Case Report

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A Rare Immune Disorder Revealed by an Unusual Chest Wall Granuloma Due to BCG Vaccination in a Child

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Abstract

Bacille Calmette-Guerin (BCG) vaccination is known to be safe. Causing tuberculous chest wall in a 12-month-old girl is certainly uncommon. This case demonstrates that such as complication may confuse with a chest wall tumor and need several investigations for the definite diagnosis. We should keep in mind a high index of suspicion of immune disorder in any infant with atypical clinical findings.

Keywords: Tuberculosis; Bacillus calmette guerin; Tumor; Mendelian susceptibility; Mycobacterial disease

Introduction

Tuberculosis (TB) is still endemic in our country. In order to prevent TB and especially severe forms, Bacillus-Calmette-Guerin (BCG) vaccination is widely used at birth in our country as a part of the vaccination program, as recommended by the World Health Organization (WHO) [1]. Although it is considered a very safe vaccine, some local adverse effects are noted such as local subcutaneous infection and regional suppurative lymphadenitis in immune competent children [2]. However, the occurrence of severe complications should lead to the search for immune disorder. Here we are reporting a rare case of chest wall granulomatous lesion developed 12 months after BCG vaccination and presenting as a tumor of the anterior chest wall. She was subsequently found to have a Mendelian susceptibility to mycobacterial disease due to mutations in IL-12Rβ1.

Case Study

A 12 month-old female child from a non-consanguineous Tunisian family, was referred to our department for a painless mass on the anterior chest wall noticed by her mother. A history of trauma or any symptoms such as fever, nocturnal sweating, or weight loss, was not reported. After 2 months of her birth, her mother was treated for tuberculous lymphadenitis. She was vaccinated with Bacillus Calmette-Guerin (BCG) at birth as it recommended in our country. Since that date, she has kept an inflammatory lesion at the inoculation site. One month ago, the mother noticed the appearance of a left axillary lymphadenopathy with fistulization on the skin [3]. During physical examination, an immobile, firm and painless mass was detected on the anterolateral side of the left chest wall, approximately 5 cm in diameter. It was associated with a chest wall veins and a homolateral axillary lymphadenopathy (**Figure 1**).

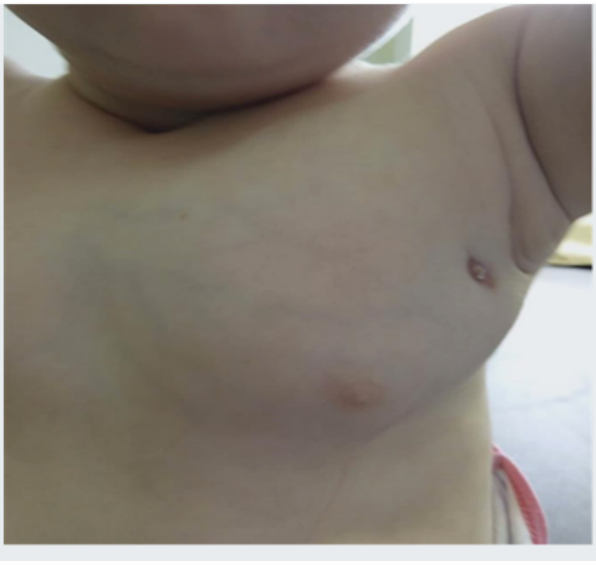

Figure 1: An immobile of the anterolateral side of the left chest wall, approximately 5 cm in diameter associated with a chest wall veins and a homolateral axillary lymphadenopathy.

Results

The laboratory evaluation demonstrated a WBC count of 8600 × 10^3/µL (42% neutrophils, 49% lymphocytes and 8% monocytes), an erythrocyte sedimentation rate (23 mm/hr) and C-reactive protein levels (3.14 mg/dl). A tuberculin skin test was positive (12 mm). Imaging including chest X-ray and abdominal ultrasonography was normal. Chest computed tomography revealed a homogeneous left p arietal m ass o pposite t he left breast region with a densification of fat under jascent and subcutaneous axillary adenomegaly. A part from this, bony structure and pulmonary parenchyma seemed normal [4]. Those findings cannot distinguish between a pyogenic chest wall abscess and a malignant tumor (**Figure 2**).

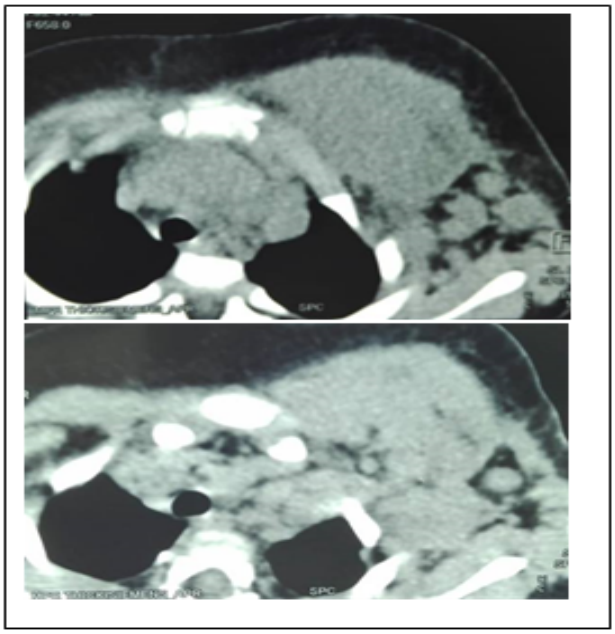

Figure 2: A mediastinal axial Computed Tomography showing an homogeneous left parietal mass opposite the left breast region with a densification of fat under jascente and subcutaneous axillary adenomegaly.

An aspiration biopsy was performed due to the presence of an index case of tuberculosis and despite the absence of its prior contagiousness, the sample was sent for both bacteriologic and histologic study. The histopathological examination indicated a granulomatous lesion, consisted of epithelioid histiocytes and Langerhans giant cells with caseous necrosis and no evidence of malignancy. Mycobacterial stains revealed acid fast bacilli and PCR analysis identified the presence of DNA of *M. tuberculosis* .

However, culture of the specimen was positive to *Mycobacterium bovis* BCG confirming the BCG infection. During follow-up, initial immunologic investigations were found normal, including the nitroblue tetrazolium test, peripheral blood lymphocyte subsets, *in vitro* lymphoproliferative response to mitogens. However, developing a rare complication to BCG vaccination induced us to look for another immune deficiency pathway. A flow cytometric analysis of interleukin-12Rb1 cell surface expression on activated T cells and interferon-gamma cell surface expression on monocytes has been realized. So, a homozygous mutation in the IL12RB1 gene causing abolished expression of IL-12Rβ1 and IL-12 response were identified. The child was treated with isoniazid, rifampicin, ethambutol and pyrazinamide during 2 months followed by isoniazid and rifampin for 4 months. She was followed up monthly and she recovered without any complication.

Discussion

Children living in the area of mild risk tuberculosis such as our country should be vaccinated as early as in the newborn period by Bacillus Calmette-Guerin (BCG) vaccine. It remains the most commonly used vaccine worldwide. It is considered a safe vaccine in immunocompetent children and could lead to some local adverse effects such us local subcutaneous infection and regional suppurative lymphadenitis [5]. Considerable risks and most serious complication following vaccination are seen as rare in individuals with immune deficiencies. Normally after vaccination, it occurs an infiltration mesearing 10 mm or less followed by a well-healed scar not more than 14 months later without association with any other clinical sign. Any evolution different from what has been described must be considered a complication which are estimated to be very rare about 0.01%-3.6% [6]. A Disseminated BCG is one of the most severe complications and it is defined as 'BCG infection being present in more than one anatomical site beyond the region of vaccination'. It occurs generally to children with severe impaired immunity. According to this definition, this case may be considered as a disseminated BCG infection occurring with immunodeficiency [7].

A localization of tuberculosis in the chest wall is rare and diagnoses in most cases are demanding and need significant effort because the lesions widely simulate pyogenic abscess or malignant tumor [8]. Because of the clinical presentation of our case as a malignant process of the chest wall, the absence of hot abscess signs and the difficulty to appreciate chest wall lesion on chest X-ray, the need of resort to an invasive examination was mandatory such us a CT scan. Computed Tomography (CT) imaging is important to assess chest wall abnormalities. Because of its good spatial resolution, the CT makes it possible to define the extent of the lesions, its relations with the neighborhood structure as well as its characteristics evoking a pyogenic abscess [9].

However, in some cases it is difficult to figure out patients the infectious origin that can appear neoplastic as in that case. Biopsy is the golden standard issue to those a confirm diagnosis. However, other less invasive and less costly means of diagnosis can help to provide diagnostic evidence and eliminate other diagnoses such as a cytology aspiration guided by ultrasonography. The proof would be either histological or bacteriolgic [10]. The PCR can be useful compared to standard diagnostics tools. It reduces the diagnostic delay and optimizes the rapid management of the patient. Culture of the mycobactirial stain was positive in 46% up to week 20 after

vaccination. After 20 weeks, no culture became positive. This was not the case of this observation. Twelve months after BCG vaccination, the culture was positive. It confirms the BCG infection.

Chest wall abscess caused by *Mycobacterium bovis* BCG is certainly rare but it must be kept in mind that having this kind of complication should prompt the physician to look for immune disorder. Several types of immune disorder witch are classified into secondary immune disorder like HIV infection and primary immune disorder such as chronic granulomatous disease, Severe Combined Immunodeficiency (SCID) or the immune disorders described as 'Mendelian Susceptibility to Mycobacterial Diseases' (MSMD). MSMD is a rare primary genetic condition increasing susceptibility of affected individuals to infection caused by weakly virulent mycobacteria such as tuberculous, nontuberculous mycobacteria and BCG due to the lack of the main role of IFN-γ axis in immunity against intra-cellular microorganisms [11]. If MSMD is suspected, other immunodeficiency disorders should be ruled out by a careful clinical history, a complete physical exam and basic immunodeficiency screening tests.

In the case of need a specific diagnosis more sophisticated laboratory investigation are required, and that was the case of this presentation. To our knowledge, this is the first case of MSMD due to IL-12Rβ1 deficiency complicated with Chest wall abscess caused by *Mycobacterium bovis* BCG and the first case of this disease to be described in Tunisia. Thus, similar cases might be existing, nevertheless misdiagnosed. There is no consensus in the treatment. The different cases reported in the literature had used different therapeutic means. Generally, the medical treatment, as in our case, was efficient. Sometimes the surgery would be necessary once to make the diagnosis and to flatten the parietal abscess especially in the case of a local complication such as bone osteomyelitis [12]. For MSMD, an appropriate antibiotic therapy is indicated according to the clinical situation. Whereas severe disease may require hematopoietic stem cell transplantation. The prognosis is most often favorable, although it depends on the diagnostic delay.

Conclusion

In conclusion, this observation highlights many particularities of clinical behavior: First, physician must consider tubercular infection even if pulmonary tuberculosis is not associated and even the absence of contagious lesion of the case index. Secondary, A chest wall complication of BCG vaccination should also be considered in the differential diagnosis of the chest wall tumor. Third, thinking to an immune disorder in the face of such complication is important.

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