

Sternum Tuberculosis Osteomyelitis in an Adolescent Patient: A Case Report and Review of Children's Literature



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Abstract

Sternum tuberculosis osteomyelitis in pediatric patients is an unusually exceptional disease. The few cases in the literature a great majority of these are from underdeveloped countries where tuberculosis is endemic. The disease, from other sites, occurs by hilar lymph nodes, hematogeneous route or lymphatic dissemination may result. A rare case of secondary tuberculosis osteomyelitis of the sternum body is described. Diagnosis in our case thorough clinical history, radiography, ultrasonography, tomography or further alternative diagnostic techniques and histopathological studies was confirmed. The patient, with anti-tubercular treatment regimen (non-operative), had a successfully decisive response and has not suffered any recurrence during the years of follow-up. The purpose of this paper was to demonstrate the rare case of sternum tuberculosis osteomyelitis disease to pediatricians, pediatric orthopedic surgeons, radiologists, and infectious disease specialists. Previous diagnosis and immediate antibiotic regimens can significantly intensify patients' outcomes.

Keywords: Tuberculosis; Sternum; Osteomyelitis; *M. tuberculosis*; Sinus

History

Hindu writings mentioned the disease, signifying "consumption" (or destruction by virtue of body deterioration, by progression of body condition). Hippocrates (460-337 BC) popularized the word for tuberculosis *phthisis* in ancient Greek, which has numerous connotations including: "to consume", "to spit", and "to waste away" [1]. John Schoenlein (1793-1864) reported, in 1839, the term tuberculosis derived from the Latin word *tubercula* (the diminutive of *tuber*), which indicates a small lump [1]. Robert Koch (1882) a German physician, bacteriologist and researcher (1843-1910) had *Mycobacterium tuberculosis* isolated [1].

Vaughan (1918) an Austrian Pole male patient aged 26, bearing clinical aspects the of the appearance of a firm woody swelling in the middle of the sternum (flat bone); a distinctly cloudy, purulent fluid serum was noted. A "woody presternal phlegmon" due to an organism of low virulence was observed.

The swelling was very hard, red and tender. The woody phlegmon was sent for microscopic examination. This was the first report of sternum tuberculosis in the literature credited [2]. Kremer and Wiese (1936) showed a picture of a young patient, with an anterior and superior swollen mass on the upper part of the sternum bone, with a fistulous lesion, diagnosed as sternum tuberculosis [3]. Olmos (1948) described two young patients with involvement of tuberculosis of the sternum. In the two cases, pulmonary lesions occurring before the age of 8 were presented [4].

Introduction

Tuberculosis (TB) continues as a multi-systemic worldwide infection unchanged in countries where it is endemic. Sternum TB in children is one of the incredibly uncommon sites for flat bone to be infected. Sternum incidence involvement constitutes approximately less than 1% of all cases of *Mycobacterium tuberculosis* osteomyelitis [5-9]. Only a limited number of articles

of sternum TB in children's literature have been reported [3,4,7,9-23]. In view of this, a representative case of sternum tuberculosis osteomyelitis in an immunocompetent adolescent male patient is described. A patient aged 15-year-10-month-old, with a cutaneous sinus formation over the anterior chest wall was ascertained on. The aim of this manuscript was to present an unusual case of tuberculous osteomyelitis of the sternum body area in a young patient with a secondary type (form) and multifocal sternum TB infection categorized [23]. The compelling diagnosis by image examinations and histopathology lesions of infected tissues was confirmed. An anti-tuberculous chemotherapy treatment consisting of rifampicin (RIF), isoniazid (INH), pyrazinamide (PZA) and ethambutol (EMB) was prescribed.

Case Presentation

A 15-year -10-month-old male adolescent patient (PHFS) from a low socioeconomic level family, with foot symptoms, was admitted from another hospital. Careful clinical history revealed initial symptoms 4 months ago after trauma to his left ankle with cast immobilization plus treatment with antibiotics (oxacillin) for 32 days, without any benefit. Past medical history was collaborative, comprising a positive history of Bacillus-Calmette-Guérin (BCG) vaccination. At admission as an inpatient at our hospital, physical examination revealed tenderness on the left foot over the medial malleolus and a purulent secretion (abscess) fistulous lesion were detected. His gait needed auxiliary crutches in conformity with his difficulty of performing flexion-extension of the ankle joint, which was limited. In the thorax over the sternum flat bone an ulcer on the middle portion was evidenced. An open chronic sinus formation or cold abscess (a skin fistula overlying the bone) with drainage of purulent material was noted.

Following informed approval by the mother, a battery of investigations to confirm the diagnosis of tuberculosis was performed. Following informed consent, HIV testing method was requested. The laboratory complementary exams asked for blood count with erythrocyte sedimentation rate (ESR), anteroposterior and lateral chest radiographs as well as routine views for left ankle X-rays, ultrasonography and tomography of the chest were requested. Bone biopsies for microscopic examination of the ankle as well as of the sternum bone were required. On histopathological examination, granulomatous inflammation composing of epithelioid histiocytes, Langhans' type giant cells and typical caseous necrosis were observed. Diagnosis of TB was corroborating upon the other histopathological macroscopic findings. A standard four-drug regimen at our center, an antituberculous chemotherapy combination consisting of rifampicin (10mg/kg per day), isoniazid (10mg/kg per day), pyrazinamide (35mg/kg per day) and ethambutol (30mg/kg per day) for two months as an intensive phase patient was received. Subsequently for order seven months rifampicin (10mg/kg per day) and isoniazid (10mg/kg per day) following the national government's (World Health Organization-WHO) guidelines were used. The patient was regularly followed by visiting the outpatient department at two-month intervals until the disease healed. The tuberculous lesions of the ankle and the sternum were completely relieved after four months of medications employed. The patient satisfactory response to treatment was demonstrated. During the follow-up period there have been no recurrences of the symptoms. Full treatment duration in our patient took nine months. The authors present after six years clinical evaluations (photograph) of the patient (PHFS), aged 21-years-10-month-old, demonstrating, that a sinus over the sternum had healed.

Image Findings (Figure 1)



Figure 1: Clinical Photograph of a 15-years -10-months-old male patient, evidence of a chronic sinus formation or cold abscess of the sternum (a skin ulcer overlying the sternum) with drainage of purulent material was observed.

Results

Laboratory Findings

Complete blood count has showed the following: red cells 5,560,000 per cubic millimeter (c.mm.), hemoglobin 12.8 gram%;

hematocrit 40%; leukocytes count 7,100/mm³; basophils 0%; eosinophils 1%; mielocytes 0%; metamielocytes 0%; band cells 2%; segmentated 73%; lymphocytes 20%; monocytes 4%; plaquets were 375.000/mm³. Erythrocyte sedimentation (ERS) rate (Westergren method) was 44 mm of mercury after one hour;

which was elevated; C-reactive protein (CRP) was positive (over 20,0 mg/dl); human immunodeficiency virus (HIV) testing was negative (nonreactive). Abnormalities in urinary findings were not evident. Purified protein derivative (PPD) was observed 11 mm (positive) 48 hours after administration.

Roentgenographic Findings (Figures 2-7)

Ultrasonography

Figure 4 Ultrasonography of the chest wall hypoechoic collection (low echogenicity) tracts communicating to the skin surface (cutaneous fistula) around the fourth rib arch is evidenced.



Figure 2: A plain chest X-ray evaluation posteroanterior incidence view, a diffuse micro nodular infiltrate, predominantly in the upper lobes of both lungs (parenchymal opacity), compatible with miliary tuberculosis is demonstrated.



Figure 3: A Radiograph of the left foot in lateral plantar flexion and neutral position views, showing diminution of the density (osteoporosis) of the hind foot bones. Adjacent soft tissue density thickness is presented.

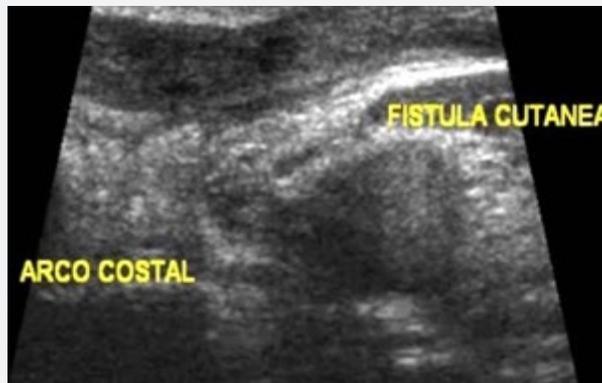


Figure 4: Ultrasonography of the chest wall hypoechoic collection (low echogenicity) tracts communicating to the skin surface (cutaneous fistula) around the fourth rib arch is evidenced.

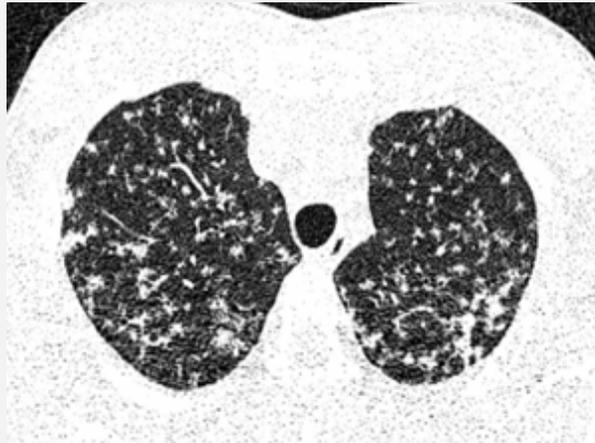


Figure 5: Chest computed tomography (CT) axial view of the lungs, observing small nodules displaying miliary tuberculosis was evidenced. Diffuse interstitial infiltrate reticle micro nodular in the lobes of both lungs is noted.

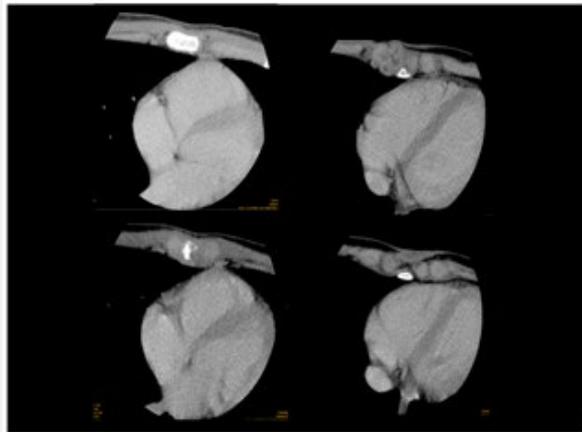


Figure 6: Computed tomography (CT) of the thorax showing bone sclerosis (dense lesion) in the body of the sternum, without mediastinal invasion is demonstrated.

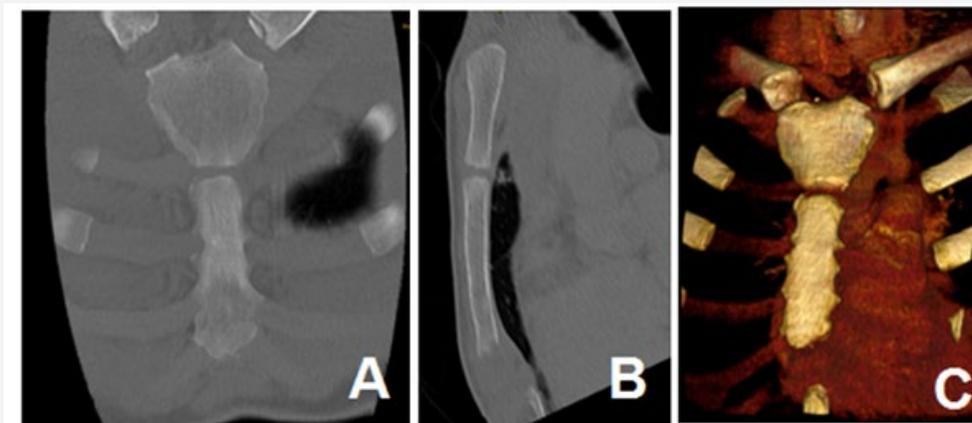


Figure 7: CT reconstruction scans of the sternum in coronal and sagittal views noting lithic (erosion) areas in the inferior part of the bone (Figures 7-A, and 7-B) Reconstruction 3D, demonstrating resorption of the sternum segments of the lower part of the bone. Complete destruction of the distal third of the sternum and the upper edge of the appendice xiphoid (xiphisternum), associated with adjacent swelling of soft tissue parts with hypodense central area, osteomyelitis and abscess is suggested (Figure 7C).

Tomography

Pathological Findings (Figures 8 & 9)

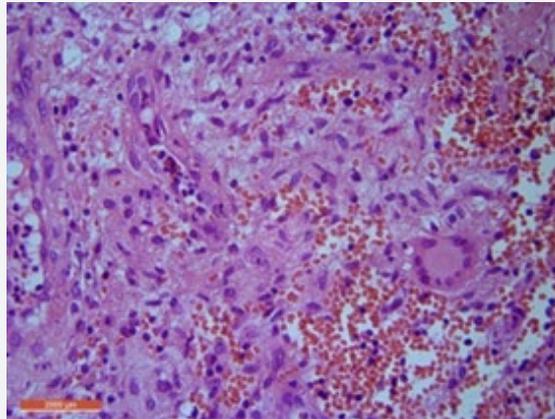


Figure 8: Granulomatous chronic inflammatory osteomyelitis process with epithelioid macrophages and Langhans' type multinucleated giant cells compatible with tuberculosis is observed H&E.

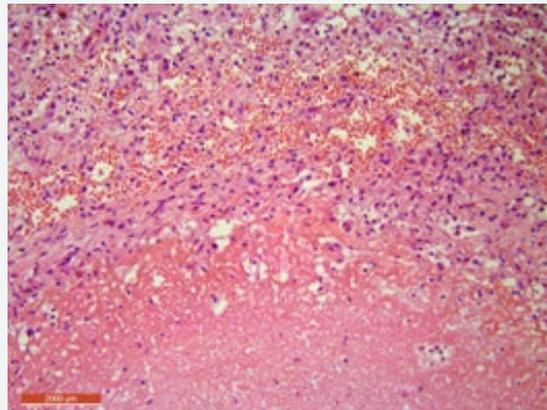


Figure 9: Chronic granulomatous inflammation with extensive caseous necrosis, by macrophages and fibroblasts is delimited. Therefore, upper blood vessels the connective tissues with sparse lymphocytes are presented H&E.

Sinus Findings after Treatment (Figure 10)



Figure 10: Clinical photograph, of the patient (PHFS) aged 21-years-10-month-old (follow-up of 6 years was identified), showing sinus overlying the cured sternum.

Discussion

In Brazil, bone tuberculosis is more common in children and corresponds to 10% to 20% of the extrapulmonary lesions in childhood [24]. Sternum tuberculosis osteomyelitis is a rare form of flat bone afflictions among pediatric populations such as in this case [20]. Sternum TB commonly occurs by reactivation of latent focuses formed throughout the course of hematogeneous or lymphatic dissemination primary tuberculosis. Classically the disease is seen in young adults, although cases in children have been reported [11,16]. A few cases in countries where TB is endemic are described [16]. Sternum TB in pediatric patients is uncommon in the literature; a small number of articles is reported [3,4,7,9-23]. Patients with pain and swelling in the sternum bone, tuberculosis should be considered [25].

Pinheiro reported a study of the records of a series of 94 child patients diagnosed with osteoarticular TB, with only one case of sternum TB observed [22]. McLellan reviewed 20 cases of sternum TB and only two cases to be from the pediatric age group found [25]. Khan et al. [9] in his work about sternum TB, three cases in children were found. Sternum TB osteomyelitis in children, comprises less than 1% of all skeletal TB cases [5-9].

Shi-Min, predominantly in adult cases, from a total of 152 patients, 76 articles on the topic TB sternum infection have been reported. According to the pathogenesis sternum *M. tuberculosis* infections can be categorized into three types: primary, secondary and postoperative. He also ascribes into peristernal and multifocal in conformity with the extent of the lesions classified [23]. Our patient can be included into secondary and multifocal classification. As stated, gender variable Shi-Min 64.9% males and 35.1% females were found [23]. Relating to young adults, males are more common than females affected [7]. According to our study in pediatric population sternum TB literature [3,4,7,9-23], gender preferences were 72% males and 28% females. A significant gender difference between male and female pediatric sternum TB patients, among 18 articles [3,4,7,9-23], at 68 patient's studies was noted. Three patients' genders were not determined [18]. The gender percentage difference among the pediatric population, according sternum TB, is similar to that of adults.

In conformity with variable age cases regarding sternum TB children, Kato et al. [11] and Aribas et al. [13] both authors in an infant 9 months of age at presentation was reported. An average age of pediatric with sternum TB, the literature [3,4,7,9-23], an 8-year-4-month-old was described. In our patient, aged 15-years-10- months-old, above the average, was ascertained. Esalatmanesh et al. [26] foot involvement accounts for less than 10% of cases of osteoarticular TB was described. In our patient with sternum TB osteomyelitis the symptoms of an open draining

on his left foot ankle was also observed. Alternative diagnostic techniques, Mantoux tuberculin skin test (purified protein derivative), radiographs, ultrasound and CT scans should be always included [23]. In our work, all variables mentioned above were followed.

It is hard to understand making a final diagnosis without a histological examination of the sternum tissue [20,26]. It is mandatory, for diagnosis, to secure important histological samples to certify diagnosis and provide drug sensitivity data [18]. The gold standard for diagnosis of osseous tuberculosis comprises of histological examination verification. A needle aspiration or excisional biopsy for histopathological diagnosis of sternum TB is obligatory [27]. Histopathology is the key suitable differential diagnosis to be confirmed [18]. In our case biopsy by an open excisional from the foot and the sternum bone has been done to ratify the diagnosis.

Treatment response depends on the stage when the disease was ascertained [28,29]. There is no consensus (empiric) on the optimal treatment of tuberculosis [21,30,31]. In our service, we follow World Health Organization (WHO) management for treating patients with active TB. Conservative treatment with competent antituberculous drugs in conformity with the research literature is the treatment of choice [3, 4,7,9-23]. We have the opinion that standard anti-tuberculous chemotherapy (conservative management) is a sufficient procedure for obtaining good results. Adequate combinations with anti-tubercular drugs provide satisfactory results [17]. Treatment duration in our patient was 9 months based on RIF + INH + PZA + EMB for two months plus seven months according to RIF + INH was prescribed. Complications of sternum TB include ulcer overlying the sternum (fistula formation) [9], spontaneous fractures of sternum bone [9], tuberculous soft tissue mass (abscess) into the mediastinum [16], secondary infection [29]. In our patient, a chronic sinus formation, with drainage of purulent material was noted.

Differential Diagnosis

Differential diagnosis of sternum bone lesions in children's literature comprise parasite infections [7], actinomycosis [7,20,32], fungal diseases [7,20,32], sternum masses mimicking benign and malignant lesions [9], chest wall tumors in children [11], sarcoidosis [15,20] and acute osteomyelitis of the sternum bone [33].

Conclusion

In conclusion, in the case of any child patient with a nonhealing ulcer or abscess in the sternum bone, reliance on physicians involved with the musculoskeletal system (infection) tuberculosis disease should be kept in mind (Figure 11).



Figure 11: Photograph of a male infant aged 2-years-11-months-old, with sternum TB, cited in Pinheiro's article [22].

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Ethical Approval

The procedures in this study involving human participants was in accordance with the ethical standards of the Institutional Jesus Children Hospital Rio de Janeiro, Brazil, and with the 1964 declaration of Helsinki (DoH) and its later amendments/clarifications have been performed. This study with under protocol nº 172/11, CAAE (Presentation Certificate for Ethical Appreciation) number 0070.0.314.000-11, approved the Research about "Skeletal tuberculosis in children".

Consent

Written informed consent from parents/guardians for publication of this report and accompanying images was obtained. Written consent on the patient's chart registration number 73777 and 127915, Departments of orthopedic, and Radiologic Services of Jesus Children's Hospital, Rio de Janeiro, Brazil is available.

Disclosure

None of the authors received payments or services, either directly or indirectly, i.e., via his or her institution of any aspect of this work. None of the authors, or their intuition(s), have had any financial relationship, prior to submission of this work, with any entity in the biomedical arena that could be perceived to influence or have the potential to influence what is written in this article. Also, no author has had any other relationships, or has engaged in any other activities, that could perceive to influence or have the potential to influence what is written in this work.

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