

# DYSPHAGIA AND LUNG ASPIRATION SECONDARY TO ANTERIOR CERVICAL OSTEOPHYTES: A CASE REPORT AND REVIEW OF THE LITERATURE

Wilfredo E. De Jesus-Monge, MD; Elsie I. Cruz-Cuevas, MD

**Introduction:** Two of the most common causes of anterior cervical bony outgrowths are diffuse idiopathic skeletal hyperostosis (DISH) and ankylosing spondylitis (AS). These osteophytes have been associated with serious complications. The objective of this case report is to highlight how commonly occurring anterior cervical osteophytes may become an uncommon cause for life-threatening dysphagia and potential lung aspiration in elderly patients.

**Case Report:** An 80-year-old man came to the hospital with dyspnea and cough productive of sputum. These symptoms were associated with progressive dysphagia for the previous 10 years, which was evaluated with a barium esophagogram that was discontinued due to aspiration of barium. The patient was placed on mechanical ventilation. Neck computed tomography showed anterior cervical osteophytes displacing the upper airway and compressing the esophagus and calcification of the posterior longitudinal ligament with a "bamboo" appearance. The patient underwent surgical removal of the osteophytes.

**Discussion:** The patient had onset of symptoms at >50 years of age, dysphagia, osteophytes in the clavicle and shoulders, anterior cervical osteophytes, calcified posterior longitudinal ligament, and spinal stenosis, all of which are associated with DISH. AS is associated with the patient's history of stooped posture, anterior displacement of the head while walking, and bamboo spine. The final diagnosis, either DISH or AS, causing these life-threatening anterior cervical osteophytes is undetermined because of the inability to evaluate for the respective diagnostic criteria. However, these osseous pathologies must be considered as causes of life-threatening dysphagia and aspiration in an elderly person. (*Ethn Dis.* 2008;18[Suppl 2]:S2-137-S2-140)

**Key Words:** Osteophyte, Diffuse Idiopathic Skeletal Hyperostosis, DISH, Ankylosing Spondylitis, AS

---

From the University of Puerto Rico, Department of Medicine (WEDM, EICC), and Master of Science in Clinical Research Program (WEDM), San Juan, Puerto Rico.

Address correspondence and reprint requests to: Wilfredo E. De Jesus-Monge, MD; Physician; University of Puerto Rico School of Medicine; 1693 Cuernavaca St; Venus Gardens; San Juan, Puerto Rico 00926-4649; 787-367-6983; 787-755-6983 (fax); wilmedresearch@yahoo.com

## INTRODUCTION

Dysphagia may have oropharyngeal or esophageal causes. Anatomic causes of dysphagia are tumors, abscesses, and cervical bony outgrowths (osteophytes). The differential diagnosis of spinal bony outgrowths is extensive, but the most common etiologies are diffuse idiopathic skeletal hyperostosis (DISH) and ankylosing spondylitis (AS). Cervical bony outgrowths are common, and most are asymptomatic. However, dysphagia secondary to compression of the esophagus by these osteophytes is unusual. The objective of this case report is to highlight how commonly occurring anterior cervical osteophytes may become an uncommon cause for life-threatening dysphagia and potential lung aspiration in elderly patients. The discussion will include diagnosis and therapy of this interesting pathology.

## CASE REPORT

An 80-year-old man with diabetes mellitus, peripheral vascular disease, arterial hypertension, congestive heart failure, and cardiac pacemaker arrived at the hospital because of incoherent speech, shortness of breath, and cough productive of whitish sputum since one day before arrival. These symptoms were associated with progressive dysphagia to solid foods for the previous 10 years but worsened in the previous two months. He developed labored breathing and cough productive of whitish sputum after ingesting a meal. He had a history of stooped posture and anterior displacement of the head while walking. He had no fever, weight change, chest pain, loss of consciousness, seizure, abdominal pain, or change in bowel movement. He was previously

evaluated for dysphagia as an outpatient by laryngoscopy and barium esophagogram, which was discontinued due to aspiration of contrast. The patient was placed on mechanical ventilation upon arrival at the hospital because of altered mental status and respiratory distress.

Physical examination showed fever, adequate blood pressure, and tachycardia. Although critically ill, he was alert and cooperative. He had heart normal sinus rhythm and rate and a grade I/VI systolic ejection murmur. His lungs were clear to auscultation bilaterally.

Laboratory tests showed a white cell count of  $19.3 \times 10^9$  per liter ( $.89 \times 10^9$  neutrophils per liter and  $.03 \times 10^9$  lymphocytes per liter). A chest radiograph showed retained barium in the lungs with atelectatic/pneumonic changes and osteophytes in the distal right clavicle (Figure 1). A head computed tomography (CT) showed no trauma, mass, or cerebrovascular accident. A neck CT showed anterior cervical osteophytes causing displacement of the upper airway and compression of the esophagus (Figure 2), calcification of the posterior longitudinal ligament with "bamboo" appearance, and stenosis of the spinal cord at C6-C7 secondary to a bulging osteophyte.

The patient was admitted and started on antibiotic therapy for pneumonia. Leukocytosis was decreasing. Cervical orthopedics service was consulted and surgical correction of cervical osteophyte was recommended. After so many years of swallowing complaints, the progressive esophageal compression induced by the anterior cervical osteophytes lead to the man's catastrophic clinical presentation to the hospital. The patient underwent surgical correction of the anterior cervical osteophytes. Shortly after surgery, the patient improved clinically, but his condition

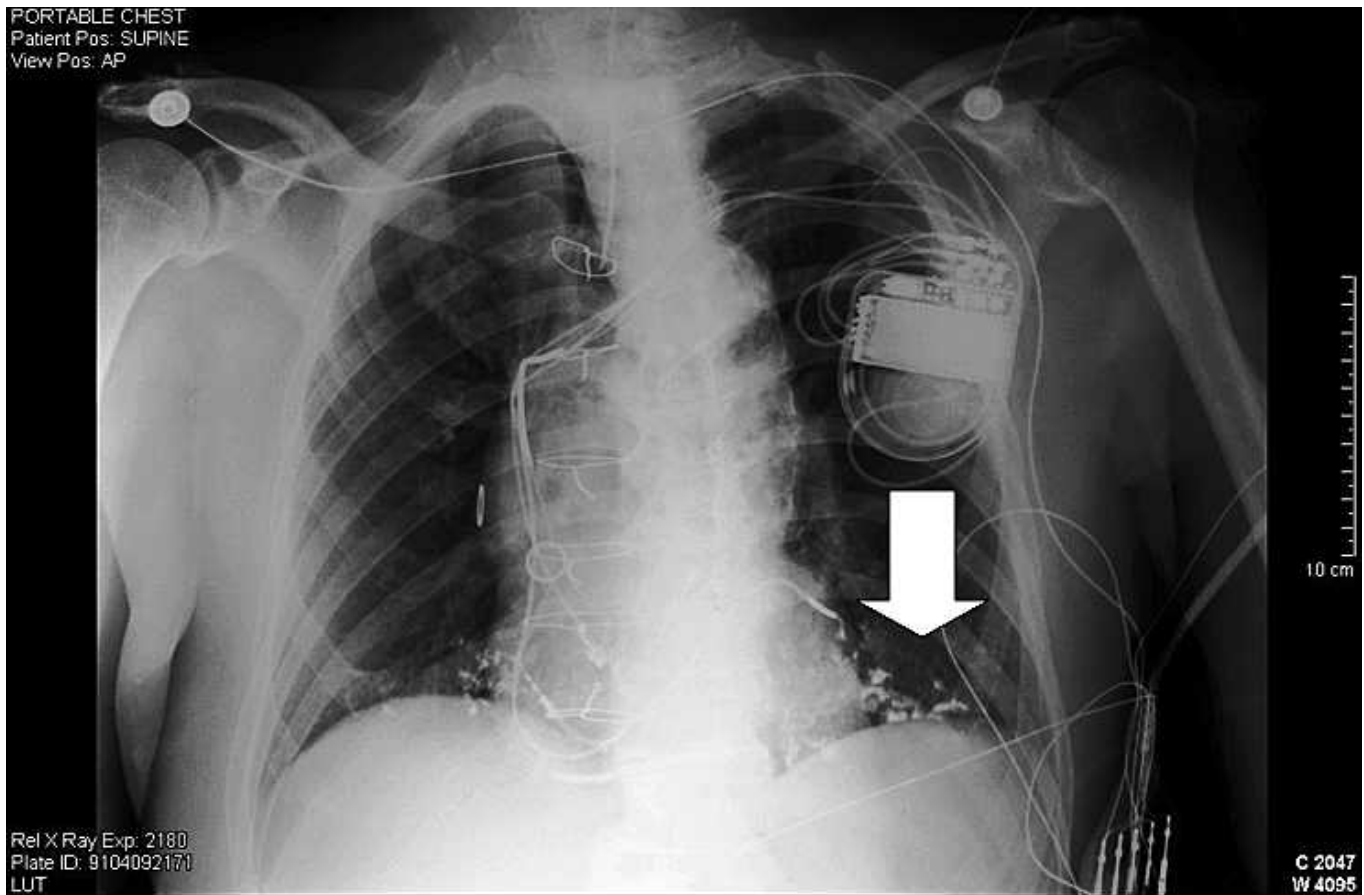


Fig 1. Chest radiograph showed retained barium in bilateral lower lobes of lungs (arrow) with atelectatic/pneumonitic changes and osteophytes in distal right clavicle and both shoulders.

deteriorated after that, and he died 13 days after surgery from aspiration pneumonia.

## DISCUSSION

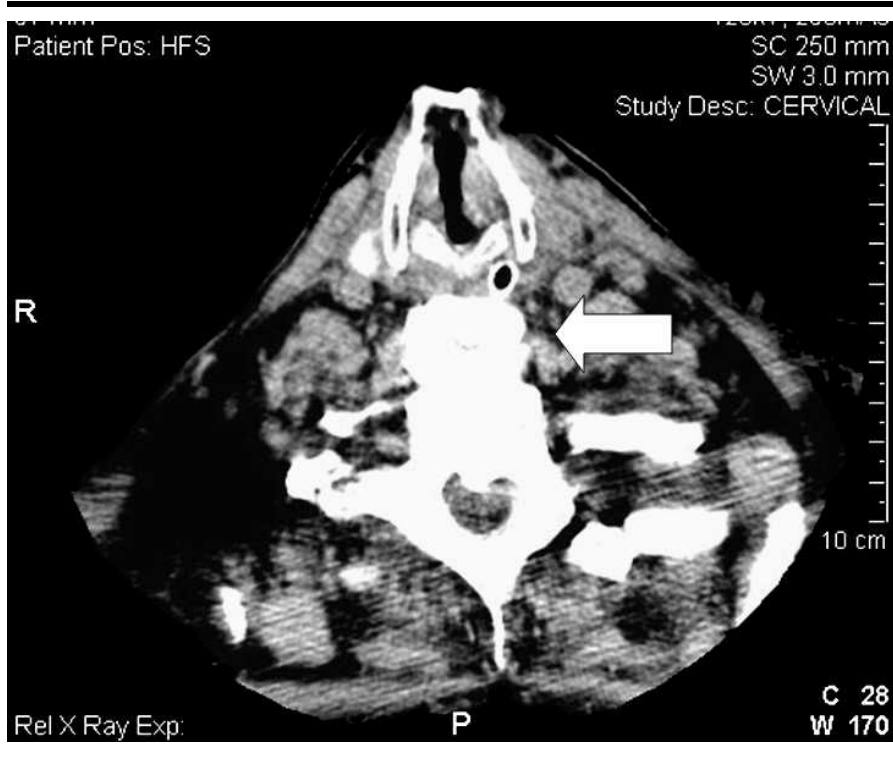
Cervical bony outgrowths are common and have a vast differential diagnosis, which includes DISH, AS, spondylosis deformans, acromegaly, hypoparathyroidism, fluorosis, ochronosis, and trauma.<sup>1</sup> Among these, the most common causes of cervical anterior osteophytes are DISH and AS.

Among 50-year-olds, DISH is more common in men than women.<sup>2</sup> The criteria for spinal involvement in DISH, all three of which must be met, are 1) presence of flowing calcification and ossification along the anterolateral as-

pect of at least four contiguous vertebral bodies, with or without associated localized pointed excrescences at the intervening vertebral body-intervertebral disc junctions; 2) presence of relative preservation of intervertebral disc height in the involved vertebral segment and the absence of extensive radiographic changes of "degenerative" disc disease; and 3) absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or intra-articular osseous fusion.<sup>1</sup> Its etiology is unknown but associated factors are co-morbid diseases (eg, endocrine and metabolic syndromes), environmental causes, toxic drug/nutritional supplementation, genetics, and growth factors, as well as cytokines posttraumatic blunt injury and surgery.<sup>2</sup> DISH is also known as spondylitis ossificans ligamentosa, spon-

dylosis hyperostotica, physiologic vertebral ligamentous calcification, generalized juxta-articular ossification of vertebral ligaments, ankylosing (senile) hyperostosis of the spine, Forestier's disease, spondylosis deformans, and vertebral osteophytosis.<sup>1</sup>

Conversely, AS has a worldwide prevalence ranging up to .9%.<sup>3</sup> Its etiology and pathogenesis are not fully understood.<sup>4</sup> AS diagnostic criteria, the modified New York Criteria, consist of one radiological (sacroilitis, at least erosion and sclerosis bilaterally or at least early ankylosis unilaterally in sacroiliac joint) and three clinical (low back pain and stiffness for more than three months that improves with exercise but is not relieved by rest, limitation of motion of the lumbar spine in both the sagittal and frontal planes, and



**Fig 2. Neck computed tomographic scan showed osteoarthritic changes consistent of anterior cervical osteophytes (arrow) causing displacement of the upper airway and compression of esophagus.**

limitation of chest expansion relative to normal values adjusted for age and sex) criteria. Diagnosis is confirmed when the radiological criterion is associated with at least one clinical criterion.<sup>5</sup> Affected joints show irregular erosion and sclerosis; tissue is replaced by fibrocartilage and becomes ossified. In the spine, the junction of the anulus fibrosus of the disc cartilage and the margin of the vertebral bone undergo irreversible damage. The outer anular fibers are replaced by bone, and the vertebrae become fused, leading to "bamboo spine." If untreated, posture changes, leading to stooped forward carriage of the neck and anterior displacement of the head while walking, can result.<sup>4</sup>

The coexistence of DISH and AS has been reported, although a meaningful association has not been documented. Diagnostic difficulties arise because of spondylitis-type changes, which may occur in some persons with DISH. A

smooth pattern of bone formation in the spine, which is occasionally observed in DISH, resembles the syndesmophytosis of AS. Para-articular or intra-articular bone ankylosis of the sacroiliac joints is evident in DISH, which may be misinterpreted as evidence of AS.<sup>1</sup>

The suggested pathogenesis of DISH indicates that ossification and new bone formation is the result of abnormal osteoblast cell growth/activity in the bony ligamentous region.<sup>6</sup> Insulin-like growth factor I stimulates alkaline phosphatase activity and type II collagen in osteoblasts, and growth hormone can induce the local development of insulin-like growth factor I and insulin-like growth factor binding proteins in chondrocytes and osteoblasts.<sup>7</sup> Patients with DISH have high insulin and growth hormone levels, which explains the osteoblast cell growth and proliferation.<sup>8</sup>

DISH is more prevalent in Caucasians, Japanese, Pima Indians, African

Blacks, and Jews in Jerusalem. It is less common in American Blacks.<sup>2</sup> A study that evaluated the prevalence of DISH in two large American Midwest metropolitan hospital populations showed that DISH was less common in the Black, Native American, and Asian populations, suggesting a genetic origin of the condition.<sup>9</sup> Another prevalence study of DISH in South African Blacks >40 years of age reported that the prevalence of DISH was 3.9% (men 3.8% and women 4.2%). Prevalence increased with age. The prevalence of diabetes mellitus was 52.4% in the 21 patients with DISH seen in a prospective substudy.<sup>10</sup>

Patients with DISH and hyperinsulinemia, with or without diabetes mellitus, have an increased prevalence of antigen HLA-B8, which is present in various endocrine disorders. Patients with DISH may have marked hyperinsulinemia after a glucose challenge, and their degree of obesity correlates with that hyperinsulinemia. Insulin itself may act as a growth factor, theoretically causing the new bone formation seen in DISH.<sup>1</sup> Coronary artery disease and hypertension may be stimuli for DISH,<sup>2</sup> resulting in early endothelial cell damage. The aggregation of platelet-derived growth factor may generate osteoblast proliferation.<sup>11</sup>

Ossification of the posterior longitudinal ligament in DISH occurs with increased frequency in  $\approx 50\%$  of patients. The cervical spine is the most commonly affected region, although occasional exceptions occur. It may explain neurologic findings in patients with DISH (muscle atrophy, sensory loss, numbness, tingling) secondary to compromised integrity of the spinal cord.<sup>1</sup>

Dysphagia in patients with DISH is occasionally apparent and is termed "DISHphagia." Difficulty in swallowing may be secondary to elongation of the styloid process at the base of the skull, calcification or ossification of the stylohyoid ligament, or both, causing

compression of the esophagus.<sup>1</sup> Typically, osteophytes that cause dysphagia are in the C5 cervical interspace. Anterior protrusion of cervical osteophytes in front of the cervical vertebrae can cause hypopharyngeal compression, sensed by the patient as a lump in the throat.<sup>12</sup> The presentation of a patient with throat pain, difficulty in swallowing, and sensation of a foreign body is known as Eagle syndrome and may be evaluated for anterior cervical osteophytes.<sup>1</sup>

The evaluation of cervical osteophytes consists of cervical spine radiograph and barium swallow to confirm their presence.<sup>12</sup> Extending the neck and swallowing a barium-coated marshmallow to reproduce dysphagia during the barium swallow may confirm the obstructive nature of the osteophytes.<sup>13</sup> Videofluoroscopy shows the sequence of muscular changes needed to transfer ingested material from mouth to upper esophagus and rules out neuromuscular causes of dysphagia.<sup>12</sup> In patients with cervical osteophytes, upper gastrointestinal endoscopy may be performed, but cautiously because of the risk of esophageal perforation.<sup>14</sup> Manometry and pH stimulation studies may help to exclude motility disorders and gastroesophageal reflux disease as a cause of dysphagia in the neck.<sup>12</sup>

Most patients with cervical osteophytes can be managed conservatively. Surgical excision is appropriate in patients with severe and progressive symptoms.<sup>15</sup> Speech and language therapies and nutritional counseling are of great importance in the assessment and management of dysphagia.<sup>16</sup>

In conclusion, our patient was a man and had onset of symptoms at >50 years of age, associated diabetes mellitus, arterial hypertension, coronary artery disease, dysphagia, osteophytes in

clavicle and shoulders, anterior cervical osteophytes, calcified posterior longitudinal ligament, and spinal stenosis, all of which are associated with DISH. AS may also be considered because the patient was a man, had a history of stooped posture and anterior displacement of the head while walking, and bamboo-appearing spine. Because of the acute and critical clinical presentation of the patient, complete evaluation for the diagnostic criteria for both disease entities could not be performed. Diffuse idiopathic skeletal hyperostosis and AS are the most common causes of bony outgrowths of the cervical spine causing dysphagia and aspiration in the elderly. Cervical anterior osteophytes are a common and easily diagnosed anatomical pathology of the elderly that must be followed very closely and treated appropriately as symptoms progress to avoid catastrophic complications.

ACKNOWLEDGMENTS

The authors thank Dr. Fanchon Rouce and Dr. Ivonne Z. Jimenez-Velazquez for manuscript review. This publication was made possible in part by grant number R25 RR17589 from the National Center for Research Resources (NCRR), a component of the National Institutes of Health (NIH). Its contents are solely the responsibility of the authors and do not necessarily represent the official views of NCRR or NIH.

REFERENCES

1. Resnick D. *Diagnosis of Bone and Joint Disorders*. 3rd ed. Philadelphia, Penn: Saunders; 1995.
2. Childs SG. Diffuse idiopathic skeletal hyperostosis: Forestier's disease. *Orthop Nurs*. 2004;23(6):375-382.
3. Braun J, Bollow M, Remlinger G, et al. Prevalence of spondylarthropathies in HLA-B27 positive and negative blood donors. *Arthritis Rheum*. 1998;41(1):58-67.

4. Sieper J, Braun J, Rudwaleit M, Boonen A, Zink A. Ankylosing spondylitis: an overview. *Ann Rheum Dis*. 2002;61 Suppl 3:iii8-18.
5. van der Linden S, Valkenburg HA, Cats A. Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York criteria. *Arthritis Rheum*. 1984;27(4):361-368.
6. el Miedany YM, Wassif G, el Baddini M. Diffuse idiopathic skeletal hyperostosis (DISH): is it of vascular aetiology? *Clin Exp Rheumatol*. 2000;18(2):193-200.
7. Vetter U, Zapf J, Heit W, et al. Human fetal and adult chondrocytes. Effect of insulinlike growth factors I and II, insulin, and growth hormone on clonal growth. *J Clin Invest*. 1986;77(6):1903-1908.
8. Atzeni F, Sarzi-Puttini P, Bevilacqua M. Calcium deposition and associated chronic diseases (atherosclerosis, diffuse idiopathic skeletal hyperostosis, and others). *Rheum Dis Clin North Am*. 2006;32(2):413-426, viii.
9. Weinfeld RM, Olson PN, Maki DD, Griffiths HJ. The prevalence of diffuse idiopathic skeletal hyperostosis (DISH) in two large American Midwest metropolitan hospital populations. *Skeletal Radiol*. 1997;26(4):222-225.
10. Cassim B, Mody GM, Rubin DL. The prevalence of diffuse idiopathic skeletal hyperostosis in African blacks. *Br J Rheumatol*. 1990;29(2):131-132.
11. Denko CW, Boja B, Moskowitz RW. Growth promoting peptides in osteoarthritis and diffuse idiopathic skeletal hyperostosis-insulin, insulin-like growth factor-I, growth hormone. *J Rheumatol*. 1994;21(9):1725-1730.
12. Srinivas P, George J. Cervical osteoarthropathy: an unusual cause of dysphagia. *Age Ageing*. 1999;28(3):321-322.
13. Davies RP, Sage MR, Brophy BP. Cervical osteophyte induced dysphagia. *Australas Radiol*. 1989;33(3):223-225.
14. Wright RA. Upper-esophageal perforation with a flexible endoscope secondary to cervical osteophytes. *Dig Dis Sci*. 1980;25(1):66-68.
15. Sobol SM, Rigual NR. Anterolateral extra-pharyngeal approach for cervical osteophyte-induced dysphagia. Literature review. *Ann Otol Rhinol Laryngol*. 1984;93(5 Pt 1):498-504.
16. Logemann JA. Multidisciplinary management of dysphagia. *Acta Otorhinolaryngol Belg*. 1994;48(2):235-238.