CASE REPORT

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38 Years of Undiagnosed Lupus Vulgarius: A Case Report

38 Yıldır Tanı Alamayan Lupus Vulgaris: Olgu Sunumu

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ABSTRACT

Tuberculosis (TB) is an infectious disease caused by the bacterium *Mycobacterium tuberculosis* that usually affects the lungs but can also affect other organs and systems in the body. One of the forms of TB with skin involvement is known as lupus vulgaris (LV) and is a rare form of skin TB. In this case report, we present a male patient with a progressive lesion with delayed diagnosis for 38 years who was diagnosed as LV by us. Dermatologic examination of the patient revealed irregular, erythematous, purple, hyperkeratotic plaques with normal skin areas and atrophic scatricial areas extending from the extensor aspect of the left arm to the elbow and forearm extensor areas. Punch biopsy of the lesions revealed caseification necrosis in the dermis and granuloma structures containing langhans type giant cells. Clinical and histopathologic findings were compatible with LV and quadruple anti-TB treatment regimen was initiated. With the treatment regimen initiated in the patient follow-up, the patient's skin lesions showed signs of improvement. The patient is still being followed up by us.

Keywords: Cutaneous tuberculosis, late diagnosis, lupus vulgaris

ÖZ

Tüberküloz (TB), *Mycobacterium tuberculosis* bakterisinin neden olduğu, genellikle akciğerleri etkileyen ancak vücuttaki diğer organları ve sistemleri de etkileyebilen bir enfeksiyon hastalığıdır. Deri tutulumu olan TB formlarından bir tanesi de LV olarak bilinir ve nadir karşılaşılan bir deri TB türüdür. Bu vakada 38 yıldır tanısı gecikmiş ilerleyici lezyonu olan tarafımızca LV tanısı konulan bir erkek hasta sunulmuştur. Hastanın dermatolojik muayanesinde sol kol ekstansör yüz dirsek ve ön kol ekstansör alana uzanım gösteren normal deri alanları ve atrofik skatrisyel alanlar içeren düzensiz yapılı, eritemli, mor renkli, üzeri hiperkeratotik görünümde olan plaklar izlendi. Lezyonlardan yapılan punch biyopsisi sonucunda histopatolojik olarak dermiste kazeifikasyon nekrozu ve langhans tipi dev hücreler içeren granülom yapıları izlendi. Klinik ve histopatolojik bulguları LV ile uyumlu olan hastaya dörtlü anti-TB tedavi rejimi başlandı. Hasta takibinde başlatılan tedavi rejimi ile birlikte hastanın cilt lezyonlarında iyileşme gösterdi. Hastanın takibi tarafımızca devam etmektedir.

Anahtar Kelimeler: Kutanöz tüberküloz, geç tanı, lupus vulgaris

Introduction

Tuberculosis (TB) is a chronic granulomatous infectious disease caused by *Mycobacterium tuberculosis* (MT), a slow-growing, rod-shaped, non-spore-forming, facultatively anaerobic bacterium (1). TB can affect the lungs but can also involve many other organs and systems in the body. One of these organs is the skin, and its classification is usually based on the route of entry of the bacillus. The most common type of cutaneous TB (CTB) is lupus vulgaris (LV) (2). The clinical presentation of LV maybe different and this may make it difficult to make the correct diagnosis. Due to the high prevalence of TB in Türkiye, the suspicion of LV should be considered in patients with chronic skin lesions. Typical lesions appear as plaques consisting of red-brown papulonodules. As the plaque enlarges, scarring often develops in the center. The head and neck region, especially the nose, cheeks, and earlobes, is the most commonly affected area. Histopathology shows tuberculoid granulomas

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consisting of epithelioid histiocytes, Langhans-type giant cells and lymphocytes in the dermis with caseating necrosis in the center (3). In this report, we present a male patient with LV on his arm for 38 years.

Case Report

A 47-year-old male patient presented with wounds on his arms that had been present for approximately 38 years. Dermatologic examination revealed irregular plaques measuring 20 × 12 cm, extending from the extensor aspect of the left arm to the extensor aspects of the elbow and forearm, including normal skin areas and atrophic cicatricial areas, with erythematous purple plaques of hyperkeratotic appearance (Figure 1). The patient's medical history indicated a diagnosis of pulmonary TB prior to the development of skin lesions; isoniazid treatment was initiated after diagnosis but was discontinued before completion. He stated that he did not receive any other treatment after this process. In addition, the patient's complaints included long-standing shortness of breath and a productive cough. No other diseases, drug or substance use, or history of TB were reported in his family or among close relatives. Systemic examination was normal, and no lymphadenopathy was detected. It was also noted that the patient had a Bacillus Calmette-Guérin vaccination scar.

Skin biopsy was performed with preliminary diagnoses of LV, deep fungal infections, atypical mycobacterial infections, and sarcoidosis based on the clinical features of the lesion and the patient's history.

Histopathologic examination of the skin punch biopsy of the lesions revealed granuloma structures consisting of epithelioid histiocytes and langhans type giant cells in the dermis (Figure 2A, B). Intense lymphocytic infiltration was observed around the granulomas. There is an amorphous, eosinophilic, caseous necrotic area in a focal region at the center of the granuloma (Figure 2C). Immunohistochemically, the granuloma structures formed by epithelioid histiocytes were visualized with CD68 stain (Figure 2D).

Purified Protein Derivative (PPD) test was performed to confirm TB infection in a patient with a prediagnosis of LV. Apositive response of 21 mm was considered as an important indicator that TB in fection might be present. Following this finding, the pulmonology department was consulted to evaluate for pulmonary TB. No findings consistent with TB were detected on chest X-ray performed in the pulmonology department (Figure 3).

Additional serologic tests ruled out other infectious diseases such as bartonellosis, syphilis, and human immunodeficiency virus infection. Tissue cultures were obtained from the lesions on the patient's left arm. No growth was observed in bacterial and fungal cultures. Ehrlich–Ziehl–Neelsen staining could not be performed because the histochemical stain was unavailable in our pathology department. TB-polymerase chain reaction testing could not be performed due to limited technical resources and the unavailability of the required equipment. Histopathological features, PPD positivity, and clinical improvement under anti-tuberculous therapy strongly support the diagnosis of CTB.

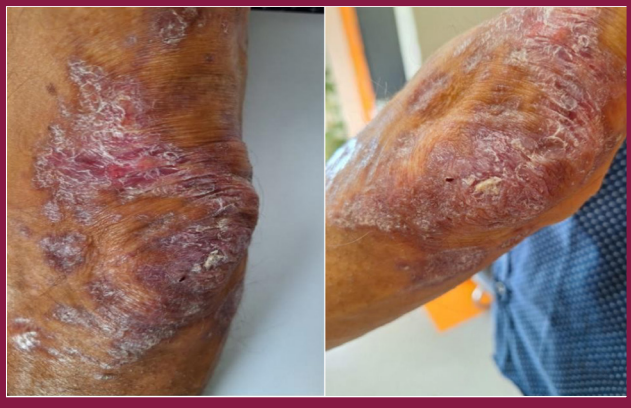

Figure 1. 20*12cm plaque-like lesions on the extensor side of the left arm, extending to the elbow and forearm extensor area, with irregularly structured erythematous purple-colored plaque-like lesions with hyperkeratotic appearance, including some normal skin areas.

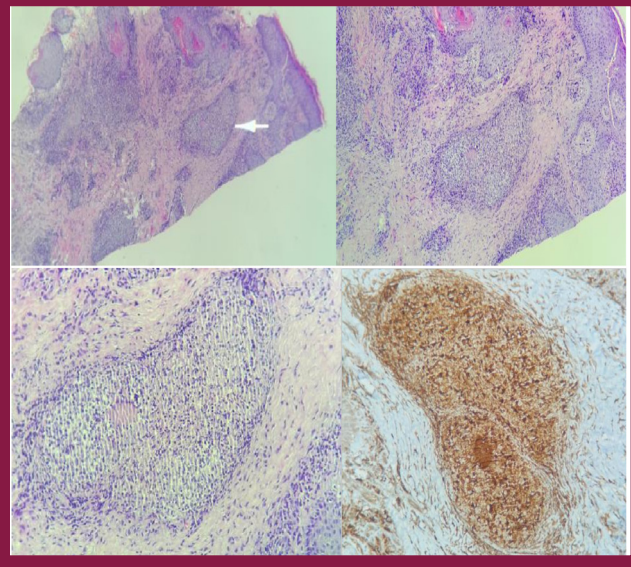

Figure 2. (a) H&E section x40; (b) H&E section x400 langhans type giant cell; (c) H&E section x200 necrotizing gralunoma structure; (d) CD68 immunohistochemical stain.
H&E, hematoxylin and eosin.

The patient's condition was evaluated in consultation with infectious diseases specialists. A quadruple anti-TB treatment regimen was initiated. This treatment regimen included isoniazid (300 mg/day), rifampicin (600 mg/day), ethambutol (1500 mg/day), and pyrazinamide (2 g/day). The patient's skin lesions started to improve with the Treatment regimen initiated during follow-up (Figure 4). The patient continues to be followed up at our clinic.

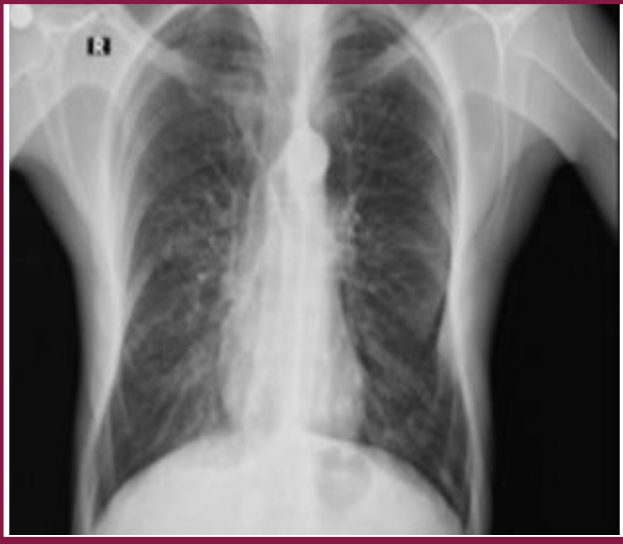

Figure 3. Posteroanterior chest radiograph demonstrating increased aeration of the upper lung zones, bilateral flattening of the hemidiaphragms, widening of the intercostal spaces, and a vertically oriented narrow cardiac silhouette ("droplet heart" appearance).

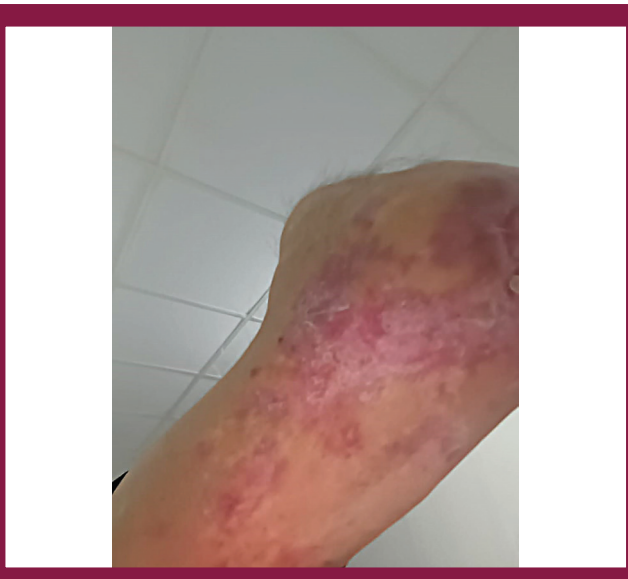

Figure 4. During treatment, the patient exhibited erythematous macules with patchy post-inflammatory hyperpigmentation.

Discussion

TB, caused by MT, is a lifelong infectious disease that can affect various organs and systems in the body. Extrapulmonary involvement accounts for approximately 20% of TB cases. CTB is a rare form of mycobacteriosis that accounts for 1% to 2% of all forms of extrapulmonary TB (4).

LV is the most common type of CTB (2). LV is more common in women than in men and affects all age groupse qually (5). LV is a type of CTB that occurs as a result of hematogenous spread, lymphatic route or direct inoculation in an individual with high immunity to the tubercle bacillus. Clinical types of LV include plaque type, ulcerative or mutilative type, vegetative type, papular or nodular type, and tumor-like type (6). Most patients with LV who have high immunity to tubercle bacilli have positive tuberculin test results. Typical lesions appear as plaques composed of red-brown papulonodules with an "applejelly" color on diascopy. As the plaque enlarges, scarring often develops in the center, which may lead to severe tissue loss over time (7). Histopathology is pathognomonic for tuberculoid granulomas consisting of epithelioid histiocytes, langhans-type giant cells and lymphocytes in the dermis with or without caseation necrosis (3). In our patient, a 21-mm positive PPD test, the characteristic apple-jelly appearance on diascopy, and compatible histopathologic findings support our diagnosis of LV. At the same time, the fact that the patient had skin lesions after being diagnosed with pulmonary TB in the past suggests that the agent probably spread from the pulmonary focito the skin by hematogenous route. The lesions in LV may not always be in typical form and may be confused with diseases such as discoid lupus erythematosus, psoriasis, sporotrichosis, actinomycosis, mycetoma, which may make the diagnosis difficult (8).

Definitive diagnosis of TB can only be made by demonstrating the presence of MT in patient samples taken from suspected sites of involvement. Culture is the gold standard. In standard settings, it can take up to 6 weeks for growth to appear in culture, which is a long time to start treatment, as in our case. Molecular and novel culture techniques accelerate diagnosis. PPD and interferon-γ release tests are commonly used. In most patients, a PPD or an interferon-γ release test is performed as part of the initial evaluation, unless there is a documented history of TB (9). It is particularly useful before cultures are available and for providing supporting evidence in culture-negative cases.

First-line treatment for drug-susceptible TB outlined by the World Health Organization includes a combination of isoniazid, rifampicin, pyrazinamide and ethambutol for at least 6 months (10). CTB is usually treated medically in

the same way as pulmonary TB. Recognition and treatment of LV are important. If left untreated, LV may lead to scars, contractures, deformities, and secondary malignancies such as squamous cell carcinoma and basal cell carcinoma.

CTB should always be considered in chronic non-healing wounds. In this report, we present a patient whose diagnosis of LV was delayed and remained untreated because it was not considered.

The patient's non-compliance with previous pulmonary TB treatment led to disease progression and complications. Awareness of the disease and accurate clinical and laboratory investigations play a critical role in the diagnosis of such cases. A multidisciplinary approach and a meticulous treatment process will increase the success of disease management. Early diagnosis, treatment, and patient adherence are of great importance in the management of rare but serious diseases such as LV, which is the cutaneous manifestation of TB.

Ethics

Informed Consent: Written informed consent was obtained.

Footnotes

Authorship Contributions

Surgical and Medical Practices: S.D., Z.M., N.D., Concept: S.D., Z.M., N.D., Design: S.D., Z.M., N.D., Data Collection or Processing: S.D., Z.M., N.D., Literature Search: S.D., Z.M., N.D., Writing: S.D., Z.M., N.D.

Conflict of Interest: No conflict of interest was declared by the author(s).

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