Young adult and giant cervical exostosis

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ABSTRACT

تؤدي التغيرات التنكسية في العمود الفقري إلى تشكل النابتات العظمية (osteophytes) على السطح الأمامي للفقرات الرقبية. وتسبب النابتات العظمية بعض الأعراض السريرية وذلك حسب موقعها، وتتمثل هذه الأعراض في عسر البلع، والبحة، والصرير. نستعرض في هذا المقال حالة مريض جاء إلى المستشفى بعد إصابته بعسر في البلع مع ألم في منطقة الرقبة. لقد أظهرت الاختبارات والتحاليل أنه كان يعاني من فرط التعظم الهيكلي والمجهول السبب. وفيما يلي سوف نقوم باختصار وصف الخواص التي ظهر بها المريض، ونتائج التصوير الشعاعي، بالإضافة إلى خيارات العلاج المقترحة.

Degenerative changes in the spine can result in the formation of osteophytes on the anterior surface of the cervical spine. Depending on their site, osteophytes can bring on clinical manifestations such as dysphagia, hoarseness, and stridor. We discuss an interesting case of a young adult patient who presented with dysphagia along with neck discomfort, and on investigation was found to be suffering from diffuse idiopathic skeletal hyperostosis. Here, we briefly portray the presenting features, radiographic findings, and management options.

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Degenerative changes in the spine can result in the formation of osteophytes on the anterior surface of the cervical spine. Due to the close relationship of the esophagus to the cervical spine, spinal disorders in the neck can interfere with esophageal function. Extrinsic compression by large anterior osteophytes may occur in cervical spondylosis and especially in diffuse idiopathic skeletal hyperostosis (DISH).[1] The posterior wall of the pharynx is separated from the vertebral column only by a thin layer of soft tissue. Below the level of the glottis, the esophagus lies in front of the vertebral bodies from C4 caudally with the trachea anterior. Thus, cervical pathology above C4 may compress the pharynx, and lesions below may cause either esophageal or pharyngeal symptoms.[2] In addition to anterior osteophytosis,[3] ossification of the annulus fibrosus and longitudinal ligament in ankylosing spondylitis can also compress the esophagus, resulting in dysphagia. Infective causes include cervical spine osteomyelitis with a pre-vertebral abscess.[4] Other differential diagnosis includes esophageal strictures, Zenker's diverticulum, motility disorders, Plummer-Vincent's syndrome, esophageal tumors, and other mediastinal mass lesions. We are reporting a case of a patient with a multiple, giant anterior osteophytes arising from cervical spine presenting as dysphagia and neck discomfort, and on investigation was found to be suffering from DISH.

Case report. A 35-year-old adult male presented to our outpatient department with a vague complaint of neck discomfort and dysphagia for the last one year. The patient described that forward bending of neck increases neck pain or discomfort. The patient also had difficulty in swallowing food, especially solids one. There were no symptoms of aspiration, neck stiffness, cough, stridor, and snoring. There was no history of any trauma, surgery, or any instrumentation prior to the onset of the symptoms. Examination revealed painful restriction of terminal flexion of cervical spine. However, no obvious mass was palpable in the neck. Radiological examination of the cervical spine was carried out, which showed giant cervical anterior osteophytes arising from C3, C4, C5, and C6 vertebrae (**Figure 1**). Radiological finding was consistent with the diagnosis of DISH. The rest of the skeletal survey was normal. This patient was advised surgery after a thorough explanation of the prognosis of the disease. The patient refused surgery and was managed conservatively with analgesics, antibiotics, short course of steroids, dietary modifications, and neck

Figure 1 - Lateral radiograph of cervical spine showing multiple, giant, anterior bridging osteophytes arising from C3 to C6 vertebra (arrow heads). Note the disc space is maintained.

physiotherapy. Following conservative treatment, the neck pain and discomfort improved, whereas dysphagia improved marginally. At the latest follow-up, the patient is doing well with dietary modifications.

Discussion. Cervical spondylosis generally presents as neck pain and radiculopathy. The close relationship between the spinal canal and its neural contents only allows limited space for small osteophytic changes without neurological impairment. Accordingly, compromise of the neural structures is the most common indication for surgery in the cervical spine.[2] Small anterior osteophytes rarely cause symptoms due to the compression of the pharynx, esophagus, or upper airways. Such symptoms more often follow local surgery.[5] However, large bridging anterior osteophytes of the cervical spine are frequently the result of DISH, and they can cause compression of the oropharyngeal swallowing structures. The DISH or Forestier's disease, or ankylosing hyperostosis is a rare clinical entity of unknown etiology seen in middle-aged and elderly patients. It is more common in males than in females. It affects up to 10% of patients older than 65 years of age.[3] Even though these patients are in general asymptomatic, there are citations of DISH patients presenting with spinal instability, upper gastrointestinal, respiratory, and neurological symptoms.[6-8] These patients can develop osteophytes, which can lead to extrinsic compression on local tissues. Depending on their site, osteophytes can bring on clinical manifestations such as dysphagia, hoarseness, and stridor.[9] Our patient presented with dysphagia and neck discomfort. The obstruction occurs most commonly at C5 and C6, and less commonly at more cephalic levels.[10]

Dysphagia is found in relation with the anterior block fusion and remodeling of several segments figuring as outgrowths from the ventral surface of the cervical spine. Giant cervical exostosis is usually more commonly seen in elderly population. Although young patients presenting with giant cervical exostosis have been reported in the literature, however, all of them were having some pre-existing predisposing factor for early degenerative changes.[11] It is probable that the existence of predisposing factors in the cervical spine leads to excessive stresses, resulting in early degenerative changes and formation of giant cervical exostosis. Fried et al[11] reported an incidence of 0.6% for formation of block vertebrae in the cervical spine. Their patients were aged between 15 and 80 years. Our patient presented with giant cervical exostosis at a young age in the absence of any pre-existing predisposing factor for early degenerative changes.

Cervical spine osteophytosis as a cause of dysphagia was first reported by Mosher in 1926,[12] and Iglauer[7] in 1938 reported the first surgical excision of a cervical spine osteophyte producing dysphagia. Thoracic spine osteophytosis hardly ever result in dysphagia as the thoracic portion of the esophagus is relatively mobile, and by virtue of its mobility, it is displaced rather than compressed by the osteophytes. Resnick et al[13] reported a 17-28% incidence of dysphagia due to cervical spine hyperostosis in patients with DISH, and surgical intervention via an anterior cervical approach was essential in 8% who failed to respond to conservative treatment. It is important to note that owing to the bone projection away from the spinal cord, it is rare for a patient to have a symptomatology that would draw initial assessment by an orthopedic surgeon.

The diagnosis of DISH is based on the following radiographic criteria: 1) flowing calcification and ossification within the anterior longitudinal ligament connecting 4 or more adjacent vertebral bodies, 2) minimal to no degenerative disc changes, and 3) absence of apophyseal joint ankylosis and sacroiliac joint erosion.[13,14] Of note among the pathological conditions that can be confused with DISH are osteophytes accompanying degenerative disc disease, and ankylosing spondylitis. These radiographic criteria allows DISH to be differentiated from ankylosing spondylitis and degenerative disc disease.

Management of patients with DISH depends on the severity of the disease and symptomatology. Most patients respond to dietary modification, swallowing therapy, non-steroidal anti-inflammatory drugs, muscle relaxants, antibiotics, and steroids.[15] Surgical excision is obligatory when symptoms are upsetting, dysphagia influences nutritional status of the patient, or the airway becomes compromised, and in this case, excision of the osteophyte via anterolateral, posterolateral, or transoral approach may be carried out.[6]

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