

Atypical sacral tuberculosis: A case report and review of treatment.

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Abstract: Extrapulmonary tuberculosis commonly involves thoracic and lumbar spine. Sacral tuberculosis is rarely reported so far. We are presenting a case of young male student with tuberculosis of sacrum. The clinical and radiological features suggested a sacral tumor instead of tuberculosis. The lesion could not be diagnosed until histopathological examination was done. The duration of antituberculous treatment in skeletal tuberculosis is controversial, ranging from six months to three years. The recent trends of treatment are briefly discussed with specific reference to our case.

Key words: Backache in young individuals, DOTS in skeletal tuberculosis, MRI in spinal tuberculosis, Sacral tuberculosis, tuberculosis mimicking spinal malignancy.

I. Introduction

Tuberculosis of the spine constitutes 50% of skeletal tuberculosis. Thoracic and lumbar spine is commonly affected, followed by cervical spine. Skip lesions are also found in 7% of spinal tuberculosis[1]. But isolated sacral involvement is rare.

MRI is the most sensitive modality used for early detection of tuberculosis of spine. MRI typically shows destruction of intervertebral disc and adjacent vertebral end plates and abscess formation, which can differentiate tuberculosis from neoplasm. But, in our case, MRI findings suggested benign or malignant tumor. Tuberculosis is diagnosed only after histopathological examination of the biopsy specimen.

The duration of antitubercular treatment (ATT) in osteoarticular tuberculosis is controversial[2]. Though DOTS is actively implemented in extraskelatal tuberculosis, it is not tested in skeletal tuberculosis. Daily therapy with ATT for one to one and half year is the preferred treatment for osseous tuberculosis. The efficacy of DOTS in treating spinal TB is shown in recent studies[3,4].

We are presenting a case of tuberculosis of sacrum in a 16 year old male. This case is presented not because of its rarity, but for its resemblance to a tumor clinically and radiologically and to show its successful management with six months of DOTS with intermittent regimen.

II. Case Report

A 16 year old male presented with low backache of six months duration. Backache started insidiously and increased gradually in intensity. It was continuously present throughout the day and was unrelated to his activities. There was no radiating pain. There was no history of tingling sensation or numbness in lower limbs. His walking was not affected and he had no bowel or bladder disturbances.

Patient had no history of fever or weight loss. He didnot receive BCG vaccination in his childhood. Patient had no obvious contact with any person suffering from tuberculosis.

On examination, an ill-defined 4*4 cm firm swelling was present over lumbosacral spine in midline. Sacral spine was tender to touch. There was no abscess or sinus anywhere. Patient had no neurological involvement in lower limbs.

Haematological investigations are within normal limits, with Hb of 13.6 gm/dl and WBC count of 6300cells/mm³. Peripheral smear showed 60% of neutrophils, 32% of lymphocytes, and 2% of monocytes. ESR was 50mm in first hour, showing a slight rise.

Radiograph of Lumbosacral spine showed radiolucent mass in first and second sacral vertebrae.



Figure 1: Initial radiographs showing lytic lesion in S₁ and upper half of S₂

MRI showed minimally hyperintense lesion in T2W images in vertebral bodies. The lesion extended upto both sacroiliac joints. Posterior elements were also involved. The mass was extending into the sacral canal and sacral foraminae, compressing the nerve roots. S₁-S₂ intervertebral disc was not affected and there was no obvious fluid collection. Subligamentous extension of soft tissue for a short distance was present anteriorly and posteriorly. Based on MRI findings, the lesion was provisionally diagnosed as sacral tumor, with a differential diagnosis of osteoblastoma, small round cell tumor and metastasis.

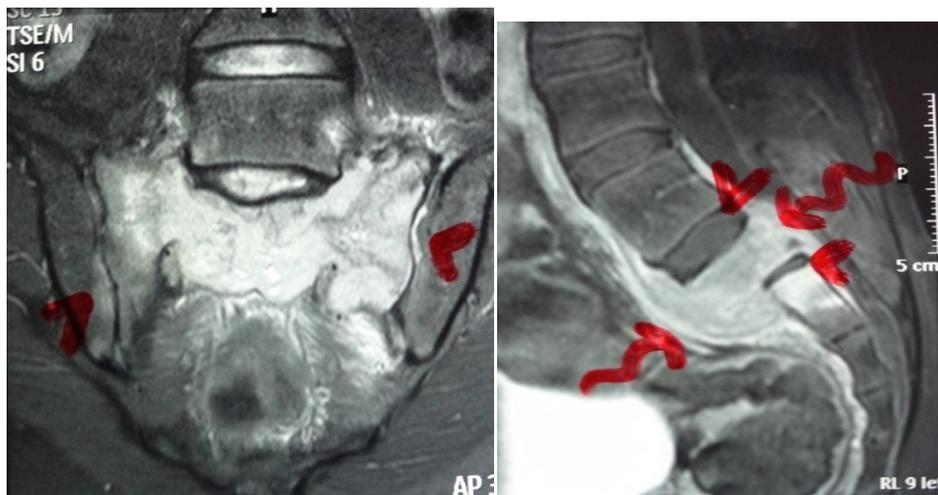


Figure 2: Coronal MRI showing the hyperintense lesion in S₁ extending into both sacro-iliac joints (arrow heads). Sagittal MRI shows subligamentous extension (curved arrows) and L₅-S₁, S₁-S₂ disc sparing (arrow heads).

CT guided FNAC was done, but it was inconclusive.

Open biopsy was done. The lesion was approached posteriorly through left paramedian incision over lumbosacral spine. Greyish white firm soft tissue mass was seen invading laminae and pedicles. S₂ nerve root could not be identified separately from the mass, hence it was resected with the biopsy specimen.



Figure 3: Paramedian operative scar

Histopathological examination showed multiple granulomas with central caseation surrounded by Langhans giant cells, epithelioid cells and lymphocytes. Tissue culture showed the growth of *Mycobacterium Tuberculosis*.

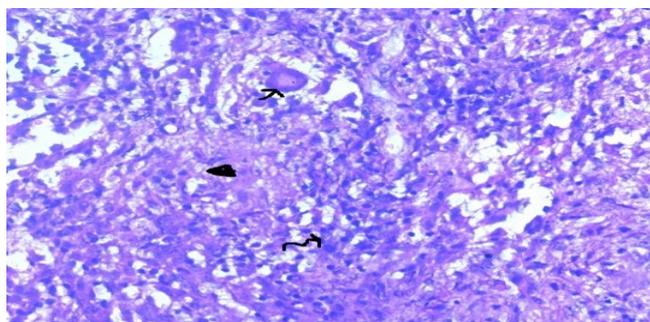


Figure 4: Histopathological examination showing Langhans giant cells (straight arrow), epithelioid cells (curved arrow) and caseous necrosis (triangle).

Post-operatively, patient developed numbness in S₂ dermatome in left leg. There was no loss of motor power in legs and no bowel or bladder disturbances.



Figure 5: Area of parasthesia due to S₂ nerve root avulsion.

Patient received six months of anti tubercular treatment as per DOTS category I. Though patient was advised bed rest for two months, patient did not comply. At the end of six months, patient was completely relieved of pain and was comfortably attending to his regular activities. ESR decreased to 20mm in first hour.

Patient has no disability at one year follow-up. His sensory loss in S₂ dermatome remained same. Follow-up radiographs show moderate remineralization.

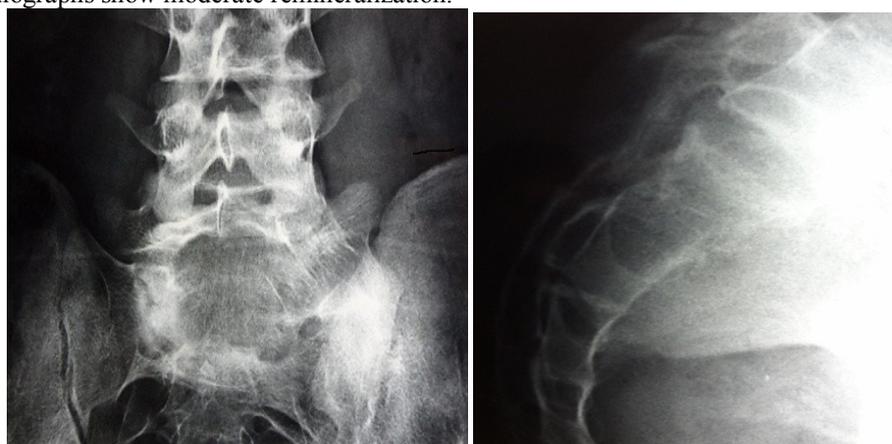


Figure 6: Post-operative radiograph after one year showing partial mineralization.

III. Discussion

Osteoarticular tuberculosis is always secondary to a primary source in lungs or mediastinal lymph nodes. Reactivation of the bacilli is more common in immunocompromised individuals. In our case, tuberculosis affected well nourished, immunocompetent individual who had no apparent contact with any individual having tuberculosis.

Though spinal tuberculosis is the most common form of skeletal tuberculosis, sacral involvement is very rare. Only isolated case reports or case studies with less than 15 patients are reported so far.

Patients usually suffer from constitutional symptoms like low grade fever, loss of weight and loss of appetite along with backache. Apart from backache, this patient did not have any other symptom suggesting inflammatory or infective disorder. Hence the diagnosis was delayed for six months.

Tuberculosis occurs in two forms[5]. One is caseous exudative type, with excessive granulation tissue, paravertebral abscess, destruction of bone and neurological signs. The constitutional symptoms and local signs of inflammation are more marked. The second granular type is a dry lesion, with less destruction of bone and no abscess formation. Ours is second type with insidious onset, no constitutional symptoms, no inflammatory signs and no abscess formation. But the disease progressed for a longer period, resulting in lysis of first and second sacral vertebrae.

Radiologically, tuberculosis of spine is classified into four types – paradiscal, central, anterior and appendicular. But a new type of spinal tuberculosis is found in recent studies[6] – spondylitis without disc involvement. In this, adjacent vertebral bodies are affected, but the intervening disc is spared. But our patient did not belong to any of these five types. He had involvement of both anterior and posterior elements of S₁ and S₂.

MRI of TB spine shows intervertebral disc destruction, inflammatory oedema, granulation tissue and multiple abscesses. Our patient had lysis of S₁ and S₂ with involvement of both sacroiliac joints. But S₁-S₂ disc was spared. Only soft tissue mass was seen and there was no inflammatory oedema or paravertebral abscess. The pathological tissue encased the nerve roots. As the disc was spared, the inflammatory tissue was less and there was lysis of S₁ and S₂ where tuberculosis is very rare, the most probable diagnosis was a neoplasm. So, clinically and radiologically, sacral malignancy was our working diagnosis.

Similarly mistaken case of sacral tuberculosis appearing as sacral tumor was reported by Chang KY et al[7].

VPS Punia et al described a case of sacral tuberculosis which mimicked cauda-conus syndrome clinically, but was diagnosed correctly on MRI with typical findings[8].

At present, tuberculosis is treated with DOTS under four categories[9]. WHO placed spinal TB in category I. In this category, intensive treatment for two months with Isoniazid, Rifampicin, Pyrazinamide and Ethambutol and continuation treatment for four months with Isoniazid and Rifampicin is given. Some authorities recommend a seven month continuation phase with Isoniazid and Rifampicin in patients with neurological signs. Tuberculosis of other bones and peripheral joints is included in category III, if the patient is not seriously ill. In this category, Ethambutol is excluded and rest of the treatment is similar to category I. A seriously ill patient with tuberculosis of any bone or joint should receive category I treatment. Drugs can be given daily or intermittently, varying their dosages. Intermittent administration of drugs is based on the discovery that when tubercle bacilli are exposed to a single dose of antituberculous drugs, there is a lag period of 24-48 hours before renewed growth of bacteria occurs. DOTS enhanced patient compliance and decreased the incidence of relapse and drug resistance.

But DOTS is not preferred by orthopaedic surgeons as ATT for 6-9 months is not thought to be sufficient to treat osteoarticular tuberculosis. The source of this appears to be the absence of radiological evidence of healing by six months[10]. A study by Rejith Valsalan et al[3] showed the efficacy of DOTS given for nine months. They achieved 92% success rate with DOTS in patients with predestructive and early destructive phases of tuberculosis. Our case indicates that short course treatment may be efficient in advanced stage of the disease also. Meta-analysis done by Van Loenhout-Rooyackers JH et al[4] showed that there is no difference in the rate of relapse between patients treated with six months of ATT and patients treated with ATT for a long duration.

Prolonged treatment is indicated only[2]

1. in paediatric patients.
2. if surgical debridement is indicated but cannot be done
3. if there is co-existent HIV infection or any other immunodeficient state.

Anil K.Jain et al[10] considered MRI findings like complete resolution of abscess collection, replacement of infected bone marrow with fat as indicators to stop ATT. Taking these as markers of healing, they evaluated 51 patients. Patients with atypical findings and sacral tuberculosis were excluded from this study. Three were unresponsive to standard drugs. Out of 48 patients, 31(65%) were healed in 18 months. But rest of the patients had taken/have been taking ATT for three years or longer. Besides being cost prohibitive, accepting MRI findings as healing markers appears to prolong the treatment for a long period.

Three phase bone scan was suggested to be useful in monitoring response to ATT by Ya-Ju Tsai et al[11]. But considering its limitations like multiple radiation exposures, high cost, false negative results in immunodeficient individuals, it doesn't appear to be of much clinical use in deciding the termination of the treatment.

We treated the patient with intermittent regimen of DOTS Category I for six months. Patient was relieved of pain within two months of treatment and remained so until now. Present radiographs at one year follow-up show partial regeneration of S₁ and S₂. Though patient has anaesthesia in S₂ dermatome in left thigh, he doesn't have any disability in attending to his daily activities and to his studies.

IV. Conclusions

1. Backache in younger individuals not reduced with conservative management for one month should be thoroughly investigated to prevent delay in diagnosing underlying condition.
2. Tuberculosis can mimic malignancy on MRI. Biopsy of the lesion is mandatory to confirm the diagnosis before planning the definitive management.
3. In immunocompetent individuals, ATT for six months may be sufficient to treat tuberculosis of spine. Prospective control studies should be done to confirm the efficacy of short term treatment.
4. Discovery of a reliable and cost-effective marker of healing in patients with skeletal tuberculosis is necessary.

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