



# Multifocal Tuberculous Spondylitis and Extensive Extrapulmonary Tubercular Osteomyelitis without Immunocompromise: Case Report and Literature Review

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## Abstract

Multifocal extensive spinal and extraspinal tuberculosis is very rare. So far, fewer than 10 cases have been reported. We hereby report two such cases to highlight their rarity, the diagnostic and therapeutic challenges they presented, and the overall prognosis of the condition. The two patients (a 19-year-old woman and another 20-year-old woman) had multifocal extensive tuberculosis involving spine and appendicular skeleton with neurological deficit. Both patients presented with back and neck pain and gradual neurological deficit. The insidious onset and malignancy-like spread pattern mimicked neoplasm. After thorough investigations with magnetic resonance imaging, positron emission tomography scan, and biopsy, antitubercular drug therapy was started, and debridement and fixation were done for significant thoracic and cervical vertebral lesions, respectively. Both patients showed excellent neurological recovery after the procedure. Early surgical treatment of the cases with large abscesses helped provide decompression and stabilization and prevented neurological deterioration and deformity. In patients with noncontiguous spinal tuberculosis, high percentage of surgical treatment may be required due to the aggressive behavior of the disease.

## Keywords

- ▶ multifocal tuberculosis
- ▶ skipped lesions
- ▶ extraspinal tuberculosis
- ▶ tubercular spondylitis
- ▶ skeletal tuberculosis

## Introduction

Multifocal skeletal tuberculosis (TB) involving both the axial and appendicular skeleton is very rare and, so far, fewer than 10 cases have been reported.<sup>1–7</sup> Skeletal TB is an uncommon form of extrapulmonary TB and accounts for 1 to 2% of all cases of TB.<sup>8,9</sup> Spinal TB accounts for 50% of skeletal TB cases. Furthermore, the incidence of multiple-level noncontiguous

spinal TB is 1 to 16% of all the cases of spinal TB.<sup>10</sup> However, extensive involvement of all spinal levels with extraspinal tubercular osteomyelitis is extremely rare,<sup>2,11</sup> especially without immunocompromised conditions and without pulmonary involvement.<sup>1</sup>

We hereby present two such cases and review the literature regarding clinical features and treatment for this atypical form of TB. To the best of our knowledge, there have been

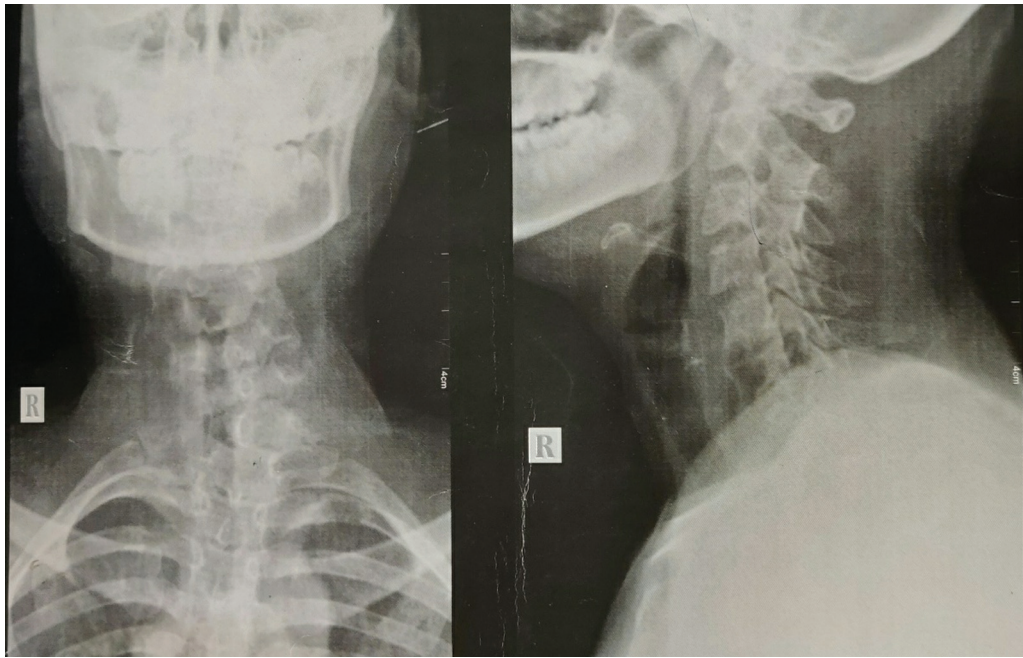
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**Fig. 1** Lytic lesions in the bodies of C6 and C7 vertebrae: anteroposterior and lateral radiographs in patient 1.



**Fig. 2** Multiple lytic lesions in the bodies of D5 and D6 vertebrae in lateral view radiograph in patient 1.

very few cases reported till now involving spinal and extra-spinal TB requiring surgery.<sup>12</sup>

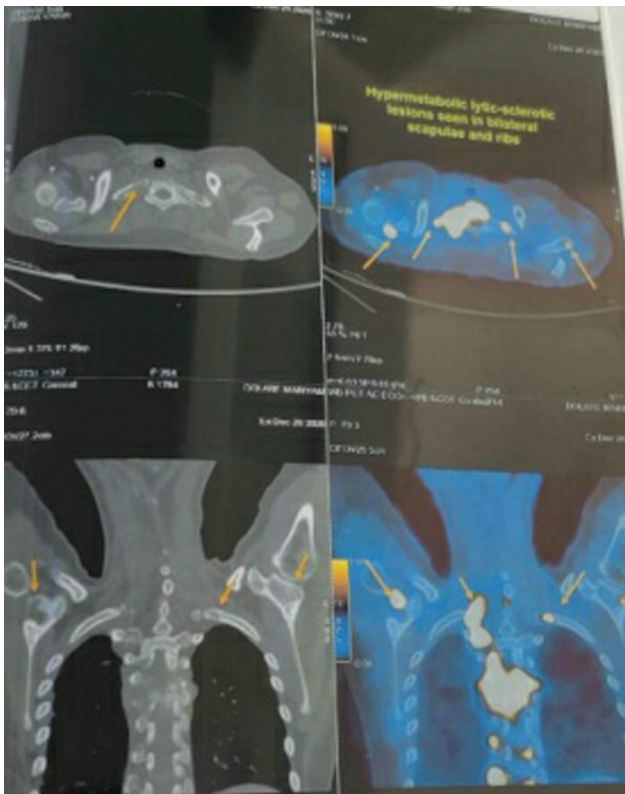
### Case Report

A 19-year-old woman came with complaints of generalized weakness and inability to walk for the last 3 days and chronic

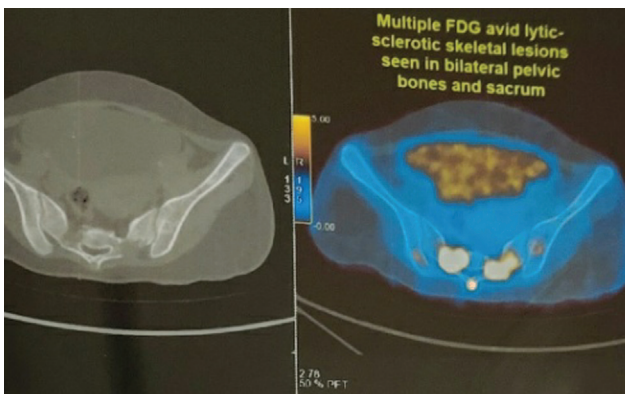
neck and back pain for the past 2 months. She gives no history of trauma but complains of significant weight loss and intermittent fever for the last 8 months. No other symptoms of TB and no exposure in family were found. On examination, there was no visible or palpable deformity over her back, but tenderness could be elicited over spinous processes of middle thoracic vertebrae.

On admission, neurological examination showed decreased power in the right upper limb at the wrist and hand (Medical Research Council Scale Grade 3/5), with normal power in the lower limb and no sensory involvement. Deep tendon reflexes were normal in the upper limb but were exaggerated in the lower limbs. Bowel and bladder were not involved. Laboratory investigations revealed mild anemia with raised erythrocyte sedimentation rate (ESR), with c reactive protein (CRP) positive and human immunodeficiency virus (HIV) enzyme-linked immunosorbent assay (ELISA) test negative.

Chest radiograph was normal. Spine radiographs (anteroposterior and lateral views) showed multiple lytic lesions with wedge collapse of C7 and D5 vertebral body (►Fig. 1 and ►Fig. 2). On whole-spine contrast-enhanced magnetic resonance imaging (MRI), multiple intercommunicating pre- and paravertebral collections from C1 to D2 and D4 to D7 and L3 to L5 were noted with spinal cord compression and cord edema. Significant destruction of bodies of D4, D5, and D6 vertebrae and vertebral bodies and posterior elements of C6, C7, and D1 was noted. Fluorodeoxyglucose-positron emission tomography (FDG-PET) scan was done, which revealed multiple lesions in bilateral iliac bones, left acetabulum and right ischium, multiple bilateral ribs, bilateral scapulae and clavicles, manubrium, body of sternum, and multiple vertebrae (►Fig. 3, ►Fig. 4, ►Fig. 5, and ►Fig. 6). A computed tomography (CT)-guided biopsy



**Fig. 3** FDG-avid lesions in scapulae and ribs in patient 1.



**Fig. 4** FDG-avid lesions in pelvis and sacrum in patient 1.

was also done from the D6 vertebral abscess, which was positive for TB by cartridge-based nucleic acid amplification test. High-resolution CT chest showed no lung parenchymal involvement.

A 20-year-old woman presented with neck pain, stiffness, headache, and right-sided upper and lower limb weakness, without any history of trauma. Symptoms were present for the past 2 weeks. No history of fever or cough was present, but she had loss of appetite and significant loss of weight. She also gave history of right supraclavicular tuberculous lymphadenopathy for which she took anti-Koch's therapy irregularly for 6 months and defaulted 2 months before presenting in the emergency room. On examination, deep tenderness on



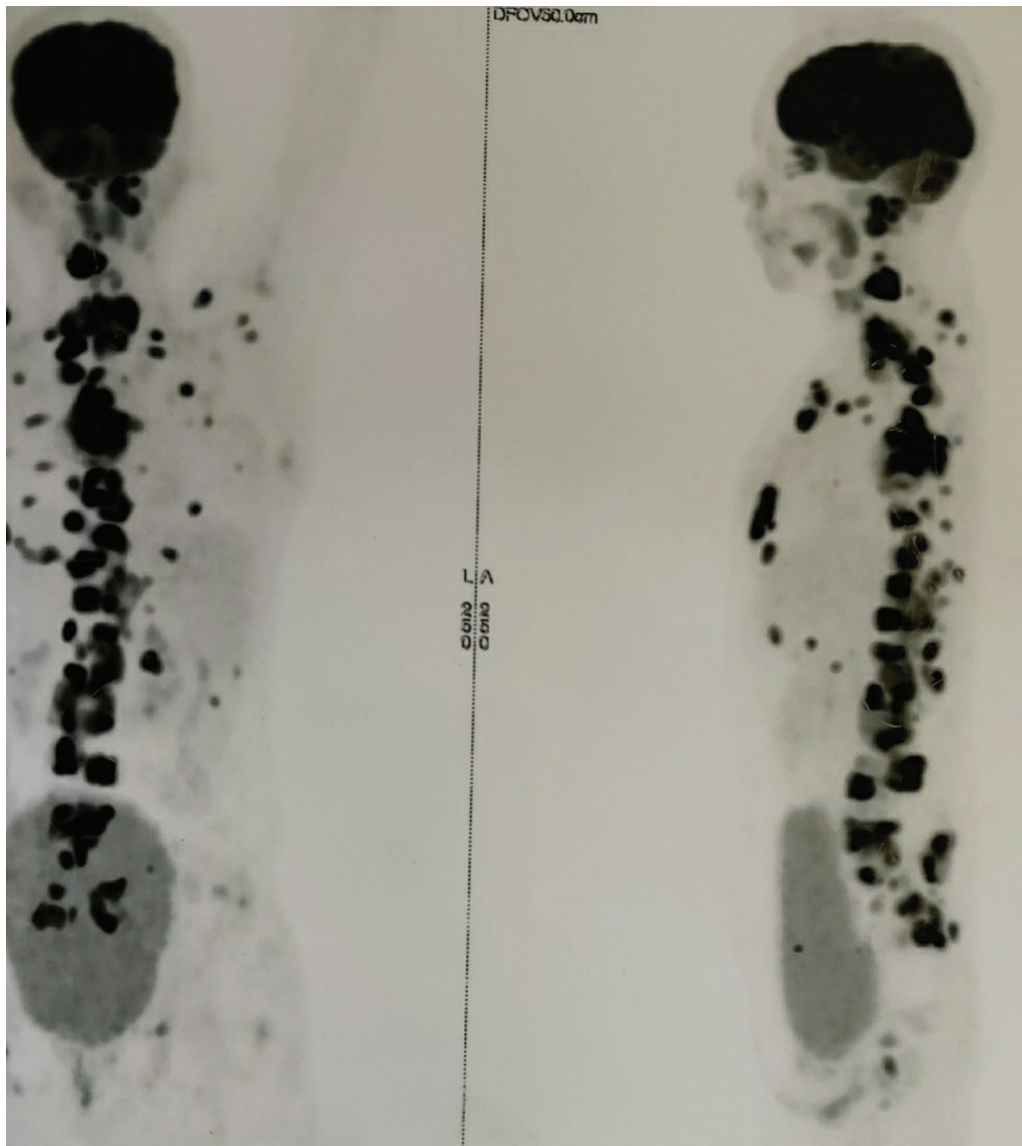
**Fig. 5** Multiple FDG-avid lesions in C7, D5, and D6 in patient 1.

palpation was noted in the midline on posterior aspect of the neck at upper cervical and at cervicodorsal junction level. Neurologically, the patient had flickering movement (Grade 1) in the right upper limb and grade 2 power in the right lower limb. Deep tendon reflexes were brisk in the right upper limb and clonus present bilaterally in the knee and ankle. Plantar reflex was extensor bilaterally and reduced sensation below C6 dermatome on the right side, with no bowel and bladder involvement. HIV ELISA blood test was negative.

Open mouth and lateral view radiographs showed C1 and C2 vertebral lesions (►Fig. 7 and ►Fig. 8). Contrast-enhanced MRI of the neck demonstrated areas of heterogeneously enhancing pre- and paravertebral abscess seen at C1 and C2 vertebral level. Dens of C2 vertebra appeared irregular and were seen displaced posteriorly with its fracture from the body. Increased atlantodental distance with spinal cord compression was seen at the C1–C2 level. C5, C7, D1, D2, and D4 vertebral bodies showed multiple focal areas of altered intensity (►Fig. 9). On PET-CT scan, hypermetabolic consolidations were seen involving the left lung, multiple bilateral lung and pleural nodules, bilateral cervical, supraclavicular, mediastinal, abdominopelvic nodes, and multiple lytic skeletal lesions (►Fig. 10 and ►Fig. 11).

## Management

Considering the risk of spinal cord compression and pathological fracture, which may cause progressive severe neurological symptoms and deformity, early fixation was done. In the first case, after confirmation of TB on biopsy, anti-TB drug therapy was started according to INDEX TB (Indian extrapulmonary TB) guidelines, in which intensive phase



**Fig. 6** Multiple FDG-positive lesions in patient 1.

was started with isoniazid (5 mg/kg/day), pyrazinamide (20 mg/kg/day), rifampicin (10 mg/kg/day), and ethambutol (15 mg/kg/day) according to the patient's weight. The first patient was operated, and debridement and decompression were done by posterior approach at D2, D3, D7, and D8, along with fixation using pedicular screws (►Fig. 12 and ►Fig. 13). The patient showed neurological improvement after 1 week of treatment. Subsequently, falling trend of ESR and CRP was noted. Postoperatively, at 1 month, the patient showed full motor recovery. In the second case, the patient was given antituberculosis drugs, and surgical decompression of the upper cervical spine and stabilization with lateral mass screws and occipital plate were done (►Fig. 14 and ►Fig. 15). The patient's neurology improved, and on postoperative day 7, the patient showed full sensory-motor recovery.

## Discussion

Spinal TB is the most common as well as dangerous form of musculoskeletal TB and accounts for 1% of all TB cases and 50% of all osseous TB. Skipped lesions in vertebrae may be due to HIV, multidrug-resistant TB, and long-standing untreated cases.<sup>2</sup> Such cases must be detected early, as a large number of the affected (40%) sites may be asymptomatic.<sup>13,14</sup> If we underestimate, such asymptomatic lesions progress to cause long-term morbidity.<sup>8</sup>

The infection typically commences at the superior and inferior vertebral end plates anteriorly, then extends by subligamentous spread over multiple vertebral ligaments.<sup>15</sup> In addition, Momjian and George demonstrated that this form of multilevel noncontiguous TB is associated with more frequent extraspinal skeletal involvement.<sup>16</sup> Such extensive



**Fig. 7** Open mouth radiograph of cervical lesions in patient 2.



**Fig. 8** Lateral view of cervical vertebral lesions in patient 2.

involvement can be seen in pyogenic/fungal infections, secondary metastatic disease, and primary tumors of the bone (osteosarcoma, chondrosarcoma, myeloma, eosinophilic granuloma, lymphoma), multiple myeloma, etc.

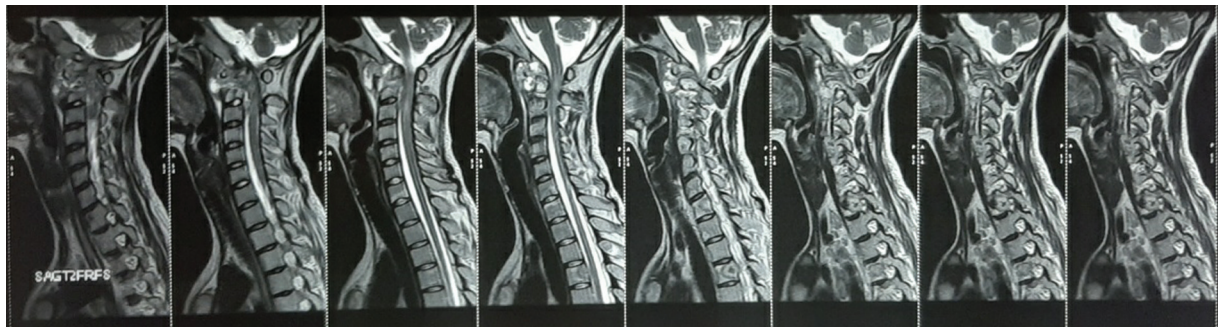
Our patients presented with neurological deficit and vertebral pain. Involvement of other bones was not associated with pathological fractures and was asymptomatic and detected on PET scan. Also, such extensive skeletal involvement is an atypical presentation of TB and led to difficulty in diagnosis.<sup>17</sup>

There are numerous similarities in the imaging and clinical manifestations between noncontiguous vertebral TB, neoplastic diseases, lymphoma, and multiple myelomas.<sup>18</sup> The specific features of lymphoma show paraspinal masses with a vertebral lesion but no extensive cortical bone destruction.<sup>16</sup> In neoplastic involvement of spine, disc spaces are usually spared and paravertebral masses are not usually seen except when solid extraosseous soft-tissue component is associated with destructed vertebral bodies.<sup>19,20</sup>

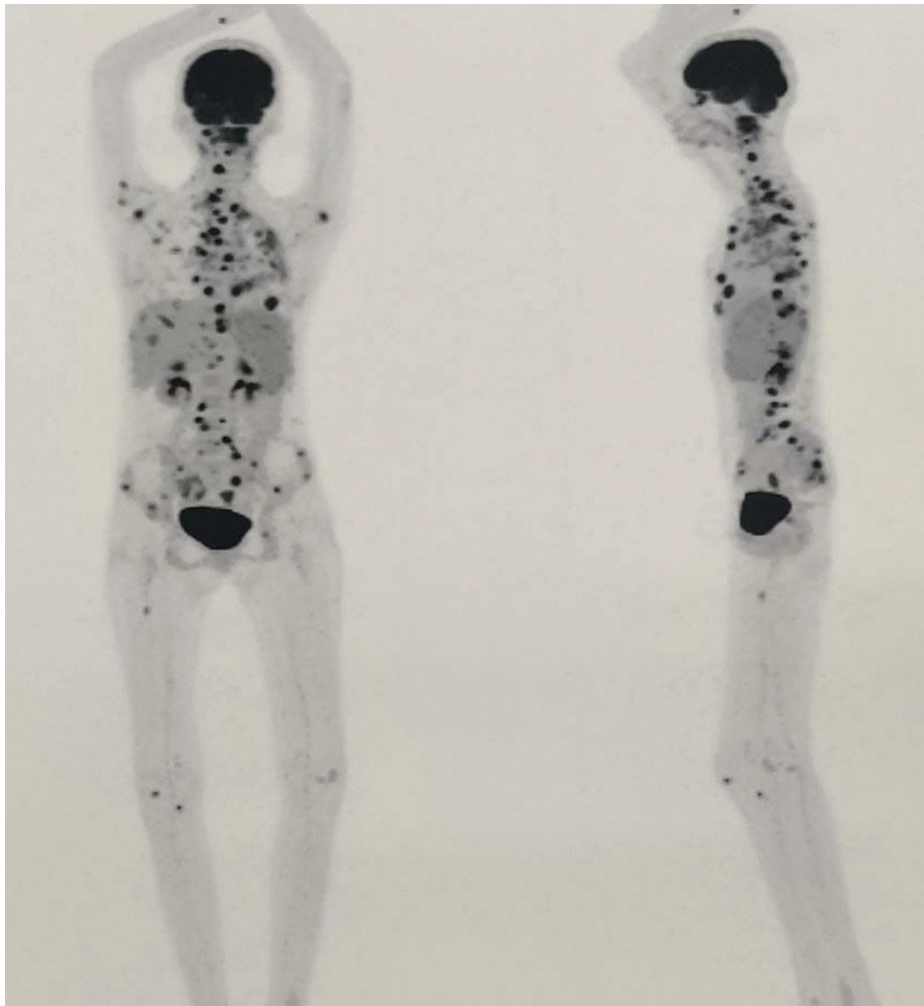
As for metastases, nearly 12% of patients with cancer present with spinal metastases, which often occur in middle-aged and elderly patients and present in lower thoracic

and upper lumbar region. On imaging, spinal metastases involve posterior elements of vertebral bodies, pedicles, and lamina, and the intervertebral discs are spared.<sup>19</sup> The gold standard for diagnosis remains histopathological examination from the biopsy of the lesion.<sup>21</sup>

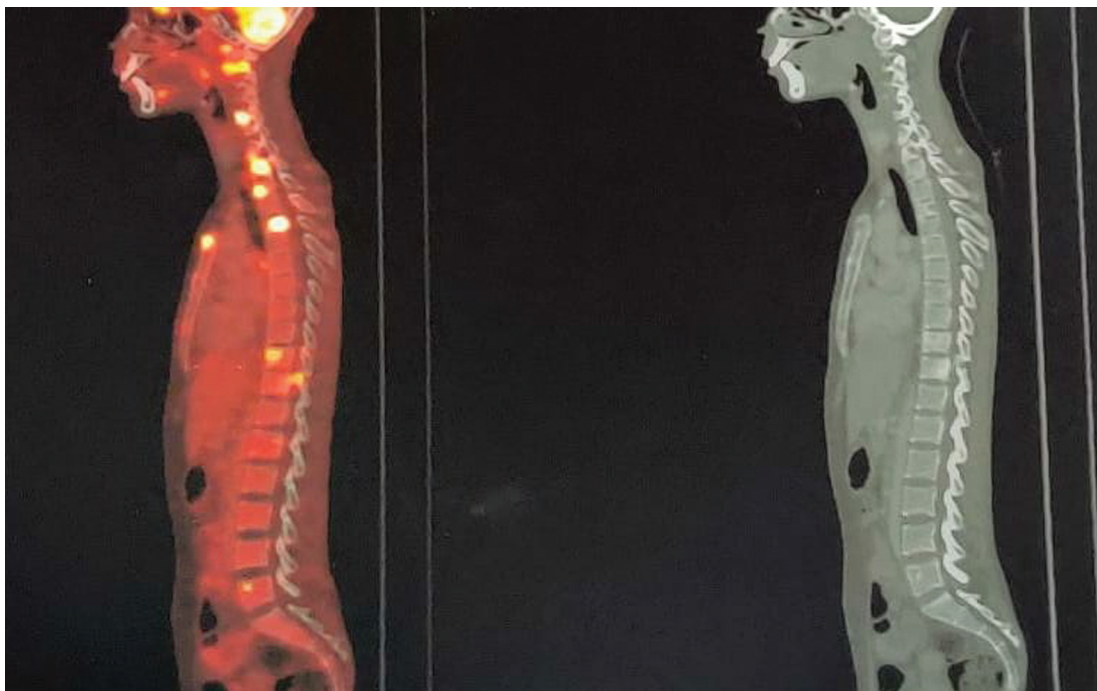
The treatment principles of multifocal extensive TB remain similar to those of typical spinal TB,<sup>13,22,23</sup> but may require surgical treatment more often due to the delay in diagnosis and aggressive behavior of the disease. Indications for surgery include no sign of progressive neurological recovery despite a fair trial of conservative/drug therapy for 4 weeks, development of neurological complications during drug therapy, large prevertebral cervical abscess, difficulty in deglutition and respiration, advanced neurological involvement with flaccid paralysis, severe flexor spasm, and bowel and bladder involvement. Also, prevention and correction of deformity should be carried out early to prevent neurological deficit.<sup>24</sup> The goals of modern surgical management of spinal TB include debridement of diseased vertebrae,



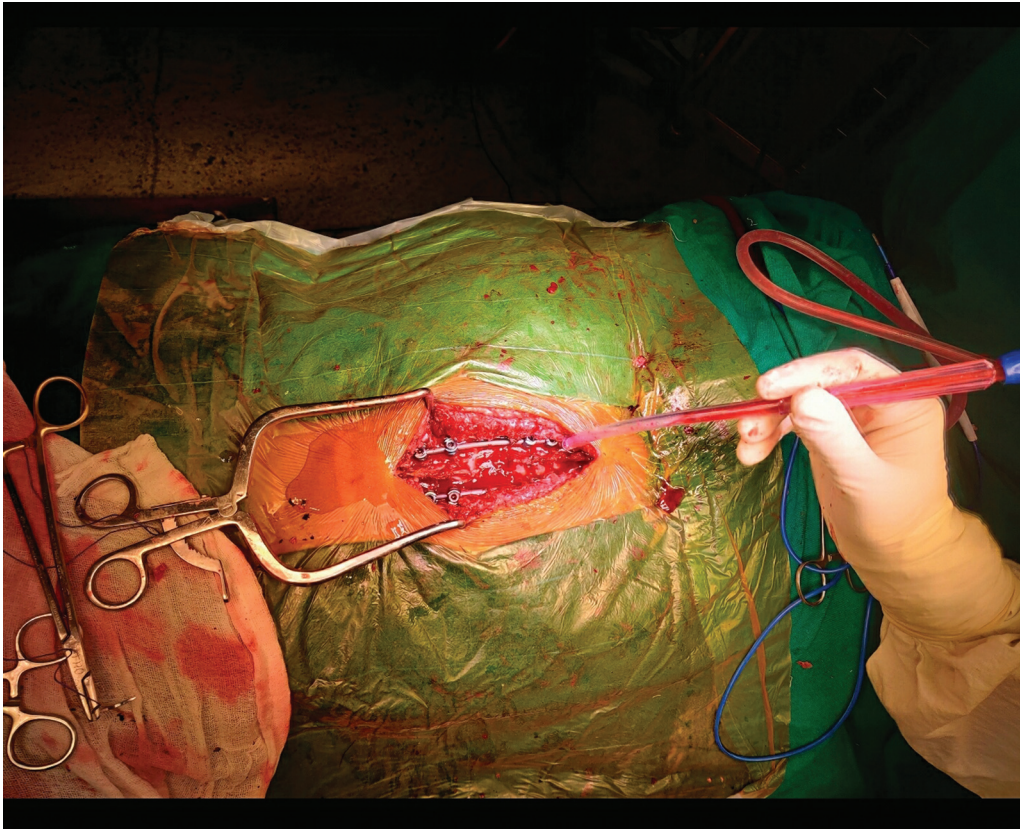
**Fig. 9** MRI showing hyperintense C1–C2 lesion in patient 2.



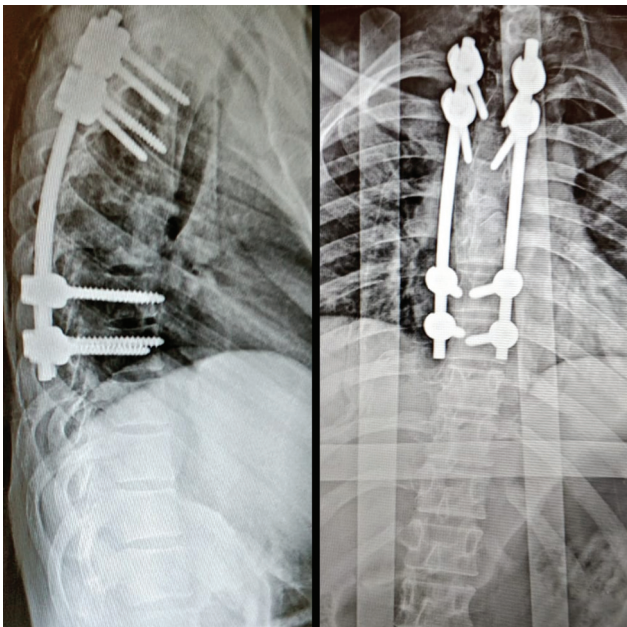
**Fig. 10** Multiple FDG lesions in the body in patient 2.



**Fig. 11** Multiple vertebral lesions in patient 2.



**Fig. 12** Intraoperative picture of pedicular screw fixation in dorsal spine in patient 1.



**Fig. 13** Postoperative radiograph of dorsal posterior spinal fusion (PSF) in patient 1.

decompression of spinal cord, correction of deformities, stabilization of spine, and further protection of spinal cord.<sup>25</sup> Both anterolateral and posterolateral approaches are good for dorsal and dorsolumbar tuberculous spine,

but the posterolateral approach allows a significant correction of kyphotic angle, better improvement of pain, and lesser duration of stay.<sup>26</sup>

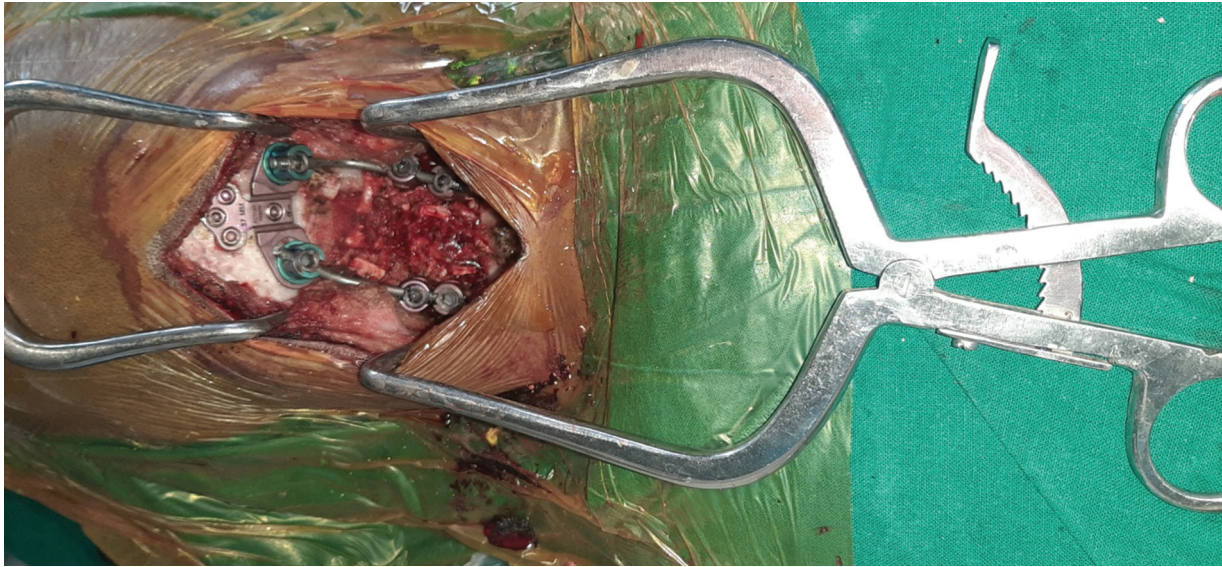
Incidence of deformity could be as high as 25 to 62%.

The treatment of early spinal TB without neurological deficit relies on mainly nonsurgical methods, based on anti-TB drugs,<sup>27</sup> but prolonged bed rest is not advisable since it is associated with a high risk of complications such as osteoporosis and hypostatic pneumonia.<sup>28,29</sup>

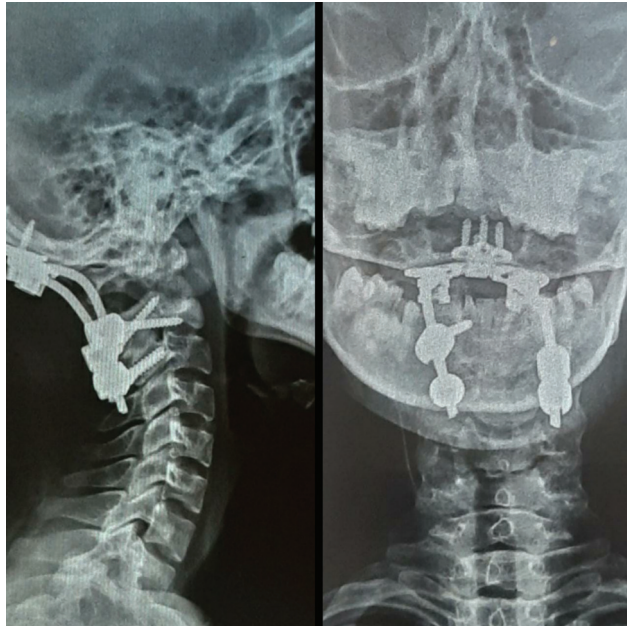
On the other hand, early surgical intervention was suggested for progressive bone destruction, high risk of bone deformity, and unrelieved pain.<sup>30</sup> In all the reported studies, treatment varied between only pharmacological and a combination of chemotherapy and surgery, all reported with a good outcome.<sup>3,22,23</sup>

## Conclusion

Possibility of TB should be considered in cases of multifocal noncontiguous skipped lesions of vertebral column, with or without extraspinal involvement. MRI and PET scan play important role in differentiating between TB and malignancy and other conditions and also in defining the extent of spread of disease in the patient. However, histopathological correlation of biopsy must be done to confirm the diagnosis. Surgical treatment of spinal TB is safe and effective with good clinical and radiological outcomes. However, early



**Fig. 14** Intraoperative picture of occipital plate in patient 2.



**Fig. 15** Postoperative radiograph of fixation of C1 and C2 lesions in patient 2.

administration of chemotherapy for TB is essential in cases of neurological deficit and control and resolution of the disease and was found effective in our patients.

#### Authors' Contributions

D.T. acquired and analyzed the data and drafted the manuscript. G.B. conceptualized and designed the study. S.K. supervised the draft and provided critical revision ideas.

#### Funding

None.

#### Conflicts of Interest

None declared.

#### Informed Consent

Written informed consent was taken from the patients for this article.

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