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Case Report

Tuberculous myositis: an unusual presentation of extrapulmonary tuberculosis

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The prevalence of *Mycobacterium tuberculosis* infection in Taiwan has increased in recent decades. Extrapulmonary infection accounts for 40% of all tuberculosis cases. Here we report a seven-year-old girl with culture-proven tuberculous myositis manifested as progressive enlargement of chest wall mass. No congenital or acquired immune deficiency could be traced at presentation. After surgical excision and completion of 6-month anti-tuberculous therapy, no recurrence of mass or any sequela was found during 6 months of follow-up.

Key words: Antitubercular agents; *Mycobacterium tuberculosis*; Myositis

Introduction

Mycobacterium tuberculosis, an aerobic and slowly growing bacillus discovered by Robert Koch in 1882, is the causative organism of tuberculosis, which is among the most devastating diseases in the world. *M. tuberculosis* initially causes chronic inflammation with granuloma formation and tissue necrosis and then fibrosis and/or calcification in hosts [1]. Although most cases diagnosed are pulmonary tuberculosis, extrapulmonary infections are also common clinically. In 2003, 22,362 reported and 14,074 confirmed cases of tuberculosis were documented in Taiwan. Among these, there were 5665 confirmed cases of extrapulmonary tuberculosis diagnosed [2].

Musculoskeletal tuberculosis represents an unusual entity of tuberculosis. In a retrospective study of 60,606 confirmed cases of tuberculosis, Farer et al found only 676 cases (1.12%) of musculoskeletal tuberculosis [3]. In another study of 2224 autopsy-confirmed cases of tuberculosis, only 4 cases (0.18%) were classified as tuberculous myositis [4]. Petter estimated the incidence of tuberculous myositis to be as low as 0.015% [5]. The majority of cases involve psoas muscles with presentation of cold abscess, protruding mass, and mild pain, extending from adjacent lumbar spine lesions [6].

We report a rare case of culture-proven tuberculous myositis involving intercostal muscle in a seven-year-old girl without documented immunodeficiency.

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Case Report

A seven-year-old girl was referred to the Division of Chest Surgery of a community hospital on December 3, 2004 and was admitted on December 8, 2004 because of a progressively enlarged mass over her right side chest wall found by her mother during bathing six months earlier. She lived in central Taiwan and was an elementary school student at presentation. After she had received vaccines as scheduled including bacillus Calmette-Guérin (BCG), there was no symptom of fever, cough, sputum production, dyspnea, headache, body weight loss, cold sweating, poor appetite, vomiting, diarrhea, abdominal discomfort, or musculoskeletal pain. No history of allergy to drug or food, congenital disease, or growth retardation could be found in this patient who had traveled neither outside central Taiwan nor abroad before. Her family, friends, and classmates were all well before and during the period of illness.

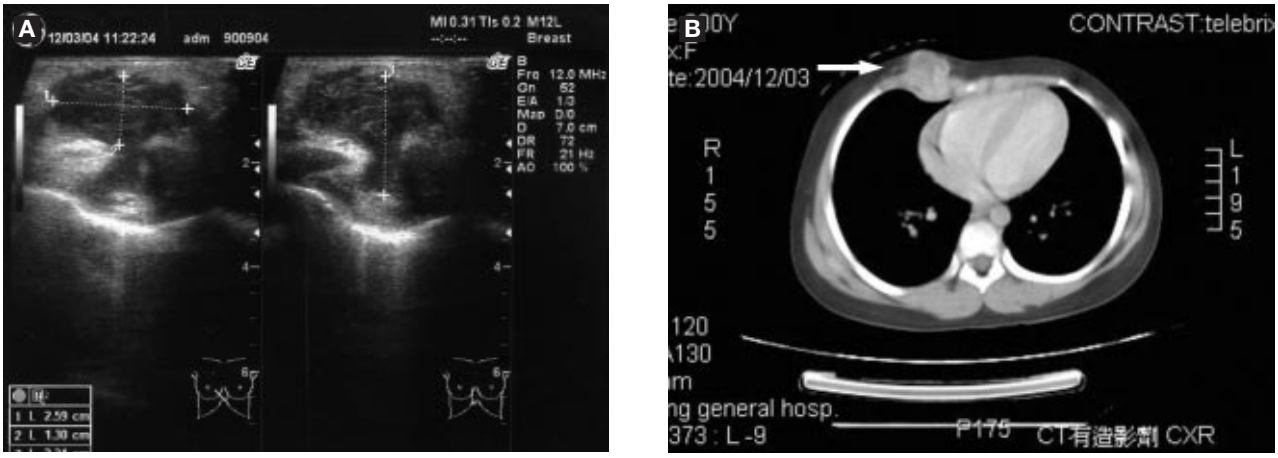

Fig. 1. Radiological examination of the patient. (A) Ultrasound image of chest wall shows a mass with soft tissue density incarcerating into right pleural cavity. (B) Computed tomography with contrast enhancement shows a soft tissue mass (arrow) near sternum involving subcutaneous tissue, intercostal muscle and parietal pleura.

During outpatient visit and admission to the hospital, she received serial examinations, including physical, laboratory and roentgenographic examinations. The physical examination revealed that the girl's height and body weight were within the 75 percentile of growth curves, and there was no significant abnormal finding except a 2×3 cm mass with soft consistence, but no tenderness or color change located in right lower chest area near the right side of the sternum. The results of blood testing and biochemistry study were all insignificant. Chest X-ray showed normal appearance. Ultrasound of thorax showed an avascular mass measuring 2.5×2.5 cm incarcerating into right pleural cavity (Fig. 1A). The computed tomography scan of thorax with contrast enhancement demonstrated a soft tissue mass near the right sternal border measuring $2 \times 2 \times 3$ cm with enhancement of mass border involving subcutaneous fat tissue, intercostal muscle, and parietal pleura with no pleural effusion or mediastinal lymph node enlargement (Fig. 1B).

The operators performed an excision biopsy of the chest mass on December 9 and found a $3 \times 3 \times 2$ cm dumbbell-shaped necrotic mass located in the right anterior chest with extension to retrosternal and retrocostal regions. She came through the operation well and no complication occurred during the postoperative period. Hematoxylin and eosin stain of the excised mass illustrated focal caseous necrosis with chronic granulomatous inflammation, epithelioid cells, histiocytes, foreign body giant cells, mononuclear cells, plasma cells, hyalinization and fibrosis (Fig. 2A). Ziehl-Neelson staining yielded few acid-fast positive bacilli (Fig. 2B). Bacterial cultures including one set of aspirated fluid from the mass and two sets of cultures

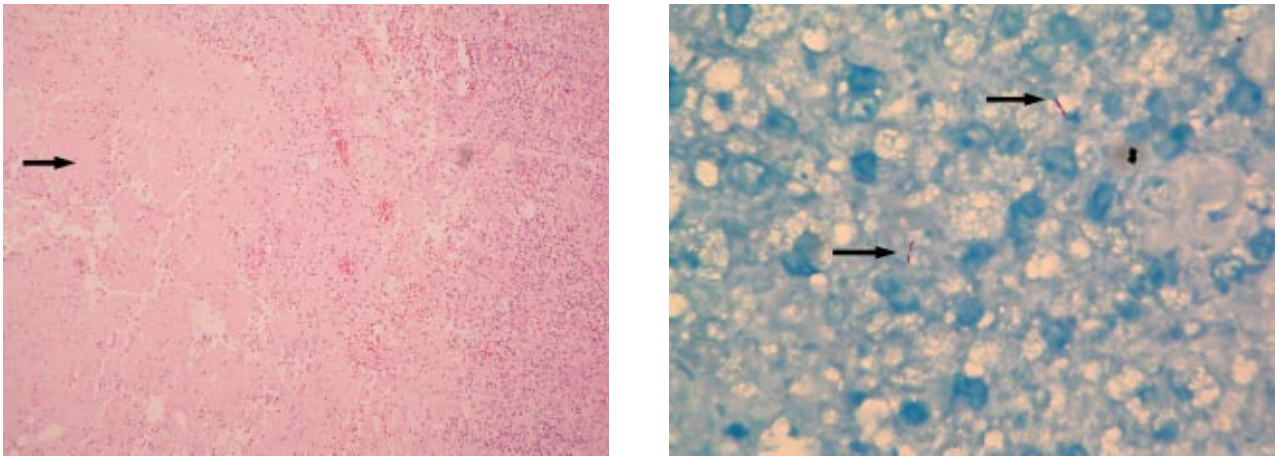

Fig. 2. Histopathology of excised mass. (A) Hematoxylin and eosin stain (× 100) shows caseous necrosis (arrow) and granuloma with inflammatory cells infiltration. (B) Ziehl-Neelson stain (× 1000) shows positive acid-fast bacilli (arrows).

of excised tissue all yielded no bacterial growth. Mycobacterial culture of excised tissue yielded *M. tuberculosis* three months after operation (reported on February 9, 2005) and drug susceptibility testing of the isolate showed susceptibility to all of the first-line anti-tuberculous regimens (isoniazid, rifampin, ethambutol, pyrazinamide and streptomycin).

The physicians suggested anti-tuberculous therapy, while the pathological results provided strong suspicion of mycobacterial infection in December, 2004. The girl's family initially refused anti-tuberculous therapy, despite the doctors' and the public health officials' consultation and recommendation. In June 2005, she started a 6-month course of anti-tuberculous therapy (isoniazid, rifampin, ethambutol, and pyrazinamide for the first two months; isoniazid, rifampin and ethambutol for the next four months) and the treatment was completed in December 2005. No recurrence of mass or any discomfort occurred during the postoperative period or anti-tuberculous therapy. Epidemiologic surveys of the patient's family, friends and classmates during the surveillance period also revealed no evidence of active mycobacterial infection in the individuals concerned.

Discussion

M. tuberculosis infections manifests with various presentations, including pulmonary and extrapulmonary infections. Musculoskeletal tuberculosis accounts for a small percentage of extrapulmonary tuberculosis and has a tendency for slow progression, with most cases diagnosed months to years after the onset of symptoms. Pain is not the most prominent symptom and limitation of muscle function is mild [7,8]. Some researchers reported that tuberculous myositis occurred more frequently in those with altered immune status [9,10], especially in patients with acquired immunodeficiency syndrome [11]. The patient reported here had no documented immune disorder or musculoskeletal dysfunction.

Diagnosis of tuberculous myositis depends on histopathologic examination (caseous necrosis, granuloma, giant cells, mononuclear cell infiltrations, etc.), and microbiologic evidence (acid-fast bacilli in microscopic examination and positive mycobacterial culture). Wang et al reported 35 cases of tuberculous myositis confirmed by mycobacterial cultures or pathologic reports of caseous granuloma over a 5-year study period [12]. Most cases were considered to be bacterial pyomyositis initially (20 patients) from adjacent infected tissue (22 patients). Only a minority of cases (10 patients) were considered to have hematogenous spread.

Radiological studies only provide limited information about tuberculosis infection, with useful findings including calcification of muscle combined with adjacent skeletal lesions in X-ray, well-defined margin of mass lesion with normal or low density in computed tomography scan, and low signal in T1 and high signal in T2 images of magnetic resonance imaging [13]. Differential diagnoses in tuberculous myositis include pyogenic myositis, actinomycosis, hematoma, sarcoidosis, benign tumors, malignant tumors, bursa or tendon cyst, and melioidosis. The tuberculous lesion of primary myositis may present as a solitary nodule with epithelioid granuloma and caseous necrosis or a cystic formation containing a gelatinoid material enclosed by a thick wall, and microscopic examination of the lesion will reveal typical tuberculous lesions [13]. Our patient had a solitary lesion in the chest wall and results of radiological examinations did not provide sufficient evidence of tuberculosis infection. The diagnosis was eventually confirmed by excision biopsy with typical pathologic findings in histologic examination and positive mycobacterial culture. Because there was no obvious abnormality in adjacent tissue found in radiological examinations, we hypothesize that the route of mycobacterial infection in this patient may have been hematogenous from an unrecognized focus (e.g., lung).

Masood reported the efficacy of aspiration cytology and mycobacterial culture in diagnosing tuberculous myositis and soft tissue infection in 11 confirmed cases. Positive acid-fast bacilli in microscopic examination and positive mycobacterial culture of aspirate alone were found in 2 and 4 cases, respectively. Both positive results of microscopic examination and mycobacterial cultures of aspirates were found in 5 patients [14]. Relatively high yield on cytologic examination and mycobacterial culture may be a potential diagnostic tool in tuberculous myositis.

Taiwan is an endemic area of tuberculosis with moderately high prevalence. Every newborn in Taiwan is scheduled to receive a single dose of BCG according to recommendations of the World Health Organization Expanded Program on Immunization. BCG was found to prevent severe forms of tuberculosis such as meningeal and miliary tuberculosis in children but proved to be less effective in prevention of pulmonary tuberculosis in adults [15]. Our patient had received BCG at birth but still developed tuberculous myositis

without evidence of preceding pulmonary infection. We suggest that BCG may have unpredictable efficacy in prevention of extrapulmonary tuberculosis.

In conclusion, myositis and soft tissue mass without rapid progression or clinical deterioration should raise the suspicion of tuberculosis infection in endemic areas. Obtaining tissue specimens for histopathologic examination, microscopic examination, and mycobacterial culture are essential in order to establish the diagnosis of tuberculosis. Surgical excision and adequate doses and duration of anti-tuberculous regimens provide the best way to eradicate tuberculous myositis.

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