Case Report

URF PUBLISHERS
connect with research world

Global Journal of Neurology and Neurological Disorders

https://urfpublishers.com/journal/neurology-and-neurological-disorders

Vol: 1 & Iss: 1

Surgical Treatment of Forestier's Disease as a Cause of Dysphagia: Case Report

Abreu Casas D, Rodriguez de la Paz NJ, Diaz Villalvilla Flores C, Lopez Ayala O and Machado C*

Department of Neurology, Institute of Neurology and Neurosurgery of Cuba, Havana, Cuba

Citation: Abreu Casas D, Rodriguez de la Paz NJ, Diaz Villalvilla Flores C, Lopez Ayala O, Machado C. Surgical Treatment of Forestier's Disease as a Cause of Dysphagia: Case Report. *Global J Neur Neurolog Dis*, 2025;1(1):11-14.

Received: 16 June, 2025; **Accepted:** 24 June, 2025; **Published:** 27 June, 2025

***Corresponding author:** Machado C, Department of Neurology, Institute of Neurology and Neurosurgery of Cuba, Havana, Cuba, E-mail: cmachado180652@gmail.com

A B S T R A C T

Introduction: Forestier's disease, also known as diffuse idiopathic skeletal hyperostosis, is a systemic, non-inflammatory disease of unknown etiology, common in men over 50 years of age and consisting of calcification and ossification of peripheral ligaments and enthesis, with great selectivity for the anterior portion of the spine.

Clinical Case: A 56-year-old male patient with a history of hypertension and type 2 diabetes mellitus, with a seven-year history of progressive dysphagia and rigidity in neck movements. Radiographic, tomographic and magnetic resonance imaging studies showed multisegmented hyperostosis of the anterior longitudinal ligament from C2 to T3, with prominent osteophytes that contact the prevertebral soft tissues, in addition to confirmation of mechanical dysphagia by esophagogram with barium contrast.

Conclusion: Surgical treatment of Forestier's disease has been shown to be highly effective in cases with severe symptoms, such as dysphagia. When conservative treatment fails, the resection of symptomatic anterior osteophytes emerges as a safe and promising procedure for long-term relief.

Keywords: Forestier's disease, Diffuse idiopathic skeletal hyperostosis, DISH, Osteophytectomy, Dysphagia

1. Introduction

Forestier's disease, also known as diffuse idiopathic skeletal hyperostosis, DISH, is a systemic, non-inflammatory disease of unknown etiology, frequently underdiagnosed, consisting of calcification and ossification of peripheral ligaments and entheses, with great selectivity for the anterior portion of the spine[1]. It was initially described in 1950 by Jacques Forestier and his student Jaume Rotes-Querol, as a form of ankylosis of the spine presented in older adults, which they named "senile ankylosing vertebral hyperostosis"[2]. Later in 1976, Resnick et al. attributed to it the term DISH, which is widely recognized today[3]. Forestier's disease typically occurs in males, in individuals over 50 years of age and is associated with risk factors such as type 2 diabetes mellitus, high blood pressure and obesity[4]. It may be asymptomatic or manifest with neck pain, pain at peripheral tendon junction sites, limitation of movement of the spine and in advanced cases in which there is great ossification of the anterior longitudinal ligament (OL) that compresses the digestive tract and airway, dysphagia and respiratory distress may appear[5]. Diagnosis is based on radiographic findings characteristic of new bone formation along the anterolateral aspect of the spine, included in Resnick's imaging criteria; 1) POT that joins at least four contiguous vertebral bodies, 2) preservation of the height of the intervertebral discs and 3) in the absence of inflammatory changes in the sacroiliac or facet joints, in order to differentiate it from ankylosing spondylitis[6]. The treatment of this entity can

vary, offering conservative treatment in asymptomatic cases or with mild symptoms or surgical treatment for those who show worsening or symptoms of compression of the digestive or respiratory tract[7].

2. Case Report

A 56-year-old male patient of urban origin, Caucasian, with a personal pathological history of arterial hypertension and insulin-dependent type 2 diabetes mellitus, monitored to date. He presents with a clinical picture of 7 years of evolution due to gradual and progressive difficulty in swallowing, mainly for solid foods, as well as having noticed a year ago, stiffness of the cervical spine that makes it impossible to perform movements of the neck in its full extension, so it limits recreational sports activities and daily life (Figure 1). He presents to the Neurosurgery consultation after noticing a worsening of his symptoms. After confirming through physical examination, the limitation of neck movement in flexion-extension, rotation and right-left lateralization, imaging studies with Computed Tomography and Magnetic Resonance Imaging are performed (Figure 2).

Figure 1: CT scan of the cervical spine, bone window, sagittal section, where signs of bone degeneration are observed, ossification of the anterior longitudinal ligament from C2 to T3, associated with prominent osteophytes at the level of C2-C3 and C4-C5, which protrude towards the prevertebral soft tissues. Hyperostosis of the anterior longitudinal ligament acquires a thickness of 7mm at the level of C3, C4 and C5.

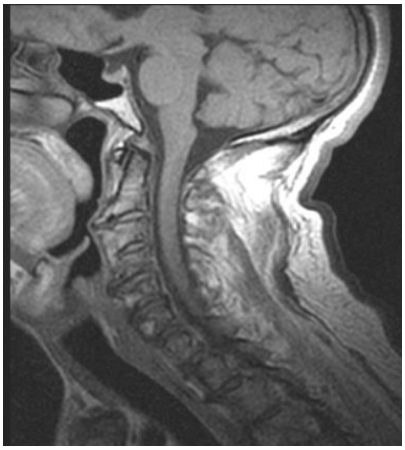

Figure 2: MRI of the cervical spine, weighted in T1, sagittal view, showing the relationship of the large cervical osteophytes at the level of C2-C3 and C4-C5, with the pharyngeal and esophageal soft tissues, respectively.

Subsequently, the patient was evaluated by the specialty of Gastroenterology and an esophagogram study with barium contrast was performed, where compression of the pharynx and cervical esophagus by osteophytes was demonstrated throughout the segment, presenting intervertebral bridges that extended to the anterior wall of the pharynx at the level of C4-C5 (Figure 3).

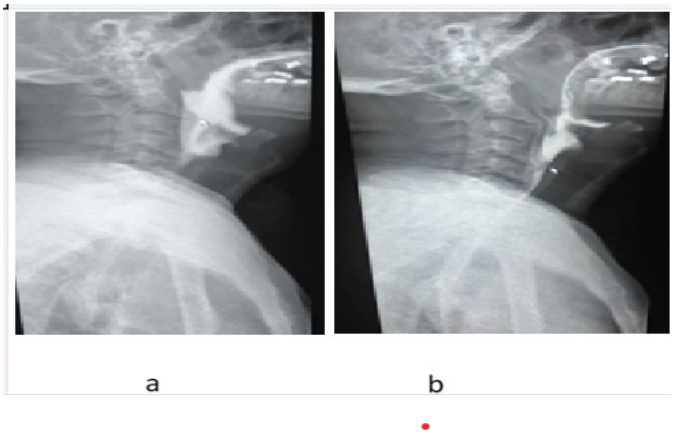

Figure 3: (a) Esophagogram with barium contrast, showing the interruption of the fluid column at the level of the pharynx due to the protrusion of osteophytes at the level C2-C3 and C4-C5. (b) The arrows point to the site of interruption of the flow.

Once the mechanical cause of the patient's dysphagia is confirmed, surgical management is decided for symptom relief.

2.1. Surgical Procedure

The patient is placed in the supine position on the operating table, with the head supported on a horseshoe headrest in hyperextension as much as the patient's cervical spine anatomy allowed, rotated 30° to the left and a scapular wedge is placed to achieve greater exposure of the anterior region of the neck to be approached. It is decided to perform a high anterolateral cervical approach, also known as a right anterior retropharyngeal approach, due to the need to address the high C2-C3 levels and sub axial C4-C5 levels. For this, a longitudinal pre- sternocleidomastoid incision is made, supported by intraoperative fluoroscopy and anatomical reference points: the angle of the mandible as the reference point for the upper end, the medial edge of the Sternocleidomastoid Muscle (SCM) and the thyroid cartilage towards the midline as the reference point for the lower end. The superficial planes of skin, subcutaneous tissue and platysma muscle are dissected in the direction of their fibers. The medial edge of the SCM is identified and the superficial aponeurosis is dissected. Blunt dissection is deepened between the visceral and vasculonervous axis, placing the digastric muscle, which is then divided. The pharyngeal, laryngeal and floor of mouth visceral elements are identified and separated medially and the vasculonervous bundle (comprising the external and internal carotid arteries, the external jugular vein and the hypoglossal nerve) is separated laterally. Subsequently, the prevertebral fascia is identified and dissected along with the longus colli muscles, exposing osteophytes and cervical hyperostosis. Self-retaining retractors and contra retractors are placed and the location of the bony elements to be addressed is confirmed through fluoroscopy. Osteophyte resection is performed at the C2-C3 level and the exposure is then extended to the C4-C5 space, repeating the resection procedure. Rigorous hemostasis is achieved and drainage is placed for 48 hours. The patient is discharged after 24 hours without immediate complications (Figures 4 and 5).

Figure 4: Trans surgical radiograph showing osteophytes at the level of C2-C3 and C4-C5.

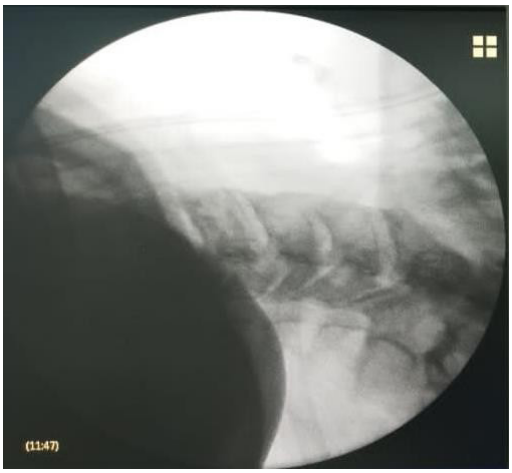

Figure 5: Trans surgical radiograph showing the removal of osteophytes at the level of C2-C3 and C4-C5.

2.2. Evolution

The patient presented a favorable postoperative evolution, showing gradual improvement of the swallowing disorder, which was corroborated in the follow-up consultations at one month, 3, 12, 18 and 24 months after surgery, confirming remission of dysphagia.

3. Discussion

Diffuse Idiopathic Skeletal Hyperostosis (DISH) is a not uncommon condition today, affecting between 5% and 35% of the population, mainly in people over 50 years of age, with a marked preference for males (six times more than in women) and in patients with metabolic syndrome[8,9]. Our patient's case is consistent with what has been reported in the literature and contributes to existing statistics.

The hyperostotic process in DISH develops along the lower half of the anterior border of the vertebral body. Its morphological aspects are described as "descending drop" or "prevertebral drop" in the front of the vertebrae. Subsequently, these types of "droplets" tend to grow, bind and expand over the upper and anterior tracts of the underlying vertebral body, leading to the formation of a "candle flame," "parrot beak," or "bridge" image. Occasionally, the hyperostotic process may involve the odontoid process and adjacent ligaments[10].

The formation of new bone components in the anterior aspect of the cervical vertebrae has been repeatedly reported as causing swallowing disorders, according to the literature review. Dysphagia typically occurs due to structural changes resulting from direct compression of the esophagus and secondarily, due to an inflammatory reaction of the adjacent soft tissue, which can lead to malnutrition and weight loss at an advanced stage. Ventral osteophytes of the cervical spine, after ruling out other potential causes of dysphagia (such as stroke, neurological diseases, tumors, diverticula, stenosis of the pharynx or esophagus), may continue to be the sole reason for a swallowing disorder in multiple cases, with the swallowing of solid food being affected first, coinciding with our case[11,12].

Radiographic, tomographic and magnetic resonance imaging studies are necessary for the diagnosis of Forestier's disease. Additionally, differential diagnostic clarification of extrinsic and intrinsic causes of dysphagia is mandatory, requiring barium swallowing tests and, alternatively, esophageal and laryngeal endoscopies if necessary[13]. Imaging diagnosis of DISH requires the presence of at least two (according to Forestier) or three (according to Resnick) contiguous intervertebral bridges. However, some authors suggest that there must be even earlier stages in which it can be suspected in a patient with a single bone bridge[14,8]. In a review of 24 related papers, the main point of debate was the number of vertebral bodies involved in new bone formation. They concluded that this disease appears to be a progressive condition that probably starts at a single level and progresses slowly to multiple levels, making the number of vertebral bodies needed to establish the diagnosis likely arbitrary[15]. In our patient, hyperostotic bridges were observed at C2-C3, C3-C4, C4-C5 and subsequently at C6-C7, C7-T1, T1-T2 and T2-T3, with a maximum thickness of hyperostosis of 7 mm. This finding aligns with the results of a study involving a series of 10 patients with DISH-related dysphagia associated with myelopathy, where the maximum thickness of hyperostosis was most found at the C4/C5 level, consistent with previous documentation[16].

Various therapeutic options have been described in the literature, considering the need for individualized treatment and the symptoms associated with dysphagia. Conservative treatment is employed in patients with mild and incipient symptoms, consisting of dietary changes, analgesics, muscle relaxants, steroids. Postural changes during swallowing and rehabilitation[7]. Conversely, in progressive or highly symptomatic cases, when the extrinsic cause of compression results in dysphagia associated with weight loss or deterioration of the patient's quality of life, surgical resection of osteophytes via an anterior approach to the cervical spine demonstrates a high success rate, with recovery in most patients. It is reported that between 8% and 10% of patients with DISH- related dysphagia require surgical treatment[8,17].

Our patient underwent surgery at two levels, C2-C3 and C4-C5, with osteophytectomy proving to be an effective, quick procedure with satisfactory results two years postoperatively. In a study of 5 patients surgically treated for DISH-related dysphagia[4], osteophytectomies were performed in all cases, involving one, two and up to four segments, with additional procedures such as discectomies, foraminotomies, intervertebral fusion and instrumentation in cases with root compressive symptoms or indicators of instability. Another study involving 6 patients[8] showed osteophytectomy to be highly effective and recommended in cases of failed conservative management, increased dysphagia with involuntary weight loss and upper

airway obstruction, with follow-up lasting up to 12 years without recurrence of osteophytes.

Among the reported risks in surgery for Forestier's disease, involving the removal of cervical osteophytes causing dysphagia, are hematomas, resection or compression of the superior and/or inferior laryngeal nerves, the hypoglossal nerve or its descending branch and the cervical sympathetic nerve, pharyngocutaneous fistula, esophageal perforation and infections (Ruetten et al., 2019). However, in our patient's case, no immediate, intermediate or late complications were observed, aligning with the low complication rate recorded in the literature.

4. Conclusion

Surgical treatment of Forestier's disease has been shown to be highly effective in cases with severe symptoms, such as dysphagia. When conservative treatment fails, the resection of symptomatic anterior osteophytes emerges as a safe and promising procedure for long-term relief. This approach not only ensures high patient satisfaction but also leads to significant improvements in quality of life.

5. References

1. Tripathi M, Rajmohan D, Quirk C, et al. Diffuse Idiopathic Skeletal Hyperostosis, Associated Morbidity and Healthcare Utilization: A University Hospital Experience. JCR: J Clin Rheumatology. 2020;26(3): 104-108.
2. Anshori F, Hutami WD, Tobing S DAL. Diffuse idiopathic skeletal hyperostosis (DISH) with ossification of the posterior longitudinal ligament (OPLL) in the cervical spine without neurological deficit - A Case report. Annals of Med Surg. 2020;60: 451-455.
3. Mader R, Verlaan JJ, Buskila D. Diffuse idiopathic skeletal hyperostosis: Clinical features and pathogenic mechanisms. Nature Reviews Rheumatology. 2013;9(12): 741-750.
4. Scholz C, Naseri Y, Hohenhaus M, et al. Long- term results after surgical treatment of diffuse idiopathic skeletal hyperostosis (DISH) causing dysphagia. J Clin Neuroscience. 2019;67: 151-155.
5. Artner J, Leucht F, Cakir B, et al. Diffuse idiopathische skelettale Hyperostose: Aktuelles zur Diagnostik und Therapie. Der Orthopäde. 2012;41(11): 916-922.
6. Mc Cafferty RR, Harrison MJ, Tamas LB, et al. Ossification of the anterior longitudinal ligament and Forestier's disease: An analysis of seven cases. J Neurosurg. 1995;83(1): 13-17.
7. Valdez C, Valdez A. Forestier's disease in patients over 65 years of age: Case report with follow-up after 6 years of conservative management. Rev Med Chile. 2018.
8. Lui Jonathan YC, Sayal P, Prezerakos G, et al. The surgical management of dysphagia secondary to diffuse idiopathic skeletal hyperostosis. Clinical Neurology and Neurosurgery. 2018;167: 36-42.
9. Adel H, Khan SA, Adil SO, et al. CT-Based Evaluation of Diffuse Idiopathic Skeletal Hyperostosis in Adult Population; Prevalence, Associations and Interobserver Agreement. J Clin Densitometry, 2020;23(1): 44-52.
10. Mader R, Baraliakos X, Eshed I, et al. Imaging of diffuse idiopathic skeletal hyperostosis (DISH). RMD Open. 2020;6(1): 001151.
11. Seidler TO, Perez Alvarez JC, Wonneberger K, et al. Dysphagia caused by ventral osteophytes of the cervical spine: Clinical and radiographic findings. European Archives of Oto-Rhino-Laryngology. 2009;266(2): 285-291.
12. Ruetten S, Baraliakos X, Godolias G, et al. Surgical treatment of anterior cervical osteophytes causing dysphagia. Journal of Orthopaedic Surgery. 2019;27(2): 2309499019837424.
13. Damade C, Masse R, Ghailane S, et al. Anterior Cervical Idiopathic Hyperostosis and Dysphagia: The Impact of Surgical Management-Study of a Series of 11 Cases. World Neurosurgery, 2020;138: 305-310.
14. Mazieres B. Diffuse idiopathic skeletal hyperostosis (Forestier-Rotes- Querol disease): What's new? Joint Bone Spine. 2013.
15. Kuperus JS, De Gendt EEA, Oner FC, et al. Classification criteria for diffuse idiopathic skeletal hyperostosis: A lack of consensus. Rheumatology. 2017;56(7): 1123-1134.
16. Glinski AV, Takayanagi A, Elia C, et al. Surgical Treatment of Ossifications of the Cervical Anterior Longitudinal Ligament: A Retrospective Cohort Study. Global Spine Journal, 2021;11(5): 709-715.
17. Kaur J, Virk JS. Dysphagia due to DISH-related anterior osteophytes: DISHphagia!! BMJ Case Reports. 2017: 222512.