

MEDICAL MEMORANDA

Granulomatous Cheilitis in Crohn's Disease

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British Medical Journal, 1974, 4, 636

Oral lesions in Crohn's disease are well recognized but involvement of the lips is less common. Such a case is reported here in which the patient presented with granulomatous cheilitis several years before evidence of ileitis developed.

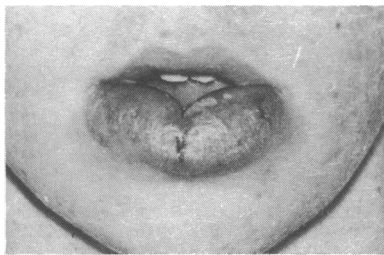
Case Report

In 1965 a 14-year-old boy developed pain and swelling of his lower lip and right cheek. This fluctuated in severity over several months and was accompanied at times by oral ulceration. Examination showed swelling and inflammation of the lower lip, a thickened ridge of tissue opposite the upper right alveolus, and induration inside the right cheek. Biopsy of the indurated area showed multiple, non-caseating, giant-cell granulomata. The lesions failed to respond to antibiotics and local corticosteroid creams but responded partially to systemic prednisone 5-10 mg daily.

In 1968 he had rectal bleeding, and several fleshy perianal skin tags were removed. Histologically these tags, which were covered with stratified squamous epithelium, showed stromal infiltration by inflammatory cells and prominent epithelioid-cell granulomata with giant cells, some of which contained birefringent material. No tubercle bacilli were identified. The diagnosis of perianal Crohn's disease was made.

In 1969 the lip swelling increased (see fig.). Excision of a median labial fissure, labial biopsy, and plastic repair were carried out. The excised labial tissue showed fissuring of the mucosal surface. In places the stratified squamous epithelium appeared normal but elsewhere it showed parakeratosis, spongiosis, and intracellular oedema. The fissures were lined by extensions of the surface epithelium. The subepithelial tissues showed fibrosis and oedema, and in addition numerous dilated vascular spaces were present. There was a diffuse infiltrate of lymphocytes, plasma cells, and histiocytes. Multiple, small, non-caseating granulomata were present. Birefringent material was present in some of the large multinucleate epithelioid cells. The picture was one of a non-specific granulomatous cheilitis with histological appearances closely resembling those of the previously removed anal skin tags. At the time the haemoglobin, white cell count, erythrocyte sedimentation rate, serum mucoproteins, and barium meal and follow-through were all normal. The Mantoux test gave a negative result.

In 1974 he developed lower abdominal pain, flatulence, nausea, and night sweats. Examination showed marked swelling of the lower lip with less swelling of the upper lip. Both lips appeared inflamed



Appearances of lower lip at age 18.

and he had angular stomatitis. Ulcers were present on the soft palate and in the lower left vestibular sulcus. He had some induration and reddening of the perianal skin. No masses were palpable in the abdomen and there was no superficial lymphadenopathy. He had a low-grade fever. Barium follow-through examination showed thickening of the ileal mucosal folds and areas of narrowing. The terminal ileum appeared abnormal with appearances suggestive of Crohn's disease. He had a mild iron-deficiency anaemia and a slightly reduced serum albumin (3.4 g/100 ml) but no other evidence of malabsorption. The serum mucoproteins had become moderately raised (195 mg/100 ml as tyrosine). There was some improvement in his symptoms after restarting treatment with prednisone (20 mg/day). Kveim's test (while on prednisone) gave a negative result.

An attempt to improve the appearance of his lip was made by local infiltration of triamcinolone, with noticeable reduction of the swelling. At the time of writing, however, it was uncertain whether this would be of lasting benefit.

Comment

This patient's chronic granulomatous condition, evolving over a period of nine years, was easily recognizable as Crohn's disease once there was evidence of involvement of the terminal ileum. The initial presentation with cheilitis, however, was atypical. In addition to simple aphthous ulcers oral lesions having typical non-caseating granulomata on histological examination are well recognized in Crohn's disease (Dudeny and Todd, 1969; Issa, 1971; Bishop *et al.*, 1972; Eisenbud *et al.*, 1972; Stankler *et al.*, 1972; Varley, 1972). Granulomata of the skin have also been described (McCallum and Kinmont, 1968). The association between granulomatous cheilitis and Crohn's disease, however, is less common.

The characteristic histological changes were found in an excised labial fissure from one of three patients with oral Crohn's disease described by Schiller *et al.* (1971). Swelling of the lips was noted in two other patients with oral Crohn's disease but the labial histological picture was not reported in these cases (Croft and Wilkinson, 1972; Ellis and Truelove, 1972). Dudeny and Todd's original patient described in 1969 was later reported to have developed lip swelling (Verbov, 1973). Swelling of the lips in Crohn's disease could result either from direct involvement of labial tissue by granulomatous inflammation or from lymphatic obstruction. There was preliminary evidence in our patient that intralesional corticosteroid injections may be valuable in treating the disfigurement produced by this condition.

I wish to thank Dr. T. H. Boon and Dr. G. Holti for allowing me to report this case, which is under their care. Thanks are also expressed to the several pathologists at the Royal Victoria Infirmary, Newcastle upon Tyne, who examined the various biopsy specimens, and to Dr. A. P. Douglas for his advice about the patient.

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