Odontoid process pathologic fracture in spinal tuberculosis

M. Ould-Slimane, T. Lenoir, C. Dauzac, D. Breitel, E. Hoffmann, P. Guigui*, B. Ilharreborde

Orthopedic Surgery Department, Beaujon Hospital, Assistance publique–Hôpitaux de Paris, Paris VII University, 100, boulevard du Général-Leclerc, 92110 Clichy, France

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Summary Craniovertebral junction tuberculosis is a rare lesion in which treatment remains controversial. Options range from conservative treatment to surgery, independently of any associated neurological threat. We here report the first case of pathologic odontoid fracture in a context of spinal tuberculosis, complicated by unusual neurological evolution. The patient presented with non-contiguous multifocal tuberculosis, of which there have previously been only 6 reported cases.

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Introduction

The incidence of spinal lesions in tuberculosis is around 2% [1]. They are usually unifocal, but 6 cases of non-contiguous spinal lesions have previously been reported [2]. Craniovertebral junction locations are especially rare, less than 1%, and are associated with high neurologic risk. No cases of pathologic odontoid fracture have, however, been reported in the literature.

We here report the atypical case of a woman presenting with pathologic odontoid fracture in a context of spinal tuberculosis, causing sudden-onset tetraparesia with multifocal non-contiguous spinal lesions.

Observation

A 37-year-old Cameroonian woman, with no particular previous history, was initially treated in internal medicine as her presenting symptom was a deterioration in general health status of 3 months’ duration, associating fever, night sweat and 5 kg weight-loss. She also complained of neck pain and medial posterior lower neck swelling. Initial imaging found costal, iliac and spinal bone lesions. Biopsy of the anterior arc of the 9th rib found multisensitive Mycobacterium tuberculosis on PCR, confirming the diagnosis of bone tuberculosis. Initial exploration found no extra-osseous (notably, pulmonary) involvement and tuberculosis culture from sputum and gastric washing was negative. Initial neurological examination results were normal, without sign of medulla compression. Spinal MRI revealed craniovertebral, cervicothoracic and thoracolumbar junction loci, apparently without immediate neurological risk (Fig. 1), the patient having been under antitubercular tetratherapy for
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Figure 1 Sagittal section cervical MRI, available at 1st consultation: retropharyngeal abscess and C2 involvement without fracture or displacement. Note: 2nd locus, facing C7 and T1 posterior arcs.

45 days. Orthopedic treatment was decided on, associating continued tetratherapy to a Minerva cervical brace with frontal band and chin support. This rigid restraint was to be worn continuously, and compliance could be ensured as the patient was at the time hospitalized in an internal medicine ward.

Evolution was at first favorable, with resolution of the general deterioration.

Ten days later, however, the neck pains suddenly recurred, despite absence of trauma. The patient was seen in emergency, and clinical examination disclosed a tetrapyramidal syndrome with a general, predominantly left upper-limb motor deficit of 3/5 associated with paresthesia in all 4 limbs and the trunk (Fränkel’s grade C) [3]. Emergency MRI and whole-spine CT scan revealed a pathologic fracture of the odontoid process with frontal displacement (Figs. 2, 3).

Emergency cervical traction by cranial halo-fixation was prescribed, and reduced the displacement and significantly alleviated the neurological disorder (Fränkel’s grade D) (Fig. 4). Surgery, after 1 week’s traction, comprised occipitocervical arthrodesis (C0–C3) without medullary decompression, using a screwed iliac graft. Postoperatively, the neurologic disorder rapidly and almost completely resolved. At last follow-up, residual intermittent paresthesia was observed in both hands, without pyramidal syndrome.

The cervicothoracic junction lesions were not destabilizing, limited to the soft parts and posterior arcs, without significant bone lysis. They thus posed no immediate neurologic risk and were managed by tetratherapy. Given the degree of regional cyphosis (22°) at the thoracolumbar locus (Fig. 5), posterolateral arthrodesis without release was performed in a 2nd step between T10 and L2.

The patient stood in the brace 2 days after the 2nd operation, and walking could be resumed without difficulty. At 1 year, evolution was satisfactory, both clinically

Figure 2 Cervical scan centered on the base of the odontoid process (axial section [a], sagittal section [b]), 10 days after 1st consultation, showing pathologic fracture with severe displacement.

Figure 3 Sagittal section cervical spine MRI, showing medulla compression, 10 days after 1st consultation.
Figure 4  Lateral X-ray view of cervical spine, after 7 days’ 3 kg traction, showing partial reduction of odontoid displacement.

Figure 5  Sagittal section CT scan, 10 days after 1st consultation, showing pathologic narrowing of the thoracolumbar junction.

and on X-ray, with odontoid and thoracolumbar spinal fusion without correction loss, and with stabilization of the cervicothoracic junction lesions (Figs. 6–8).

Discussion

This clinical case has the originality of being the first report of pathologic C2 fracture, complicated by rapidly progressive tetraparesia, in a context of multifocal spinal tuberculosis.

Epidemiology and pathogenesis

Tuberculosis affects nearly 30 millions people, mainly in developing countries, although it has been seen to be spreading again in the West since the 1980s, in parallel to the onset of the HIV pandemic [4]. Spinal lesions affect only 1% of tuberculosis patients and 6% of those with extrapulmonary lesions; only 1% of these spinal lesions involve the upper cervical spine [5].

Spinal tuberculosis of the craniovertebral junction classically begins as a retropharyngeal abscess, evolving into adjacent bone and/or ligament lesions and progressive atlantoaxial dislocation [6].

Figure 6  Wide lateral standing view, at 1 year’s FU.

Figure 7  Sagittal section CT scan of occipitocervical lesion, at 1 year’s FU, showing odontoid consolidation.
Several contiguous vertebrae are usually involved, due to hematogenic spread of the *Bacillus* by the segmentary arteries which each vascularize 2 vertebrae [2]. Only 6 cases of non-contiguous multifocal spinal involvement have been reported, considered by the authors as an aggressive form of the pathology [2]. The present case was multifocal, with 2 non-contiguous loci in the cervical (C1—C2) and thoracolumbar spine (T11, T12 and L1).

**Diagnosis and evolution**

Diagnosis is usually based on an association of clinical signs (general health status deterioration, fever, night sweats, neck pain and dysphagia), a biological inflammatory syndrome, and suggestive imaging. Standard X-ray classically shows prevertebral soft-tissue thickening around the upper cervical spine, with C0—C1 and C1—C2 narrowing. Spinal scan and MRI reveal retropharyngeal abscess and joint and odontoid osteolysis or C1—C2 dislocation [7]. Response to specific treatment also confirms diagnosis.

Scan-guided or surgical biopsy is often necessary to isolate the *Bacillus*, although pathognomonic alcohol-acid resistant bacilli (AARB) are by no means consistently found on direct biological examination [8]. Histology is less specific, revealing caseous necrosis and epithelioid and giant-cell granuloma.

The existence of non-contiguous multifocal forms, however rare, should entail systematic whole-spine MRI or CT scans as part of initial assessment for spinal tuberculosis.

Neurologic disorder, associated with 10 to 46% of cases of Pott’s disease, usually involves the thoracic spine. It may be associated with active tuberculosis or with spinal sequelae. Onset tends to be insidious, following the progression of the abscess and caseous necrosis in the spinal canal [9]. More rarely, it may be associated with progressive upper cervical spine dislocation, although not all such patients show neurologic impairment.

After a few months’ adapted antibiotherapy, an increase in abscess volume and osteolyses is classically observed on X-ray and MRI, despite satisfactory clinical improvement [4]. The present case is rare inasmuch as our patient had been under adapted treatment for 6 weeks and the neurologic impairment set in within a week despite conservative management by brace. The onset of this complication would have been hard to predict. Compliance with immobilization could be ensured by surveillance in the hospital setting, but MRI had presumably underestimated the instability of the initial lesions. It showed the paravertebral and epidural soft-tissue infiltration and the inflammatory bone edema with precision, but it was the CT scan that assessed lysis and hence bone instability [7]. Both types of imaging need to be associated for initial lesion assessment.

**Treatment**

Treatment remains controversial (see Table 1). Advocates of conservative medical management point out that direct peroperative bacteriological examination is of low negative predictive value [8]. Moreover, 30 to 35% of patients presenting associated neurologic disorder show complete spontaneous recovery within 3 to 4 weeks of enriched diet, adapted antibiotherapy and strict dorsal decubitus [9]. Chadha et al. [1] recently reported a series of 13 craniovertebral tuberculosis patients (including 3 with neurologic impairment), managed medically: consolidation was achieved in all cases, without neurologic deteriora-

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<th>Table 1</th>
<th>Literature series reporting craniovertebral spinal tuberculosis.</th>
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<td>Number of patients</td>
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<tr>
<td>Teegala et al., 2008 [12]</td>
<td>71</td>
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<td>Chadha et al., 2007 [1]</td>
<td>13</td>
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<td>Moon et al., 2007 [5]</td>
<td>54</td>
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<td>Gupta et al., 2006 [8]</td>
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<td>Sinha et al., 2003 [13]</td>
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<td>Behari et al., 2003 [14]</td>
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<td>Arunkumar et al., 2002 [15]</td>
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tion, after 3 months’ cervical traction in dorsal decubitus with associated antibiotherapy. The literature now accords no role to corticotherapy in initial management of cervical spine tubercular lesions, even in case of associated neurologic disorder [4,5,8,9].

Certain authors, in contrast, recommend surgery on both diagnostic and therapeutic grounds. It enables not only bacteriological sampling but also medulla decompression and spinal stabilization, thereby optimizing neurologic and functional recovery. According to Anil and Jain, surgery moreover enhances local access for antibiotics, by removing a fibrous barrier [9]. Use of instrumentation in such a context of infection would not seem to be problematic: M. tuberculosis is less adherent to steel than are other bacteria [10]; although this characteristic remains to be confirmed as regards titanium, there have been no reports of local septic relapse following instrumental surgery [11].

Surgery, at least cervical, was more straightforwardly indicated in the present case, given the degree of C2 displacement and of rapidly progressive associated neurologic disorder. Emergency traction succeeded in partially reducing the displacement and neurological problems. Almost complete reduction was subsequently obtained thanks to the cranial halo, enabling occipitocervical arthrodesis without medullary decompression. A preliminary step was envisaged for the thoracolumbar locus, to avoid disassembly or correction loss. However, the tendency of tubercular lesions to resolve spontaneously and the moderate degree of anterior bone loss led us to adopt a long posterior assembly, avoiding anterior approach morbidity. This strategy would seem to have been effective, since imaging at 1 year’s FU found no loss of correction (Figs. 6–8).

Conclusion

We here report the 1st case of pathologic odontoid process fracture in a context of spinal tuberculosis. The lesion was a sign of the severity of the pathology, especially as it occurred in a patient under treatment with non-contiguous multifocal involvement. Initial assessment should include whole-spine CT and MRI scans. Strict clinical and X-ray monitoring is indicated in case of conservative management of spinal tuberculosis of the cervicocranial junction.

Conflict of interest statement

None.

References