

A misleading appearance of a common disease: A unique presentation of extra pulmonary and multifocal tuberculosis: case report and literature review

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Abstract

Tuberculosis (TB) continues to be a common cause of infectious disease, and it's a common illness for vulnerable populations in resource-limited settings. Extra Pulmonary Tuberculosis (EPTB) accounts for about 20% of TB cases worldwide. Until now, the diagnostic of ETB is not initially considered especially in the setting of a vague clinical presentation, particularly when it is a multifocal localization defined as the presence of lesions, affecting at least two extrapulmonary sites, with or without pulmonary involvement. Elsewhere multifocal forms are exceptional even in endemic countries and affect mainly immunocompromised patients. Here, we report an uncommon case of extra pulmonary and multifocal tuberculosis, with vertebral, digestive and lymph node involvement in a young immunocompetent patient. Diagnosis was confirmed by pathology after the surgery.

Key words: Tuberculosis, Extra pulmonary, Spine, Infection, Bone

Introduction

Tuberculosis (TB) continues to be a common cause of infectious disease afflicting up to one-third of the world's population¹. An estimated 1.67 million people died from TB in 2016². Africa accounts for 3 in 10 cases of TB worldwide and about 4 in 10 cases of TB mortality globally³. The causative organism *Mycobacterium tuberculosis*, which is predominantly air-borne, affects the lung causing pulmonary TB. When TB is bacteriologically confirmed or clinically diagnosed in other parts of the body other than the lung such as the abdomen, meninges, genitourinary tract, joints, bones, lymph nodes and skin it is classified as EPTB. Various risk factors reported to be associated with EPTB include immunosuppression, HIV

infection, male gender and younger age⁴. On the other hand, other studies have found females and increasing age to be more associated with EPTB⁴.

Despite all technological advances, the diagnosis of tuberculous spondylitis remains a clinical challenge since it depends on a high grade of clinical suspicion. This case report shows the importance of taking into consideration a possible TB aetiology even when lesions are observed far away.

Case report

A 16-year-old Algerian man, presented with a 3 month history of permanent lower lumbar pain without radicular distribution. He had a history of asthenia with weight loss of 20kg over the past 12 months with fever, and diarrhoea. He had a colonoscopy which showed segmental areas of inflammation in ileum, pathology showed severe lymphoplasmacytic infiltration, marked architectural distortion, and chronic inflammation without granulomas. The patient was diagnosed with Crohn's disease and was treated with prednisone and mesalazine. However, his symptoms progressively worsened over the next three months, with installation of low back pain. At the physical examination, he had showed good general state of health, tenderness over the lumbar spine without limitation, and no neurological signs. Heart and lung auscultation were normal with no hepatosplenomegaly or adenomegalies. Laboratory findings showed a normocytic and normochromic anaemia (haemoglobin 8.7 g/dL), and thrombocytosis (platelets: 618 x 10⁹/L). C-reactive protein levels of 26.9 mg/dL and an erythrocyte sedimentation rate of 81 mm/h. The tuberculin test was phlyctenular. Computed Tomography (CT) of the thorax, abdomen and pelvic (Figure 1) revealed deep abdominal lymphadenopathy, with ostéomyelitis ivory in appearance at multiple dorsal-lumbar and sacral vertebrae contiguous and noncontiguous.

Figure 1: Computed Tomography: ostéomyelitis ivory in appearance at multiple dorsal-lumbar and sacral vertebrae contiguous and noncontiguous



Histopathological examination of spine bone biopsy and ileum revealed granulomatous lesions with caseous material and multinucleated giant epithelioid cells in favor of spinal tuberculosis. The diagnosis of Crohn's disease was no longer considered and removed from his medical history but considered intestinal tuberculosis. At his last clinic visit, six months into his treatment without any complaints, the treatment will be maintained for 12 months.

Discussion

Despite being a curable disease, TB remains a major public health problem worldwide and one of the diseases with higher mortality. Ohene *et al.*⁴ reported that proportion of EPTB among TB patients was 21.8%, fell within the range of what has been reported for other countries such as Swaziland (18.4%), Cameroon (19.4%) and Botswana (25%).

The proportion of EPTB varies from region to region, reflecting HIV prevalence such as Benin, with a low HIV prevalence (1%) compared to Botswana, which reported an EPTB prevalence of 9%, and in Tanzania EPTB accounted for 15% in one study, and amongst them 58.3% were HIV positive⁵⁻⁷. Despite the low rate of HIV infection in north of Africa, EPTB rates are even higher in this countries with Morocco and Algeria reporting 44.4% and 60% respectively, and 56.9% in Tunisia⁸. Female gender, socio-demographic data, and younger patients represents a major risk factor for EPTB according to several published reports in Turkey, USA, Asia, Egypt and North Africa⁸⁻¹².

Multifocal TB is characterized by the presence of large multifocal tuberculous areas in the same or different organs. On the other hand, disseminated haematogenous TB is characterized by the presence of large numbers of tubercle bacilli throughout body sites, resulting in large numbers of tiny tubercular lesions (1-3 mm in diameter). This entity is usually referred to as miliary TB and has a variable clinical presentation¹³.

Multifocal skeletal TB is defined as osteoarticular lesions that occur simultaneously in two or more locations, with or without pulmonary involvement. It is uncommon, with a reported incidence of 7–10%, and is usually associated with disseminated disease¹⁴. Multifocal intestinal TB is less defined and may refer to multiple liver lesions in the presence of miliary TB. Multifocal systemic TB may merely be referred to as multifocal TB¹³. The term multifocal systemic disease is preferable, because the entity is characterized by the presence of two or more lesions in extra-pulmonary sites, with or without pulmonary involvement.

As in this case described, it is often difficult to promptly diagnose multifocal systemic tuberculosis. Of importance, diagnostic delay, often linked to non-specific symptoms, can have a significant impact on disease progression, favoring the spread of TB to other organs or the impairment of organs already affected by TB. The most common presenting complaint of spinal TB is back pain¹⁵.

Our patient presented two atypical features which delayed the diagnosis, the first one is the intestinal tuberculosis which has been confused with Crohn's disease. The second one is the bone tuberculosis with uncommon contiguous and noncontiguous spinal tuberculosis with ivory aspect.

The intestinal tuberculosis is rare in developed countries and accounts for less than 1% of all cases of abdominal tuberculosis¹⁶. However, its prevalence is significantly higher in countries where tuberculosis is endemic, such as India, African, and Southeast Asia.

However the intestinal tuberculosis and Crohn's disease, frequently present with similar clinical symptoms of weight loss, abdominal pain, fever, bowel obstruction, and bloody diarrhoea, endoscopic findings of skip lesions, ulcerations, and terminal ileum involvement and pathological features, it is occasionally difficult to distinguish between them, thus resulting in a misdiagnosis

as described in our clinical case¹⁷. The histologic hallmark of intestinal tuberculosis that best distinguishes it from Crohn's disease is confluent caseating granulomas, within the submucosa with positive acid-fast bacilli staining.

Vertebral involvement is also particular in our case, because usually, two or more contiguous vertebrae are involved in spinal tuberculosis owing to haematogenous spread through one intervertebral artery feeding two adjacent vertebrae¹⁸ and despite the typical presentation of spinal TB, multiple atypical features have been reported in the literature¹⁹. These forms are indistinguishable from metastasis or lymphoma.

The characteristics uncommon in children with atypical spinal TB include the lack of atypical spinal TB are mainly involvement of the posterior elements of the vertebrae, no intervertebral disc involvement, and extradural spinal cord compression without bony involvement, and the association of multifocal systemic tuberculosis and spondylitis is uncommon in children²⁰.

Our patient had the tuberculous involvement in almost all spinal levels. There were multiple contiguous and noncontiguous lesions in thoracic, thoracolumbar, lumbar and sacral vertebrae without paravertebral abscesses, and without primary lung tuberculosis infection. What was interesting in our patient was the condensing aspect with ivory aspect of the vertebrae instead of a rather lytic aspect. While the condensing aspect giving the ivory aspect of the vertebrae secondary to tuberculosis is found in 10%²¹.

In this patient the source of infection was probably the intestinal infection, since the chest scanner was revealed without anomalies. Turgut²² put forward pelvic infection as a source of spinal tuberculosis in his patient and Kulali *et al.*¹⁸ reported that they could not find any source in their patient. Other multifocal noncontiguous spinal tuberculosis cases in the literature are the cases in small series, and we could not reach any information about their infection sources.

Wang *et al.*¹⁹ reported eight patients with single noncontiguous, multi-segmental, atypical spinal TB with no intervertebral disc involvement. To our knowledge, no case similar to ours has been described, to know a multifocal systemic tuberculosis combining a deceptive intestinal and vertebral form with lymph node involvement without pulmonary involvement. The goal of multifocal systemic tuberculous treatment is to eradicate infection and to treat and prevent different complications as neurological complications or spinal deformities. Pharmacological treatment should be initiated as soon as the diagnosis is confirmed, with 2 months of HRZE (intensive phase) followed by 4 to 7 months of HR (continuation phase). The duration of treatment remains controversial. Due to difficulties in assessing response and risk of relapse, most experts recommend 9 to 12 months of treatment, and in situations of slow radiological resolution as case 2, 12 to 24 months of treatment should be considered²³.

Conclusion

TB may cleverly mimic many diseases and affect multiple organ systems and sites. Thus a high level of suspicion for TB should be maintained in patients with multiple sites of involvement, especially in countries where TB is endemic.

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