Hypertrophic cervical osteophytes causing dysphagia
A review

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Problems associated with osteophytes of the spine are frequently called to the attention of the neurosurgeon. Diffuse idiopathic skeletal hyperostosis (Forestier's disease) is a common disorder of the spine; a small but important number of these patients present with spondylitic dysphagia. Anterior cervical decompression restores esophageal function. A patient with Forestier's disease is reported, and Forestier's disease is compared and contrasted with other disorders of the spine.

KEY WORDS • diffuse idiopathic skeletal hyperostosis • Forestier's disease • spondylitic dysphagia • cervical osteophyte • ankylosing hyperostosis

Asymptomatic hypertrophic spurs of the anterior margins of the cervical vertebrae may occur in 20% to 30% of the population. Occasionally, dysphagia or even dyspnoea may be caused by such cervical osteophytes pressing against the esophagus, trachea, or nearby soft tissue. Seven of the 116 patients in Saffouri and Ward's series with anterior cervical osteophytes had dysphagia. Although this entity is well recognized, the presence of anterior cervical osteophytes is obscured by confusing terminology: cervical exostosis, cervical arthritis, cervical arthrosis, cervical spondylisis, ankylosing spondylisis, degenerative disc disease, spondylisis deformans, and Forestier's disease (also known as ankylosing hyperostosis or diffuse idiopathic skeletal hyperostosis [DISH]).

Some authors have denied a cause and effect relationship between the anterior cervical osteophytes and the development of dysphagia. Others have acknowledged the entity of spondylitic dysphagia, but have recommended no treatment at all or treatment with steroids or other anti-inflammatory agents. The issue of etiology and treatment has been further confused by reports of associated vertebral or esophageal tumors. Although effective treatment clearly involves neurosurgical intervention, most discussions of the subject have taken place in the non-neurosurgical literature (especially otorhinolaryngological literature).

Recently, we managed a case of spondylitic dysphagia, and thus had occasion to review the literature. After discovering the controversy, we thought it worthwhile to call this entity to the attention of our neurosurgical colleagues, and to review for them the pertinent literature. In addition, we would like to add our case of DISH to those already reported in the world literature.

Case Report

This 77-year-old black woman was healthy until 7 years before admission, when she noticed difficulty in swallowing solid foods. Symptoms progressed until she experienced difficulty in swallowing liquids, 4 years prior to admission. She was seen at another hospital, 1 month before admission, for weakness, dizziness, and difficulty in swallowing; she reported losing 50 lb or more during the last 1 to 3 years. She was advised that she had arthritis and was given follow-up appointments to a neurology clinic. The patient presented to the New York Hospital emergency ward because of progressive fatigue and inability to swallow. She denied significant head, neck, chest, or abdominal pain and discomfort. General review of
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systems and pertinent review of possible neurological problems were unremarkable except for the inability to swallow. The patient's past medical history was unremarkable. The patient took aspirin occasionally for her arthritis and regularly took a "one-a-day vitamin."

Examination. The patient was a pleasant, cooperative woman who drooled oral secretions. Blood pressure was 170/80 mm Hg, pulse 64/min and regular, weight 64 kg, respiration rate 16/min, and temperature 37°C. General physical examination was remarkable for bilateral arcus senilis, left to right deviation of the trachea, generalized decrease in the range of motion of the cervical spine, and an inability to swallow oral secretions. Neurological examination was within normal limits.

Routine cervical spine films revealed prodigious osteophytes of the cervical spine (Fig. 1) involving the bodies of C-3, C-4, and C-5. Esophagram revealed an anterior and rightward displacement of the esophagus and trachea with nearly complete occlusion of the lumen of the esophagus between C-3 and C-6, although motility otherwise appeared normal (Fig. 2). Films of the thoracic, lumbar, and sacral spine were compatible with DISH (Fig. 3). Hemogram revealed a hypochromic microcytic anemia; hemoglobin electrophoresis revealed 29.9% hemoglobin F. The remainder

![Fig. 1. Magnified lateral view of the cervical spine. Prominent osteophytes are present along the anterior surface of the bodies of C-3, C-4, and C-5.](image)

![Fig. 2. Preoperative esophagram. Left: Lateral view. Cervical osteophytes impinge upon and displace the esophagus; the passage of barium is severely compromised. Right: Anteroposterior view. The entire diameter of the esophagus is distorted, and function is altered within the midcervical segment.](image)
Fig. 3. Lateral view of the thoracic spine. Flowing calcification and ossification are seen within the anterior longitudinal ligament, involving the lower thoracic spine (arrowheads).

Fig. 4. Intraoperative cervical spine film. Osteophytes have been surgically decompressed, and the anterior borders of the affected bodies appear smooth.

of the admission laboratory data was essentially normal except for some cardiomegaly demonstrated on the admission chest x-ray film. Stool was negative for occult blood.

Operation. After receiving digitalis for incipient heart failure, the patient underwent anterior cervical exploration for removal of the osteophytes at C-3, C-4, and C-5, and decompression of the esophagus and trachea. The esophagus and trachea were carefully freed from the impinging osteophytes and then gently retracted in order to drill off the offending osteophytes with the Hall drill. Adequate decompression was verified by a lateral cervical spine x-ray film (Fig. 4) obtained in the operating room. A Foley catheter with a 30-cc balloon was placed down the esophagus, the balloon was inflated, and the catheter withdrawn to ensure patency of the esophagus.

Postoperative Course. The patient was given nothing by mouth for 48 hours; she handled oral secretions without difficulty. On the 3rd postoperative day, she had a normal esophagram (Fig. 5) and was started on a liquid diet. Thereafter she progressed to a regular diet. The patient has done well since, gaining 7.5 kg in the first 3 months postoperatively and regaining her old level of energy and sense of well being. She no longer has dizzy spells.

Discussion

Zahn recognized cervical osteophytes causing dysphagia as early as 1904. However, the first two cases of spondylitic dysphagia were reported by Mosher in 1926. Iglauer reported the first surgical excision of an osteophyte 12 years later, in 1938.

Anatomically, the esophagus is fixed by the cricoid cartilage and the diaphragm as the esophagus passes from the neck to the abdomen. Thus, pathophysiologically, the esophagus is most likely to be compressed in the neck by an anterior vertebral mass in the area of the cricoid. Hilding and Tachdjian, in reviewing the literature up to 1960 (36 cases), found that the C5–6 vertebral level was involved in 40% of the cases, the C4–5 level was involved in 23% of the cases, and the C2–3 and C3–4 levels were each involved approximately 14% of the time. When multiple levels were involved, the most common levels involved were C3–5. To date, slightly more than 60 cases have been reported, and these cervical localizations still hold true.

The differential diagnosis of dysphagia includes such entities as tumors of the larynx, esophagus, lung, mediastinum, or spine, esophagitis, esophageal motility disorders, esophageal stricture, aberrant vessels (dysphagia lusoria), Zenker's diverticulum,
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Phemmer-Vinson syndrome, or even globus hysterius. When anterior cervical osteophytes are present on spine films, dysphagia secondary to mechanical obstruction from the projecting bone can be suspected. Caution must be exercised, however, because of the possibility of coexisting, unrecognized esophageal carcinoma. Such bone prominences may result from congenital bone bars, atlantoaxial spurs, osteochondromas, spondylosis deformans, degenerative disc disease, rarely from frank anterior herniation of calcified disc material, and from diffuse idiopathic skeletal hyperostosis.

Diffuse idiopathic skeletal hyperostosis (DISH) is an entity well described in the rheumatology and radiology literature, and in particular by Resnick, *et al.* It is relatively unfamiliar to many clinicians, although it is common in the general population, having been observed in 6% to 12% of routine autopsy specimens. Previously, DISH has been noted in the literature under a variety of names: Forestier's disease, physiological vertebral ligamentous calcification, ankylosing hyperostosis, vertebral osteophytosis, and spondylitis ossificans ligamentosa. It has been confused with ankylosing spondylitis, spondylosis deformans, and degenerative disc disease (Table 1).

The hallmark of this disorder is the intense and luxuriant calcification of the anterior longitudinal ligament, but with only minimal spinal pain or stiffness. Radiographic criteria for diagnosis, as suggested by Resnick, *et al.* are as follows: 1) Flowing calcification and ossifications within the anterior longitudinal ligament of at least four contiguous vertebral bodies, and most commonly involving the lower thoracic spine. Four is an arbitrary figure designed to differentiate DISH from spondylosis deformans, a distinction that may prove to be artificial. 2) In general, a paucity of radiographic signs of degenerative disc disease, although of course these may coexist. 3) Absence of apophyseal joint ankylosis and sacroiliac joint erosion, sclerosis, or intra-articular osseous fusion. However, bridging para-articular osteophytes may occur in DISH over the upper third or fibrous portion of the sacroiliac joint.

Although the radiographic appearance may be quite striking, there is essentially no decrease in spinal mobility on physical examination, although up to 80% of afflicted patients may complain of some mild morning stiffness. Another peculiar manifestation of this disease is the tendency for calcification at sites of ligament and tendon attachments to bone, occurring in extraspinous as well as axial locations. Patellar, olecranon, and calcaneal spurs are found, and there is associated peripheral bone and joint pain in 33% of cases.

Dysphagia was found to be a prominent symptom in patients with DISH in the series reported by Resnick, *et al.* They found dysphagia in 17% to 28% of the histories, and in 8% it required surgical removal. It is highly probable that the great majority of the cases of cervical dysphagia reported in the older literature represented patients with DISH, or a forme fruste thereof, since the largest anterior osteophytes are found in this disorder (Table 1).

Once the anterior osteophyte is large enough to compress or displace the esophagus (and/or the trachea), patients may complain of dysphagia, dysphonia, a sensation of a foreign body in the throat, or a constant urge to clear the throat. Occasionally, patients complain of pain in the throat, or pain referred to the shoulders, arms, or hands, or even dyspnea. On examination, the trachea may be displaced from the midline with a hard palpable mass between the soft tissue of the sternocleidomastoid laterally, and the trachea and esophagus medially. Horner's syndrome or recurrent nerve paralysis has been reported in some cases. Indirect laryngoscopy with a tongue depressor and mirror discloses high cervical lesions. Lower lesions of the cervical spine are more simply elucidated with lateral spine films and an esophagram. Although some clinicians have recommended endoscopy, this may in fact prove dangerous and has been a common cause of esophageal perforation.

A filling defect visible on esophagraphy has misled some into thinking that the lesion was a tumor of the esophagus, when in fact an osteophyte was involved or vice versa. It would seem that careful examination of the patient, his history, and his diagnostic radiographic studies (both plain films and esophagram) should obviate such errors.
# TABLE 1

**Differential diagnosis of anterior vertebral osteophytes***

<table>
<thead>
<tr>
<th>Site</th>
<th>DISH</th>
<th>Spondylosis Deformans</th>
<th>Ankylosing Spondylitis</th>
<th>Intervertebral Osteochondrosis</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Radiographic Features</strong></td>
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<tr>
<td>vertebral bodies</td>
<td>“flowing” anterior ossification involving four or more contiguous vertebrae</td>
<td>large anterior osteophytes (not as large as in DISH)</td>
<td>thin syndesmophytes</td>
<td>sclerosis of superior &amp; inferior surfaces</td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td>Schmorl’s nodes</td>
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<tr>
<td>intervertebral discs</td>
<td>normal or only mild decrease in height</td>
<td>normal or only mild decrease in height</td>
<td>normal or convex in shape</td>
<td>moderate to severe decrease in height</td>
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<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>“vacuum phenomenon”</td>
</tr>
<tr>
<td>apophyseal joints</td>
<td>normal or only mild sclerosis with occasional osteophytes</td>
<td>normal or only mild sclerosis with occasional osteophytes</td>
<td>erosions, sclerosis, bone ankylosis</td>
<td>almost all show the secondary changes of osteoarthritis</td>
</tr>
<tr>
<td>sacroiliac joints</td>
<td>para-articular osteophytes (only in the upper 1/3, the fibrous portion)</td>
<td>normal</td>
<td>erosions, sclerosis, bone ankylosis (starts in the lower 2/3, the synovial portion)</td>
<td>normal</td>
</tr>
<tr>
<td>peripheral skeleton</td>
<td>ossification at tendon insertions</td>
<td>normal</td>
<td>“whiskering”</td>
<td>normal</td>
</tr>
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<tr>
<td><strong>Clinical Features</strong></td>
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<tr>
<td>spinal pain &amp; stiffness</td>
<td>mild in about ¼, asymptomatic in the rest</td>
<td>usually mild</td>
<td>severe</td>
<td>mild to severe</td>
</tr>
<tr>
<td>etiology</td>
<td>HLA-B27 antigen in 34% (present in 8% of controls)</td>
<td>a tear in Sharpey’s fibers at edge of annulus with resulting prolapse of relatively nondegenerated disc causing traction osteophytes to form in anterior longitudinal ligament; associated with mechanical stress, as in heavy manual labor or on concave side of a scoliotic spine</td>
<td>HLA-B27 antigen present in 88%-95%</td>
<td>dessication</td>
</tr>
<tr>
<td></td>
<td>may represent an advanced or severe form of spondylosis deformans</td>
<td></td>
<td></td>
<td>progressive loss of chondroitin sulfate</td>
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<td>incidence</td>
<td>6%-12% of autopsy series, more common in males, mean age 65-67 yrs</td>
<td>60% of women &amp; 80% of men by 49 years</td>
<td>1 to 3 per 1000, young males</td>
<td>increases with age, 100% over 90 years</td>
</tr>
<tr>
<td>sedimentation rate</td>
<td>mild increase in 22%</td>
<td>normal</td>
<td>elevated in 80% initially &amp; during exacerbations</td>
<td>normal</td>
</tr>
</tbody>
</table>
The exact pathophysiology of DISH is unclear, although alterations in tissue elasticity and osteoelasticity have been invoked, as well as local factors to explain this lesion. In addition, while it has been suggested, no particular role by either trauma or generalized metabolic (endocrine) disturbance has been clearly demonstrated.10,11,32

Piquet26 reported radiotherapy useful in treating dysphagia due to osteophytes, although most authors probably would not agree with this. Similarly, steroids or other anti-inflammatory agents have been recommended by some, but these appear effective only in reducing edema of the trachea, larynx, esophagus, or soft tissue of the neck without frankly treating the osteophyte itself.37 Osteophytic dysphagia has been treated not at all or with gastrostomy;9,10,29 however, thoughtless manipulation of the trachea or esophagus during the procedure might certainly lead to perforation of either of these structures, since the walls are frequently thinned out and weakened by the local long-standing compression.1 Subsequent fistula or abscess formation could result. Careful dissection and retraction avoids this complication.

References

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