Isolated oral mucosal leishmaniasis

Sir,

A 34-year-old man from Bihar, which is a state endemic for kala-azar in India, had a one-and a half year history of a fleshy swelling inside the oral cavity accompanied by a change in his voice and difficulty in deglutition. The swelling began as an approximately 1 cm sized nodule on the hard palate that increased progressively in size. The patient was not a smoker or alcoholic and was not promiscuous. He gave no history of any surgery, blood transfusions or drug abuse.

A biopsy had been taken from the mass and the report stated that there were numerous ill-formed granulomas with overall features of chronic inflammation without significant atypia. Based on the biopsy report, the patient was prescribed anti-tubercular treatment (ATT) by his physicians. The patient was referred to

New Delhi when no improvement was noted after two months of treatment.

On examination, a proliferative fleshy growth with a cauliflower-like surface was noted within the buccal cavity involving the entire hard and soft palate and both the tonsils and extending to the posterior pharyngeal wall [Figure 1]. The lesion was confined to the oral cavity. There were no skin lesions. The jugulo-digastric lymph nodes on both sides and multiple lymph nodes in the posterior triangle were enlarged. Routine laboratory tests were within normal limits. Contrast enhanced CT scan of the neck showed a homogeneous, lobulated, strongly enhancing mass lesion involving the hard and soft palate and extending into the nasopharyngeal air column. A soft tissue mass was also seen at the posterior end of the nasal septum and within the right nostril. A dense, lobulated, soft tissue mass filling up the right pyriform fossa, pharyngeal mucosal spaces, vallecula and also pre- and para-laryngeal fat spaces was noted.

A repeat biopsy was taken from the mucosal mass. Impression smears from the biopsy were sent for parasitological workup to the Microbiology department, All India Institute of Medical Sciences, New Delhi. Giemsa and acridine-orange stained smears revealed numerous Leishman-Donovan (LD) bodies. The serum aldehyde test was positive and serum total protein was 8.7 g%, with albumin being 3.9 g% and globulin 4.8 g%. The test for serum antibodies against *L. donovani*-specific kinesin antigen (rK-39) was positive by an immune-chromatography test (InBios International, Washington 98104, USA).

A diagnosis of mucosal leishmaniasis caused by *L. donovani* was made and the patient was subsequently treated with intramuscular sodium stibogluconate 800 mg daily for 20 days. There was prompt improvement with clearance of the larynx and disappearance of the growth [Figure 2]. At follow up, microscopy of a biopsy from fibrotic remnants of the lesion was negative for LD bodies and histopathological examination revealed fibrotic and mild inflammatory changes. The serum rK-39 antibody test, however, continued to remain weakly positive.

Visceral leishmaniasis is endemic in some states of India; small outbreaks of cutaneous leishmaniasis due to L. donovani were also reported from India and Sri Lanka but mucosal leishmaniasis is exceedingly rare in the subcontinent.[1]

Mucosal leishmaniasis is most commonly associated with *Leishmania braziliensis*, and is limited mostly to South America. There are rare reports of mucosal involvement by *L. donovani* in Sudan.[2] Mucosal leishmaniasis does not heal spontaneously; secondary bacterial infections and other complications are common making the disease potentially fatal.

In the presence of immune deficiency/suppression, *L. donovani* can manifest in an unusual manner but in an immunocompetent person, leishmaniasis presenting as an isolated mucosal lesion without any cutaneous manifestations is exceedingly rare.[3-6] Naik *et al*. in 1978, reported a case of leishmaniasis in which a huge nasopharyngeal tumor was the presenting complaint; however, a final diagnosis of visceral

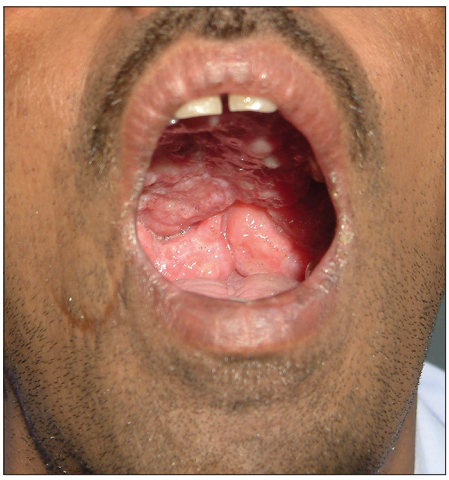

Figure 1: Before treatment, proliferative growth in the buccal cavity involving the hard palate, soft palate, tonsils and posterior pharyngeal wall

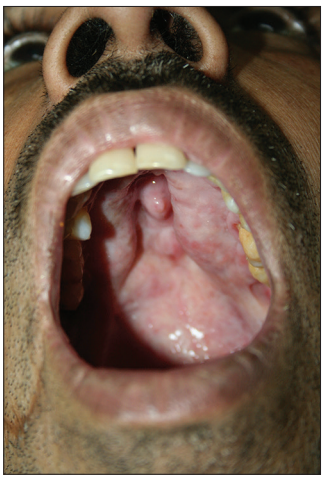

Figure 2: After treatment, complete regression of the mass barring the small fibrotic remnant persisting in the hard palate

leishmaniasis was made because of accompanying systemic manifestations and demonstration of LD bodies in the splenic aspirate.[7] Cases with atypical clinical manifestations can be initially missed as happened in the present case. Specific diagnosis followed by appropriate treatment eventually resulted in cure of the disease. A high index of clinical suspicion for leishmaniasis is warranted in patients coming from endemic areas.

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