

Cheilitis Glandularis Apostematosa in a Female Patient – a Case Report

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Abstract

Cheilitis is an inflammatory condition of the vermilion border of the lips, which is the junction between the skin and the mucosa. Cheilitis may arise as a primary disorder of the vermilion zone; the inflammation may extend from the nearby skin, or less often from the oral mucosa. Primary cheilitis lesions are either superficial or deep. Deep types include cheilitis glandularis (inflammatory changes and lip gland swelling), and granulomatous cheilitis (chronic swelling of the lip due to granulomatous inflammation mostly of unknown origin). Cheilitis glandularis is a rare condition that mostly affects the lower lip and it is characterized by nodular enlargement, reduced mobility and lip erosion. Based on clinical presentation, cheilitis glandularis may be classified into three subtypes: simplex (described as Puente and Acevedo), superficial suppurative (described by Baelz-Unna), and the most severe type – deep suppurative, also known as cheilitis glandularis apostematosa (Volkmann's cheilitis) characterized by deep-seated inflammation forming abscesses and fistulous tracts.

This is a case report of a female patient with a deep suppurative type of cheilitis affecting both lips. Treatment with systemic antibiotics (using antibiogram tests), corticosteroids and topical therapy resulted in significant improvement.

Key words

Cheilitis + diagnosis + etiology + classification + therapy; Disease Progression; Prognosis; Treatment Outcome

Cheilitis is an inflammatory condition of the vermilion border of the lips, which is the junction between the skin and the mucosa. Cheilitis may arise as a primary disorder of the vermilion zone, the inflammation may extend from the nearby skin, or less often from the oral mucosa (1, 2). Cheilitis may represent a focal inflammatory process or a manifestation related with diseases of other systems or organs (3). The primary cheilitis lesions are either superficial or deep (4). Superficial cheilitis can be classified into: exfoliative (factitious); postmenopausal; actinic (solar); allergic (contact); eczematous; angular and abrasive precancerous (Manganotti's). Deep types of cheilitis include glandular and granulomatous C.

Glandular cheilitis (GC) is a rare condition characterized by inflammatory changes and swelling of salivary glands in the lips. It commonly affects

the lower lip with clinical manifestations of nodular enlargement, reduced mobility and lip inversion (5). It is most commonly seen in adult males (6), but it has also been described in females (7, 8), and children (9). Cases involving the upper (10) or both lips (11) have also been reported.

The etiological factors are sometimes hard to determine. Some causes or predisposing factors include bacterial infections (mostly *Staphylococcus aureus*), syphilis, actinic radiation, smoking, poor oral hygiene, compromised immune system, but also genetic transmission (autosomal dominant transmission is suggested) (11, 12, 13). Leao (14) described a case of GC in a HIV-infected patient, with an explanation that it was probably a coincidence.

Based on clinical presentation, glandular cheilitis can be classified into subtypes: simplex (described by

Puente and Acevedo) (15), superficial suppurative (described by Baelz-Unna) (16, 17), and a more severe deep suppurative type, also known as myxadenitis labialis or cheilitis glandularis apostematosa (Volkman's cheilitis) (18), characterized by deep-seated inflammation forming abscesses and fistulous tracts.

Von Volkman (18) was the first to describe cheilitis glandularis apostematosa in 1870, as a chronic suppurative inflammation of the lower lip characterized by swelling of the mucus glands and the mucopurulent discharge through the dilated ductal openings.

We report a patient with deep suppurative type of cheilitis of both lips. The treatment with systemic antibiotics (using antibiogram tests), corticosteroids and topical therapy resulted in significant improvement.

Case Report

A 61-year-old village housewife claimed that the first changes occurred on the right half of her lower lip at the age of 56 in the form of prominent redness, bumps and wetting. During the next year, the changes affected the entire lower lip. At the age of 60, the initial wetting was followed by purulent discharge, with scales and squamous lesions. She was treated by a dermatologist, a dentist and an ENT (ear, nose and throat) specialist. Various drugs were applied, mostly topically: antibiotics, antimycotics, interferon and acyclovir. The treatment provided only mild, temporary improvement.

Clinical status at first examination (the first contact with the patient)

Both lips and the vermilion border were covered with thick, adherent scales and squamous crusts; purulent hemorrhagic discharge was seen under pressure (Figure 1); lesions were painful, especially sensitive to touch, while normal functions such as speaking, eating and chewing were compromised.

Clinical status after crust removal

Both lips were enlarged, extremely erythematous, infiltrated, with erosions and superficial shallow ulcerations and fissures; the erythema and infiltration spread along the vermilion; the corners of the mouth



Figure 1. Both lips and the vermilion border are covered with thick, adherent scales and squamous crusts with purulent hemorrhagic lesions underneath

were not affected (Figure 2); the lips were of hard-elastic consistency to touch and granular in structure; extreme sensitivity caused hemorrhagic or purulent discharge; the regional lymph nodes were not enlarged; the tongue was unaffected, while the teeth were neglected and mostly missing.

Internist examination

The internist examination showed normal findings.

Laboratory tests

The relevant hematological and biochemical parameters were within physiological levels; the serologic test for syphilis and enzyme-linked immunosorbent assay



Figure 2. Both lips are enlarged, extremely erythematous, infiltrated, with erosions and superficial shallow ulcerations and fissures; erythema and infiltration spread along the vermilion; corners of the mouth not affected

(ELISA) for human immunodeficiency virus (HIV) antibodies were negative; bacteriological examination of lesion specimens showed *Staphylococcus alpha haemolyticus* and *Neiseria catharalis*.

Histopathological analysis

Probatory excision was performed 5 years earlier at the Ear, Nose and Throat Clinic in Belgrade: histological findings were consistent with inflammatory leukoplakia; the affected area showed folliculitis, and there were erosions of the vermilion lip. Repeat biopsy was rejected by the patient.

Treatment

The therapy included oral ciprofloxacin (500 mg twice a day) according to antibiogram during 10 days; 15 mg prednisone per day during 3 months; boric acid and antiseptic solutions were used to remove crusts and squamous lesions, which was followed by application of antibiotic ointments (garamycin and later chloramphenicol).

Local status after therapy

The lips were less infiltrated and erythematous without layers of crusts and squamous lesions with some erosions of the central lower lip (Figure 3); repeated antibiotic and corticosteroid therapy resulted in significant improvement (Figure 4).

Discussion

The classification of GC into three subtypes was done regarding the severity of inflammation, presence of bacterial infection and lip enlargement (5, 7, 19, 20). The simplex GC is characterized by multiple painless lesions with central depression and dilated canals, as well as mucous secretion which may occur spontaneously or under pressure. The superficial suppurative type of GC presents swelling of the lip, induration and areas of ulcerations and crusting with secretion of clear or viscous exudates from the salivary duct openings. Deep suppurative type of glandular cheilitis or cheilitis glandularis apostematosa is characterized by formation of deep abscesses and fistula tract that eventually heal by scarring. Episodes of suppurative discharge are spontaneous.

Many believe these subtypes probably represent a continuation of the same disease process, i. e., if the simple type is not treated properly, it becomes secondarily infected and progresses to the next type and then to the next (3). It is possible that the excessive salivary secretion from minor salivary glands represents an unusual response to irritation of the lip caused by other reasons, for example actinic damage or repeated licking (2). The disease progression in our patient has proven this assumption. The first symptoms were typical for GC simplex, probably caused by actinic irritation without data on hereditary



Figure 3. After treatment, lips are less infiltrated and erythematous without layers of crusts and squamous lesions with some erosions of the central lower lip



Figure 4. Repeated antibiotic and corticosteroid therapy resulted in significant improvement

burden. The subsequent bacterial infection, probably caused by poor oral hygiene, led to the development of GC apostematosa.

Based on literature data, there is a difference in the definition of the disease. There is a disagreement regarding the obligatory hyperplasia of local salivary glands. While Von Volkmann (14) described cheilitis as swelling of the mucous glands, many authors (4, 6, 8, 11) point to the hyperplasia of minor salivary glands or dilated ductal canals, and some others point to inflammation and swelling (3, 13, 20, 21). This disagreement is based on different histopathological findings: some authors (6, 7, 11) found hyperplasia of minor salivary glands, whereas others did not (3, 9, 12, 14, 21 -29). Based on histopathological findings, it prevails that hyperplasia of salivary glands in GC is not typical; chronic sclerosing sialadenitis and scarring are predominant, whereas ductal ectasia is a dominant histopathological and clinical finding (3). In general, histopathological findings of dense chronic inflammatory infiltrate are found only in more severe types of GC, while genuine hyperplasia of salivary glands or/and ductectasia are rather rare (2).

Differential diagnosis includes angioedema (no swelling between attacks), exfoliative C (persistent scaling), granulomatous C (histological changes are not always conspicuous or specific), elephantiasis nostras (3), irritant or contact cheilitis as well as plasma cell cheilitis (circumscribed, flat or elevated

patches of erythema with dense plasma cell infiltrates) (29).

The treatment of GC depends on the type; it may include systemic corticosteroids, but also extensive surgical resections (3). The reduction or elimination of predisposing factors (sun or wind exposure) is the first step in the treatment, followed by photoprotection and use of emollients (30). Apart from topical use, corticosteroids may be used as intralesional and systemic. The treatment may also include anticholinergics, antihistamines and antibiotics (3, 9, 29, 31, 32, 33). Radiation therapy and surgical procedures: cryosurgery, vermilionectomy and/or labial mucosal stripping, may be used as well (33).

After application of local antiseptic and antibiotic ointments, our patient received systemic corticosteroids and antibiotics (according to an antibiogram), which led to initial improvement. Due to some deterioration, the therapy was repeated resulting in significant improvement.

The prognosis for *quo ad sanationam* was unfavorable. Although cases of spontaneous remission (11) have been reported, the treatment outcome is uncertain. The possibility of malignant alteration should not be ignored. Patients with GC, especially those with deep suppurative type, should be followed-up due to the risk of squamous cell carcinoma (SCC) (21, 31). Nico et al. evaluated 22 patients diagnosed

with CG and reported three cases of superficially invasive carcinoma on the lower lip, out of which two were albino. This points to the adverse effects of sun exposure on the development of CG and the possibility of malignant alteration (19, 22), especially in cases of deep suppurative type of CG (30, 31). In some series, 18 – 35% of cases progressed to SCC (22). The reason for this probably lies in the higher susceptibility of the inverted lip to all risk factors for the development of SCC, rather than in GC being a premalignant condition *sui generis*. The majority of reported cases had deep suppurative type of the disease requiring surgical intervention and regular follow-up (3).

Conclusion

This is a report of a female patient with a severe type of glandular cheilitis affecting both lips, with a progressive course and good response to combined antibiotic and corticosteroid therapy.

Abbreviations

- C - cheilitis
- GC - glandular cheilitis
- HIV - human immunodeficiency virus
- ENT – ear, nose and throat
- Elisa - enzyme-linked immunosorbent assay
- SCC - squamous cell carcinoma

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Cheilitis glandularis apostematosa kod osobe ženskog pola – prikaz slučaja

Sažetak

Heilitis (*Cheilitis*) inflamatorno je oboljenje rumene zone usana (vermilion) koja se nalazi na prelazu kože u sluzokožu. Heilitisi koji nastaju kao samostalna oboljenja mogu biti površni ili duboki. Duboki su *Cheilitis glandularis* i *Cheilitis granulomatosa*. *Cheilitis glandularis* (CG) retka je bolest koja najčešće zahvata donju usnu i karakteriše je nodularno uvećanje, redukovani mobilitet i everzija usne. Kliničke varijante

su *CG simplex* (Puente and Acevedo), *CG suppurativa superficialis* (Baelz-Unna) i *CG suppurativa profunda seu CG apostematosa* (Von Volkmann).

Mi prikazujemo bolesnicu sa dubokom supurativnom formom heilitisa na obema usnama, kod koje je sistemska primena antibiotika, prema antibiogramu, i kortikosteroida, uz lokalnu terapiju, dovela do znatnog poboljšanja.

Ključne reči

Cheilitis + dijagnoza + etiologija + klasifikacija + terapija; Tok bolesti; Prognoza; Ishod lečenja