Atypical Presentation of Lupus Vulgaris: A Case Report

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Abstract: Lupus vulgaris is the most common form of cutaneous tuberculosis. It is acquired by inoculation from injury or by hematogenous spread from an infective focus. Clinically, it varies tremendously from erythematous plaque to destructive and perforating lesions. Thus the diagnosis of lupus vulgaris becomes a challenge to clinician at times. Atypical forms can mimic various other dermatological conditions leading to delay in diagnosis and increased morbidity. Here we report a rare presentation of lupus vulgaris which was mimicking mycetoma or osteomyelitis. Strong clinical suspicion, histopathology and response to anti-tubercular treatment led to the diagnosis. In this report, emphasis on the early diagnosis and prompt treatment is made.

Keywords: Lupus vulgaris, mycetoma, osteomyelitis, atypical form, histopathology.

INTRODUCTION

Cutaneous tuberculosis (TB) forms an important domain of extra pulmonary TB [1]. The incidence of cutaneous TB is around 5.9 per 1000 population. Lupus vulgaris (LV) is the most common morphological variant of cutaneous TB and constitutes 74% of total cutaneous TB cases [1]. Although the incidence of cutaneous TB has fallen from 2% to 0.1% among skin outpatient departments, atypical forms with varied manifestations are being reported from all over the world [2]. These atypical forms pose a great challenge for diagnosis due to paucity of bacilli. This is usually observed in developing countries where confirmatory tests like polymerase chain reaction (PCR) are not easily available which makes it extremely difficult to diagnose LV [3]. Here we report an atypical presentation of LV that masqueraded as mycetoma or osteomyelitis and detected by histopathology. Aim of this presentation is to emphasize high index of clinical suspicion, histopathology and empirical anti-tubercular treatment in this chronically scarring but definitely curable disease.

CASE REPORT

A 60-year-old housewife from urban area presented with a soft tissue swelling over dorsal aspect of left hand of 6 months duration. Onset was insidious with history of preceding trauma 2 months prior to appearance of skin lesions. The swelling slowly grew in size with formation of multiple sinuses exuding serous fluid (Figure **1**). Tenderness was present. Potassium hydroxide (KOH) preparation for fungus was negative.

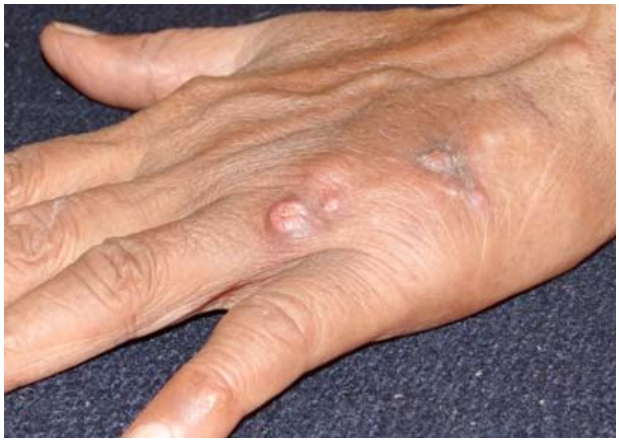

Figure 1: Soft tissue swelling over dorsum of left hand with multiple discharging sinuses.

Culture for fungus showed no growth. X-ray of the part was normal. She was treated with systemic antibiotics with no relief of symptoms. Hematological and biochemical profile and chest X-ray were within normal limits. Blood test for human immunodeficiency virus (HIV) was nonreactive. Mantoux test was negative. There was no history of fever, loss of weight or appetite. Skin biopsy was taken with 4mm punch. Histopathological examination showed dense tuberculoid granulomas in mid-dermis with Langhans giant cells without caseation (Figure **2**). Stain for acid fast bacilli (AFB) was negative. Periodic acid Schiff stain for fungus was negative. Empirical short course regimen of antitubercular therapy (ATT) consisting of rifampicin, isoniazid, pyrizinamide and ethambutol for two months and only rifampicin with isoniazid for four months was started with excellent results. The mass reduced in size and sinuses healed during the course of ATT and skin lesions (Figure **3**) as well as granulomas (Figure **4**) resolved completely after completing 6 months of ATT. Depending on histopathology and response to ATT, our final

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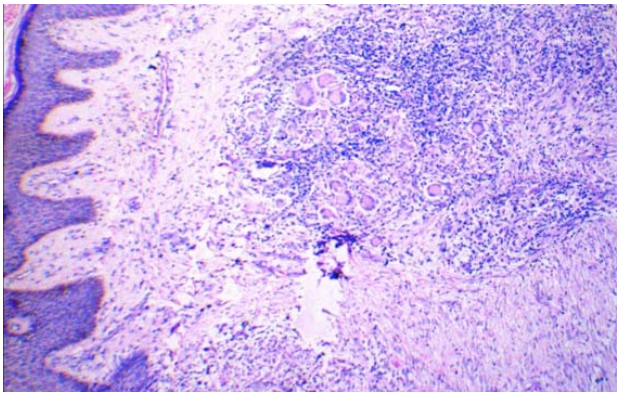

Figure 2: Tuberculoid granulomas in mid-dermis with Langhans giant cells (H&E x10).

diagnosis was lupus vulgaris. Mode of spread may be primary inoculation due to injury.

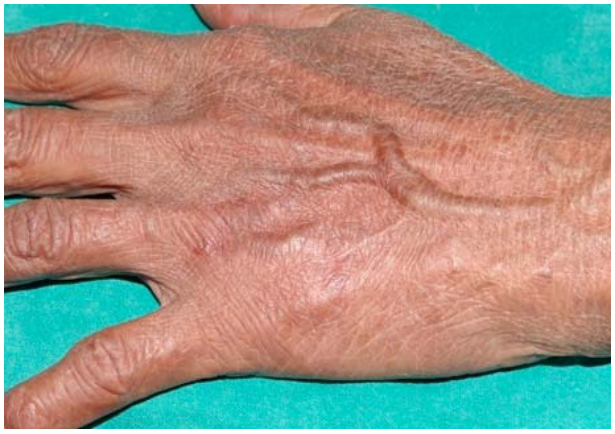

Figure 3: Complete remission of skin lesions after anti-tubercular treatment.

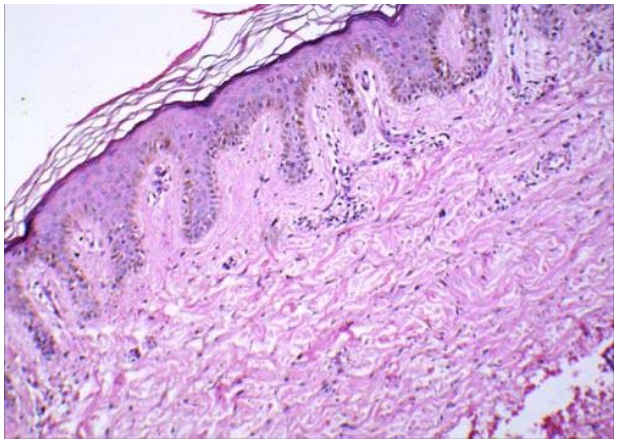

Figure 4: Disappearance of tuberculoid granulomas after treatment (H&E x10).

DISCUSSION

Erasmus Wilson coined the term “Lupus” to emphasize ulcerating and devouring character of lesions comparing the lesions to the ravages of wolf [1]. LV can be acquired either exogenously by direct inoculation or endogenously by hematogenous or lymphatic spread from an underlying infected focus [2]. Gorpade reported two cases of LV occurring at site of tattoo as primary inoculation [4]. Our patient gives history of trauma at the site prior to appearance of these lesions and no focus of TB was detected anywhere else in the body suggesting spread by primary inoculation.

Various unusual forms of LV are reported in literature and these forms are frequently elusive as it mimics a wide differential diagnosis. Heo *et al.* described a case of LV which was misdiagnosed as tinea and treated for 10 years without relief [5]. Hence misdiagnosis of LV occurs sometimes due its sporadic presentation in atypical forms. Saritha *et al.* described three cases of LV mimicking actinomycosis and mycetoma which were diagnosed by histopathology and lesions resolved completely with ATT [6]. Our patient presented with lesions resembling mycetoma or osteomyelitis affecting the left hand, however, histopathology and excellent response to ATT led to the confirmation of diagnosis.

LV destructed partial nose and caused endophthalmitis in a 17-year-old emaciated girl who had not received Bacillus Calmette Guerin (BCG) vaccine during childhood [7]. This clearly indicates the importance of unusual forms of LV as well as early diagnosis and prompt treatment. Jain *et al*. presented a case with giant sized hypertrophic LV covering almost left half of the chest and accounted for the importance of histopathology in such unusual presentations of LV [8]. Tissues smears and cultures from LV lesions are usually negative due to paucibacillary nature of disease and PCR tests must be employed where ever appropriate [2, 3]. Hsiao *et al*. described the utility of PCR in detection of *Mycobacterium tuberculosis* in tissues showing granulomas without demonstrable AFB in immunocompromised patients, however, its usefulness in immunocompetent patients is controversial [9]. In developing countries like India such tests are not easily available and not affordable, therefore clinicians must rely on strong clinical suspicion, histopathology and positive response to ATT to confirm diagnosis [10].

CONCLUSION

Although LV is common, its atypical and protean forms can challenge the diagnostic skills of treating clinician. The discharging sinuses in our patient were

mimicking osteomyelitis or mycetoma, making it difficult to diagnose. Histopathology proved the diagnosis. Hence high index of suspicion, histopathology and response to ATT play a vital role in confirmation of diagnosis when PCR is not available in such case scenario.

What is known- Lupus vulgaris can present with various atypical forms.

What is new- Lupus vulgaris can also present with discharging sinuses which is extremely rare and skin biopsy should be done for histopathological confirmation of diagnosis.

CONFLICT OF INTEREST

Authors declare no conflict of interest.

Human studies were conducted with the patient's informed written consent.

REFERENCES

[1] Bhandare CA, Barad PS. Lupus vulgaris with endophthalmitis - A rare manifestation of extrapulmonary tuberculosis in India. Indian J Tuberc 2010; 57: 98-101.

[2] Yates VM, Rook GAW. Mycobacterial infections. In: Burns T, Breathnach S, Cox N, Griffiths C, eds. Rook's Textbook of Dermatology, 7th edn. London: Blackwell Science Limited 2004; pp. 28.1-28. 39.

[3] Gerhard T. Tuberculosis and infections with atypical mycobacteria. In: Wolff K, Goldsmith LA, Katz SI, Gilchrest BA, Paller AS, *et al.* Eds. Fitzpatrick's Dermatology in General Medicine, 7th ed. New York: McGraw-Hill 2008; pp. 1768-78.

[4] Ghorpade A. Tattoo inoculation lupus vulgaris in two Indian ladies. J Eur Acad Dermatol Venerol 2006; 20: 476-77. http://dx.doi.org/10.1111/j.1468-3083.2006.01471.x

[5] Heo YS, Shin WW, Kim YJ, Song HJ, Oh CH. Annular lupus vulgaris mimicking tinea cruris. Ann Dermatol 2010; 22: 226-28. http://dx.doi.org/10.5021/ad.2010.22.2.226

[6] Saritha M, Parveen BA, Anandan V, Priyavathani MR, Tharini KG. Atypical forms of lupus vulgaris – A case series. Int J Dermatol 2009; 48: 150-53. http://dx.doi.org/10.1111/j.1365-4632.2009.03863.x

[7] Padmavati L, Laksmanarao L, Ethirayan M, Krishnaswamy B. Ulcerative lupus vulgaris of the face-An uncommon presentation in India. Indian J Tuberc 2007; 54: 52-54.

[8] Jain VK, Agarwal K, Jain S, Singh S. Hypertrophic lupus vulgaris: An unusual presentation. Indian J Drmatol 2009; 54: 287-89. http://dx.doi.org/10.4103/0019-5154.55644

[9] Hsiao PF, Tzen CY, Chen HC, Su HY. Polymerase chain reaction based detection of Mycobacterium tuberculosis in tissues showing granulomatous inflammation without demonstrable acid-fast bacilli. Int J Dermatol 2003; 42: 281-86. http://dx.doi.org/10.1046/j.1365-4362.2003.01461.x

[10] Sehgal NV, Sardana K, Sehgal R, Sharma S. The use of anti-tubercular therapy (ATT) as a diagnostic tool in pediatric cutaneous tuberculosis. Int J Dermatol 2005; 44: 961-3. http://dx.doi.org/10.1111/j.1365-4632.2004.02327.x

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