case report

Lupus vulgaris in a young girl

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With the estimated global burden of TB being 8.8 million incident cases and 1.1 million deaths from TB in HIV-negative cases and additional 0.35 million deaths in HIV-associated cases,[1] the total number of cutaneous TB cases (<1-2 % of total cases) becomes significant. With the WHO setting up public-private mix partnerships and a millenium development goal of a 50% reduction in the total number of incident cases, the case detection and reporting of unusual cutaneous TB cases becomes very important. We present a case of lupus vulgaris in a young girl with rapid progression of a large plaque with hypertrophic features in the periphery. The case is unusual due to its rapid progression, unusual site and extensive giant form which have never been reported previously.

Lupus, meaning wolf-like, initially referred to any ulcerative lesion, reminiscent of a wolf bite. In 1803, Robert Willand used the word lupus as a reference to the latter stages of facial cutaneous TB. This clinical description of facial lupus was the origin of similar terms such as lupus erythematosus and lupus pernio (a variant of sarcoidosis). Also called as tuberculosis luposa, it is a post-primary, paucibacillary variant developing in individuals with moderate immunity and a high degree of tuberculin sensitivity. It originates by hematogenous, lymphatic or contiguous spread from TB elsewhere in the body, the most common sites of latter being cervical adenitis or pulmonary TB. Rarely, it follows BCG vaccination. We report a case of giant lupus vulgaris of the plaque type in a young girl with hypertrophic variant in the periphery.

CASE

A 17-year-old girl presented with asymptomatic, slowly progressing lesions present bilaterally on the submandibular areas, neck, anterior chest, axillae, back and occipital area for the previous 5 to 6 years. The lesion progressed from a small papule and gradually enlarged to involve all the above areas.

On examination, a well-defined, hypopigmented, single large plaque (45×30 cm approximately) with central atrophy, telangiectasia and islands of normal appearing skin were present. The periphery showed a gyrate outline with crusted, verrucous appearing lesions and erythema (**Figure 1A**). The left axillary region and submandibular region showed scarring signifying adenitis in the past. The back lesion extended up to the lower back. The lesions started 5 to 6 years before for which she took medication (probably antibiotics also), the details of which were unavailable.

The hemogram and biochemical investigations were all within normal limits apart from relative lymphocytosis and raised a erythrocyte sedimentation rate of 70 mm in the first hour. The Mantoux test was strongly positive being a 17 mm induration in greatest diameter. The chest x-ray showed no significant active lesion or changes. She had no cough and sputum for acid-fast bacilli (AFB) was negative. ELISA for HIV was non-reactive. PCR for *M tuberculosis* was negative. Biopsy of the peripheral crusted lesion showed hyperkeratosis, acanthosis and papillomatosis. Tuberculoid granulomas composed of epitheloid and Langhans giant cells were seen predominantly in the upper dermis. Associated lymphocytic infiltrate was also seen. AFB staining was negative for tubercle bacilli (**Figure 2**).

Because of the large plaque size and rapid progression, she was put on category I anti-tubercular treatment under DOTS with $2HRZE_3$, isoniazid, rifampicin, pyrazinamide and ethambutol given three times weekly for 2 months followed by $4HR_3$ given three times weekly for 4 months. At follow up after 3 months

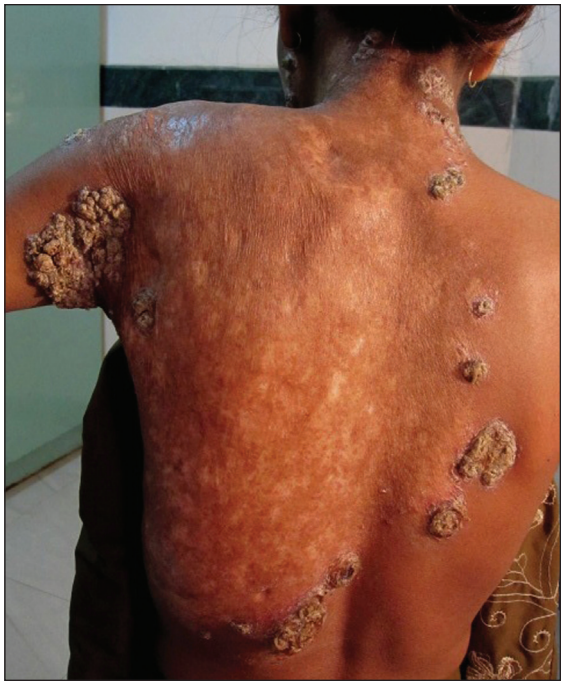

Figure 1A. The periphery showed gyrate outline with crusted, verrucous appearing lesions and erythema.

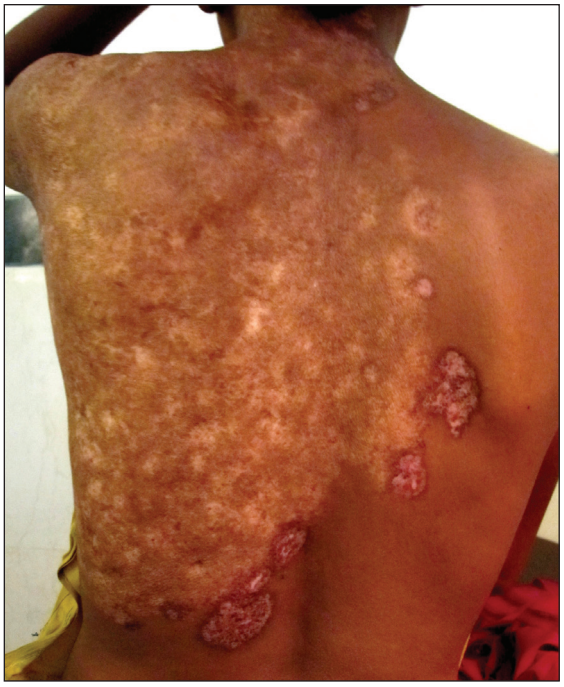

Figure 1B. The follow up after 3 months of therapy showed approximately 90% clearance of the lesions.

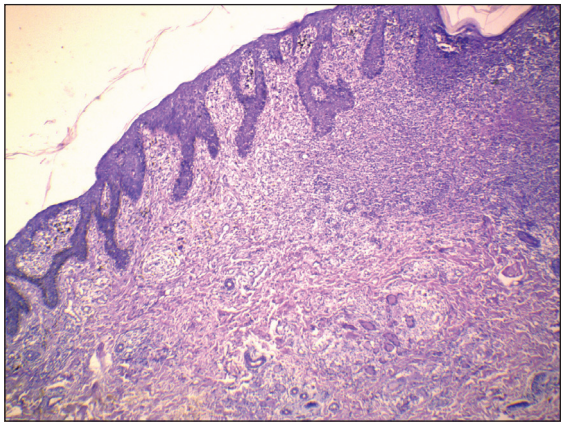

Figure 2. Tuberculoid granulomas composed of epitheloid and Langhans giant cells were seen predominantly in upper dermis. Associated lymphocytic infiltrate was also seen. AFB staining was negative for *tubercle bacilli.*

of therapy there was approximately 90% clearance of the lesions (**Figure 1B**).

DISCUSSION

Lupus vulgaris is the most commonly encountered variant of cutaneous TB in adults in most studies.[2,3] Atypical localization and clinical presentations may occur and lead to difficulty in diagnosis and treatment.[4-7] Some unusual sites involved by this variant have been extremities, trunk, buttocks, the perianal region and even the scalp.[6,7] The characteristic morphological patterns are papular, plaque, nodular, tumid, atrophic and ulcerative forms. Atypical clinical forms such as cellulitis, lichen simplex chronicus, sporotrichoid, frambesiform, lichenoid, gangrenous, ulcero-vegetant and tumor-like have also been rarely encountered.[2,6-12] The gold standard for establishing the diagnosis is culture of tubercular bacilli.

The case is worth presenting due to the large size of the plaque, the rapid progression, the presence at a young age and the large central atrophic area with peripheral hypertrophic features of lupus vulgaris. Despite all the advances in microbiology, including sophisticated techniques such as PCR, the sensitivity of new methods are no better than the gold standard, that is, the isolation of *Mycobacterium tuberculosis* in culture. Even now, we rely on old methods as the intradermal reaction purified protein derivative standard test and therapeutic trials, as diagnostic tools. Also with the recent WHO 'negative' policy recommendation on TB regarding serological commercial tests, it is important to recognize the many presentations of cutaneous tuberculosis to prevent missed or delayed diagnoses by having a strong clinical suspicion and supporting tests.

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