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Diffuse Idiopathic Skeletal Hyperostosis: A Case with Dysphonia, Dysphagia and Myelopathy

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Male, 66
Final Diagnosis: Diffuse idiopathic skeletal hyperostosis
Symptoms: Dysphagia • dysphonia • myelopathy
Medication: —
Clinical Procedure: X-ray computed tomography
Specialty: Orthopedics and Traumatology

Objective: Rare disease
Background: Diffuse idiopathic skeletal hyperostosis (DISH) is characterized by the ossification of soft tissues, primarily the ligaments and entheses. Exuberant osteophyte formation of the anterior longitudinal ligament of the spine is usually found. Among the reported complications of cervical osteophyte, dysphagia is the most frequent symptom, and dysphonia is rare.

Case Report: A 66-year old male was suffering from progressive dysphonia, dysphagia, and myelopathy. Anterior cervical osteophytes and ossification of the posterior longitudinal ligament (OPLL) was shown on x-ray and computed tomography (CT). He was diagnosed with DISH and the osteophytes were resected. The patient's symptoms gradually improved.

Conclusions: DISH may induce varying symptoms and surgical intervention is a good way to relieve these symptoms. We rarely see the symptoms of dysphonia, but we should consult with other professionals, such as otolaryngologist and dietician, when treating DISH patients.

MeSH Keywords: Cervical Vertebrae • Deglutition Disorders • Dysphonia • Hyperostosis, Diffuse Idiopathic Skeletal • Ossification of Posterior Longitudinal Ligament • Spinal Cord Compression

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/913792>

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Background

Diffuse idiopathic skeletal hyperostosis (DISH) is a common disorder of unknown etiology that is characterized by osteophyte formation of ligament, tendon, or joint capsule insertion, and can cause back pain and spinal stiffness.

Dysphagia is the most frequently reported complications of DISH and occurs in about 30% of patients with cervical osteophytes. Other symptoms, such as a cough, sore throat, and sleep apnea, are also described in these patients, but dysphonia is a rare complication.

We herein report a rare Japanese case of DISH with progressive dysphonia, dysphagia, and myelopathy. Surgical treatment was performed via the anterior approach and showed good results.

Case Report

A 66-year-old male with a history of brain infarction, arrhythmia, hypertension, and sleeplessness presented with a 2-month history of progressive dysphonia, dysphagia, and gait disturbance. He only made conversation by means of writing. An examination revealed hyper-reflex of both extremities; however, there was no muscle weakness or sensory disturbance. He felt slight gait disturbance and was afraid of falling down, so a walking stick or cane was indispensable in his daily life.

Neck x-ray and computed tomography (CT) showed a massive osteophyte of the anterior longitudinal ligament at C4 to T1 and compressing the pharynx at the C3/C4 level (Figures 1, 2).



Figure 1. X-ray showed anterior cervical osteophytes at the C3/C4 level.

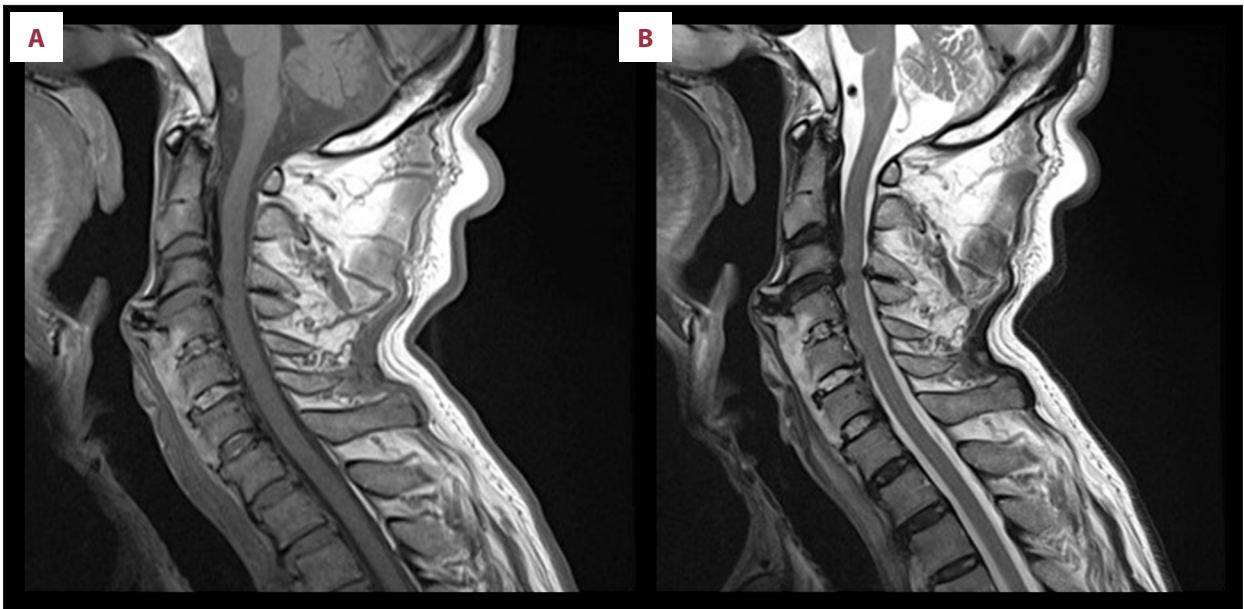
Ossification of the posterior longitudinal ligament (OPLL) was also noted in the spinal canal. Unfavorable instability was also found in this place. Magnetic resonance imaging (MRI) showed slight compression of the cervical cord at the C3/C4 level by OPLL (Figure 3A, 3B). A fiberscope examination by an otolaryngologist revealed protruding submucosal mass in the posterior pharyngeal wall and vocal cord dysfunction (Figure 4). The patient had a high risk of aspiration regardless of meal form, so it was hard for him to take anti-inflammatory drugs.

An operation to remove the anterior osteophyte and a discectomy at the C3/C4 level and fusion were performed to reduce the pharynx compression and diminish the unfavorable movement of the OPLL. The patient was placed in the supine position with neck extension. A collar incision was made at the left side by retracting the omohyoid muscle and pharynx, and the C3/C4 osteophyte was easily palpable just posterior to the pharynx. The osteophyte from C3 to C4 was resected to make a smooth surface, and bleeding from the bone was stopped with bone wax (Figure 5). Anterior cervical discectomy and fusion using a stand-alone anchored spacer (Stryker® 7 mm, 4°, fixed with 12 mm screw) with local bone grafting was performed at C3/C4. The operation time was 114 minutes, and the estimated blood loss was 10 g.

Dysphagia and dysphonia were improved by day 3 after surgery, although an x-ray showed massive swelling in the retropharyngeal space (Figure 6). The patient's myelopathy, such as clumsy hand movements and walking difficulty, was also improved. The subjective symptoms of dysphagia and dysphonia were improved by day 20 after surgery, and food intake was started with a dysphagia diet after confirming that deglutition



Figure 2. Computed tomography scan showed anterior cervical osteophytes at the C3/C4 level.



Figures 3. (A, B) Magnetic resonance imaging showed slight compression of the cervical cord at the C3/C4 level by OPLL. OPLL, ossification of the posterior longitudinal ligament.

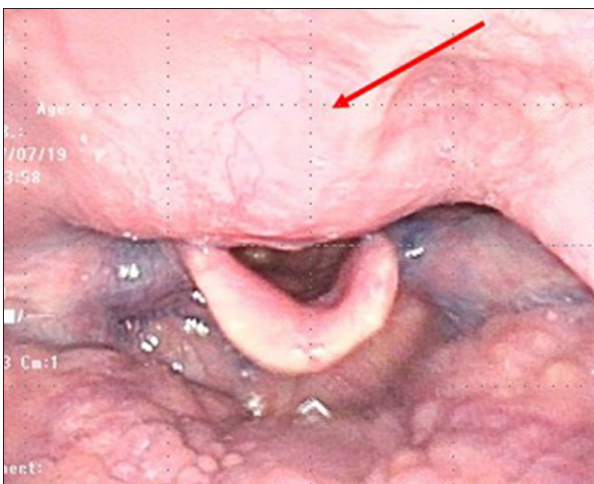


Figure 4. Protruding submucosal mass in posterior pharyngeal wall and vocal cord dysfunction is revealed by fiberoptic examination.

(swallowing) was not a problem through video-fluoroscopic examination, although x-ray and a fiberoptic examination still showed swelling in the retropharyngeal space (Figure 7A, 7B).

Discussion

This case report describes a patient who presented varying symptoms with DISH. DISH tends to occur more often in elderly men with metabolic syndrome, diabetes mellitus, and obesity. Growth hormone and insulin-like growth factor are said to promote bone growth [1].

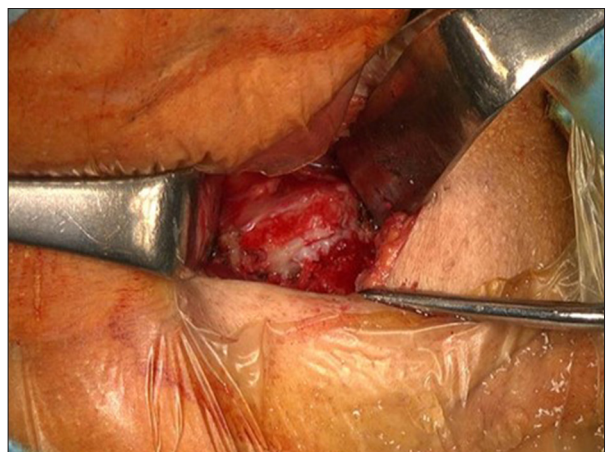


Figure 5. Osteophytes at C3 to C4 was resected to make smooth surface.

DISH is distinguished from ankylosing spondylosis and other degenerative disease. DISH is characterized by 3 criteria identified by Resnick [2]: 1) calcification and ossification along the anterolateral paravertebral ligaments at least 4 vertebral bodies; 2) relative preservation of the intervertebral disc height in contiguous body; and 3) absence of apophyseal ankylosis or erosion of sacroiliac fusion.

The thoracic spine is most frequently affected, causing back pain and stiffness. The symptoms of DISH depend on its localization. In the cervical spine, the formation of a large osteophyte can result in dysphagia due to esophageal compression.



Figure 6. X-ray showed massive swelling in the retropharyngeal space.

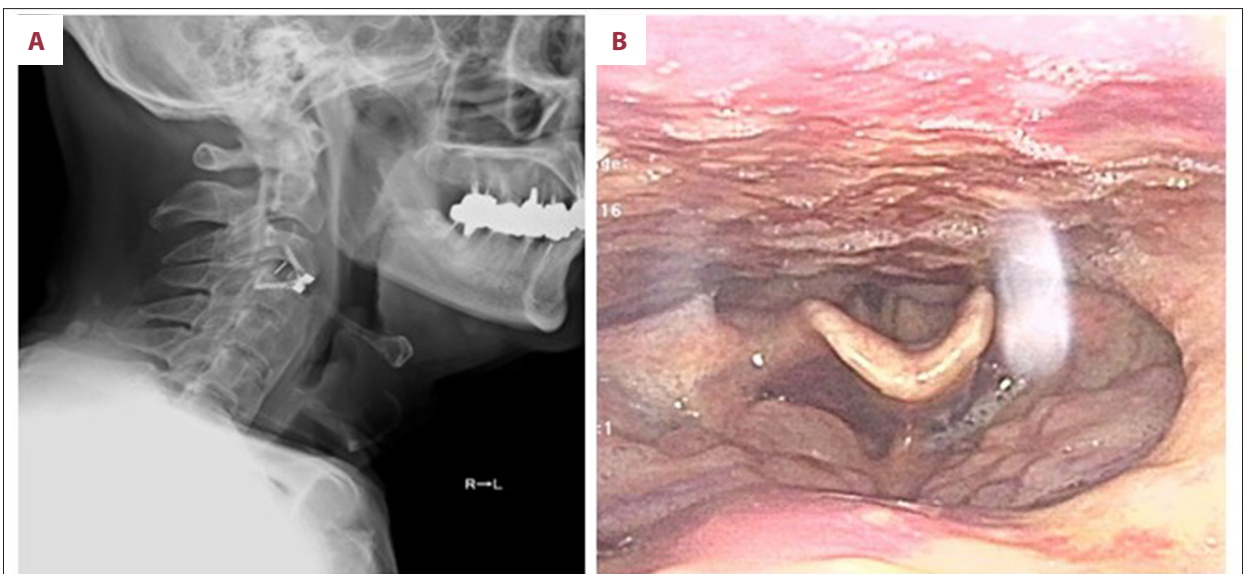
Exuberant osteophyte formation of the anterior longitudinal ligament sometimes induces dysphagia in DISH patients. The mechanisms underlying dysphagia in DISH, as suggested in the literature, includes restriction of epiglottis mobility, incomplete glottal closure, and restriction of the movement of larynx [3]. Patients rarely have dysphonia [4]. Dysphonia may be attributed not only to the mechanical obstruction of the larynx but also to a reduction in glottal mobility by retro-cricoid inflammation. Direct compression of the osteophyte can cause recurrent laryngeal nerve paralysis [5]. There are a few reports that have suggested cases of bilateral vocal cord paralysis due to compression of a retro-cricoid lesion associated

with exuberant osteophyte formation [6,7]. This mechanism can also cause dyspnea and sometimes needs immediate surgical interventions for emergency airway opening, but increased formation of osteophytes can sometimes interfere with tracheal intubation.

Our patient presented with progressive dysphonia, dysphagia, and myelopathy of over 2 months history. CT showed a huge osteophyte compressing the pharynx and the OPLL compressing the cervical cord at the C3/C4 level. A fiberscope examination prior to the operation showed severe oropharyngeal dysphagia and vocal cord dysfunction. Surgical treatment is considered a reasonable treatment option [8]. After the removal of the osteophyte and anterior decompression and fusion, the patient's dysphonia and dysphagia gradually improved. Myelopathy also improved by stabilizing the OPLL. However, despite symptom resolution, the swelling in the retropharyngeal space persisted for about a month, which was a relatively long time in this case. This may have been due to pre-existing inflammation induced by the protruding osteophyte. Some cases have shown that despite a successful surgery, the lack of swallow coordination may continue resulting in a patient unable to eat on their own any longer. Patients should be followed-up for several years in terms of symptom recurrence as the long-term outcomes of post-operative recurrence remains unclear [9].

Conclusions

We must remember DISH may induce varying symptoms: dysphagia, gait disturbance, sleep apnea, and so on. However, dysphonia due to mechanical obstruction, such as was found



Figures 7. (A, B) Swelling in the retropharyngeal space was remaining.

in our case, is rare. Radiological examinations, such as x-ray and CT-scan, are a simple and easy way to investigate the situation. However, when we come across these patients, cooperation with otolaryngologists and dieticians with various expertise is also important before a patient's symptoms worsen.

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Conflict of interest

None.