Tuberculous osteomyelitis of the pubic symphysis – a case report of a rare entity mimicking spondyloarthritis

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Abstract

Tuberculosis (TB) osteomyelitis of the pubic symphysis is an extremely rare diagnosis. Axial spondyloarthritis (SpA) is characterized by inflammatory back pain and enthesitis, and involvement of pubic symphysis is very unusual at presentation.

A 36-year-old female patient with a history of inflammatory back and pubic pain was referred to Rheumatology. She had a pelvic magnetic resonance imaging (MRI) suggestive of osteitis pubis. She was started on etoricoxib 90mg/day as axial spondyloarthritis was suspected, with no improvement. Pelvic MRI was repeated and showed osteomyelitis of the iliopubic branches. An ultrasound-guided biopsy was performed, and culture was positive for *Mycobacterium tuberculosis*. Further imaging studies revealed small cavitations and several centrilobular micronodules with a tree-in-bud pattern in the upper lung lobes and in the upper segment of the lower left lobe. She was started on anti-tuberculous treatment for 1 year and had a good clinical and radiological response.

TB osteomyelitis of the pubic symphysis is a rare entity and has seldom been reported. However, this is the first case, to our knowledge, where the clinical picture mimicked an itself unusual presentation of SpA.

Keywords: Spondyloarthropathies (including psoriatic arthritis); Spondylarthritis; Spinal disorders; Infectious and arthritis; Bacteria.

Introduction

Tuberculosis (TB) is an infectious disease caused by *Mycobacterium tuberculosis*.(1) TB can involve extrapulmonary sites in up to 20% of patients, most often affecting lymphatics, pleura and bone and jointsa[2, 3]. Skeletal TB can present in several ways, most common being spondylitis (Pott disease), comprising almost half of skeletal TB cases, with much rarer forms being TB arthritis and osteomyelitis[3, 4]. Although TB osteomyelitis can occur in any bone, it usually presents in a single bone, like hip, knee and near the ankle[3]. TB osteomyelitis of the pubic symphysis is an extremely rare diagnosis[3]. Diagnosis of TB osteomyelitis is supported by imaging, and radiography, computerized tomography (CT), and magnetic resonance imaging (MRI) are important tools for the diagnosis, even though there are no pathognomonic findings[1, 5]. Thus, the diagnosis is only possible with the identification of the causative organism through biopsy and culture of the lesion[6, 7]. Treatment is based on antimicrobial therapy as used for pulmonary TB, usually for 6 to 9 months[8] but surgery may be needed in some instances[9].

Axial spondyloarthritis (SpA) is a group of diseases characterized by inflammatory back pain and enthesitis with a diverse clinical presentation, classically affecting sacroiliac joints and spine, but peripheral joints as well[10, 11]. Even though it is a recognized site of possible inflammation in SpA, the involvement of pubic symphysis is usally a late onset manifestation, being associated with male gender, older age and elevation of inflammatory markers[10, 11]. Classification criteria by Assessment of Spondyloarthritis International Society (ASAS) are a valuable aid in the diagnosis of SpA[12]. Imaging plays a very important role, with spine and sacroiliac radiography showing typical changes, and MRI being able to demonstrate pre-radiographic changes[12].

We report the case of a young woman with pelvic pain suggestive of an unusual presentation of SpA, which in turn mimicked a very rare manifestation of TB.

Case Report

We report a case of a 36-year-old female patient who was referred to the General Rheumatology Clinic due to inflammatory back and pubic pain, to evaluate the possible diagnosis of SpA. The patient presented with a 3-month history of inflammatory back pain and pubic pain, with significant morning stiffness lasting for more than 30 minutes. There was no arthralgia or enthesitis, gastrointestinal symptoms, uveitis, previous infectious episodes, personal or family history of psoriasis or inflammatory bowel disease, or family history of

inflammatory rheumatic disease. She also denied fever, anorexia, weight loss, cough, or genitourinary symptoms. On physical examination, Schober test was 4 cm, palpation of the pubic symphysis was frankly painful, there was discomfort with adduction of the thighs against resistance, but sacroiliac joint palpation was painless, and Patrick and Volkmann's maneuvers were negative; there was no peripheral arthritis, joint mobility was preserved and painless, and no skin lesions were seen. She had already undergone a pelvic magnetic resonance imaging (MRI), with the report showing a significant T1-weighted signal change on both sides of the pubic symphysis, but especially on the left with irregularity of the cortical bone and bone marrow edema, suggestive of osteitis pubis. Due to the presence of inflammatory back and pubic pain lasting more than 3 months with onset at an age younger than 45 years-old, mildly limited spinal mobility and osteitis pubis depicted in the MRI, axial spondyloarthtritis was suspected and a trial of a non-steroidal anti-inflammatory drug (NSAID, etoricoxib 90 mg/day) was started, with only slight improvement of the clinical picture. Laboratory tests were requested and revealed raised erythrocyte sedimentation rate (ESR, 71 mm/1st h) and C-reactive protein (CRP, 25 mg/L), and a mild leukocytosis (12.200/µL) and thrombocytosis (434.000/µL); HLA-B27 antigen was negative. Sacroiliac joint MRI was normal, without any changes suggestive of sacroiliitis.

NSAID was changed to acemetacin 210 mg/day. Laboratory tests were repeated, showing further elevated inflammatory markers, with ESR 86 mm/1st h and CRP 80.4mg/L. Pelvic radiography was repeated and showed a slight enlargement of the pubic symphysis with bone irregularities (Figure 1). A pubic symphysis ultrasound-guided biopsy was performed – while cultural results and direct mycobacterial results were negative, culture for *Mycobacterium* revealed *Mycobacterium tuberculosis*, establishing the diagnosis of TB osteomyelitis.

Pelvic MRI was repeated and showed a clear bone marrow alteration of the iliopubic branches, with edema, inflammatory changes, loss of T1-weighted signal, and clear heterogeneity of the pubic symphysis, with an inflammatory collection lateralized to the left, measuring 34x15mm, very suggestive of osteomyelitis of the iliopubic branches, with epicenter in the pubic symphysis (Figure 2).

She was admitted to the Rheumatology inpatient department and was started on ethambutol 1200 mg/day, isoniazid 300 mg/day, rifampicin 600 mg/day, and pyrazinamide 1500 mg/day. Sputum collection was performed and was positive for *Mycobacterium tuberculosis*. A computerized tomography (CT) scan of the spine was normal; a thoraco-abdomino-pelvic scan showed opacities in both lung apices, with small cavitations and several centrilobular micronodules with a "tree-in-bud" pattern in the upper lobes and in the upper segment of the lower left lobe, very suggestive of pulmonary TB (Figure 3).

Diagnosis of disseminated TB (pulmonary and bone) was made. She was transferred to the Infectious Diseases inpatient department but was soon discharged to continue treatment as an outpatient, after being evaluated by Orthopedic Surgeons that decided to continue the anti-tuberculous treatment only. During the next few months, she underwent surgery, with curettage and extraction of a small fragment (2 cm) of bone sequestration in the right iliopubic branch (Figure 4a). She was on anti-tuberculous treatment for 1 year and had a good clinical and radiological response, without the need for additional orthopedic procedures (Figure 4b).

Discussion

TB can be classified according to the sites involved in pulmonary and extrapulmonary TB. TB osteomyelitis of the pubic symphysis is a rare entity and has seldom been reported, especially in the era of effective anti-tubercular therapy[3]. However, this is the first case, to our knowledge, where the clinical picture mimicked an itself unusual presentation of SpA, since the patient presented as well with inflammatory back pain, which turned out not to be involved by the infectious process, according to the imaging studies carried out. Other case reports have been published with TB mimicking SpA, but usually with involvement of the sacroiliac joints, which is far more common in SpA patients[13, 14]. Nonetheless, despite being a rare form of clinical presentation, imaging studies have found that symphysis pubis involvement during disease course can affect 47% of patients[11]. Interestingly, some studies have reported higher inflammatory markers in SpA patients where pubic symphysis is involved and active lesions where seen, and these patients are usually older[10]. Osteitis pubis is a common process seen in this anatomical area, but is a usually self-limiting process seen after trauma, pregnancy or in sports athletes[3], which is not the case in this patient, and tends to respond to NSAIDs[3]. Former reported cases of TB osteomyelitis of the pubic symphysis show that surgery may be needed in about half of the situations[3] as it was seen with this patient.

In conclusion, the authors highlight that there is a need to properly evaluate patients who present with inflammatory pubic and back pain, especially when there are high inflammatory markers and little or no response to NSAIDs is seen. In this case, the patient had an unusual presentation compatible with SpA, but a thorough evaluation, with repeated MRI and tissue biopsy made the diagnosis of a rare form of extrapulmonary TB.

Figures

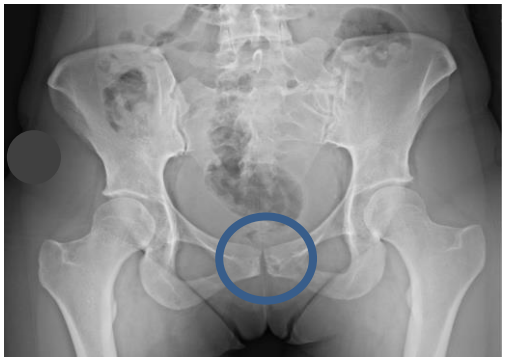

Figure 1. Pelvic radiography showing slight enlargement of the pubic symphysis with bone irregularities.

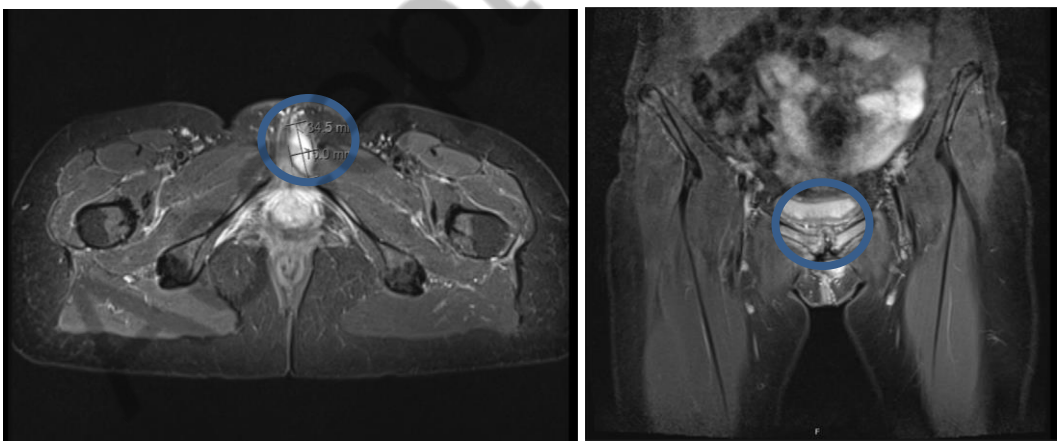

Figure 2. T1-weighted pelvic MRI showing pubic symphysis and an inflammatory collection of the left iliopubic branch.

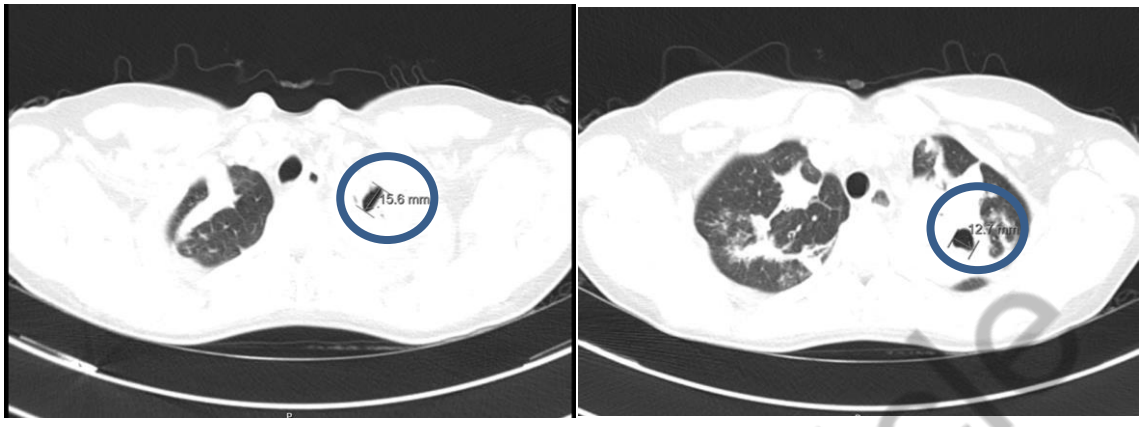

Figure 3. Chest CT scan showing consolidations with cavitations in the upper left lobe suggestive of pulmonary tuberculosis.

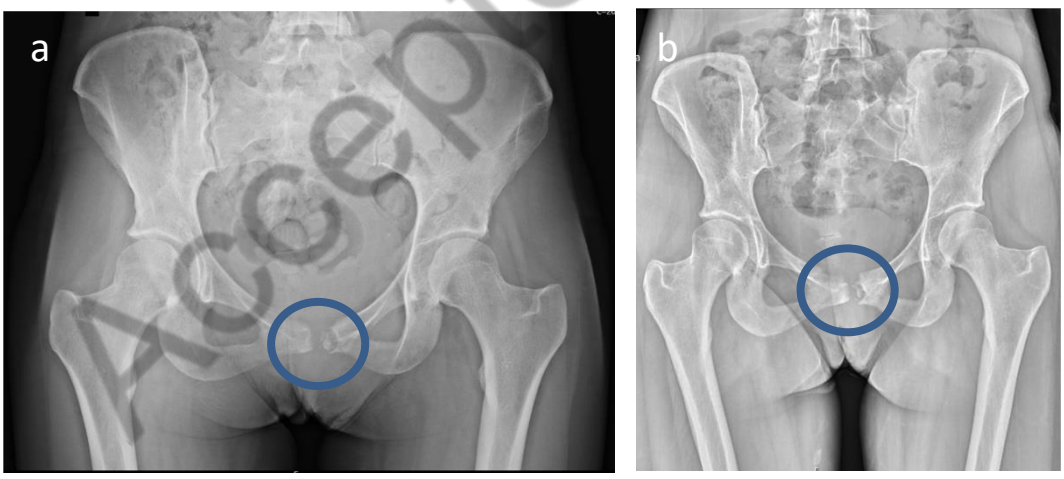

Figure 4. Pelvic radiography after removal of bone fragment (a) and 1 year after diagnosis (b).

References

1. Marais S, Roos I, Mitha A, Mabusha SJ, Patel V, Bhigjee AI. Spinal Tuberculosis: Clinicoradiological Findings in 274 Patients. Clin Infect Dis. 2018;67(1):89-98.

2. Peto HM, Pratt RH, Harrington TA, LoBue PA, Armstrong LR. Epidemiology of extrapulmonary tuberculosis in the United States, 1993-2006. Clin Infect Dis. 2009;49(9):1350-7.

3. Gothwal S, Varshney P, Mathur S, Songra B. Tuberculosis of the pubic symphysis. BMJ Case Rep. 2014;2014.

4. Vohra R, Kang HS, Dogra S, Saggar RR, Sharma R. Tuberculous osteomyelitis. J Bone Joint Surg Br. 1997;79(4):562-6.

5. Shanley DJ. Tuberculosis of the spine: imaging features. AJR Am J Roentgenol. 1995;164(3):659-64.

6. Merino P, Candel FJ, Gestoso I, Baos E, Picazo J. Microbiological diagnosis of spinal tuberculosis. Int Orthop. 2012;36(2):233-8.

7. Colmenero JD, Ruiz-Mesa JD, Sanjuan-Jimenez R, Sobrino B, Morata P. Establishing the diagnosis of tuberculous vertebral osteomyelitis. Eur Spine J. 2013;22 Suppl 4(Suppl 4):579-86.

8. Nahid P, Dorman SE, Alipanah N, Barry PM, Brozek JL, Cattamanchi A, et al. Official American Thoracic Society/Centers for Disease Control and Prevention/Infectious Diseases Society of America Clinical Practice Guidelines: Treatment of Drug-Susceptible Tuberculosis. Clin Infect Dis. 2016;63(7):e147-e95.

9. Jutte PC, Van Loenhout-Rooyackers JH. Routine surgery in addition to chemotherapy for treating spinal tuberculosis. Cochrane Database Syst Rev. 2006(1):Cd004532.

10. Kang Y, Ahn JM, Lee E, Lee JW, Kang HS. Active inflammatory changes around the pubic symphysis in patients with axial spondyloarthritis: Magnetic resonance imaging characteristics and association with clinical factors. Eur J Radiol. 2020;124:108802.

11. Koo BS, Song Y, Joo KB, Lee S, Kim TH. Radiologic Changes in the Symphysis Pubis of Male Patients with Ankylosing Spondylitis. J Rheumatol. 2016;43(2):330-4.

12. Mandl P, Navarro-Compán V, Terslev L, Aegerter P, van der Heijde D, D'Agostino MA, et al. EULAR recommendations for the use of imaging in the diagnosis and management of spondyloarthritis in clinical practice. Ann Rheum Dis. 2015;74(7):1327-39.

13. Huyge V, Goldman S, Soyfoo MS. Disseminated tuberculosis mimicking ankylosing spondylitis. Case Rep Rheumatol. 2011;2011:195085.

14. Prakash J. Sacroiliac tuberculosis - A neglected differential in refractory low back pain - Our series of 35 patients. J Clin Orthop Trauma. 2014;5(3):146-53.