

Surgical Treatment for Cervical Diffuse Idiopathic Skeletal Hyperostosis as a Cause of Dysphagia

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Abstract:

Introduction: The majority of diffuse idiopathic skeletal hyperostosis (DISH) involving the anterior margin of the cervical vertebrae is asymptomatic, but it can cause dysphagia. Improvements in swallowing after surgical treatment have been reported in several case series. However, the appropriate amount of osteophyte resection for this disease in terms of the pathophysiology of dysphagia is still unknown. The current report describes the appropriate surgical procedure for dysphagia secondary to anterior cervical hyperostosis, and discusses the etiology of dysphagia.

Methods: This is a retrospective review of four patients who presented with complaints of dysphagia secondary to anterior cervical hyperostosis. All patients underwent videofluoroscopic esophagrams (VFEs) to identify the specific region associated with the dysphagia. Esophageal obstruction was present at C3-4 in two patients and at C4-5 in two patients. Three patients underwent localized and limited resection of the anterior cervical osteophytes. One patient underwent total resection of the anterior cervical osteophytes, because re-ossification had occurred after a previous resection.

Results: Postoperative VFE demonstrated an improvement in swallowing in the three patients who underwent limited resection of the osteophytes. The patient who underwent total resection of the osteophytes did not experience a full recovery of normal swallowing function. We concluded that the dysphagia was caused by both osteophyte obstruction and neuropathy resulting from the previous surgery or inflammation secondary to osteophyte irritation.

Conclusions: Localized and limited resection of anterior cervical osteophytes is recommended and should be considered for patients with dysphagia from anterior cervical hyperostosis.

Keywords:

Diffuse idiopathic skeletal hyperostosis (DISH), Ossification of the anterior longitudinal ligament (OALL), Dysphagia

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) involving the cervical spine, also known as Forestier's disease¹⁾, is a noninflammatory enthesopathy of unknown etiology. The diagnosis of DISH is established when the soft tissues of the anterolateral thoracolumbar spine are ossified in at least four contiguous segments, with flowing vertebral bony bridges and without obvious signs of intervertebral or apophyseal degenerative changes²⁾. The vast majority of patients with cervical osteophytes are asymptomatic. In 1926, Mosher was the first to report a case of dysphagia due to a cervical spine osteophyte³⁾. In 1938, Lglauer reported the first surgical excision of a cervical spine osteophyte that was causing

dysphagia⁴⁾. Dysphagia appears to be the most common presentation, though only a subset 0.1%-6% develops dysphagia due to cervical osteophytes⁵⁻⁷⁾. However, a comprehensive discussion of the optimal surgical procedures according to pathogenesis is yet lacking.

The aim of our study was to evaluate the appropriate extent of surgical resection of DISH-associated anterior osteophytes causing dysphagia using preoperative and postoperative videofluoroscopic esophagram (VFE) findings.

Subjects and Methods

Four patients with DISH and anterior cervical osteophytes presenting with the chief complaint of dysphagia are re-

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Table 1. Data of the Investigated Patients.

Case	Age	Level of obstruction	Disease duration	Dysphagia	Cough reflex	Pharyngeal movement	Epiglottic movement	Videofluoroscopic assessment			Surgery
								vallecula	pyriform sinuses	esophageal inlet	
1	72	C3/4	2 years	liquid/solid bolus	decrease	n.p.	n.p.	retention	n.p.	n.p.	partial osteophytes resection
2	68	C4/5	1 year	liquid/solid bolus	n.p.	n.p.	n.p.	n.p.	retention of the right side	n.p.	partial osteophytes resection
3	68	C4/5	2 years	solid bolus	n.p.	n.p.	n.p.	n.p.	n.p.	retention	partial osteophytes resection
4	61	C3/4	8 months	liquid/solid bolus	absence	n.p.	impaired movement	retention	retention of the right side	retention	extensive osteophytes resection

ported herein (Table 1). All four patients were men ranging in age from 61 to 72 years (mean age, 67.3 years). All patients complained of gradually progressing dysphagia for more than one year. Nonoperative management using a restricted diet of mainly soft foods and liquids was attempted in all patients; however, their complaints persisted. Patients were then referred to the authors' institute for possible spinal surgical evaluation. All patients were also preoperatively assessed for gastroenterological or otolaryngological issues in order to exclude any intrinsic factors. In all four patients, lateral plain radiographs of the thoracic spine showed flowing ossification of the anterior longitudinal ligament along the anterior vertebral bodies involving four or more contiguous levels, consistent with the diagnosis of DISH. Lateral plain radiographs of the cervical spine and sagittal computed tomography (CT) reconstruction showed protruding beak-like osteophytes at C3/4 in one patient, and at C4/5 in two patients. Case 4 involved the postsurgical recurrence of an osteophyte, which was most protruding at C3/4. Ossification of the posterior longitudinal ligament (OPLL) was identified by CT scans of the cervical spine in all patients. MR imaging demonstrated compression of the spinal canal in two patients. One of these patients (case 2) presented with mild clumsiness of the hands. VFE showed mechanical obstruction at the level of C3/4 in two patients (cases 1, 4), C4/5 in two patients (cases 2, 3). Although three cases revealed normal pharyngeal movement and normal laryngeal elevation, one case (case 4) demonstrated impaired laryngeal elevation. All four patients underwent surgical osteophyte resection.

Results

Three patients underwent localized and limited resection of the anterior beak-like osteophytes at the level of the obstruction. Osteophylectomy was performed using a combination of osteotomes and diamond burs. The goal of reducing the mass was not to match the native anterior vertebral contour, but instead to match the level of the adjacent osteophytes. Postoperative VFE at postoperative week two or

three demonstrated remarkable improvement in swallowing function in these three patients. There were no complaints of obstruction in the transit of both solid and liquid boluses. Alternatively, one patient underwent total resection of the anterior cervical osteophytes from C2 to T1 because of reossification subsequent to a previous partial resection. However, this patient did not experience a full recovery of normal swallowing function.

Case 1

A 71-year-old male presented with a one-year history of slowly progressing dysphagia. He complained of difficulty in swallowing both liquid and solid diets with concomitant weight loss. Reduced mobility of the cervical spine was revealed on physical examination and X-ray. X-ray and CT reconstruction showed an anterior hyperostosis from C2 to C7 (Fig. 1a). The largest bony excrescences were 31 mm thick at the level of C3/4. VFE showed contrast medium residue in the valleculae at the level of C3/4 (Fig. 1b), but no retention in the pyriform sinuses or esophageal inlet. Pharyngeal movement and laryngeal elevation were normal. We concluded that the dysphagia was caused by mechanical obstruction due to the beak-like osteophytes at the level of C3/4. The patient underwent localized and limited resection of the anterior beak-like osteophytes from C3 to C4 (Fig. 1c). Postoperative VFE showed no remaining obstruction and he had no complaints of dysphagia during the swallowing of both solid and liquid boluses at follow-up (Fig. 1d).

Case 4

A 62-year-old male presented with recurrence of dysphagia seven years after initial osteophyte resection. He complained of difficulty in swallowing both liquid and solid diets with concomitant weight loss for four months. Reduced mobility of the cervical spine was revealed on physical examination and X-ray. X-ray and CT reconstruction showed an anterior hyperostosis from C2 to T3, especially

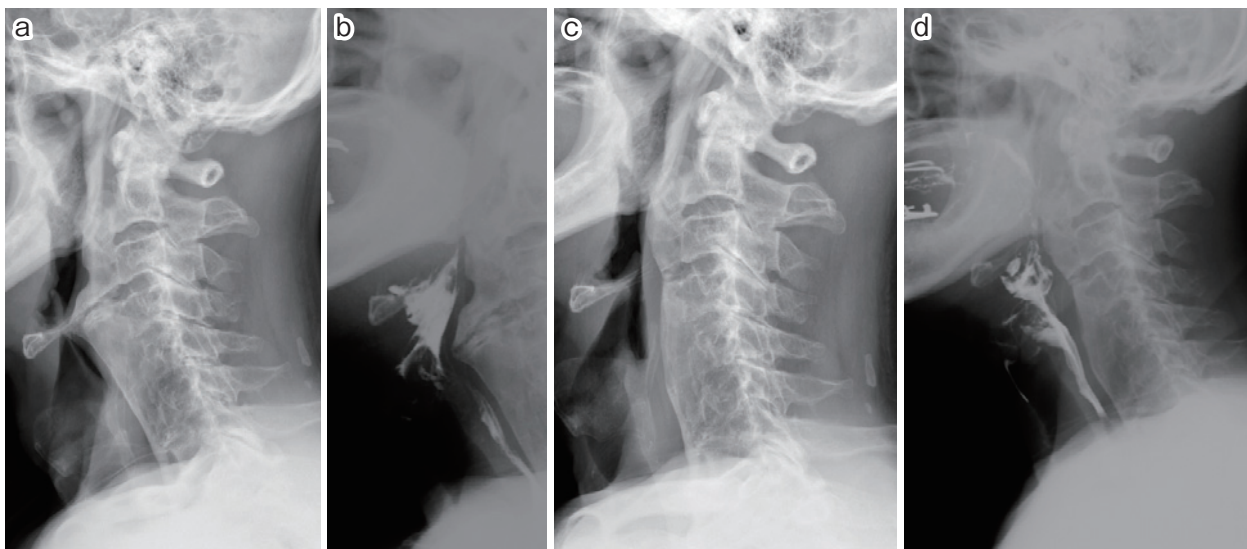


Figure 1.
 Preoperative lateral radiograph of the cervical spine (a).
 Preoperative VFE shows obstruction at C3/4 (b).
 Postoperative lateral radiograph of the cervical spine (c).
 Postoperative VFE shows no obstruction (d).

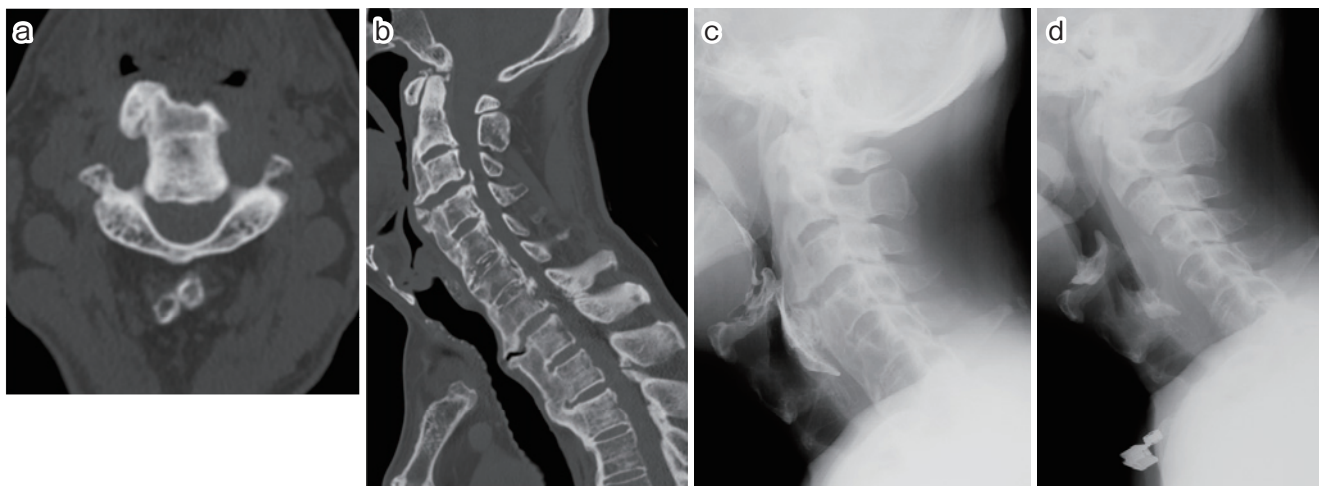


Figure 2.
 Preoperative axial cervical CT (a).
 Preoperative sagittal cervical CT (b).
 Preoperative VFE shows obstruction at C3/4 (c).
 Postoperative VFE revealed residue in the right side pyriform sinus, and esophageal inlet (d).

on the right side (Fig. 2a, b). VFE showed contrast medium residue in the valleculae, right side pyriform sinus, and esophageal inlet (Fig. 2c). He had normal pharyngeal movement but impaired laryngeal elevation. We concluded that the dysphagia was caused by two pathologies: First, neuropathy induced by the previous surgery or inflammation secondary to osteophyte irritation; and second, mechanical obstruction due to regrowth of the osteophyte on the right side. Complete surgical removal of the osteophytes was performed from C2 to T1 because of the recurrent nature of the case. VFE revealed no obstruction at the level of C3 to C4 (no contrast medium residue in the valleculae), and reduc-

tion in contrast medium residue in the right side pyriform sinus was observed three weeks postoperatively (Fig. 2d). However, a slight increasing of contrast medium residue in the esophageal inlet was identified. The patient achieved a slight improvement of the dysphagia one year after surgery. His dysphagia remained unchanged for the next year.

Discussion

The pathogenesis of dysphagia caused by anterior cervical hyperostosis is hypothesized to be the result of direct compression of the pharyngoesophageal segment. Seidler et al.

reported that osteophytes larger than 10 mm thickness lead to symptoms⁸). However, case 1 had no complaint until the size of osteophytes progressed to 31 mm. This observation indicated that there is no direct correlation between the size of osteophytes and the severity of symptoms. Rather, the osteophyte-size that causes dysphagia differs by site and individual. Furthermore, there are other mechanisms that cause dysphagia. Several possible explanations other than direct obstruction have been put forward: Several studies suggested neuropathy due to recurrent laryngeal nerve injury as a result of ongoing inflammation and fibrosis of the esophagus secondary to osteophyte irritation as a cause⁹⁻¹⁴). One case report indicated a palsy of the left vocal cord due to pressure on the recurrent laryngeal nerve¹⁵). Verlaan et al. analyzed 118 publications with a total of 204 patients between 1980 and 2009¹⁶) and reported that swallowing dysfunction remained in only 13% of the patients after surgery. Likewise, in our case series, subjective symptoms showed complete improvement in three cases, with only case 4 showing persistent dysphagia following surgery. Preoperative VFE of case 4 revealed impaired laryngeal elevation. In postoperative VFE of case 4, impaired epiglottic movement was still observed, which suggested that pre-existing neuropathy was a cause of dysphagia. No contrast residue was observed in the valleculae and less contrast medium at the pyriform sinuses. This suggested that osteophyte was the mechanical cause of dysphagia. However, there was slight worsening of dysphagia in the esophageal inlet. Therefore, we hypothesized that extensive, total resection of the anterior cervical osteophytes may increase the risk of damage to pharyngeal branch of the vagus nerve, which, in our case, already had pre-existing neuropathy as a sequelae of the previous surgery that created prolonged inflammation and adhesion. Given the possibility that there is a factor other than mechanical compression that caused persistence of the symptoms even after surgical removal of the osteophytes, careful preoperative evaluation of pharyngeal movement, laryngeal elevation, and other factors is essential, even in the case of large, evident osteophytes.

There is a controversy in the literature concerning the appropriate treatment of dysphagia due to Forestier's disease¹⁰). The recommended first-line approach has been observation or diet modification and a regimen of anti-inflammatory medications¹⁷). Resection of the offending anterior osteophytes has represented the typical surgical intervention¹⁶). However, previous reports did not address the extent of the osteophyte resection in detail^{16,18}). Some surgeons prefer the complete resection of all cervical spine osteophytes¹⁹⁻²¹). However, it was also reported that dysphagia persisted unchanged following surgery with extensive C3-6 osteophyte resection²⁰). We contend that extensive osteophyte resection is more disadvantageous than it is beneficial for the following two reasons: First, there is a possibility of neuropathy because the nerve is compressed for an extended period of time. Second, dysphagia tends to be more frequent in the C3 to C4 and 4 to C5 regions. Thus, we advocate for the local-

ized and limited resection of osteophytes based on VFE as the most appropriate treatment.

Miyamoto et al. reported that patients with DISH who were treated with surgical resection of the anterior cervical osteophytes without fusion developed radiological recurrence¹²). However, only two patients became subsequently symptomatic, both with a minimum follow-up period of 10 years. On the other hand, Urrutia reported no radiological recurrence after anterior cervical osteophyte resection without fusion at one to nine years follow-ups²²); however, in this previous study the cause and pathology of dysphagia was not discussed in detail. Several reports have insisted that appropriate management of this condition requires cervical fusion^{12,23}), as ligamentous ossification is believed to progress until rigidity is established. Nicolas H et al. also reported six cases of osteophylectomy and fusion²⁴). However, the possibility of adjacent segment osteophyte development after fusion surgery remains unclear.

In the elderly population, osteophytes tend to be slow growing, usually asymptomatic, and accompanied by dysphagia. It is necessary to accurately identify the site of mechanical obstruction using preoperative VFE if mechanical compression from anterior cervical osteophyte is the cause of dysphagia. Our first choice of strategy is partial osteophyte resection of the causative site without fusion. Osteophyte resection with fusion may be considered for younger patients with intervertebral mobility as a risk factor for recurrence.

Conclusion

We reported four cases of dysphagia secondary to anterior cervical hyperostosis. We recommend the localized and limited resection of anterior cervical osteophytes according to pathogenesis. Wide surgical exposure and extensive anterior cervical osteophyte resection carry the risk of the pharyngeal branch of the vagus nerve damage due to the possibility of neuropathy from long term suppression. In such cases, VFE is necessary to accurately evaluate the cause of dysphagia.

Conflicts of Interest: The authors declare that there are no conflicts of interest.

Author Contributions: Katsuhito Yoshioka wrote and prepared the manuscript, and all of the authors participated in the study design. All authors have read, reviewed, and approved the article.

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