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Papulonecrotic Tuberculide: A Case Report

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Abstract:

A 20 year old lady presented with asymptomatic papulo-pustular eruption over upper, lower extremities and the abdomen. There was no manifestation of tuberculosis elsewhere in the body. Investigations were suggestive of papulonecrotic tuberculide.

Key words: *Tuberculide, Papulonecrotic*

Introduction

Tuberculosis is still a major cause of morbidity in developing countries, like Bangladesh. Cutaneous tuberculosis is therefore not uncommon in this community. The concept of "tuberculide" was first introduced by Darier in 1896. Later pautrier established the fact that papulonecrotic tuberculide was a tuberculosis associated condition. It represents cutaneous autoimmune type IV hypersensitivity reaction to the bloodstream dissemination of mycobacteria. In most of the cases evidence of tuberculosis is seen elsewhere in the body.

Three particular conditions are suggested as jenuine tuberculides. These are erythema induratum of bazin (EIB), papulonecrotic tuberculide (PNT) and lichen scrofulosorum(LS). We present a case of papulonecrotic tuberculoses in a young adult lady without any underlying tuberculous focus elsewhere.

Case Report

A 19 year old female was first seen in the outpatient department of a private hospital of Dhaka, Bangladesh in September 2012 with numerous active papulonecrotic lesions, together with old healed scars of similar origin on thighs, legs, forearms and anterior abdominal wall. These lesions were present for about three months and followed an acute episode of high fever in mid August. There was no constitutional symptom or respiratory problem at the time of presentation. There was associated pain and fever, but no respiratory problem at the time of presentation. There was no history of drug intake, nor there any personal or family history of tuberculosis. She had not been vaccinated with BCG.

On clinical examination, the lesions seemed papulopustular, most of which have undergone necrosis to form deep ulcers (Fig.1-4). The lesions were symmetrically distributed and there size ranged from 5-25 mm in diameter. Lesions were

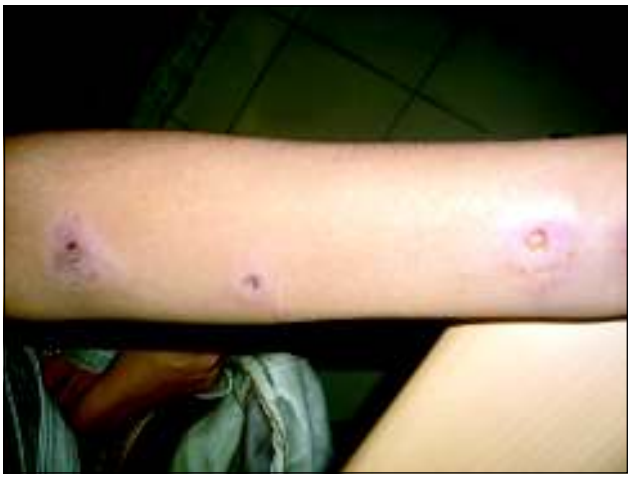

Fig.-1: *Papulonecrotic lesions on flexor surface of forearm*

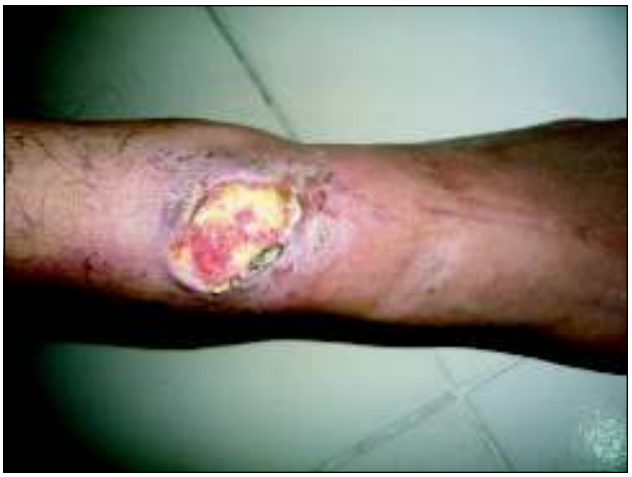

Fig.-2: *Deep ulcer on the left foot.*

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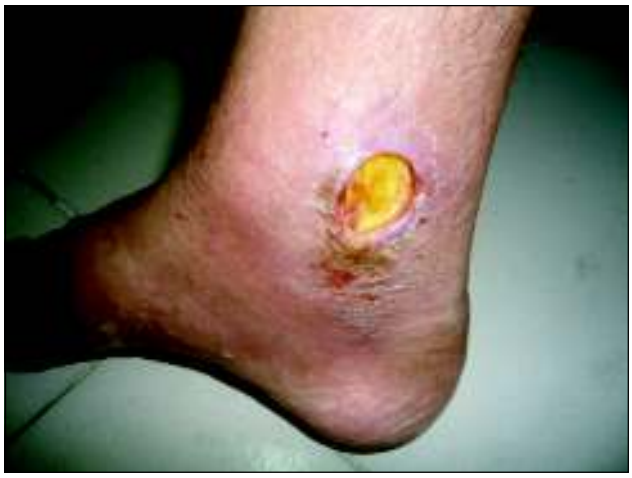

Fig.-3: *Ulcer in the right foot.*

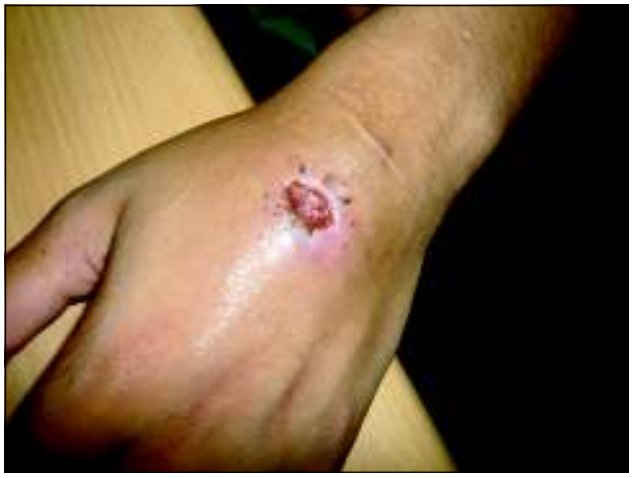

Fig.-4: *Ulcer in the left hand.*

found in both flexor and extensor surfaces. A few lesions were seen in the anterior abdominal wall just above the umbilicus. There was no significant inguinal or axillary lymphadenopathy. Systemic examinations were normal.

The hemogram revealed a raised erythrocyte sedimentation rate (42 mm in first hour) and neutrophilic leukocytosis. Culture from purulent discharge was negative for bacterial pathogen or fungus. Antinuclear antibody test was negative, C-reactive protein was 9.39 mg/L and random plasma glucose was 5.90mmol/L. VDRL and HIV serology was non-reactive. Chest x-ray was unremarkable. The Manteaux test was strongly positive (16 mm). PCR test for mycobacterium tuberculosis was not performed however because of its high cost.

Aspirate from a soft nodule in left forearm revealed features of tubercular chronic granulomatous inflammation.

A full thickness incisional biopsy was performed from a lesion on dorsal aspect of her right hand. The epidermis was deeply ulcerated. The dermis and superficial subcutis showed a well circumscribed zone of necrosis and mixed infiltration with lymphocytes and polymorphs. Well established granulomas composed of epithelioid histiocytes are seen at the periphery (Fig 5&6). Few multinucleated Langhan's type giant cells are seen as well. No distinct perivascular inflammatory feature is seen in adjacent areas. The histological diagnosis was granulomatous dermatitis consistent with papulonecrotic tuberculide. Special stain however failed to demonstrate tubercle bacilli.

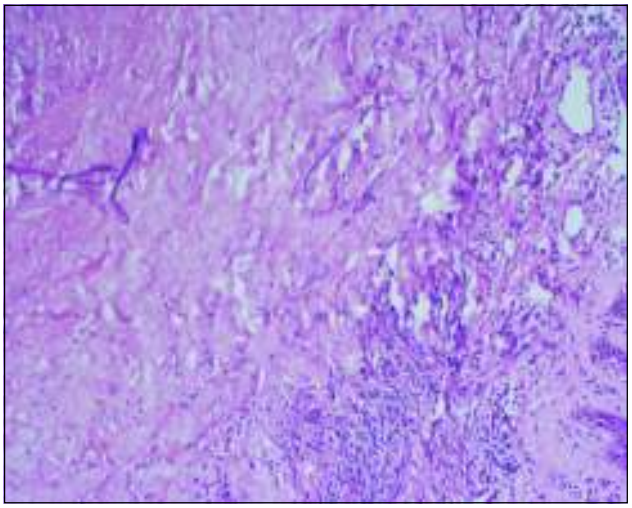

Fig.-5: *Wide caseation necrosis involving deeper dermis*

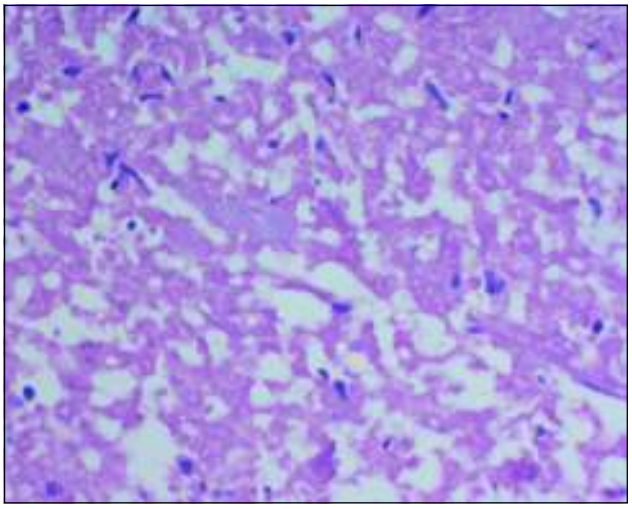

Fig.-6: *Caseation necrosis with scattered epithelioid cells*

Patient showed unequivocal response to antituberculous therapy. Her lesions started healing very fast and by three months of therapy almost all of her lesions were healed by a depressed scar formation.

Discussion

Tuberculides are the cutaneous manifestations of cell-mediated (Type IV) hypersensitivity reactions to an internal

focus of tuberculosis or disseminated mycobacteria in a patient with moderate to high degree of immunity. The usual feature is symmetrical eruptions in the extensor surfaces of the extremities. The sites of predilection are the elbows, knees, legs, hands, feet etc. Ears, face, buttocks and penis can be rarely involved.[1]

Papulonecrotic tuberculide is a rare condition and can be seen in areas where mycobacterium tuberculosis infections are prevalent. e.g., South Africa, India, Southeast Asia. The basic diagnostic criteria for papulonecrotic tuberculide are –

- A strongly positive tuberculin test.
- A tuberculoid histology with endarteritis and /or thrombosis of dermal vessels.
- Resolution of the lesion with anti-tuberculous therapy.

Histological findings may however be inconclusive and a range of patterns including acute inflammatory cellular infiltrate with nonspecific vasculitis to well developed palisaded granuloma formation. The tubercle bacilli can not be identified on tissue staining and cannot be cultured either. But PCR of affected tissue can detect DNA of the bacilli. In our case, light microscopy revealed triangular area of necrosis surrounded by epithelioid histiocytes. Occasional langhan's giant cells are also noticed. No associated vasculitis is seen however.

Clinically, the lesions usually present as ulcerated pustules occurring primarily on extensor surfaces of upper and lower extremities.[2] our case presented with symmetrical lesions in both upper & lower extremities but the lesions are largely distributed in flexor surfaces and in addition anterior abdominal wall was also involved. Some of the lesions healed by formation of a depressed scar. Ost authors have described 80% of the lesions to present in extremities only.[2]

Some case reports described solitary lesions on penis appearing as ulcerations or nodules without ulceration.[3,4] PNT (Papulonecrotic tuberculide) is often reported in adults than in children. There was however a study reporting PNT in eight children of south Africa.[5] In Hong Kong, a ten year retrospective study showed that PNT accounted 4% of all cases of cutaneous tuberculosis diagnosed during this period and all of them(n=7) were within the age range of 18-43 years.[2]

True cutaneous tuberculosis other than tuberculides include Lupus vulgaris (LV), Tuberculosis verrucosa cutis(TVC) and Scrofuloderma (SFD). Coexistence of different forms of cutaneous tuberculosis had also been reported by some authors.[6,7] Simultaneous occurrence of two tuberculides were also reported.[8] In adult onset type of PNT a concomitant occurrence of EIB and LV can be documented in the same patient.[9]

PNT cases are frequently associated with extra cutaneous tuberculosis. In some series, 40% of cases had extra cutaneous involvement, of which 27% had tuberculous lymphadenitis.[10] our case however no extra cutaneous manifestation had as per reports of other relevant investigations. Nor was there any association with other forms of cutaneous tuberculosis or tuberculide.

In some of the early lesions, as reported by some authors, a full-fledged granulomatous lesion can not be identified.[11] The histological picture may be of a nonspecific necrotic focus or in some cases may mimic granuloma annulare.[11] in our case; the initial suggestion came from cytological examination which revealed collections of epithelioid cells against mixed inflammatory cellular and necrotic background. Subsequent histopathology revealed a typical triangular necrotic area surrounded by well developed granuloma.

Our patient had a brief episode of fever followed by skin eruptions which probably indicate a bacteremia with mycobacterium tuberculosis initiating the hypersensitivity response. The differential diagnosis on the basis of this clinical presentation was erythema induratum. The tissue diagnosis however confirmed the lesion as an unequivocal case of tuberculosis.

PCR is a very useful test for diagnosis of mycobacterial DNA in clinical specimens. Though a negative result does not rule out this disease but positive result can decrease the likelihood of missing the diagnosis and subsequently causing delay in appropriate treatment and thus increasing patient morbidity. Keeping under consideration the FNA report our patient was advised for PCR for m tuberculosis.

As the patient could not afford the test, a prompt incision biopsy in an outdoor setup was done for coming into a conclusive diagnosis. In order to avoid an invasive procedure like biopsy, a combination of cytology (FNA or scraping) and PCR analysis may be advisable for starting an anti-tuberculous therapy in earliest possible time. A good anti-TB drug response further establishes the diagnosis.

Conclusion:

Treatment of PNT should be a combination of anti-tuberculous drug therapy such as that received in our case with an excellent outcome. Nowadays PNT is a rare occurrence; physician however should remain alert for this disease as a sign of tuberculosis especially in countries where tuberculosis is endemic.

Conflict of Interest : None

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