Case report
Tuberculous Spondylitis involving Sacrum- a rare location and presentation of Pott’s disease: Case report and review of the Literature

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Abstract
Background: Spinal tuberculosis is a common form of extrapulmonary manifestation of tuberculosis. Among them tubercular involvement of the lumbosacral region or isolated involvement of sacrum is very rare presentation.

Aim of the work: The aim of this case presentation is to describe two rare cases of Pott’s disease which clinically mimicked spinal tumor. Case report: A 60-year-old hypertensive, diabetic male patient admitted with low back pain radiating to both lower limbs for 7 months along with low grade fever, generalized weakness and weight loss for the same duration. Clinically, patient had features of flaccid paraparesis, tenderness at the lumbosacral junction, restricted SLR on both sides. There was no features of autonomic dysfunction. MRI of lumbosacral spine showed infective spondylodiscitis at L4 to S1 level with collapse of L5 vertebrae along with paravertebral and epidural abscess. Diagnosis was confirmed by CT guided FNAC from the lesion. Next case was a 23-year-old male patient presented with back pain and walking difficulty for 5 months with some constitutional symptoms. MRI of the dorsal spine with whole spine screening showed multifocal skipped lesion involving whole spine with presence of huge presacral collection. Percutaneous transpedicular aspiration of the purulent material under C-Arm guidance was done and tuberculous infection confirmed by Gene Xpert. Conclusion: Though sacral tuberculosis is rare presentation of Pott’s disease but it should be considered as one of the differential diagnosis in any lytic lesion involving the sacrum in perspective of our country.

Keywords: Tuberculous Spondylitis, Sacrum, Anti-TB therapy.

Abbreviation:
CT : Computed Tomography
FNAC : Fine Needle Aspiration Cytology
MRI : Magnetic Resonance Imaging
SLR : Straight Leg Raising Test
TB : Tuberculosis

Introduction
Spinal tuberculosis is the most common form of extra-pulmonary tuberculosis, accounting for approximately 50% of allcases¹. Dorsal and dorsolumbar junction is the most frequently involved site²-⁶. Tuberculosis involving cranio-

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vertebral junction, cervical spine, lumbosacral region considered to be rare presentation. Among them, isolated form of tuberculosis affecting the sacrum are considered to be extremely rare. The usual presentation is low back pain in adult along with presence of a fistula or abscess, specially in children with or without neurologic deficit. Despite the advancement of imaging modalities, extra-pulmonary tuberculosis is difficult to diagnose due to wide variability of the presentations. In this presentation, we report two cases of sacral tuberculosis which clinically mimicked spinal tumor, later on confirmed by cytological examination and Gene Xpert. Both patients advised to continue anti TB regimen for 18 months and due to marked clinical improvement after initiation phase of anti TB therapy, none of them required operative intervention.

Case 1:
Clinical history and physical examination
A 60-year-old hypertensive diabetic patient presented with sudden onset of back pain with progressive weakness of both lower limbs for 7 months. However, autonomic functions were intact. Patient also complained about evening rise of temperature and weight loss for the same duration. On query, there was no history of trauma, tuberculosis, UTI, prolong use of steroid etc. Physical examination demonstrated presence of spine tenderness at the lumbosacral junction. Neurological examination of the lower limb revealed, power was 4/5 in both sides, jerks were normal, plantar was bilaterally flexor. Pain, touch and temperature sensations were intact in all four limbs.

Laboratory investigations
Complete blood count revealed red blood cells count, hemoglobin level, and white blood cells count were all within a normal range, except high erythrocyte sedimentation rate which was 76 mm in 1st hour. Mountoux test was positive. Induration was 10 mm.

Radiological investigations
Chest X-ray revealed no pulmonary lesion. MRI of the lumbosacral spine with screening of whole spine revealed complete collapse of L5 body with formation of paraspinal abscess with epidural extension resulting spinal canal stenosis. After giving contrast, there was heterogenous contrast enhancement of the lesion. (Figure 1A, 1B, 1C). CT scan of the sacral region revealed destruction of the vertebral body (Figure 1D).

Management
From history, physical examination and radiological investigation, patient started anti TB therapy empirically. After 3 months, there was marked improvement of neurological status. He was discharged to home with the advice to continue 18 months regimen of anti TB therapy. During hospital course, patient received anti TB chemotheraphy, including rifampicin (450mg/day), isoniazid (300mg/day), ethambutol (750mg/day), pyrazinamide (750mg/day). Repeat laboratory test results showing his ESR is reduced to 18 mm in 1st hour, hemoglobin level 12.0 g/dl and significant improvement of systemic TB symptoms within 3 months of starting anti-tubercular therapy.

Case 2:
Clinical history and physical examination
A 23-year-old man presented with insidious onset of back pain for 6 months and walking difficulty for 2 months. For last 1 month, he developed urinary incontinence. Patient also complained about evening rise of temperature and weight loss of about 5 kg for last 6 months. On query, there was no history of trauma to the back, systemic illness, cough, hemoptysis, exposure to known TB patient, including his family members. He was non smoker, non alcoholic and no history of drug abuse. Physical examination demonstrated presence of gibbus at the level of D5 and spine tenderness at the same level. Neurological examination of the lower limb revealed, power was 4/5 in both sides, jerks were exaggerated, plantar was bilaterally extensor. Pain, touch and temperature sensations were diminished below D7, which was more marked on left side.

Laboratory investigations
Complete blood count revealed red blood cells count, hemoglobin level, and white blood cells count were all within a normal range, except high erythrocyte sedimentation rate which was 92 mm in 1st hour. Mountoux test was negative. Aspirated pus was sent for biochemical, cytological, microbiological examination and also for GeneXpert. Pus was milky white in colour, total cell count was 60000 cells/cu. mm, neutrophils: 95%, lymphocytes: 05%, protein: 68 gm/L, sugar: 3 mmol/L. Ziehl-Neelsen Staining showed Acid fast bacilli. GeneXpert for pus examination revealed Mycobacterium tuberculosis.

Radiological investigations
Chest X-ray revealed no pulmonary lesion but presence of abnormal paravertebral soft tissue shadow. Plain x-ray of the dorsolumbar spine revealed collapse of the D5 vertebral body. MRI of the dorsolumbar spine with screening of whole spine revealed complete collapse of D5 body with
formation of epidural abscess causing compression of the spinal cord. At this level, there was a small intramedullary tuberculoma (Figure 2A). There was huge pre-vertebral collection extending from L4-S3 (Figure 2B) with erosion of anterior part of L4-S1 vertebral bodies. After IV contrast, all of the collections showed heterogenous marginal contrast enhancement (Figure 2C).

**Management**

From history, physical examination and radiological investigation, patient started anti TB therapy empirically. Shortly thereafter, percutaneous transpedicular aspiration of pus done from lumbosacral region. About 80 ml of milky white pus was aspirated and sent for biochemical, cytological, microbiological examination and GeneXpert. After 2 months, repeat MRI of lumbosacral spine with screening of whole spine done. It showed marked reduction of the size of pre-sacral abscess (Figure 2D). Due to marked clinical, serological and radiological improvement, patient was discharged to home with the advice to continue 18 months regimen of anti TB therapy, mobilization with Taylor brace and regular follow-up with Plain x-ray of dorsolumbar spine, MRI of the dorsolumbar spine with whole spine screening to detect aggravation of kyphotic angle and state of cord compression. During hospital course, patient received anti TB chemotherapy, including rifampicin (450 mg/day), isoniazid (300 mg/day), pyrazinamide (750 mg/day) and Inj. Streptomycin (1 gm)-IM daily. Repeat laboratory test results showing his ESR is reduced to 21 mm in 1st hour, hemoglobin level 12.2 g/dl and significant improvement of systemic TB symptoms within 2 months of starting anti-tubercular therapy.

**Discussion**

Tuberculosis is one of the major health issues in the developing countries like Bangladesh. Through hematogenous route spine can be involved secondarily from pulmonary or extra-pulmonary sites. The condition may be seen even in the absence of a documented pulmonary infection. At first, infection begins in subchondral bone and spreads slowly to the adjacent vertebral body through subligamentous spread of pus. Involvement of two adjacent vertebral bodies deprive the nutritional support of the intervertebral disc which ultimately leads to destruction of the disc. The dorsolumbar region is the frequent site of involvement. Involvement of the lumbosacral junction by tuberculosis is quite rare and occurs in only 1 to 2% of all cases of spinal tuberculosis. In comparison, isolated sacrococcygeal or coccygeal tuberculosis is much rarer. The most common presenting symptoms are non-specific low back pain and swelling with or without neurological deficit. Ocasially, patient may present with only features of non specific constitutional symptom like low grade fever, weakness and weight loss. So, the diagnosis can be delayed because of this non-specific character of the clinical features that mimics numerous other disease entities like prolapsed lumbar intervertebral disc, spinal tumor in particular malignancy. The time interval between onset of symptoms so that of diagnosis was 6.5 \pm 2.5 months with a range from 3 to 12 months in the previously reported literature. Isolated sacral tuberculosis is extremely rare but it should be considered as one of the differential diagnosis in the presence of atypical clinical and radiological features of a sacral lesion particularly in developing countries. In the presence of a presacral mass along with lytic lesion involving the sacrum, sacral tuberculosis mimics neoplasias like chordoma and osteoclastoma. So, it is recommended by the Authors that CT guided FNAC should be considered in all cases. Percutaneous aspiration can be done in selected cases to establish the diagnosis before planning for operative procedure. In our reported series, percutaneous aspiration done in the 2nd case due to presence of huge pre sacral abscess after which diagnosis was confirmed by GeneXpert. 1st case underwent CT guided FNAC which gives the definitive diagnosis. However, sometimes there is diagnostic dilemma even after doing all relevant investigation. In that case, sometimes empirical anti TB started and the response monitored in clinical, biochemical and serological parameters.

Conservative treatment involves a combination of four drugs: rifampicin, isoniazide, pyrazinamide and ethambutol in the first three months (initiation phase), followed by rifampicin and isoniazide for next 15 months (continuation phase). Regarding treatment there are different opinions in the literature. Many clinician advised anti TB chemotherapy for 6 months while some continue it for 9-18 months. In our reported series, total duration of antituberculous therapy was planned for 18 months in both patients with regular follow-up. Surgical management can be avoided if there is satisfactory response after institution of chemotherapy. Surgery is necessary if there is nerve root compression during extensive destruction of several vertebral bodies with spinal deformity or to evacuate an abscess that is resistant to medical treatment. The prognosis of sacral tuberculosis is favourable, if diagnosis can be made at the initial
stage of the disease. This pathology should always be suspected in any lytic lesion involving sacrum or coccyx, especially in endemic areas of tuberculosis which helps to reduce the morbidity of this disease.

**Conclusion**

In context to our country, even if there is no radiological evidence of pulmonary TB, sacral TB should be considered as the differential diagnosis for any lytic lesion involving the sacrum-specially if it is associated with paraspinal and epidural collection, which strongly suggest tuberculosis. Both CTscan and MRI study of lumbosacral spine are recommended for the assessment of bone and soft tissue involvement. Authors recommend to do a CT guided FNAC before starting empirical anti TB therapy which helps in definitive diagnosis when features mimics spinal tumor specially in elderly population.

**Declarations:**

**Authors’ contributions:**
Conception, diagnosis and design, Radiological diagnosis and Final approval of manuscript: Dr. Shahidul Islam Khan, Prof. Dr. Rafiquul Islam, Dr. Md. Kamrul Ahsan.
Manuscript preparation, Technical revision, Manuscript editing and revision: Dr. Asifur Rahman, Dr. Nazmin Ahmed
Literature search: Dr. Nazmin Ahmed, Dr. Abdullah Al Mahmud

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**Ethics approval:** There is no ethical issue in this paper.

**Legends of Figures**

Figure 1A: MRI of the lumbosacral spine T2WI sagittal section showing collapse of D5 vertebra with infective spondylodiscitis at the level of L4, L5, S1. 2A showing scoliosis at the lumbosacral region with paravertebral collection. 2C showing heterogenous contrast enhancement of the lesion.
Figure 1d: CT scan of the sacral region axial section showing destruction of S1 vertebral body.

Figure 2a: MRI of the dorsolumbar spine with contrast showing collapse of D5 vertebrae with prevertebral and epidural abscess formation. 2b showing huge prevertebral abscess at the lumbosacral region. 2c showing irregular marginal contrast enhancement. 2d: After percutaneous aspiration, there was marked reduction of prevertebral abscess.
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References: