



Case Report

Surgical management of craniocervical junction tuberculosis with atlanto-axial dislocation – A case report and review of the literature

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ABSTRACT

Background: There are few guidelines on how to best manage craniocervical junction (CVJ) tuberculosis (TB). Certainly, timely tissue diagnosis, immobilization of the neck, and decompression of CVJ with appropriate stabilization are the mainstays of treatment for TB at the CVJ.

Case Description: Three patients, ages 16–68, presented with CVJ TB with atlanto-axial dislocation responsible for progressive quadriparesis/plegia. Based on X-rays, magnetic resonance, and computed tomography studies, patients underwent timely decompressions and fusions followed by antitubercular drug treatment.

Conclusion: Early diagnosis, proper decompression with fusion, treated with anti-TB drug for proper period were keys to managing TB involving the craniocervical junction in these three patients.

Keywords: Anti-tubercular therapy, Atlanto-axial dislocation, Craniocervical junction, Kyphotic deformity, Tuberculosis

INTRODUCTION

The incidence of tuberculosis (TB) in the spine is <1%.^[2] Isolated involvement of the craniocervical junction (CVJ) (i.e., including the atlas, axis, and basiocciput) is even less frequently encountered and may involve a 0.3–1% risk of spondylodiscitis.^[4,5] To prevent long-term neurological deficits the timely diagnosis and surgical management (i.e., decompression/fusion) of TB at the CVJ level is critical along with adjunctive anti-tubercular therapy.^[3-7]

CASE 1

History and presentation

A 25-year-old Asian male presented with 2 months of severe neck pain (i.e., focally tender on exam), a progressive bilateral quadriparesis; 3/4 bilateral motor function, bilateral diffuse

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hyperreflexia/Hoffman's Signs, Babinski responses, and a definite C2 sensory level (American Spinal Injury Association, ASIA D). Cervical X-rays showed an osteolytic C1 lesion with instability of the Atlanto-Axial dislocation (AAD). The magnetic resonance imaging (MRI) confirmed CVJ instability while also demonstrating a pre-vertebral collection contributing to marked anterior cord compression [Figure 1]. Lab studies showed an elevated erythrocyte sedimentation rate (ESR) of 110 mm, and C-reactive protein (CRP) of 68. The chest X-ray was normal, there were no palpable lymph nodes, and the tuberculin test was negative.

Surgery

Prone skeletal tong traction corrected the AAD (Atlanto-Axial-Dens) malalignment. At surgery, following the occiput to C2 exposure, the C1 lateral masses were clearly eroded. With minimal distraction, pus immediately extruded from the prevertebral space; it was washed and followed by an occipito-cervical fusion (i.e., including the use of C2/C3 lateral mass screws). He wore a rigid cervical collar for 1 month, followed by a soft collar for 2 months. Within 2 postoperative weeks, he could walk independently, and at 2 postoperative months was neurologically intact.

CASE 2

History and presentation

A 16-year-old female presented with severe neck pain for 1-month, low grade fever, and 2 weeks of progressive spastic quadriparesis (motor 4/5) with sensory loss to pin from C2 to C4 (ASIA D). The cervical X-rays and computed tomography (CT) scan showed an osteolytic C2 lesion (i.e., involving the C2 body and pedicle) with kyphosis and Atlanto-Axial instability. The MRI showed an extradural mass anterior to C1/2 compressing the cord spinal cord; it extended into the retropharyngeal space [Figure 2]. Her ESR was 90 mm in the 1st h and her CRP

was 65. The chest X-ray was normal, and the tuberculin test was negative.

Surgery

Surgery was performed in two stages. The first was transoral decompression; the second was posterior stabilization [Table 1]. Her motor exam markedly improved within 10 postoperative days. Further, the histopathology, postoperative antibiotic TB treatment regimen, use of Pyridoxine, cervical collar use, and entire postoperative course were similar to case 1 [Table 1].

CASE 3

History and presentation

A 68-year-old male presented with severe neck pain for 1 month, respiratory distress, quadriplegia/sphincter dysfunction, a C2 complete sensory level, and bladder-bowel dysfunction (ASIA A). After immediate placement in skeletal (tong) traction, his motor exam improved to the 2/5 level. Cervical X-rays of the CVJ confirmed atlanto-axial instability. The MRI revealed destructive changes involving the C2 and C3 intervertebral discs/vertebral bodies along with an extradural anterior C1–C3 retropharyngeal collection that markedly compressed the cord. The CT confirmed the destruction of dens [Figure 3].

Surgery

After 2 days of skeletal (tong) traction, through a midline posterior occiput-C2 incision, the C1/2 lateral joint space was identified and pus was spilling out from it anteriorly. Granulation tissue was debrided lateral to the C2, and C3 lateral masses. The screw was placed from C1 to C3. Postoperatively, the treatment with a collar for 3 months, the antibiotic regimen, the diagnostic studies, and the postoperative lab studies were all similar than the prior cases [Table 1].

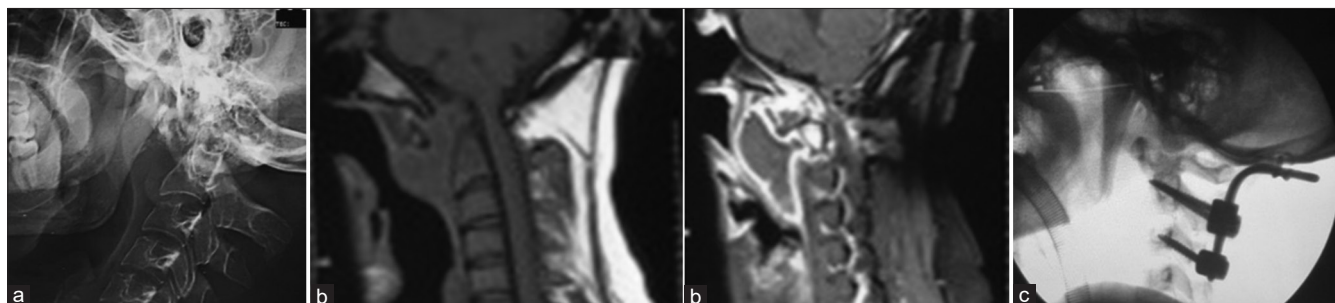


Figure 1: (a) X-ray cervical spine lateral view showing increased atlanto dental interval (13 mm) with radio dense shadow in between. (b) Magnetic resonance imaging cervical spine in sagittal T1-weighted image and contrast showing prevertebral retro-oropharyngeal abscess. (c) Immediate postoperative cervical X-ray days showed beautiful correction.



Figure 2: (a) X-ray cervical spine lateral view showing increased atlanto dental interval (16 mm) with radio dense shadow in between, (b and c) computed tomography (CT) cervical spine lateral (b), coronal (c) view showing osteolytic lesion at C2 body. (d and e) Magnetic resonance imaging (MRI) cervical spine in sagittal T2-weighted image (T2WI) (d) and T1-weighted image (e) showing prevertebral retropharyngeal abscess. (f and g) Postoperative cervical X-ray after 15 days, (h) CT cervical spine sagittal after 1 month, and (i) MRI of the cervical spine in sagittal T2WI after 3 months showed complete resolution of abscess, correction of kyphotic deformity.

Table 1: Craniocervical junction TB (Summary of three cases).

S. No.	Age (years)	Gender	Duration of symptoms	Vertebra involved. (Radiologically)	ASIA impairment scale	Management	Diagnosis	Follow up
1.	25	Female	2 months	O-C2	D	OCF+Anti TB-18 months	Histopath+Gene Xpert	2 years
2.	16	Female	1 month	O-C2	D	AD+C1-C4 fixation+anti TB-18 months	Histopath+Gene Xpert	3 years
3.	68	Male	1 month	O-C3	A	Tongs+C1-C3 fixation+anti TB-18 months	Histopath+Gene Xpert	2 years

O: Occipital bone, OCF: Occipito-cervical fixation, AD: Anterior decompression, ASIA: American Spinal Injury Association. All patients got anti-tubercular therapy for 18 months, TB: Tuberculosis

Histopathology and antibiotic treatment

Histopathology showed granulomatous inflammation suggestive of TB. The GeneXpert (A cartridge based nucleic acid amplification test rapid diagnostic test for TB detection

as well as Rifampicin resistance in direct smear negative cases) was positive for TB. The patient was given 18 months of anti-tubercular therapy; 3 months of 4 drugs followed by 15 months of 2 drugs. He was also given, pyridoxine tablets for 18 months.

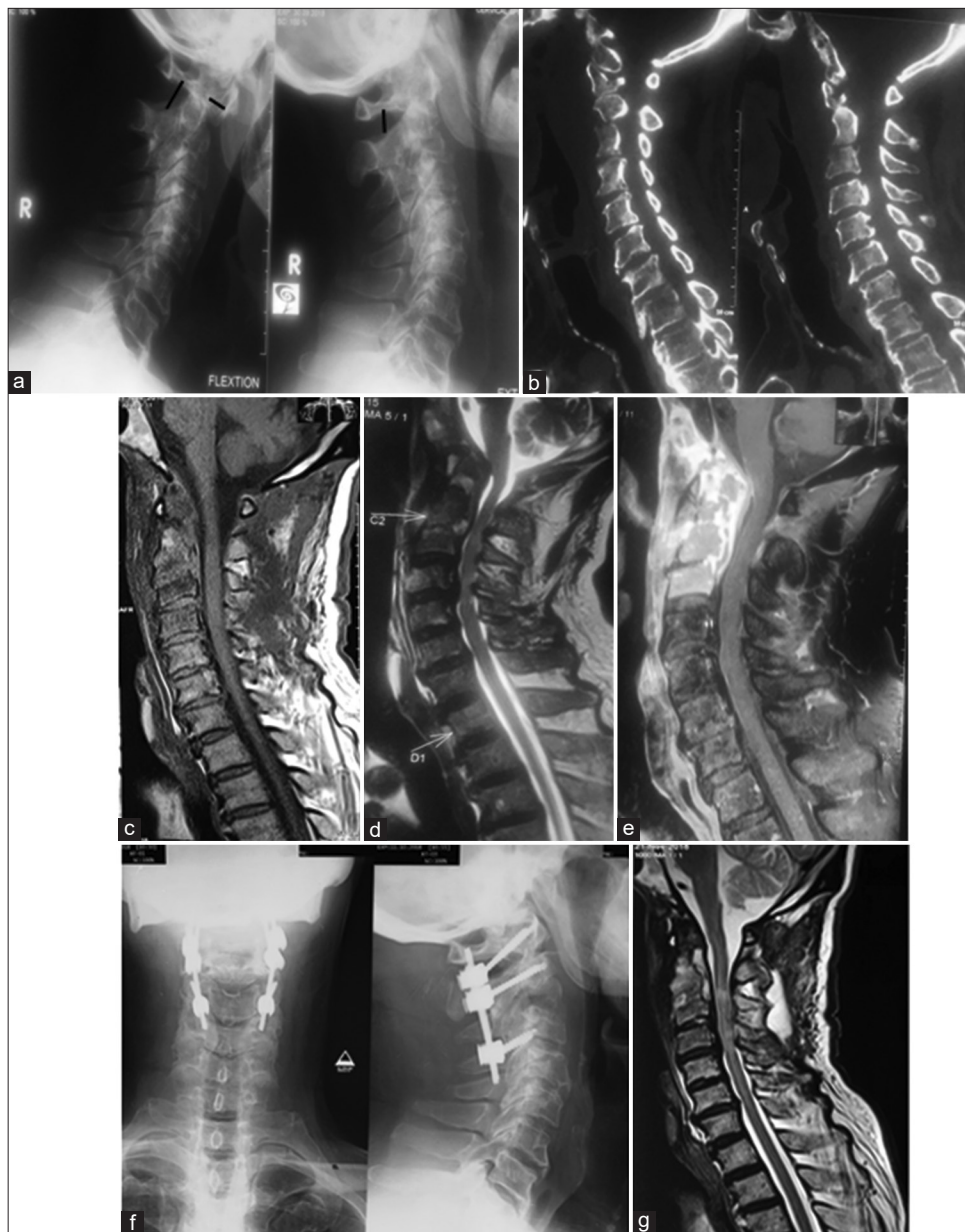


Figure 3: X-ray cervical spine dynamic view showing C1/2 instability, (b) Computed tomography (CT) cervical spine in sagittal view, showing partial erosion of C1 lateral mass and C2 body, (c, d, e) Magnetic resonance imaging (MRI) of the cervical spine, contrast enhancing extradural mass at C1–3 levels causing obliteration of the anterior CSF column and prevertebral collection of abscesses. (f) Postoperative X-ray of the cervical spine at 1 month, showing spinal fixation between C1 lateral mass, C2 pedicle, and C3 lateral mass with screws and rods, (g) MRI of the cervical spine at 6-month complete resolution of abscess and significant decompression of cord.

Neurodiagnostic follow-up

X-rays were done on postoperative days 1, 7, and 15 and every month thereafter looking for progressive infection/deformity, and/or pseudarthrosis [Figure 1]. Complete blood count and CRP were also followed on post-operative day 1, 15 days later, and at 1 month intervals screening for disease progression.

DISCUSSION

Spinal TB rarely involves the cervical spine (i.e., 3–5%) and even more infrequently occurs at the CVJ.^[7,8] When diagnosed with dynamic X-rays, MR, and CT studies, TB typically results in the destruction of vertebra/discitis at multiple contiguous levels. The resultant instability must be immediately stabilized, the patients should undergo emergent spinal cord

Table 2: Reported series of tuberculous CVJ TB.

Author	Year	No of the patient	Management*
Allali <i>et al.</i> ^[2]	2000	2	Minerva jacket+PF (n=1), PF (n=1)
Gupta <i>et al.</i> ^[12]	2006	16	Halo
Behari <i>et al.</i> ^[9]	2003	17	Minerva jacket (n=8), TOD+PF (n=2), PF (n=7)
Arunkumar and Rajshekher ^[5]	2002	6	Transoral drainage+PF (n=2), TOD+PF (n=4)
Edwards <i>et al.</i> ^[11]	2000	1	Transoral drainage
Shukla <i>et al.</i> ^[14]	2005	14	TOD+PF (n=9), PF (n=5)
Sinha <i>et al.</i> ^[15]	2003	18	Anterior transcervical retropharyngeal decompression+PF

*All patients underwent antituberculous medication. TOD: Transoral decompression, PF: Posterior fusion, AF: Anterior fusion. CVJ: Craniovertebral junction, TB: Tuberculosis, n: Number of the patient

as well as paraspinal/retropharyngeal decompression/abscess debridement.^[10,11,13,14] Within some cases CT angiography and/or MR angiography of the cervical spine are warranted to evaluate the course of the vertebral arteries and avoid a vascular catastrophe during surgical decompression/fusion.^[3,4,9,10,12-14] Many authors recommend posterior fusion and fixation with or without anterior decompression, and with or without irreducible AAD [Table 2].^[1,2,4] Here, we presented three cases of CVJ TB who underwent early diagnosis and management with decompression and CVJ stabilization followed by appropriate long-term 18-month postoperative anti-TB therapy.

CONCLUSION

In three patients with TB at the CVJ patients underwent early diagnosis (X-rays, MR, and CT) and surgical decompressions/fusions to prevent further neurological deterioration and offer the potential for neurological recovery.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

- Akhaddar A, Chakir N, El Hassani MY, El Quessar A, Jiddane M, Boukhrissi N. Sub-occipital Pott's disease. Diagnostic imaging in 2 cases. *J Neuroradiol* 2000;27:144-9.
- Allali N, El-Quessar A, Melhaoui A, El-Hassani MR, Chakir N, Jiddane M. Sub occipital Pott's disease: Report of 8 cases. *J Neuroradiol* 2004;31:72-3.
- Allali F, Benomar A, El Yahyaoui M, Chkili T, Hajjaj-Hassouni N. Atlantoaxial tuberculosis: Three cases. *Joint Bone Spine* 2000;67:481-4.
- Arora S, Sabat D, Maini L, Sural S, Kumar V, Gautam VK, *et al.* The results of nonoperative treatment of craniovertebral junction tuberculosis: A review of twenty-six cases. *J Bone Joint Surg Am* 2011;93:540-7.
- Arunkumar MJ, Rajshekhar V. Outcome in neurologically impaired patients with craniovertebral junction tuberculosis: Results of combined anteroposterior surgery. *J Neurosurg* 2002;97:166-71.
- Augier A, Zrig H, Roqueplan F, Brauner M, Dumas JL. MR and CT features of craniocervical junction tuberculosis: A report of 5 cases. *J Radiol* 2008;89:585-9.
- Bapat MR, Lahiri VJ, Harshavardhan NS, Metkar US, Chaudhary KC. Role of transarticular screw fixation in tuberculous atlanto-axial instability. *Eur Spine J* 2007;16:187-97.
- Belanger E, Levi AD. Tuberculosis of the axis in a patient with systemic sarcoidosis: Technique of posterior open biopsy of the dens: Case report. *Neurosurgery* 2000;47:969-72.
- Behari S, Nayak SR, Bhargava V, Banerji D, Chhabra DK, Jain VK. Craniocervical tuberculosis: Protocol of surgical management. *Neurosurgery* 2003;53:1010.
- Dhaon BK, Jaiswal A, Nigam V, Jain V. Atlantoaxial rotatory fixation secondary to tuberculosis of occiput: A case report. *Spine (Phila Pa 1976)* 2003;28:E203-5.
- Edwards RJ, David KM, Crockard HA. Management of tuberculomas of the craniovertebral junction. *Br J Neurosurg* 2000;14:19-22.
- Gupta SK, Mohindra S, Sharma BS, Gupta R, Chhabra R, Mukherjee KK, *et al.* Tuberculosis of the craniovertebral junction: Is surgery necessary? *Neurosurgery* 2006;58:1144-50.
- Mohindra S, Gupta SK, Mohindra S, Gupta R. Unusual presentations of craniovertebral junction tuberculosis: A report

- of 2 cases and literature review. *Surg Neurol* 2006;66:94-9.
14. Shukla D, Mongia S, Devi BI, Chandramouli BA, Das BS. Management of craniovertebral junction tuberculosis. *Surg Neurol* 2005;63:101-6.
15. Sinha S, Singh AK, Gupta V, Singh D, Takayasu M, Yoshida J. Surgical management and outcome of tuberculous

atlantoaxial dislocation: A 15-year experience. *Neurosurgery* 2003;52:331-8.

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