

**Erosive cervico-occipital arthritis caused by enthesitis in a patient with axial
spondylarthritis**

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Abstract

The occipito-cervical junction may be affected during the course of spondyloarthritis (SpA). However it is not usual be an inaugural manifestation of SpA. We report herein a case of axial SA in a young patient discovered by O-C1 arthritis.

Case report:

A 29 years-old man with no particular past medical history of trauma, psoriasis, tumor, inflammatory bowel disease, eye redness or inflammatory rheumatic disease in family complained about persistent discomfort of the upper cervical spine for 2 years. Physical examination revealed a limited range of motion in rotation of the cervical spine with no other abnormalities. X-rays of the cervical spine and of the cranio-cervical hinge were unremarkable. MRI showed a low signal intensity on T1-weighted imaging around lateral facets of O-C1 joints and high signal intensity on STIR sequences. Searching for etiology, plain radiography of the pelvis revealed sarco-iliitis grade IV of New York's criteria and the patient was also positive for HLA B27. The diagnosis of axial SpA was retained.

Conclusion :

This is the second case of SpA revealed by O-C1 arthritis. As the craniocervical junction is not well analyzed in X-rays, persistent upper cervical pain should be investigated by MRI. Diagnosis of SpA should be considered and discovering of O-C1 arthritis should lead to performance of a pelvis X-rays.

Introduction:

31 Generally, inflammatory involvement of the upper cervical spine in rheumatoid arthritis (RA) and
32 spondyloarthritis (SpA) consists on atlanto-axial (C1-C2) subluxation or spondylodiscitis. However,
33 involvement of the atlanto-occipital (O-C1) joint has not been described previously not as a complication nor as
34 a circumstance of discovery of SpA. We report herein a case of axial SA in a young patient discovered by O-C1
35 arthritis.

36 **Case report:**

37 The patient was a 29 years-old man with no past medical history of trauma, psoriasis, tumor, inflammatory
38 bowel disease, eye redness or inflammatory rheumatic disease in family. He complained about persistent
39 discomfort of the upper cervical spine for 2 years. There was no Arnold's neuralgia or paresthesia of upper
40 extremities. Three months before consulting, the patient presented an inflammatory back and buttock pain
41 without peripheral arthralgia, unsuccessfully treated with diclofenac (100mg/day). Physical examination
42 revealed a limited range of motion in rotation of the cervical spine. The lumbar spine was not limited with
43 Shober index at 4 cm. Sacro-iliac joints were not painful at pressure or mobilization. Peripheral joints and
44 enthesitis examination was unremarkable. There was no motor or sensory deficit of upper extremities. X-rays of
45 the cervical spine and of the cranio-cervical hinge were unremarkable for O-C1 joint and did not reveal a C1-C2
46 subluxation. MRI showed a low signal intensity on T1-weighted imaging around lateral facets of O-C1 joints
47 and high signal intensity on STIR sequence (fig 1). There was no associated signal abnormalities elsewhere on
48 the spine. Besides, no signs of abscess, tumoral expansion or myelopathy were found. CT-scan of the cervical
49 spine was performed to better analyze bone erosions on O-C1 (fig 2). Plain radiography of the pelvis revealed
50 sacro-iliitis grade IV of New York's criteria (fig3). The C-Reactive Protein rate (CRP) was 5mg/l, the
51 Erythrocyte Sedimentation Rate (ESR) was 15mm the first hour and the Cell Blood Count was unremarkable.
52 The absence of inflammatory biological syndrome was supporting absence of infection. The diagnosis of axial
53 SpA was retained based on ASAS classification satisfying an inflammatory back pain for 3 months and
54 radiographic sacroiliitis on a patient aged less than 45 years-old. The patient was also positive for HLA B27. As
55 his disease was active with an ASDAS-CRP at 3.2 and ASDAS-ESR at 2.7, diclofenac was switched to
56 indomethacin (100 mg/day) associated to gastric protection and a rigid neck collar with good outcomes.
57 Rehabilitation exercises were prescribed. Because of gravity of the localization of the cervical spine
58 involvement we established a tight control of the disease and a pre-biotherapy assessment was prepared.

59 **Discussion:**

To our knowledge, this is the first case of O-C1 arthritis which revealed an axial SpA in a young man. Numerous cases of traumatic O-C1 joint involvement were reported [1] and the issue was often fatal. However, non-traumatic O-C1 joint involvement has been rarely described: infection especially tuberculosis [2], tumor metastasis [3], myeloma [4] or RA [5]. Cases of osteoarthritis were also reported; they occurred more often in elderly patients and can be source of cord compression although the canal is wide in that region [6]. Metabolic cases were not reported since chondrocalcinosis affects mostly C1-C2 joint [7].

Plain radiographies are usually not contributive for the etiological diagnosis of upper cervical spine pain since the O-C1 joint is not well visualized with antero-posterior, lateral and open-mouth incidences. MRI seems to be most performant since it allows a good soft tissue analysis and can detect bone edema [8]. In our case X-rays were unremarkable, but MRI showed arthritis of the O-C1 joint and did dismiss septic and tumoral causes.

According to previously cases in literature, O-C1 arthritis in RA was always associated to a C1-C2 subluxation [9] suggesting a spread of inflammation to the upper joint. In our case of SA, involvement of O-C1 was not associated to a C1-C2 pannus. But it may be explained by an enthesitis of the atlanto-occipital membrane and ligaments.

Conclusion:

This is the first case of SpA revealed by O-C1 arthritis. As the cranio-cervical junction is not well analyzed in X-rays, persistent upper cervical pain should be investigated by MRI. Diagnosis of SpA should be considered and discovering of O-C1 arthritis should lead to performance of a pelvis X-rays.

Ethical standards: informed consent has been obtained from the patient before submission.

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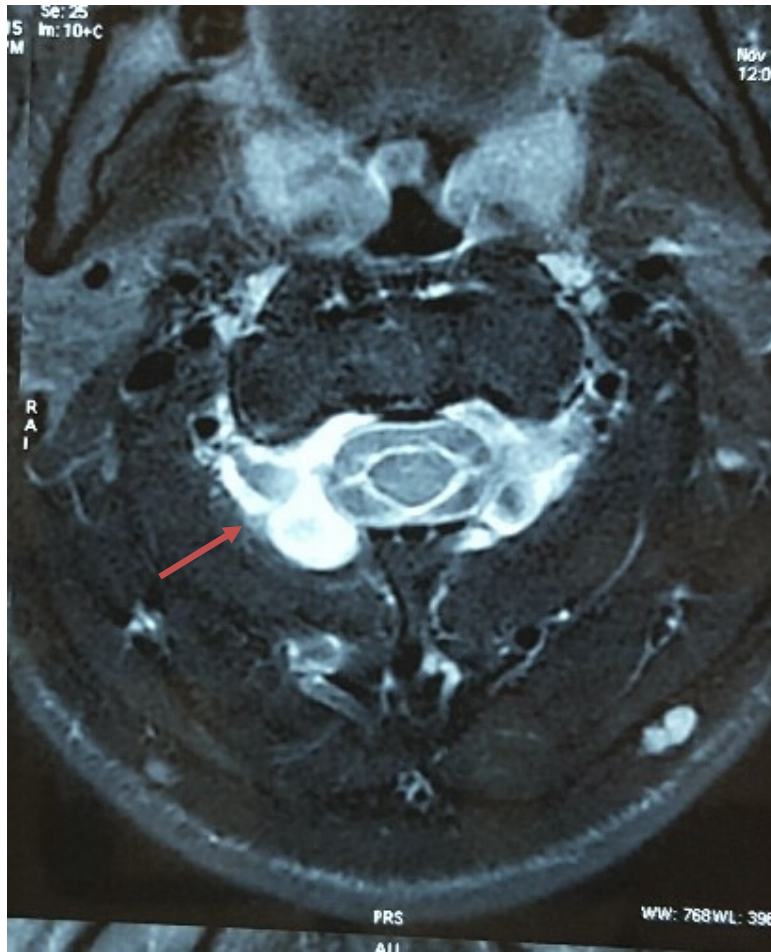
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102 **Figures caption:**

103 **Fig. 1** MRI axial view of the occipito-cervical hinge showing a high intensity signal on T2 sequence



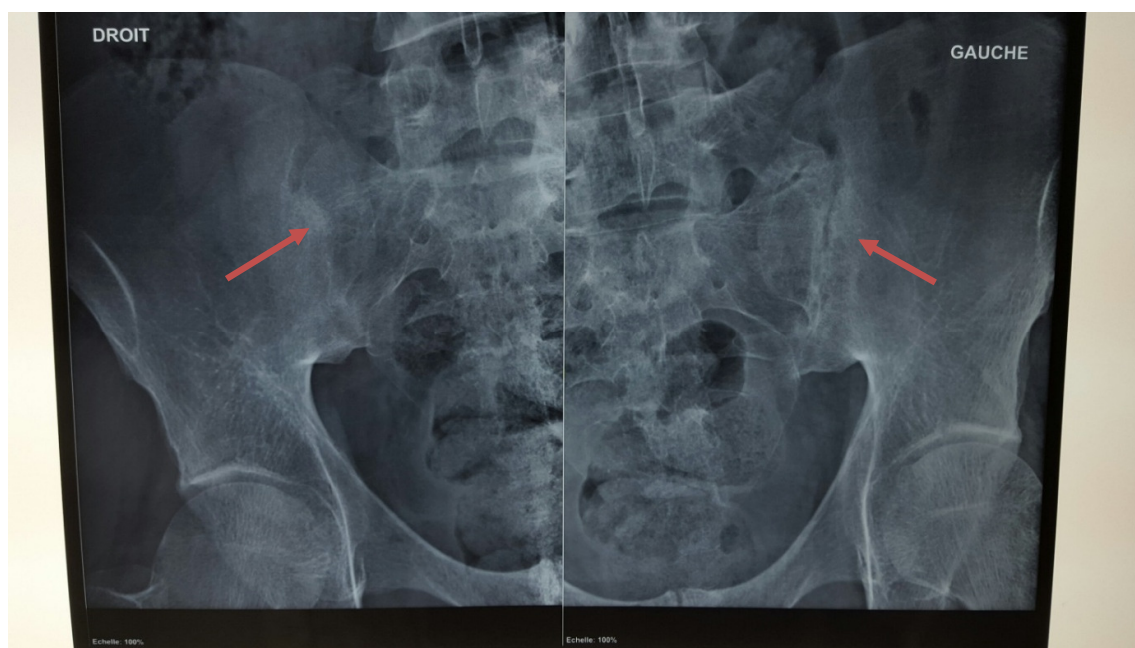
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105 **Fig. 2** Transversal CT view of the cervical spine showing erosions in the C0-C1 joint



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107 **Fig. 3** Plain radiography of the pelvis showing sacroiliitis grade 4 of New York's and Forestier criteria



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