Retro-odontoid pseudotumor: a rare complication of Diffuse Idiopathic Skeletal Hyperostosis

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INTRODUCTION

Diffuse Idiopathic Skeletal Hyperostosis (DISH) is a proliferative, mostly asymptomatic bone disease, featuring diffuse calcification and ossification of ligaments and entheses. DISH classically affects the anterolateral aspects of the thoracic spine, but the cervical and lumbar segments can also be involved by marked ligament ossification. Clinical manifestations include progressive restriction of spinal movements, with little or no associated pain, and symptoms related to cervical ossification, as dysphagia and airway obstruction. Neurologic manifestations are uncommon and mainly caused by mechanical enthesophyte myelocompression[1]. The formation of retro-odontoid masses (pseudotumors) is a rare cause of myelopathy in DISH and has been seldomly reported[2,3].

CASE REPORT

A 76-year-old male was referred to our clinic complaining of progressive limitation of spinal movements in the past ten years. He referred intermittent cervical mechanical pain but no limb numbness or weakness. He was being treated for Arterial Hypertension and Diabetes Mellitus type 2 for the past twenty years and had a sedentary lifestyle. Physical examination showed cervical kyphosis and lumbar rectification. The spinal range of motion was extremely limited but painless in all segments, particularly in the cervical spine. The neurological and further musculoskeletal exam was unremarkable. Plain radiographs of the spine showed massive ossification of the anterior longitudinal ligament in the cervical spine (Figure 1), suggesting DISH. Coarse osteophytes were also prominent in the thoracic and lumbar segments. Cervical magnetic resonance imaging (MRI) was performed to study the atlantoaxial transition. A retro-odontoid pseudotumor of fibrous aspect was evident, with no clear neural compromise (Figure 2). The patient was referred to the neurosurgery department for monitoring, as a surgical approach might be needed in the case of pseudotumor progression conditioning myelocompression.

FIGURE 1. Plain radiograph of the cervical spine (lateral incidence). Diffuse ossification of the anterior longitudinal ligament (arrows).

DISCUSSION AND CONCLUSIONS

Retro-odontoid pseudotumors are uncommon non-neoplastic masses whose pathophysiology is mainly linked to chronic atlantoaxial instability. The increased motion of the craniocervical junction generates micro-

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FIGURE 2. T2-weighted magnetic resonance imaging of the cervical spine. A low-signal intensity retro-odontoid mass (arrow) slightly reduces the canal's patency by an impression of the anterolateral right side of the cranial cervical cord but without clear neural compromise. The retro-odontoid pseudotumor is hypointense in T1 and T2 sequences, suggesting a fibrous nature.

injuries and a consequent reparative process with fibrocartilaginous metaplasia of the peri-odontoid soft-tissues[4]. In DISH, C1-C2 transition becomes hypermobile in patients with extreme rigidity of the more ossified lower cervical spine. Although rare in DISH, retro-odontoid pseudotumors can lead to severe complications as tetraparesis due to medullar compression[2,3]. Special attention must be given to the detection of early signs and symptoms suggesting myelopathy in all patients with advanced DISH.

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