

# Case Report

## Mucocutaneous leishmaniasis: does it really exist in Pakistan?

Arfan ul Bari\*, Amir Manzur\*\*

\* Pak Field Hospital-IV, UNAMSIL, Kenema, Sierra Leone (West Africa).

\*\* Department of Dermatology, PAF Hospital, Sargodha, Pakistan.

**Abstract** Mucocutaneous leishmaniasis is considered to be a disease of the New World. It has rarely been reported from our part of the world. A 2-year-old child reported from an endemic area of cutaneous leishmaniasis with clinically suggestive lesions of CL over cutaneous as well as mucosal surfaces of the lip and nose. Diagnosis was confirmed on slit skin smear preparation and he was treated with intramuscular injection of meglumine antimonite (glucantime). A brief review of the condition is also presented along with the case report.

**Key words**

Mucocutaneous leishmaniasis, cutaneous leishmaniasis, *Leishmania* parasite, LT (*Leishmania* trophozoite) bodies.

### Introduction

Leishmaniasis is a group of parasitic conditions due to a flagella protozoan of the *Leishmania* genus and transmitted to man by *Phlebotomine* sandflies. Cutaneous leishmaniasis (CL) exists all over the world. Those of the Old World are, in the vast majority of cases, purely cutaneous and spontaneously regress in several weeks to several months.<sup>1</sup>

Mucocutaneous leishmaniasis (MCL) is predominantly a New World disease and

may not manifest clinically until years after localized cutaneous disease apparently has healed. In a poorly understood manner, certain species of *Leishmania* migrate to the upper respiratory tract where relentless destruction of the oropharynx and nose ensues. In New World it is caused by *L. mexicana*, *L. amazonensis*, *L. braziliensis*, *L. guyanensis*, and *L. panamensis* and may simulate paracoccidioidomycosis, histoplasmosis, syphilis, yaws, rhinoscleroma, squamous cell carcinoma, and midline granuloma of the face. These species of parasite provoke mutilating and disfiguring lesions, resistant to treatment.<sup>1-3</sup> MCL is found infrequently in Old World and is usually due to *L. major* (a dermatropic parasite that is responsible for CL) and very rarely caused by *L. aethiopica* and is characterized by less extensive mucosal involvement, absence of mutilating lesions and the excellent response to treatment.<sup>4,5</sup> Clinically, MCL may be divided into three

---

**Address for correspondence**

Dr. Arfan ul Bari,  
Consultant Dermatologist,  
Pak Field Hospital-IV,  
UNAMSIL, P.O. Box 4670,  
(United Nations Mission in Sierra Leone)  
New York 10163 – 4670  
USA, C/O. CAPO - Q  
Ph # 0023230264187  
E mail: albariul@yahoo.com  
albariul@gmail.com

stages: (1) the primary or cutaneous, (2) the cicatricial, and (3) the secondary or mucocutaneous stages. The primary stage can be further subdivided into lymphoplasmocytic, tuberculoid, and diffuse cutaneous phases and secondary stage can also be subdivided into edematous, granulomatous proliferative, and granulomatous necrotizing on the basis of histopathology.<sup>6</sup> The most common symptom of MCL is persistent nasal congestion, for which the differential diagnosis is broad. Progressive MCL lesions destroy upper respiratory tract mucosa over months and years. Common sites are the turbinates and nasal septum, where erosion of underlying tissue and cartilage may result in perforation. Progressive tissue destruction at the nasal mucocutaneous junction in advanced disease may cause marked disfigurement, requiring reconstructive surgery. Lesions may also affect the palate, pharynx, and larynx, causing palatal dysfunction, dysphagia, dysphonia, and aspiration. Bony structures are not involved.<sup>2,3,6,7</sup> MCL can present in most atypical form when associated with HIV infection.<sup>8</sup>

Diagnosis of MCL, like of CL, is established by demonstrating the presence of *Leishmania* parasites in infected tissues. Serology is rarely helpful except in advanced MCL. Punch biopsies should be taken from the raised, indurated edge of the lesions and sent for microbiological and parasitological examination and in formalin for histopathological assessment. Examination of Giemsa-stained impression smears or slit skin smears for the presence of the intracellular form of the parasites (amastigotes) may quickly yield a diagnosis

in some patients. Amastigotes or leishmania trophozoite (LT) bodies may also be seen in histopathological sections. The flagellate form of the parasite (promastigote) may be cultured from biopsies on modified Novy-McNeal-Nicolle medium incubated for up to three weeks. MCL requires prolonged parenteral treatment with pentavalent antimonials (treatment of choice). Other therapeutic options, mostly do not deliver satisfactory results.<sup>1-3</sup>

### Case history

A 2-year-old child reported in PAF Hospital, Sargodha, from a village in the outskirts of District Khushab with two months history of crusted and slightly oozing lesions over her lip and nose. The lesions started as small papules on the margin of lip and nose and gradually progressed to form a large crusted plaque. The child became irritable and her dietary intake was reduced, possibly due to pain and discomfort associated with the lesions. She was initially managed with conventional oral and topical antibiotics but the lesions continued to progress. On examination she was found to have two isolated crusted indurated plaques. One was on mucocutaneous junction of lower lip and the other one was located on the right ala of the nose and was extending inward to the mucosal surface of the nostril (**Figures 1 and 2**). Submental or cervical lymph nodes were not palpable. Systemic examination was unremarkable. Considering the nature of the lesions and origin of the patient from a geographical area (endemic focus for CL), she was provisionally diagnosed as a case of leishmaniasis with mucocutaneous involvement. Impression skin smear with Giemsa stain was made and luckily it



**Figure 1** Two indurated, nonhealing crusted plaques over lower lip and right nostril margin.



**Figure 2** Further close up of the same lesions.

revealed LT bodies and thus confirmed the diagnosis. She was treated with Inj meglumine antimonite (Glucantime) 10 mg/kg intra muscular (gluteal regions) daily. The lesions started regressing after three injections and substantially healed after 10 days. She could not continue with the injections after 10 days (non compliance due to pain of injections), and was advised oral rifampicin 5 mg/kg daily before breakfast.

## Discussion

MCL is endemic in much of South America and is mostly caused by *L. brasiliensis*. The disease usually begins as typical sores of localized CL. Mucosal lesions may be present with cutaneous lesions, but more

commonly mucosal disease appears years after the skin lesions have healed.<sup>2,3</sup> Mucosal disease usually presents with chronic nasal symptoms that may include increased secretions, epistaxis, pain, and other evidence of inflammation. These lesions are particularly destructive and painful in New World MCL. They erode underlying tissue and cartilage. The precise mechanisms by which the parasites disseminate from the skin and localize within macrophages in the naso-oro-pharyngeal membranes are unknown, as is the reason why mucosal disease subsequently develops in only a few patients with cutaneous disease. Fever, weight loss, anemia and secondary infections may be associated with mucosal disease.<sup>2,3,7</sup>

In addition to South and Central America, MCL affecting nose and mouth is fairly common in Ethiopia and this resembles South American leishmaniasis but probably results from direct extension from skin lesions rather than from metastatic spread of organisms.<sup>4</sup> It has infrequently been reported from Middle East, Far East and India.<sup>5,9,10</sup> We could not find any such case of MCL in our local literature. This case was unusual from commonly occurring cases of CL, as the lesions were at mucocutaneous junctions involving both the cutaneous as well as mucosal surfaces but it was not as classical as most cases of New World MCL where disease is caused by secondary dissemination of the parasites and is potentially more destructive and mutilating.<sup>2,3,7</sup> Probably this was either a case of primary CL caused by dermatropic *L. major* or *L. tropica* with extension to mucosa or it was the result of simultaneous bites of the sandfly over cutaneous and

adjacent mucosal area. This was more likely as the child was too young to have any protection against sandfly bites and she also belonged to an area where there were cluster of cases of CL.

### Conclusion

Although MCL seems to be non-existing in Pakistan, we should be forewarned due to ever increasing cases of CL with varied clinical presentation, more frequent traveling and possibility of emergence of new mucotropic strains of parasite.

### References

1. Herwaldt BL. Leishmaniasis. *Lancet* 1999; **354**: 1191-9.
2. David C, Dimier-David L, Vargas F *et al*. Fifteen years of cutaneous and mucocutaneous leishmaniasis in Bolivia: a retrospective study. *Trans R Soc Trop Med Hyg* 1993; **87**: 7-9
3. Ahluwalia S, Lawn SD, Grant JKH, Lockwood DNJ. Mucocutaneous leishmaniasis: an imported infection among travellers to Central and South America. *BMJ* 2004; **329**: 842-4.
4. Barnetson RS, Ridley RS, Wheate HW. A form of muco-cutaneous leishmaniasis in the Old World. *Trans R Soc Trop Med Hyg* 1978; **72**: 516-8.
5. Al-Gindan Y, Omer AH, Al-Humaidan Y. A case of mucocutaneous leishmaniasis in Saudi Arabia caused by *Leishmania major* and its response to treatment. *Clin Exp Dermatol* 1983; **8**:185-8.
6. Sanguenza OP, Sanguenza JM, Stiller MJ, Sanguenza P. Mucocutaneous leishmaniasis: a clinicopathologic classification. *J Am Acad Dermatol* 1993; **28**: 927-32
7. Weller PF, Durand ML, Pilch BZ. A 35-year-old man with nasal congestion, swelling, and pain. *N Eng J Med* 2005; **352**: 609-15.
8. Calzaa L, D'Antuono A, Marinacci G *et al*. Disseminated cutaneous leishmaniasis after visceral disease in a patient with AIDS. *J Am Acad Dermatol* 2004; **50**: 461-5.
9. Kumar MKA; Raghunandanan SG; Kurian CA. Mucocutaneous leishmaniasis. *Indian J Otolaryngol Head Neck Surg* 1996; **48**: 238-40.
10. Kharfi M, Fazaa B, Chaker E, Kamoun MR. Mucosal localization of leishmaniasis in Tunisia: 5 cases. *Ann Dermatol Venereol* 2003; **130**: 27-30.