

## Case Report



# Swallowing Difficulty in Diffuse Idiopathic Skeletal Hyperostosis with Metabolic Syndrome

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### Conflict of Interest

The authors have no financial conflicts of interest.

## ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) is a disease of unknown etiology developing following ossification of the antero-lateral ligaments of the spine. Mostly, prevailing elderly adult males, it is an uncommon cause of dysphagia and dysphonia. We report three cases of DISH with metabolic syndrome. They were complained of neck movement restriction and dysphagia. At first, They all visited ear, nose, and throat outpatient department. The initial impression was gastroesophageal reflux, and an endoscopy excluded esophageal lesion. Cervical spine radiologic imaging revealed ossification of the cervical anterior longitudinal ligament with large, conspicuous osteophytes from cervical spine lesion, producing compression of pharyngoesophagus and upper airway; these images corresponded to DISH. Cervical osteophyte surgical removal resulted in a complete alleviation from dysphagia for the patient. DISH should be considered in the differential diagnosis of dysphagia.

**Keywords:** Diffuse idiopathic skeletal hyperostosis; Deglutition disorders; Spine; Osteophyte

## INTRODUCTION

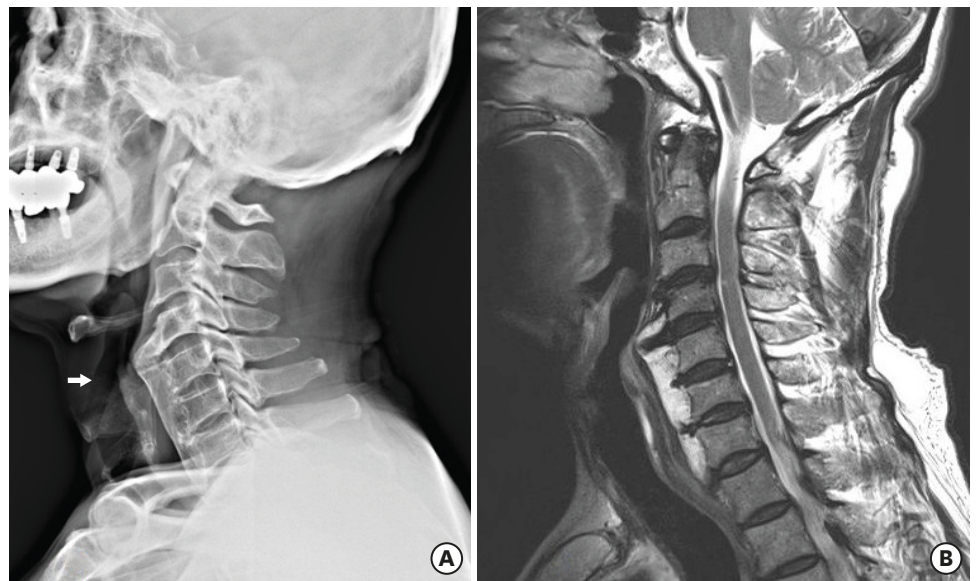
Anterior cervical osteophytes take place in 20–30% of the elderly population and generally appear asymptomatic.<sup>9)</sup> Dysphagia may occur in about 17% to 28% of patients with cervical ossification.<sup>13)</sup> Dysphagia in cases of cervical hyperostosis may be associated with direct impingement, fibrotic adhesions, or epiglottic mobility disorder.<sup>6,17)</sup> Oropharyngeal dysphagia is the most perpetual symptom in patients with diffuse idiopathic skeletal hyperostosis (DISH) localized to the cervical spine.<sup>15)</sup>

## CASE REPORT

### Case 1

A 65-year-old male went to the ear, nose, and throat outpatient department with the disturbance of dysphagia to solids for 5 months. Dysphagia was proceeded and he localized the blockage to neck. He had a 10 kg weight loss for 6 months and was only able to eat soft diet, but not liquid due to aspiration. He had no experience of odynophagia, heartburn

and regurgitation of foods. He had hypertension, hypertriglyceridemia, and low high density lipoprotein cholesterol levels, accordingly it met the diagnostic criteria of metabolic syndrome. On physical examination, oral and oropharyngeal check-up was trivial. Indirect laryngoscopy and fibreoptic examination of laryngopharynx didn't indicate any pathology. Cranial nerve examination was normal. The patient had rather limited neck movements and neck pain. A video fluoroscopic swallowing test was done and revealed blockage to the passage of dye opposite to junction of fourth and fifth cervical vertebra (**FIGURE 4A**). We also identified bony bridging anterior to body of fourth, fifth and sixth, seventh cervical vertebra (**FIGURES 1 & 2**). We diagnosed to have DISH as a grounds for dysphagia and did operation. An anterior neck approach was used. Under general anesthesia, the patient was placed supine with sandbag under the shoulders. A horizontal neck incision was made starting in the midline at the level of the thyroid cartilage and extended laterally up to sternocleidomastoid



**FIGURE 1.** Lateral cervical spinal X-ray (A) and a T2-sagittal cervical magnetic resonance imaging (B) revealed giant cervical osteophytes at the ventral portion of C4–C5, preserving the disc space, anteriorly displacing laryngeal air shadow (arrow) and contiguous bulky ossification of anterior longitudinal ligament of anterior cervical vertebral bodies.

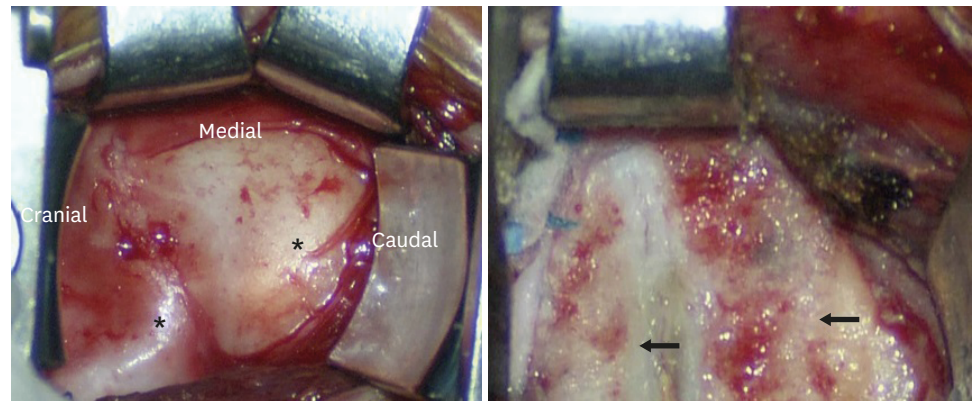


**FIGURE 2.** Pre-operative cervical spine computed tomography shows anterior osteophyte (asterisk) with esophageal indentation external esophageal compression by the large osteophytes (arrow).

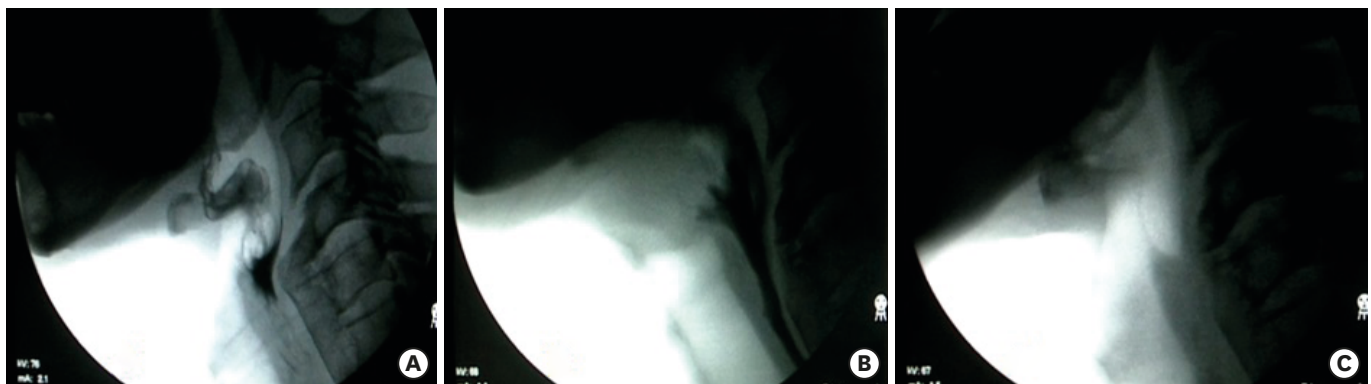
muscle on the right side. Platysmal muscle was dissected. Dissection was carried deeper and medial to sternocleidomastoid muscle and lateral to strap muscles. Internal jugular vein and carotid sheath were identified and retracted laterally. By blunt dissection medial to carotid sheath, the esophagus was identified and retracted anteriorly. The hardening prevertebral fascia was identified and the longus colli muscle was separated for placing retractor. Calcified anterior longitudinal ligament overlying C4–C5 vertebrae was seen and were removed with the rongeurs and drilling of high speed drill (**FIGURE 3**). A surgical placed over the raw bone to prevent damage to the esophagus. The draining tube was inserted in operative site. The patient was tolerable in postoperative state (**FIGURE 5**). There was mild aspiration after surgery, but no lesions such as pneumonia, hoarseness and esophageal related problem were observed. There was no cervical spine instability. The patient was followed up video fluoroscopic swallowing test at 3 weeks after surgery and reported remarkable reduction in dysphagia (**FIGURE 4B & C**). He was followed up monthly for the next 6 months and was completely alleviated of dysphagia. He gained 5 kg body weight, too.

### Case 2

A 64-year-old male was complained with swallowing difficulty and hoarseness a 3 months ago. He was able to eat the soft diet. He had complication of paraparesis (3/5), hypthesia below T4 due to spinal cord injury, ossification of the posterior longitudinal ligament, 10 years ago. At the time, neuroplasty on C5, C6, C7 was done. After operation, he was



**FIGURE 3.** Operative finding showing before (asterisks) and after (arrows) removal of osteophyte.



**FIGURE 4.** Preoperative esophagogram shows obstruction to the dye passage opposite C4 and DISH bridging C4–C6 vertebrae. (A) Narrow upper esophageal passage, dye retention in pyriformis sinus on swallowing test before surgery, postoperative esophagogram (B) shows widening upper esophageal passage after operation, (C) no food dye retention on swallowing test after operation.



**FIGURE 5.** Postoperative lateral cervical spinal X-ray shows blunted cervical osteophyte (arrow).

undergoing rehabilitation. But he was stayed in bed ridden state and was overweight. He visited another hospital for checking up. The cervical spine X-ray (**FIGURE 6**) was revealed anterolateral paravertebral ossification at C2–C3, C4–C7. For operation, he visited our clinic.

The cervical spine computed tomography (CT), video esophagogram (**FIGURE 7**) was evaluated. On the blood test, he had hypercholesterolemia and diabetes mellitus which he didn't know. Under general anesthesia, the patient was positioned in supine. The head was slightly turned to left side. And then the transverse skin incision was made on right side anterior neck along skin crease from midline to the lateral border of the sternocleidomastoid muscle at the level of the thyroid notch. The platysma muscle was also incised transversely. Dissection between the internal carotid sheath and midline visceral structure along the medial border at sternocleidomastoid muscle was performed. The C4, C5, C6, C7 vertebral body was exposed and the level was confirmed with C-arm. After retraction of paravertebral



**FIGURE 6.** Preoperative lateral cervical spinal X-ray shows blunted cervical osteophyte.





**FIGURE 7.** Preoperative cervical spinal computed tomography sagittal view revealed giant osteophyte along the anterior cervical vertebral body from C2–C7. Preoperative esophagogram shows obstruction of dyes at the C4 level. From the C4 level, prominent diffuse idiopathic skeletal hyperostosis was located.

structure with cloward retractor, prevertebral ossification was removed with drill and punch forceps. There was no associated morbidity and complication.

Time right after surgery, the dysphagia was dramatically improved. Before the surgery, he could not eat the solid diet. He managed to eat the rice porridge. After the surgery, he could eat the solid diet and the hoarseness was improved, too. He denied any symptoms suggestive of cervical radiculopathy or myelopathy. There was no active lung lesion.

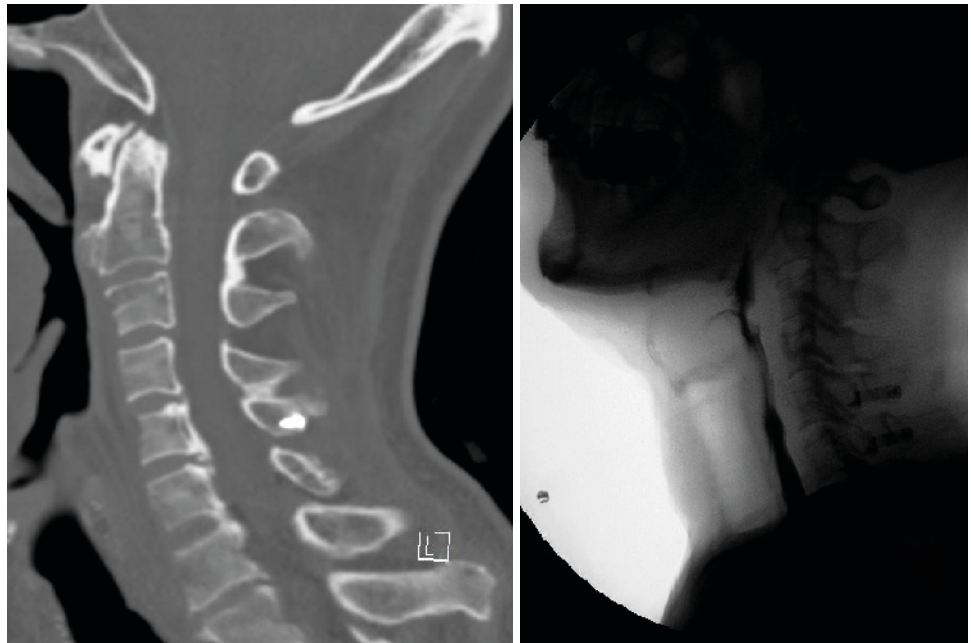
For the evaluation of spine lesion and swallowing, The cervical CT and video esophagogram (**FIGURE 8**) was checked.

We found the improvement of swallowing and interval between the esophagus and cervical spine.

### Case 3

A 82-year-old male had a complicated medical history which included hypertension, adult onset diabetes mellitus. He was not well compliant of hospital and economically vulnerable class. At first, he came to the hospital because of swallowing difficulty, anorexia, and weight loss. He lost 8 kg for 2 weeks, could not eat general diet.

On blood tests done by internal medicine, HbA1c was 10.7. Barium swallow showed posterior esophageal compression in cervical tract of esophagus. On the esophageal endoscopy, There was a esophageal stricture. So, esophageal balloon dilatation was performed about 8 mm to 10 mm. For the conservative treatment, he was prescribed diabetes medicine, proton pump inhibitor and nonsteroidal anti-inflammatory drugs (NSAID). After 2 days, he drank sips of water but was still not swallowed. For the cervical spine evaluation. He was transferred to our department.



**FIGURE 8.** Postoperative computed tomography and esophagogram. All anterior cervical osteophytes were removed. Esophagogram shows improvement of esophageal pathway as compared with pre-operation.

The cervical spine CT was evaluated. A large osteophyte were grown in front of the C3, C4, C5, compressing the esophagus. So we decided to remove it surgically. Under general anesthesia, the total cervical osteophytes removal was done in C3, C4, C5 through anterior cervical approach.

Before the surgery, his weight was -1.5 standard deviation of the standard weight.

After BST control, the weight was -1.3 standard deviation. His anorexia, swallowing difficulty was consisted. After the surgery, his diet continued to increase and dysphagia, anorexia was resolved. Postoperative 6 months later, his weight improved to a normal range.

## DISCUSSION

Dysphagia can be divided into oropharyngeal and esophageal dysphagia. Esophageal dysphagia can be owing to motility disorders, inadequate oropharyngeal bolus transport, inability to pressurize the pharynx, inability to elevate the larynx, discoordination of pharyngeal contraction and cricopharyngeal relaxation; and decreased implementation of the pharyngoesophageal segment secondary to muscle pathology.

Pharyngoesophageal swallowing disorders are generally due to acquired disease involving the central and peripheral nervous system. This includes cerebrovascular accidents, brain stem tumors, poliomyelitis, multiple sclerosis, Parkinson disease, pseudobulbar palsy, peripheral neuropathy, and operative damage to the cranial nerves involved in swallowing. Muscular diseases such as radiation-induced myopathy, dermatomyositis, myotonic dystrophy, and myasthenia gravis are less common causes. Rarely, extrinsic compression by thyromegaly, cervical lymphadenopathy, or hyperostosis of the cervical spine can cause

pharyngoesophageal dysphagia.<sup>1)</sup> DISH is an idiopathic rheumatologic abnormality in which confluent ossification occurs along throughout the body, but most significantly the anterior longitudinal ligament of the spine. DISH is a degenerative musculoskeletal disease that is common in elderly adults, especially 6th to 7th decades in men, often in association with insulin resistance conditions like diabetes and metabolic syndrome, leading in turn to an increased cardiovascular risk.<sup>5,11)</sup> The exact cause is unknown. Dysphagia due to osteophytic lesion was first verified in 1905 by Zahn,<sup>19)</sup> and the first surgical elimination of the osteophyte was accomplished in 1926 by Mosher.<sup>14)</sup>

The dysphagia is usually marked, present for solid foods, improved by flexion of the neck, and worsened by extension of the neck. Combined symptoms may include a foreign body sensation, odynophagia, salivary stasis, dysphonia, dyspnea, and aspiration.<sup>10)</sup> There are some potential mechanism that DISH causes dysphagia: 1) large osteophytes may lead to dysphagia by direct mechanical obstruction; 2) smaller osteophytes may impinge at sites of associated immobility of the esophagus since anatomically the esophagus is anchored at the level of the cricoid cartilage and the diaphragm; 3) inflammatory response of soft tissue nearby the esophagus; 4) esophageal and pharyngeal narrowing due to sore muscle spasm<sup>12)</sup>; and 5) recurrent nerve palsies triggered by the hyperostosis.<sup>3)</sup> The most common level of involvement related to dysphagia is C5–C6 followed by C4–C5, C2–C3 being the least common level affected.<sup>7)</sup>

Plain radiographs are usually adequate to make the diagnosis, but a CT scan may show the extent of the hyperostosis and its location relative to the esophagus. For the swallowing disorder evaluation, Video fluoroscopy (video esophagogram) is considered the gold standard.<sup>16)</sup>

The severity of dysphagia may need surgical treatment.<sup>2,4)</sup> Therapy for DISH is chiefly conservative, nonsteroidal anti-inflammatory drugs and muscle relaxants are used. However in refractory cases there are two surgical approaches to remove osteophytes, by lateral cervical or peroraltranspharyngeal ways.<sup>18)</sup> The management of dysphagia due to Diffuse Idiopathic Skeletal Hyperostoses, in mild cases, it can be managed with NSAIDs and bisphosphonates, but in severe cases required surgery. Anterior neck approach had a good outcome.

Stabilization of the spine is not recommended. Large osteophytes also present a risk of esophageal injury during the operative exposure. The esophagus may be difficult to mobilize and in some degree adherent to other cervical fascia due to local inflammatory reaction. Esophageal and recurrent laryngeal nerve injury are the complications of the surgery. Re-ossification with new osteophyte formation may be on rare occasion and repeat surgery may be indicated.<sup>8)</sup> Recurrence of dysphagia after operation was minimal and requires revision surgery.

The prevalence of DISH as a cause of dysphagia is low.

DISH may be caused by aging like other degenerative lesions, but the associated cause is different from other common benign spinal diseases caused by repetitive mechanical stress, microtrauma.

DISH may be caused by uncontrolled metabolic syndrome (ex. hypertension, diabetes mellitus, hyperlipidemia). Coincidentally, all three of our cases were patients with medically underlying disease and those who complained of swallowing disorders. This could suggest that DISH, which has no clear etiology, may be associated with underlying medical disease.

## CONCLUSION

DISH is a rheumatologic abnormal condition with exuberant ossification. Dysphagia is perceived as a common aspecific clinical finding of painful or difficult swallowing. Thus, this diagnosis should be doubted in specific clinical settings, like as old males with metabolic disorders and with nonetheless unaccounted for dysphagia. Besides, surgical removal of only large osteophytes combined with NSAID therapy may relieve symptoms.

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