Giant Lupus Vulgaris Presenting as Two Ulcerative Plaques in a Filipino Female: A Case Report*

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ABSTRACT

Lupus vulgaris is a form of cutaneous tuberculosis (CTB) caused by the bacterium *Mycobacterium tuberculosis* (MTB). It is characterized by a usually solitary, long-lasting skin lesion that most commonly develops on the head or neck, especially the nose, cheek, earlobe, or scalp. A 69-year-old Filipino female presented with a 20-year history of progressively growing erythematous ulcerative plaques on the right arm and ear, with associated mild pruritus and pain. She appeared to be immunocompetent with no clinically apparent underlying focus of TB infection. Tuberculin skin test (TST) was done, which showed a positive reaction (an induration of 11mm), and histopathologic examination revealed a chronic granulomatous dermatitis that is focally positive for acid-fast bacilli. She was given anti-tuberculosis therapy (ATT) with subsequent resolution of the lesions.

Keywords: Lupus vulgaris, Cutaneous tuberculosis, Giant lupus vulgaris

INTRODUCTION

Lupus vulgaris (LV) is a chronic and progressive form of cutaneous tuberculosis (CTB) caused by *Mycobacterium tuberculosis* (MTB). It is usually characterized by the development of skin lesions on the head and neck region and typically begins on the nose, cheek, earlobe, or scalp, with gradual spread to the adjacent areas.[1] In most cases, lesions are solitary, but multiple areas can be affected. Although this is considered a rare form of tuberculosis, its prevalence has been increasing in recent years, particularly in immunocompromised individuals.[2]

Typically, the characteristic lesions manifest as a discrete red-brown, friable macule or papule with a smooth or hyperkeratotic surface. As the condition advances, these papules may transform into atrophic, ulcerated plaques and develop crusted nodules that are usually less than 10cm in size.[3] If left untreated, LV progresses over several years and can lead to noticeable disfigurement.[4] Diagnosis requires a high index of suspicion, as it can mimic various dermatological conditions such as Spitz nevus and lupus erythematosus in early stages, rosacea, port-wine stains, sarcoidosis, deep fungal infections, and leprosy.[5] Thorough clinical examination, histopathological analysis, and microbiological investigations are essential for accurate diagnosis. Skin biopsy specimens typically demonstrate granulomatous inflammation with caseating necrosis, which is characteristic of tuberculosis (TB) infection.[6] The management includes a standard multidrug anti-tuberculosis therapy (ATT). The duration of treatment varies based on the severity of the disease and the individual's response to therapy. Surgical measures such as excision of affected tissue or laser therapy may be necessary for extensive or aesthetically concerning lesions.[5,7,8]

In this case report, we present a detailed clinical and histopathological description of a patient with giant LV, highlighting the challenges encountered in diagnosis. Through this report, we also aim to contribute to the limited pool of existing literature on multiple LV and emphasize the importance of early recognition and appropriate treatment to prevent complications and improve patient outcomes.

CASE REPORT

A 69-year-old Filipino female presented with a few erythematous plaques with well-defined borders on her right proximal arm and right ear. Twenty years prior to consult, she noted two discrete asymptomatic erythematous papules on her right arm with no recalled

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history of trauma. No fever, cough, nor joint pains were noted at that time. No consults and no interventions were done. There was no known history of contact with an active case of TB. General and systemic examinations were normal. Interval history showed a gradual increase in the size of the lesions with peripheral expansion, which coalesced to form an erythematous plaque.

One year prior to consult, the plaque progressed and developed white to greenish foul-smelling discharge and was associated with mild pruritus graded 3/10. She sought consult with multiple private physicians and was given unrecalled topical and oral antibiotics with no subsequent improvement.

One week prior to consult, the patient noted a new onset solitary erythematous patch on the right ear that progressed to a moist plaque now also associated with purulent foul-smelling discharge with pruritus (3/10) and persistent dull pain (3/10). Persistence of symptoms prompted consult at our institution.

On physical examination, there were two well-defined, irregularly-shaped erythematous, moist, ulcerative plaques with hemorrhagic crusts on the right ear and infra-auricular area measuring 11 x 7 cm, and right deltoid measuring 25 x 17 cm (FIGURE 1). Diascopy was positive for the 'apple-jelly' sign. No mucosal lesions or regional lymphadenopathies were noted. Dermoscopic evaluation of the lesions revealed yellow-orange clods and fine telangiectasias with pinkish red background and yellow-white scales (FIGURE 2).

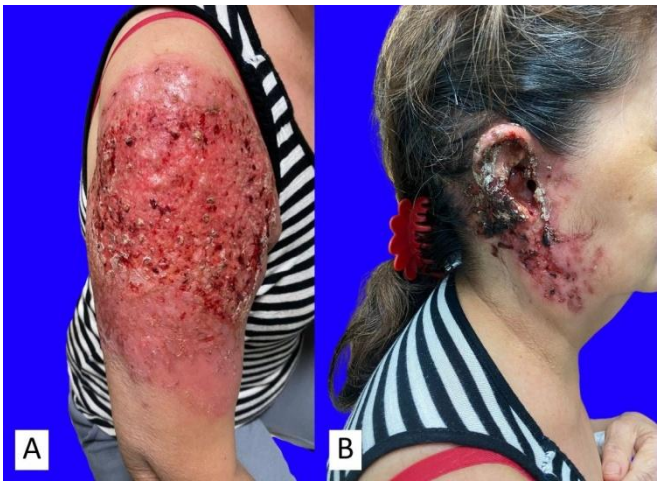

FIGURE 1. Few well-defined irregularly-shaped erythematous, moist ulcerative plaques with hemorrhagic crusting on the A) right proximal arm measuring 25 x 17 cm, and B) on the right ear measuring 11 x 7 cm.

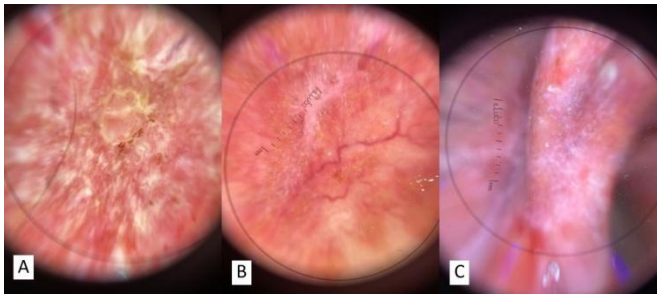

FIGURE 2. Dermoscopy of: A) and B) the right proximal arm with yellow-orange clods and fine telangiectasias with pinkish red background and yellow-white scales, C) the right ear with yellow-orange clods.

A 4mm skin punch biopsy performed on her ear and her right arm revealed orthokeratosis, pseudoepitheliomatous hyperplasia, and nodular granulomatous infiltrates consisting of lymphocytes, histiocytes, plasma cells and Langhans giant cells in the superficial and mid-dermis (FIGURE 3A). Fite Faraco staining of the right arm showed acid-fast bacilli (FIGURE 3B). Periodic Acid Schiff stain was negative for fungal spores and hyphae. The tuberculin skin test (TST) was positive with an induration of 11mm. Wound acid-fast bacilli smear and sputum Gene Xpert were both negative. QuantiFERON-TB Gold (QFT-G) was positive. Polymerase Chain Reaction (PCR) for MTB DNA and tuberculosis culture were not done.

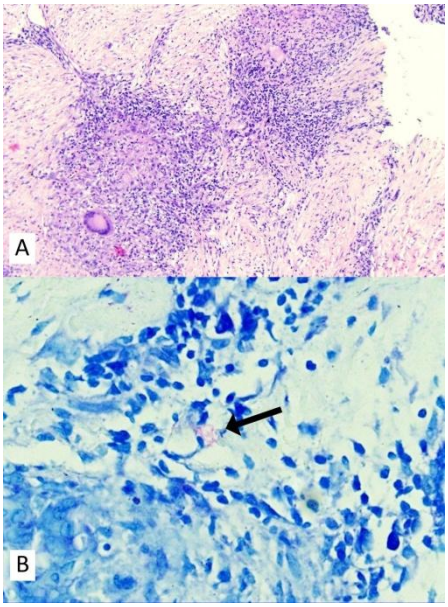

FIGURE 3. Hematoxylin and eosin stain from the right arm showing A) nodular granulomatous infiltrates with Langhans giant cells in the superficial and mid-dermis, B) Positive for acid-fast bacilli (Fite Faraco stain).

Screening for extracutaneous focus of TB infection was negative. Clinical, dermoscopic, and microscopic features were consistent with the diagnosis of LV. She was started on ATT to complete six months of treatment. She was also instructed on daily wound care and to apply mupirocin 2% ointment using sandwich dressing twice daily for one month. On one month of follow-up, there was a noted decrease in size and discharge of the lesions, with resolution of pruritus and pain (FIGURE 4A, 4B, & 4C). Her response to treatment further strengthened the diagnosis. After completion of treatment, there was resolution of lesions with only hyperpigmentation and atrophic scarring present on both her right arm and ear (FIGURE 4D, 4E, & 4F).

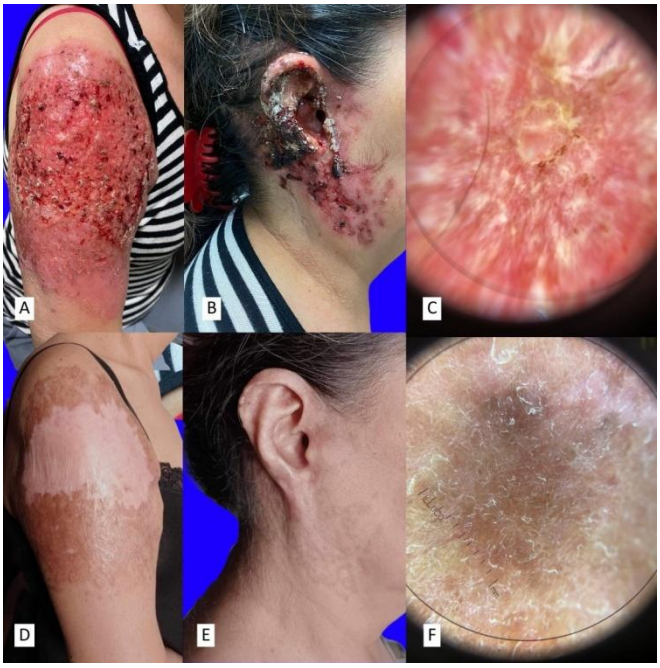

FIGURE 4. Comparative photos of the patient on A), B), and C) one month after initiation of therapy, and D), E), and F) after completion of therapy.

DISCUSSION

LV is a form of CTB that has a chronic and progressive course. It usually occurs in individuals with moderate to high immunity against the bacillus. A prior study reported an incidence of 8.6 cases per 100,000 in a Philippine tertiary care center, with the highest rates in men aged 11-20.[9] LV has been observed to occur in individuals who were previously diagnosed with TB verrucosa cutis, scrofuloderma, and, less commonly, following BCG vaccination or primary inoculation.[8,10] This patient was a 69 year-old female with an unrecalled history of BCG vaccination.

The usual belief is that LV emerges from a primary focus in either lymph nodes, bones, or joints, commonly associated with lymph node, joint, or bone TB.[5,11] Alternatively, it can result from the reactivation of a latent cutaneous focus due to previous silent bacteremia, direct inoculation, or from lympho-hematogenous extension.[11,12] However, in this case, no focus of underlying TB infection was detected since the patient denied joint pains, cough, dyspnea, or mucosal lesions, and had negative screening tests for extracutaneous TB infection. Similar to this patient, most case reports found no internal focus of infection upon screening.

In the early stages, the skin lesions may be seen as discrete, reddish-brown papules that coalesce to form an asymptomatic plaque. As the disease progresses, the lesions may become larger with a slightly raised border, central atrophy, and a soft, gelatinous surface. Dermoscopy can be used to identify well-focused, linear-branching telangiectasias on a diffuse or localized, structureless, yellow to orange background, and whitish, reticular streaks. However, these features are not specific to LV and can also be seen in cutaneous sarcoidosis.[14] Upon blanching using diascopic pressure, the lesions usually reveal a characteristic brownish-yellow color or "apple-jelly" color.[14] Similar to this, our patient initially presented with an erythematous papule on the right deltoid and eventually the right ear that gradually progressed to an ulcerative plaque with dermoscopic features of yellow-orange clods on an erythematous background with apple-jelly color upon application of diascopic pressure.

LV lesions are most commonly found on the head and neck, particularly on the nose and cheek. However, recent studies have shown that the predilection sites can vary by locality. In Europe, over 80% of the lesions are found in the head and the neck.[15] They are rarely found on the extremities, buttocks, or trunk.[16,17] However, according to another study done by Bilen et al (2000), lesions on the legs and buttocks were found to be a common occurrence in Asia.[17] More specifically, lesions are more often seen on the trunk, elbows, and knees in India and the Philippines.[9,18] LV commonly presents as a single lesion, with few case reports with findings of multiple lesions.[19,20,21,22,23,24] To the best of our knowledge, there have been limited case reports of solitary plaques affecting only the ear,[27] and no case reports have been identified that involve both the ear and the unilateral proximal arm.

Regarding the size of the lesion, it is usually small in size, and gradually grows peripherally to reach 0.5cm–10cm.[3,19] The classification of giant LV based on size lacks consensus or standardization but is usually greater than 10cm.[8,20,27] Considering the measurements reported in previous studies for giant LV lesions, and with this patient's lesion measuring 25 x 17cm, we believe that this case qualifies as giant LV as well.

Diagnosis usually relies on clinical suspicion and histopathological features.[8,28,29] The definitive diagnosis of LV is the isolation of MTB from a culture of the skin lesion. However, this is often difficult to achieve, as the lesions are usually paucibacillary.[18] Positive PCR results for M. tuberculosis can strengthen the diagnosis of tuberculosis even in paucibacillary forms.[8,30] Interferon-gamma release assays (IGRAs) measure the immune response to MTB antigens and are more accurate than other tests in areas where TB is common and in people who have been vaccinated with BCG.[31] QFT-G is increasingly being used in the Philippines to diagnose LV because it is more sensitive than the PPD test, and it can also detect tuberculosis in patients who have been vaccinated against BCG.[32] Previous studies found that QFT-G had a sensitivity of 89% and a specificity of 99% in the diagnosis of CTB.[33] These findings suggest that QFT-G is a valuable tool for the diagnosis of CTB. In this case, the patient was positive for both the PPD and QFT-G tests.

Histopathologically, the epidermis may be atrophic or hypertrophic, with acanthosis, papillomatosis, and even pseudoepitheliomatous hyperplasia. The reticular dermis may contain well-formed tuberculous granulomas, often with Langhans giant cells, or foreign body-like granulomas. There may also be sarcoidosis-like granulomas. The lymphocytic infiltrate is dense, and caseous necrosis is rare, but it may be present in small foci in the center of the granuloma. Acid-fast bacilli are not readily found even with culture due to its paucibacillary nature.[12,19,20] In line with this, our patient presented with nodular granulomatous infiltrates with Langhans giant cells in the superficial and mid-dermis. Interestingly, she was focally positive for acid-fast bacilli with Fite-Faraco stain which is rarely seen in this condition.

Current literature found that CTB typically responds well to standard ATT, with clinical improvement expected after 4 to 6 weeks.[8,34] However, it is essential to tailor the treatment to the patient's individual requirements, taking into account their overall health, immune status, type of CTB, severity of cutaneous involvement, disease stage, medication compliance, and side effects. If left untreated, LV lesions can remain for years, gradually increasing in size. This can lead to scarring, deformation, and disfigurement that can be severe.[5,26] The risk of malignant transformation of lesions into squamous cell carcinoma (SCC) is estimated to be between 0.5% and 10.5%, typically occurring after 25-30 years of untreated LV.[22,35] In line with this, our patient had a 20-year history of two gradually enlarging and ulcerating plaques that did not show signs of malignant transformation to SCC.

DATA PLAN

All data obtained from this study will be stored for 10 years in the private clinic of the patients' dermatologists in case the investigators need to review the source data should there be questions about the results. These data will be used solely for the study and the principal investigator and co-authors will have access to this data. We will destroy the data after the said duration. Data collection will commence after approval of the IRB and will last for the 4 month duration of the study.

ETHICAL CONSIDERATION

This case report involves no more than minimal risk and will use only the medical data of patients. Secured data acquisition will be implemented involving only the authors and the private dermatologists in charge of the patient's records. Only necessary information such as diagnosis, ICD-10 codes used, family history and treatment outcome will be collected and no other revealing personal information will be recorded throughout the study. Only redacted data were recorded under a non-disclosure agreement. The protocol was submitted to and approved by the Institutional Review Board. No code names were used for the patient as the authors only included the patient's race, age and sex.

CONCLUSION

We report a case of LV in a 69-year-old immunocompetent Filipino female, which presented with an uncommon size and an unusually large size. The patient had two well-defined, irregularly-shaped ulcerative plaques located on the right deltoid and right ear, extending to the right infra-auricular area, measuring 25 x 17 cm and 11 x 7 cm respectively. Interestingly, these lesions had progressed over a period of twenty years without a clinically apparent underlying focus of infection. By contributing our case to the limited pool of information on giant LV, we can potentially enhance the understanding of this disease and aid in establishing more standardized criteria for its classification, especially in terms of lesion size. Additionally, this case underscores the importance of maintaining a high index of clinical suspicion, particularly when lesions occur in atypical

locations. Clinical and histopathological correlation remains essential for early diagnosis of this disease. In this regard, the QFT-G can serve as a helpful adjunctive test to strengthen the diagnosis in challenging cases. Early detection and treatment are crucial to prevent complications such as permanent disfigurement and the risk of malignant transformation.

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