

Nasal polyp associated with mucocutaneous leishmaniasis. Report of two cases

LUIS VALDA-RODRIGUEZ, JEAN-PIERRE DEDET, PIERRE RAVISSE, RENATO BUSTILLOS, LAURE DIMIER-DAVID, FÉLIX ROLLANO, HEIDY MONASTERIOS

Mucocutaneous leishmaniasis (MCL) can manifest in different forms and affects skin as well as mucosae. Nasal polyps are rare. We report the cases of two male patients with nasal polyps. One of them with cutaneous ulcers and a nasal polyp with the presence of leishmanias detected by pathological examination. In the second case, polyps on the uvula and nasal mucosa appeared during treatment by Amphotericin B. In both cases, the continuation of antileishmanial treatment led to the disappearance of the polyps.

ucocutaneous leishmaniasis (MCL) is relatively frequent in the tropical and sub-tropical zones of various American countries, including Bolivia where it occurs in about two thirds of the territory [1].

After a primary cutaneous lesion, mucosal involvement can subsequently appear. The nasal-buccal-pharyngeal mucosae are mostly affected, with possible extension to the larynx and trachea within a proliferating or ulcerating process. This form of disease is particularly common and severe in Brazil, Bolivia, Peru and Ecuador, where the species Leishmania braziliensis braziliensis predominates [2].

The disease shows a great variety in cutaneous as well as in mucosal lesions which has led physicians to propose numerous classifications based on the clinical aspects of the

Polypous intranasal lesions during mucocutaneous leishmaniasis have rarely been reported in the literature.

Case reports

Case 1

A 29-year-old Bolivian man, was hospitalized in March 1989. He had ulcers on the left buttock and the right thigh and, for two months, nasal obstruction caused by a mass located on the mucosa of the left nasal fossa. These lesions had been treated previously without success.

In the anterior part of the right nasal fossa, was a polypoid mass arising from the inferior part of the septum and e ding to the floor of the right vestibule, partly obstructing nasal fossa (Fig. 1). This polypoid mass reddish grey, sessile, with a smooth surface and a broad base, had approximately the size of a pea, and bled to the touch. There was no ulceration of perforation of the nasal septum. The left nasal fossa was apparently intact, with only slightly hyperemic mucosa. The Montenegro skin test was positive (10 mm), and the indirect immunofluorescent test (IFI) with *Leishmania* antigen was

positive (1/80).

The histopathology of a biopsy of the nasal polyp showed an ulcerated mucosa with fibrin and granulation tissue. Deeper, there was a dense infiltrate of plasma cells, lymphocytes and histiocytes, with formation of epithelioid cell granulomas. These contained scanty intracellular leishmanias (LD bodies, about 5-6 per section; Fig. 2 and 3).
The patient was treated with Meglumine antimoniate (20 mg

Sb⁵⁺/kg/day) plus allopurinol (20 mg/kg/day) for 30 days. At the end of this period the cutaneous and mucosal lesions had healed, and the polyp had disappeared.



Figure 1. Polypoid formation of the inferior part of the nasal floor and extended in part to the floor of the nasal vestibule (Case 1).

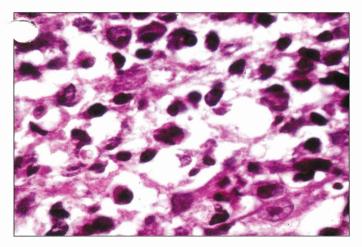


Figure 3. **Lympho-plasmo-histiocyte infiltrate.** An isolated Leishmania is visible at the center. HES × 1,200.

Case 2

5-year-old Peruvian man, with mucosal leishmaniasis resistant to treatment with Meglumine antimoniate for 8 years. At the time of his hospitalization in November 1990, he presented a swelling on the back of his nose and erosive-crusty lesions of the nasal mucosa with perforation of the nasal septum. The oral mucosa, particularly at the level of the velum of the palate and the posterior wall of the pharynx showed an infiltrate with intact uvula. The ENT examination revealed the presence of active granulomatous lesions in the nasal fossae, soft and hard palate and larynx.

The laboratory findings showed a 8 mm Montenegro skin test, IFI positive for leishmaniasis (1/160). Smears for Leishmania were negative. The patient was treated with Amphotericin B in slow intravenous perfusions of 5% dextrose with dexamethasone. An initial dose of 25 mg Amphotericin B was given and later increased to 50 mg, every other day. This was well-tolerated, the side effects were minimal, and the progress favorable until dose 17 of the treatment, when we observed the appearance of a hypertrophy of the uvula, and at dose 20 a polyp formation in the right nasal fossa which in the ENT examination was shown to be a polypoid mass, situated in

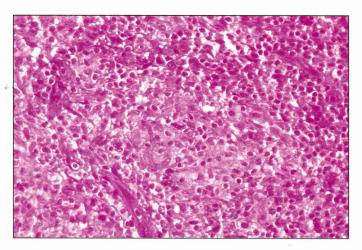


Figure 2. **Epithelioid granuloma infiltrated by some lymphoplasmocytes.** *HES* × 480.

the head of the right inferior turbinate body which caused nasal obstruction and spontaneous epistaxis. The size was approximately that of a pea, of a greyish pink color and partly pedunculated.

Biopsy of the nasal polyp showed partial ulceration. It was very vascular and fibrotic, with plasma cells, lymphocytes and polymorphs, but few histiocytes. No LD bodies could be found

Treatment with Amphotericin B was continued for 30 foses (a total of 1,500 mg), achieving the healing of the mucous lesions and the total disappearance of the nasal polyp.

Discussion

The common evolution of MCL occurs currently in facial mucosae. While the presence of leishmaniae in normal nasal mucosae has been described, the lesions are generally obvious. The mucosal involvement starts gradually in the nose, where the mucosa becomes inflamed, edematous and at a later stage ulcerated. In a second stage, the cartilage of the nasal septum is invaded and then perforated. Gradually, the lesion develops resulting in a total destruction of the nasal floor and consequently a fall of the nasal pyramid, giving the classical aspect of "Tapir Nose" [3]. Sometimes the destruction involves not only the nasal floor but also the nasal fossae, exposing the bony structures of the nose which are generally spared. The resulting sequelae from the nasal lesions in leishmaniasis, bring about various deformities [4].

As part of the proliferating or ulcerating process, the lesions extend to the mucosae of the lips, palate, tonsils, pharynx, larynx and trachea, consequently interfering with phonation, deplitition and recognisation.

deglutition and respiration.

The polypoid form of mucous leishmaniasis is rarely described in the literature and its existence was seriously questioned when the first cases were described. An extensive description of this rare mucosal form of leishmaniasis was made in Brasil by Mangabeira-Albernaz [5] who reported 28 cases of polyps until 1947. Two of these cases showed leishmanias in the pathologic study.

In 1954, Jaffe in Panama [6] described a new case, with a quite illustrative photograph of the protruding character that can develop in this clinical form of nasal leishmaniasis.

Recently in Bahia (Brasil), Marsden [2] reported the presence

of polypoid formation n 8% of patients with unique mucous lesions, usually located on the cartilaginous septum and therefore accessible to examination with a simple nasal speculum. The clinical form, generally unilateral, upon examination usually appears to be a genuine polyp of the nasal septum. The surface is lobulated or smooth, the color varies from yellowish pink to reddish violet, the consistency is soft or hard and a touch may produce bleeding; the base is sessile or pedunculated and the size variable, in some cases completely obstructing the nasal fossa and projecting outside [3, 5]. The observation of numerous cases has permitted Mangabei-

ra-Albernaz to establish the existence of four different types of polypoid mucous leishmaniasis:

(1) a fibrous sessile type,

2) a pedicular mucous type (infrequent),

(3) a more frequent pendulum type, in which the tumor oscillates inside the perforation of the septum,

(4) a diffuse hyperplastic type. According to this author, these four macroscopic types, all with a common histologic aspect, would constitute phases of one same process conditionned by the natural organic resistance or influenced by the treatment.

The existence of this rare clinical variation of mucous leishmaniasis stresses a careful examination of the nose and mouth, which would permit the detection of new cases in the futu-

re.

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L. Valda-Rodriguez, J.P. Dedet, P. Ravisse, R. Bustillos, L. Dimier-David, F. Rollano, H. Monasterios: Departments of Dermatology and Otorhinolaryngology, Hospital de Clínicas, the Instituto Boliviano de Biologia de Altura, Casilla 7426, La Paz, Bolivia, and the Institut Pasteur, 25, rue du Docteur Roux, 75015 Paris, France.

Reprints: L. Valda.

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