

Tuberculous Spondylitis of Craniovertebral Junction of a 70 Years Old Female: Case Report and Review of the Literature

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Abstract

Craniovertebral junction tuberculosis is rare, accounting for 0.3 to 1% of all tuberculous spondylitis cases. MR imaging is the modality of choice to detect bone involvement, abscess formation, pre and paravertebral collection, sub ligamentous spreading of the pus and to differentiate from other lesions affecting the craniovertebral junction. It is also helpful to determine the modality of treatment and monitor the efficacy of treatment. Surgical treatment of patients with craniovertebral junction tuberculosis has been associated with high mortality rate ranging up to 10% and recurrence rate ranging up to 20%, conservative treatment is the modality of choice for most of the cases. We present a case of craniovertebral junction *Mycobacterium tuberculosis* infection diagnosed with clinical, serological and radiological evidence. A Philadelphia collar was applied for external immobilization and anti-tuberculous treatment with rifampicin, isoniazid, pyrazinamide and ethambutol was initiated. After completion of the initiation phase, the patient was neurologically improved. MRI showed marked resolution of the craniovertebral mass. Anti-tuberculous treatment was advised to continue for next 15 months.

Keywords: Tuberculous Spondylitis; Craniovertebral Junction; Anti-TB Therapy.

Introduction

Craniovertebral junction TB (CVJ TB) is a rare disease, accounting for less than 1% of tuberculous spondylitis [2, 3]. Clinical manifestations of CVJ-TB may range from mild neck pain along with nonspecific constitutional symptoms to severe craniovertebral junction instability leading to severe neurological deficit and even death of the patient. The diagnosis and decision to choose treatment modality of CVJ-TB are challenging as there are no well-defined guideline. Patient's neurological status, associated co-morbid conditions gives an arbitrary guideline whether patient will receive conservative management or surgical decompression with or without fixation. Surgical options includes posterior occipito-cervical fusion along with anti-tuberculous

medication or transoral decompression followed by posterior occipito-cervical fusion in the same sitting or two sittings. Sometimes, there is delay in diagnosis as well as treatment which can lead to cervicomedullary compression from atlantoaxial dislocation (AAD). Direct extension of the abscess or granulation tissue and localized angulation can result in significant neurological deficits or even death [4]. We report on a 70-years old woman presenting with CVJ TB without pulmonary involvement. The pathogenesis, diagnosis, and management options are discussed.

Case Report

A 70-years-old normotensive, non-diabetic woman was admitted with progressive weakness of all four limbs for 1 month with evening rise of temperature and weight loss for the same duration. She had no history of tuberculosis or contact with any TB patient. On admission, her vitals were within normal limit. There was no palpable lymph nodes. On neurological examination, power was 3 in both upper limbs and 4 in both lower limbs. Jerks were exaggerated and plantar response was bilaterally

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extensor. There was no definite sensory level. She was catheterized. Neck movements could not be evaluated properly due to pain. Laboratory results including serological tests, tumor markers, and rheumatoid factors were not significant except for hemoglobin: 9 mg/dl, marked elevation of erythrocyte sedimentation rate (ESR) which was 110 mm of hg in 1st hour. Plain x-ray chest showed no evidence of pulmonary lesion. Gadolinium-enhanced magnetic resonance imaging (MRI) study of the cervical spine showed a craniovertebral mass extending from the level of foramen magnum down to C2 with prevertebral and paravertebral collection. There was also presence of epidural abscess with compression of cervicomedullary junction (Fig. 1,2).

Sputum examination showed no AFB. A 10-mm induration was noted in response to an intradermal tuberculin test. Given the risk of aggravating spinal cord compression by extension of the infection, needle biopsy of the craniovertebral mass was considered hazardous. Empirical anti-TB therapy started including isoniazid (INH) 300 mg/day, rifampicin (RFP) 450 mg/day, ethambutol (EMB) 750 mg/day, and pyrazinamide (PZA) 1,000 mg/day, and a hard cervical collar foreexternal immobilization. On followup MRI study 1 month after the initiation of the treatments, the craniovertebral mass had significantly decreased in size, leading to resolution of the spinal cord compression. The same treatment is continued and is scheduled for an 18-months course.

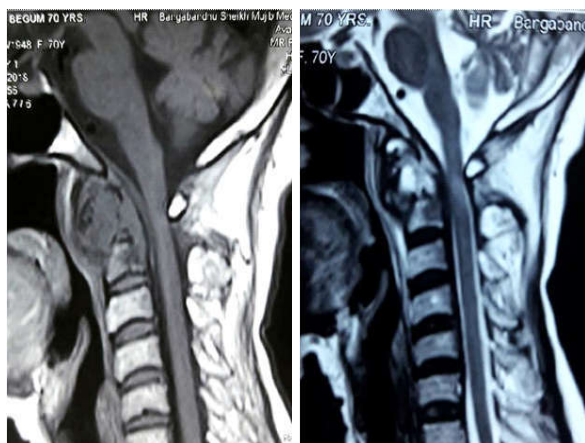


Fig. 1: MRI of the cervical spine with craniovertebral junction, T1WI sagittal section showing iso to hypo intense lesion with destruction of C1 anterior arch and odontoid process of C2 (A), the lesion is heterogeneously hyper intense with formation of epidural abscess- causing compression of the cervicomedullary junction with intrinsic hyper intense signal change (B).

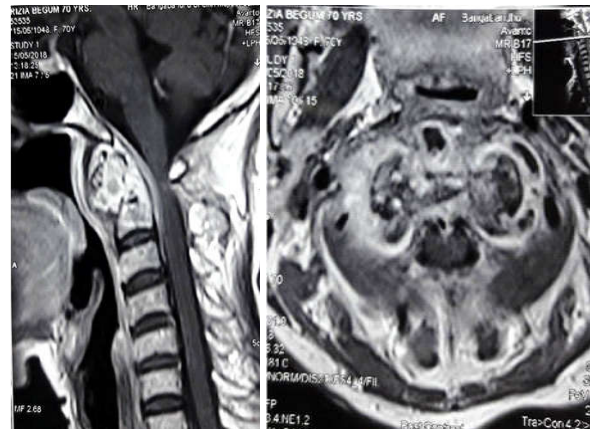


Fig. 2: MRI of cervical spine sagittal section-post contrast sequence showing heterogeneous contrast enhancement with sub ligamentous spread of the pus causing narrowing of oropharyngeal lumen (A), T2WI axial section showing both pre and paravertebral soft tissue collection (B).

Table 1: Stages of Craniovertebral Junction Tuberculosis [4]

Stage	Radiological Criteria
Stage 1	Minimal Bony And Ligamentous Destruction without AAD
Stage 2	Minimal Bony And Ligamentous Destruction with AAD
Stage 3	Extensive Bony And Ligamentous Destruction with AAD

Discussion

CVJ-TB is a rare entity accounting only 0.3 to 1% of tuberculosis of the spine [1]. There is slight female preponderance. CVJ TB may occur as secondary infection from hematogenous spread from pulmonary TB or from direct extension of infection from suppuration of retropharyngeal tubercular lymphadenitis [2,3].

In our reported case, chest X-ray was normal, so retropharyngeal extension of infection may be responsible for the involvement of CVJ.

Dasari et al. reported 16 cases of CVJ tuberculosis which showed diverse clinical manifestations of tuberculous spondylitis of CVJ. Most common manifestations are neck pain and quadriplegia. Other reported features were restricted neck movements, paresthesia, drop attacks and cranial nerve palsies in descending order of frequency [4].

The initial symptoms are usually insidious in onset and nonspecific which is responsible for delay in diagnosis. Most common clinical presentations are neck pain with restriction of neck movement, along with low-grade fever, malaise, weight loss, and loss of appetite [5].

The development of neurological deficits depends on the grade of cervico-medullary compression either from epidural abscess or angulation formed by destroyed bone fragments. Neurological deficits resulted from more than 60% narrowing of the spinal canal diameter [6].

In tubercular spondylitis, it has been stated that, more than 50% of the vertebral destruction brought changes in the plain x-ray [3]. After initial evaluation with plain x-ray, both CT scan and MRI study are ideal diagnostic tools for CVJ TB [7].

The characteristic features of CVJ TB are multiloculated para spinal abscesses with enhancing irregular rim in the presence of vertebral body fragmentation, differentiating it from other lesions at the CVJ, such as rheumatoid arthritis, brucellosis, sarcoidosis, fungal infection, lymphoma, or chordoma [5].

In the present case, MRI of the craniovertebral junction with cervical spine showed multiloculated mass at the CVJ with evidence of destruction of anterior arch of atlas and odontoid tip. There was formation of epidural abscess which was responsible for compression of cervicomedullary junction.

Radiologically craniovertebral junction tuberculosis has been graded by Lifeso et al (Table 1) [4]. According to this grading our present case had features of bone destruction with both prevertebral, paravertebral and epidural abscess formation but there was no evidence of atlanto axial instability, evidenced from X-ray cervical spine lateral view (flexion-extension sequence).

The definitive diagnosis tuberculous spondylitis is isolation of AFB or histological findings consistent with TB pathology. In the craniovertebral junction, specimens are obtained by CT guided FNAC from a retropharyngeal abscess, infected vertebrae, or excisional biopsy from palpable cervical lymph nodes.

In comparison to other spinal levels, microbiological evidence of AFB is less common in CVJ tuberculosis [5], so the diagnosis often made initially by clinico-radiological evidence and sequential follow-up of patient with empirical anti TB therapy with adequate response in follow-up clinical examination, serological results-like hemoglobin level, ESR and state of resolution, documented from MRI scan.

In our reported case, patient denied any invasive procedure, so we gave empirical anti TB therapy after initial evaluation and applied Philadelphia collar to restrict mobility. After one month, follow-up examination and MRI scan showed improved neurological status and state

of resolution of pus. She had been advised to complete 18 months anti TB therapy.

Conclusion

In the elderly people even if there is no radiological evidence of pulmonary lesion, CVJ TB should be considered as the differential diagnosis especially in context to Bangladesh where tuberculosis is one of the most prevalent disease. Both CT scan and MRI study of craniovertebral junction are recommended for evaluation of both bone and soft tissue involvement, also extent of instability which may necessitate for surgical decompression. Considering patient's age, Authors recommend for conservative management if there is no evidence of AAD.

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Conflicts of Interest

There are no conflicts of interest.

Patient Consent

An informed written consent was obtained from the patient.

Ethics Approval

There is no ethical issue in this paper.

Abbreviation

AAD	: Atlantoaxial Dislocation
CT	: Computed Tomography
CVJ	: Craniovertebral Junction
MRI	: Magnetic Resonance Imaging
TB	: Tuberculosis

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