

Case Report



A Case Report of Early Surgical Intervention for Diffuse Idiopathic Skeletal Hyperostosis of the Cervical Spine: Challenges in Management

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ABSTRACT

Background and Aim: Diffuse idiopathic skeletal hyperostosis (DISH) can compress the trachea and esophagus when located in the cervical spine. In this report, we investigated whether it is preferable to perform the early surgical intervention in symptomatic patients or to wait and administer supportive care and perform late surgical intervention regardless of whether symptoms progress or not.

Case Presentation: We present the case of a 70-year-old patient with diffuse idiopathic skeletal hyperostosis (DISH) causing significant dysphagia and unilateral vocal cord paresis, resulting in dyspnea and stridor. Imaging diagnostics revealed large osteophytes anterior to the cervical spine from C3 to C6 compressing the cervical spine. Significant clinical improvement was observed following the anterior resection of the patient's osteophytes.

Conclusion: In order to achieve higher success and less recurrence, it is preferable to perform surgical intervention earlier in the disease's progression. However, more studies are necessary to confirm this because most of the present results are from case report articles and have less evidence.

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Highlights

- Diffuse idiopathic skeletal hyperostosis (DISH) is asymptomatic, although cervical DISH may present with secondary dysphagia.
- The objective of this study was to evaluate whether early surgical intervention in symptomatic patients, particularly in patients with secondary dysphagia, results in improved clinical outcomes.
- Further comprehensive investigations in the future could provide insight into the optimal timing for surgical intervention to manage cervical DISH.

Plain Language Summary

Diffuse idiopathic skeletal hyperostosis (DISH) can cause breathing and swallowing problems when it affects the neck. This report examines whether it is better to do surgery early on in patients with symptoms or to wait and provide supportive care until later. The report describes a case of a 70-year-old patient with DISH who had trouble swallowing and breathing due to bone growths in their neck. Surgery to remove the growths improved their symptoms. Doing surgery earlier can be better but more research is needed to confirm this.

1. Introduction

Diffuse idiopathic skeletal hyperostosis (DISH, or Forestier's disease) is characterized by bony hardening at sites of tendon and ligament insertion around the vertebral column [1]. Resnick and Niwayama's diagnostic criteria for DISH include "four continuous levels of ossification, relative preservation of disc spaces, and the absence of apophyseal joint ankylosis, and sacroiliac sclerosis" [2].

DISH is typically asymptomatic and discovered incidentally on spine radiography [3]. However, cervical DISH can cause annoying symptoms for the patient related to airway or neural compression at the spine, such as dysphagia, dysphonia, paralysis of the vocal cords, dyspnea, snoring, or decreased range of motion [3-5]. Anterior cervical osteophyte resection is indicated in patients with airway obstruction and/or dysphagia in whom conservative treatment fails.

The following is a report of a rare case in which early surgical intervention in a patient with cervical DISH yielded favorable outcomes with minimal complications. Based on previously published research, we investigated whether it is preferable to perform the early surgical intervention in symptomatic patients, especially patients with secondary dysphagia, or to wait and administer supportive care and perform late surgical intervention regardless of whether symptoms progress.

2. Case Presentation

A 70-year-old man presented to our clinic complaining of neck pain, swallowing difficulties, and voice changes. According to his medical history, he had arterial hypertension and hypercholesterolemia. In addition, he had no other co-morbidities and his diabetes test was negative. He had experienced mild symptoms, such as dysphagia for several years. His dysphagia and hoarseness worsened in the last two months. In addition to experiencing painful swallowing, the patient had also lost a significant amount of weight over the past month. On physical examination, movements of the cervical spine, such as lateral bending and axial rotation, were painful, and the patient complained of muscle stiffness. No motor or sensory deficits were present, and deep tendon reflexes were normal.

Concerning the patient's trouble speaking and other issues, a laryngoscopy was performed on him, confirming the diagnosis of unilateral left-side vocal cord paresis (Figure 1). We obtained a computed tomography (CT) scan (Figure 2), radiographs, and magnetic resonance imaging (MRI) of the cervical spine (Figure 3). Normal bone density and the presence of new bone formation at C2-C3, and C4-C6 vertebrae with anterior fusion appeared on the lateral cervical X-ray (Figure 4). The cervical spine was scanned using T1 and T2 images in the sagittal and axial planes. The radiologist noted disc bulging with moderate compression effect on the central canal and neural foramina at different levels from C1 to C6; two enormous osteophytes, one measuring ap-



Table 1. Summary of related articles

Study	Year	TNP	LSI	LSI.R	ESI	ESI.R	Design	Country	Mean Age (y)	Mean Follow-up (mo)	Male/Female	Complications
Mattioli et al. [9]	2020	21	9	5	12	0	Retrospective	Italy	70.6	66	18/3	2 Hemorrhages 1 Tracheotomy 4 Wound Infection
Jonathan et al. [15]	2018	6	1	0	5	1	Retrospective	UK	59	42.3	6/0	1 Hematoma
Myamoto et al. [12]	2009	7	2	1	5	0	Prospective	Japan	65	108	6/1	N/S
Yoshioka et al. [18]	2018	4	3	1	1	0	Retrospective	Japan	67.3	N/S	N/S	N/S
Nelson et al. [16]	2006	5	2	0	3	0	Retrospective	USA	78.8	12	4/1	N/S
Scholz et al. [17]	2019	5	3	3	1	1	Retrospective	Germany	61.6	70.3	5/0	1 Hoarseness



Abbreviations: TNP: total number of patients; LSI: late surgical intervention; LSI.R: late surgical intervention with remaining of symptoms after operation; ESI: early surgical intervention; ESI.R: early surgical intervention with remaining of symptoms after operation; N/S: not specified

proximately 25.3 mm on C2-C4 and the other measuring approximately 10.3 mm on C5-C6, and a moderate to severe anterior compression effect on the pharynx region, consistent with a DISH diagnosis of disc dehydration and degenerative changes. However, no proof of cervical myelopathy was found. After consulting with a gastroenterologist to check the differential diagnoses of secondary dysphagia, the cervical DISH diagnosis topped the list of differential diagnoses. He was referred to the neurosurgery clinic for a surgical consultation and future care.

Surgery

The surgical technique was done using the Smith-Robinson approach. We used a generous horizontal mid-cervical skin incision. Platysma was split longitudinally. This allowed generous access from C3 down to C6. We

were able to achieve satisfactory exposure through a horizontal incision. By elevating the longus colli, the lateral aspects of the osteophytes were exposed. No adhesions to the esophagus existed. To remove osteophytes, we used a high-speed drill. Standard suturing was used. The procedure was performed by a neurosurgeon with 10 years of experience in spine surgery. The day after the operation, the patient reported a significant improvement in swallowing and a reduction in local pain in the cervical spine area.

Quality of life (QoL)

We followed up the patient at intervals of 2, 6, and 12 months after surgery. A lateral cervical radiograph was performed for the patient, which showed no evidence of regrowth. The neurological examination was normal, and the patient’s dysphagia and sleep quality had also

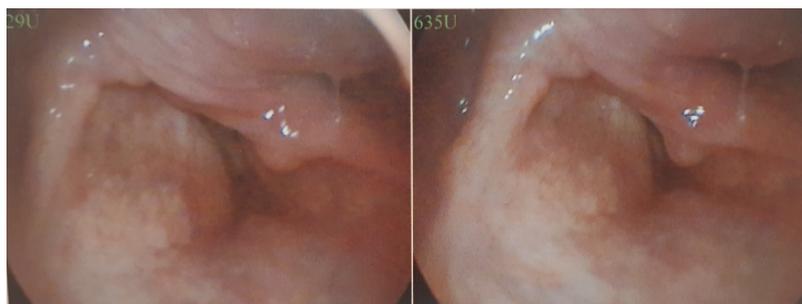


Figure 1. Concerning the patient’s trouble speaking and other issues

Laryngoscopy was performed on him, confirming the diagnosis of unilateral left side vocal cord paresis.





Figure 2. Patients computed tomography (CT) scan pointing to the pathology

improved significantly. According to him, he had gained a little extra weight, he was happier and did not wake up with shortness of breath. He is now more satisfied with his life. The patient reported a significant improvement in swallowing, breathing, and cervical spine range of motion.

3. Discussion

Clinical manifestations of DISH may include musculoskeletal symptoms, such as neck and back pain, cervical involvement symptoms like dysphagia, hoarseness,



Figure 4. Normal bone density and the presence of new bone formation at C2-C3, C4-C6 vertebrae with anterior fusion on the lateral cervical X-ray

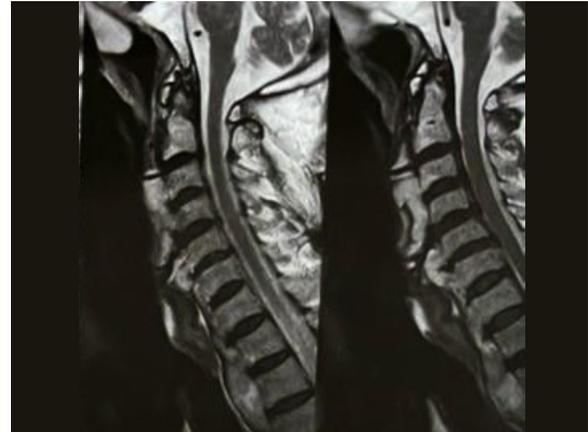


Figure 3. Patients cervical magnetic resonance imaging (MRI)

sleep apnea, and neurological defects due to ossification of the posterior longitudinal ligament (OPLL) [6]. Mechanical factors, some dietary factors, such as retinol and its medical derivatives, high levels of osteocalcin, and lower levels of dickkopf-1 (DKK1) are suspected to be involved in the development of DISH. Spondylosis deformans, ankylosing spondylitis, and OPLL are conditions that present similarly to DISH and should be carefully evaluated [7].

The incidence of dysphagia in DISH patients ranges from 0.2% to 28.2%. DISH occurs between 2% and 4% of the time in patients older than 40 years, and this rate reaches 28% in patients older than 80 years old [8]. When mild symptoms, such as mild pain, stiffness, and decreased range of motion are present, DISH can be managed pharmacologically with non-steroidal anti-inflammatory drugs (NSAIDs) and local glucocorticoid injections, and conservatively with physical therapy [9]. Certain conditions necessitate a surgical consultation to determine if an intervention is appropriate [10-13]. These conditions include dysphagia resulting from cervical osteophytes and spurs, OPLL, and neurological defects resulting from nerve root compression or thoracic outlet syndrome [10-12, 14].

According to previously published studies, this study was conducted to determine whether or not early surgical intervention in symptomatic patients, particularly in patients with secondary dysphagia, yields better clinical outcomes than late surgical intervention when symptoms have progressed to OPLL or thoracic outlet syndrome (TOS) or become more severe. Julian L. Genreau et al. [8] conducted a systematic review and meta-analysis of 22 papers reporting 119 DISH patients with dysphagia. Most papers included case reports or case

series. According to their findings, after surgery, 89% of patients were relieved of their symptoms. In the remaining 11% of patients, failure to alleviate dysphagia after the surgical intervention was associated with a longer duration of preoperative symptoms ($P < 0.01$) and increased severity of preoperative symptoms ($RR = 2.86$, 95% CI: 1.19-6.09, $P = 0.02$). Their findings were consistent with those of Kei Miyamoto et al. [12], who investigated the recurrence of osteophytes in seven DISH patients with dysphagia. In their study, patients with more severe symptoms and a longer postoperative period did not experience relief. They also concluded that as symptoms progress, the likelihood of osteophyte recurrence after surgery increases. Therefore, it is preferable to monitor patients for at least 10 years after surgery.

Additionally, Francesco Mattioli et al. [9] examined 41 DISH patients who underwent surgical intervention. In their study, patients with more severe and advanced dysphagia did not benefit as much from the intervention as patients with milder symptoms. They concluded that early surgical intervention has a higher success rate and therefore benefits patients.

As described in Table 1, patients who were symptomatic for longer before surgical intervention also experienced more post-operative complications. Since the resection of osteophytes is a simpler surgical intervention, it seems that surgical intervention in the early stage of cervical DISH and before the appearance of neurological deficits or respiratory symptoms can be associated with a lower rate of post-operative complications [9, 12, 15-18].

4. Conclusion

According to our experience and a review of the relevant literature, to achieve a higher success rate and less recurrence, it is preferable to perform surgical intervention earlier in the disease's progression. But more studies are necessary to confirm this as most of the present results are from case reports and have a lower level of evidence.

Ethical Considerations

Compliance with ethical guidelines

Written informed consent was obtained from the patient before the operation and for the rest of the procedure.

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Authors' contributions

Conception and design: Misagh Shafizad, Rostam Poormousa; Data Analysis and interpretation: Misagh Shafizad, Pooria Sobhanian; Drafting the article: Pooria Sobhanian, Erfan Ghadirzadeh; Critically revising the article: Pooria Sobhanian, Erfan Ghadirzadeh; Reviewing submitted version of manuscript: Rostam Poormousa, Gholam-ali Godazandeh; Approving the final version of the manuscript: Misagh Shafizad, Gholam-ali Godazandeh.

Conflict of interest

The authors declared no conflict of interest.

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