CASE REPORT

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Cutaneous Leishmaniasis with Unusual Psoriasiform Presentation

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ABSTRACT

Cutaneous leishmaniasis (CL) is still an important public health problem in many countries, especially in developing countries. The disease can manifest itself in a broad spectrum of clinical presentations. In this case report, we describe a 3-year-old patient with a 12-month history of non-healing psoriasiform lesions on her right leg. It was diagnosed as CL and the lesion almost completely resolved after 5 sessions of intralesional glucantime.

Keywords: Antimony compounds, Cutaneous leishmaniasis, Intralesional therapy, Psoriasiform

Introduction

Leishmaniasis is caused by protozoa of genus Leishmania through the bite of the female phlebotomine sand fly. Depending on the type of Leishmania species and the host's immune response, infection results in cutaneous, mucocutaneous or visceral disease. Cutaneous leishmaniasis (CL) is the most common form of leishmaniasis; affecting 600,000-1 million people each year [1]. The disease can manifest itself in a broad spectrum of clinical presentations. Classical CL lesions evolve from papules to nodules to ulcerative lesions, with a central depression and a raised, indurated border, and eventually, over months to years, to atrophic scars [2]. Most cases do not cause any diagnostic difficulties; however, some patients present with very unusual morphological forms of CL, which make the diagnosis even more challenging.

Case Report

A 3-year-old girl presented to our clinic with a growing lesion on her right thigh and leg. The patient's mother reported that the lesion started as a small papule 12 months earlier and grew without any response to topical or systemic antibiotics. She also did not complain about pain or pruritus. Her personal history of illness and family history was unremarkable. The patient's family was residing in Ankara, Turkey and had a history of travel only to Corum province 18 months ago. Dermatological examination revealed multiple slightly erythematous scaly papuloplaques arranged in a linear and annular pattern (Figure 1).

A 4-mm punch biopsy was performed considering granuloma anulare, zosteriform lichen planus and inflammatory linear verrucous epidermal nevus as the differential diagnosis. Histological examination showed orthohyperkeratosis, focal parakeratosis and focal atrophy; dense lymphoplasmacytic infiltration accompanied by giant cells and histiocytes. Infiltrates and histiocytes contained small oval parasitic organisms known as amastigotes (Figure 2). Giant cells along with lymphohistiocytic cell infiltration were observed (Figure 3).

In accordance with the findings, the patient received a diagnosis of CL and treated with intralesional meglumine antimoniate injections weekly. After 5 sessions, a noticeable improvement was noted (Figure 4). Informed consent was taken from the patient's mother for possible case report publication.

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Discussion

CL is a serious public health problem in the eastern Mediterranean region including Turkey. The disease had been widespread in our country before 1950; then it became limited to the Southeastern Anatolia region with the use of intense dichlorodiphenyl trichloroethane against malaria [3]. Unfortunately, there have been increased number of CL cases reported from both endemic and non-endemic regions, mainly due to the immigrants from Syria, recently [4]. Besides, it is noticed that there are also inhabitant CL cases reported from non-endemic regions in Turkey [5]. While travelling to endemic regions is considered to be the major reason for inhabitants, ecological studies have revealed some phlebotomine species that can be carriers for Leishmania species, living in non-endemic regions as well. For example, although Corum and Ankara province are located in Central Anatolia and regarded as non-endemic regions for CL, according to these studies, Phlebotomus transcaucasicus and Phlebotomus tobbi were found in Corum and Phlebotomus perfiliewi was reported in Ankara [6,7]. Interestingly, our patient's mother gave a history of visit to Corum province 18 months ago. The patient may have contracted this disease in Ankara or during this trip; this situation remains uncertain.

The typical presentation of CL is ulcerations on the exposed areas of the body such as face, arms and legs. However, it may present to our clinics as the 'great imitator' on occasion. The resemblance to other skin diseases, such as verruca vulgaris (warty), herpes zoster (zosteriform), psoriasis (psoriasiform), lupus (lupoid), erysipelas (erysipeloid) and sporotrichosis (sporotrichoid) may cause a challenge for an accurate clinical differentiation.

There may be multiple erythematous nodules mimicking cutaneous lymphoma or pseudolymphoma [8]. To our best knowledge, there are four cases of CL with psoriasiform presentation, reported in the literature. In 1997, a 28-year-old, human immunodeficiency virus-positive visceral leishmaniasis patient with widespread psoriasiform plaques was reported [9]. Moreover, Schepis et al. [10] reported a 68-year-old CL patient, with erythema and desquamation on

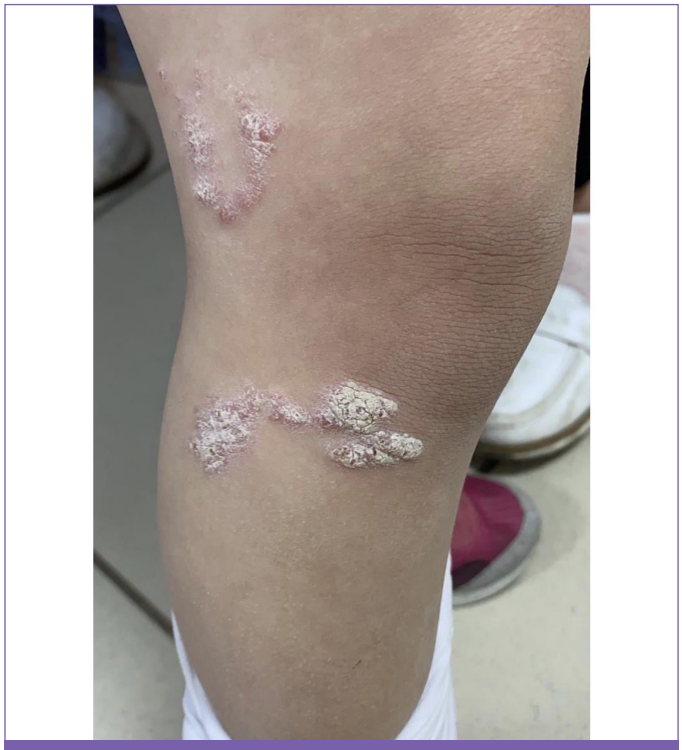

Figure 1. Linear and annular psoriasiform papules

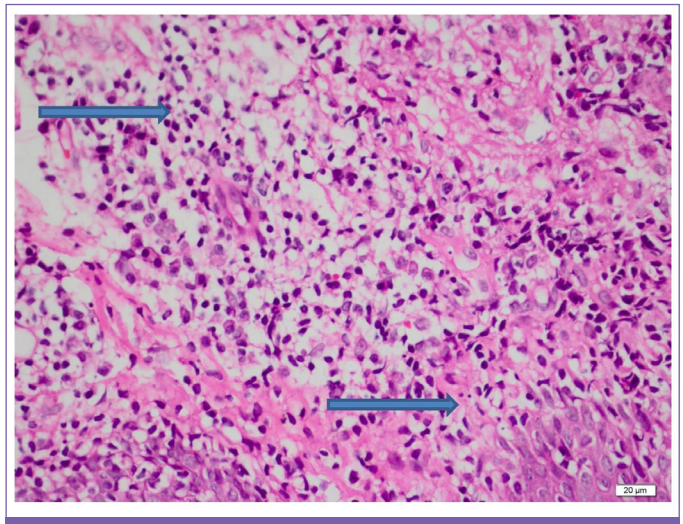

Figure 2. Circular, crumb-shaped structures compatible with leishmania are observed, in the cytoplasm of histiocytic cells (H&E x400)

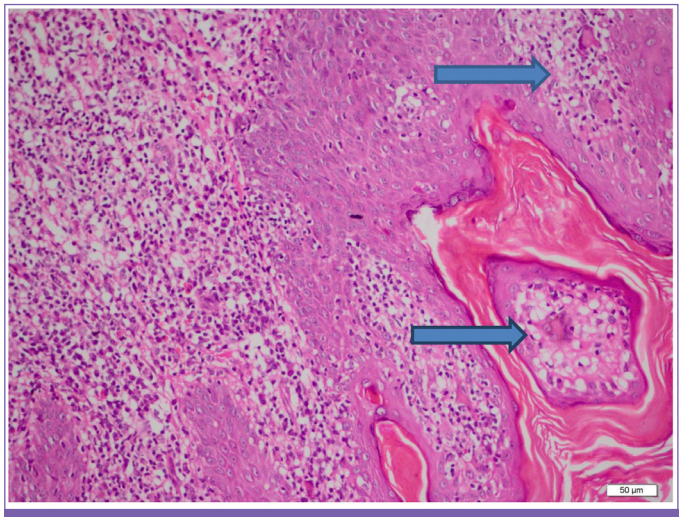

Figure 3. Giant cells indicated by blue arrows between lymphoplasmacytic cell infiltration (H&E x200)

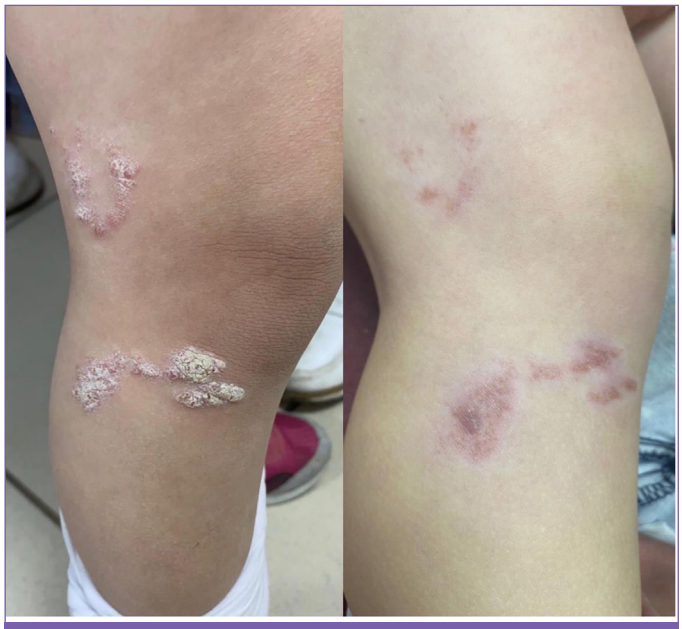

Figure 4. Before and after 5 sessions of meglumine antimoniate injections

the scalp and face resembling sebopsoriasis. Veraldi et al. [11] reported a 54-year-old CL patient who was misdiagnosed as psoriasis and was unresponsive to topical corticosteroid and vitamin D analogues [10,11]. The patient had scaly, erythematous-infiltrated lesion with well-defined borders. In addition, a study which was recently published, collected cases of CL with atypical clinical features. Out of 27 atypical CL patients, only one patient had psoriasiform lesion on the elbow [12]. Interestingly, our patient is the only pediatric case reported up to now. She was diagnosed with CL histopathologically and responded well to the intralesional glucantime treatment.

The reason for pleomorphism of CL is not fully understood; but variations in parasite virulence and host factors, abnormal host immune response, malnutrition, and immunosuppression have been suggested as the possible reasons. Although the usual clinical presentations of leishmaniasis are easily diagnosed by clinicians in endemic regions, the unusual forms may give rise to difficulties in diagnosis, delaying the diagnosis and appropriate treatment for several months. Thus, all cases of atypical CL should be confirmed by demonstration of the parasite in a Giemsa-stained smear. When the parasite cannot be demonstrated in a smear, histopathological examination should be used. If the microscopic examination result is negative, polymerase chain reaction appears to be the most sensitive diagnostic test for the identification of parasites; in the event that the microscopic examination result is negative.

These atypical forms are observed in only 2 to 5% of all affected patients and may cause a delay in the diagnosis and treatment. Although not life-threatening, it is important to diagnose and treat CL because it can be associated with permanent scarring, decreased quality of life, stigmatization and long-term psychologic consequences [1]. It also remains as an important public health problem in endemic regions. In case of an increase in cases, it is important to apply prompt therapeutic and preventive interventions. Consequently, our case is worth reporting, since it is seen in a non-endemic region, with a very rare clinical presentation.

Ethics

Informed Consent: Informed consent was taken from the patient's mother for possible case report publication.

Peer-review: Internally and externally peer-reviewed.

Authorship Contributions

Data Collection or Processing: E.P.E., Analysis or Interpretation: N.K., Literature Search: İ.K., E.K.N., Writing: İ.K.

Conflict of Interest: No conflict of interest was declared by the authors.

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