

## Case reports

# Cutaneous leishmaniasis mimicking granulomatous cheilitis and treated successfully with oral fluconazole in a boy

Gamze Serarslan, Metin Aksakal

Department of Dermatology, Faculty of Medicine, Mustafa Kemal University, İskenderun St., Hatay 31100, Turkey

Corresponding author: Gamze Serarslan; e-mail: serarslang@yahoo.com

**ABSTRACT.** Cutaneous leishmaniasis is a protozoan infectious disease and widespread in Mediterranean basin including Turkey. Lesions usually start with erythematous papules, gradually enlarges and afterwards it ulcerates. We present a 12-year-old boy with diffuse persistent lip swelling mimicking granulomatous cheilitis. Systemic glucantime was started. However, severe hypotension and bradycardia was developed after injection. Oral fluconazole was started and the lesion resolved completely. Cutaneous leishmaniasis can have varied clinical manifestations and should be suspected especially in endemic areas. Oral fluconazole seems to be safe and effective treatment modality in paediatric cases.

**Key words:** cutaneous, leishmaniasis, glucantime, fluconazole

### Introduction

Cutaneous leishmaniasis (CL) is a protozoan infectious disease, caused by the flagellate protozoan genus, *Leishmania*. This disease is transmitted through the bite of an infected female sandfly. Leishmaniasis is widespread in most countries in the Mediterranean basin, including Turkey [1]. We present an unusual case of CL with diffuse lower lip swelling without nodule or ulcer and responded well to oral fluconazole treatment.

### Case history

A 12-year-old boy presented with 5 months

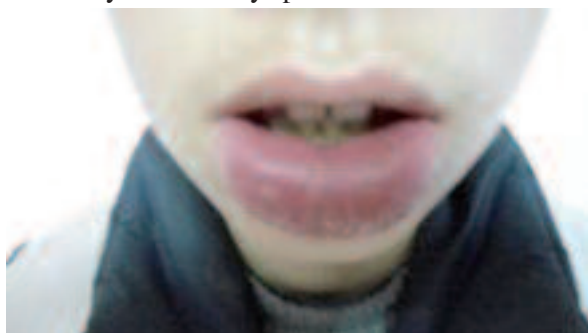


Fig. 1. Diffuse swelling of the lower lip



Fig. 2. Nodular lesion on the upper extremity

history of painless persistent swelling of lower lip and a papule on his right upper extremity. On examination, diffuse firm swelling of lower lip was present (Fig. 1). There was also a 2 cm in diameter, pink-violaceous papule on his right upper extremity (Fig. 2). Systemic examination was normal, with no evidence of organomegaly or lymphadenopathy. Laboratory tests were all within normal ranges.

A punch biopsy was taken from the lower lip and upper extremity. Histopathological examination revealed the presence of Leishman bodies. Meglumine antimonate therapy (glucantime) was started intramuscularly. However, hypotension and bradycardia developed right after the injection.



Fig. 3. Three weeks after therapy

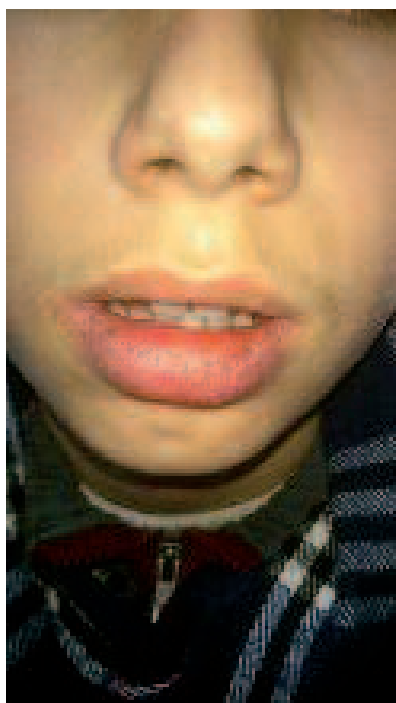


Fig. 4. Six weeks after therapy

Therefore glucantime was stopped and 200 mg/day (5 mg/kg) oral fluconazole was started. The patient was continued the therapy for 6 weeks. Laboratory findings including complete blood count, liver and kidney function tests were within normal limits

during the treatment. At the end of six weeks swelling of the lip and the lesion localized on the upper extremity was regressed (Fig. 3,4).

### Discussion

Leishmaniasis is one of the top 5 diseases targeted by the World Health Organization (WHO) Special Program for Research and Training in Tropical Diseases. Approximately 1.5 million new cases are documented each year and more than 350 million people live in areas of active parasite transmission [2]. Leishmaniasis is endemic in the Mediterranean basin and represent a public health problem in most countries [1]. *Leishmania tropica* is reported to be primarily responsible for CL in Turkey [1,3].

In typical cases of CL, inflammatory lesions are initially papular and enlarge into nodules and/or ulcerations after 3 to 4 months. Eventually the lesions become crusted. Spontaneous healing usually occurs without treatment, however leaves an atrophic scar [4,5]. The typical form of CL with a crusted papule on the upper extremity was present in our patient.

Although CL localized on the lip is not rare, a nodule is often present within the swelling: it may undergo an ulceration which may be covered by crusts and scaling [6–8]. However, diffuse swelling of the entire lower lip without crust or ulceration was present in our patient. Nodular crusted papule localized on the upper extremity was typical for CL and it was the clue for us that lip lesion could also be a *Leishmania* lesion.

There are several factors that affect the choice of the treatment such as duration of the disease, age of the patient and location, number and size of the lesion. The pentavalent antimony compounds are often recommended first line treatment for Old World cutaneous leishmaniasis [9]. In certain cases, when there are few lesions, the lesions are small (4–5 cm), lesions are not localized on joints or aesthetically compromised areas (eyelashes, lips), and there are no signs of lymphangitic dissemination or immunosuppression, local therapies such as cryotherapy, local infiltration with antimonials are good options [10]. As our patient had diffuse lower lip involvement, we preferred to treat with systemic glucantime. However, bradycardia and hypotension was developed right after the injection. Therefore we started to oral fluconazole. At the end of 6 weeks treatment, swelling of the lip was

regressed and papular lesion localized on the upper extremity was also regressed with an atrophic scarring. Oral fluconazole had been reported to be successful in several studies and case reports [5,11–13]. Alrajhi et al. [11] and Emad et al. [12] showed that oral fluconazole was efficacious in patients with CL and reported that it was a suitable alternative therapy for CL. These studies had been performed in adult group. There were a few case reports who had been treated with oral fluconazole in paediatric patients. Toubiana et al. [13] used successfully oral fluconazole treatment in an 11-year-old child and reported that oral fluconazole was safe in children with CL. In another case report, a 2,5-year-old girl with CL had been treated with oral fluconazole [5].

In conclusion, the patient is important in two points: 1. diffuse lip involvement without ulceration or nodule is rare and it is important to differentiate from other diseases that cause lip swelling such as granulomatous cheilitis; 2. oral fluconazole is safe and can be used successfully in paediatric patients. However there is still no consensus on systemic treatment for paediatric patients.

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