Lupoid cutaneous leishmaniasis due to Leishmania major, a great mimicker of Lupus Vulgaris in Sanliurfa, Turkey: a case report

Nebiye Yentur Doni[1], Gulcan Gurses[2], and Isa An[3]

[1]Harran University Faculty of Medicine
[2]Harran University Vocational School of Health Services, Department of Microbiology and Parasitology
[3]Sanlıurfa Training and Research Hospital

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Abstract

Lupoid cutaneous leishmaniasis is identified by a broad range of clinical presentation with brown to red, yellowish-brown, apple-jelly papules. These papules enlarge to plaques, develop into granulomatous, crusted ulcers, which are as similar as cutaneous lesions of lupus vulgaris. Herein, we report a patient with lupoid CL mimicking lupus vulgaris

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INTRODUCTION

Cutaneous leishmaniasis (CL) is a dermal infection caused mostly by*Leishmania major* , *Leishmania tropica* , and *Leishmania infantum* , is transmitted to humans by the bite of sandflies. Cutaneous leishmaniasis is polymorphic and may result in lesions characterized by different clinical features. Cutaneous leishmaniasis is also characterized by acute or chronic course. Lupoid cutaneous leishmaniasis (LCL) is a clinical form of CL characterized by a chronic relapsing course, unusual clinical features, an unique granulomatous lesion[1]. Lupoid cutaneous leishmaniasis is identified by a broad range of clinical presentation with brown to red papules, yellowish-brown papules and, apple-jelly papules. These papules enlarge to plaques, and develop into granulomatous and crusted ulcers, which are as similar as cutaneous lesions of lupus vulgaris. Herein, we report a patient with lupoid CL mimicking lupus vulgaris to highlight the unusual clinical spectrum of CL.

Key words: Lupoid cutaneous leishmaniasis, lupus vulgaris,*Leishmania major* , Turkey

Key Clinical Message

Many unusual lesions might be caused by *Leishmania.*

Case Report

Clinical Findings

A 24 year old man presented presented with a 3-5 months history of a well-defined erythematous plaque extended to involve the whole left hand middle finger, resembling lupus vulgaris. He also presented 1 ulcerated lesion on his left shoulder, and 1 papulonodular lesion on his left arm. He was a textile worker.

He could not use his left hand due to the huge lesion on his middle finger. While, his general physical examination and systemic examination were normal.

The patient had no familial history for the development of CL. He had no travel to other CL endemic provinces. The patient had applied to different medical clinics in non-endemic provinces for CL for the diagnosis of the lesions he had. He was diagnosed as "lupus vulgaris" but he had not been treated for lupus vulgaris or CL. Then the patient applied to our dermatological clinic in Sanliurfa, a hyper-endemic province for CL.

Histopathological Findings

Direct smears from the exudate of fluid of the lupoid lesion were stained with special stains Gram, Erlich-Ziehl-Nielsen (EZN), Periodic Acid-Schiff (PAS), Giemsa for detection of bacteria, mycobacteria, fungi and *leishmania* amastigotes, respectively. A punch biopsy aspiration fluid from lesions was used, and a part of the exudate of fluid of the lupoid lesion was used for the identification of*Leishmania* species by Real Time PCR. Application of real time PCR that targeted the ITS1 region of *Leishmania* species using the DNAs obtained from the lesion of the patient. In this real time PCR, melting temperature of *L. major* reference control and sample is 85°C as described previously[2].

Results

Written informed consent to enroll the study and use clinical images in case report was obtained from the patient.

Granuloma structures characterized by epithelioid histiocytes in the dermis were observed in the prepared sections. Langerhans-type multinucleate giant cells were noteworthy which were found in granulomas suggestive of leishmaniasis, tuberculosis with Hematoxylin-eosin staining (200X) (Figure 1). Intense mononuclear inflammatory cells surrounding the granuloma infiltrate the granuloma were also noteworthy. Granulomas were mostly observed in the dermis. The histomorphological findings seemed to be compatible with lupus vulgaris. However, a definitive differential diagnosis could not be performed.

No bacteria was observed with Gram staining, no Mycobacteria was observed with EZN staining and no fungal organism was observed with PAS staining.

A few amastigotes of *Leishmania* were observed with Giemsa staining. Detection of *Leishmania* amastigotes by the microscopic examination of the direct smears exudative fluid of the lesions strictly supported the definitive differrantial diagnosis of lupoid CL. It has been reported that *Leishmania* amastigotes are usually absent on microscopy, thus, PCR is especially recommended for diagnosis of CL to prevent the delayed diagnosis or clinical and histological misdiagnosis often with Lupus vulgaris[3]. In light of this data**,** Real Time PCR was used to identify the *Leishmania*species, which was found to be *L. major* (Figure 2).

Figure 1. Non-caseified granulomas including giant cells (green arrow) and *Leishmania*amastigotes (red arrow) were seen in dermis (Hematoxylin-eosin, X200)

Figure 2. Real-time PCR analysis of *Leishmania major* infection in lesion of the patient

Identification of polymerase chain reaction products with melting temperature analysis of two *L. major* samples and two samples of*Leishmania major* belonged to the patient. The melting temperature is 85^0 C for *L. major* MHOM/SR/2015/HRURFA012.

The patient with lupoid CL (Figure 3) was treated with intralesional meglumine antimoniate (Glucantime®) injections twice a week (Monday-Thursday) for four weeks. After four weeks, the patient was called monthly for control the healing of the lesion for one year, it was seen that the lesion was regressed (Figure 4).

The plaque type occurred over the finger and presented clinically as well-defined erythematous plaques that sometimes extended to involve the whole finger resembling lupus vulgaris.

Figure 3. Lesion of the lupoid CL before treatment

Figure 4. Lupoid CL, after treatment of the lesion with intralesional meglumine antimoniate (Glucantime®)

Discussion

Leishmaniasis is a neglected vector-borne disease caused by protozoan parasites belonging

to *Leishmania* genus, and endemic in all WHO regions, including Turkey. There are 4 main clinical classifications of the leishmaniasis, visceral, post-kala-azar dermal, cutaneous (CL) and mucocutaneous. Turkey is considered to have a high burden country for CL[4]. Although a National control programme has been established, CL is still a serious public health problem with increasing the number of CL cases due to the Syrian conflict in Turkey, particularly in Southeastern and Southern regions of the country. Sanliurfa is a hyper-endemic province of CL in Southeastern region of Turkey. Various clinical presentations of CL including papule, nodule, ulcerative, noduloulcerative, and recidivans forms have been reported in Sanliurfa[5-8]. Noduloulcerative form, classically known as oriental sore, is the most common form in study area.

To the best of our knowledge, lupoid CL mimicking lupus vulgaris has not been reported from Sanliurfa, Southeastern Region of Turkey. However, a few lupoid CL cases have been reported in different provinces of Turkey[9; 10]. Of all CL cases the incidence of Leishmaniasis recidivans has been reported with 0,5%-6,2% in the Middle East and Afghanistan[11]. In a recent study carried out in Tunisia, lupoid CL represented 9% of cases in adults[12]. It has been reported that in lupoid CL, the*Leishmania* amastigotes are usually absent or rare on a microbiological smear [13-15]. The paucity of*Leishmania* amastigotes in the lesion samples and in direct smears might lead to misdiagnosis with lupus vulgaris[11; 13-15]. Since, it has been reported that clinically and histologically, lupoid leishmaniasis is similar to lupus vulgaris, which is considered as the most important differential diagnosis[16; 17]. Contrary to other studies[13-15], we detected fortunately *Leishmania* amastigotes in direct smears of the lesion of the patient. This can be explained by the short duration of the patient's lesions (approximately 3-5 months) at the moment of diagnosis. We detected also *Leishmania* DNA by ITS 1 PCR and identified by Real time PCR. We found that L. *major* is responsible agent for this LCL case, which is compatible with the other studies[14; 18; 19]. *Leishmania tropica* is considered to be the most common, and *L. major* is the rare causative agent of lupoid CL[11; 14; 15; 20]. In a study it has been reported species-specific PCR analysis is sensitive in cases of acute cutaneous leishmaniasis, but in lupoid leishmaniasis it is less sensitive[14].

There is no standardized treatment for this condition and thus multiple treatments have been reported with varying degrees of success. Treatment options include cryotherapy, topical antimonial compounds and intralesional pentavalent antimony. This patient was treated with intralesional meglumine antimoniate (Glucantime®) injections twice a week for four weeks with marked improvement of clinical features.

Conclusion

Lupoid CL can be manifested as an uncommon form that can mimic many other skin disorders as lupus vulgaris. Hence, the physicians living in nonendemic areas should never overlook that many unusual lesions might be caused by *Leishmania.* Any ignoring may be resulted in missed and delay the precise diagnosis, submitting the patients to inappropriate prescribing of drugs, increased resistance to treatment, worsening the clinical presentation, and contributing to the transmission chain of the parasite. Correct and reliable identification of parasite might contribute for breaking the chain of transmission of the parasite and decrease the prevalence. Diagnosis of CL should be performed in all patients from endemic areas of Leishmaniasis by both microscopic examination of Giemsa stained smears and PCR and identified of *Leishmania* species by using a PCR-RFLP technique.

Author Contribution

Conceptualization: Nebiye Yentur Doni, Gulcan Gurses, İsa An.

Data curation: Nebiye Yentur Doni, Gulcan Gurses.

Formal analysis: Nebiye Yentur Doni.

Funding acquisition: Nebiye Yentur Doni.

Investigation: Nebiye Yentur Doni, Gulcan Gurses.

Methodology: Nebiye Yentur Doni, Gulcan Gurses, İsa An.

Project administration: Nebiye Yentur Doni

Resources: Nebiye Yentur Doni.

Software: Nebiye Yentur Doni, Gulcan Gurses, İsa An.

Supervision: Nebiye Yentur Doni, Gulcan Gurses

Validation: Nebiye Yentur Doni, Gulcan Gurses, İsa An.

Visualization: Nebiye Yentur Doni, Gulcan Gurses.

Writing ± original draft: Nebiye Yentur Doni.

Writing ± review & editing Nebiye Yentur Doni, Gulcan Gurses, İsa An.

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