The Journal of Laryngology & Otology
July 2005, Vol. **119,** pp. 567–569

Mucocutaneous leishmaniasis presenting as facial cellulitis

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Abstract
We report a case of mucocutaneous leishmaniasis caused by *Leishmania viannia braziliensis*. Despite several courses of both oral and intravenous antibiotics no improvement was seen. This case highlights the importance of taking a thorough history, including details of recent travel, and considering rarer causes when no improvement with antibiotics is seen. Our patient was infected with a particularly virulent strain and destruction of the mucous membranes is not uncommon. Rapid diagnosis and treatment are therefore crucial.

Key words: Cutaneous Leishmaniasis, Nasal Cavity; Antimicrobial Chemotherapy

Case report

We report a case of mucocutaneous leishmaniasis. An 18-year-old girl presented to our dermatology department with a seven week history of erythema and swelling of her nose (Figure 1). Prior to this, she had been treated with two courses of oral antibiotics by her general practitioner and one course of intravenous antibiotics by the local ENT surgeons for a presumed facial cellulitis. Six weeks before the onset of symptoms, she had been on a two-week school trip to Belize, Central America. She did not recall any bites to the face, but had toured through the rainforest as well as spending a few nights by the beach in a hut with no windows or doors.

The lesion started as a papule that discharged and crusted over. Eventually, the erythema and swelling spread to involve the right side of her face and upper lip. She remained afebrile throughout her presentation. She was investigated with numerous blood tests (Table I).

No other members of the group were affected. She failed to respond to oral antibiotics, but she did initially respond to intravenous antibiotics, but relapsed as soon as she was switched to oral antibiotics. When she failed to respond to the courses of antibiotics that she had been prescribed, a biopsy was taken from the cutaneous septum under local anaesthetic. Histological examination revealed numerous granulomas (Figure 2), but no organisms were identified on staining. In view of the history and the possibility of leishmaniasis, a second biopsy was taken from the nasal turbinate, which on giemsa staining revealed leishmania amastigotes. PCR performed on the sample was positive for *Leishmania viannia braziliensis*.

TABLE I
INVESTIGATIONS PERFORMED DURING THE PATIENT'S ILLNESS

FBC (incl. differential)	Normal	ANA/ANCA	Normal
U&Es	Normal	Blood cultures	Negative
ESR/CRP	Normal	Skin swab	Negative
TSH	Normal		

After the diagnosis was established, she was admitted to the Hospital for Tropical Diseases for a four-week course of intravenous sodium stibogluconate (20mg/kg/day). Following this, she made a good recovery and 12 weeks post-treatment her nose has returned to normal (Figure 3).

Discussion

New world mucocutaneous leishmaniasis is caused by either the *Leishmania braziliensis* complex or the *Leishmania mexicana* complex, and is transmitted by the bite of infected lutzomyia sandflies. New world leishmaniasis is more severe and aggressive than old world leishmaniasis, and it usually affects parts of the body that are exposed and prone to insect bites, e.g. the face, neck and arms.

There are an estimated 400 000 new cases of leishmaniasis occurring worldwide each year.[1] The incidence of each subtype has not been calculated. Leishmaniasis can be simply divided into three broad classifications depending on the different species:

(1) Cutaneous (oriental) leishmaniasis caused by *Leishmania tropica* in Asia and Africa and by *Leishmania mexicana* in Central and South America
(2) Mucocutaneous (American) leishmaniasis caused by *Leishmania braziliensis*
(3) Visceral leishmaniasis (kala-azar) caused by *Leishmania donovani*

This is an oversimplification as a great deal of overlap exists between the various forms.[2]

Unfortunately there is no gold standard investigation for diagnosing the condition, all the techniques available produce false negatives, and therefore ideally the following four should be positive to make the diagnosis:

(1) Direct visualization of the parasite using either giemsa or leishman stain
(2) Culture of affected material

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This case was presented at the Royal Society of Medicine, Dermatology Section, in London on 20 March 2003.
Accepted for publication: 31 January 2005.

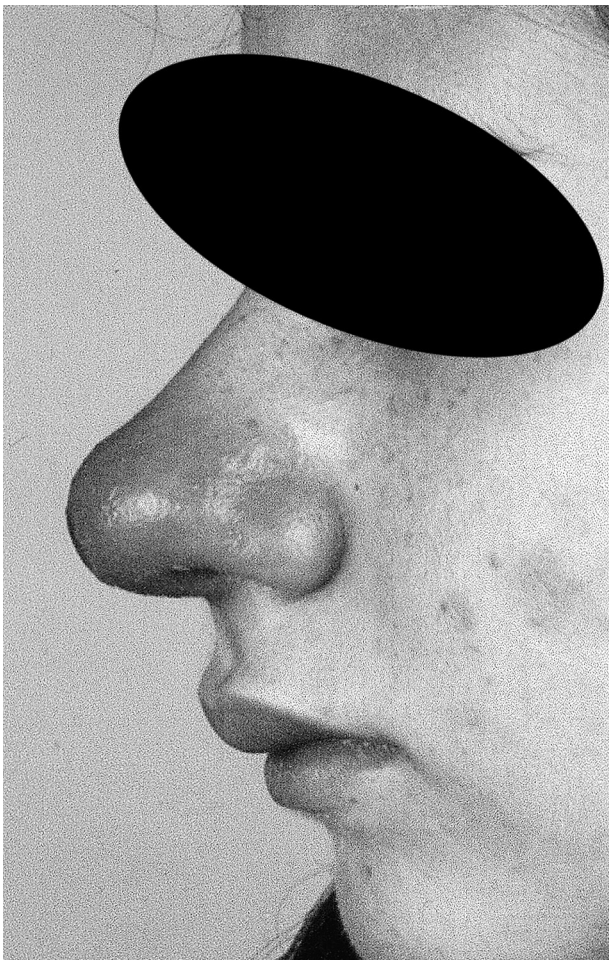

FIG. 1
At presentation to the dermatology department

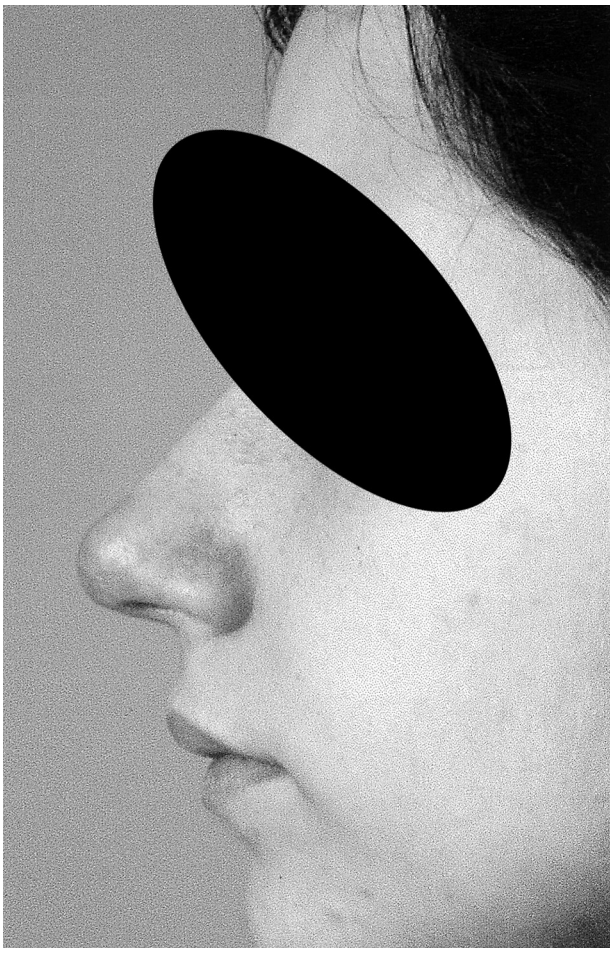

FIG. 3
Twelve weeks after treatment, the lesion has essentially resolved

(3) Histology that classically shows a massive dermal infiltrate of lymphocytes, parasitized macrophages, epitheloid cells and occasional giant cells and plasma cells
(4) PCR to identify the species of leishmaniasis

Although recovery from an infection confers lifelong immunity against re-infection with the same species of parasite, that immunity does not develop early enough or adequately to prevent the blood-borne metastatic spread of parasites of the *Leishmania braziliensis* complex to the mucosa of the nose, mouth, palate or larynx. Here, they may multiply and be recognized immunologically, and cause severe destructive lesions known as espundia. Up to 40 per cent of patients with sores due to *Leishmania braziliensis* may develop mucosal lesions. Approximately 50 per cent of mucosal lesions will develop within two years of the appearance of the skin lesions and 90 per cent within 10 years, although delays of 35 years have been recorded.[3]

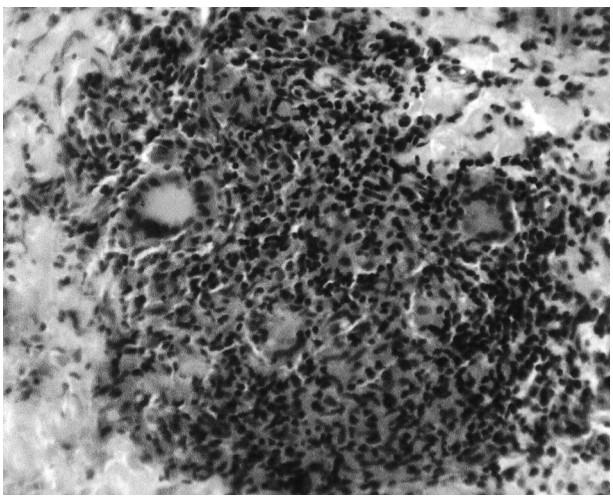

FIG. 2
High power examination of the biopsy showing numerous giant cells within the granuloma. (Haematoxylin and eosin stain; original magnification x 100)

Systemic treatment is required for mucocutaneous leishmaniasis. Topical treatment is not effective and is generally only used for old world cutaneous leishmaniasis. The treatment of choice is pentavalent antimonials such as sodium stibogluconate, used once daily for three to four weeks intravenously. Resistance to sodium stibogluconate has been reported and alternative treatment options include amphotericin B, which is required for at least two months. In patients with chronic or recurrent leishmaniasis, unresponsive to either sodium stibogluconate or amphotericin B, there have been promising results with a combination of pentavalent antimonials and pentoxifylline.[4]

This case highlights the difficulties facing clinicians in diagnosing a rare, predominantly tropical disease. It reiterates the importance of taking a foreign travel history. With leishmaniasis this is especially important, as the travel to an endemic area can have been several years before the onset of symptoms and signs.

https://doi.org/10.1258/0022215054352252 Published online by Cambridge University Press

The case also highlights the fact that if what appears to be a straightforward case of facial cellulitis does not respond to conventional treatment, then atypical causes must be sought.

Acknowledgements

We would like to thank Dr Vega Lopez from the Hospital of Tropical Diseases for his help with this case.

- **Mucocutaneous leishmaniasis can present in the head and neck**
- **This case report illustrates the importance of an awareness of this condition, particularly with a history of recent foreign travel**
- **The clinical features and treatment are discussed**

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Dr R Clayton takes responsibility for the integrity of the content of the paper.
Competing interests: None declared.