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Case Report

Cervico-occipital junction infectious osteoarthritis complicated with atlantoaxial dislocation, cervical abscesses and epiduritis: A case report and review of the literature ☆,☆☆

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ABSTRACT

Craniocervical junction infections are considered a rare condition and can be associated with degradation of the odontoid ligaments and, the risk of a subsequent atlantoaxial subluxation or dislocation. Osteomyelitis at this level is often seen in immunocompromised patients with, intravenous drug use or infective endocarditis. Atlantoaxial subluxation associated with pharyngeal infection or its surrounding tissues is called Grisel's syndrome. We report the case of a 29-year-old man diagnosed with infectious cervico-occipital osteoarthritis complicated by atlantoaxial dislocation, abscesses located in the retropharyngeal space, and the prevertebral space as well as upper cervical epiduritis. Our purpose is to highlight the role of radiologists in diagnosing this rare yet deadly condition.

Introduction

Craniocervical junction lesions are congenital or acquired abnormalities of the occipital bone, foramen magnum, or first 2 cervical vertebrae. These abnormalities can be responsible for decreasing the space for the lower brain stem and cervical cord. Considered a rare condition, upper cervical spine infections can be associated with degradation of the odontoid ligaments and, the risk of a subsequent atlantoaxial subluxation or dislocation. The prevalence of osteomyelitis at this level has increased significantly over the past decades primarily due to immunocompromised hosts, intravenous drug use, and infective endocarditis [1].

Atlantoaxial subluxation associated with pharyngeal infection or its surrounding tissues is called Grisel's syndrome [2]. In this paper, we report the case of a 29-year-old man, with an extremely uncommon case of infectious cervico-

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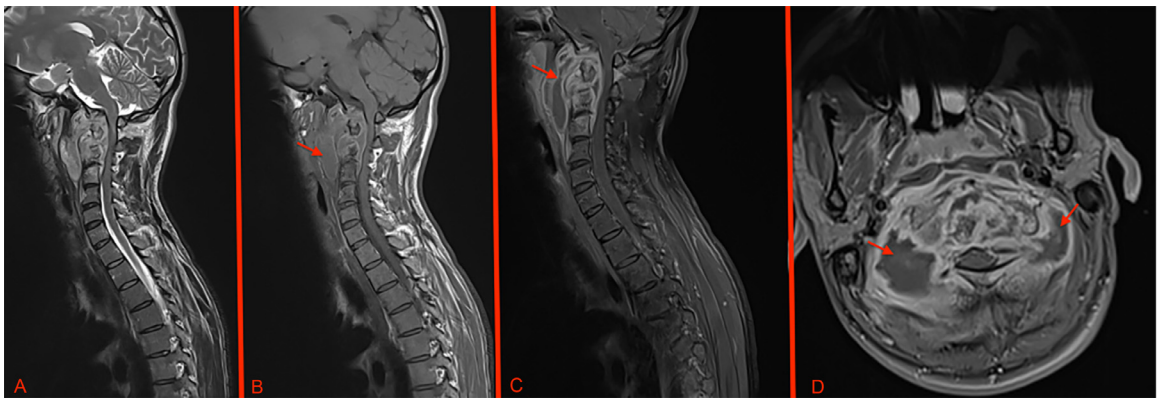

Fig. 1 – Sagittal T2 weighted images (A) / Sagittal T1 weighted images without (B) and after injection of Gadolinium with FAT SAT (C) / Axial T1 weighted images after injection (D), showing multiples diffuse collections (red arrows) in the prevertebral space with a thick wall, peripherally enhancing infiltration the paravertebral muscles.

occipital osteoarthritis complicated by atlantoaxial dislocation, abscesses located in the retropharyngeal space, and the prevertebral space as well as upper cervical epiduritis [3].

Case report

We report the case of a 29-year-old man, initially treated for **chronic** suppurative **otitis media,** presenting to our department for febrile torticollis going back 3 days ago with inflammatory retro-auricular swelling.

Clinical examination found a patient with spastic tetraparesis without sensory disturbances or associated dysfunctions. There was a swelling in the retro-auricular space associated with redness and pain. He was noted on admission to be febrile (T 39.0°C), with a high initial white blood cell (WBC) of 14.52 × 10^9/L with 75.7% neutrophils and a C-reactive protein (CRP) of 170 mg/L. **Magnetic resonance imaging (MRI)** of the brain and the cervical spine was performed due to the acute onset of tetraparesis. It showed an enlargement of the C1-C2 joint space measured at 13 mm associated with large diffuse collections extended to the peri vertebral spaces, in hyposignal T1 and heterogeneous intermediate signal in T2 sequences, enhanced in the periphery after injection of Gadolinium, indicating abscesses. These collections extend to the atlanto-occipital joints as well as the trapezius muscle and the right subcutaneous soft tissue (Fig. 1). It includes the IJV which has a filiform appearance and is the seat of endoluminal material, in favor of thrombosis.

These collections also invade the retro and parapharyngeal spaces but remain limited by the long muscles of the head, and extend to the posterior soft tissues (Figs. 2 and 3).

Furthermore, the abscesses extend in the spinal canal creating a circumferential epiduritis compressing the cervical cord which is the seat of hypersignal T2 lesion extended until C4 (Fig. 3).

These abnormalities were responsible for an anteroposterior and superior atlantoaxial dislocation compressing the bulb and the bulbo-medullary junction with a basilar impression (Fig. 4). We also noted a lytic clivus and right otomastoiditis (Fig. 5).

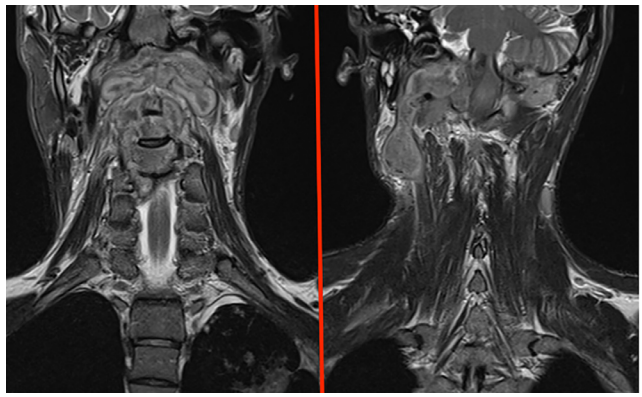

Fig. 2 – Coronal T2 weighted images showing the collections having heterogeneous signals with a thick wall, infiltrating the para vertebral spaces.

We concluded to a cervico-occipital infectious osteoarthritis with dislocation and basilar impression, retro-pharyngeal, and perivertebral abscesses as well as circumferential epiduritis and myelopathy, all associated with partial thrombosis of the right IJV.

The patient was then referred to the neurosurgical department where he was also reviewed by the ENT and the diagnosis was confirmed. He was then placed on cervical collar immobilization.

A surgical drainage of the retropharyngeal abscess was initially performed with an initial course of IV antibiotics administrated afterward. The neurosurgical then team opted for a posterior approach with C0-C5 fusion. A C2-C3 laminectomy was also realized with a C3-C4 corpectomy and expandable cage. The patient remained in intensive care after the intervention and unfortunately passed away after 15 days of ventilation complications.

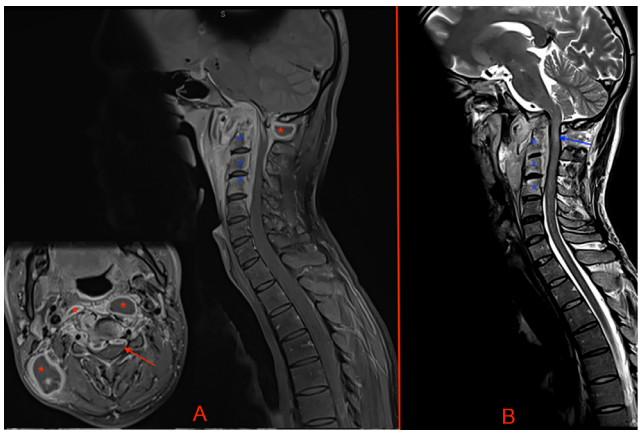

Fig. 3 – (A) Axial and sagittal T1 weighted images after injection of Gadolinium with FAT SAT and (B) sagittal T2 weighted images, showing retropharyngeal and latero-cervical collections (red asterisk) extending to the epidural space realizing a circumferential epiduritis (red arrow). The latter extends until C4, compressing the cervical spinal cord which is the seat of a hypersignal abnormality (blue arrow). Also, note the T2 hyper signal of C2, C3, and C4 vertebral bodies with enhancement after Gadolinium (blue asterisk), which is a sign of osteitis.

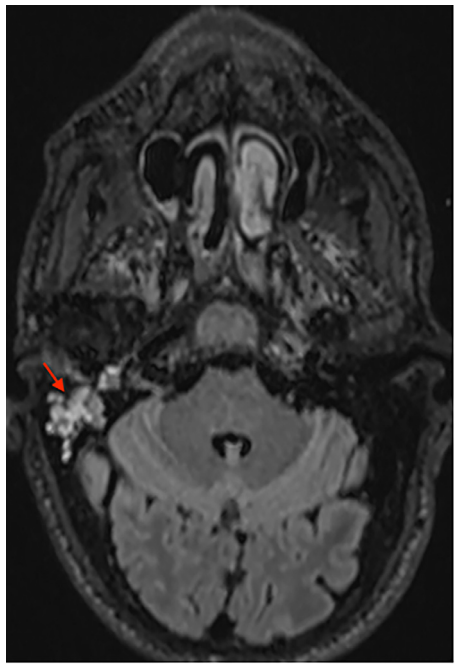

Fig. 5 – Axial FLAIR images showing right otomastoiditis (red arrow).

Discussion

Cervico-occipital junction infectious osteoarthritis can be presented with a triad of pain, neurologic deficit, and fever [4]. This pathology usually gets rapidly complicated by an upper cervical epidural abscess. The latter seems to initially present with neck pain, neck stiffness, and/or fever as shown in the study by Al-Hourani and Al [5]. Some patients may present insidious symptoms such as headaches, sore throat, and odynophagia with a highly variable onset time for symptoms. A full neurologic examination including cranial nerves is necessary. It may highlight a sensorimotor deficit. However, we cannot exclude the diagnosis if our neurologic examination is normal. A nose, throat, and ear examination should not be excluded. It may identify a potential etiology such as suppurative otitis or tonsillitis. A lymph node examination should also be done to highlight any inflammatory lymphadenopathies or swelling. Atlantoaxial dislocation is a dreadful complication of upper cervical osteoarthritis [6]. Respiratory compromise may occur with sometimes other serious sequelae including myelopathy, vertebral artery dissection, and rarely quadriplegia or death if left untreated [7,8]. In our

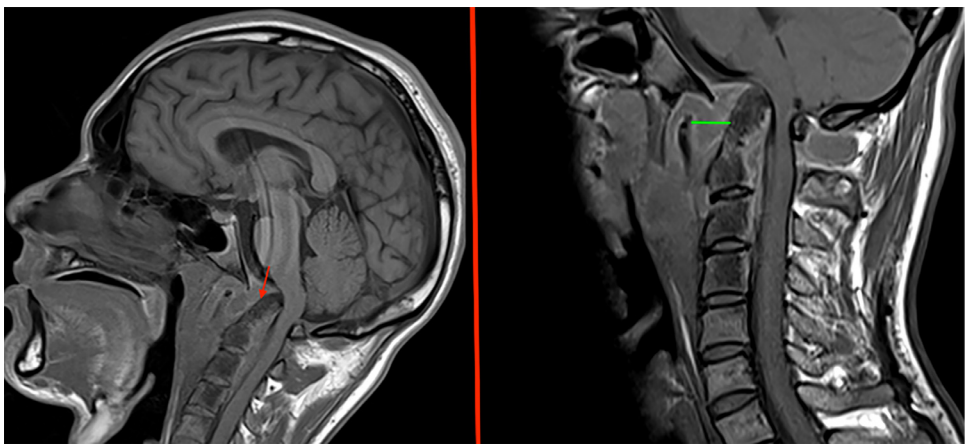

Fig. 4 – Sagittal T1 weighted images showing anteroposterior and superior atlantoaxial dislocation compressing the bulb and the bulbo-medullary junction (red arrow) as well as the enlargement of the C1-C2 joint space measured at 13 mm (green line).

Table 1 – Arguments in favor of atlantooccipital dislocation [10,11].

Widened unilateral atlanto occipital interval (AOI)	*The normal range being 0.5-1.8mm in adults (which is wider in children).*
Condylar sum > 4.2 mm	*Has 100%sensitivity and 69% specificity, and 76% accuracy.*
Midline occiput to C1 spino-laminar line > 4.2 mm	*Which may be artifactually shortened due to extension positioning in collar.*
Further arguments to look for:	
A widened interval basion-den (BDI) > 8.5 mm.	
- an anterior or posterior position of C1 relative to basion,	
An incongruity of occiput-C1 facet joints	*Well seen on sagittal images*

case, the patient presented with fever, torticollis, and retroauricular swelling. The examination found an inflammatory retro auricular lymphadenopathy and tetraparesis which coincides with our review of the literature. Inflammatory markers and white blood cell count should be ordered as part of the evaluation. These markers are not specific but can remain supportive of a diagnosis.

Initial imaging should include plain radiographs to eliminate fractures causing neck pain and also assess for other common causes such as cervical spondylosis. It may also show signs of vertebral osteomyelitis such as vertebral collapse or bony erosions. If osseous changes in the upper cervical spine are noted, we may go for an odontoid view and/or flexion and extension views. ***Computed tomography (CT) scan*** is useful in showing variable bone destruction involving the anterior arch of C1, odontoid, and body of C2. It may identify soft tissue mass adjacent to bone lesions within prevertebral space, variable extension into epidural space, and enhancement of soft tissue component [9]. **Arguments in favor of atlantooccipital dislocation are summed up in** Table 1 [10,11].

MRI remains the modality of choice and has the best accuracy. A sensitivity value of up to 95% and a specificity of over 90% were reported [4]. T1-WI can show a low signal mass centered at C1-C2 with variable involvement of odontoid and lateral masses. It may also show enlarged atlanto dental interval, prevertebral increased soft tissue/edema, or epidural mass with thecal sac/cord compression [12]. With T2 sequences, we can have a diffuse increased signal from vertebral bodies or a soft tissue mass. We may also identify restricted diffusion of abscess components. Gadolinium enhancement can differentiate between an abscess within a prevertebral region or epidural space, and surrounding neurologic structures [13,14]. Since dislocations are a common complication, we should always look for ligamentous injuries (best shown in STIR and T2 weighted images) where tectorial membrane disruptions can be found [15].

In up to 75% of cases, it is possible to identify the causative germ with CT-guided biopsy, which is decisive in the diagnostic pathway [5]. We can also obtain cultures surgically from a direct biopsy of tissue, transoral/retropharyngeal biopsy, or following incision and drainage of the abscess. Ideally, the identification of the causative organism should be done as soon as the diagnosis is confirmed on imaging [16].

Open surgical decompression/stabilization with subsequent long-term IV antibiotics remains the favored option. Variable approaches are possible: anterior debridement and posterior cervical-occipital arthrodesis. Successful medical management is reported with an initial course of IV antibiotics if the patient is neurologically stable followed by prolonged oral antibiotic administration [17,18].

Author contributions

Yahya El Harras was responsible for Manuscript Concept, design as well as editing and literature search. Ola Messaoud and KENZA BERRADA helped in manuscript editing and literature search, Manuscript editing, and Manuscript review. Meryem Fikri and Najwa Ech-Cherif Kettani contributed to the acquisition, analysis, and interpretation. critically revised the manuscript and gave final approval. Mohamed Jiddane critically revised the manuscript and gave final approval. Firdaous Touarsa contributed to acquisition, analysis, and interpretation; critically revised the manuscript and gave final approval.

Patient consent

Written informed consent was obtained from a legally authorized representative for anonymized patient information to be published in this article.

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