

Diffuse Idiopathic Skeletal Hyperostosis Causes Acute Airway Obstruction Following Upper Respiratory Tract Infection: A Case Report

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Diffuse idiopathic skeletal hyperostosis (DISH) is a progressive, systematic, non-inflammatory disease of unknown origin that causes ossification of ligaments and tendons attached to bone. It is most commonly observed in the spine, particularly at the lower thoracic and cervical levels¹⁾. While DISH is usually asymptomatic, it may occasionally cause compression myelopathy, dysphagia, and dyspnea. To our knowledge, however, there are only a few previous reports of DISH-associated acute airway obstruction requiring surgical treatment²⁻⁴⁾. Here we describe an extremely unusual case of DISH wherein the patient developed acute airway obstruction after upper respiratory tract infection.

A 65-year-old man who was unable to breathe was admitted to our hospital emergency department after complaining of cold-like symptoms such as fever and cough along with dyspnea for several days. His past medical history included posterior cervical laminectomy and laminoplasty performed for compression myelopathy following ossification of the posterior longitudinal ligament (OPLL), with a prolonged history of mild dysphagia. Computed tomography (CT) scanning revealed spinal ankylosis due to either DISH or OPLL. Significant anterior protrusion of the ossified anterior longitudinal ligament at the C4-5 level was also observed (Fig. 1A). The surrounding soft tissues at the level of the hyoid bone were markedly swollen, and the airway was obstructed (Fig. 1B, C). Laryngofiberscopy performed by an otolaryngologist showed a normal epiglottis and surrounding mucosal surface, with the posterior pharyngeal wall largely protruding. There was no vocal cord paralysis in the glottis. Emergency orotracheal intubation was performed to secure

the airway. We concluded that the airway obstruction was triggered by soft-tissue swelling following an upper respiratory tract infection in the presence of the huge osteophytes. The next day, the patient underwent anterior drilling of the extensive osteophytes on the anterior longitudinal ligament (Fig. 2). The patient was extubated the day after the surgery, and bronchial fiberscopy was performed to confirm that the airway was not obstructed. Despite presenting with transient dysphagia, the patient was discharged without symptoms 21 days after admission.

Although it varies among ethnicities, the prevalence of DISH increases with age. DISH is common in men and has been suggested to be associated with obesity and abnormal glucose metabolism. Even then, no clear cause has been determined. It is commonly known that ossification of the anterior ligament in patients with DISH causes dysphagia and dyspnea⁵⁾, and that these symptoms progress slowly in most cases. However, the development of acute airway obstruction is extremely rare. According to Rojas et al⁶⁾, the upper limits of the normal prevertebral soft-tissue thickness measured using multidetector CT in the neutral position in adults is 8.5, 6, and 7 mm at C3, C4, and C5, respectively. In our case, the cause of the huge osteophytes was probably the mechanical stress concentrated at C4/5, the border of the ankylosed spine. Moreover, the soft-tissue edema following the upper respiratory tract inflammation triggered airway obstruction at that level. The indications for surgical intervention for this pathology have not been clearly defined. Matan et al reported that airway obstruction resulting from DISH can be treated by surgical excision of osteophytes, according

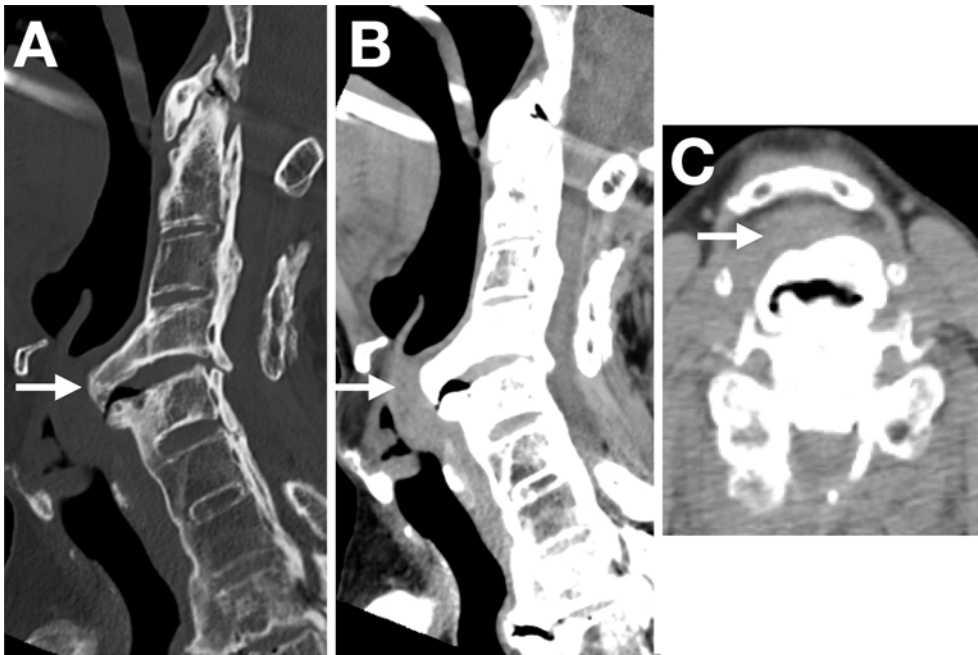


Figure 1. (A), (B) Sagittal computed tomography (CT) scan of the neck shows an ankylosed cervical spine secondary to ossification of the anterior and posterior longitudinal ligaments. Laminectomy at the C2, C5, and C6 levels and laminoplasty at the C3 and C4 levels had been performed in the past. (C) Axial CT scan of the neck at the C4/5 level shows airway obstruction due to extensive anterior hyperostosis and thickening of the surrounding soft tissues (white arrow).

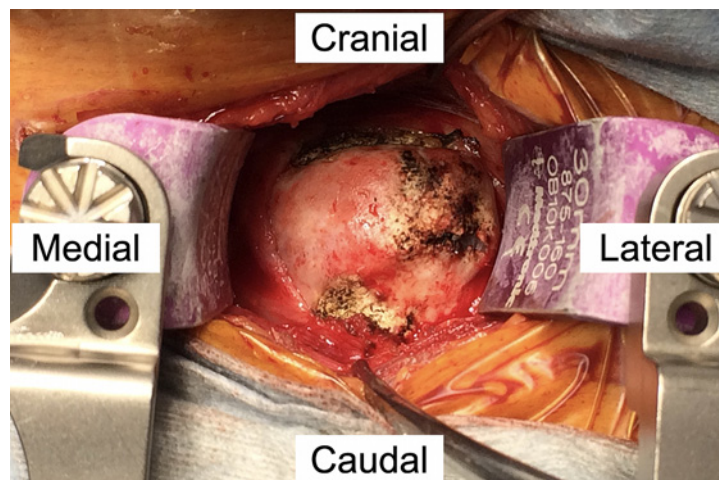


Figure 2. Intraoperative photograph. An extensive osteophytic mass involving the anterior longitudinal ligament has been easily exposed, with no peripheral adhesions.

to the same principles used for the treatment of dysphagia³). We planned immediate removal of the ossified portion of the anterior longitudinal ligament without cervical fusion because of the emergency situation. This treatment successfully relieved the severe respiratory distress in the short term. However, mechanical stress remained at the site, which could lead to re-enlargement of the osteophytes. In fact, lateral radiography of the cervical spine demonstrated slight enlargement of the osteophytes 3 years later (Fig. 3),

and we continued to monitor the patient carefully.

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

Ethical Approval: unnecessary

Informed Consent: Informed consent was obtained from all participants in this study.

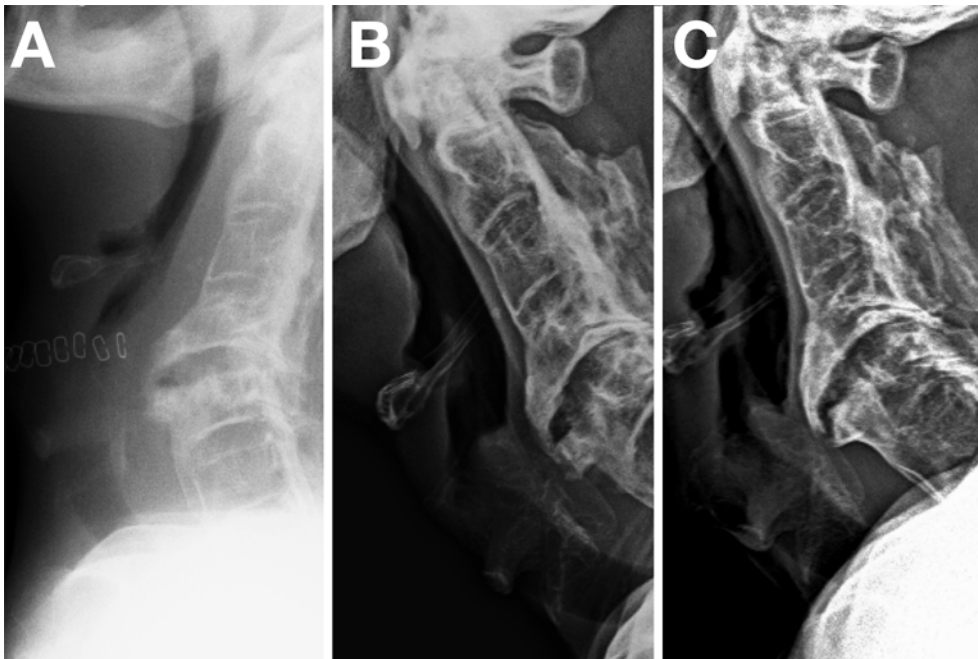


Figure 3. Lateral radiographs of the cervical spine. (A) Five days after surgery, the soft-tissue swelling has reduced and no airway narrowing can be recognized. (B) Six months after surgery. (C) 3 years after surgery, the osteophytes have slightly enlarged.

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