ORAL LEISHMANIASIS: REPORT OF THREE CASES - WITH DIFFERENTIAL DIAGNOSIS

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SUMMARY

Three cases of visceral leishmaniasis with oral manifestations are described. Two of these cases presented with chronic non-healing ulcers. The third case presented with characteristic erythematous, swollen and bleeding gingiva. The clinical appearance of both forms of presentation is discussed, and the importance of including oral leishmaniasis in the differential diagnosis of similar oral lesions highlighted.

Key words: Leishmaniasis, Syphilis, Leukemia, Acute Necrotizing Ulcerative Gingivitis

INTRODUCTION

The term leishmaniasis refers collectively to various clinical syndromes that are caused by obligate intracellular protozoa of the genus leishmania. They cause a wide range of infectious processes, from asymptomatic to cutaneous, mucosal or visceral manifestations depending on the species of the parasite and the type of immune response induced. In mucocutaneous leishmaniasis patients may present with a painless nodule or ulcer with oedematous areola and the formation of the high edges at the site of infection. The pharynx, palate, larynx and upper lip are the usual sites affected. In cutaneous leishmaniasis, the involvement of the lip and perioral tissues has been noted. The lesion involves the area immediately below the vermilion border, and no solely mucosal lesion are seen. Recently, involvement of the mouth proper has been reported to occur in some cases of visceral leishmaniasis. The patient may present with swollen and bleeding gingiva, loosening of the teeth, erythematous palate, and even gangrenous stomatitis.

The diagnosis of leishmaniasis in endemic areas is presumptive and is early made by the classical clinical presentation of the disease. However, in areas in which leishmaniasis is not usually found, unfamiliarity with the disease makes diagnosis difficult. The diagnosis is confirmed by identifying leishmania sp. amastigotes in tissue or by growing promastigotes in culture. Three cases of visceral leishmaniasis with oral manifestations are presented here. Their importance lie in the differential diagnosis which includes: squamous cell carcinoma, tuberculosis, syphilis and leukaemia.

CASE 1

A 44-year-old patient from Gedariff State (an area in eastern Sudan that is endemic for visceral leishmaniasis) was referred to our department with an oral ulcer (Fig. 1). The ulcer had appeared three months previously and failed to heal despite the use of many medications. On examination the patient looked well and healthy apart from a slight asymmetry of the face showing some swelling of the left cheek. Regional lymph nodes were palpable and tender. Intra-oral examination revealed a deep ulcer in the mucogingival fold extending from the midline backward up to the first premolar tooth. The edges of the ulcer were raised and irregular. No induration was felt around the ulcer. The gingiva was red in color, and easily bleeding. A...
Fig. 1 A 44-year old patient with a deep ulcer in the muco gingival fold — lower jaw
differential diagnosis of squamous cell carcinoma, tuberculosis, syphilis and leishmaniasis was made. Full blood count (FBC) and ESR were normal. A smear biopsy taken from the inflamed gingiva and stained using the Gimsa method demonstrated the leishmania parasite. The patient was then referred to the leishmaniasis ward at Soba University Hospital for management.

CASE 2

A 47-year old patient was referred to our department with an oral ulcer of long duration (Fig.2). The condition started as a small nodule which then ulcerated. The patient appeared healthy and was from an endemic area for visceral leishmaniasis (Sennar). Examination revealed a 2x2 cm ulcerated area in the labial mucosa opposite to the lower right canine. The ulcer had an irregular but raised margin, the floor of which was coated with a yellowish material. The gingiva and the remaining oral soft tissues were within normal limits. A differential diagnosis of tuberculosis, squamous cell carcinoma, syphilis and leishmaniasis was made. Both the polymerase chain reaction and tissue culture were positive for the leishmania donovani bodies. The patient was then referred to the leishmaniasis ward at Soba University Hospital for further management.

CASE 3

A 32-year old patient from the south of Sudan presented to our department with chronic diffuse form of gingivitis (Fig 3). The patient was from a region known to be endemic for leishmaniasis. The condition had started more than 6 months previously and since then the patient had difficulty in eating and drinking. He had also complained of fever, diarrhea and weight loss. Examination revealed severely inflamed gingiva, which was oedematous, red and sponge-like. There was spontaneous bleeding. The palate was also erythematous and inflamed. The regional lymph nodes were enlarged and tender. A differential diagnosis of leishmaniasis, acute necrotising ulcerative gingivitis (ANUG), and leukaemia was made. A full blood count revealed patient was anaemic as well as thrombocytopenic (9g/dl & 50X10/1 respectively). The histopathology examination using the Gimsa method confirmed the diagnosis of leishmaniasis. The patient was then referred to the leishmaniasis ward at Soba University Hospital for further management.

DISCUSSION

Visceral leishmaniasis is endemic in large parts of rural Sudan. The symptomatic patient usually complains of fever, discomfort from abdominal swell-
ing, weight loss and diarrhoea. The symptoms may relate to the nasal and oral cavities, and also to the nervous system.9,11,16,20

All three patients reported herein were from areas endemic for visceral leishmaniasis. Two presented with oral ulcerations and the third presented with swollen and bleeding gums. Apart from the oral ulceration, the first two patients were symptomatic. Their infection appeared to be confined to the lip mucosa and not accompanied by visceral involvement.5,26

Ulcerative lesions are commonly encountered in patients attending the oral surgery clinic. Their etiology ranges from reactive to neoplastic oral manifestation of systemic diseases. In the first two patients the differential diagnosis included squamous cell carcinoma, tuberculosis, syphilis and leishmania. The oral leishmanial ulcer is difficult to differentiate from the malignant ulcer on history or clinical appearance, particularly in the asymptomatic patient. A biopsy is therefore essential. The parasite can be identified by different methods including isoenzyme characterization, serologic analysis using monoclonal antibodies and recombinant deoxyribonucleic acid (DNA) technology.13,16,23 On the other hand, oral cancer in Sudan is not infrequent, particularly carcinoma of the labial mucosa and the gingiva.9 This is generally linked to snuff dipping in the labiogingival sulcus of the lower lip.24 The clinical picture varies from a white patch to a non-healing ulcer to an exophytic lesion.7,28,29 The oral leishmanial ulcer should also be differentiated from a tuberculous ulcer. The latter usually follows implantation of M, tuberculosis from infected sputum in the oral mucosa, and the tongue and the palate are favoured locations.29 The typical lesion is an indurated, chronic, non-healing ulcer that is usually painful. The edges are thin, undermined and often bluish.4,14,28,29 A high ESR and a+ve mantoux test are highly suggestive of the disease and confirmation can be obtained by a sputum culture or Ziehl-Nielsen stain. Oral leishmaniasis should also be differentiated from syphilis. The latter disease was described as the great mimicker or imitator because of its resemblance to many other unrelated conditions including leishmaniasis.29 In the mouth, syphilis manifest itself as a primary chancre which has indurated base that feels like a button, as snail-track ulcers which characterise the secondary stage of the disease and found mainly at the mucocutaneous junctions, or as a gummatous ulcer which is invariably met with in the palate and which usually has the characteristic features of the punched-out edge and the yellowish-Grey floor.7,14,28,29 The diagnosis of syphilis is confirmed by serological examination and the disease in general responds well to penicillin in contrast to other chronic ulcers. Based on clinical signs and symptoms only, the oral leishmanial ulcer can not be differentiated from several other conditions; therefore careful history and examination are important. Patients coming from endemic areas are highly suspect, though laboratory investigations are essential to confirm the diagnosis.

The oral manifestation of leishmaniasis in the third patient was different. The sponge —like appearance, the scarlet colour and the generalised oedema of the gingiva as well as that of the palate are typical features of the disease. The differential diagnosis of this case included leukaemia and acute necrotising ulcerative gingivitis (ANUG). In the acute myelomonocytic type of leukaemia, the gingiva is infiltrated with leukaemia cells. These leukaemia deposits occasionally cause gingival swelling, and the patient may present with bluish haemorrhagic swelling of the gingiva, invariably those of the lower jaw.4,31 However, the condition can be ruled out or confirmed by laboratory investigations including full blood count (FBC) and biopsy. In ANUG, painful ulcerations of the interdental papillae and the marked bleeding tendency of the ulcerated tissues are typical features. In addition there is often an accompanying halitosis and in severe cases the infection may involve the mucosa of the soft palate or tonsils.29,31 In contradistinction to oral leishmaniasis, in ANUG relief is often immediate and marked following local debridement, and oral hygiene care supplemented with a 7-day regimen of metronidazole therapy.

The drug of choice for leishmaniasis is pentavalent antimony sodium stibogluconate and different regimes are under continuous evaluation.3,8,15,19,27 This drug was recommended by WHO at a dose of 20 mg/kg per day over a period of 4 weeks.33 All three patients reported here received sodium stibogluconate (Pentostam R, Welcome lab, UK) 20 mg/kg per day for 28 days. The drug was well tolerated. Response to therapy was judged by the improvement in general condition of the patient, the regression of the splenomegaly and the rise in the haematological indices. Healing of the mouth ulceration and regression of the oral and gingival swelling had occurred by the end of the treatment.

CONCLUSION

Visceral leishmaniasis may manifest itself in the mouth as a chronic non-healing ulcer, or as a form of
stomatitis. The disease must be included in the differential diagnosis of chronic lesions affecting the oral mucosa, particularly in areas known to be endemic. The oral leishmanial ulcer usually has an irregular margin and slightly raised edge. The ulcer needs to be differentiated from the tuberculosis and syphilitic ulcer as well as from squamous cell carcinoma. In cases of the stomatitis, the gingiva are always hypertrophied, reddish in colour and easily bleeding, and the palate may also be involved. The condition must be differentiated from leukaemia and necrotising ulcerative gingivitis. In both forms of oral manifestations the diagnosis of oral leishmaniasis should be confirmed by laboratory investigations.

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REFERENCES


24. Idriss AM. A study investigating the etiological association between oral cancer and use of toombak in the Sudan A thesis submitted for the degree of Doctor of philosophy (PhD). Dept. of Pathology, Faculty of Medicine, University of Khartoum, 1991.


