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European Journal of Cardio-thoracic Surgery 21 (2002) 352–354

EUROPEAN JOURNAL OF CARDIO-THORACIC SURGERY

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Case report

Cold abscess of the chest wall as an unusual complication of BCG vaccination

Olgun Kadir Aribas[a,*], Fikret Kanat[b], Niyazi Gormus[c], Emel Turk[d]

[a]*Department of Thoracic Surgery, Meram Medical School of Selcuk University, Konya, Turkey*
[b]*Department of Chest Diseases, Meram Medical School of Selcuk University, Konya, Turkey*
[c]*Department of Cardiovascular Surgery, Meram Medical School of Selcuk University, Konya, Turkey*
[d]*Department of Infectious Diseases, Meram Medical School of Selcuk University, Konya, Turkey*

Received 27 September 2001; received in revised form 13 November 2001; accepted 17 November 2001

Abstract

Bacillus-Calmette-Guérin (BCG) vaccination often results in local adverse effects; however, serious or long-term complications are rare. The involvement of sternum among skeletal BCG osteomyelitis is a rarely seen complication of BCG vaccination. Such a complication may confuse with a chest wall tumor and a surgical intervention may be needed for the definite diagnosis. A 9-month-old infant who had a parasternal cold abscess in the anterior chest wall and sternal osteomyelitis of tuberculosis in the late period of BCG vaccination of whom the etiological diagnosis was histopathologically confirmed after surgery is presented and the preoperative diagnostic problems are discussed.

Keywords: Osteomyelitis; Tuberculosis; Extra-pulmonary tuberculosis; Sternal tumor; Bacillus-Calmette-Guérin vaccination; Complications

1. Introduction

Bacillus-Calmette-Guérin (BCG) vaccination is widely used in our country as in the whole world to prevent tuberculosis (TB) and though its safety profile is high, sometimes a variety of complications may develop. The most frequent complications encountered are the local subcutaneous abscess and the regional suppurative lymphadenitis [1]. It may also cause a disseminated disease with fatal prognosis in infants having especially immune deficiency [2]. Bone osteomyelitis as a complication of BCG vaccination is rarely seen mostly involving the epiphysis of long bones [3]. Sternal osteomyelitis with abscess was seldom reported in the literature [4].

2. Case report

A nine-month-old male infant was admitted with a swelling on the anterior chest wall noticed 1 month ago and growing gradually. Physical examination revealed a mass on the left of sternum, approximately 3 cm in diameter, with elastic consistency and pain on pressure, and without hyperemia. The other systems and laboratory investigations were normal. No lymphadenopathies were palpated. Chest X-ray and chest computerized tomography (CT) revealed an oval shaped $2 \times 3 \times 3$ cm mass with soft tissue density to the left of sternum (Fig. 1). Pulmonary parenchyma seemed to be normal.

The patient underwent an operation via vertical incision on sternum. Chondral parts of third and fourth ribs with the adjacent segment of sternum and associated pectoralis major muscle adherent to the chondrosternal junction appeared to be destroyed. In addition, we observed a parasternal abscess with a caseous necrosis, 3 cm in diameter, located posterior to the pectoralis muscle and lying in the anterior mediastinum. Cartilages of the left third and fourth ribs, involved sternum and pectoralis muscle were removed completely. Intact segments of corpus sternum were primarily sutured. The pectoralis muscle flap was then joined to the sternal periosteum in the midline, advancing the flap inferiorly to cover the defect, 3×4 cm, remained in the space of excised cartilages. Parasternal abscess was completely drained and associated anterior mediastinum was cleaned. A chest tube was placed in the anterior mediastinum and a redon drain was brought through the skin flap to the left of the sternum and placed over the pectoralis muscle in the left parasternal position. On histopathological examination,

* Corresponding author. Selcuk Universitesi Meram Tip Fakultesi, Gogus Cerrahisi Anabilim Dali, 42080, Meram, Konya, Turkey. Tel.: +90-332-3232600x1844; fax: +90-332-3232643.
E-mail address: olgun@selcuk.edu.tr (O.K. Aribas).

1010-7940/02/$ - see front matter
PII: S1010-7940(01)01103-4

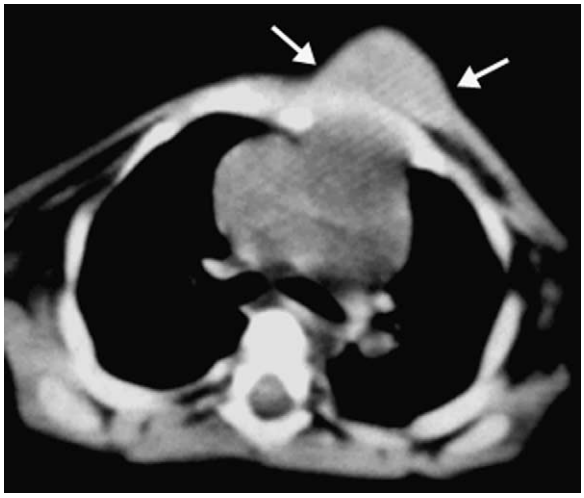

Fig. 1. CT scan of the chest disclosed 2 × 3 × 3 cm in size, oval mass in soft tissue density to the left of sternum (white arrows).

granulomatous inflammation consisting of epitheloid histiocytes, Langhans' type giant cells and typical caseous necrosis were present (Fig. 2). Both histopathological and intraoperative macroscopic findings, confirm the diagnosis of TB on the 2nd postoperative day, and antituberculous therapy with isoniazid, rifampin and streptomycin was started. The abscess content was negative for acid-fast bacilli. PPD in early postoperative period was positive. The culture of tissue specimens was also negative for any microorganisms. We found no contact to the patient with TB in the family. It was considered that the disease resulted from the BCG vaccine complication, demonstrating subacute character. We did not come across any postoperative complications and adverse drug effects and the patient's chemotherapy was terminated after 6 months. He is now 5 years old in good health without any thoracic wall deformity.

3. Discussion

TB is an important public health problem associated with socioeconomic level, nutrition and general hygienic conditions. Although the incidence of TB had significantly been decreased, it has tended to rise especially since 1985. This rise is particularly expressed in extra-pulmonary TB, which is 17.5–32% of all TB cases [5,6]. A synchronicity with pulmonary TB was reported only in 6% of such cases [5]. Thus, the definite diagnosis in these cases may be usually possible with surgical interventions [3,6,7]. The most frequent localization of extra-pulmonary TB is the lymph nodes [6]. Sternal tuberculous osteomyelitis, as in our case, comprises only 1% of all skeletal TB [8]. Although sternal osteomyelitis is a serious complication of BCG vaccination, in our patient the subacute presentation was seen. Although we did not isolate *Mycobacterium tuberculosis*, as there was no index case for TB that could infect the patient in the family or family relatives and neighbours and at birth, for the subclinical character of the disease, because of the regressing ipsilateral cervical and axillary lymphadenopathies explored in the investigation of patients history and more than that the histopathology consistent with TB were the evidences supporting our diagnosis, a complication of BCG vaccination. Hengster et al. [1] reported that in the first 20 weeks of vaccination *M. tuberculosis* could be grown in culture in only 46% of the patients and after that period even if it was cultured no growth would be seen. The negativity of our culture resulting in specimens of operative material taken after 9 months of vaccination is in parallel with that finding.

On BCG vaccination, first an ulceration and later granulation tissue develop at the inoculation site and a primary complex occurs as soon as it reaches regional lymph nodes via lymphamatous dissemination [3]. When it passes through lymph node obstacle hematogenous dissemination develops [2]. Gormsen [9] reported that granulomatous lesions might occur in remote organs such as liver, lungs, spleen and kidneys following BCG vaccination in a postmortem study of children who died of other causes, that could be explained by the hematogenous spread of inoculated bacilli. Sternal osteomyelitis after BCG vaccination may also occur hematogenously. However, in our case, the clinical picture and the parasternal cold abscess associated within the anterior mediastinum as revealed on chest CT suggest that this might well be due to a cold abscess in the internal mammary nodal chain.

The BCG vaccine complications are met in 3.3% of patients and generally occur after 6–9 months of vaccine administration [1]. Consistent with the literature, vaccine complication in our case was detected 9 months after vaccination. Easton and Hershfield [10] published BCG lymphadenitis that developed even 11 and 18 years after vaccination. Lymphadenopathies less than 1.5 cm usually regress spontaneously [1] as in our patient's history. However, lymphadenopathies more than 1.5 cm in diameter

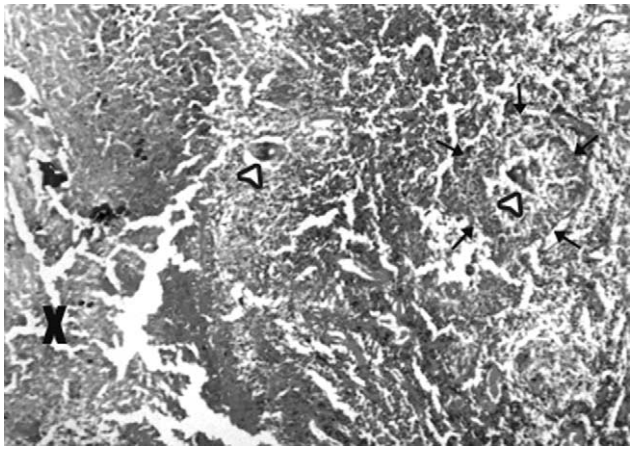

Fig. 2. Histopathological examination of the specimen revealed granulomas (black arrows) including epitheloid histiocytes, Langhans' type giant cells (white arrows) and a caseous necrosis (×). (HE × 20).

have been noted usually fistulizing and not showing regression [1–10].

Vaccine complications categorized to four groups by Talbot et al. [2] include: (1) regional disease, (2) extra-regional localized disease, (3) disseminated disease, and (4) the other BCG syndromes. According to this classification, our patient may be thought to be in the second group.

Osteomyelitis secondary to the BCG vaccine is frequently seen in the epiphysis of long bones [3,4]. BCG osteomyelitis and subcutaneous abscess have been reported 1/3000–5000 in Finland and Sweden [4]. This high incidence has been associated with the method of vaccination and the more virulent vaccine strains [3]. The most serious complication of BCG vaccination is disseminated BCG infection having guarded prognosis [2].

In conclusion, the BCG vaccine complication should be considered in such cases even without bacterial isolation, because of the subacute course of disease, and the occurrence within 1 year of BCG vaccination, and the absence of pulmonary foci, a contact to the patient with TB, and the proof of TB histopathologically. In addition to that we should always remember sternal osteomyelitis with abscess formation in the differential diagnosis of chest wall tumors in children.

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