

**Multifocal Tuberculosis with Rare Cervical and Navicular Bone Localization**

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**Abstract:** Tuberculosis (TB) is a public health problem in Morocco; multifocal disease is a serious form usually affecting immunocompromised patients who already have pulmonary localization. However, it can affect immunocompetent individuals outside of any pulmonary localization. While the association of ganglionic and osteoarticular tuberculosis is infrequent, Tuberculosis of navicular bone is a rare entity. It is important to recognize skeletal tuberculosis at an early stage, as early treatment can effectively eliminate long-term morbidity. The clinical symptoms are often insidious, the diagnosis can be guided by radiographic imaging, the bacteriological and histological examination of the tissue allows the isolation of the pathogen, the identification of the bacillus and the sensitivity of the strain to antibacillary treatment. We report the case of an immunocompetent 55-year-old woman with multifocal ganglionic tuberculosis with a rare attack of navicular bone with good clinical improvement under anti-tuberculosis treatment. It is therefore necessary to systematically carry out an exhaustive assessment of the spread of tuberculosis germs for better management. The prognosis is often favorable, depending on the type of involvement and the early onset of anti-tuberculosis treatment.

**Keywords:** Multifocal tuberculosis, cervical lymphadenopathy, navicular involvement, histology.

**INTRODUCTION**

Tuberculosis caused by Mycobacterium infection remains a public health problem in developed and developing countries [1, 2]. Morocco is an endemic country; about 27,324 new cases of tuberculosis, all forms are diagnosed each year [3].

Osteoarticular tuberculosis represents 2 to 5% of all tuberculosis and 11 to 15% of extra-pulmonary tuberculosis; localization to the midfoot is rare [4]. In addition, tuberculosis of the feet represents only 5 to 10 % of patients with osteoarticular tuberculosis [5]. The diagnosis of tuberculosis of the foot is usually late due to the non-specificity of signs and symptoms [6].

Tuberculosis may involve virtually any organ, tissue or bone in the body. The early diagnosis and prompt treatment is of utmost importance for good clinical outcome. Multifocal tuberculosis is defined as the involvement of at least two extra-pulmonary sites with or without pulmonary involvement. Multifocal forms are rare and represent 9 to 10% of extra-pulmonary locations. Their prognosis is poor with a mortality rate of 16 to 25% according to the authors [7].

Multifocal tuberculosis usually occurs in immunocompromised patients but may affect

immunocompetent. In this connection, we report the case of multifocal TBC with rare bone and ganglionic localization with a review of the literature.

**CASE PRESENTATION**

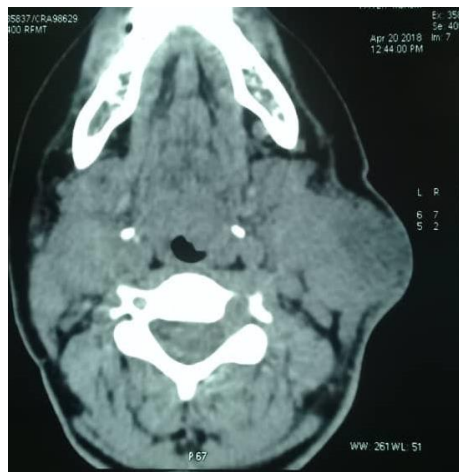
A 55-year-old woman with no particular history was admitted to the otolaryngology department for cervical lymphadenopathy, whose onset was 6 months old, with fever sweating at night, associated with chronic pain and swelling of the tarsal part of her body His foot.

On the cervical plane, there was cervical lymphadenopathy, left territory IIB, painful; with inflammatory sign next, nasal endoscopy was normal. The appearance of mechanical pain in the tarsus of the right foot started 6 months before cervical lymphadenopathy and was treated with analgesics.

The physical examination of the tarsal revealed pain in support, palpation and walking. This pain becomes mixed after a few weeks. Cervical CT showed a left sub-angulomaxillary mass in contact with the sterno-cleido-mastoidian muscle, whose limits are poorly defined.

This mass has a subcutaneous necrotic component with adherence to the cutaneous plane and

deletion of the celulo fatty tissue opposite. Measuring 44×33 mm in its larger axes (Figure 1)



**Fig-1: Cervical tomodensitometry showed a left sub-angulomaxillary mass subcutaneous necrotic component with adherence to the cutaneous plane**

The lymphadenopathy cervicotomy was performed. The histopathological examination of the cervical mass lesion revealed a giant cell granulomatous epithelioid process with caseous necrosis compatible with tuberculosis.

X-rays showed marked navicular osteitis and tarso-metatarsal osteoarthritis with pronounced lysis and sequestration. The computed tomography (CT) confirmed osteolysis of the foot (Figure 2, 3).

The chest x-ray was normal. The biological samples showed a leukocytosis of 14,000 / ml and a CRP of 80 mg / l. Other laboratory tests were within normal limits. The tuberculin skin test was positive.

#### **A biopsy of the bone lesion was performed**

The bacteriological analysis of the bone fragment after Ziehl Neelsen staining was negative. However, the culture in Bactec MGITTM liquid 960

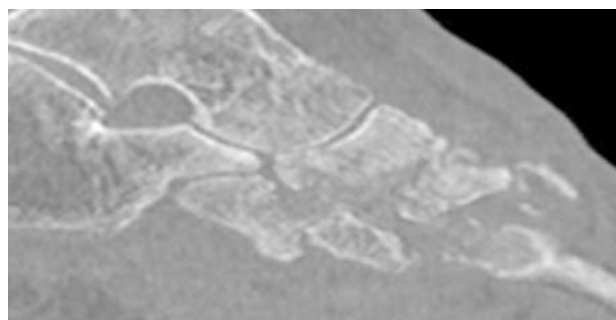
medium was positive after 10 days of incubation.

Identification by molecular biology confirmed the presence of *Mycobacterium tuberculosis* susceptible to rifampicin and isoniazid.

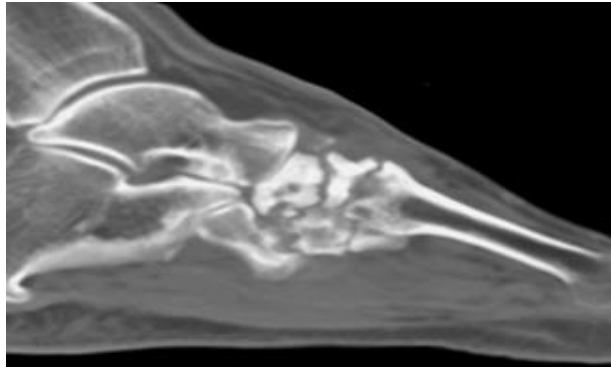
Direct examination of sputum (three samples taken on consecutive days) and urine using Auramine dyes and Ziehl Neelsen were negative for bacillus tuberculosis.

#### **The crops were also negative**

The patient was treated with four-drug antituberculous drugs (rifampicin, isoniazid, pyrazinamide and ethambutol) for 4 months and the combination of rifampicin and isoniazid for 10 months. Clinical improvement occurred after a period of 2 months as he reported decreased pain and swelling of the foot and improved gait.



**Fig-2: CT scan of the foot showing lytic lesion of navicular bone and metatarsals**



**Fig-3: C T foot scan, 4 months after antituberculous chemotherapy: showing the decrease in navicular osteolysis and tarso arthritis**

## DISCUSSION

The incidence of tuberculosis has increased, even in developed countries. Lack of BCG vaccination, trauma or immunodeficiency, and low socio-economic status are risk factors that can be considered responsible [8]. Osteoarticular and lymphatic tuberculosis are the most prevalent localizations of tuberculosis after pulmonary, urogenital, it remains endemic in developing countries[5].

It has been clearly demonstrated that the risk of developing extra-pulmonary lesions is proportional to the degree of immunodeficiency [9] Several hypotheses have been advanced to explain the occurrence of this severe form of tuberculosis in immunocompetents, as in our case.

Some authors implicate malnutrition as a contributing factor, other studies have been able to establish a relationship between diffuse tuberculosis and the intensity of community transmission [10] and others have described the susceptibility syndrome. Mendelian to mycobacterial infections by the existence of defects of the interleukin 12-interferon gamma axis and exposing to diffuse tuberculosis [11].

Tuberculosis of the feet is a fairly rare form of osteoarticular tuberculosis, with a prevalence rate of 5 to 10%. The tarsal joints and the calcaneus bone are the two preferred sites, although navicular and cuneiform tuberculosis is rarely observed [12].

It can be in the form of arthritis or osteomyelitis, involving a single bone, or can be multifocal. The bones involved in tuberculosis are usually the calcaneus, the talus, the first metatarsal, the navicular bone, and the medial and intermediate cuneiform. Due to the existence of intercommunicating synovial spaces, tuberculosis in the middle part of the foot spreads rapidly too many joints [13]. Cuneiform lesions are also described in the tarsal localization of extrapulmonary TB, while 3 cases of navicular bone TB have been reported to date [13-15].

The most common symptom of osteoarticular tuberculosis is the pain often associated with fever with night sweats. Evolution is often chronic and insidious. Our patient complained of inflammatory tarsal pain relieved by analgesics. The diagnosis was discovered following the biopsy excision of cervical lymphadenopathy revealing tuberculosis. Skin fistulization occurs at late stages [16]. Intradermoreaction is positive in 90% of immunocompetent patients but its negativity does not exclude the diagnosis [17]

Standard bone radiography remains nonspecific. Soft tissue swelling and decalcification may be present before late signs of bone destruction. Diagnosis is based on computed tomography. MRI is the baseline examination in early stages where standard radiography and CT can be normal [18].

However, MRI images of osteolysis may suggest other diagnoses such as chronic osteomyelitis, brucellosis, tumors such as osteoid osteoma or chondroma, and metabolic diseases such as hyperparathyroidism and Paget's disease [19].

Histopathological and microbiological examination of the lesion is crucial because it allows the isolation, identification and study of antimicrobial sensitivities. The culture may take 6-8 weeks [5]. Polymerase chain reaction (PCR), is useful for the early and rapid diagnosis of extrapulmonary tuberculosis that can be established within 24 hours with a sensitivity of up to 70-83% [20].

Sensitivity is higher in spinal samples than in non-spinal samples; 90% and 62.5%, respectively. Histopathology remains the gold standard in bone samples from other regions than the spine, which reveals classical tubular tubercle granulomas [21].

According to the studies, because the Koch bacillus locates the periphery of the bone lesion, the tuber may be absent from many samples. For this reason, a surgical biopsy is preferable to a percutaneous

puncture to improve the sensitivity of this technique [22].

#### CONCLUSION

Tuberculous navicular bone is a very rare condition that is difficult to diagnose because clinical symptoms are insidious and standard non-specific imaging is difficult. The histological study after surgical biopsy remains the gold standard for diagnosis.

It can be conservatively treated unless associated with significant osteolysis or other complications. Anti-tuberculosis treatment has excellent results without any complications. We recommend that in this case screening for tuberculosis should be suspected in all cases of chronic osteolysis and / or foot arthritis, especially in countries where tuberculosis is a problem endemic.

Treatment is based on antituberculous chemotherapy for 9 to 12 months. Our patient reported improvement in gait, in cases where bone destruction and functional disability is important an orthosis or surgery for severely deformed foot joints is indicated [23].

#### REFERENCES

1. Subbian S, Tsenova L, Kim MJ, Wainwright HC, Visser A, Bandyopadhyay N, Bader JS, Karakousis PC, Murrmann GB, Bekker LG, Russell DG. Lesion-specific immune response in granulomas of patients with pulmonary tuberculosis: a pilot study. *PLoS One*. 2015 Jul 2;10(7):e0132249.
2. WHO. Global Tuberculosis Report 2015.
3. Bulletin of Epidemiology and Public Health (October 2017), Ministry of Health Directorate of Epidemiology, Kingdom of Morocco
4. Hachimi H, Tahiri L, Kadi N, Ibrahim A, Elmrimi A, Harzy T. Case Report-Tuberculose du médio-pied, une localisation inhabituelle: à propos d'un cas. *Pan African Medical Journal*. 2012;11(1).
5. Dhillon MS, Aggarwal S, Prabhakar S, Bachhal V. Tuberculosis of the foot: An osteolytic variety. *Indian journal of orthopaedics*. 2012 Mar;46(2):206.
6. Dahuja A, Dahuja G, Kaur R, Bansal K. Isolated tuberculosis of talus: a case report. *Malaysian orthopaedic journal*. 2014 Mar;8(1):61.
7. Denis D, Merrien D, Billaud E. Tuberculose multifocale: à propos de 47 cas. *Pathol Biol (Paris)*. 1998 Jun;46(6):375.
8. Habib M, Tanwar YS, Jaiswal A, Arya RK. Tubercular arthritis of the elbow joint following olecranon fracture fixation and the role of TGF-beta in its pathogenesis. *Chinese Journal of Traumatology*. 2013 Oct 1;16(5):288-91.
9. Bodur H, Erbay A, Bodur H, Yilmaz O, Kulacoglu S. Multifocal tuberculosis presenting with osteoarticular and breast involvement. *Annals of clinical microbiology and antimicrobials*. 2003 Dec;2(1):6.
10. Boukeroui L, Nafti S. La tuberculose multifocale chez l'immunocompétent est-elle liée à l'intensité de la transmission dans la collectivité?. *Revue des maladies respiratoires*. 2012 Jan 1;29:A205.
11. Catherinot E, Fieschi C, Feinberg J, Casanova JL, Couderc LJ. Genetic susceptibility to mycobacterial disease: Mendelian disorders of the interleukin-12-interferon-gamma axis. *Revue des maladies respiratoires*. 2005 Nov;22(5 Pt 1):767-76.
12. Liu Y, Chen C, Yang H, Gao H. Case Report Navicular and cuneiform tuberculosis: a rare localization of tarsal tuberculosis. *Int J Clin Exp Med*. 2016;9(11):22486-93.
13. Tuli SM. Tuberculosis of the skeletal system (bones, joints, spine and bursal sheaths). Second ed. New Delhi: Jaypee Brothers Medical Publishers (P) Ltd, 1991:3-122.
14. Yombi JC, Vandercam B, Cornu O, Lecouvet F, Leemrijse T. Tarsal osteoarthritis: a rare localization of tuberculosis. *Rev Chir Orthop Reparatrice Appar Mot* 2007;93(November (7)):740-5.
15. Birjandinejad A, Parsa A, Ebrahimzadeh MH. Isolated tuberculosis of the talonavicular joint in a child. *The Foot*. 2012 Sep 1;22(3):255-7.
16. Zacharia TT, Shah JR, Patkar D, Kale H, Sindhvani V. MRI in ankle tuberculosis: Review of 14 cases. *Australasian radiology*. 2003 Mar;47(1):11-6.
17. Nielsen FF, Helmig O, de Carvalho A. Case report 533. *Skeletal radiology*. 1989 Apr 1;18(2):153-5.
18. Pittet-Barbier L. les infections ostéoarticulaires, neurodiagnostic, neuroradiologie-appareil locomoteur. *Encycl Med Chir*. 1995;10:31-218.
19. Murray RO, Jacobson HG. The radiology of skeletal disorders. 1st ed. Edinburgh: Chrchill Livingston. 1977. p. 559-61.
20. Pandey V, Chawla K, Acharya K, Rao S, Rao S. The role of polymerase chain reaction in the management of osteoarticular tuberculosis. *International orthopaedics*. 2009 Jun 1;33(3):801-5.
21. Urbanczik R. Present position of microscopy and of culture in diagnostic mycobacteriology. *Zentralblatt für Bakteriologie, Mikrobiologie und Hygiene. Series A: Medical Microbiology, Infectious Diseases, Virology, Parasitology*. 1985 Aug 1;260(1):81-7.
22. Lemaitre F, Damade R, Pouchot J, Barge J, Boussougant Y, Vinceneux P. Osteoarticular tuberculosis. Diagnostic contribution of local sampling. *La Revue de medecine interne*. 1995;16(3):191-4.
23. Pertuiset E. Tuberculose osseuse et articulaire des membres. *EMC Rhumatol Orthop*. 2004;1:463-86.