Cheilitis Glandularis: A Case Report

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Sir,

Cheilitis glandularis (CG) is a rare disorder characterized by hyperplasia of the labial mucous glands and of heterotopic mixed salivary glands as well as by ectasia of glandular ducts (1). Patients suffering from CG can develop CG apostematosa – a severe deep-seated inflammation forming abscesses and fistulous tracts (1 – 3). CG predominantly affects males (3), but here we report on a female patient who presented with CG apostematosa.

CASE REPORT

A 37-year-old woman gave an 8-day history of herpes labialis, with clear vesicles located at the lower lip, which she had herself topically treated with an acyclovir cream. Because she did not notice any signs of recovery, she visited her dermatologist 4 days preceding hospital admission. The dermatologist prescribed a glucocorticosteroid ointment, but with this treatment she developed an increasing swelling of the lower lip. In an outpatient management, the patient was treated for acute allergic contact dermatitis intravenously with 250 mg prednisolone on 4 consecutive days and topically with gentamicin cream until the day before hospital admission. The treatment resulted in only temporary improvement.

Upon hospitalization the patient had a massive swelling and eversion of the lower lip as well as seropurulent and haemorrhagic crusts on its surface. The crusts were carefully removed with physiologic saline. Multiple interconnecting fistulous tracts were observed from which pus was expelled on slight pressure (Fig. 1). Oral mucosa was not affected and no lymphadenopathy was noticed. Laboratory investigations disclosed a slightly elevated ESR.

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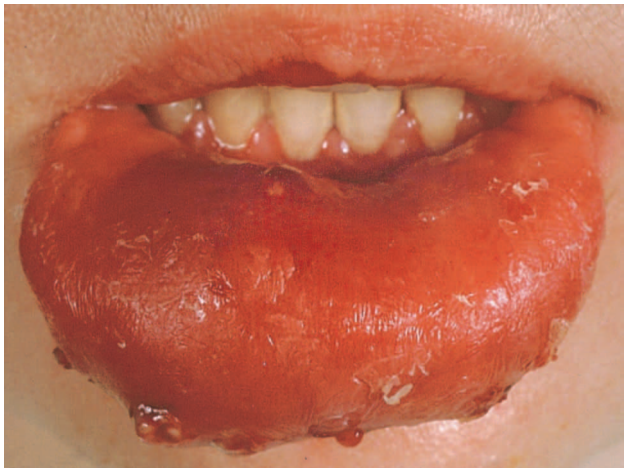

Fig. 1. Clinical findings on hospital admission: impressive swelling of the lower lip and multifocal discharge of pus.

Blood count parameters were normal, as were C-reactive protein, serum-glucose, hepatic and renal parameters, serum protein-electrophoresis, the Treponema pallidum hemagglutination test (TPHA), the HIV-screening test and serum antistreptolysin-titre. Herpes simplex serotesting revealed an elevated IgG-titre (1:3400); no specific IgM was detectable.

The Gram stain of a smear taken from the lower lip showed numerous leucocytes and staphylococci and a few epithelial cells. Staphyloccocus aureus was identified in the bacterial culture. Mycologic culture was negative. A patch test performed with the aciclovir preparation used by the patient showed positive (+) reaction at the 72-h reading according to the criteria of the ICDRG (4).

The patient was treated surgically: two longitudinal incisions were performed followed by necrosectomy and insertion of broad gauze, which was left for several days. The abundant abscessing inflammation did not permit taking an intraoperative representative biopsy for histopathologic investigation of the lip glands. Additionally, clindamycin (600 mg/8 h) was administered intravenously. The condition of the patient quickly improved, with swelling of the lower lip abating within several days.

DISCUSSION

In the literature, CG has been classified in three forms: cheilitis glandularis simplex (Puente and Acevedo), cheilitis glandularis purulenta superficialis (Baelz-Unna) and cheilitis glandularis apostematosa (von Volkmann) (1–3). CG predominantly affects middle-aged or elderly men and is usually located on the lower lip. However, children and, infrequently, women can also be affected; occurrence on the upper lip has also been reported (2, 3, 5, 6). An analogous disease pattern of the oral mucosa has been described as stomatitis glandularis (7).

CG simplex is the most common type (2). It is characterized clinically by purpuric red pinhead-sized papules of the margin of the lips and the adjacent mucosa. The papules have a central depression from which a thick, viscous fluid may be milked (1–3, 7). On palpation, the lip is irregularly indurated and, with progression, becomes enlarged, firm and everted (1, 3, 7). CG purulenta superficialis and CG apostematosa are believed to evolve from CG simplex due to bacterial superinfection (1, 3). Patients with CG purulenta superficialis initially exhibit indolent swelling and induration of the lip, crusts and ulcers (1, 2). CG apostematosa is characterized by a deep, abscess-forming inflammation and fistulous tracts (1, 2). The course of the disease is chronic and according to the literature the development of CG apostematosa from CG simplex is generally slow (1). In contrast, the patient reported here showed a rapidly progressive disease; the abscessing cheilitis developed within a few days. Presumably the glucocorticosteroid treatment of the acute contact dermatitis due to the aciclovir cream caused the rapid spreading of the staphylogenous superinfection.

In the German-speaking literature, an affection of the lower lip termed "acute ectropionizing bacterial cheilitis" has been described (8) which marks a differential diagnosis of CG. According to Bork et al. (8) this uncommon disease is caused by exacerbation of a folliculitis or furuncle affecting the adjacent skin. Acute ectropionizing bacterial cheilitis begins with an oedema of the lower lip. Then within a few hours inflammatory infiltration, eversion and crusts develop on the mucosal part of the lip. In contrast to CG, abscess-forming or phlegmonous inflammation is not observed (8).

CG has been challenged as being a separate entity. Swerlick & Cooper (9) examined 5 own cases of CG and reviewed 48 published cases. Histopathologically, the labial glands of their 5 patients with CG did not differ in size, depth or histologic appearance from those seen in 10 controls. Swerlick & Cooper (9) concluded that CG is an unusual reaction pattern in response to chronic irritation, but conceded, reviewing the literature, that few patients of CG had been reported in whom sialadenitis of the enlarged glands and prominent duct ectasia had been documented. Bork et al. (8) assumed that some cases of CG represent a cheilitis granulomatosa.

In summary, pathogenesis of CG is not yet fully understood, but bacterial superinfection, especially due to staphylococci, seems to be crucial for progression of CG simplex into the suppurative forms.

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