ORIGINAL RESEARCH PAPER

Otorhinolaryngology

KEY WORDS: dysphagia, forestier, hyperostosis, DISH, Resnick and Niwayama criteria, osteophyte

WHEN THE SPINE SAYS NO: DYSPHAGIA AS FORESTIER'S UNSEEN SIGNATURE

Dr. Rameeza Bee Z Junior Resident 3, Department of ENT, Sapthagiri NPS University, Bangalore, Karnataka

Dr. Naveen Kumar AG Professor & HOD, Department of ENT, Sapthagiri NPS University, Bangalore, Karnataka

ABSTRACT

Background: Forestier's disease, or Diffuse Idiopathic Skeletal Hyperostosis (DISH), is a non-inflammatory condition characterized by ossification of ligaments, particularly the anterior longitudinal ligament of the spine. While often asymptomatic, it can present with dysphagia when cervical spine involvement impinges on adjacent soft tissues.**Case Presentation:** A 68-year-old male, presented with progressive difficulty in swallowing solid foods over the past few months. X ray and cervical CT imaging, demonstrated prominent ossification from C3 to C6 impinging upon the pharyngeal wall suggestive of Forestier's disease.**Intervention And Outcome:** Conservative management was initiated, including dietary modifications, anti-inflammatory medications, and physiotherapy targeting cervical mobility. Over several weeks, patient reported marked improvement in swallowing and overall comfort.**Conclusion :** This case underscores the importance of considering Forestier's disease in elderly patients presenting with unexplained dysphagia.Timely imaging and multidisciplinary management can lead to symptomatic relief and prevent complications.

INTRODUCTION:

Diffuse idiopathic skeletal hyperostosis (DISH) is a rheumatologic disease, is a idiopathic condition, characterized by spinal osteophyte formations resulting from the ossification of the paravertebral ligaments and muscles. The most common site is anterior longitudinal ligament and the frequency of the disease increases after the 5th decade. Most common regions involved are lower cervical segments leading to dysphagia.1

It can be associated with several metabolic disorders, such as obesity, type 2 diabetes, and highly prevalent among elderly. Forestier- Rotes-Querol disease may be asymptomatic and only diagnosed in the imaging, or it can be associated with clinical conditions like stiffness, pain and reduced spine mobility.There is no specific treatment guidelines at present.2

When it affects the cervical spine, it can result in important ENT manifestations like dysphagia and is usually this condition is undiagnosed. It is most common in elderly male.3

It was first described by Forestier and Rotes-Querol in 1950. Dysphagia results from compression of the tethered portion of the esophagus between the third and seventh cervical vertebrae. It can also cause laryngeal stridor, dyspnea, hoarseness, snoring, neurologic deficits, and aspiration, the latter occurring when the osteophytes are larger than 10mm in size4

In a study by Strasser et al.,1.7% of patients with osteophytes had dysphagia. If dysphagia occurs, the C5-C6 vertebrae are most often involved, followed by C4-C5, C2-C3 AND C3-C4. Surgical resection of hyperplastic osteophytes is an effective method for treating severe dysphagia.5

Diagnosis is usually made with conventional radiography, based on the Resnick and Niwayama criteria: flowing osteophytes over at least four contiguous vertebral bodies, the preservation of intervertebral disk space, absent facet and costovertebral joint ankylosis, and absent sacroiliac joint abnormalities. A "melted candle wax" appearance along the spine is typical of the advanced disease.6

Spine radiography is the investigation of choice for identifying characteristic bone formation. Management of DISH requires a multidisciplinary approach and may include non - pharmacological approaches (eg. Patient education, self management, physiotherapy) pharmacological therapies, and surgery.7

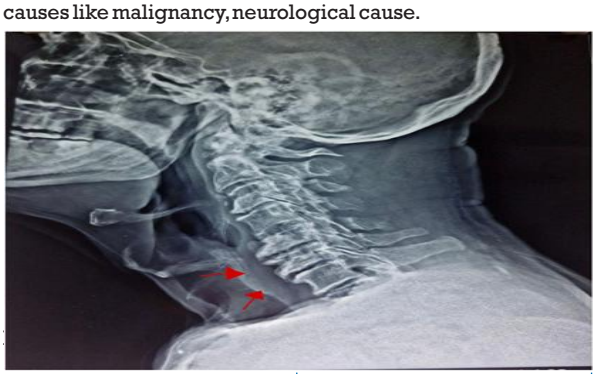
This condition is also called as ankylosing spinal hyperostosis.8

Other important symptoms associated with Forestier syndrome are stiffhess and pain in the back, pain related to tendinitis, myelopathy related to core compression associated with the ossification of the posterior longitudinal ligament, and pain related to vertebral complications such as fracture or subluxation. 9

Surgical treatment is with respect to specific symptoms or complications and generally involves osteophytectomy through the anterior approach. 10

CASE REPORT: A 68-year-old male, presented to ENT OPD, with progressive difficulty in swallowing solid foods in the last three months, no difficulty swallowing liquids or saliva. It was not associated with painful swallowing, difficulty opening of mouth, any swelling in the neck or increased salivation. No significant weight loss, hoarseness of voice, or neurological deficits were reported. On physical examination, patient is moderately built and nourished. On oral cavity examination, loose teeth present, rest of oral cavity normal. Oropharynx normal findings. Videolaryngoscopy showed bilateral vocal cords mobile on phonation and respiration, bilateral pyriform fossa normal, no growth or mass noted. Upper GI endoscopy showed normal study. Routine investigations ruled out esophageal and neurological causes. Patient also had neck pain for which lateral cervical spine X-ray revealed flowing ossifications along the anterior aspect of the vertebrae, suggestive of Forestier's disease. This was confirmed by cervical CT imaging, which demonstrated prominent ossification from C3 to C6 impinging upon the pharyngeal wall.

Diagnosis was done accidentally and after ruling out all causes like malignancy, neurological cause.



vertebrae in which the red arrows depict the ossification of and skeletal outgrowths in the C5 and C6 vertebrae

INTERVENTION AND OUTCOME: Conservative management was initiated, including dietary modifications, anti-inflammatory medications, antacids and physiotherapy targeting cervical mobility. Over several weeks, patient reported marked improvement in swallowing and overall comfort. Continuous monitoring of lifestyle and dietary modification was done with pharmacological therapy. Patient was compliant and hence there was marked improvement in swallowing. Surgical intervention was not needed in this case as the patient had significant improvement and no complications were noted as airway compromise. Counseling of patient was done and reassurance was given.

DISCUSSION

Forestier disease, also known as Diffuse Idiopathic Skeletal Hyperostosis (DISH), is a non-inflammatory systemic disorder characterized by ossification and calcification of ligaments and entheses, particularly the anterior longitudinal ligament of the spine. First described by Forestier and Rotes-Querol in 1950, it predominantly affects elderly males and is frequently associated with metabolic disorders such as diabetes mellitus, obesity, hyperuricemia, hyperlipidemia, and hyperinsulinemia.

Although DISH is relatively common in the elderly population, most patients remain asymptomatic and the condition is often detected incidentally on radiological examination. Symptomatic cervical involvement is uncommon, and dysphagia is the most frequently reported otolaryngological manifestation. Cervical osteophytes causing dysphagia have been reported in only a small proportion of patients with DISH, making Forestier disease an unusual but important differential diagnosis in elderly patients presenting with progressive dysphagia.

The pathophysiology of dysphagia in Forestier disease is multifactorial. Direct mechanical compression of the pharynx and esophagus by large anterior cervical osteophytes is considered the principal mechanism. Additional contributing factors include local inflammation, periesophageal edema and fibrosis, cricopharyngeal spasm, and restriction of epiglottic movement leading to impaired swallowing dynamics. The most commonly affected cervical levels are C3–C6, where osteophytes can significantly compromise the hypopharyngeal and upper esophageal lumen.

Patients typically present with gradually progressive dysphagia, initially affecting solid foods and later progressing to liquids. Associated symptoms may include foreign body sensation, odynophagia, hoarseness, dysphonia, chronic cough, aspiration, dyspnea, stridor, and obstructive sleep apnea. In the present case, the progressive nature of dysphagia and radiological evidence of anterior cervical osteophytes were consistent with previously reported cases in the literature.

Radiological imaging plays a pivotal role in diagnosis. The Resnick criteria remain the standard diagnostic criteria for DISH and include flowing ossification along the anterolateral aspect of at least four contiguous vertebral bodies, preservation of intervertebral disc height, and absence of sacroiliac or apophyseal joint ankylosis. Lateral cervical radiographs and computed tomography are particularly useful for demonstrating the extent of osteophyte formation and degree of aerodigestive tract compression. Barium swallow studies may further delineate the site and severity of esophageal obstruction.

Management depends on symptom severity. Conservative treatment consisting of dietary modification, anti-inflammatory medication, swallowing therapy, and treatment of associated reflux may be adequate for mild symptoms. However, surgical excision of cervical osteophytes remains the treatment of choice for patients with severe dysphagia, significant weight loss, aspiration, airway compromise, or failure of conservative therapy. Most reported cases demonstrate excellent symptomatic improvement following osteophytectomy, although recurrence of osteophytes has been described during long-term follow-up.

This case highlights the importance of considering Forestier disease in the differential diagnosis of unexplained dysphagia in elderly patients. Awareness of this rare entity among otorhinolaryngologists is essential to avoid diagnostic delay and ensure appropriate management. Early recognition through clinical suspicion and radiological evaluation can lead to effective treatment and significant improvement in quality of life.

CONCLUSION

Forestier disease is an uncommon but treatable cause of progressive dysphagia. Cervical osteophyte formation secondary to DISH should be suspected in elderly patients presenting with unexplained swallowing difficulties. Radiological imaging is crucial for diagnosis, and surgical osteophytectomy provides excellent symptomatic relief in appropriately selected patients. This case underscores the importance of considering Forestier's disease in elderly patients presenting with unexplained dysphagia. Timely imaging and multidisciplinary management can lead to symptomatic relief and prevent complications. Awareness of this underdiagnosed entity is essential for clinicians evaluating atypical dysphagia.

REFERENCES:

1. Karaarslan N, Gürbüz MS, ÇalıSkan T, Simsek AT. Forestier syndrome presenting with dysphagia: case report of a rare presentation. Journal of Spine Surgery. 2017 Dec;3(4):723.
2. Oliva F, Agnes MN, Strollo F, Danieli R, Maffulli N. The Diffuse Idiopathic Skeletal Hyperostosis (DISH): A Literature Review on the Rehabilitation Treatment. Muscles, Ligaments & Tendons Journal (MLTJ). 2023 Oct 1;13(4).
3. Pulcherio JO, Velasco CM, Machado RS, Souza WN, Menezes DR. Forestier's disease and its implications in otolaryngology: literature review. Brazilian journal of otorhinolaryngology. 2014;80:161-6.
4. Khashab MA, Liangpunsakul S. Forestier disease: a rare cause of dysphagia. Clinical Gastroenterology and Hepatology. 2009 Mar 1;7(3):e16.
5. Li C, Luo W, Zhang H, Zhao J, Gu R. Case Report: Diffuse idiopathic skeletal hyperostosis with ossification of the posterior longitudinal ligament in the cervical spine: A rare case with dysphagia and neurological deficit and literature review. Frontiers in surgery. 2022 Aug 9;9:963399.
6. Cellina M, Bongetta D, Martinenghi C, Oliva G. Diagnosing Dysphagia in Forestier Syndrome: A Dynamic Digital Radiology Application. Diagnostics. 2025 Nov 27;15(23):3020.
7. Le HV, Wick JB, Van BW, et al. Diffuse idiopathic skeletal hyperostosis of the spine: pathophysiology, diagnosis, and management. J Am Acad Orthop Surg. 2021 Dec 15;29(24):1044-51.
8. Kawauchi E, Yamagata T, Tohda Y. A case of Forestier disease with obstructive sleep apnea syndrome. Sleep and Breathing. 2012 Sep;16(3):603-5.
9. Aydin E, Akdogan V, Akkuzu B, KirbaSI, Ozgirgin ON. Six cases of Forestier syndrome, a rare cause of dysphagia. Acta oto-laryngologica. 2006 Jul 1;126(7):775-8.
10. Balta B, OnbaSı IM, Bilgin B, Akıncı AT. Forestier syndrome causing dysphagia: a case report. Turkish Medical Student Journal. 2024 Feb 22.