



EXTRA PULMONARY TUBERCULOSIS IN THE ILIUM - CASE REPORT AND REVIEW OF THE LITERATURE

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Article Info	ABSTRACT
<p>Received 23/03/2016 Revised 27/03/2016 Accepted 15/04/2016</p>	<p>A rare case of extra pulmonary tuberculosis located in the ilium is presented. The incidence of tuberculosis in the Western European countries is decreasing. However, this decline is stagnating. This is probably due to a changing demography, increased intercontinental travelling possibilities and increasing immigrant flow to the Western European countries. The presented case was managed with multi-drug anti-tuberculosis therapy and several surgical interventions. When tuberculosis is diagnosed in an advanced stage abscess formation and cortical destruction are often present. These findings are related with a poor clinical outcome. The management of isolated osseous tuberculosis should be antibiotics with or without surgical interventions, but surgery can never be a substitute for medicinal therapy. Osseous tuberculosis should still be in every differential diagnosis when a patient, in particular an immigrant, has pain in the hip.</p>
<p>Key words: Osseous, Multidrug anti-tuberculosis therapy, Surgery.</p>	

INTRODUCTION

The incidence of tuberculosis (TB) is increasing worldwide, however the incidence in developed countries is low and decreasing [1,2]. In the Netherlands TB mainly affects first generation immigrants, which account for 73% of newly diagnosed infections. Somali immigrants show the highest incidence [1-3]. In newly diagnosed TB the manifestation of pulmonary and extra pulmonary TB is equal [1,4]. Osseous TB accounts for about 1 to 3% of extra pulmonary TB and usually affects the spine, knee or hip [5,6]. Clinical presentation of extra pulmonary TB is varied and related to its location. Osseous TB is usually secondary to a primary focus which can be detected in about 50% of the patients [7]. When the disease is diagnosed in an advanced stage abscess formation and cortical destruction are present more often. These findings are related with poor clinical outcome. TB is not often included in the differential diagnosis for an immunocompetent patient with hip or pelvic pain.

Moreover, TB in the ilium has only been reported a few times in current literature [8]. We report a case of an immunocompetent Somalian immigrant diagnosed with TB in the ilium.

CASE REPORT

A 27-year old male visited our outpatient clinic with pain in his right hip. The pain had an insidious onset and started eight months ago. A weight loss of nine kilograms was noticed in the last eight months and there was a progressive painful swelling in his groin region. Night sweats or fever did not occur. No other joints were involved. NSAIDs relieved symptoms temporarily. Seven months prior to presentation the patient migrated to the Netherlands from Somalia. In Somalia he never underwent surgery. Medical history was blank and HIV tests were negative. Physical examination showed a deviant gait pattern and weight bearing aggravated complaints. Internal



and external rotation of the hip as well as flexion and extension was painful. Rotations were restricted, but flexion and extension were not. Basic neurological examination was normal. Laboratory testing showed increased inflammatory parameters: C-reactive protein (CRP), 65; erythrocyte sedimentation rate (ESR), 92; white blood cell count (WBC), 5.8. Chest and spine x-rays were normal and a conventional pelvic x-ray revealed a large radiolucent lesion in the right iliac bone. (Fig. 1).

Osseous TB is often secondary to another primary focus; therefore, skeletal scintigraphy was performed. It showed an increased uptake in the right pelvic region suggesting pathology, however no other skeletal lesions were detected. (Fig. 2).

Additional Magnetic Resonance (MR) imaging showed cortical destruction, abscess formation and a large soft-tissue mass. MR imaging did show an involvement of the hip joint. (Fig. 3).

The patient underwent three surgical procedures in four weeks. During the first surgical procedure a straight lateral approach (Hardinge approach) was used. When opening the fascia lata a large amount of purulent discharge was seen. The lesion was estimated at the size of a tennis ball and was surrounded with multiple abscesses. The lesions were extensively rinsed. After rinsing, the cavity was inspected and it extended from the fascia lata to the joint capsule. However, the joint capsule was intact and was left untouched. The surrounding soft tissue as well as the bony lesion in pelvic bone (ilium) was debrided. Samples of the debris and fluid were collected for microbiological and histopathological examination including Polymerase Chain Reaction (PCR), Ziehl-Neelsen (ZN) stain and culture on Mycobacterium tuberculosis. Empirically, multiple beads impregnated with gentamicin were placed into the bony lesion and the surrounding soft tissue. Postoperatively empirical antibiotics were started (Amoxicillin/ Clavulanic acid 1000/200mg four times a day intravenously). Histopathological examination showed granulomatous inflammation accompanied by necrotic tissue. This result increased the likelihood the lesions are caused by M.tuberculosis, therefore after two days treatment was adjusted to a multi-drug anti-tuberculosis therapy. The infection parameters decreased but were still elevated at

the time of the second surgical procedure ten days after the first. In this procedure the same incision and approach was used and again a large amount of purulent discharge was seen, although it was less than in the first procedure. The bony lesion was situated in the ilium and involved the acetabulum but there still was a thin layer of bone in the acetabular roof. The lesions were extensively rinsed again. The gentamicin beads were replaced because the diagnosis of tuberculosis was not confirmed at the time of the second surgical procedure. Twenty-six days after the first procedure a third procedure was performed. By the time of the third procedure the infection parameters returned to normal and the patient only had some minor complaints by weight bearing. Again the same incision and approach was used. In this procedure there was not any discharge apparent. The bony lesion in the ilium was rinsed again and the hip joint still was not affected.

PCR detected M.tuberculosis. After 19 days cultures were also positive for M.tuberculosis, specie that was sensitive to all used antibiotics. The ZN stain remained negative. The National Institute of Health and Environment (RIVM) confirmed the diagnosis and antibiotic treatment was consisted of a multi-drug anti-tuberculosis therapy. A combination of rifampicin, isoniazid, ethambutol and pyrazinamide were taken for 2 months and rifampicin and isoniazid were continued for in total nine months. After a 30-day hospital stay the patient was discharged. The Dutch Municipal Health Services (GGD) was contacted to observe and perform a Directly Observed Treatment (DOT) during the following nine months to increase the compliance [9].

After completion of the treatment our patient was seen again at the outpatient clinic. He still complained of some minor pain at night and weight bearing and walking caused pain. Physical examination revealed a persisting deviant gait pattern and a flexion contracture of 10 degrees. Internal and external rotation was painless and unrestricted. The swelling in his groin region was diminished and palpation was not painful. Infection parameters were normalized: CRP, 4; ESR, 4; WBC 4.4. Conventional x-rays and MR images of the pelvis showed a radiolucent lesion that was reduced in size as compared to images 9 months earlier. (Fig. 4 and 5).

Table 1. Published literature on extrapulmonary tuberculosis located in the ilium

Referentie	Number of patients	Age (in years)	Duration of symptoms (in months)	Therapy	Duration of therapy (in months)	Outcome
Trikha et al 2005 ⁵	2	10 and 25	2 and 3	Surgical procedures and multi-drug anti-tuberculosis therapy	15	Full recovery
Mogha et al 2013 ⁷	1	42	3	Surgical procedures and multi-drug anti-tuberculosis therapy	15	Full recovery
Chaudhary et al 2005 ¹³	1	35	2	dressings and antituberculous	NA	NA



				chemotherapy and drainage		
Dhingra et al 2008⁸	1	20	NA	multi-drug anti-tuberculosis therapy	22	Full recovery
Current report	1	27	8	Surgical procedures and multi-drug anti-tuberculosis therapy	9	Flexion contracture and night pain

Fig 1. Anteroposterior x-ray of the pelvis shows a lytic region in the right iliac bone.



Fig 2. Anteroposterior skeletal scintigraphy image showing the increased activity in the right pelvic region



Fig 3. Anteroposterior T2 TIRM MR image of the pelvis showing cortical destruction (*), abscess formation (**), and a large soft-tissue mass in the right hemi pelvis (***)

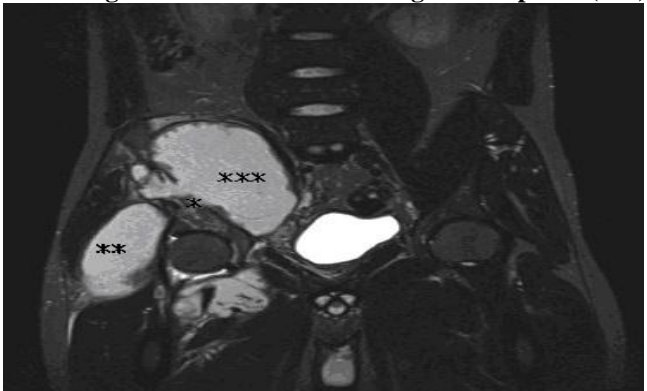
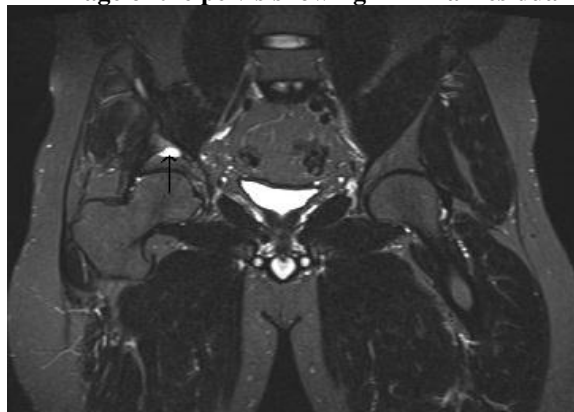


Fig 4. Anteroposterior x-ray of the pelvis, nine months postoperatively, shows a residual radiolucent lesion of the right pelvis (arrow)



Fig 5. Anteroposterior T2 TIRM MR image of the pelvis showing minimal residual abscess formation (arrow)



DISCUSSION

An extensive search of three relevant electronic databases (Pubmed, Web of Science and Google Scholar) was performed with search terms including; tuberculosis, osseous/ skeletal, ilium/ pelvis/ iliac and immunocompetent. In 1892 extra pulmonary tuberculosis located in the ilium in an immunocompetent patient was reported for the first time. Since then only few case reports have been published in medical literature [8].

Presentation and differential diagnosis

The clinical presentation of extra pulmonary TB is often atypical and shows a wide variation in presentation in correlation with its localization [7]. This review of the literature includes four case reports in which five patients are presented with tuberculosis located in the ilium. All patients had at time of presentation complaints at the hip and weight bearing was painful in all patients. These patients had small lesions with a maximum size of 40 mm; these lesions existed only for a shorter period (3-4 months). When looking at the differential diagnosis in an immunocompetent patient with this presentation it should include malignancies such as chondroblastoma or sarcoma, Brodie's abscess or chronic osteomyelitis and the Hand-Schüller-Christian syndrome (including lytic bone lesions, exophthalmos and diabetes insipidus) which is part of the Langerhans cell histiocytosis spectrum. Cultures and histopathological tests should differentiate between these above mentioned diagnosis [10].

Diagnostics

Diagnosing extra pulmonary TB can be challenging, particularly when a patient has such an atypical localisation and presentation. The golden standard for diagnosing TB is culturing of *M.tuberculosis* from taken samples. The Mantoux test and the tuberculin test often show false-negative results, especially in the early stages of the disease. In patients with extra pulmonary TB the ZN stains are positive in approximately 64%. The additional value of PCR has to be determined [11,12]. Of the available additional imaging techniques, MR imaging is the preferred tool because of its ability to evaluate abscess formation and soft-tissue masses and it is able to estimate the degree of cortical destruction. Diagnosing osseous TB at an early stage is critical for the clinical outcome. In a more chronic stage joint contractures and limited clinical improvement are more likely to occur. Abscess formation is associated with a longstanding lesion. When a joint is affected the joint is prone to develop a secondary osteoarthritis [12].

Treatment

Of the included case reports, four patients were treated with a combination of surgical procedures and multi-drug anti-tuberculosis therapy [5,8,13]. Only one

patient was treated with multi-drug anti-tuberculosis therapy solely [14]. (Table 1) The patients in the included case reports had a much less extensive and longstanding lesion than our patient. In the presented cases the patients had an elevated ESR and no other abnormalities in their laboratory tests. On the contrary our patient also had elevated CRP and had a leucocytosis. In three patients the duration of the medical therapy was longer as can be seen in table 1. In one case report it is not clear what medication was prescribed [5]. It is unclear whether abscess formation was present and if they had performed one or more surgical procedures. All patients had a symptom free recovery after completion of the therapy. It was unclear in the mentioned literature in which country the patients were born and if they migrated prior to their presentation.

CONCLUSIONS

Treatment should involve multi-drug anti-tuberculosis therapy whether or not in combination with surgical procedure [2,3]. The optimal duration of therapy has not been clarified yet [7]. Most practitioners favour a prolonged course for at least a period of nine months. The antibiotic therapy can be carried out with or without surgical procedures, but surgery can never substitute the drug treatment. To optimize the compliance patients should be observed frequently by a Municipal Health Services, using DOT. This also minimalizes the risk developing a multi-drug resistant TB [4,7-9].

A physician should still be aware for TB in patients, mainly immigrants, with atypical hip pain. Tuberculosis should always be included in the differential diagnosis. This case demonstrates that if the disease is not diagnosed at an early stage, and signs of cortical destruction and abscess formation are apparent, a symptom free recovery is hard to achieve. Therefore, it is essential to diagnose it at an early stage and start the treatment immediately and aggressively [1,2].

ACKNOWLEDGEMENT

None

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

ETHICAL APPROVAL

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards."

INFORMED CONSENT

Informed consent was obtained from all individual participants included in the study



REFERENCES

1. <http://www.rivm.nl/cib/infectieziekten/>,
2. <http://www.nationaalkompas.nl>
3. <http://www.kncvtbsurvey.nl/>, last visited on: 06-10-2015
4. Beek LAM, Werf MJ, Richter C, Borgdorff MW. (2006). Extrapulmonary Tuberculosis by Nationality, the Netherlands, 1993-2001. *Emerging Infectious Diseases Journal*, 12, 1375-1382.
5. Trikha V, Varshney MK, Rastogh S. (2005). Tuberculosis of the ilium: is it really so rare? *Acta Orthopaedica Belgica*, 71, 366-368.
6. Vanhoenacker FM, Sanghvi DA, Backer AI. (2009). Imaging features of extraaxial musculoskeletal tuberculosis. *Indian Journal of Radiological Imaging*, 19(3), 176-186.
7. Murtza C. (2005). Study of non-spinal, extra-articular, osseous tuberculosis, its modes of presentation, diagnostic pitfalls and treatment. *Biomedica*, 21(2), 1-5.
8. Mogha A, Verma S, Dhingra M, Kushwaha N, Gupta O. (2013). Isolated Left Ilium Bone Tuberculosis: A Case Report. *The Internet Journal of Orthopedic Surgery*, 8(2), 200.
9. <http://www.who.int/gtb/dots/>.
10. Haider AM. (2007). Bones and Joints Tuberculosis. *Bahrain Medical Bulletin*, 29(1), 1-9.
11. World Health Organisation. (2003). Treatment of tuberculosis; Guidelines for national programme, 313.
12. Centers for Disease Control and Prevention. (2003). Treatment of Tuberculosis, American Thoracic Society, CDC, and Infectious Diseases Society of America, 52-68.
13. Chaudhary IA, Mallhi S, Mallhi AA. (2005). An unusual presentation of Tuberculosis of Iliac Bone. *Pakistani Journal Medical Science*, 21(4), 489-490.
14. Dhingra VK, Rajpal S, Mittal A, Gupta UA, Arora VK. (2008). Tuberculosis of the ilium: a case report. *The Journal of Communicable Diseases*, 40(1), 79-82.

