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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.

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].

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

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

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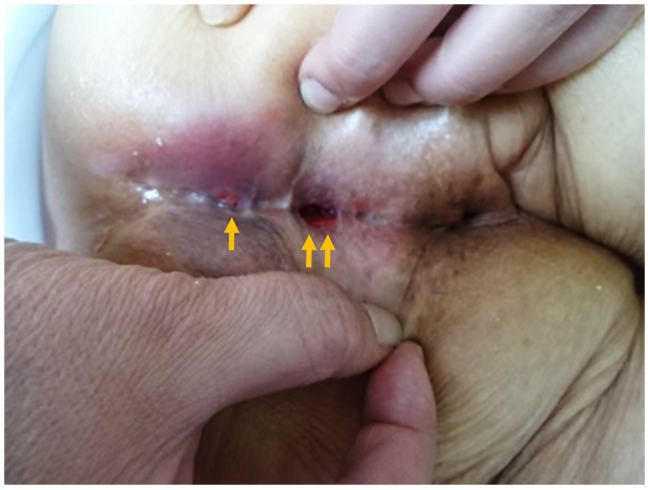

Fig. 1. Peri-anal inspection during initial visit. There were two fistulous orifices between the anus and coccyx (arrows).

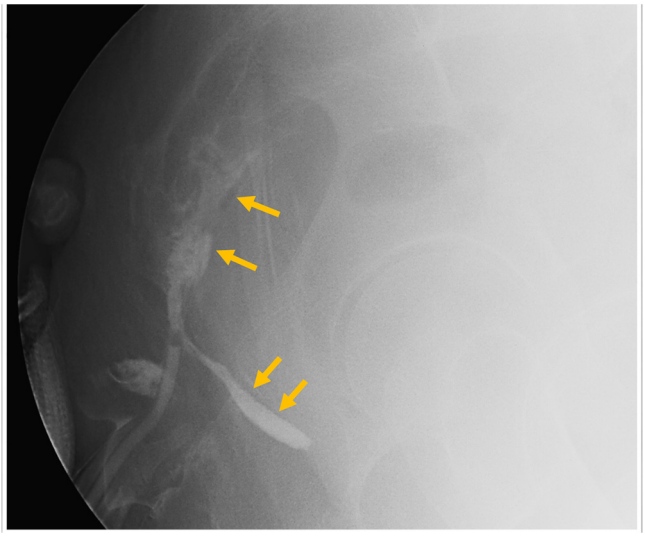

Fig. 2. Fistulography showed a complex supra-elevator track that is connected to the pre-sacral area and another track ending blindly behind the rectum (arrows). No connection between the track and the rectum or anus was identified.

tomography (CT) scan of the whole body reveled 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 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

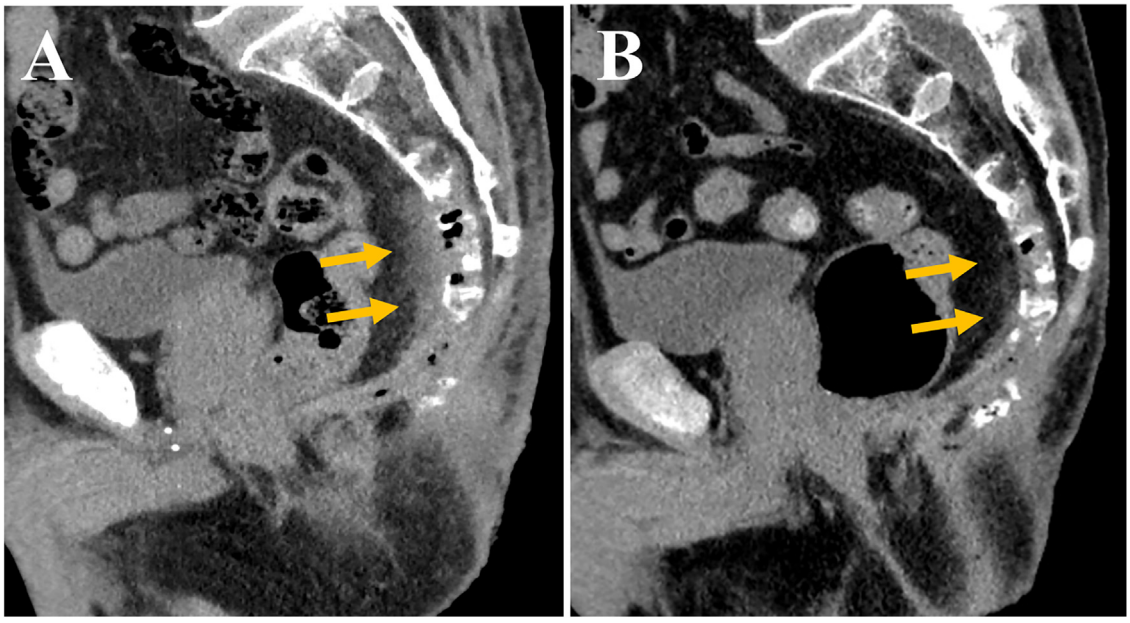

Fig. 3. (A) Sagittal CT image before treatment. CT scan showed a lytic destruction of the sacrum and coccyx, with a low-density area in the pre-sacrococcygeal area, suggesting inflammatory fluid collection. (B) The sagittal CT image after treatment. Fluid collection in front of the sacrum was disappeared.