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Diffuse idiopathic skeletal hyperostosis causing progressive dysphagia

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Abstract

Introduction: Diffuse idiopathic skeletal hyperostosis (DISH) of the cervical and upper thoracic spine, a relatively common finding in spinal imaging, is rarely associated with upper esophageal pathology. We present a case of progressive dysphagia due to DISH.

Case Presentation: A 64-year-old male with a history of alcohol abuse and smoking presented to the emergency department with a three-day history of chest pain, cough, and emesis associated with oral intake. He noted increased weight loss and progressive inability to swallow liquids and solids. Esophagogastroduodenoscopy was concerning for external compression of the esophagus at the upper esophageal sphincter. Imaging of the spine revealed bulky, flowing osteophytes in the cervical and thoracic spine consistent with diffuse idiopathic skeletal hyperostosis. Dynamic swallow study showed the epiglottis abutting the posterior pharyngeal wall at C2-3 causing incomplete inversion and silent aspiration. He remained nil per os with nutrition supplementation through a nasogastric tube with outpatient neurosurgical follow up.

Conclusions: Diffuse idiopathic skeletal hyperostosis is an abnormal calcification of the anterolateral aspects of the spinal ligaments, and less commonly, the appendicular skeleton. Patients with DISH are usually asymptomatic, but can present with limited cervical mobility, or neck and back pain. Dysphagia is a relatively uncommon finding in DISH. Mild cases are treated conservatively with physical therapy and pain control; however, progressively worsening symptoms or focal deficits require surgical management. Studies have shown that recurrence of osteophytic lesions after surgical management is common.

Background

- Prevalence of dysphagia in persons over 50 years of age is thought to be greater than 20% and as high as 40-60% for those residing in assisted living facilities or nursing homes
- Dysphagia may be classified anatomically as either oropharyngeal or esophageal
 - Oropharyngeal dysphagia is related to the initiation of the swallow from the soft palate to the hyoid bone
 - Esophageal dysphagia relates to any process from the body of the esophagus to the lower esophageal sphincter
- Swallowing is normally controlled by the various cranial nerves described in three distinct phases: oral, pharyngeal, esophageal
 - Oral (divided into preparatory and propulsive) is the volitional component which prepares and propels the food (bolus) into the pharynx
 - Pharyngeal involves closing the airway and projecting the bolus into the esophagus
 - Esophageal phase further transits the bolus into the stomach via peristalsis
- Etiologies of dysphagia include structural (strictures, compression, etc), neurologic (neurodegenerative, CVA, myopathy), and infectious (CMV, HSV, *Candida albicans*)
- Dependent on the specific etiology, symptoms associated with dysphagia can include odynophagia, dysphonia, emesis, aspiration, heartburn, and retrosternal chest pain

- Diffuse idiopathic skeletal hyperostosis, first described in 1950, is a particular type of ankylosing hyperostosis of the spine associated with ossification of the anterior and lateral aspects of the vertebral ligaments, most commonly in the cervical and thoracic spine
- Though less common, DISH can also present with extraspinal hyperostosis, which presents as calcific enthesopathy
- Common diagnostic criteria (Resnick and Niwayama) state that ossifications must span at least four contiguous vertebrae without gross degeneration of the intervertebral disc
 - Lateral radiographs and sagittal CT images of the spine are sufficient for diagnosis
- Prevalence of DISH is debated due to alternative proposed diagnostic criteria, though it is estimated to be 12-28%. Studies have shown males, and those over 40 are at highest risk.
- While most cases of DISH are asymptomatic, common presenting symptoms include limited spinal mobility and pain in the associated region

Case Presentation

- A 64-year-old male presents to the emergency department with a three-day history of chest pain, cough, and emesis with inability to tolerate oral intake
 - He admitted to an extensive history of alcohol abuse with current everyday use and a 30-year smoking history. He noted increased weight loss over the past couple months with concurrent progressive inability to swallow liquids and solids, increasing hoarseness, and limited neck mobility
- Initial cardiac workup was negative for any acute abnormalities. Follow up right upper quadrant ultrasound and HIDA scan were negative for acute cholecystitis.
- Esophagogastroduodenoscopy was concerning for external compression of the esophagus at the upper esophageal sphincter (Figure 4) and esophagitis in the mid- and distal aspects of the esophagus
- CT imaging of the spine revealed bulky, flowing osteophytes in the cervical and thoracic spine consistent with DISH and the following:
 - Left bulky osteophyte at C2-C3 indenting the posterior aspect of the airway (Figure 1)
 - Bulky osteophytes at T2-T3 and T4-T5 indenting the posterior esophagus (Figure 2)
- Dynamic swallow study showed delayed swallow initiation, incomplete epiglottic inversion, incomplete laryngeal vestibule closure, reduced laryngeal sensation in the presence of bolus material, decreased base of tongue approximation to the posterior pharyngeal wall, and reduced pharyngeal stripping
 - Epiglottis appeared to abut the posterior pharyngeal wall at C2-C3 (Figure 5)
- MRI Brain was negative for any acute or chronic infarcts. EMG was negative for any neurodegenerative or neuromuscular disorders.
- The patient remained nil per os with nutrition supplementation through a nasogastric tube with outpatient neurosurgical follow up for further management
- Of note, the patient had a similar episode of dysphagia secondary to compression from osteophytes that required C5-C6 osteophyctomy four years prior

Images

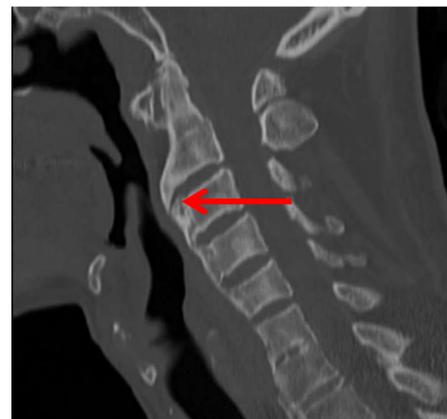


Figure 1: Sagittal CT of the cervical spine showing anterior osteophyte formation indenting the posterior pharyngeal wall



Figure 2: Sagittal CT of the thoracic spine showing anterior osteophyte formation indenting the esophagus



Figure 3: EGD of a normal upper esophageal sphincter



Figure 4: EGD showing external compression at the upper esophageal sphincter

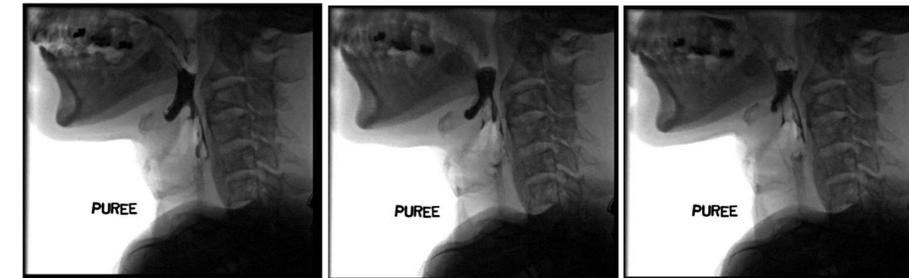


Figure 5: Sequential dynamic swallow study images show the epiglottis abutting the posterior pharyngeal wall during the swallowing mechanism leading to incomplete inversion and silent aspiration.

Discussion

- Diagnostic Algorithm of Dysphagia:
 - If concern includes oropharyngeal dysfunction, ENT examination and dynamic swallow studies should be performed first for determining abnormalities
 - If concern includes esophageal dysfunction, contrast esophagram and upper endoscopy may be more beneficial for diagnosis
- Exact pathogenesis of DISH is still unknown. Some postulate that dietary factors, environmental exposures, vitamin A derivatives, and genetic factors involving HLA genes could all play a role in disease development
- Though similar to ankylosing spondylitis (AS), there are key differences between the two
 - DISH primarily affects the cervical and thoracic spine, while AS targets the lumbar spine
 - DISH is seen in those over 40, while AS affects adolescents and young adults
 - Fusion of the facet, costovertebral, and sacroiliac joints are characteristic of AS
 - While the etiology of DISH is still unknown, HLA-B27 antigen is a key factor in development of AS
- Mechanism of dysphagia associated with DISH focuses on cervical osteophytes, most commonly from C3-C6
 - Protrusions can lead to impairment of epiglottic motility, distortion the laryngeal cartilages, or esophageal obstruction
 - Epiglottic tilting, occurring during the pharyngeal phase of swallowing, is hindered due to mechanical impingement leading to incomplete closure of the laryngeal inlet as the epiglottis cannot fully invert
 - Osteophytes at C3-C4 result in laryngeal dysfunction and increased risk of aspiration
 - Osteophytes at C5-C6 can impact upper esophageal sphincter function and lead to mechanical obstruction
- Mild cases of DISH can be managed with physical therapy and pain control. Progression of disease with increasing symptom severity requires surgical management of protruding osteophytes with osteophyctomy.
- Due to increased recurrence of osteophytes after resection in those with DISH-associated dysphagia, patients should have surgical follow up for up to 10 years post-resection

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