

Case Report

Tuberculosis of the ribcage by dissemination of Koch bacilli

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Abstract

Introduction: Tuberculosis (TB) is an infectious, transmissible and immune disease caused by the *Mycobacterium tuberculosis*-complex (MTBC). Although osteoarticular tuberculosis (OATB) has been widely described, the ribcage variety remains a rare form.

Case report: A thirteen-month-old male and a twenty-month-old female, both with pain and increased volume of anterolateral left rib cage were described. Physical examination revealed the presence of a soft consistent mass at the level of the 9th and 5th costal arches in the male and female patients respectively. Upon clinical evaluation, tuberculosis was suspected, which was confirmed by X-ray and histopathological studies. After confirmation, the management, based on anti-tuberculosis therapy was started as follows: nine months of anti-tuberculosis therapy for the male patient and fourteen months for the female. The outcomes were favorable for both patients. However, further interventions, consisting of abscess drainage in the male patient and excisional biopsy in the female patient were necessary. With these therapeutic interventions, to date, the patients are without any evidence of active TB.

Key words: children; Koch bacilli; *Mycobacterium*; rib cage; tuberculosis.

J Infect Dev Ctries 2022; 16(7):1221-1225. doi:10.3855/jidc.14062

(Received 03 October 2020 – Accepted 06 March 2021)

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Introduction

Tuberculosis (TB) is an infectious and transmissible disease of essentially immune nature that is caused by the *Mycobacterium tuberculosis*-complex (MTBC), a group of species belonging to the genus *Mycobacterium*. TB infections have strikingly continued to pose a public health challenge. The worldwide incidence covers a large population group, predominating in marginalized areas and among vulnerable groups [1,2]. The existing epidemiological data depict an alarming global picture. The World Health Organization (WHO) 2017 report estimated that approximately one-third of the world population have latent infection by members of the MTBC [3]. Although its magnitude is unknown, it is estimated that childhood tuberculosis represents approximately 6% of all new cases, and mainly concentrated in countries with a high burden of the disease. There are many factors impeding the correct estimation of tuberculosis in the population of children. Among these factors are the difficulty of establishing a definitive diagnosis, the presence of extrapulmonary disease (which makes it necessary to consult a specialist), its low priority in public health

(childhood tuberculosis is rarely bacilliferous), and the lack of communication between private sector pediatricians and national tuberculosis programs. Children could suffer from tuberculosis at any age, but the most frequently affected group is between 1 and 4 years. Case notifications depend on the intensity of the epidemic, the age structure of the population, available diagnostic tools, and the extent to which contact tracing is routinely practiced.

There is limited data on childhood tuberculosis. The available data are mostly from adults, thus, leaving childhood tuberculosis underreported worldwide [4].

The infection is predominant in the marginalized areas of the world, especially in low-income countries. The extensive spread of TB is associated with migration, untimely diagnosis, inadequate medical management of diagnosed infections, poor patient compliance effort, and inadequate training and poor knowledge on the disease among general practitioners [5].

Ribcage TB is a rare form of osteoarticular TB. However, most of the available literature on this form of TB report cases on a wide age range. Well-defined,

age-group incidence epidemiological data, particularly, in the pediatric population, is lacking. In this communication, two cases of rib cage TB, in a thirteen-month-old male and in a twenty-month-old female, were reported.

Description of cases

Case 1

A thirteen-month-old Mexican male with complete vaccination record that included Bacillus Calmette-Guérin (BCG) vaccine administered at birth was admitted to the National Institute of Pediatrics, Mexico City (INP Spanish acronym). The child arrived with a history of swelling in the anterolateral left rib, six months prior to the admission. Upon evaluation, the swelling was appreciated on the level of the 9th costal arch and measured approximately 4×4 cm in size. There was no change in skin color of the area, nor was there localized increase in temperature. The mass had a soft consistence and was painful on touch. At the time of admission, there was no previous reference to fever. A tomography, performed on admission (Figure 1), corroborated the presence of expansive lytic lesion on the costal arch at the level of osteochondral joint of the 9th arch, surrounded by an extensive zone of necrotic

Figure 2. Rib cage (anterolateral view) showing partial damage of 10mo of the end-portion of left costal arch (first patient).

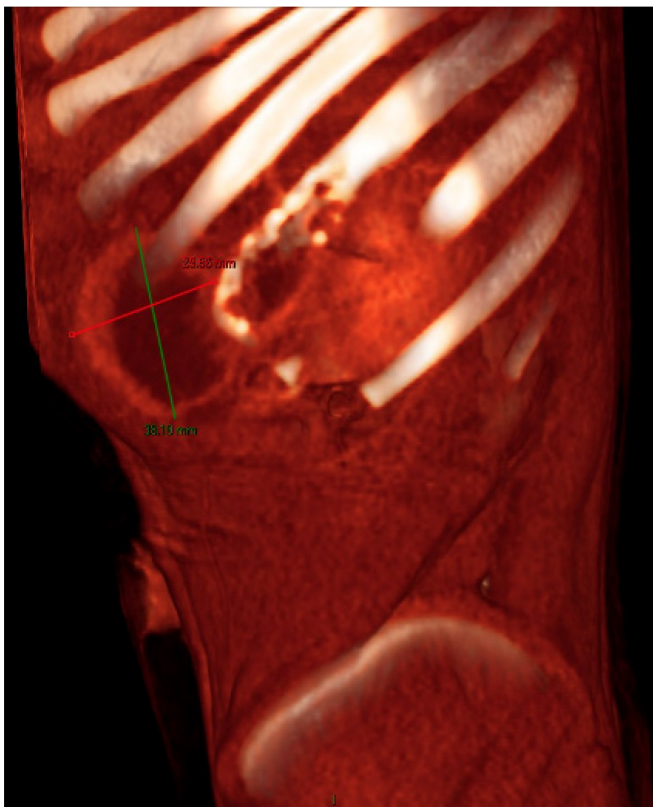


Figure 1. Ossifluent abscess surrounding 10mo of left costal arch (first patient).



tissue (Figure 2). Biopsy of the lesion was performed, extracting 15 mL of cheese-like secretion. The bacterial culture was negative and pathological examination showed areas of chronic granulomatous inflammation with giant multinucleate cells and cheese-like necrosis compatible with TB. Ziehl Neelsen staining revealed a large number of acid-fast bacilli (AFB). A two-month intensive phase of oral anti-tuberculosis therapy based on isoniazid, rifampicin, pyrazinamide and ethambutol was started followed by a five-month maintenance phase with isoniazid-rifampicin. Post-treatment outcome was favorable during five months of follow-up. To date, the patient remains asymptomatic.

Case 2

A twenty-month-old Mexican female with incomplete vaccination record and with BCG vaccine administered at birth arrived to the hospital with an increase in the volume of the left anterior costal region at the level of 5th arch, which began a week prior to her arrival. The mother did not report any known exposure to TB or existing history of TB in the family. She recalled a fever of 39 °C on one occasion. Evaluation of the swelling revealed a mass with hard consistent and painful to touch. The mass measured approximately 4×4 cm in size. The area was discolored with erythematous pigments. Infection by facultative bacteria was suspected, and the patient was administered a seven-day antibacterial treatment, which included clarithromycin and clavulanic acid. However, there was no improvement in her condition. Paraclinical tests performed did not show leukocytosis and the acute phase reactants were negative. Chest X-ray was requested and this indicated an opaque zone, adjacent to a lytic lesion, in the upper anterior portion of the left

5th costal arch. Computerized tomography (Figure 3) showed a heterogeneous mass of soft tissue with extensive areas of necrosis adjacent to the expansive lytic lesion on the 5th costal arch. The lesion had a funnel shape, which extended beyond the rib cage, that compressed and displaced the pleura without invading lung tissue. In a biopsy, a purulent material was obtained. Histological examination indicated necrosis in 80% of the tissue with chronic granulomatous inflammation. A test for the presence of AFB was positive, and osteomyelitis with destruction of costal arch was observed. A diagnosis of rib cage TB was concluded. A fourteen-month anti-tuberculosis therapy based on isoniazid, rifampicin, pyrazinamide and ethambutol was prescribed. The outcome was favorable. The patient is currently asymptomatic without evidence of active disease.

Ethics approval and consent to participate

The study was approved by the Academic Committee of the National Institute of Pediatrics, Mexico City. All procedures were performed in accordance with all studies involving human participants, the ethical standards of the institutional and/or national research committee, and the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Discussion

Tuberculosis is an important cause of morbidity and mortality, particularly, in developing countries. In the case of pediatric patients, the typical and characteristic dissemination route of TB outside the lung is the lymphatic-hematogenous pathway. Through this route, the tubercle bacilli could spread to other parts of the body. This makes them difficult to be isolated in corporal secretions. Although not culture-active, purulent samples containing Koch bacilli were obtained and observed in the two cases presented here [6].

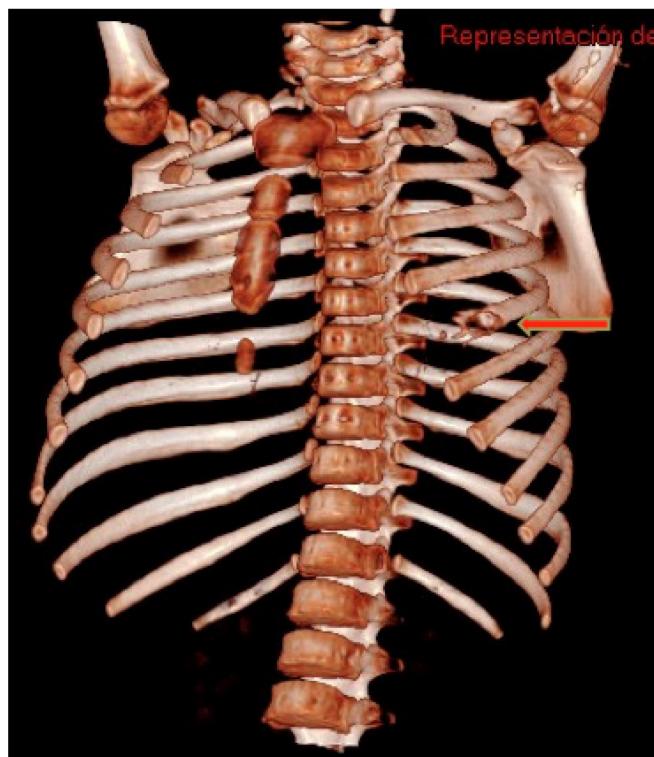
In Mexico, the notification and registry of the Epidemiological Monitoring System for Tuberculosis has reported a total of 18,848 new cases of TB. This is an indication of the high morbidity and gravity of Koch bacilli infection in the country and the burden it entails in the national health budget.

Mycobacterium tuberculosis can affect any part of the body, and the lung is the most frequently affected site, where bacillus infection represents 80-85% of all TB cases [7]. The national incidence rate of extrapulmonary TB in our country is 2.8/100,000 inhabitants (15%). Out of this, 1-5% are osteoarticular tuberculosis (OATB) [6]. The osteoarticular

affections occur through lymphatic-hematogenous dissemination of a primary pulmonary focal infection or by contiguity with a paravertebral lymphatic ganglion. The epiphyseal and metaphyseal areas of high weight-bearing bones and high blood-flow demand with bounteous bone marrow, abundant lymphoid cells and relatively scant macrophages are the most frequent sites. Bones and joints are affected in about 5% of TB patients and the predilection sites are the vertebral bodies and large synovial joints.

TB of the rib cage is an unusual form and occurs by direct inoculation from the lymph nodes or via hematogenous dissemination [6,8]. It represents 1-5% of the cases of musculoskeletal tuberculosis and about 1-2% of all cases of TB infection [9]. It is twice more frequent in men than in women, and is more frequent in the age range 15-30 years. In contrast, other types of osteoarticular TB are more common in children of 2-10 years of age [10]. Indigenous people, immigrants, rural populations, children below 5 years of age, and immunodeficient individuals are the vulnerable groups. The low incidence of ribcage TB could be attributed to its insidious and localized form of presentation that could be misdiagnosed as benign or malignant bone tumor leading to a delay in its treatment. Moreover,

Figure 3. An adjacent ossifluent abscess surrounding the mentioned rib, which extends into the thorax with compression of the pleura, without lung involvement or pleural effusion (second patient).



bone diseases in children are not always detected in routine X-rays. Therefore, the diagnosis is generally late and is usually made when bone destruction has already occurred or when there is growth of a mass in the chest wall, which occurs without local inflammation (cold or hot abscess) [11]. It has to be noted that when *Mycobacterium tuberculosis* is present in the body, its slow growth generates organic affection as well as the activation of immunological processes, which make the damage to be more pronounced.

In rib cage TB, the site of the mass is not classical and any part of the rib could be involved. The mass could be cystic or solid, painful or unpainful. Secretion drainage has been reported in 25% of the cases; however, this happens at a later stage. Findings from X-ray, depending on the part of rib affected, are divided into four categories: costovertebral (35%), costochondral (13%), rib body (61%) and multiple cysts. Usually in lytic lesions, rib widening with periosteal reaction and arrest suggest tuberculosis. Faure *et al* [12], found that ribcage TB generally involves only one rib, and that the most frequent site is the body of the same rib, which is in conformity with the two cases presented in this report. In a study by Chang *et al* [13], TB was confirmed in all the cases following surgical resection and this led the authors to conclude that surgery is the final diagnostic and therapeutic option in ribcage TB. In this report, definitive diagnosis was established based on histopathological findings, which is contrary to what was reported by Faure *et al* [12]. These authors were not able to establish TB diagnosis before surgery in majority of their cases. Preoperative diagnosis of rib cage TB is often complex. Thus, open or guided biopsy is deemed the Gold Standard.

Tuberculosis is the second most common cause of destructive rib cage lesions, preceded by primary or metastatic neoplasia [14]. Other infections associated with rib cage destruction are typhoid or paratyphoid fever, actinomycosis, syphilis, coccidioidomycosis, blastomycosis and brucellosis. A previous history of TB infection could help in the diagnosis of osteomuscular TB; nevertheless, such history is not always present as was observed in the cases of rib cage TB in the patients of this report.

The treatment of rib cage TB is similar to that of bone TB and consists of two-month intensive phase of anti-tuberculosis therapy with isoniazid, rifampicin, pyrazinamide and ethambutol. This is followed by maintenance phase of 9-12 months with a combination of isoniazid-rifampicin. Complete surgical resection is controversial; though, treatment with anti-tuberculosis

drugs could be insufficient for cure in the case of only histopathological diagnosis. There are reports indicating that surgery is the best option. This argument is based on the fact that radical surgery results in a complete removal of the affected area and allows histopathological diagnosis as well as post-operative neovascularization that improves the optimal distribution of anti-tuberculosis drugs and hence, increases the efficacy and response to such treatment.

Conclusions

The patients in this report were histopathologically diagnosed and successfully managed with anti-tuberculosis therapy without relapse. This result could be associated with early diagnosis of the condition. The present cases illustrate that rib cage TB could be histopathologically diagnosed with success. In addition, the results of the present report indicate that the disease should be suspected in patients from endemic areas who present destructive lesions of the rib.

Acknowledgements

We thank Dr. Cyril Ndidi Nwoye Nnamezie, an expert translator and native speaker, for helping in preparing this manuscript. We are very grateful to the National Institute of Pediatrics, Mexico City, for the provision of Federal Resources to carry out the work.

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Conflict of interests: No conflict of interests is declared.