The Journal of Laryngology & Otology (2009), ***123***, *915–918.*

doi:10.1017/S0022215108003782

Clinical Record

Auricular leishmaniasis mimicking squamous cell carcinoma

M-T Khorsandi-Ashtiani, M Hasibi*, N Yazdani, J A Paydarfar†, F Sadri‡, F Mirashrafi, A Kouhi

Abstract

Objectives: We report a rare case of auricular involvement by leishmaniasis, in order to demonstrate the importance of thorough investigation of cutaneous head and neck lesions, and also the importance of inclusion of infections such as leishmaniasis in the differential diagnosis of auricular lesions, especially in endemic areas.

Case history: A 42-year-old man with multiple lesions on his head, neck and hands was referred to our centre. He had the following lesions: a painful, crusted, 8 × 8 cm plaque with indurated margins on the left parotid region and auricle; a red papule on the right temporal region; an ulcerative lesion on the skin overlying the proximal interphalangeal joint of the fifth finger of the right hand; and a bluish papule on the neck. Although histopathological examination of the Geimsa-stained specimen was misleading, a direct smear prepared from biopsies showed amastigotes, and therapy resulted in complete recovery.

Conclusion: Leishmaniasis can be both under- or over-diagnosed. Especially in endemic areas, parasitic causes of chronic infections should always be kept in mind.

Key words: Leishmaniasis; Skin; External Ear

Introduction

The aetiology of a chronically painful, red, swollen skin lesion may include infection, inflammatory disease or neoplasia. In the head and neck region, neoplasia is the diagnosis most likely to be considered first. Most bacterial and fungal causes of such lesions are rare, and usually affect other parts of the body. The same is also true of the protozoal infection leishmaniasis, which tends to involve skin away from the head and neck. Ear involvement by this disease is extremely rare, and chronic infection may resemble other infections such as tuberculosis.

According to the World Health Organization, leishmaniasis is endemic in 88 countries, with a total of 350 million people at risk.[1] There are about 1.5 million new cases of cutaneous leishmaniasis reported each year worldwide, mostly from Afghanistan, Iran, Iraq, Algeria, Saudi Arabia, Peru and Pakistan.[2] The condition is also endemic in the Mediterranean basin, particularly in the rural areas of Sicily where almost 60 per cent of Italian cases occur.[2]

The presentation of leishmaniasis varies, from self-limited and even self-healing cutaneous forms to fatal, systemic disease. The lesions of cutaneous leishmaniasis may occur anywhere on the body, but the most likely sites are exposed skin areas. Here, we present an abnormal manifestation of cutaneous leishmaniasis.

Case history

A 42-year-old man with a history of diabetes mellitus and hypertension had initially presented to another hospital in southern Iran with a one-month history of an ulcerative, purulent, tender plaque on the left auricle. He had been admitted with a diagnosis of perichondritis. His diabetes had been aggressively treated, and he had been commenced on intravenous antibiotics. However, after two weeks his condition had not improved, and he underwent surgical debridement of the necrotic and infected tissue. Despite this, there had been no significant improvement, he was referred to Amir-A'lam Hospital in Tehran, a tertiary centre for the treatment of otolaryngology disorders in Iran.

Upon admission, the patient was noted to have multiple lesions: an 8 × 8 cm, crusted, painful plaque with indurated margins on the left parotid region and auricle; a red papule on the right temporal region; an ulcerative lesion on the skin overlying the proximal interphalangeal joint of the fifth finger of the right hand; and a bluish papule on the neck skin (Figure 1). On examination of the ear, there was significant asymmetry and infiltration of the pinna, suggesting a neoplastic process. The external auditory canal was filled with debris and secretions; however, the tympanic membrane was normal. There was no significant lymphadenopathy.

Audiologic evaluation showed normal auditory thresholds. Investigation, including blood cultures and tuberculin testing, was negative. A full-thickness biopsy from an infiltrated margin of the lesion was taken and divided into three parts for impression smear, histological examination and culture. Leishman bodies were seen in the direct smear of the hand and ear lesions. However, histological

From the Department of Otolaryngology, Head and Neck Surgery, Otorhinolaryngology Research Center, and the *Department of Infectious Disease, Amir-A'lam Hospital, Medical Sciences/University of Tehran, the ‡Department of Parasitology, Razi Hospital, Medical Sciences/University of Tehran, Iran, and the †Otolaryngology Department, Dartmouth Hitchcock Medical Center, Lebanon, NH 03756, USA.
Accepted for publication: 24 July 2008. First published online 28 October 2008.

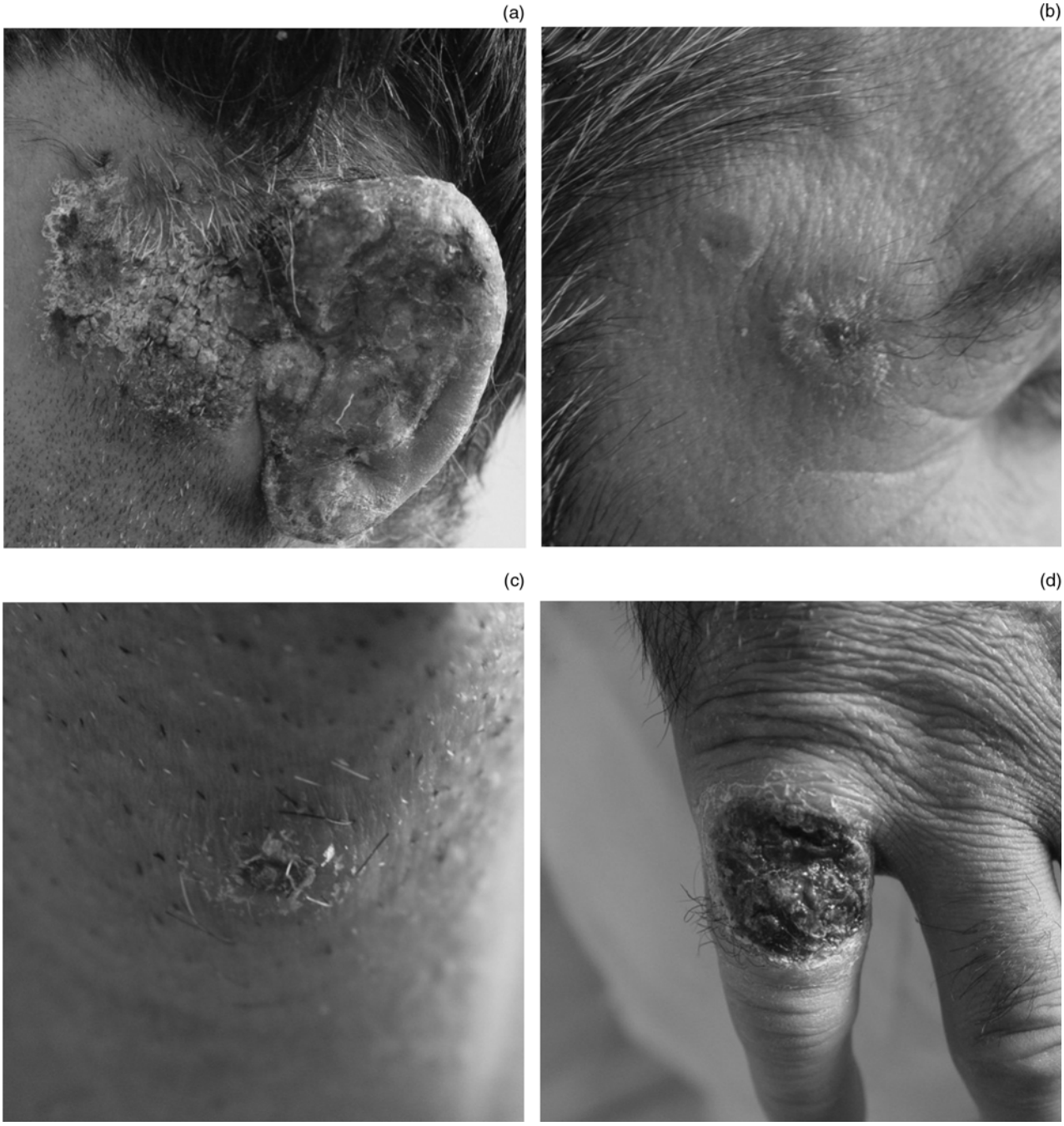

FIG. 1
Initial appearance of multiple lesions on the patient's head, neck and hands. (a) Left auricle; (b) right temporal region; (c) anterior neck; (d) proximal interphalangeal joint of right fifth finger.

examination of the biopsy from the pinna demonstrated hyperplastic mucosa and neoplastic cells invading the stroma, which was interpreted as squamous cell carcinoma.

Given the similar appearance of the multiple lesions, the positive direct smear, the non-malignant appearance of the other biopsies and the fact that the patient was from an area endemic for leishmaniasis, his pathology was thought most likely to be leishmaniasis rather than squamous cell carcinoma. Therefore, he was commenced on anti-leishmania therapy, with Glucantime (a pentavalent antimonial; Aventis, Lyon, France), as two courses at a dose of 20 mg/kg/day intramuscularly for three weeks interrupted by a two-week rest period.

After the second course of anti-leishmania therapy and wound care, the skin lesions were noted to have improved significantly and the pinna size had normalised (Figure 2). The patient was monitored until the lesions had healed fully and the infiltration had resolved. After nearly nine months' follow up, there had been no recurrence. The patient was warned that relapse was possible, and instructed to observe his scars for any sign of thickening, crusting or ulceration.

https://doi.org/10.1017/S0022215108003782 Published online by Cambridge University Press

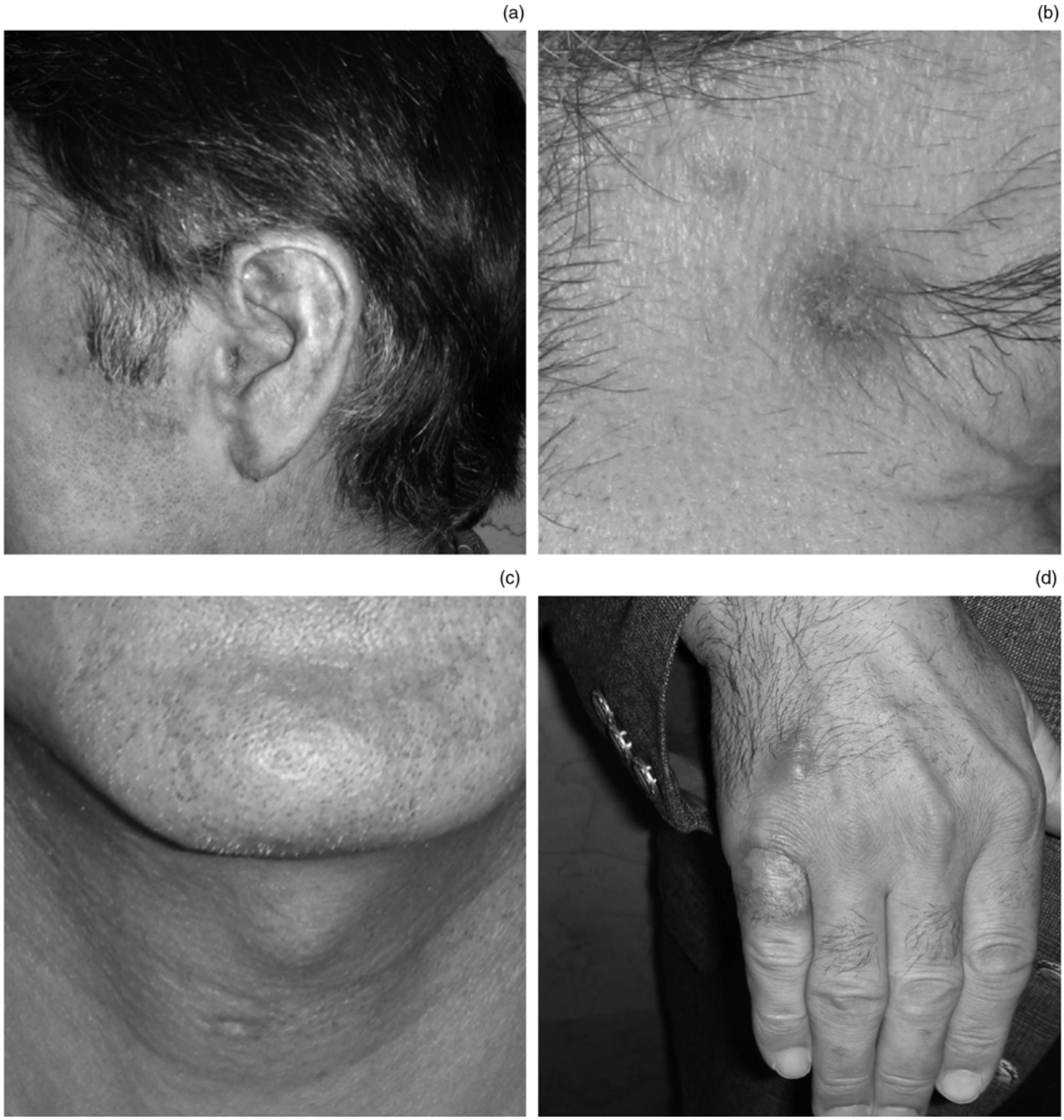

FIG. 2
After two courses of anti-leishmania therapy, the lesions were noted to have improved significantly. (a) Left auricle; (b) right temporal region; (c) anterior neck; (d) proximal interphalangeal joint of right fifth finger.

Discussion

Leishmaniasis is caused by protozoal parasites of the species *Leishmania infantum*, which are carried by the sandfly species *Phlebotomus perniciosus* and *P perfiliewi perfiliewi*.[2] These protozoa are intracellular (i.e. amastigote) in rodents and dogs, which act as reservoirs; there is also an extracellular form (i.e. promastigote) in sandflies, which act as vectors.[3]

The muco-cutaneous form of leishmaniasis, which is almost exclusively restricted to South America, Spain and the Middle East, involves primarily the face and nose;[4,5] to our knowledge, this form of leishmaniasis has not previously been reported to involve the external ear. Leishmaniasis is still an important public health problem, due not only to environmental risk factors (such as large-scale migration, urbanisation, deforestation and new irrigation schemes) but also to individual risk factors (such as human immunodeficiency virus, malnutrition, and genetic and iatrogenic immunodeficiency).[1,6]

Typically, cutaneous leishmaniasis lesions are 0.5–3 cm in diameter; however, in the current case they were much larger. Multiple lesions may be due to multiple sandfly bites.[7]

The diagnosis is often made by the trained eye of a careful physician, based on a typical lesion appearance in conjunction with an appropriate history of exposure,

https://doi.org/10.1017/S0022215108003782 Published online by Cambridge University Press

usually in an endemic area. However, there are a number of mimics to trap the unwary; leishmaniasis may be under- or over-diagnosed and treated (or mis-treated) unnecessarily. The treatment is toxic, so pathological confirmation should be sought, preferably by demonstrating the organism in both tissue and culture.[8] Unfortunately, this is not always possible in clinical practice. The parasite may not be found by the most adequate methods, as was the case with the biopsies taken in the current case. Another problem is pseudo-epithelialisation, seen in many chronic infections, which may be mistaken for neoplasia.

The tissue impression smear technique, with direct examination for amastigotes on Giemsa-stained preparations, has been reported to have a high sensitivity, comparable with conventional histopathological examination. It is easy to perform and particularly useful in cases of multiple suspect lesions.[9] Histological examination was of no assistance in our case, confirming the important role of direct smear examination.

Polymerase chain reaction (PCR) based methods can have markedly great sensitivity in the diagnosis of leishmaniasis.[10] The specificity of this technique is close to 100 per cent.[11] Thus, PCR may be very effective in diagnosing leishmaniasis especially when other methods fail, e.g. microscopic examination as in our case. However, as our patient responded to anti-leishmania treatment and as PCR was unavailable in our hospital, we unfortunately did not perform this test.

- **This paper describes a rare case of auricular involvement by leishmaniasis**
- **After the second course of anti-leishmaniasis therapy and wound care, the skin lesions had improved significantly and the pinna size had normalised**
- **Leishmaniasis can be both under- and over-diagnosed. Especially in endemic areas, parasitic causes of chronic infections should always be kept in mind**

Although it is rare, some leishmaniasis lesions resolve spontaneously with expectant management.[12] In most cases, however, effective active therapy is needed. Antimonial compounds are the first-line agents and are given intralesionally. Cryosurgery, local heat treatment and surgery have also been used successfully.

Our patient was followed up for nearly nine months. Most relapses occur within three to six months of successful treatment, so a follow up visit at six months is considered appropriate.

Acknowledgement

We are grateful to Dr Farshid Mahbubi for help in patient visits.

References

1 Desjeux P. Leishmaniasis: current situation and new perspectives. *Comp Immunol Microbiol Infect Dis* 2004;**27**:305–18
2 Bettini S, Gramiccia M, Gradoni L, Biggio P, Loi R, Cottoni F *et al*. Leishmaniasis in Sardinia. IV. Epidemiological appraisal of cutaneous leishmaniasis and biochemical characterization of isolates. *J Trop Med Hyg* 1990;**93**:262–9
3 Lainson R, Shaw J. Evolution, classification and geographic distribution. In: Peters W, Killick-Kendrick R, eds. *The Leishmaniases in Biology and Medicine*. Orlando: Academic Press, 1987;**1**:1–120
4 Tomson N, Symonds R, Moir A, Kendall C, Wiselka M. New World leishmaniasis from Spain. *Postgrad Med J* 2002;**78**:757–8
5 Clayton R, Grabczynska S. Mucocutaneous leishmaniasis presenting as facial cellulitis. *J Laryngol Otol* 2005;**119**:567–9
6 Pittalis S, Nicastri E, Spinazzola F, Ghirga P, De Marco M, Paglia M *et al*. *Leishmania infantum* leishmaniasis in corticosteroid-treated patients. *BMC Infect Dis* 2006;**6**:177
7 Khan S, Muneeb S. Cutaneous leishmaniasis in Pakistan. *Dermatol Online J* 2005;**11**:4
8 Kubba R, Al-Gindan Y, El-Hassan A, Omer A. Clinical diagnosis of cutaneous leishmaniasis (oriental sore). *J Am Acad Derm* 1987;**16**:1183–9
9 Bahamdan K, Khan A, Tallab T. Value of touch preparations (imprints) for diagnosis of cutaneous leishmaniasis. *Int J Dermatol* 1996;**35**:558–60
10 Safaei A, Motazedian MH, Vasei M. Polymerase chain reaction for diagnosis of cutaneous leishmaniasis in histologically positive, suspicious and negative skin biopsies. *Dermatology* 2002;**205**:18–24
11 Tordini G, Giaccherini R, Pacenti L, Miracco C, Zazzi M, Zanelli G. Cutaneous leishmaniasis: usefulness of PCR on paraffin-embedded skin biopsies as part of routine investigation. *Ann Trop Med Parasitol* 2007;**101**:745–9
12 White J, Salisbury J, Jones J, Higgins E, Vega-Lopez F. Cutaneous leishmaniasis: three children with leishmania major successfully treated with itraconazole. *Pediatr Dermatol* 2006;**23**:78–80

Address for correspondence:
Dr Ali Kouhi,
Otorhinolaryngology Research Center,
Amir-A'lam Hospital,
South Sa'adi Ave,
PO Box 11457-65111,
Tehran, Iran.

Fax: +98 21 66760269
E-mail: akouhi@razi.tums.ac.ir

Dr A Kouhi takes responsibility for the integrity of the content of the paper.
Competing interests: None declared