Case Report

The youngest patient of lupus vulgaris; A cutaneous tuberculosis case report

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ABSTRACT

Tuberculosis, which may involve most organs, is still a major health problem in developing countries. Despite a high and increasing frequency of tuberculosis, cutaneous tuberculosis (CT) is an uncommon form. CT may develop due to Mycobacterium tuberculosis, Mycobacterium bovis, and the Bacille Calmette-Guérin (BCG). CT may have various clinical forms. The most frequent form of CT is lupus vulgaris (LV). LV originates from inactive tuberculosis focus in the body and spreads by hematogenous or lymphatic way and by direct or exogenous inoculation. A diagnosis of LV was made based on clinical and histopathological examination. The lesions regressed after treatment with 3 antituberculous drugs. CT must be considered in cases with chronic skin lesions because tuberculosis prevalence is high in our country. Early diagnosis and treatment of patients with CT is extremely important in order to prevent complications. We report, to the best of our knowledge, the youngest CT affecting case.

KEY WORDS: Cutaneous tuberculosis, Childhood, Youngest.

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INTRODUCTION

Both in developing and already developed countries, tuberculosis (TB) has a high and increasing prevalence.[1] Cases with childhood TB include 5-15% of all TB cases. Pulmonary TB is the most frequent type in children as in adults, and extrapulmonary TB constitutes just 20% of all childhood TB cases.[2] Cutaneous tuberculosis (CT) constitutes approximately 1.5% of all extrapulmonary TB cases in childhood.[3] CT is often seen with malnutrition, low socioeconomic environments, and in crowded societies. Extrapulmonary tuberculosis is very common in early adulthood, and lymph nodes are the most common localizations.[4]

The clinical and pathologic lesions are varying from scrofuloderma to lupus vulgaris (LV) in CT,[6] but the most common form of CT is LV.[5] The most significant problem for the diagnosis of CT is the low positive cultures results.[5] Here, we describe a young girl with LV involving the left face and arm, and she may be the youngest case with the diagnosis of CT in the literature.

CASE REPORT

An 18-month-old nonimmunized girl presented with a six month history of pink plaques appearing and progressing slowly on the left face and the posterior surface of the left arm (Fig 1 and 2). Clinical

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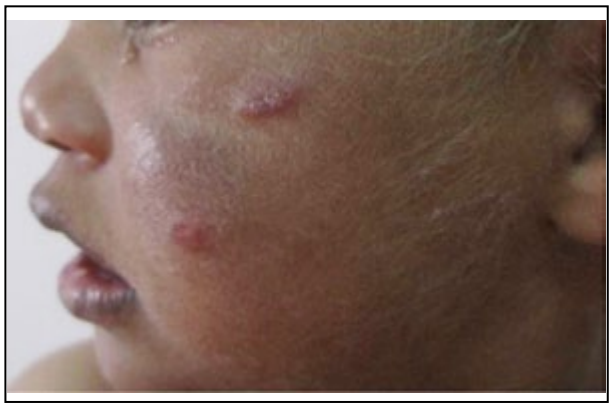

Fig.1: The lesions of lupus vulgaris on left face.

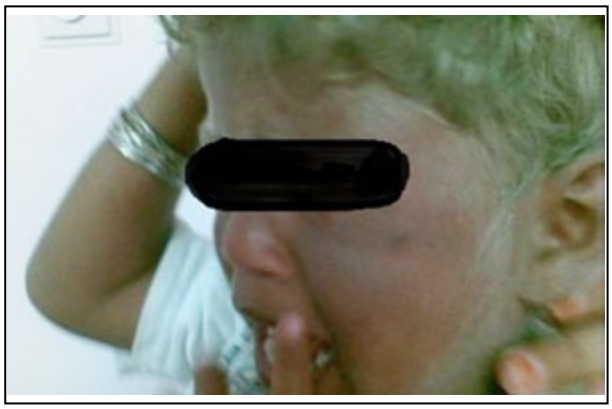

Fig.2: End of treatment.

examination revealed regularly bordered, pink lesion on the left face and posterior of the left arm. Apple-jelly color was seen when examined by diascopy. There was no regional lymphadenopathy, and systemic examination revealed no abnormalities. No other family members had similar lesions.

Routine biochemical analysis, complete blood count, and urine microscopy were all normal, Chest radiograph and computed tomography findings were normal, and no sign of pulmonary tuberculosis was present. The purified protein derivative test (Mantoux test) showed normal reactivity with a 18mm in duration after 48 hours. Histopathological examination of the biopsy samples showed normal epidermis and caseating tuberculoid granulomas containing epithelioid histiocytes and Langhans giant cells in the papillary dermis (Fig.3).

The standard short-course chemotherapy for treatment of cutaneous tuberculosis which involves the administration of three antituberculous drugs for the first two months (isoniazid 10 mg/Kg, rifampicin 10 mg/Kg, pyrazinamide 30 mg/Kg), followed by four months of isoniazid and rifampicin was started. Marked improvement of the lesions with hyperpigmentation was seen by the end of six months treatment (Fig.2).

DISCUSSION

The infection mechanisms of CT are direct inoculation, local invasion, or hematogenous dissemination, and these infections are classified as multibacillary and paucibacillary.[6] Although, LV is the second most frequent form in the children, it is the most common clinical form of CT in adults.[6,7] On the other hand, Ramesh et al have found as the most common form of CT in children (63.5%).[4,5,8] Our patient may be the youngest case of CT in the literature. Most cases, especially children with primary TB, carry the lesions on extremities and on face following scratching, bruising, lacerations, pin-pricks, impetigo, boils, piercing, tattoos, and circumcision.[6] LV might occur at the site of BCG vaccination suggesting exogenous inoculation of the infection.[8] Our patient had no BCG scar. Therefore, we concluded that the patient was not immune to TB previously.

Many types of infections have been reported including classic plaque or keratotic type, hypertrophic, ulcerative, atrophic, and planar. Keratotic type of them is the most frequent whereas ulcerative and atrophic forms are the least common in children.[8,9] Type of the cases lesion was hyperkeratotic and ulcerative. Although involvement of regional or systemic lymph nodes is commonly seen in LV and scrofuloderma,[6] there was no local or systemic lymphadenopathy in our case. Kumar et al[3] and Pandhi et al[8] have observed very high rates of Mantoux test positivities in their

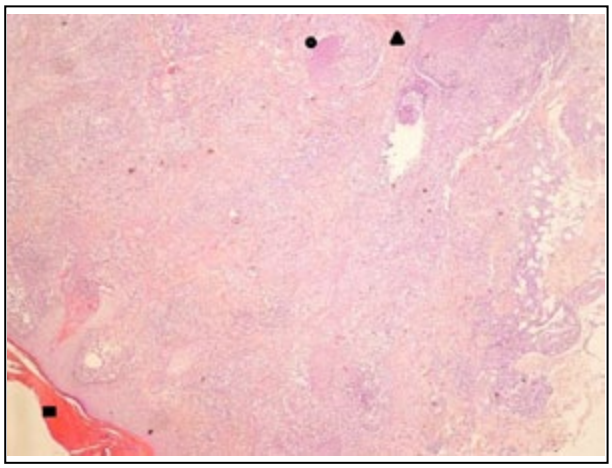

Fig.3: Structure of hyperkeratosis (■), epithelioid hystiocytes (▲), granuloma and caseification necrosis (●).

series (97.1% and 91.8%, respectively). Mantoux test of the patient showed positively reactivity with a 18 mm indurations after 48 hours. The histopathologic examination showed the tubercules which are hallmarks of cutaneous tuberculosis. They consist of accumulations of epithelioid histiocytes with Langhans giant cells and varying amount of caseation necrosis in the center.[9] In our case, incisional biopsy specimen showed normal epidermis with caseating tuberculoid granulomas consisting of epithelioid histiocytes and Langhans giant cells in the papillary dermis.

The initial treatment should be three or four drugs combinations including isoniazid, rifampicin, pyrazinamide or/and ethambutol, and the clinical healing is initiated to be seen in 4-6 weeks.[10] Five weeks of therapy may be tried in cases of strongly suspected CT.[10] Our treatment was three antitubercular drugs for the first two months (isoniazid 10 mg/Kg, rifampicin 10 mg/Kg, pyrazinamide 30 mg/Kg), followed by four months of isoniazid and rifampicin was started. Partial remission was seen after three weeks, and marked improvement of the lesions with hyperpigmentation was seen by the end of six months in our case.

In conclusion, the diagnosis of CT is based on clinical features, demonstration of acid-fast bacilli on smear, tissue culture, skin biopsy, and PCR in recent years. However, the diagnostic value of culture and PCR is less and diagnosis may be dependent on clinical features, histopathological findings, and retrospective review of response to treatment. The purpose of this case report was to emphasize that the diagnosis of LV depends chiefly on clinical suspicion and histopathological features even in children.

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