



Isolated Sacral Tuberculosis in an Immunocompetent Patient Presenting As a Presacral Mass – A Rare Case

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ABSTRACT:-

Introduction:- Tuberculosis (TB) is still the second most frequent infectious disease after malaria on a worldwide basis and remains a major cause of skeletal infection in many parts of the world. Vertebral tuberculosis, the most common form of osseous compromise, is responsible for approximately 50% of cases that present with bone destruction. The lower thoracic and upper lumbar spines are the usual affected regions in such patients. Isolated sacral tuberculosis, however, is a rare condition, with scant reports in medical literature.

Case report:- A 25 year old Indian female computer professional was referred to us with chief complains of low back pain radiating to her left thigh region since 9 months. The other associated symptoms were tingling sensations and numbness in posterior aspect of her left thigh region, generalised weakness and intermittent constipation. On physical examination, patient had tenderness over sacral region on deep palpation without any swelling, sinus or spinal deformity and reduced sensation in S2 dermatome. Her laboratory investigations showed lymphocytosis with raised erythrocyte sedimentation rate and low haemoglobin. Magnetic resonance imaging (MRI) showed isolated presacral mass suggestive of infective etiology. Computed tomography guided biopsy showed typical caseating granuloma on histopathological examination confirming tuberculous origin. Patient was treated with extended WHO-DOTS therapy for 8 months on ambulatory basis and patient recovered completely within a year. A two year follow up of patient with magnetic resonance imaging showed complete resolution of presacral mass due to tuberculosis.

Conclusion:- Sacral tuberculosis is a rare disease, unusually responsible for refractory low back pain and atypical neurologic symptoms. The diagnoses suspicion is essential in endemic countries like India, thus diagnostic work up and treatment might be performed as soon as possible.

KEYWORDS:- Sacrum, Skeletal tuberculosis, WHO-DOTS, Low back pain, MRI, Caseating granuloma.

I. INTRODUCTION

Tuberculosis (TB) is still the second most frequent infectious disease after malaria on a worldwide basis and remains a major cause of skeletal infection in many parts of the world. Each year, 3.8 million new cases of tuberculosis are reported globally, the vast majority in the developing countries. Tuberculosis of bones and joints accounts for approximately 10% to 15% of all extrapulmonary forms of tuberculosis.¹ The proportion of spinal tuberculosis to all TB cases varied from 1% to 5%. Spinal tuberculosis is the most common form of skeletal tuberculosis, constituting approximately 50% of all cases. The most frequent region of the vertebral column involved by tuberculous infection is the dorso-lumbar region.² However, Sacrum is a rare site for tuberculosis. With the rise in the number of AIDS patients and emergence of multidrug resistant tuberculosis, tuberculosis should be considered as one of the possibilities in lesions present in odd areas of the human body.³

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Studies on cases with isolated form of tuberculosis affecting the sacrum are rare.² Hereby, we are reporting an interesting case of isolated sacral tuberculosis with a presacral mass in an immunocompetent patient which was treated with chemotherapeutic treatment successfully.

II. CASE REPORT

A 25 year old Indian female computer professional was referred to our outpatient department with chief complains of low back pain radiating to her left leg since 9 months which was dull aching in nature , continuous, aggravated while sitting or squatting, radiating to left thigh region over posterior aspect till knee and partially relieved with analgesics. Patient had previous multiple consultations with other clinicians but had no relief. The other associated symptoms were tingling sensations and numbness in posterior aspect of her left thigh region, generalised weakness and intermittent constipation. She also gave history of weight loss of about 12 kilograms in last 3 months. Patient had no history of fever, chronic cough, night sweats or contagion with TB patient. The patient had received the Bacillus Calmette– Gue ´rin (BCG) vaccine against TB as a routine vaccination during childhood. Patient had no previous history of any immunocompromised conditions like HIV infection, diabetes mellitus, steroid intake, radiation or cancer. On physical examination, patient was afebrile, normotensive with pallor, without generalised or local lymphadenopathy. Her respiratory and abdominal examination found to be normal. On local examination, patient had tenderness over sacral region on deep palpation without any swelling, sinus or spinal deformity. Patient had no impaired motor power or reflexes in lower limbs, though she had altered sensation for touch and temperature in S2 dermatomal distribution. On further examination, patient had none signs of nerve root tension or sacroiliac joint involvement. Roentgenographic evaluation of lumbosacral region of patient did not reveal any abnormality . However, her laboratory investigations showed total leucocyte count of 10,600 cells/mm³ with neutrophils (42%) and lymphocytes (57%) , while Haemoglobin was 7.8 gm/dl and ESR was 90 mm/1st hour. Markers for HIV and hepatitis B surface antigen were negative but her Mantoux test was positive with an induration of 20×15 mm. Her altered laboratory investigations gave impetus for further investigation in form of magnetic resonance imaging (MRI) which revealed hypointense signal on T1W and hyperintense signal on T2W and STIR images in S1 to S3 vertebral bodies. It was associated with presacral collection measuring 8.1 (Tr) × 2.1(AP)× 3.2(SI)cm in transverse, anteroposterior and superoinferior dimensions respectively suggestive of infective etiology [Figure 1 & 2]. There was no evidence of any skip lesion or disc bulge/protrusion/destruction. CT(Computed Tomography) guided biopsy was done, which was positive for multiple granulomas and central caseation surrounded by Langhans giant cells, epithelioid cells and lymphocytes on histopathological examination confirming tuberculous origin.

Thus, according to RNTCP(Revised National Tuberculosis Control Programme, India) protocol , patient was provided with Extended WHO-DOTS CAT I regimen including Rifampicin (450 mg) , Isoniazid (300 mg), Pyrizinamide (1500mg) and Ethambutol (1200 mg) thrice a week for 2 months in intensive phase and 6 months of continuation phase with Rifampicin(450mg) and Isoniazid (300mg) on ambulatory basis. Patient had been regularly followed up at monthly interval with routine investigations of Total leucocyte counts and ESR for therapeutic response which showed progressive improvement along with clinical improvement of patient’s pain and paraesthesia and overall well being. Over two months of DOTS therapy , patient had relief of low back pain and pain radiating to left thigh region. At end of therapy , patient had no symptoms of low back pain or leg pain or numbness in thigh region and was able to carry out her deskjob of long hours comfortably. Magnetic resonance imaging done at one year and two years of follow up showed complete resolution of presacral mass/collection with no altered signals in sacral region. [Figure 3 & 4]

III. DISCUSSION

The tubercle bacillus has co-existed with Homo sapiens since time immemorial. The Rig Veda, Atharva Veda (3000 - 1800 BC) and Samhita of Charaka & Sushruta (1000 & 600 BC) recognized the disease as “Yakshma” in humans, which by its symptoms and signs could only be tuberculosis of the lungs. Tuberculous lesions have been found in Egyptian mummies and the Greco Roman civilisation recognised phthisis or consumption as a problem of the lungs.⁵ Spinal tuberculosis remains a frequent disease in developing countries. The dorsolumbar spine is the seat of choice (95%), whereas the cervical spine is affected in only 5% of the cases . Tuberculosis of the sacrum is rarely reported. The diagnosis can be easily delayed because of non-specificity of clinical signs and can suggest numerous other disease entities in particular malignancy. The time lapsed till TB diagnosis was 9 months in our case. In the literature the mean of this time was 6.5 ± 2.5 months with a range from 3 to 12 months.⁶⁻⁸ The most common presenting symptoms are non-specific pain and swelling. Consequently, skeletal tuberculosis frequently mimics neoplasias like chordoma and osteoclastoma or sometimes metastasis leading to incorrect initial diagnosis and delay in the institution of treatment.² The first case of sacral tuberculosis described in medical literature was reported by Campbell in 1917. His patient was a 3-year old boy who had tubercular lesions in lower lumbar region and sacrum. In 1998, Rajshekar reported a 15-year follow-up of 53 patients. Of these only 12 had spread disease inferiorly to

lumbosacral junction. Pun reported 20-year follow-up of 26 patients of tuberculosis of lumbosacral junction. In majority of patients, the disease had started in lumbar spine and descended to the sacrum. In 1985, Dayras reported a case of isolated sacral tuberculosis with lower back pain. In 2004, Wellons reported a case of sacral tuberculosis with lower back pain and difficulty in walking.⁷ Our case was an isolated form of sacral tuberculosis without any skip lesions with S2 sensory involvement. This might have resulted from involvement of left lumbosacral plexus. Sament et al⁹, have described a case report of monoparesis with isolated sacral tuberculosis showing similar involvement of lumbosacral plexus.

In literature, it was stressed that MRI is more sensitive than radiography and more specific than scintigraphy in diagnosis of spinal tuberculosis.^{4,10} In our case, radiography was inconclusive while MRI helped us clinch a diagnosis. Thus, MRI by far remains non invasive investigation of choice in diagnosis of Spinal tuberculosis. According to Patankar et al¹¹, S2 vertebra was always involved in pathology in his all 15 cases of isolated sacral tuberculosis. We share his finding too in our case.

Medical treatment involves a combination of four drugs: rifampicine, isoniazide, pyrazinamide and ethambutol, during 2 months, followed by bitherapy. Regarding treatment there are different opinions in the literature. Many workers prescribe chemotherapy for 6 months while some continue it for 9–18 months.² The total duration of antituberculous therapy was 8 months in our patient with a favorable evolution under extended DOTS (Directly Observed Treatment, Short course) chemotherapy supported by World health organisation (WHO).

Isolated sacral tuberculosis is a very rare condition, and to the best of our knowledge, its presentation as S2 sensory deficit with presacral mass has not been reported in literature so far. A case of sacral tuberculosis presenting with such presentation is not only atypical, but also it can cause difficulty and delay in making a diagnosis.

IV. CONCLUSION

Tuberculosis of bones has been known to present itself in a variety of atypical clinical forms. This interesting case of isolated sacral tuberculosis with sensory deficit was unique and emphasizes importance of atypical presentation of bony tuberculosis. In conclusion, isolated sacral tuberculosis is rare but it should be the first and foremost differential diagnosis in the presence of atypical clinical and radiological features of a sacral lesion particularly in developing countries like India.

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Clinical Message

Sacral tuberculosis is a rare disease, unusually responsible for refractory low back pain and atypical neurologic symptoms. The diagnoses suspicion is essential in endemic countries like India, thus diagnostic work up and treatment might be performed as soon as possible.

Abbreviations

TB- Tuberculosis , AIDS – Acquired immunodeficiency syndrome, HIV- Human immunodeficiency virus, RNTCP- Revised national tuberculosis control programme, CT – computed tomography, WHO-DOTS- World health organization – Directly observed therapy ;Short course, BCG – Bacillus Calmette– Gue´rin , MRI – Magnetic resonance imaging , T1W and T2W- T1 weighted and T2 weighted.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

UGW- Searching Literature. Writing case report and discussion sections. Collecting radiological investigation. Surgeon for CT guided biopsy. **AK**- Searching Literature. Editing case report and discussion. **PR** - Contributor in writing case report. Editing case report. Searching Literature. **NR**- Searching Literature. Writing case report and discussion sections. **RMP**- Searching Literature. Writing case report and discussion sections

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Conflict of Interest

Nil.

Illustrations and Figures



Figure 1 - Sagittal STIR (Short Tau Inversion Ratio) MRI of Lumbosacral Lesion showing presacral mass with altered marrow signal in S1 to S3

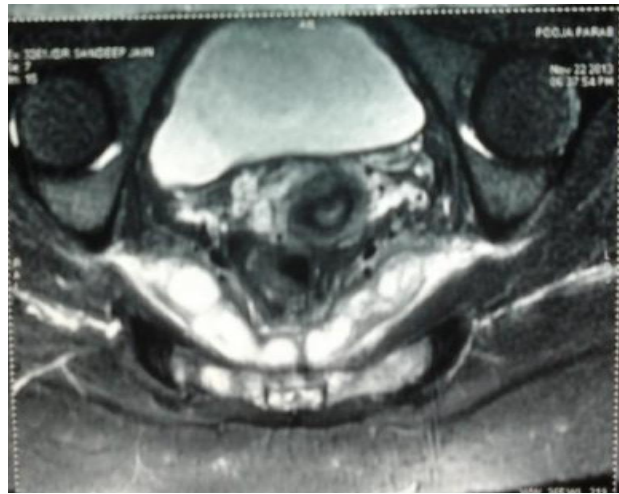


Figure 2 – Coronal STIR MRI through S2 level showing loculated presacral mass.

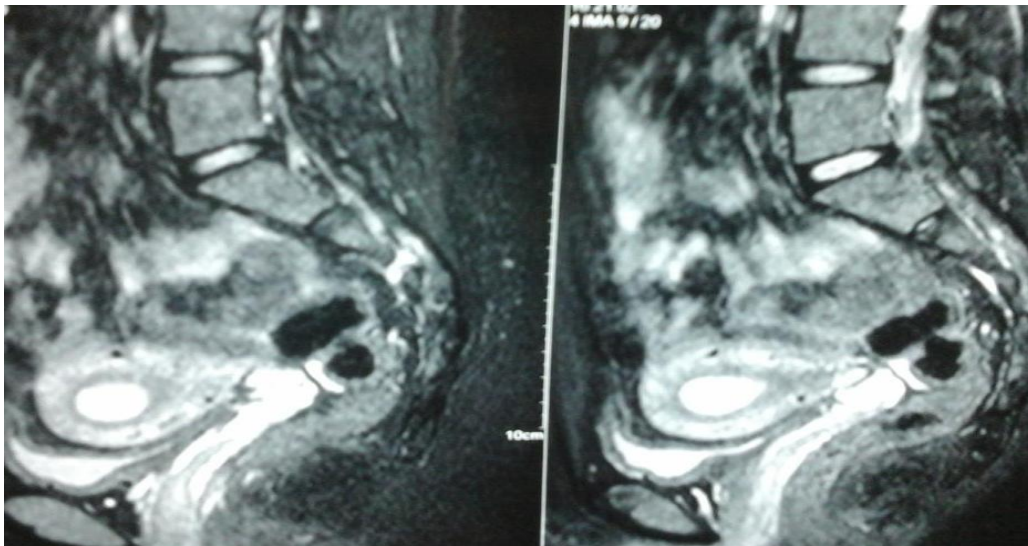


Figure 3- Post 1 year follow up STIR MRI images showing complete resolution of presacral mass with reduced oedema signal in S1 and S2 vertebra correlating with resolution of symptoms of patient.

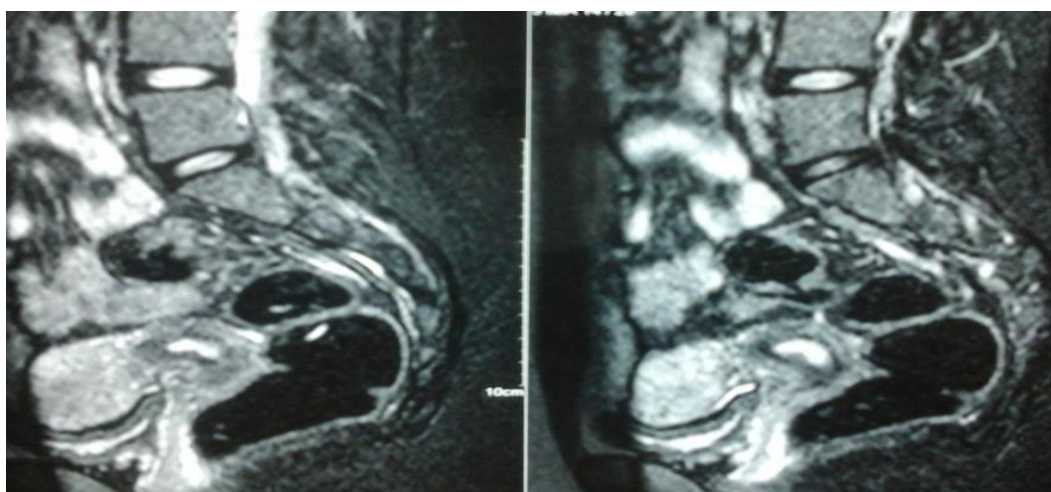


Figure 4 – Post 2 year follow up STIR MRI images showing complete resolution of presacral mass with no bony destruction.