

Radiological approach to Forestier Rotes Querol disease. A case report

Enfoque radiológico de la enfermedad de Forestier-Rotes-Querol. Presentación de caso

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Key words (MeSH)

Hyperostosis

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Palabras clave (DeCS)

Hiperostosis

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Summary

The case of a 64-year-old female with associated comorbidities is presented, who attends the Honduran social security institute due to chronic cervical pain and stiffness. Magnetic resonance studies show bone protrusion with formation of anterior bridges of vertebral bodies that is consistent with the alterations visualized in tomography and cervical radiography. Idiopathic diffuse skeletal hyperostosis is a poorly recognized pathology of the spine that can be asymptomatic and unnoticed. In case of cervical pain, spinal tomography is the initial and ideal study method for bone assessment in case of ossifying and degenerative pathologies that, when complemented with magnetic resonance imaging, helps to make an accurate diagnosis and thus evaluate and decide between surgical treatment or conservative management.

Resumen

Se presenta el caso de una paciente femenina de 64 años de edad, con comorbilidades asociadas, quien acude al Instituto Hondureño de Seguridad Social por dolor y rigidez cervical de evolución crónica. En estudios de resonancia magnética se observa protrusión ósea con formación de puentes anteriores a los cuerpos vertebrales, que concuerda con las alteraciones halladas en tomografía y radiografía cervical. La hiperostosis esquelética difusa idiopática, o enfermedad de Forestier-Rotes-Querol, es una patología de la columna poco reconocida que puede cursar asintomática y desapercibida, pero ante dolor cervical la tomografía de columna es el método de estudio inicial e ideal para la valoración ósea en caso de patologías osificantes y degenerativas, que al complementar con resonancia magnética ayuda a realizar un diagnóstico preciso, para decidir entre un tratamiento quirúrgico o un manejo conservador.

Introduction

In 1950, Forestier and Rotes-Querol published a series called “senile vertebral ankylosing hyperostosis”. Later, in 1971, Forestier and Lagier described spinal hyperostosis in young adults omitting the term senile and distinguished it as a separate nosological entity from spondyloarthritis and ankylosing spondylitis (1).

Diffuse idiopathic skeletal hyperostosis (DISH) is an idiopathic form of degenerative arthritis that typically affects individuals over 60 years of age (with an average prevalence of approximately 10% in people over 50 years of age), of unknown etiology. The worldwide prevalence ranges from 2.9% to 42% (2).

This pathology is more frequent in men than in women, with a 2:1 ratio, a difference that decreases with age; it is limited to the spine but can also affect multiple peripheral sites independently.

Extraspinous entheses ossifications are frequent, and when found in isolation may lead to the diagnosis of DISH. Coexistence of this entity with ankylosing spondylitis has been reported (3).

Although most patients with cervical involvement are asymptomatic or have only limited mobility or nonspecific pain, there are several reports of patients with symptoms secondary to compression of the pharynx and airway, such as dysphagia, stridor, dysphonia and difficulties with intubation or endoscopy, so the diagnosis of this entity should be suspected as a possibility in cases of dysphagia, once other more common causes of dysphagia have been ruled out (1, 4).

The symptoms of the disease are usually determined by ossifications of the periosteum, most frequently in the areas of insertions of ligaments and tendons; the most common localization is in the thoracic region of the spine, infrequently in the cervical area (5).

The excellent spatial resolution of computed tomography (CT) allows an accurate evaluation of the facet joints, which is an important aspect to differentiate DISH from ankylosing spondylitis.

The case presented here is intended to provide insight into the different diagnostic methods for this spinal pathology and, in this particular case, the com-

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plementation of CT and magnetic resonance imaging (MRI) will help in the discrimination of sea-discal pathology.

Case presentation

A 64-year-old female patient with a history of diabetes, arterial hypertension and hypothyroidism presents with symptoms characterized by cervical pain and stiffness of the cervical spine of chronic evolution.

Physical neurological examination revealed a right lateralization gait, cervical pain and stiffness with limited mobility. Osteotendinous reflexes, vibration and sensibility were preserved in the upper limbs. Radiological, CT and MRI studies were performed to rule out degenerative discocervical pathology.

The cervical CT with sagittal reconstruction and axial sections showed ossification along the anterior aspect of the vertebral bodies from C2 to C7 with preservation of the intervertebral spaces, which was in agreement with what was visualized in the cervical X-ray in sagittal projection (figure 1).

The MRI of the cervical spine showed bone growth in the anterior border of the vertebral bodies that protrudes to the prevertebral space and that is posterior to the pharynx and larynx, corresponding to bridges between the vertebrae, which do not compromise the intervertebral space and without evidence of disc or radicular pathology (figure 2)

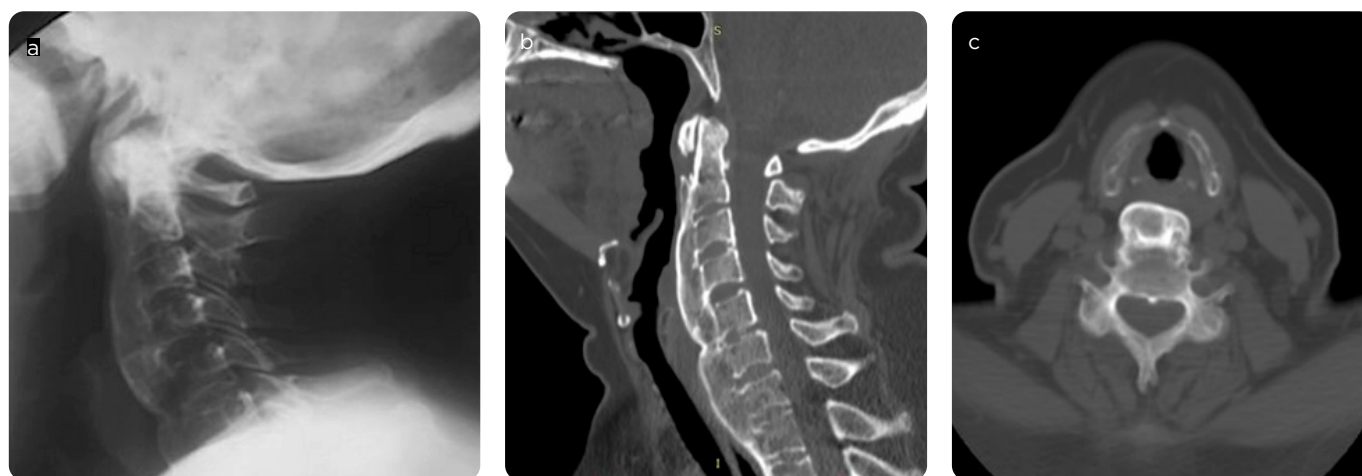


Figure 1. a) X-ray of the cervical spine, lateral projection: loss of lordosis and extensive hyperostosis anterior to the anterior longitudinal ligament. b and c) CT of the cervical spine sagittal reconstruction and axial section: ossification along the anterior aspect of the vertebral bodies from C2 to C7 with preservation of the intervertebral spaces.

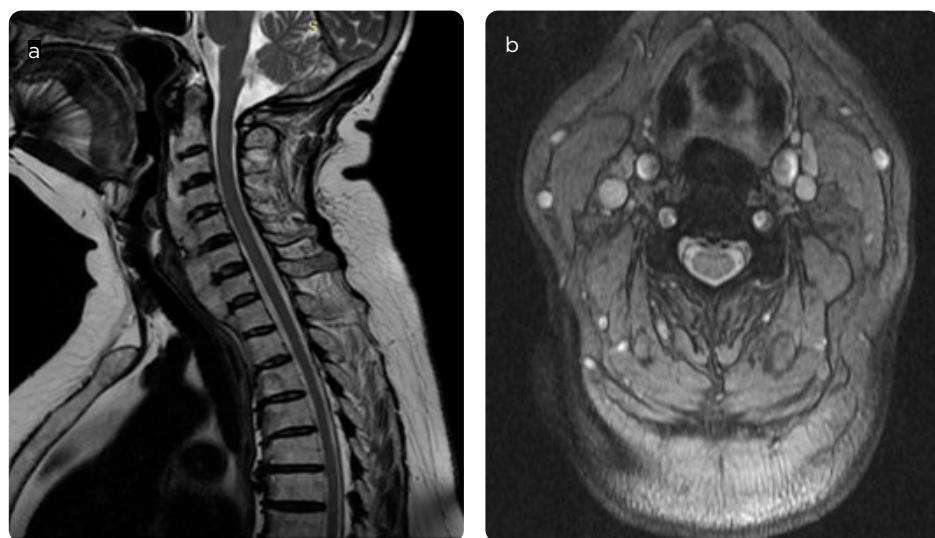


Figure 2. a and b) MRI Sequence with sagittal T2 information and axial MERGE (multiple echo for recombined gradient echo) sequence. Bone growth on the anterior border of the vertebral bodies that protrudes into the anterior prevertebral space and is posterior to the pharynx and larynx. It corresponds to bridges between vertebrae that do not compromise the intervertebral space, without evidence of disc pathology, spinal cord compression or radicular.

Discussion

Diffuse idiopathic skeletal hyperostosis (DISH) is a disorder characterized by ossification and calcification along the anterolateral aspect of the vertebral bodies and peripheral entheses (6).

Several causative agents have been suggested, such as diabetes mellitus, obesity, old age, hyperuricemia, arterial hypertension, hyperinsulinemia and insulin-like growth factor. Some papers have also cited trauma, occupation, fluorosis, infectious diseases, autoimmune origin (HLA-B27, HLA-B5, HLA-A11), neuroarthropathy and hyper-*vitaminosis A* as primary causes (2, 7).

In 1976, Resnick and Niwayama (8) proposed diagnostic criteria for DISH in the spine based on radiologic features: 1) ossification of at least four contiguous vertebral bodies; 2) relative preservation of intervertebral disc space; and 3) absence of either apophyseal joint ankylosis and joint erosion, sclerosis or fusion either intra-articular or sacroiliac.

The first two criteria help to distinguish it from spondylosis and the third from ankylosing spondylitis. Although in recent studies of patients with DISH undergoing lumbar spine surgery, anterior bone bridging and sacroiliac joint ankylosis were more frequent.

Differential diagnosis is sometimes difficult because both pathologies may be present. Hyperostosis can also be observed in the ankle, in the posterior aspect of the calcaneus and in the elbow, in the insertion of the triceps in the olecranon (2, 6).

Spine CT shows ossification along the anterior aspect of the vertebral bodies, and reconstructions in coronal and sagittal planes usually show the classic pattern of ossification and bone bridges. MRI is generally not indicated initially in DISH. However, it can be valuable when ossification of the posterior longitudinal ligament causes neurological symptoms, as it allows determination of the extent of such ossification, the effect of the mass on the thecal sac and compression of the spinal cord (2).

In the case studied here, the patient had some risk factors and the hyperostosis was located in the cervical region, the site of least occurrence; but there are cases in the literature of greater complexity, such as the one described by Ito et al, where the patient presented hyperostosis of the anterior and posterior longitudinal ligament with stenosis of the spinal canal, as well as hyperostosis of the ligaments between the spinous apophyses and anterior osteophytosis which made swallowing difficult even after the first surgical intervention (7).

The differential diagnosis includes spondylosis deformans, ankylosing spondylitis, psoriatic arthritis, fluorosis, osteomalacia, acromegaly, pachydermoperiostitis, hypophosphatemia and hypoparathyroidism. Initial management of DISH should be conservative; it consists of dietary changes, anti-inflammatory drugs, muscle relaxants, corticosteroids and postural changes during the swallowing process. Surgery, for progressive or very symptomatic cases, is usually performed by anterior approach with favorable results, but has the disadvantage that the disease may recur, especially when performed in the more mobile segments (8-10). The disease in general has a good prognosis; surgery is required in those cases with cervical involvement involving the digestive or upper respiratory tract.

Conclusion

Forestier-Rotes-Querol disease, an unusual cause of neck pain, may go unnoticed and be an incidental finding on imaging studies; however, the radiologic approach is ideal for ruling out degenerative and other causes and is a tool for rapid and effective diagnosis. Among the diagnostic modalities, CT as an initial method helps to evaluate bone involvement, to consider conservative or surgical treatment. MRI is complementary to evaluate the involvement of other structures and to give a broad view to the treating physician.

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