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Rare case of sacrococcygeal tuberculosis mimicking as an anal fistula

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ABSTRACT

INTRODUCTION: Spinal tuberculosis (TB) is the most common manifestation of extra-pulmonary TB. TB of the lumbosacral junction is rare and occurs in only 1 to 2% of all cases of spinal TB. Moreover, isolated sacrococcygeal TB is extremely rare. Herein, we report a rare case of sacrococcygeal TB, which was difficult to distinguish from complex anal fistula.

CASE PRESENTATION: A 93-year-old man presented with sacral pain and peri-anal discharge. He had pulmonary TB at 25 years of age. Fistulography revealed an abnormal tract that connected to the pre-sacrococcygeal area, which was not connected to the rectum. Computed tomography scan showed fluid collection in front of the sacrum, with a lytic destruction of the lower sacrum and coccyx. Cold abscess aspiration cytology was negative for acid-fast bacilli. However, real-time polymerase chain reaction was positive for *Mycobacterium tuberculosis*. His symptoms resolved immediately after the initiation of anti-TB chemotherapy.

CONCLUSION: This case highlights the importance of considering tuberculosis as a diagnosis if the unusual sites are involved.

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1. Introduction

Despite the improvement in public health policies and the availability of effective antibiotics, tuberculosis (TB) remains a major health problem worldwide, particularly in developing countries. Although Japan is a developed country, it is still categorized as TB middle-burden country with a notification rate of 13.9 per 100,000 population [1]. This is mostly attributed to an aging population who become infected during his or her young age, and are now developing disease due to a comprised immune system.

Among the various forms of the disease, vertebral involvement represents the primary manifestation of extra-pulmonary TB [2]. The dorsolumbar is the most frequently involved region of the vertebral column in TB infection, and infection of the lumbosacral junction is relatively rare [3]. Moreover, isolated sacrococcygeal TB is extremely rare, and only few cases are reported in the literature [4–7].

Abbreviations: PCR, polymerase chain reaction; CT, computed tomography; MRI, magnetic resonance imaging.

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Herein, we report a rare case of isolated sacrococcygeal TB with peri-anal discharge, which was difficult to distinguish from recurrent complex anal fistula. This work has been reported in line with the SCARE criteria [8].

2. Presentation of case

A 93-year-old Japanese man who presented with gluteal and coccygeal pain was referred to our hospital. Furthermore, discharging were observed in his fistula in the posterior peri-anal region. He had neither fever nor respiratory symptoms.

He had history of pulmonary TB at 25 years of age, and underwent surgery for the removal of complex anal fistula at 75 years of age at another institution.

Based on clinical examination, there were two fistulous orifices between the anus and coccyx with purulent discharge (Fig. 1). He did not present with cough, fever, weight loss, and anorexia. The chest radiograph result was normal, and hematologic examination showed no abnormalities, except for a slight elevation in C-reactive protein level.

Fistulography showed a complex supra-elevator track that is connected to the pre-sacral area and another track ending blindly behind the rectum. The connection between the track and the rectum or anus was not identified on fistulography (Fig. 2). A computed



Fig. 1. Peri-anal inspection during initial visit. There were two fistulous orifices between the anus and coccyx (arrows).

tomography (CT) scan of the whole body revealed a lytic destruction of the sacrum (S3–5) and coccyx, with a low-density area in front of the sacrum, suggesting fluid collection (Fig. 3A). No abnormality was found in the lungs and other vertebrae on CT scan. Colonoscopy findings showed normal colorectal mucosa and no evidence of fistula.

Based on these radiological findings and the past medical history of the patient, the recurrence of complex anal fistula with osteomyelitis, or metastatic bone tumor of unknown origin, was suspected.

Bacterial examination of the fistula discharge showed normal bacterial culture and was negative for acid-fast bacilli (AFB). However, real-time polymerase chain reaction (PCR) using COBAS TaqMan MTB test (Roche Molecular Systems, the USA) was used to test the cold abscess fluid, and results showed that the patient was positive for *Mycobacterium tuberculosis*. AFB test and PCR were negative for sputum. However, the T-SPOT.TB (Oxford Immunotec, the

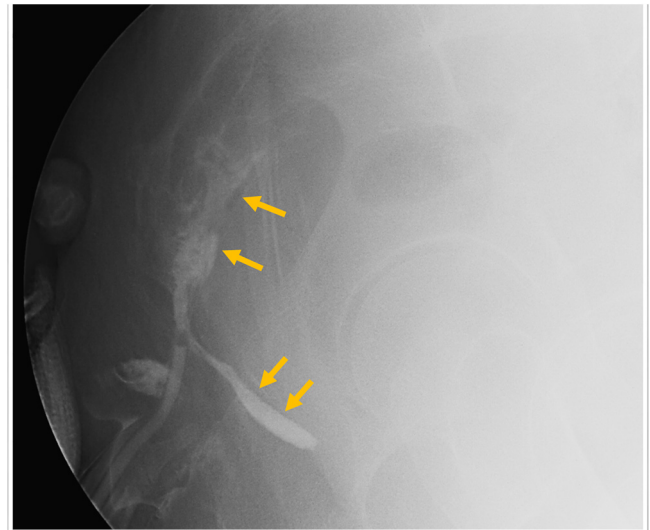


Fig. 2. Fistulography showed a complex supra-elevator track that is connected to the pre-sacral area and another track ending blindly behind the rectum (arrows). No connection between the track and the rectum or anus was identified.

UK) blood test result was positive. Thus, the diagnosis of isolated sacrococcygeal TB was confirmed.

The patient was treated with rifampicin, isoniazid, and ethambutol for 2 months, followed by rifampicin and isoniazid for 7 months. The patient's pain resolved, and the purulent drainage stopped within the first 2 months of treatment. Based on the physical findings, closure of the fistula was not achieved. However, the PCR test result for PCR DNA turned to be negative. A follow-up CT scan was carried out during treatment, and results showed that fluid collection in front of sacrum had disappeared (Fig. 3B).

3. Discussion

TB remains a rampant infectious disease of global importance, and it is still a threat to both clinicians and radiologists due to its

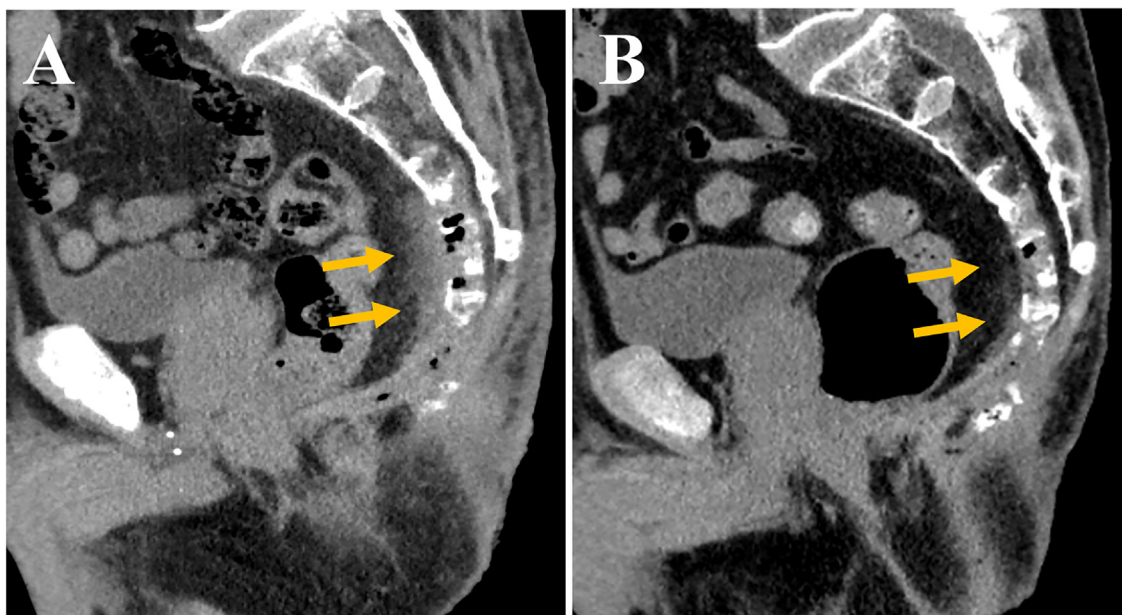


Fig. 3. (A) Sagittal CT image before treatment. CT scan showed a lytic destruction of the sacrum and coccyx, with a low-density area in the pre-sacrococcygeal area, suggesting inflammatory fluid collection. (B) The sagittal CT image after treatment. Fluid collection in front of the sacrum was disappeared.

Table 1
Reported isolated sacrococcygeal tuberculosis in the literature.

Author	Published year	Country	Gender	Age	Chief complaint	Diagnostic method	Time between onset and diagnosis
Kumar	2006	India	F	42	Coccygeal pain and anal fistula	Needle biopsy	6 months
Kim	2012	Korea	M	35	Coccygeal and gluteal pain	CT-guided biopsy	3 months
Osman	2016	Tunisia	F	55	Lower back pain	Open biopsy	12 months
Our case	2017	Japan	M	94	Coccygeal pain and anal fistula	PCR of discharge	2 months

often non-specific and diverse clinical manifestations. Moreover, rare symptoms and unusual location can cause untimely and inaccurate diagnosis and management of the condition.

The sacrum is not a common site in TB involving the spine. Moreover, isolated sacrococcygeal or coccygeal TB is extremely rare, and only 3 cases are reported in the literature (Table 1). Kumar et al. reported the case of an isolated TB of the coccyx that presented as an anal fistula [5]. Therefore, this is the second case of isolated sacrococcygeal TB mimicking an anal fistula. The diagnosis of all 3 cases involved an invasive procedure (open or needle biopsy). However, our is the first case involving isolated sacrococcygeal TB that was diagnosed through a non-invasive procedure.

Concomitant active pulmonary TB is observed in approximately 50% of spinal TB cases [9]. However, in our case, the patient did not present with pulmonary symptoms, and the involvement of an extremely rare site was observed. Clinicians find it difficult to initially consider TB infection. In fact, other infectious diseases or neoplasms are more likely suspected.

Therefore, the usefulness of the PCR test is worth emphasizing because its less invasiveness and has a high specificity for TB. PCR was significantly more sensitive than histopathology in detecting TB [10,11]. Histopathology might not accurately diagnose TB in several patients, leading to the recurrence of the disease. As in our case, the patient had an intractable anal fistula 25 years previously, and he underwent several surgeries for the complex anal fistula with the removal of coccyx. Although it was hypothesized, preexisting TB involvement might not be addressed in a previous treatment.

Treatment guidelines for lumbosacral TB are not yet established. According to a recent study, the most currently used chemotherapy regimen is similar to that indicated for pulmonary TB, which includes rifampicin, isoniazid, pyrazinamide, and ethambutol for 2 months, followed by rifampicin and isoniazid for 4 months [12]. However, there is no consensus on the duration of the treatment. In this case, PCR has significantly helped in confirming the appropriate treatment.

Our case was involved spinal TB, which was difficult to distinguish from complex anal fistula. Meanwhile, anoperineal TB is also a rare disease but should still be considered. Anoperineal TB is considered different from normal anal fistula, which developed from crypto-glandular theory. However, due to the similar clinical presentations, anoperineal TB cannot be distinguished from those of crypto-glandular origin. Data have suggested TB as a relevant factor for the recurrence of anal fistula. Therefore, surgeons must be alert in suspecting such cases, which require imaging studies (CT and MRI) and bacilloscopy with appropriate staining and culture, using all collected materials, such as sinus discharge, skin, or excised fistulous tract. However, in negative cases wherein suspicion for the diagnosis persists, a PCR test must be carried out.

4. Conclusion

Isolated sacrococcygeal TB is an extremely rare condition. However, atypical abscess with osteolytic changes must be considered. In our case, the importance of considering TB as a diagnosis when

an unusual site is involved is emphasized. Such presentations of TB must be kept in mind to ensure a prompt and accurate diagnosis.

Conflict of interest

The authors have no conflicts of interest.

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Ethical approval

Ethical approval was not required by our institution and patient identifying knowledge was not presented in the report.

Consent

Written informed consent has been obtained from the patient for the publication of this case report and any accompanying images.

Author contribution

YT participated in treatment of the patient, collected case details, literature search and draft the manuscript. MF, KO, and SK participated in treatment planning of the patient. HN, HE, and HO participated in treatment planning of the patient and helped to draft the manuscript. All authors read and approved the final manuscript.

Registration of research studies

Not applicable.

Guarantor

Yuji Takakura.

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