The Journal of Laryngology and Otology
September 1993, Vol. **107**, pp. 834–836

Nasal leishmaniasis

G. A. VERNHAM, B. M., F.R.C.S.(Ed.),* H. SADIQ, F.R.C.S.(Ed.),‡ E. A. MALLON, M.B.Ch.B., M.R.C.Path.†
(Glasgow)

Abstract
Leishmaniasis is an uncommon condition in Western Europe, except around the Mediterranean coast. However, it may occasionally be seen in the United Kingdom, in patients who acquired the infection in foreign lands. An unusual case of localized cutaneous leishmaniasis recidiva affecting the nose after septal surgery is presented.

Key words: Nose; Leishmaniasis

Introduction

Leishmaniasis is a communicable disease, caused by protozoa of the genus *Leishmania* (a zoonosis), which are endemic, and sometimes epidemic, in many parts of the world. The condition is rarely seen in the British Isles (Jaffe, 1952; Emslie, 1962), but as illustrated by the case described, it must be considered in patients presenting with granulomatous nasal disease.

Case report

A 40-year-old housewife of Pakistani origin, but resident in the United Kingdom for the last 25 years, presented with a history of nasal blockage. The only significant past history was of a nasal polypectomy four years previously. However, examination revealed a deviation of her nasal septum, and a septoplasty was therefore subsequently performed. She made an uneventful post-operative recovery and was discharged home. However, when reviewed two months later she was complaining of nasal discomfort, with crusting and occasional bleeding. A diagnosis of vestibulitis was made, but treatment, initially with chlorhexidine and neomycin cream, and later with framycetin and gramicidin spray together with steam inhalations failed to produce any improvement. Culture of a nasal swab revealed no growth and treatment with oxytetracycline and hydrocortisone ointment plus systemic amoxycillin and clavulanic acid also failed to produce any clinical improvement. A foreign body was suspected, but the patient declined surgical re-exploration and while on a visit to Saudi Arabia was treated with minocycline without any benefit. Indeed, induration spread over the skin of the nasal tip and she developed a butterfly wing erythema with slight infiltration of the cheeks (Figure 1). Close questioning at this stage revealed a history of an insect bite many years previously which had given rise to a lesion on the tip of the nose at that time. In retrospect, this information was significant, although its importance was not initially appreciated.

Eleven months after her septoplasty, during which time the patient had temporarily returned to Pakistan, intranasal crusting and induration of the nasal tip persisted unabated and a perforation of the cartilaginous septum had developed: a further nasal swab produced coagulase-positive *Staphlococcus*, but treatment with fucidin ointment produced no benefit. X-rays revealed no abnormality of the facial bones or soft tissues. A chest X-ray was

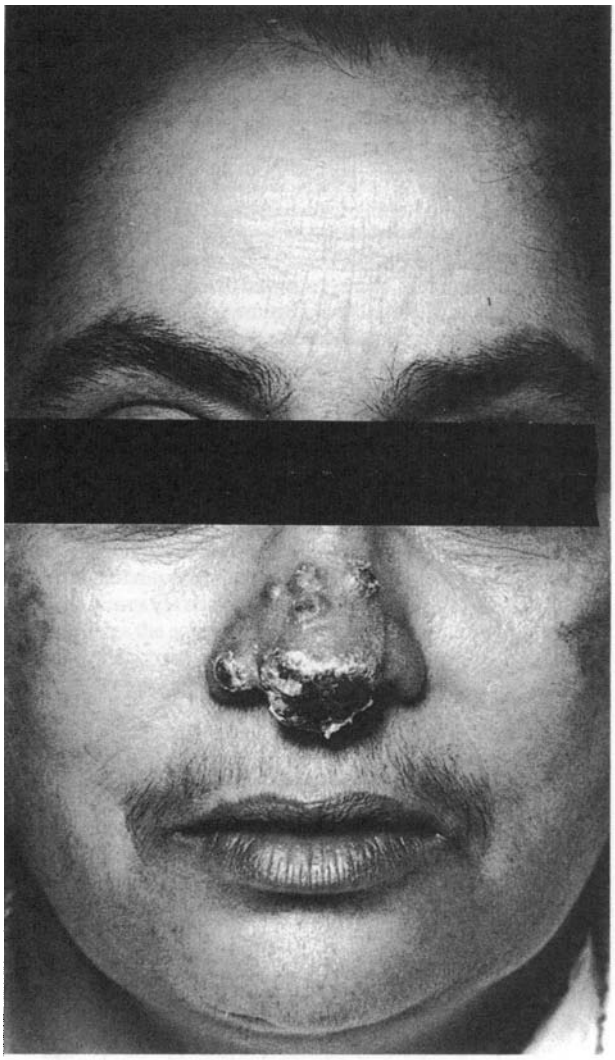

Fig. 1
Induration of the skin of the nasal tip with butterfly wing erythema of the face.

From the Department of Otolaryngology,* and Histopathology,† Victoria Infirmary, Glasgow and the Department of Otolaryngology,‡ Gartnaval General Hospital, Glasgow.
Accepted for publication: 30 May 1993.

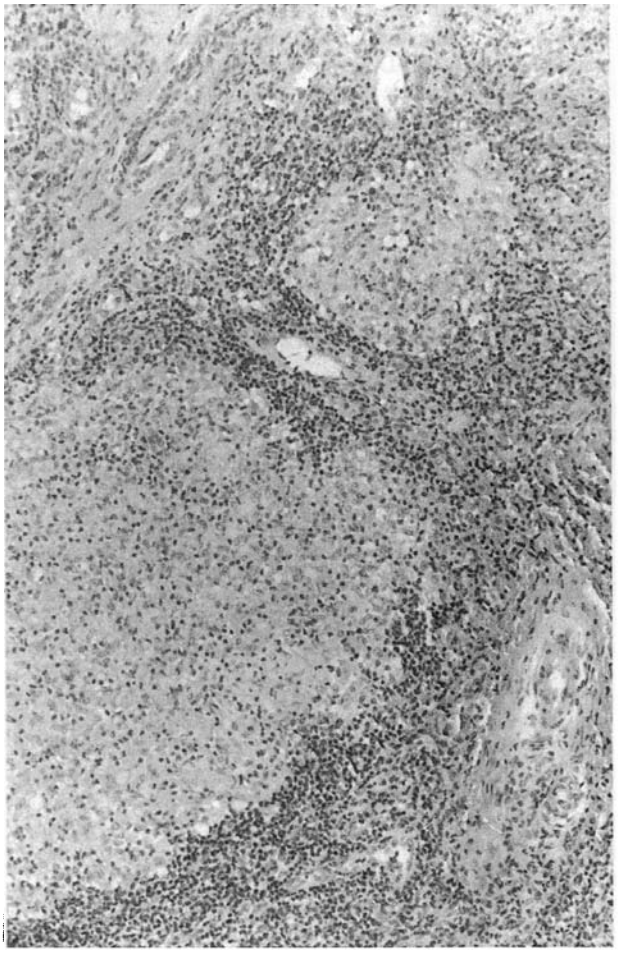

Fig. 2

Biopsy of nasal lesion: a granulomatous lesion with tubercle type follicles without caseation. A marked non-specific chronic inflammatory infiltrate permeates the entire dermis.

also normal. ESR was 3 mm in the first hour. Full blood count revealed some decrease in MCH and MCV, but no significant abnormality. Blood film was normochromic and showed some macrocytes and anisocytosis. Kveim test was negative. A Mantoux 1 in 1000 was positive and biopsy revealed a granulomatous lesion with well formed tubercle type follicles without caseation involving the mid and deep dermis. There was also a marked non-specific chronic inflammatory infiltrate permeating the entire dermis (Figure 2). Staining for tubercle bacilli and for leprosy and atypical mycobacteria was negative, but a diagnosis of tuberculosis was considered probable and anti-tuberculoid therapy commenced (rifampicin, isoniazid and pyrazinamide). However, three months on this regimen produced no improvement and examination of the nose under anaesthetic plus further biopsy was performed.

Lymphoma was considered in the differential diagnosis, but anti-CD3 and pan-B markers produced no evidence for this. Rare fungal and yeast infections were also considered, but all were excluded. Rhinosporidiosis typically presents as a bleeding polyp, spores being found on microcopy; in phycomyocoses, hyphae can be identified in multinucleate giant cells; blastomycosis rarely affects the nose, and presents with cysts containing polymorphonuclear leucocytes surrounding the organism; cryptococcosis usually infects other areas by dissemination from pulmonary infestation; histoplasmosis usually gives rise to hepatosplenomegaly and lymphadenopathy, the yeast organism being identifiable histologically.

Mucocutaneous leishmaniasis was eventually diagnosed by culture of promastigotes of *Leishmania* from biopsy specimens submitted to the Department of Parasitology, Hospital for Tropical Diseases, London (Figure 3). In addition her leishmanin skin test produced an intense, although delayed inflammatory reaction. She was treated by a 10-day course of sodium stibogluconate, which duly produced a rapid resolution of the nasal lesion.

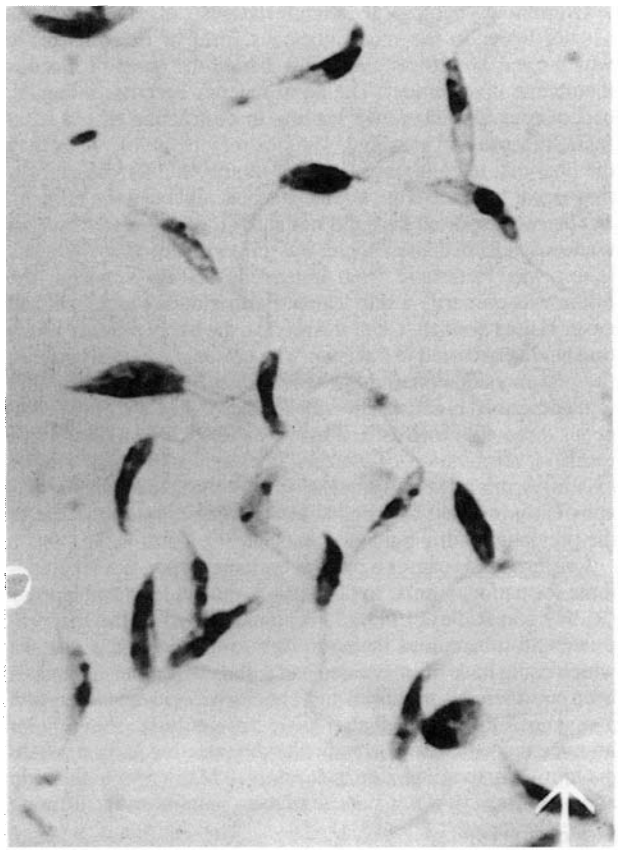

Fig. 3

Promastigotes of *Leishmania*.

Discussion

Leishmaniasis is endemic, and sometimes epidemic, in many parts of the world. *Leishmania* occurs as an intracellular (amastigote) form in man and other mammals, and as an extracellular (promastigote) form in the sandfly. The main reservoir is in rodents and dogs. The classic Leishman–Donovan body is a leishmanial amastigote, an ovoidal or rounded structure, 2–3 μm in length, which occurs intracellularly in monocytes, polymorphonuclear leucocytes, and endothelial cells.

Leishmaniasis is transmitted to man by the bite of the sandfly vector, and occurs in three main clinical forms (Farah, 1979; Bryceson, 1987):

(1) The cutaneous form gives rise to single or multiple lesions affecting only the skin. It is caused by *L. tropica* in the Middle East, Central Asia, and around the Mediterranean coast, and by *L. mexicana* in the rain forests of South and Central America.

(2) The mucocutaneous form, caused by *L. braziliensis*, presents with cutaneous lesions followed by metastatic mucous membrane infection. It occurs in Central and South America.

(3) Systemic leishmaniasis (kala-azar) is a disease of the reticuloendothelial system caused by *L. donovani*. It usually has a protracted course, the main clinical features being fever, emaciation, hepatosplenomegaly, lymphadenopathy, leukopenia, anaemia, and oedema. It is prevalent in India, China, Indochina, around the Mediterranean coast and in parts of Western Africa. In the absence of successful treatment, it is ultimately fatal.

https://doi.org/10.1017/S0022215100124569 Published online by Cambridge University Press

Otorhinolaryngological manifestations of leishmaniasis usually occur in the mucocutaneous form of the disease, in which nasal symptoms commonly herald the onset of mucous membrane involvement. The nasal mucosa becomes inflamed, oedematous and ulcerative leading to destruction of the nasal fossa, mucosa and cartilage. The lips may be involved, as may the pharynx, tonsillar area, floor of mouth, and tongue. Spread may occur to involve the larynx, trachea, and even the bronchi.

The case reported here did not show the features of mucocutaneous leishmaniasis: there was no evidence that the nasal lesion was metastatic from cutaneous disease – indeed, the lesion was primarily a skin lesion. Furthermore, the patient had never visited South or Central America, the likely primary infection having occurred in Pakistan. The prolonged period between the first innoculation and the development of the nasal granuloma described is consistent with an unusual variant of the cutaneous disease, known as leishmania recidiva, which occurs as a result of reactivation of intracellular organisms in or around existing scars or healed lesions. In this case, reactivation was apparently initiated by surgical intervention close to the site of the previous sandfly bite.

Leishmaniasis causes a cellular immune response with granuloma formation, similar to that seen in tuberculosis and leprosy (Ridley and Ridley, 1983). This similarity led to the inappropriate anti-tuberculous therapy given to this patient, a mistake which could have been avoided had a diagnosis of leishmaniasis been considered in the absence of conclusive evidence to support a diagnosis of tuberculosis. As in tuberculosis, the cellular immune response usually eradicates the infection and a permanent immunity to reinfection is developed. Many people living in endemic areas develop such immunity without ever suffering clinically apparent disease.

If a diagnosis of leishmaniasis is suspected, the leishmanin skin test is often useful; 0.1 ml of a suspension of washed promastigotes in 0.5 per cent phenol saline is injected transcutaneously. The result is read after 48–72 hours as for a tuberculin reaction. Histopathological findings in chronic cutaneous leishmaniasis and leishmania recidiva are often inconclusive due to a sparcity of Leishman–Donovan bodies, together with the aforementioned resemblance to tuberculosis and leprosy – tubercles and Langhan's giant cells are present. Diagnosis often depends upon biopsy culture to produce a growth of promastigotes.

Acknowledgements

We wish to thank Mr D. Crossan, Dr P. L. Chiodini and the Medical Illustration Department, Gartnaval General Hospital for their expert help with the illustrations, and Mr E. A. Osborne for permission to report on a patient under his care.

References

Bryceson, A. D. M. (1987) Leishmaniasis. In *Oxford Textbook of Medicine*. 2nd Edition. (Weatherall, D. J., Ledingham, J. G. G., Warrell, D. A. eds.), Oxford University Press, Oxford, pp 5.524–5.533.

Emslie, E. S. (1962) South American leishmaniasis in London. *British Medical Journal* **5274:** 299–300.

Farah, F. S. (1979) Protozoan and helminth infections. In *Dermatology in Clinical Medicine*. 2nd Edition. (Fitzpatrick, T. B., Eisen, A. Z., Wolf, K., Freedberg, I. M., Frank Austen, K., eds.), McGraw-Hill Incorporated, USA, pp 1641–1647.

Jaffe, L. (1952) Probable polypous form of leishmaniasis of the nose. *Practical Oto-rhino-laryngology* (Basel) **14:** 14–18.

Ridley, D. S., Ridley, M. J. (1983) The evolution of the lesion in cutaneous leishmaniasis. *Journal of Pathology* **141:** 83–96.

Address for correspondence:
Mr G. A. Vernham,
Department of Otolaryngology,
Victoria Infirmary,
Langside,
Glasgow G42 9TY.