

# Double Cervical Bird Beak Sign Resulting in Dysphagia

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## ABSTRACT

**Objective:** The authors report a rare case of ossification of the anterior longitudinal ligament in the cervical spine underlying dysphagia. **Case Report:** We report the case of a 50-year-old male presenting with difficulty swallowing and choking. CT of the cervical spine demonstrated anterior longitudinal ligament hypertrophy with osteophytes worst at C1-C2 and C6-C7, without spinal cord compression. A videofluoroscopic swallow study revealed reduced epiglottic inversion and hyolaryngeal elevation resulting in incomplete clearance from the pharynx, as well as compression of the esophagus. Subsequently, the patient underwent osteophylectomy. There were no intraoperative or postoperative complications. Two weeks after the surgery, videofluoroscopic swallow demonstrated improved function and reduced compression. Four weeks postoperatively, the patient completely regained swallowing function without pain.

**Keywords:** Dysphagia; Anterior Longitudinal Ligament; Cervical Spine; Diffuse Idiopathic Skeletal Hyperostosis; Ossification; Calcification

## 1. Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a degenerative disease characterized by excessive ligamentous calcification and ossification, most commonly affecting spinal and extraspinal locations [1,2]. Also named ankylosing hyperostosis and Forestier's disease, it has specific radiological criteria for diagnosis and should not be confused with ankylosing spondylosis or cervical degenerative disease. The diagnostic criteria established by Resnick are 1) flowing calcification and ossification along anterolateral aspect of at least two contiguous vertebral bodies, 2) relative preservation of intervertebral disc height and no disc generation in affected areas, and 3) absence of apophyseal joint ankylosis and sacroiliac joint fusion [3].

DISH commonly occurs in the elderly population and twice as frequently in men [3]. Though radiological imaging can detect distinct paravertebral masses anterior to the vertebrae, ordinary physical examination cannot [2]. In the cervical spine, excessive calcification and ossification of the anterior longitudinal ligament can result in dysphagia, and less commonly, dyspnea and dysphonia, because of the compression of anterior structures [4].

The authors present a rare case of ossification of the anterior longitudinal ligament in the cervical spine underlying dysphagia and treated with osteophylectomy.

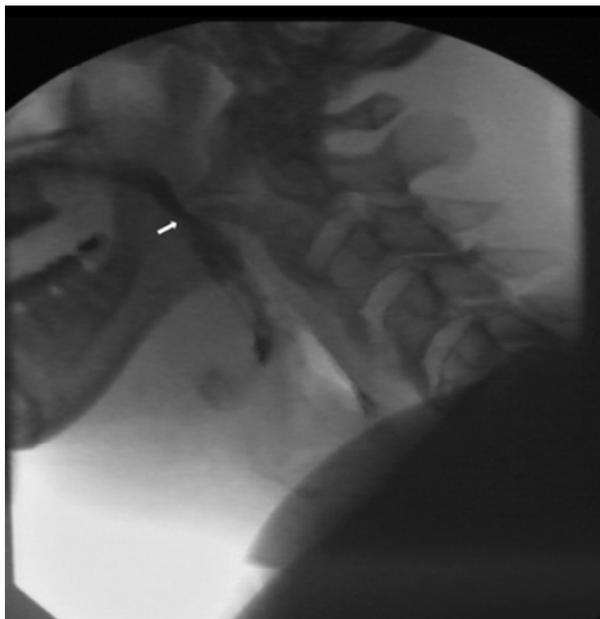
## 2. Case Report

A 50-year-old man presented to the emergency department with severe neck pain and difficulty swallowing; specifically, his neck pain radiated down the bilateral upper extremities in a C3-C4 distribution, and difficulty swallowing was associated with pain and choking. Approximately three months earlier, this patient was admitted to the emergency department after a fall, at which time he complained of neck pain with radiation into the left shoulder without any numbness or tingling, but was discharged for the non-acute nature of the findings.

Two weeks after his most recent emergency department visit, the patient came for additional imaging and follow-up, and reported worsening neck pain, significantly increased difficulty swallowing, and increased choking. He denied having arm pain, weakness, or numbness, and his examination was unremarkable. CT of the cervical spine demonstrated anterior longitudinal ligament hypertrophy with osteophytes worst at C1-C2 and C6-C7, without spinal cord compression (**Figure 1**). A videofluoroscopic swallow study revealed reduced epiglottic inversion and hyolaryngeal elevation resulting in incomplete clearance from the pharynx, as well as compression of the esophagus (**Figure 2**). He underwent a C2-C7 anterior osteophylectomy. Though it was difficult to reach up to C1 and remove the entire osteophyte,



**Figure 1. Preoperative CT scan demonstrates anterior longitudinal ligament hypertrophy with largest osteophytes at C1-C2 and C6-C7, without spinal cord compression.**



**Figure 2. Preoperative videofluoroscopic swallow study demonstrates reduced epiglottic inversion and hyolaryngeal elevation resulting in incomplete clearance from the pharynx, as well as compression of the esophagus.**

it was drilled down sufficiently. The patient tolerated the procedure well. There were no intraoperative or signifi-

cant postoperative complications.

Post-op CT revealed significantly decreased osteophytes (**Figure 3**). Two days after surgery, the patient had persistent difficulty and pain swallowing resulting from soft tissue edema. A videofluoroscopic swallow study demonstrated incomplete pharyngeal clearance of large solid boluses. He was recommended a pureed diet with thin liquids, crushed medications in applesauce, and a follow-up videofluoroscopic swallow study. The patient was neurologically and physically stable, and discharged two days post-op.

Two weeks post-op, videofluoroscopic swallow study revealed improved epiglottic inversion and hyolaryngeal elevation, as well as reduced esophageal compression (**Figure 4**). Four weeks after surgery, the patient completely regained functional swallowing with no pain.

### 3. Discussion

DISH, a condition characterized by calcification and ossification in the axial and peripheral enthesial locations, was first described by Forestier *et al.* in 1950 [1,4,5]. Despite its longstanding documentation in the medical community, its pathology remains unknown. However, DISH has been correlated with diabetes, obesity, hypercholesterolemia, and gout [1,6].

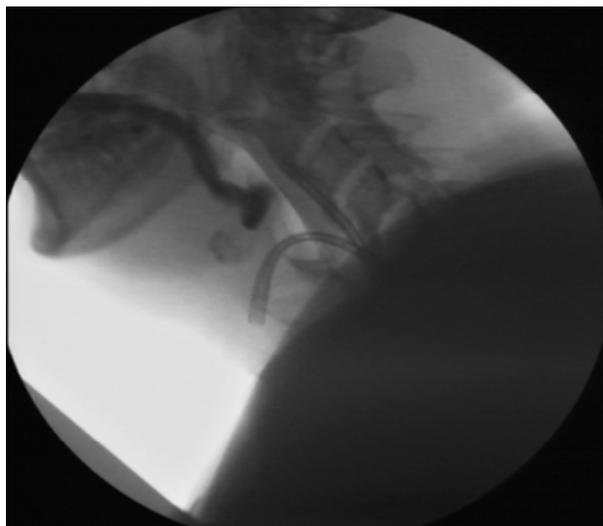
DISH patients are typically asymptomatic, though upper gastrointestinal, respiratory, neurological, and spinal instability complications have been documented [3]. The thoracic spine is the most commonly affected region in DISH patients, and this region generally presents with back pain and stiffness. Of DISH patients, 76% of patients have cervical involvement [7]. Further, dysphagia resulting from DISH-related mechanical compression in the cervical spine occurs at rate of 28% [8].

DISH is easily diagnosed using X-Ray, though CT may be useful in showing the size and shape of osteophytes with respect to relevant structures such as the esophagus [1]. Further, barium swallow videofluoroscopy is useful in confirming esophageal compression and obstruction. For those with dysphagia, endoscopy is an effective tool to rule out other potential causes such as tumors, motility disorders, esophageal strictures, esophagitis, esophageal webs, or candidiasis, though it risks perforation [9]. Other dysphagia-inducing conditions that should be considered include achalasia, motor neuron disease, Parkinson's disease, pharyngeal pouch, and stroke [2].

The management of dysphagia associated with DISH is broad; conservative treatment includes modification of diet, non-steroidal inflammatory medications, corticosteroids, and myorelaxants, while aggressive treatment involves osteophylectomy [1]. When conservative therapies fail and the patient experiences increased complica-



**Figure 3.** Postoperative CT scan demonstrates significantly reduced osteophytes.



**Figure 4.** Postoperative videofluoroscopic swallow study demonstrates improved epiglottic inversion and hyolaryngeal elevation, as well as reduced esophageal compression.

tions, surgery can prove to be an effective solution [10, 11]. Fortunately, our patient recovered within a few weeks and completely regained swallowing function; however, it is possible for patients to exhibit uncoordi-

nated swallowing and a high rate of aspiration even one month postoperatively [2].

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