Case Report

Tuberculous Empyema Necessitatis with Osteomyelitis, a Rare Case in the 21st Century

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Empyema necessitatis refers to empyema that extends into the extrapleural space through a defect in the pleural surface. Tuberculous empyema necessitatis is a rare complication of tuberculosis. We experienced a 21-month-old boy with tuberculous empyema necessitatis with osteomyelitis in the right 7th rib. He presented with a mass on the right lateral chest wall, which was soft and nontender, enlarging for one month. He also had mild fever. The plain radiograph of his chest revealed soft tissue swelling and calcified lymph node on the left axilla, and his PPD skin test was positive. CT scan of the chest showed empyema necessitatis at the right lower chest and upper abdominal walls with osteomyelitis of the right 7th rib. He did not have concurrent pulmonary tuberculosis. Surgery was performed for diagnosis and treatment. In histopathologic findings, chronic granulomatous inflammation with caseation necrosis was shown and was positive for acid fast bacilli stain. In addition, *M. tuberculosis* complex was found as etiology by polymerase chain reaction. The patient has been treated with anti-tuberculous medication without any specific complication. **(Korean J Pediatr Infect Dis 2011;18:80-84)**

Key Words : Empyema, Pleural, Tuberculosis, Osteomyelitis

Introduction

Empyema necessitatis is a collection of inflammatory tissue that usually extends directly from the pleural cavity into the thoracic wall, forming a mass in the extrapleural soft tissues of the chest[1]. It was an uncommon complication of tuberculosis even in the pre-antibiotic era. Moreover, most of its cases have been reported in adults, and pediatric cases have been extremely rare[2, 3].

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It was first described by Gullan de Baillon in 1640 when he reported a woman who had a left-sided pulsating tumor associated with pleurisy[4]. In 1940, Sindel published a comprehensive review of 115 empyema necessitatis cases[5]. Since 1966, only 26 cases have been reported[2]. Among them, less than 10 pediatric cases have been reported since the first case by Laënnec in 1819[6].

We present a 21-month-old boy with tuberculous empyema necessitatis with osteomyelitis on the right 7th rib.

Case Report

A 21-month-old boy presented with an enlarging mass for one month on the right lateral chest wall.

He had complained of pain and mild swelling in the right chest wall since he slipped down one month ago but there was no external wound. He was diagnosed with rib fracture and treated conservatively for one month. However, his chest wall mass increased in size and he showed other symptoms such as mild fever and cough wax and wane for one month. He had no familial and environmental exposure history of tuberculous diseases, and had received BCG vaccination (subcutaneous multipuncture type) at infancy.

Vital signs on admission were temperature 37.9 ℃, pulse rate 124 beats per minute, and respiratory rate 32 breaths per minute. Physical examination revealed a 3×3 cm subcutaneous mass without crepitus in the right lateral chest wall between the 6th and 9th ribs, which was erythematous, soft and not tender. The right lower chest was dull to percussion, and breath sounds decreased in the right lung.

The plain radiograph showed soft tissue swelling in the right chest wall and calcified lymph node in the left axilla (Fig. 1). He showed a positive result (15 mm of induration) in Mantoux tuberculin skin test. Computed tomography (CT) scan of the chest revealed empyema necessitatis at the right lower chest and upper abdominal walls (Fig. 2). It was accompanied by osteomyelitis of the right 7th rib with cortical destruction. In addition, surrounding subcutaneous tissue showed diffuse inflammatory change which was extended to pleura. He did not have concurrent pulmonary tuberculosis.

Laboratory values revealed a white blood cell count of 13,900/mm^3 with 45% neutrophils and 52 % lymphocytes. Hemoglobin was 9.7 g/dL and platelets were 441,000/mm^3. C-reactive protein was 2.43 mg/dL, erythrocyte sedimentation rate was 44 mm/hr, and lactate dehydrogenage was 239 U/L. Blood culture performed on the 1st hospital day revealed no growth of general organisms. His serum immunoglobulin (G, A, M) levels were within the normal range and C3, C4 and CH50 levels were normal. The number and function of phagocytes by respiratory burst test were normal.

For diagnosis and treatment, surgical debridement and rib resection were performed. The lesion was a round mass and 7 cm in diameter, filled with pus-like materials. The 7th rib was destructed at 1.5 cm from the costochondral junction. In the histopathologic examination of the resected rib, chronic granulomatous inflammation with caseation necrosis was shown and it was positive for acid fast bacilli stain. The cultures of pus and tissue were negative for acid fast bacilli. Etiologic diagnosis was confirmed as *M. tuberculosis* complex, not *M. bovis* BCG in pus and tissues by polymerase chain reaction (PCR) and other molecular methods.

The patient has been treated with anti-tuberculous medication for 12 months (a regimen of isoniazid and rifampin supplemented in 1st 2 months by pyrazinamide and streptomycin) without any specific

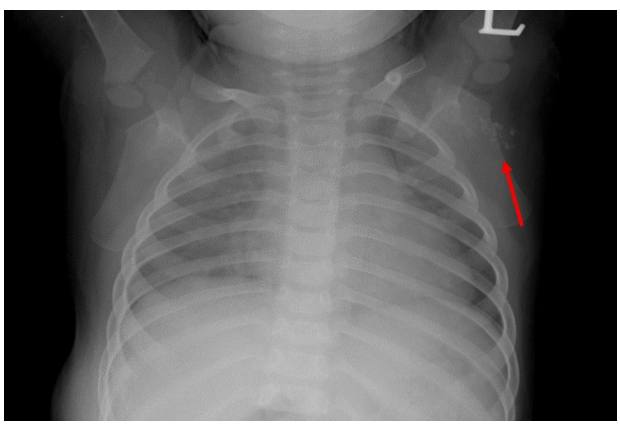

Fig. 1. A plain radiography revealed a mass in the right lower chest wall with osteomyelitic change of the right 7th rib and left calcified axillary lymph node.

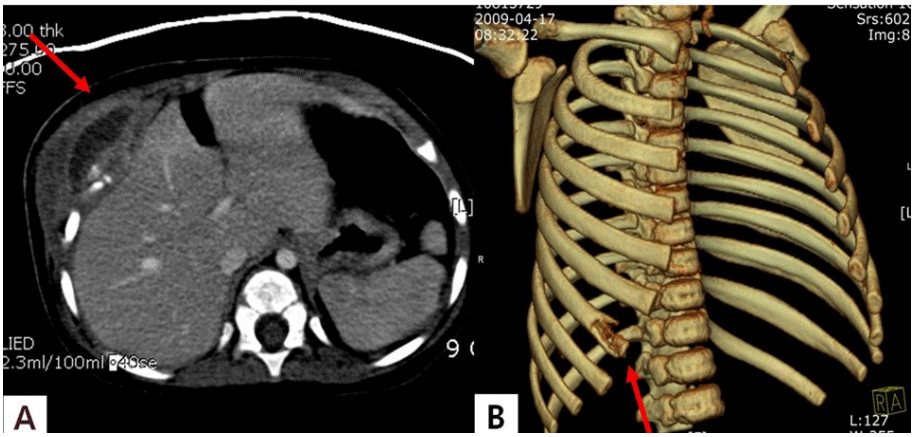

Fig. 2. CT scan of the chest showed empyema necessitatis at the right lower chest and upper abdominal walls with osteomyelitis of the right 7th rib. (A) Axial scan of the chest and upper abdomen, (B) 3D- reconstruction of the bony chest wall.

complication.

Discussion

Empyema necessitatis is an extremely unusual complication of empyema, and most of its patients were described in the pre-antibiotic era. In addition, most of the cases over the past 30 years have reported in immunocompromised patients[1].

Empyema necessitatis is a collection of inflammatory tissue that ruptures spontaneously through a weakness in the chest wall to surrounding soft tissues[1]. Increased pressure within the loculation, chronic inflammation, and necrosis with erosion and fluid extension contribute to this disease[7]. It is characterized by the extension of suppuration from the pleural space, typically through the thoracic wall, to form a subcutaneous abscess. In addition to the thoracic wall, less commonly it may involve the bronchus, esophagus, diaphragm, breast, pericardium, mediastinum, even retroperitoneum and groin[2-4]. However, the most common site is the anterior chest wall between the midclavicular and anterior axillary line and between the 2nd and 6th intercostal spaces[5].

This complication can occur by a number of infectious agents, but indolent infections such as tuberculosis and actinomycosis are most likely related. The chronic nature of these infections is closely correlated with the typical slow progression of the disease. Of the infectious agents, *Mycobacterium tuberculosis* is most frequently implicated. Among the 115 cases described by Sindel, 84 cases (73%) were caused by tuberculosis and 31 by pyogenic infections, mainly *Streptococcus pneumoniae* [5]. In 2004, Freeman et al. represented in the review of published cases since 1966 that tuberculosis was the leading cause of empyema necessitatis (50%, 13 of 26 cases), followed by actinomycosis (24%, 7 of 26 cases)[2]. Other causative agents were *S. pneumoniae, Staphylococcus aureus, Streptococcus milleri, Fusobacterium nucleatum,* and *Mycobacterium avium-intracellulare*[2]. According to Sindel's report, the etiology of empyema necessitatis was more often pyogenic bacteria in younger patients and tuberculosis in older patients possibly because of

the chronicity of tuberculosis[5]. It was very rare that *M. tuberculosis* was the causative agent in a young boy who did not have the history of exposure to tuberculosis. Therefore, we performed PCR for differentiating from *M. bovis* BCG.

In our case, it is difficult to determine whether his lesion was from the osteomyelitis of the 7th rib or pleural empyema. In 1999, Mehndiratta et al. presented a 60-year-old male with empyema necessitatis following osteomyelitis of rib in India, which was caused by *S. aureus*[8]. Like this, empyema necessitatis might have originated from rib osteomyelitis in our case. Empyema formation following osteomyelitis of rib is basically due to the extension of infection to the pleura. If the suppurative process is uncontrolled and undrained, it may present as empyema necessitatis.

Tuberculous rib osteomyelitis is rare and its incidence has been reported to be less than 0.1% of all tuberculosis cases[9]. Occasionally, osteomyelitis may develop as a complication of rib fracture. Trauma may cause local edema and may alter the blood flow, and hematoma seems to provide a good local environment for bacterial proliferation[10]. In our patient, trauma may be a predisposing factor.

Differential diagnosis of empyema necessitatis is various. Neoplasms are important since they can present in a very similar way with extension from one anatomical plane to another. Not only primary pulmonary neoplasm but also a systemic disease such as lymphoma is a possible cause. Benign conditions such as Wegener's granulomatosis and sarcoidosis, infective endocarditis and septic embolisation should be considered[11]. Thus, histopathologic findings through biopsy are important for the accurate diagnosis of etiology.

Treatment of empyema necessitatis includes surgical drainage and antimicrobial medication depending on the organism. Although conventional wisdom states that 12 to 18 months of treatment is required for skeletal tuberculosis, newer evidence recommends that 6 to 9-month regimens containing rifampin are sufficient[12, 13]. Because we were concerned about infection by a resistant organism and our patient was very young, streptomycin was included in the treatment regimen instead of ethambutol.

In the pre-antibiotic era, mortality from empyema necessitatis was 66%[5]. However, with the advent of antibiotic therapy, the mortality has decreased markedly[2]. Moreover, there have been no reported deaths since 1966[2]. However, delay in diagnosis and treatment may result in the progression of the disease process and significant morbidity and mortality.

In conclusion, empyema necessitatis caused by osteomyelitis has not been reported previously in Korea. Because of its insidious symptoms, early lesions may be neglected. Thus, physicians should be aware of enlarging chest wall mass and local signs of inflammation and a high index of suspicion is very important. Early diagnosis and surgical intervention followed by proper antibiotic medication can achieve the best outcome.

요 약

늑골 골수염에 동반된 흉벽 천공성 농흉 1례

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흉벽 천공성 농흉(Empyema necessitatis)은 농흉이 흉막 외의 공간으로 확장되어 나간 것을 말한다. 결핵성 흉벽 천공성 농흉은 결핵의 드문 합병증으로서 특히 소아에서는 더욱 드물다. 본 저자들은 결핵에 노출된 병력이 없던 21개월 남자아이에서 7번째 늑골의 골수염에 동반된 결핵성 흉벽천공성 농흉을 경험하여 이에 대해 보고한다. 우리는 환자의 진단과 치료를 위해 수술적 치료를 시행하였고, 조직을 이용한 PCR 및 분자생물학적 검사에서 *M. tuberculosis* complex를 확인하여 항결핵제를 이용해 합병증 없이 치료하였다.

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