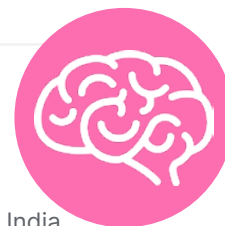


Isolated Sacral Tuberculosis: A Case Report and Review of Literature of this Rare Sacral Pathology

Authors:

Harshit Arora,¹ *Vasudha Sharma,² Waryaam Singh,³
Lavanya Arora,⁴ Sumer Singh Nanda,⁵ Rajesh Pasricha⁶



1. Brigham and Women's Hospital, Harvard Medical School, Boston, Massachusetts, USA
 2. Dayanand Medical College & Hospital, Ludhiana, Punjab, India
 3. Mayo Clinic, Rochester, Minnesota, USA
 4. Teerthanker Mahaveer University, Moradabad, Uttar Pradesh, India
 5. Sri Guru Ram Das Institute of Medical Sciences, Amritsar, Punjab, India
 6. Satyam Hospital & Trauma Centre, Jalandhar, Punjab, India
- *Correspondence to poster_de_vasu@outlook.com

Disclosure:

The authors report there are no competing interests to declare. The patient has provided consent to publish their case, and the identifiers have been taken out of the case so as to protect the patient's interests.

Received:

14.04.23

Accepted:

06.06.23

Keywords:

Neuroinfection, rare location, sacral pathology, sciatica, spinal tuberculosis (TB).

Citation:

EMJ Neurol. 2023; DOI/10.33590/emjneuro/10300316.
<https://doi.org/10.33590/emjneuro/10300316>.

Abstract

Introduction: Tuberculosis (TB) of the spine is one of the rare secondary manifestations of the disease, while isolated TB of the sacrum is an even rarer finding of the disease.

Case: The authors present the case of a male patient in their late 20s who was suffering from lower back pain radiating to the leg, on and off fever for 5 months, and stiffness of the lower back. X-ray and MRI showed a sacral mass-like lesion, which led to the suspicion of spinal TB. This diagnosis was later confirmed through adequate microbiological testing.

Methods and Materials: A review of literature by a thorough search of the PubMed and Google Scholar databases was carried out, and a total of 42 patients with isolated TB of the sacrum were studied.

Results and Conclusions: Sacrum in isolation is a rare location for spinal TB, often overlooked by primary physicians. A prompt diagnosis and early treatment of the disease will mitigate the risk of developing complications.

Key Points

1. Pott's spine (tuberculosis of the spine), a presentation of secondary tuberculosis with huge morbidity in India, rarely presents isolated to the sacral spine.
2. Pott's spine presents variably with a spectrum of symptoms, including high fever, weight loss, radiculopathy, and back pain, and it requires thorough clinical and radiological investigations to reach a definitive diagnosis.
3. The treatment modalities are dependent on the extent of spread of the disease, and include surgical intervention, as well as medical treatment in form of anti-tubercular therapy, with regular follow-up to prevent morbidity.

INTRODUCTION

Tuberculosis (TB) is an infectious disease caused by the transmission of *Mycobacterium tuberculosis* bacillary complex, which usually involves multiple systems if not recognised and treated early. TB of the spinal region is categorised as secondary TB, involving the spinal vertebrae and the intervertebral discs.¹ The burden of spinal TB is 5% of total TB cases around the globe; it is higher in developing countries, with the Indian subcontinent housing a majority of its cases. The overall risk of spinal TB increases with co-infection with HIV.²

The involvement of the spine in the case of TB generally occurs after the haematogenous spread of the bacteria from a primary region, usually the lungs. The spread occurs through the anterior part of the vertebral bodies, or may involve the central portion of the vertebrae by communicating through the valveless venous plexus or the Batson plexus.¹ Patients usually present with fever, night sweats, pain in the lower region of the back radiating to the legs, and weight loss.¹ X-ray is usually the primary imaging modality, while MRI of the spine is more specific. Biochemical and microbiological analysis of the biopsy or aspirate confirms the diagnosis.² It is pharmacologically treated by anti-tubercular therapy, supplemented by surgical management if the spread leads to physical deformities or complications.^{1,2}

In the wide spectrum of spinal TB, isolation to the sacrum is a rare presentation. Amongst various sacral pathologies diagnosed on imaging, TB of the sacrum is one of the rare conditions that need adequate clinical, biochemical, and microbiological analysis to be ruled as the

disease, which is why the authors believe a thorough review of the same is necessary.

CASE REPORT

A male patient in their late 20s presented to the outpatient department of a tertiary care hospital in Northern India with insidious, progressive pain in the lower back for the past 1.5 years, specifically in the right lower paraspinal region. The pain varied in intensity, radiating to the right lower leg, and was associated with weakness of lower limbs bilaterally. The pain was aggravated by walking and sitting, and relieved by taking analgesics. The patient had no seasonal or diurnal variation of the pain. The patient also had a low-grade fever, on and off for the past 5 months, which they managed with over-the-counter analgesics.

There was no reported previous history of any trauma to the back, or any similar complaints. There was a history of right inguinal hernia 6 years before presentation, treated with hernioplasty, and history of abscess in the scrotum 3 years before presentation, treated with incision and drainage of the abscess. The patient noticed no changes in their sleep pattern, appetite, bowel movements, or urination. The patient was a carpenter by profession, and would frequently lift heavy weights. There was no other relevant past, family, or personal history.

On examination, the patient was calm, conscious, co-operative, and well oriented to time, place, and person. There was no swelling or localised tenderness present in the lower back region. There was restricted bending of the lower back and the straight leg raise test was limited at 60° bilaterally. Normal power, tone, sensations,

and reflexes were observed in both legs with no significant signs. The higher motor, cerebellar, gastrointestinal, respiratory, cardiovascular, and urological examinations were normal.

Routine investigations, such as complete blood count and urine analysis, were within normal limits. Antero-posterior and lateral X-ray of the lower back showed suspicious lesions in the sacral area. Erythrocyte sedimentation rate and C-reactive protein levels were significantly elevated. An MRI of the spine showed a mass-like lesion in the sacral vertebrae, along with osteolytic lesions. A biopsy of the same ruled out any suspicion of malignancy. Ziehl-Neelsen staining of the same yielded acid-fast bacilli and a PCR confirmed the presence of *Mycobacterium* TB in the lesion. Chest X-ray was normal and showed no signs of infection. Human leukocyte antigen B27 was negative.

A diagnosis of isolated TB of the sacrum was reached, and the condition, treatment, and prognosis were explained to the patient. The patient was initiated on anti-tubercular therapy (ATT), consisting of rifampicin, isoniazid, and ethambutol for a period of 9 months, and pyrazinamide, streptomycin, and ofloxacin for a period of 3 months, along with pyridoxine and analgesics. The patient was transferred to the Directly Observed Therapy (DOT) governmental department, which ensured compliance and monitored cure and side effects.

METHODOLOGIES

Informed consent was taken from the patient to publish the case report, and all the personal identifiers were removed from the case report.

The authors carried out a thorough search of the PubMed and Google Scholar databases for cases of isolated sacral TB, using the keywords “sacral tuberculosis,” “tuberculosis of the sacrum,” and “sacrum tuberculosis.” Ultimately, the authors were able to find a total of 26 articles,³⁻²⁸ including one retrospective analysis of 15 cases,⁵ a case series comprising three cases,¹⁷ and 24 others classified as case reports. Articles without abstracts or available literature were excluded. Articles without isolated sacrum involvement, namely sacrococcygeal, lumbosacral, or multifocal spinal TB were also excluded.

DISCUSSION

The authors' patient presented with pain in the lower back, radiating to the lower limbs bilaterally with weakness and intermittent fever. The involvement of the lumbosacral region was speculated and the differential diagnosis of osteomyelitis, tumour of the local area, and lumbar disc herniation was taken into consideration.²⁹ An X-ray of the lumbosacral region was taken, which showed suspicious lesions. This was followed by an MRI to understand the extent of these lesions. As the patient had osteolytic lesions in the anterior sacral region, an histopathological investigation was advised to differentiate the infectious causes from neoplastic ones. After adequate biochemical and microbiological investigations, a diagnosis of sacral TB was established.

After a thorough review of the available medical literature, the authors isolated a total of 42 cases mentioned across 26 research articles. This case might be the 43rd case of isolated sacral TB reported across the aforementioned databases. The sample included 18 males and 23 females, and ranged from age 5.00–73.00 years (mean: 30.97 years). In their case report, Meurice et al.³ chose not to disclose the gender identity of the youngest patient. Patanakar et al.⁵ noted a much more severe disease presentation (abscesses and sinuses) in younger patients compared with older ones after reviewing 15 cases. The authors' patient, a male in their late 20s, fitted the typical presentation.

Sacral TB presents with a wide spectrum of symptoms, including fever, night sweats, pain in the lower back and legs, and weight loss, with the intensity ranging from very severe to mild or minimal. Lower back pain was the most common symptom, present in 34 patients, including the authors' case.^{4,5,7,9-23,25-28} This also includes the three cases in the case series by Lazrak et al.,¹⁷ and 10 cases in the observational study.⁵ Fever due to underlying inflammation^{5,16,17,19,21,27,28} and weight loss^{13,16-19,22,25,27,28} was reported in 10 cases. Rigidity and stiffness of the lower back were reported in five cases.^{4,5,9} Mild swelling of the local region was present in three cases,^{15,18,21} of the right buttock in one case,²⁰ and of the thighs bilaterally in a case reported by Djaja et al.²⁴

Night sweats, a common symptom of TB, were reported in four patients.^{7,16,17,28} The presence of neurological symptoms is a common feature in sacral TB due to the local involvement of the nerve plexus. Wellons et al.⁷ and Sament et al.¹⁶ both reported cases of numbness of the lower limbs with associated weakness in the latter. Patanakar et al.⁵ reported a case of paraparesis in their observational study. Lower limb functional loss was reported by both Meurice et al.³ in the left leg, and Lmejjati et al.⁶ in the right leg. Khosla et al.⁸ reported a case of a female who reported no symptoms of sacral TB during their pregnancy, but presented with obstructed labour due to a swelling of the sacral region obstructing the vaginal canal.

Sacral TB usually presents with localised symptoms, with very few cases diagnosed prior to presentation in another system of the body. The case reported by Meurice et al.³ presented as a recurrence post-9 months. Wellons et al.⁷ reported a positive purified protein derivative skin test in a patient in their 30s, 3 years prior to presentation. Shah and Kulkarni¹² reported a patient in their 20s who presented with lumbar TB 11 months prior to presentation. A patient in their 50s reported by Djaja et al.²⁴ had presented with TB of the lymph nodes 25 years prior to presentation. There was no diagnosed prior presentation of TB in the authors' patient.

On laboratory investigations, patients usually have elevated erythrocyte sedimentation rate and C-reactive protein levels due to the underlying infectious pathology,¹ while leukocytosis may or may not be present in the patient, similar to the presentation of the authors' case. Chang et al.¹⁰ reported a case of sacral TB mimicking a neoplasm with elevated levels of carbohydrate antigen 19-9. A mandatory chest X-ray should be done in all cases of sacral TB so as to rule out pulmonary TB. Of the 42 cases available, an abnormal chest X-ray with the presence of pleural effusion and atelectasis of the right side was present just in one case.²⁸

An initial diagnosis may be made using a radiograph of the spinal region, but the definitive diagnosis and the extent of the disease are better understood via MRI of the spinal region, as MRI is more sensitive than a spinal radiograph, and more specific than a CT scan of the same region.¹ The most common MRI finding is the

presence of osteolytic lesions in the sacrum, followed by a mass-like presentation that could be mistaken for cancerous growth.³⁻²⁸ In a few cases, the infection might extend to the piriformis muscles, leading to the formation of abscesses and collections in the same.^{16,20,21,24,25} The tissue diagnosis can be confirmed by aspirating tissue and analysing histopathology (showing acid-fast bacilli), Lowenstein Jensen culture, and PCR.^{1,14}

Adequate treatment for sacral TB includes primary therapy ATT, consisting of isoniazid, rifampin, pyrazinamide, and ethambutol. The World Health Organization (WHO) recommends ATT for a period of 9 months for patients who have TB in bones and joints,³⁰ while the American Thoracic Society (ATS) recommends ATT for 12 months for children and 6 months for adults.³¹ The duration of ATT can vary depending on the local guidelines, as well as the severity of the disease. The reviewed cases had ATT prescribed varying from 6⁷ to a maximum of 18 months.^{13,16,19} Surgical intervention was required in a female patient in their 20s with a severe destructive lesion of the S1 body, requiring a lumbopelvic fixation, along with S1 reconstruction using an allograft.¹² Surgical debridement was carried out for a female patient in their 50s who had developed submuscular gluteal abscesses bilaterally as a complication of sacral TB.²⁴

In the absence of early diagnosis and adequate treatment, complications may occur, such as structural and postural deformities, permanent neurological complications, and the extension of the disease to the neighbouring structures, leading to the formation of cold abscesses in the gluteal and piriformis muscles.^{1,2,21,24} Surgical intervention might be required in patients who develop structural complications, such as bone insufficiency, abscesses, and spinal deformities.¹

The prevalence of TB is higher in lower-middle income countries, and India suffers from a significant burden of the disease owing to the associated morbidity and mortality.³² To address this issue, it is crucial to prioritise the primary and secondary prevention of the disease. This involves implementing measures by identifying individuals at high risk, such as immunodeficient individuals, young individuals, and patients suffering from chronic diseases of diabetes, chronic kidney disease, and HIV.³³

Additionally, early detection and identification of the disease is necessary. Tuberculin skin test and interferon- γ release assays serve as valuable methods for detection of the disease.³⁴ Timely diagnosis and anti-tubercular therapy administration can potentially reduce the morbidity and complications associated with the disease.³⁵

Sacral TB might extend to the other surrounding structures. Lumbosacral TB is common in comparison to isolated sacral TB, and sacrococcygeal TB is rarer.^{36,37} Sacrococcygeal TB presents as lower back pain³⁶ or gluteal and coccygeal pain with anococcygeal fistula and discharge.³⁷

CONCLUSION

Sacrum in isolation is a rare location for spinal TB, often overlooked by primary physicians. A prompt diagnosis and early treatment of the disease will mitigate the risk of developing complications. The authors hope that this review helps physicians and surgeons to consider TB of the sacrum as one of the key differential diagnoses in patients exposed to the endemic region presenting with a sacral lesion.

References

1. Khanna K, Sabharwal S. Spinal tuberculosis: a comprehensive review for the modern spine surgeon. *Spine J.* 2019;19(11):1858-70.
2. Garg RK, Somvanshi DS. Spinal tuberculosis: a review. *J Spinal Cord Med.* 2011;34(5):440-54.
3. Meurice JC et al. Ostéite tuberculeuse sacrée. *Rev Mal Respir.* 1994;11(4):418-20.
4. Raymond F et al. Tuberculose osseuse sacrée chez un enfant de 6 ans. *Arch Pediatr.* 1994;1(5):489-92.
5. Patankar T et al. Imaging in isolated sacral tuberculosis: a review of 15 cases. *Skeletal Radiol.* 2000;29(7):392-6.
6. Lmejjati M et al. Tumor-like tuberculosis of the sacrum. *Joint Bone Spine.* 2000;67(5):468-70.
7. Wellons JC et al. Sacral tuberculosis: a case report and review of the literature. *Surg Neurol.* 2004;61(2):136-9.
8. Khosla AH et al. Obstructed labour due to sacral TB. *Trop Doct.* 2005;35(4):236-7.
9. Kumar A et al. Unusual presentation of isolated sacral tuberculosis. *Joint Bone Spine.* 2006;73(6):751-2.
10. Chang KY et al. A case of sacral tuberculosis mimicking metastatic bone tumor with elevated CA 19-9. *J Yeungnam Med Sci.* 2011;28(2):196-201.
11. Coffman GJ. Presentation of a rare sacral tuberculosis in an otherwise healthy patient: diagnostic challenge and review of treatment. *J La State Med Soc.* 2012;164(2):67-9.
12. Shah SP, Kulkarni AG. S1 tuberculosis treated with segmental lumbopelvic fixation: a case report. *Spine (Phila Pa 1976).* 2012;37(14):E866-9.
13. Shantanu K et al. Tuberculosis of sacrum mimicking as malignancy. *BMJ Case Rep.* 2012;2012:bcr0720114505.
14. Huang YH et al. Polymerase chain reaction as the sole diagnostic tool for sacral-spinal tuberculosis. *Surg Infect (Larchmt).* 2013;14(2):225-8.
15. Rekha YB et al. Atypical sacral tuberculosis: a case report and review of treatment. *IOSR-JDMS.* 2013;10(1):41-5.
16. Sament R et al. Isolated tuberculosis of sacrum with monoparesis: an atypical presentation. *Asian Spine J.* 2013;7(4):351-4.
17. Lazrak F et al. A rare location of sacral tuberculosis: a report of three cases. *Eur J Rheumatol.* 2014;1(2):78-80.
18. Oniankitan O et al. Sacrum Pott's disease: a rare location of spine tuberculosis. *Egypt Rheumatol.* 2014;36(4):209-11.
19. Dharmshaktu GS et al. Isolated sacrum tuberculosis with presacral and paraspinal abscess: case report of a common disease at uncommon site. *Int J Res Orthop.* 2015;1(1):28-30.
20. Okuyama Y et al. Sacral Tuberculosis. *Intern Med.* 2016;55(14):1939-40.
21. Khariwal V et al. A rare case report at rare site tuberculosis of sacrum sacroiliac joint with gluteal abscess indication for drainage of cold abscess which usually treated by conservative treatment. *Int J Orthop.* 2017;3(2):885-9.
22. Munakomi S. Isolated sacral and pelvic TB abscess—an enigmatic tale of a ubiquitous pathogen. *Oxf Med Case Reports.* 2017;2017(5):omx019.
23. Cavalcante RA et al. [Sacral Tuberculosis in the Elderly]. *JBNC.* 2013;24(4):335-9. (in Portuguese).
24. Djaja YP et al. Submuscular gluteal abscess: an unusual presentation of rare sacral tuberculosis. *Int J Surg Case Rep.* 2019;54:55-9.
25. Mirah FAS et al. Sacral tuberculosis: an atypical manifestation. Abstract 33. Medical Research Symposium Kulliyah of Medicine, 29 August, 2018.
26. Lekha AK, Krishnakumar J. A rare of sacral tuberculosis in an immunosufficient woman: a case report. *Drug Invent Today.* 2019;12(11):2775-7.
27. Sinaga PL et al. Tuberculosis of sacrum: a rare location of spinal tuberculosis. *AANHS-J.* 2019;DOI: 10.32734/aanhs-j.v1i1.1127.
28. Khoury NC et al. A rare case of sacral Pott's disease. *IJID.* 2021;2(1):21-2.
29. Jensen RK et al. Diagnosis and treatment of sciatica. *BMJ.* 2019;367:l6273.
30. World Health Organization (WHO). Treatment of tuberculosis guidelines, fourth edition. 2010. Available at: <https://apps.who.int/iris/rest/bitstreams/52536/retrieve>. Last accessed: 21 March 2023.
31. Bass JB Jr et al. Treatment of tuberculosis and tuberculosis

- infection in adults and children. American Thoracic Society and the Centers for Disease Control and Prevention. *Am J Respir Crit Care Med.* 1994;149(5):1359-74.
32. Prathiksha G et al. Active case-finding for tuberculosis in India. *Natl Med J India.* 2019;32(2):90-5.
33. World Health Organization (WHO). Latent tuberculosis infection: updated and consolidated guidelines for programmatic management. 2018.
- Available at: <https://apps.who.int/iris/bitstream/hand>. Last accessed: 21 March 2023.
34. Salazar-Austin N et al. Seventy years of tuberculosis prevention: efficacy, effectiveness, toxicity, durability, and duration. *Am J Epidemiol.* 2019;188(12):2078-85.
35. Balcells ME et al. Isoniazid preventive therapy and risk for resistant tuberculosis. *Emerg Infect Dis.* 2006;12(5):744-51.
36. Osman W et al. A rare case of tuberculosis with sacrococcygeal involvement miming a neoplasm. *Case Rep Orthop.* 2016;2016:7286806.
37. Takakura Y et al. Rare case of sacrococcygeal tuberculosis mimicking as an anal fistula. *Int J Surg Case Rep.* 2018;49:74-7.