CASE REPORT

A persistent case of tuberculosis of the pubic symphysis and pubic bone

Ulrike Dapunt,¹ Alexander Mischnik,² Stephan Goeppinger,³ Burkhard Lehner¹

¹Department of Orthopaedics and Trauma Surgery, Heidelberg University, Heidelberg, Germany

²Department of Infectious Diseases, Medical Microbiology and Hygiene, Heidelberg University, Heidelberg, Germany

³Department of Pathology, Heidelberg University, Heidelberg, Germany

Correspondence to

Dr Ulrike Dapunt,
ulrike.dapunt@med.uni-heidelberg.de

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SUMMARY

A case of a patient suffering from tuberculosis of the pubic symphysis and pubic bone is presented. There were no symptoms other than pain in the right groin area for 12 months. An X-ray of the pelvis showed an osteodestructive lesion of the pubic bone, and an MRI revealed an abscess formation of the pubic symphysis. Tissue samples were collected via CT-guided needle biopsy. Histological evaluation of tissue and analysis by PCR prompted the diagnosis of musculoskeletal tuberculosis. Despite antituberculous chemotherapy according to the current guidelines, the osteodestructive lesion progressed. This case highlights the difficulty of treating bone infections in general. Moreover, *Mycobacterium tuberculosis* as a rare causative agent of bone infections is discussed.

Figure 1 An X-ray of the pelvis showing an osteodestructive lesion of the pubic bone.

BACKGROUND

Although there has been a decrease of incident cases since the 1990s and the treatment success rate has improved, tuberculosis (TB) remains a major global health issue particularly with regard to the high incidence of coinfection in HIV-positive people and the emergence of multidrug-resistant strains of *Mycobacterium tuberculosis*. Since musculoskeletal TB is rare, difficult to diagnose and requires an extensive antibiotic therapy, failures of treatment occur easily. Therefore, we would like to draw attention to the clinical course of a rare case of TB of the pubic symphysis and pubic bone.

CASE PRESENTATION

A patient in his mid-70s presented to his general practitioner and reported the presence of pain in his right groin area on weightbearing during the past 12 months. There was no hyperthermia, swelling or erythema detectable and the patient reported pain on 30° abduction in both hip joints. There was no history of trauma, no signs of an inguinal hernia and the patient did not report fever, night sweats, weight loss or cough in the past months. The laboratory diagnoses were normal except for a mildly elevated C reactive protein concentration (13.9 mg/L). No history of illnesses except for scarlet fever in his childhood years was reported. Besides a prostate hyperplasia, the patient was completely healthy and denied any trips overseas.

An X-ray of the pelvis showed a diffuse osteodestructive lesion of the pubic bone (figure 1) and a contrast-enhanced MRI revealed fluid collection with an enhancing rim in the pubic symphysis. The patient was admitted to the orthopaedic

department and a CT-guided needle biopsy was performed under the suspicion of an infectious genesis of the osteolytic lesion. Samples were sent to the microbiology department and pathology department for evaluation. The cultures revealed *Staphylococcus lugdunensis* and the patient was started on an antibiotic treatment with cefuroxime and rifampicin according to the susceptibility testing and because staphylococcal osteitis was suspected. The histological results revealed a caseating granulomatous inflammation which was indicative of TB of the bone (figure 2). A Ziehl-Neelsen stain tested negative for acid-fast bacilli, but PCR was positive for *M. tuberculosis*. This prompted the diagnosis of musculoskeletal TB. Following this diagnosis, the patient was referred to the pulmonology department to rule out concurrent pulmonary TB. The tuberculin skin test was positive, but bronchoscopy, cultures of sputum and imaging of the lungs were negative for TB. An examination of urine and kidneys to rule out genitourinary TB was not performed.

TREATMENT

The patient was started on an antituberculous chemotherapy with isoniazid, rifampicin, ethambutol and pyrazinamide for 3 months. Antibiotic treatment was continued for another 6 months with isoniazid and rifampicin. After completing the antibiotic therapy the patient's symptoms slowly recurred. An X-ray of the pelvis showed an increase of the osteodestructive lesion in the pubic bone which further progressed in the following 3 months (figure 3). An MRI of the pelvis showed unaltered fluid collection with contrast rim enhancement in and around the pubic symphysis (figure 4).

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Figure 2 Histological analysis of tissue samples revealing caseating granulomas.

OUTCOME AND FOLLOW-UP

Because of the progressive osteodestructive lesion in the pubic bone, the patient is now possibly awaiting surgical intervention to prevent further bone degradation.

DISCUSSION

Osteomyelitis, in general, is difficult to treat and cases of persisting bone infections are well described. The treatment is even more problematic when the infection is associated with a foreign body, that is, an implant. Therefore, bone infections remain one of the biggest challenges in the field of orthopaedic surgery. Staphylococci species are prevalent in bone infections, but other species and also multispecies infections can occur.¹⁻⁴

TB of the bone, which is most commonly caused by the bacillus *M tuberculosis*, is a rare form of osteomyelitis. In the majority of cases an infection occurs in the lungs, but also other sites (extrapulmonary TB) such as the musculoskeletal system can be affected. In general, more men than women, predominantly in the economically productive age groups are affected by this disease.⁵⁻⁶ The probability of developing TB is much higher among immunosuppressed people, for example, in HIV-infected population, and therefore TB remains a major global health problem. According to the latest estimates published by the WHO, there were 8.6 million new TB cases in 2012 and 1.3

million TB deaths (including 320 000 deaths among HIV-positive people).⁶⁻⁷ After a resurgence of cases of TB in the mid-1990s, the WHO introduced a 20-year programme to reduce incidence, prevalence and mortality of TB worldwide by 2015. So far, there has been a 45% reduction in mortality rate globally and the treatment success rate was improved from 69% to 87%.⁶ Clearly, the number of TB infected people varies widely among countries and the lowest numbers are found in high-income countries such as Western Europe. For example, in Germany specifically, the incidence of TB has been steadily decreasing between 1990 and 2012 from 21 to 5.6 cases per 100 000 population.⁶

However, since TB is a rare form of osteomyelitis, the possibility of a mycobacterial infection is often overseen and the diagnosis is often delayed by up to 16–18 months.⁸ Moreover, diagnosing TB can be tedious and time-consuming. Smear microscopy is still one of the most rapid and most inexpensive ways to diagnose TB. Normally, the predictive value for *M tuberculosis* in expectorated sputum is >90%.⁹ However, the reliability is highly dependent not only on the technician's experience, but also on the number of acid-fast bacilli present in the sample. Only 60% of the smears are positive if 10⁴ acid-fast bacilli/mL are present.¹⁰ For cultures, the number of mycobacteria required for diagnosis is usually lower than by staining with Ziehl-Neelsen or Kinyoun. Culture can detect as few as 10–100 viable organisms/mL of specimen and is also more sensitive than PCR.¹¹ The disadvantage of mycobacterial culture is that it has to be held for 6–8 weeks before being discarded as negative. In the present case, there was no initial suspicion of mycobacterial infection, and therefore no samples were cultured for mycobacteria. Mycobacteria could not be grown and susceptibility testing was not possible. The cultures tested positive for *S lugdunensis* and the patient was treated accordingly for staphylococcal-induced osteitis. Several reports have been published on the possibility of another infectious agent masking musculoskeletal TB and therefore mycobacteria have to be kept in mind.¹²⁻¹³ However, the histological evidence of caseating granulomas and a positive TB-PCR which was performed in the pathology department were sufficient to make the diagnosis. This further underlines that it is mandatory to send in a biopsy for histological analysis.^{5-6 14-16}

Musculoskeletal TB is reported to account for 1–3% of all forms of TB and the spine is mostly affected.⁵⁻¹⁴ TB of the symphysis and pubic bone, as in our case, is very rare, though it

Figure 3 After cessation of antituberculous chemotherapy, the osteodestructive lesion further progressed.

Figure 4 Contrast-enhanced MRI of the pelvis showing fluid collection with an enhancing rim of the pubic symphysis.

has been previously reported in the literature.^{13 17 18} Clinical symptoms of musculoskeletal TB are often unspecific.⁸ Most patients report pain in the affected area, swelling of the joint and a limited range of motion. A cold abscess is highly suggestive of TB.^{5 14 15} Systemic symptoms include weight loss, night sweats and fever, but there have been reports that 31.5% of patients with musculoskeletal TB have no systemic symptoms.¹⁴ Because of the frequent association of musculoskeletal TB with pulmonary or genitourinary TB, cultures of sputum and urine should be performed.^{5 6 14}

Currently, the German Central Committee against TB and the WHO recommend a 2-month regimen of four first-line drugs (isoniazid, rifampicin, ethambutol and pyrazinamide) followed by a 7-month treatment with isoniazid and rifampicin for susceptible cases of musculoskeletal TB.^{6 5 19} In our case, the patient received a 3-month course of four first-line drugs, followed by 6 months of isoniazid and rifampicin treatment, which should have been sufficient according to the latest guidelines. However, there have been other published articles, according to which a prolonged antibiotic treatment for 12–18 months of TB cases with an osseous involvement has been recommended.⁵

Patients have to be closely monitored for adverse side effects of the antibiotic treatment. Ethambutol can cause retrobulbar neuritis, and visual acuity tests should be performed before and during the antituberculous chemotherapy. Hepatotoxicity is a major concern of treatment with isoniazid and attention should be paid to existence of hepatitis or excessive alcohol consumption. Rifampicin can cause gastrointestinal upset and a mild jaundice.

When treatment is initiated early and correctly, the majority of cases of musculoskeletal TB can be successfully treated with antibiotics alone. A surgical intervention is usually only necessary to drain large abscesses or to obtain tissue samples. When the osseous involvement of the infection is severe and compromises stability, surgical debridement needs to be performed. Implanting osteosynthetic material should be avoided,

if possible, for a minimum of 3 months.⁵ There have been reports, that in cases of articular TB, at least 3 months of antituberculous chemotherapy should be applied before performing arthroplasty and a prolonged antibiotic treatment should follow arthroplasty, although few authors suggest waiting up to 12 months prior to joint replacement.^{5 20 21}

In conclusion, this case highlights the difficulty of treating bone infections in general and the possibility of rare causative pathogens, such as *M tuberculosis*. When a mycobacterial infection is suspected, specimens should be sent in for histological analysis and for culture to confirm species and perform susceptibility testing.

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Learning points

- ▶ Musculoskeletal tuberculosis is a rare disease and is therefore often misdiagnosed.
- ▶ If available, diagnostic methods should always include standard cultures, staining for acid-fast bacilli, histological evaluation and PCR analysis to increase the chances of detecting mycobacteria or mycobacteria-induced inflammatory response.
- ▶ Because of the association of musculoskeletal tuberculosis with pulmonary or genitourinary tuberculosis, sputum and urine samples should also be evaluated.
- ▶ In cases of persisting joint and bone infections with an identified causative agent, the possibility of a coincidental mycobacterial infection needs to be considered and diagnostic tests should be conducted accordingly.
- ▶ When treated adequately, a surgical intervention is usually not required in musculoskeletal tuberculosis. Surgery only needs to be performed when draining of large abscesses is necessary, bone damage is extensive or stability is at risk.

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