Case Report



Uncommon Site of Primary Tuberculosis: Sternum

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Abstract

Primary tuberculosis (TB) of the sternum is uncommon even in endemic countries. It can present as swelling, discharging sinus, erosion and spontaneous fracture of sternum, and with secondary infection (abscess). A proper history with physical examination, computed tomography, Ziehl-Neelsen staining of aspirate, and histopathology of biopsy are the keys to forming a suitable differential diagnosis. Its treatment consists of standard antituberculous therapy with or without antibiotics as done for other forms of extra-pulmonary TB. We report one case of primary tubercular osteomyelitis of the sternum causing its destruction without any underlying pulmonary lesion. The patient was successfully managed with debridement and with second-line anti-tubercular therapy (ATT).

Key words: Tuberculosis, sternum, erosion of sternum, anti-tubercular therapy

Introduction

Primary tuberculous (TB) sternal osteomyelitis is a rare clinical entity. The diagnosis is usually made at the advanced stage of the disease, as it is a slow-growing process. Primary sternal osteomyelitis accounts for approximately 0.3% of all types of osteomyelitis and 1% of all skeletal TB cases [1]. Approximately 36 cases have been reported so far in the literature [2]. This disease is common in relatively young people mostly with underlying disease such as pleuropulmonary focus or mediastinal tubercular lymphadenitis. Hereby we report one case of multidrug resistant tuberculosis (MDR-TB) of the sternum (without underlying disease) that was treated with second-line anti-tubercular therapy (ATT). ¹Muzaffarnagar Medical College Opposite Begrajpure Industrial Area Delhi-Dehradun Highway Muzaffarnagar (UP) India

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A 12-year-old girl presented with 9 months' history of pain and swelling over the center of the upper chest wall. Pain was insidious in onset. It was localized and used to subside by taking analgesics. It was aggravated by physical activity and coughing. A history of loss of appetite and weight was present. She also had a history of malaise, fatigue, and evening temperature rise. There was no history of cough or shortness of breath. Physical examination showed pulse rate of 84/min, blood pressure 110/70 mm of Hg and respiratory rate of 22/min. Slight pallor was present.

On her chest examination a diffuse swelling was present over the center of the upper chest (Figure 1). It was tender on deep pressure but the local temperature was not raised. No axillary or cervical lymphadenopathy was found. There was no tenderness over the spine. Abdominal examination showed no organomegaly or any ascites. Laboratory investigations showed Hb 9 gm%, total leukocyte count (TLC) 7600/mm-3 and erythrocyte sedimentation rate (ESR) 60 mm in the first hour. A Montoux tuberculin skin test (purified protein derivative 5 tuberculin unit) was positive (20–22 mm) after 48 hours of test dose. Polymerase chain reaction (PCR) for TB was positive. Chest radiograph was normal. Computed tomography (CT) scan revealed a small cystic lesion (1.96 x 1.42 cm) with enhancing wall, over the upper left half of the 1st portion of the sternum, with its destruction (Figure 2,3). The rest of the lung parenchyma was normal.

Fluid was aspirated from the swelling and it was negative for gram stain but smear was positive for acidfast bacilli (AFB). Fine needle aspiration slides showed clusters of epitheloid cells and giant cell of Langerhan's (Figure 4a,4b). Mycobacterial culture by radiometric method (BACTEC-450) was positive. Drug sensitivity by radiometric method showed bacilli were resistant to isoniazid (H), rifampicin (R), pyrazinamide (Z), ethambutol (E), and streptomycin (S) but were sensitive to kanamycin, ethionamide, ofloxacilin, cycloserin, and para-aminosalicylic acid (PAS). The diagnosis of secondary MDR-TB of the ribs was established. Debridement was done and the patient was put on second-line drugs with 750 mg kanamycin and 1500mg Z daily in the morning; 250mg ethoinamide and 400mg ofloxacillin twice daily and 3 g PAS four times daily, as per



Figure 1. Showing swelling over upper chest.

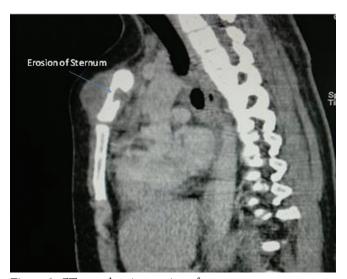


Figure 2. CT scan showing erosion of sternum.



Figure 3. CT scan showing erosion of sternum.

World Health Organization (WHO) guidelines which recommend use of Z in all reserve regimes irrespective of sensitivity result [3]. After three months of treatment, the patient was clinically improved with reduction of swelling. Treatment was continued for 1.5 years. The patient is doing well at 9 months follow up.

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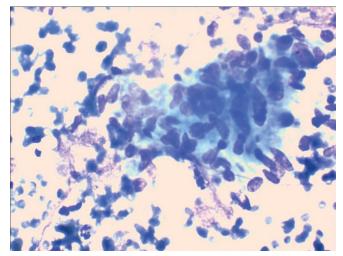


Figure 4a. Slide showing epitheloid granuloma.

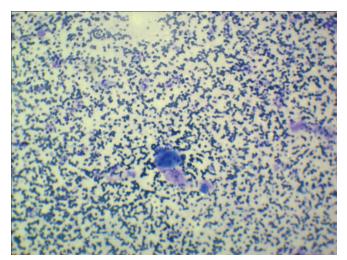


Figure 4b. Slide showing Langerhan's giant cell.

Discussion

Thoracic TB disease may involve the ribs but rarely involves the sternum [4]. Sternal TB is a rare even in endemic countries. This disease is common in younger people but is uncommon in the pediatric age group [5]. In the post-antitubercular treatment period, approximately 25 cases have been reported to date. In one study, out of more than 4000 patients with tuberculosis, only two cases were diagnosed with sternal tuberculosis [6].

Sternal tuberculosis can occur due to direct extension of the disease from pleuro-pulmonary foci or mediastinal lymph nodes, hematogenous spread from a sub-clinical focus in the body, or lymphatic dissemination from other sites, and rarely following BCG vaccination in children [7]. Rarely, post-cardiac surgery sternotomy wounds can get infected with TB due to reactivation of pulmonary tuberculosis [8]. TB is mostly found at the margins of the sternum (as in our case). Incidence of TB of the sternum is common in males and occurs most commonly between 18 to 59 years of age.

Sternal TB osteomyelitis presents as soft tissue swelling (81%) (as in our case), bone or sinus pain Constitutional symptoms like evening rise of temperature, night sweats, or loss of weight and appetite, are uncommon but were present in our case. Complications of sternal TB are superimposed pyogenic infection, sinus formation, erosion and sternal fracture, pressure on large thoracic blood vessels, on the trachea, and/or tracking of tuberculous abscess into the mediastinum, pleural cavity, or subcutaneous tissues and rarely it can spread to bone marrow [2].

Pyogenic abscess of the chest wall, bone destruction due to lymphomas or secondaries of breast or lung cancer are the differential diagnoses for TB of the sternum [9]. Cartilage destruction is slow during TB and starts in the periphery due to lack of proteolytic enzyme. Thus, TB of the sternum can present with a slow, painless course without constitutional symptoms, or as a more aggressive course causing painful bone destruction with constitutional symptoms (as in our case) [10]. Involvement of the lungs presents in 30% of cases and that of the lymph nodes is in approximately 40% of cases of sternal TB [4]. An extrasternal source of TB infection could not be located in our case.

The tuberculin skin test is positive in approximately 70% cases of sternal TB (as in our case). CT can show the extent of bone and joint destruction. In enhanced CT-scan, lesions appear as well-defined juxtra costal soft tissue masses with non-enhancing central low attenuation (due to caseation) and peripheral rim enhancement with frequent evidence of sternum destruction. MRI defines the abscesses and bone marrow involvement in a better way. MRI plays an important role in early detection of doubtful cases [4,10,11].

Radiological changes due to tuberculosis mimic bone destruction due to malignancy and so cannot pinpoint the cause. A needle aspiration or excisional biopsy is crucial for histopathological confirmation. Culture for mycobacterium is positive in 50% of cases. The incidence of positive cultures is similar between both types of biopsies. The biopsy or aspirate material can be 260

soft or friable, light grey in color with a cheese-like consistency. PCR is a reliable and rapid test for recognition of mycobacteria (PCR was positive in our patient). As MDR-TB cases are increasing, culture sensitivities for mycobacterium should be carried out [4,8].

ATT is the basis of treatment for sternal TB although there is no common guideline to the exact regimen and duration of treatment for sternal TB. Extra-pulmonary TB is generally treated with a 6 to 9 months' regimen (2 months of isoniazid, rifampicin, pyrazinamide, and ethambutol, followed by 4 to 7 months of isoniazid and rifampicin), unless the organisms are resistant to these first-line drugs. Directly observed therapy (DOT) is strongly recommended to ensure better medication compliance [12]. Clinical response, side effects, and culture and sensitivity results help determine a suitable drug regimen. Clinical improvement is quicker than radiological alterations, and the pain usually disappears within 2 to 6 months before radiographic improvement [10].

Surgical debridement, reconstruction with pectoralis major muscle flaps, and ATT are necessary to avoid recurrence or sinus formation [8,13,14].

Recently, cases of MDR-TB have increased in India. MDR-TB, described as bacilli resistant to both H and R with or without other drugs, is basically a man-made disease (acquired drug resistance) [3]. Rarely MDR-TB can occur in persons without history of anti¬tubercular therapy intake (primary drug resistance) due to infection with resistant strains (as in our case). Treatment of MDR-TB is not very easy and can only be done with second-line drugs. These drugs are very costly and toxic in nature and have feeble anti-mycobacteriocidal activity. Surgical excision and adding up of fluroquinolone gives better results in the treatment of MDR-TB [3].

Conclusion

Our patient presented with an extremely rare manifestation of tuberculosis (TB of the sternum). A proper history with clinical examination, histopathology, and CT scan are key to forming a suitable diagnosis. Such cases highlight the diagnostic dilemma and generate an awareness regarding MDR-TB of extrapulmonary sites.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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