Review

Tuberculous gummas with sporotrichoid pattern in a 57-year-old female: A case report and review of the literature

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ABSTRACT

Sporotrichoid tuberculosis is a rare form of cutaneous tuberculosis; it primarily affects children after a post-traumatic inoculation. The diagnosis is often difficult and based on a set of arguments; it should be considered in any sporotrichoid lesion, especially in tuberculosis endemic countries. The following describes a new case of Mycobacterium tuberculosis skin infection with an unusual sporotrichoid clinical appearance in a healthy woman, emphasizing the diagnostic difficulties with a review of literature.

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Introduction

Tuberculosis is a bacterial infection caused by Mycobacterium tuberculosis (MTB); it is endemic in several countries, including Morocco (25,000 new cases/year). Cutaneous tuberculosis is the fifth most common form after pleuropulmonary,
Table 1 – Details of patients with sporotrichoid cutaneous tuberculosis in literature.

<table>
<thead>
<tr>
<th>Age (years)/sex</th>
<th>Clinical features</th>
<th>Satellite lymphadenopathy</th>
<th>Duration of disease</th>
<th>Entry mechanism</th>
<th>Investigations</th>
<th>Treatment</th>
<th>Evolution</th>
</tr>
</thead>
<tbody>
<tr>
<td>Premalatha [2]</td>
<td>17/F</td>
<td>Crusted plaques/ right lower limb</td>
<td>+</td>
<td>6 months</td>
<td>ND</td>
<td>TST = 25 m Histo = EGGC, NC+, AFB+</td>
<td>Streptomycine, INH, Ethambutol</td>
</tr>
<tr>
<td>Remenyik [3]</td>
<td>12/F</td>
<td>2 Nodules/ right upper limb</td>
<td>–</td>
<td>6 months</td>
<td>Minor trauma</td>
<td>TST = 13 mm Histo = EGGC, CN–, AFB–, PCR+</td>
<td>INH, Rifam Ethambutol</td>
</tr>
<tr>
<td>Ramesh [1]</td>
<td>11/M</td>
<td>Plaques/ right lower limb</td>
<td>+</td>
<td>2 years</td>
<td>ND</td>
<td>TST = 16 mm Histo = EGGC, AFB–</td>
<td>DOTS(INH, Rifam PZA)</td>
</tr>
<tr>
<td></td>
<td>20/F</td>
<td>3–4 ulcers/ left lower limb</td>
<td>+</td>
<td>3 months</td>
<td>ND</td>
<td>TST = 20 mm Chronic inflammatory infiltrate Few epitheloid cell granulomas AFB–</td>
<td>DOTS</td>
</tr>
<tr>
<td>10/F</td>
<td>Keratotic plaques/ left lower limb</td>
<td>+</td>
<td>1 year</td>
<td>ND</td>
<td>ND</td>
<td>TST = 10 mm, AFB+, Histo = granulomatous inflammation</td>
<td>DOTS(INH, Rifam PZA)</td>
</tr>
<tr>
<td>Gökay [4]</td>
<td>87/F</td>
<td>Ulcerative nodules/ right upper limb</td>
<td>ND</td>
<td>1 year</td>
<td>ND</td>
<td>TST = 10 mm, AFB+, Histo = granulomatous inflammation</td>
<td>Levofoxacine Clarithromycine</td>
</tr>
<tr>
<td>Wilson [5]</td>
<td>13/M</td>
<td>2 Ulcers/ right lower limb</td>
<td>+</td>
<td>1 month</td>
<td>Visit to Ethiopia</td>
<td>TST = 19 mm, Histo = EGGC + necrosis Culture+</td>
<td>Streptomycine INH Ethambutol</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Fluctuant masses/ left upper limb</td>
<td>+</td>
<td>ND</td>
<td>ND</td>
<td>MRI upper limb = intraosseous abscess Computed tomography of the chest = lymphadenopathy neck, mediastinum MRI of the spine = signal abnormalities</td>
<td>ATT</td>
</tr>
<tr>
<td>Zarra [7]</td>
<td>7/F</td>
<td>Gums/ right upper limb</td>
<td>+</td>
<td>7 months</td>
<td>Spread from visceral localization</td>
<td>TST phlyctenular AFB– Histo = non specific Chest X rays = pleural effusion</td>
<td>ATT</td>
</tr>
</tbody>
</table>

F = female, M=male; + present; – absent; ND = non determined; TST=tuberculine skin test; histo = histopatholgy; EGGC = epithelioid cell granuloma gigantocellular; CN = caseation necrosis; AFB = acid-fast bacilli; INH = isoniazid; PZA = pyrazinamide; PCR = polymerase chain reaction; DOTS = Directly Observed treatment, Short course; ATT = antitubercular treatment; MRI = magnetic resonance imaging; TTE = Transthoracic echocardiography.
glandular, digestive and urogenital tract tuberculosis. The main clinical presentations are scrofuloderma and gumma. Sporotrichoid form is an exceptional form. This study reports a case of sporotrichoid cutaneous tuberculosis with review of the literature (see Table 1).

Case report

A 57-year-old woman without medical history was hospitalized for gummas of the right lower member lasting for 2 years, starting at the back of the foot, with no notion of past trauma and extending upward to the root of the thigh. The examination revealed multiple erythematous subcutaneous nodules with ulceration and purulent discharge by location, along the right lower extremity (Figs. 1–3) and ipsilateral inguinal lymphadenopathy. The sputum smear microscopy was negative for Acid Fast Bacilli (AFB); TST (tuberculin skin test) was positive (23 mm); skin biopsy showed an epithelioid cell granuloma gigantocellular with suppuration (Figs. 4 and 5); and PAS (Periodic Acid Schiff) staining did not reveal fungal elements. A repeated smear microscopy in search of bacteriological, parasitological and mycological elements with culture in special medium remained negative. Bone radiography of the lower right limb was normal and HIV serology was negative. The patient received an anti-tubercular treatment (2 months of Rifampicin, Isoniazid and Pyrazinamide and 4 months of Rifampicin and Isoniazid) with draining lesions after 2 months, regression of lymphadenopathy and complete healing after 5 months, and a follow-up period of 6 months. In this case, the diagnosis of tuberculous gummas with sporotrichoid pattern was based on the epidemiological situation, the TST, demonstration of granulomatous dermatitis, and regression following anti-tubercular therapy.

Discussion

Tuberculous gummas with sporotrichoid pattern are exceptional [1]. This form of tuberculosis was described for the first time by Premalatha [2] in 1987 in India, and then it has been reported in other countries (Hungary [3], India [1], Turkey [4], Canada [5], and Tunisia [6]). A review of the literature found only 10 cases of sporotrichoid tuberculosis, including the patient in this study, with a female predominance (8 female patients/2 male). The majority of patients (6 patients) were children, and 4 adults with a mean age of 28.1 years. The point of entry in sporotrichoid tuberculosis is often a post-traumatic skin wound [5]. Lesions involved the upper limbs and lower limbs equally (5/5) with a predominance of the...
right side (6/10) and may take the form of plaques [1] [2], nodules [3,4], or gummas [6]; moving by the classic stages of crudity, softening fistula/ulceration with possible progression to scarring after several months, they have the particularity to have a linear distribution along a lymphatic pathway [7], often with satellite lymph nodes (8/10). Sporotrichoid cutaneous tuberculosis, as its name indicates, mimics the clinical aspects of sporotrichosis; the main differential diagnoses are: leishmania, sporotrichosis, nocardiosis, atypical mycobacteria (Mycobacterium marinum), pyogenic infections (Staphylococcus aureus, Streptococcus), and deep fungal infections [3,8].

The diagnosis is difficult to make, and it is based on a set of arguments. The bacteriological examination can isolate the germ in only one third of cases; the culture on Lowenstein-Jensen medium is often negative. Histological study does not always provide definitive diagnosis [7], and the culture of biopsy specimen is recommended [5]. TST is often positive or phlyctenular, which is the same as the case of multifocal tuberculosis reported by Zarra [6] from Tunisia. The new genomic amplification techniques (PCR) now allow the rapid identification of the organism, but they are rarely realized in this context because of their cost, as is the case in this study.
Several therapeutic protocols are available; the most used is DOTS (directly observed treatment, short course), with a minimum of 6 months of treatment. A correct anti-tubercular treatment gives generally good results.

Dissemination in cases of sporotrichoid cutaneous tuberculosis is often made from a post-traumatic skin wound [5], but some authors speak of forms called segmental or inverse sporotrichoid [1], when the proximal half of the lower or upper limb was affected due to retrograde lymphatic spread from the lymph nodes or any other endogenous sites [5].

This form of tuberculosis is more common in cases of immunosuppression, in children and young adults, because of the efficient lymphatic drainage, their high physical activity which makes them more prone to trauma [1], but which was not the case in this patient.

Conclusion

This case illustrates the diagnosis difficulties caused by the sporotrichoid cutaneous tuberculosis; however, this diagnosis should be considered with any lesion showing a linear arrangement along the lymphatic vessels, especially in tuberculosis endemic countries.

Conflict of interest

None declared.