



CASE STUDY

COEXISTENCE OF TUBERCULOSIS AND MYELOMA CAUSING ATLANTO-AXIAL INSTABILITY

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ABSTRACT

Instability of atlantoaxial joint secondary to Tuberculosis or Myeloma is a rare entity. Biopsy along with stabilisation is a described operative procedure. We presented a case of 47 years old male with C1-2 affection treated with occipitocervical stabilisation and biopsy proved as tuberculosis. Antituberculosis treatment was started and clinical improvement was apparent in 5 months. Aggressive progression of diseases after 5 months prompted us to investigate further. Reviewing the biopsy slides with repeat transoral biopsy confirmed features of myeloma which finally responded to chemotherapy. Patient improved significantly (neck disability index = 24%) at the follow-up of 3 years. Hence, coexistence of both tuberculosis and myeloma very rare but should be acknowledged.

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INTRODUCTION

Upper cervical spine tuberculosis (TB) and multiple myeloma (MM) are a rare entity. The coincidental presentation of both MM and spinal TB is reported to be very rare (Yeshurun *et al.*, 2002). Considering that the atlantoaxial joint is extensively mobile, the possibility of instability through the disease process is a common feature (Yeshurun *et al.*, 2002; Goel, 2015). Loss of stability requires surgery and biopsy decides the further management (Valaskatzis, 1996). The distinction between TB and malignancies of spine on the imaging studies can be difficult and often warrants a histopathological confirmation (Gupta, 1996). Further, existence of dual pathology may mislead the correct diagnosis and there is high chance of masking of other diseases behind the primary pathology. In the context of differentiating these two conditions, the importance of carefully planned biopsy not has been over emphasised. In reporting this rare case, we hope to increase the high index for suspicion of tumours in upper cervical spine coexisting with tuberculosis.

Case Report

A 47 year old male presented with complains of severe neck pain and restricted neck movements for 2 months. He had to hold head for walking and could sit with support for half an

hour. He had a Neck Disability Index (NDI) of 94% and VAS Score for axial neck pain was 9 out of 10. Log roll was painful and was associated with disturbed sleep. There was no history suggestive of cranial nerve dysfunction, altered sensations or disturbances of bladder and bowel. On examination, motor power, sensations and reflexes were normal in both the extremities. Laboratory investigations demonstrated haemoglobin of 9.1 gm/ dl, total leucocytes count of 12, 100 per cc and Erythrocyte sedimentation rate of 85 mm/1st hour. Plain radiographs (Fig.1A, B) and CT scan (Fig.1 C, D) of cervical spine showed osteolytic lesion in C2. C1-2 involvement with prevertebral soft tissue mass and no cord signal intensity changes were noted on MRI (Fig. 1 E-H). In view of the suspected instability due to the destruction around the atlantoaxial region a posterior occipitocervical fusion was performed via posterior approach (Fig.1 I, J). Biopsy was taken with a disc forceps via interfacetal route from C2 vertebral body on left side under fluoroscopy guidance. Three samples were taken with evident macroscopically pathological tissue grayish white which were histopathologically confirmed as tuberculosis. Review reporting of slides was seconded by another pathologist to confirm the diagnosis. Patient was started with antituberculosis treatment (standard recommended regime WHO for extrapulmonary TB). Post-operatively patient was started mobilised with philadelphia collar. At follow up at 2 months, he improved symptomatically and was able to perform his routine daily activities. At 5 months, patient was brought in a stretcher as he was not able to walk because of increased neck pain since 3 days.

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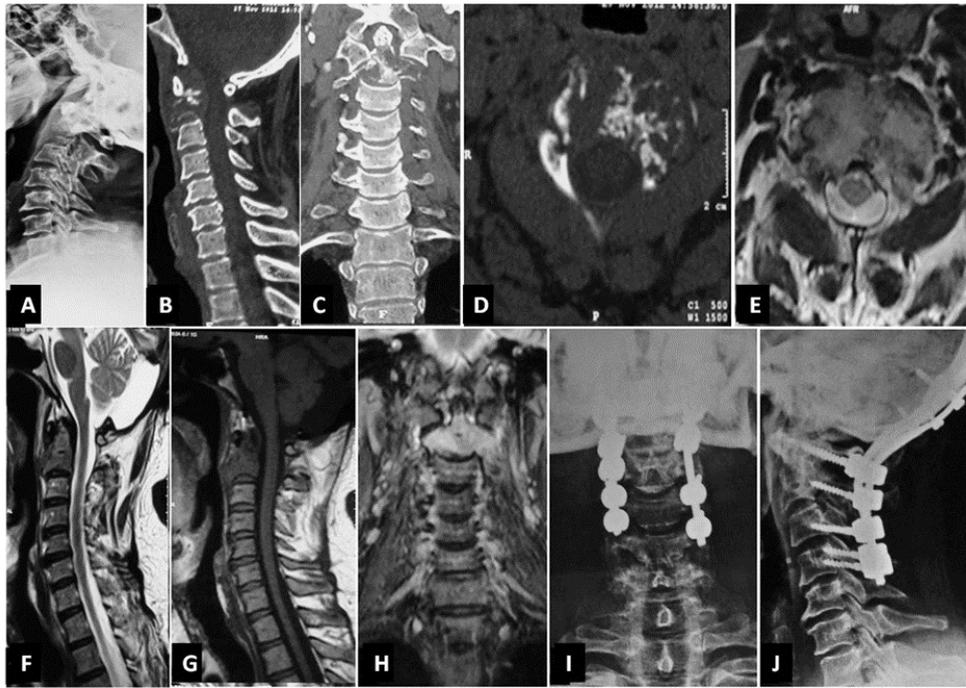


Fig. 1. A Radiograph cervical spine showing osteolytic lesion involving C2 vertebra with destruction of anterior cortex. (B,C,D) left para-sagittal, coronal and axial section of CT scan showing irregular expansile lytic destruction of C2 vertebral body and lateral mass left side. (E,F,G,H) T2 axial, T2 sagittal, T1 sagittal and coronal section of MRI showing of heterogenous lesion affection without cord compression. (I,J) Occipitocervical fixation with skipped C2 screw

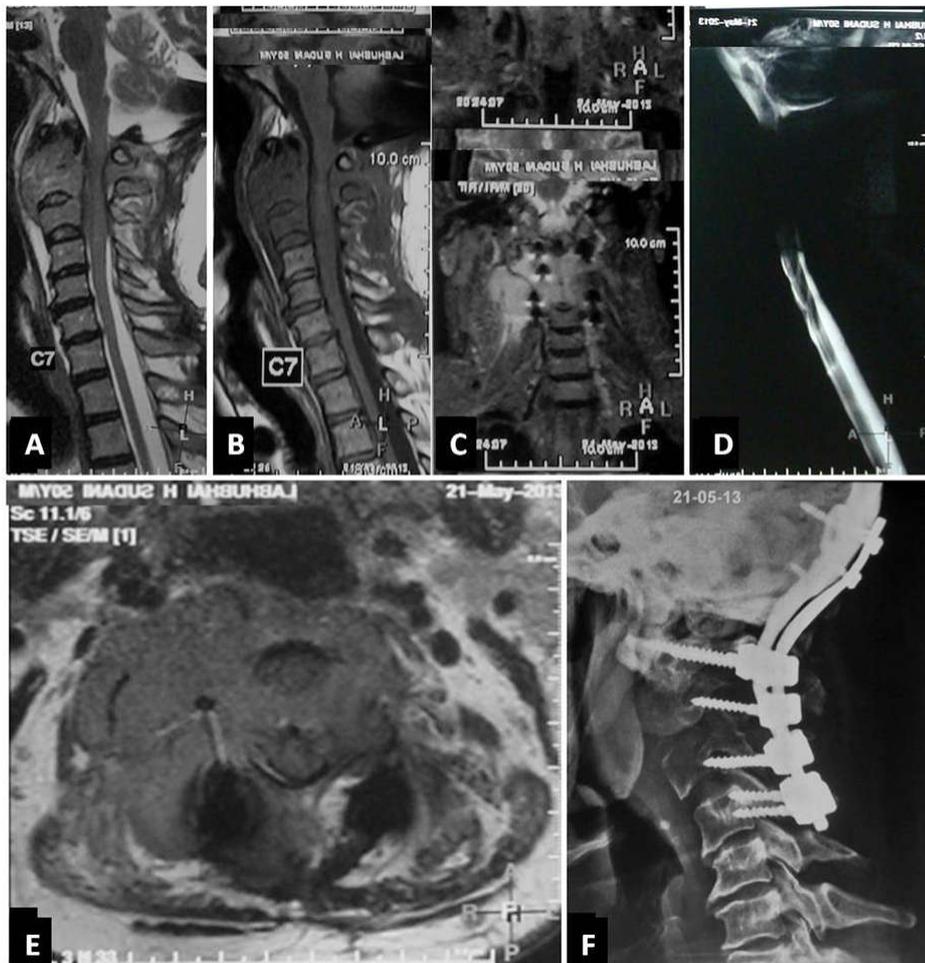


Fig. 2. A,B,C T2, T1 weighted Saggital MRI at 5 months follow up with aggressive lesion anterior and posterior extension, C3 involvement and gross destruction. (D) MR myelogram showing complete block. (E) Axial T2 image showing homogenous lesion with cord compression and with right side more affection, contrasting to preoperative left side affection (F) Lateral Radiograph showing affection with pre-vertebral soft component. In situ fixation with probable loosening at middle segment

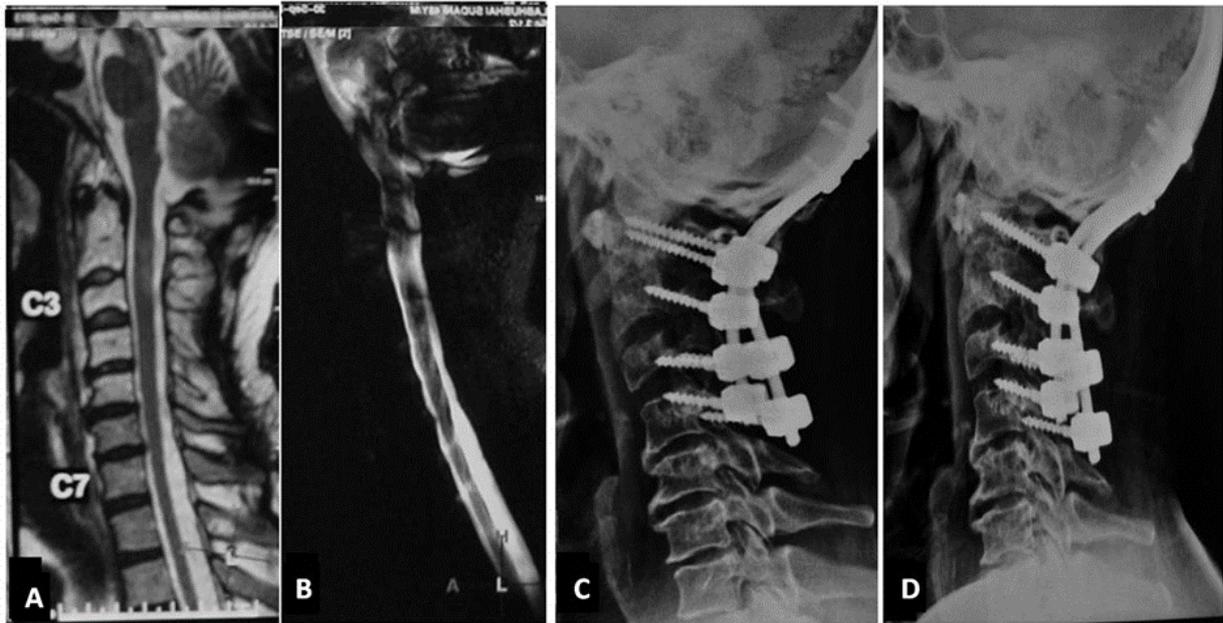


Fig. 3. A T2 saggital MRI image at 8 months with 2 months chemotherapy complete resolution of lesion. (B) MR myelogram showing no compression.(C) 8 months radiograph showing loosening of implant one side.The patient ambulatory with NDI 32%.(D) Radiograph at 3 year with arrested loosening and stable construct with NDI 24%. Normalised soft tissue shadow

Neurology examination revealed within normal limits. Laboratory investigations demonstrated haemoglobin of 10gm/dl, total leucocytes count of 9, 100 per cc and Erythrocyte sedimentation rate of 100 mm/1st hour. Further imaging showed gross destruction of C1-2 and increase in the soft tissue mass on MRI (Fig. 2 A-F). Biopsy slides were reviewed again at two other centres that reported findings consistent with tuberculosis. CT guided transoral biopsy for reconfirmation was done. Biopsy turned out to be myeloma. Further investigations were carried that included skeletal survey radiographs of skull, chest, pelvis and Ultrasound abdomen was normal. Bone marrow biopsy showed 30% malignant plasma cells and serum immunoelectrophoresis was abnormal (IgG kappa monoclonal gammopathy, presence of M band and altered globulin levels - low alpha one, high alpha 2 and high gamma). Patient was referred to oncologist and treated with 6 monthly cycles of chemotherapy with zoledronic acid, dexamethasone and thalidomide. In follow-up, patient recovered gradually and MRI at 2 months post chemotherapy (Fig. 3 A-C). At the end of 6 months of treatment NDI was 56% and VAS was 5/10. Serial radiographs were taken and the end of follow-up was at 3 year (NDI 24% and VAS 3/10) with near normal attainment of bone density of C1-2 (Fig.3 D).

DISCUSSION

The co-existence of these two diseases, MM associated with spinal TB is unusual and the diagnosis is challenging. Both diseases are considered differential diagnosis, as the presentations are similar (George and Sadovsky, 1999). A review of the medical literature revealed paucity of information regarding coincidental presentation of myeloma and spinal tuberculosis (TB) due to the rare occurrence. MM of spine is not rare. However, it is uncommon to involve the high cervical spine (Yeshurun *et al.*, 2002). Solitary myeloma especially in the C1-2 region is rare and as a potential cause of atlanto-axial dislocation, seldom reported in the literature (Goel, 2015). Multiple myeloma is a condition of malignant plasma cell proliferation derived from a single B-cell lineage (George and Sadovsky, 1999). These cells produce monoclonal immune-

globulins, most commonly either immunoglobulin G (IgG) or immunoglobulin A (IgA). Making the diagnosis includes demonstrating these M-proteins in either serum or urine, proving the presence of more than 10% of these malignant plasma cells in the bone marrow and observing the clinical manifestations of the disease (George and Sadovsky, 1999). Cervical and upper thoracic spine is rarely affected, whereas vertebrae of lower thoracic spine are more likely to be involved (Waldestrom, 1970; Schiller, 1993). Spinal TB accounting for about 2% of all TB cases, results mainly from the reactivation of quiescent vertebral foci produced during an earlier infection (Nussbaum *et al.*, 1995). TB of the craniovertebral region is very rare and is reported in less than 1% of all cases of spinal tuberculosis (Tuli, 1974). Between 30-70 % of the patients of the craniovertebral junction have been reported to present with neurological deficit (An *et al.*, 1991; Pandya, 1971). In this case the patient had no neurological deficit but significant affection of C1-2 needing stabilisation. MRI report suggested TB and it was confirmed by open biopsy as well. Immediate outcome after posterior stabilisation was good as seen by improvement in NDI score and general condition. But increased neck pain and failure to improve at 5 months prompted to investigate further and repeat MRI suggested aggressive progression of pathology. The reason for failure of diagnosis has been hypothesised due to either improper biopsy specimen or coexisting TB and MM or MM triggering a latent TB. Qualitatively or Quantitatively inadequate biopsy sample has been reported a reason for failed diagnosis. Open biopsy or definitive surgical biopsy have better yield and positivity in comparison to fluoroscopically guided biopsy and CT guided biopsy (Ghelman, 1998). In this case though an adequate sample has been taken with macroscopically looking abnormal tissue, the sample showed only one lesion. It was taken from the centre of the lesion thus would have yielded active tuberculous tissue, where the infective disease might have lysed the tumour tissue. On recurrence of the lesion, review of the biopsy slides was done by two different pathologists who reconfirmed the TB reporting. The second biopsy at 5 months which was taken transorally, reported the diagnosis of plasmacytoma but did not revealed TB. This would have been

due to the 5 months ATT responsive TB spine. The myeloma part progressed and was seen in the sampling.

There is a paucity of literature on coincidental presentation of both MM and TB. Yeshurun *et al* reported first time, spinal cord compression by dual aetiology, MM and spinal tuberculosis at dorsal spine (Yeshurun *et al.*, 2002). Shyam *et al* observed that tuberculosis was the commonest infectious disease among SLE patients (Berker *et al.*, 2001). Coincidental presentation of osteosclerotic myeloma and spinal tuberculosis with neurological involvement is rare, has been reported by Caroline *et al.* (Omoti *et al.*, 2008). Delayed diagnosis of tuberculous spondylitis masked by concomitant methicillin resistant staphylococcus aureus infection has been reported by Kim *et al.* (2010). It is to be noted that the detailed MRI of the primary lesion was of cervical spine and only T2 sagittal screening of the dorsolumbar spine was done. MM commonly affects dorsolumbar region (Waldestrom, 1970) and only marrow abnormalities are diagnosed in T1 and STIR images. To be diagnosed a MM aggressive lesion in T2 weighted image the lesion should have progressed to the lytic stage. Four patterns of marrow involvement have been identified. A normal marrow appearances present at diagnosis in 50–70% of untreated Durie/Salmon stage 1 and in 20% of untreated Durie/Salmon stage 3 (Herneth *et al.*, 2005). Various patterns of marrow involvement have been identified that include normal, focal, diffuse and a variegated appearance thus creating a lack of specificity. The distinction of TB and malignancies has always been difficult to differentiate (An, *et al.*, 1991). Especially MM comes only as a differential diagnosis in developing nations and with decreasing incidence the clinical expertise and index of suspicion becomes low. Certain drawbacks exist about the case report. Though in these non approachable areas, it becomes difficult to take ideal biopsy sample. An alternative approach may have prevented this happening. A transoral CT guided biopsy which could procure multiple biopsy samples at different angles would have yielded the diagnosis as a primary option. This case emphasized that indications for tissue culture should always be liberal, otherwise, one can never know what may be masked behind the primary pathology.

Conclusion

Both solitary myeloma and TB especially in the region of C1-2 is rare. Imaging characteristics may be a typical and biopsy is the main stay of diagnosis. Proper selection of sample site and adequate quantity is mandatory for prevention of errors in diagnosis and treatment. Review of the slides by second pathologist and repeat biopsy should be done in non improving scenarios.

Disclosure of interest

The authors declare that they have no competing interest.

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