org/0000-0002-0208-1647>

References

- Holton KF, Denard PJ, Yoo JU, et al. *Diffuse Idiopathic Skeletal Hyperostosis and Its Relation to Back Pain Among Older Men: The MrOS Study*. Elsevier; 2011:131-138.
- Resnick D, Niwayama G. Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology*. 1976;119(3):559-568.
- Mader R. *Clinical Manifestations of Diffuse Idiopathic Skeletal Hyperostosis of the Cervical Spine*. Elsevier; 2002:130-135.
- Rosenthal LHS, Benninger MS, Deeb RH. Vocal fold immobility: a longitudinal analysis of etiology over 20 years. *Laryngoscope*. 2007;117(10):1864-1870.
- Sebaaly A, Boubez G, Sunna T, et al. Diffuse idiopathic hyperostosis manifesting as dysphagia and bilateral cord paralysis: a case report and literature review. *World Neurosurg*. 2018;111:79-85.
- Aydin K, Ulug T, Simsek T. Bilateral vocal cord paralysis caused by cervical spinal osteophytes. *Br J Radiol*. 2002; 75(900):990-993.
- Dunya G, Orb QT, Smith ME, Marie J-P. A review of treatment of bilateral vocal fold movement impairment. *Curr Otorhinolaryngol Rep*. 2021;9(1):7-15.
- Harlianto N, Westerink J, Foppen W, et al. Visceral adipose tissue and different measures of adiposity in different severities of diffuse idiopathic skeletal hyperostosis. *J Pers Med*. 2021;11(7):663.
- Young D, Saxby K, Beard M, et al. Acute bilateral vocal cord paralysis (BVCP) in a patient with undiagnosed diffuse idiopathic skeletal hyperostosis (DISH). *Chest*. 2021;160(4): A890-A891.
- Jain S, Singh P, Agrawa G, Kumar S, Banode P. Arytenoid sclerosis in diffuse idiopathic skeletal hyperostosis presenting with acute stridor in elderly: chance or association. *J Laryngol Voice*. 2016;6(1):14-17.
- Goico-Alburquerque A, Zulfiqar B, Antoine R, Samee M. Diffuse idiopathic skeletal hyperostosis: persistent sore throat and dysphagia in an elderly smoker male. *Case Rep Med*. 2017;2017:2567672.

12. Allensworth JJ, O'dell KD, Schindler JS. Bilateral vocal fold paralysis and dysphagia secondary to diffuse idiopathic skeletal hyperostosis. *Head Neck*. 2017;39(1):E1-E3.
13. Certo F, Sciacca G, Caltabiano R, et al. Anterior, extracanal, cervical spine osteochondroma associated with DISH: description of a very rare tumor causing bilateral vocal cord paralysis, laryngeal compression and dysphagia. Case report and review of the literature. *Eur Rev Med Pharmacol Sci*. 2014;18(1 suppl):34-40.
14. Cherfane P, Smailly H, Khalaf MG, Ghaoui N, Melkane AE. Otolaryngologic manifestations of diffuse idiopathic skeletal hyperostosis (Forestier's disease): a systematic review of the literature. *Joint Bone Spine*. 2021;88(6):105218.
15. Presutti L, Alicandri-Ciufelli M, Piccinini A, et al. Forestier disease: single-center surgical experience and brief literature review. *Ann Otol Rhinol Laryngol*. 2010;119(9):602-608.
16. Harlianto NI, Kuperus JS, Mohamed Hoesein FAA, et al. Diffuse idiopathic skeletal hyperostosis of the cervical spine causing dysphagia and airway obstruction: an updated systematic review. *Spine J*. 2022;22:1490-1503.
17. Benninger MS, Gillen JB, Altaian JS. Changing etiology of vocal fold immobility. *Laryngoscope*. 1998;108(9):1346-1350.
18. Kibel SM, Johnson PM. Surgery for osteophyte-induced dysphagia. *J Laryngol Otol*. 1987;101(12):1291-1296.
19. Rihn JA, Kane J, Albert TJ, Vaccaro AR, Hilibrand AS. What is the incidence and severity of dysphagia after anterior cervical surgery. *Clin Orthop Relat Res*. 2011;469:658-665.
20. Bazaz R, Lee MJ, Yoo JU. Incidence of dysphagia after anterior cervical spine surgery: a prospective study. *Spine*. 2002;27(22):2453-2458.
21. Riley LH, Skolasky RL, Albert TJ, Vaccaro AR, Heller JG. Dysphagia after anterior cervical decompression and fusion: prevalence and risk factors from a longitudinal cohort study (presented at the 2004 CSRS Meeting). *Spine*. 2005;30(22):2564-2569.