A 33-year-old male farmer from eastern Iran presented with progressive gingival swelling and bleeding of several months duration. His past medical history was unremarkable. Oral examination revealed gingival hypertrophy, especially of the upper jaw and several plaques and papules (cobblestone appearance) of the hard palate (Figures 1, 2). Systemic examination did not reveal any additional cutaneous lesions, enlarged lymph nodes, or hepatosplenomegaly. The complete blood count, serum electrolytes, urea, liver function tests and chest radiography were within normal limits.

• What is the likely cause of the oral lesions?
• How could the diagnosis be confirmed?

(Answer on page 268)
Diagnosis: Oral leishmaniasis

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The differential diagnosis of gingival hyperplasia with or without hyperplasia of the palate mucosa includes inflammatory gingival hyperplasia, Dilantin hyperplasia, inflammatory papillary hyperplasia, and leukemia and lymphoma. Inflammatory hyperplasia of the gingiva usually results from prolonged chronic inflammation of gingival tissue, which could be associated with Vitamin C deficiency, endocrine imbalance, Crohn's disease, or infection, including mucosal leishmaniasis. Gingival hyperplasia usually begins 2 to 3 months after Dilantin therapy and is almost entirely confined to the gingival tissue surrounding the teeth. On rare occasions hyperplasia may occur in areas apart from the gingiva, such as the palate, in patients wearing a prosthetic appliance. Papillary hyperplasia occurs predominately in edentulous patients with dentures. Gingival hyperplasia may be an early finding in patients with leukemia (e.g., AML) or lymphoma, secondary to infiltration of the gingival mucosa.

A biopsy was performed from the oral mucosal lesion. Histological examination revealed pseudoepitheliomatous hyperplasia of the squamous epithelium with dense inflammatory infiltrates in the dermis composed of lymphocytes, plasma cells and histiocytes containing numerous amastigote forms of Leishmania (Figure 3). Bone marrow aspiration and trephine examination showed no evidence of dissemination of leishmaniasis. A diagnosis of mucosal leishmaniasis was made and treatment was started with intramuscular meglumine antimoniate (20 mg/kg/day for 20 days). The patient showed a dramatic response and the oral lesions almost completely disappeared.

Discussion

Leishmaniasis is infection by protozoans of the genus Leishmania. It is one of the neglected diseases that usually afflict the world’s poorest people. Leishmaniasis is transmitted by the bite of sandfly and presents in three forms: cutaneous, mucocutaneous and visceral. Early descriptions of the parasite in cutaneous lesions were done by Cunningham, Borovsky and Wright between 1885 and 1903. Other forms of leishmaniasis were described later. The clinical manifestation of leishmaniasis depends on the interaction between the characteristic virulence of the species and host immune response. Mucosal leishmaniasis is a chronic infection of the mucosal membranes, which in most cases is primary but may develop during or after an attack of visceral leishmaniasis. There are at least 30 species of Leishmania, of which 12 named and several unnamed species affect man. Leishmania live two quite separate lives—one in the sandfly, the other in mammals. In the sandfly, the organism exists as the promastigote (Leptomonad), and in tissue as the amastigote (leishmanial or aflagellar form). These parasites are endemic to the tropics of the Americas, parts of Asia, Europe and tropical Africa north of the equator. There are reports of mucosal leishmaniasis from different parts of the world, which almost in all cases...
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Figure 3. Histology of the oral lesion showing macrophages containing numerous Leishman bodies (H&E stain, X400).

References


In Iran, infection by protozoa usually leads to visceral or cutaneous disease. Primary mucocutaneous or mucosal leishmaniasis is very rare. Our case is among the few reports of this condition from Iran. The clinical picture of this case appears to be more similar to the cases reported from Sudan. More cases need to be reported from Iran to be able to present the various characteristics, which may be unique to mucosal leishmaniasis in Iran.