

## Case Report

# Cheilitis Glandularis of the Lower Lip

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**Abstract:** Cheilitis glandularis is an uncommon disease most commonly affecting the lower lip caused by hyperplasia of minor salivary glands with the presence of inflammatory component. UV rays have been considered the primary cause for CG. It has categorized into 3 clinical types: simple, superficial suppurative, and deep suppurative. [1]. The lack of sufficient data makes the diagnosis and treatment of the disease cumbersome. Hereby we report a case of actinic cheilitis of the lower lip and discuss its diagnostic and therapeutic aspects.

**Keywords:** Cheilitis Glandularis(CG), Actinic Cheilitis, Baelz's Disease, Lip, Salivary Glands, Malignancy, Inflammation

## 1. Introduction

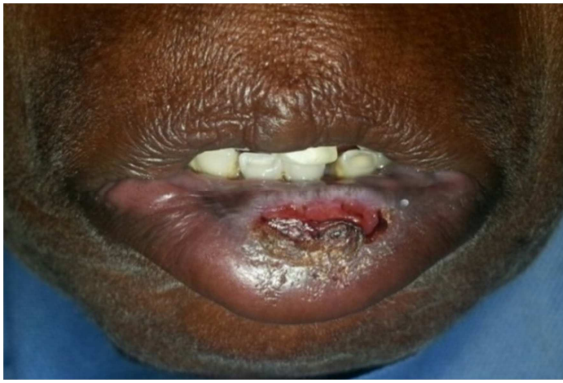
The term 'Cheilitis Glandularis' was first coined by Volkmann in 1870. Cheilitis Glandularis (CG) is a disorder that presents with a chronic, suppurative inflammation characterized by swelling of the mucous glands, dilated openings and mucopurulent discharge process that affects the lower lip in 95% of the cases. It is caused by chronic exposure to sunlight or artificial ultraviolet radiation [2].

## 2. Case Report

A 50-year-old female patient, who is a farmer by occupation reported to the department of oral medicine and radiology with the chief complaint of swelling in the lower lip since 3 years (figure 1). The patient gives a history that the swelling was initially small before 3 years which has gradually increased to the current size over the years. Patient further complains of burning sensation while having food, especially spicy food which relieves on applying cream given by the physician. There was no significant medical history or habit history elicited.



*Figure 1. Patient picture.*



**Figure 2.** Lesion of the lower lip.

On extra oral examination, a well defined solitary ulcerative lesion was seen on the lower lip at the midline measuring about 2\*1 cm. There were encrustations seen with an erythematous base with the eversion of the lower lip. Further on palpation the swelling was tender & firm in consistency (figure 2). Lymph nodes were not palpable. Intra orally, the Incisal and the labial surface of 11 and 21 appeared to be abraded and were sharp on palpation (figure 3).



**Figure 3.** Abraded labial surface of maxillary anteriors.

A provisional diagnosis of Actinic cheilitis of the lower lip was rendered.

Complete Blood Count and Random Blood Sugar were found to be within the normal range. Hence an excisional biopsy was planned and carried out. The specimen was sent for histopathological analysis.

The biopsy specimen showed parakeratotic, hyperplastic to atrophied stratified squamous epithelium showing features of

mild dysplasia. The underlying connective tissue showed sub-epithelial chronic lymphoplasmacytic infiltrate and numerous dilated capillaries. The submucosa showed mucous minor salivary glands showing atrophy of acini, ductal ectasia and chronic lymphoplasmatic infiltrate replacing the glandular parenchyma suggestive of chronic sialadenitis. A Histopathological diagnosis of cheilitis glandularis was given.

However, the lesion recurred 15 days after the excision.

### 3. Discussion

Cheilitis glandularis is an unusual inflammatory condition of the minor salivary glands, which evolves in response to chronic irritation, most commonly affecting the lower lip, but there are reports of upper lip and even palatal involvement. It causes obliteration of the mucosal-vermilion interface due to progressive enlargement and eversion of the lower labial mucosa. The chronic exposure of the delicate labial mucous membrane to external environment leads to erosion, ulceration, and crusting of the labial mucosa [2, 3].

The etiology of CG is still not understood, however it has been hypothesized as an autosomal dominant hereditary disease. UV rays have been considered the primary cause for CG. Other predisposing factors include poor oral hygiene, tobacco, bacterial infections, syphilis, possibly heredity, wind, smoking, and an immunocompromised state has also been proposed. The possibility of a genetic predisposition for cheilitis glandularis has been raised by some authors. Parmar and Muranjan, among others, described a genetic syndrome involving "double lip" of both lips in conjunction with ptosis and other physical abnormalities. In our case, chronic sun exposure and abraded sharp incisal tips seems to be the causative factor. Recent studies have shown that an alteration in the function of some of the aquaporin proteins in the minor salivary glands leads to alteration in water flow mechanism and subsequent alteration in salivary composition leading to the characteristic thick saliva [2-6].

It is more apparent in fair-skinned adults and albino patients. It has been most commonly reported in middle-aged and elderly men between the fourth and seventh decades of life; however, the age range is wide, with only a few cases reported in women and children [4, 5].

**Table 1.** Differences between Actinic Cheilitis and Cheilitis Glandularis.

	<b>Actinic Cheilitis</b>	<b>Cheilitis Glandularis</b>
Type	Premalignant lesion	Inflammatory disease
Etiology	Due to sun exposure (ultraviolet radiation)	Sun exposure, bacteria, tobacco, poor oral hygiene, hereditary factors
Age and gender	Mostly in men (M;F- 10:1), White people Below 45 years age	Mostly men, Black people, 40-70 years
Pathology	Mutations in p53 tumor suppressor genes	Not clear
Appearance	Scaly, rough, Dry areas which can be peeled off with some difficulty	Multiple painless dilated salivary ducts with thick mucous secretion
Histology	Thick keratin layer, inflammatory infiltration & perivascular inflammation	Atrophy of acini, chronic inflammatory infiltration, areas of fibrosis
Malignancy rate	20%	18-35%

Neither there is sufficient nor any reliable data, which can be attributed to the relatively small number of reported cases of CG. A history of chronic sun exposure exists, strongly indicates a need for biopsy to rule out actinic cheilitis or carcinoma [7]. The differential diagnosis of CG includes actinic cheilitis, chronic sialadenitis of the minor salivary glands. A list of differentiating features is included in table 1 between actinic cheilitis and cheilitis glandularis[3].

CG has categorized into 3 clinical types: simple, superficial suppurative, and deep suppurative.

In simple type, multiple painless lesions, dilated ductal openings and numerous small nodules that may be palpable but a lack of inflammation and the excretion of mucinous material may be appreciated. Further infection of the simple lesions may result in progression to the superficial or deep suppurative types. Superficial suppurative type is characterized by superficial ulceration, painless crusting, swelling, and induration of the lip; a mucinous exudate is apparent at the ductal openings. In the deep suppurative type, infection of the deeper tissues with abscess formation and fistulae is seen. The deep suppurative type has also been variously referred to as myxadenitis labialis or cheilitis apostematosa, and the superficial suppurative type has been termed Baelz disease. [1-10].

Further some of them have hypothesized that these subtypes possibly represent a continuation of the same disease process, wherein if the simple type is not treated properly, it might become secondarily infected and progresses to the subsequent type i.e superficial type and then further to the deep suppurative variant of CG. The probability 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 [7].

As it is self-healing, the patient does not seek treatment. However the treatment may vary depending on the severity of the condition. A malignancy rate of 18-35% has been reported [3, 9]. The conservative therapy includes the use of lip balms, sunscreens, topical steroids, intralesional steroids, systemic antihistamines, and/or antibiotics [11]. The next line of treatment would be surgical resection or vermillionectomy. The malignant potential of CG especially with deep suppurative type into squamous cell carcinoma should be always considered and never ignored [2, 5].

## 4. Conclusion

Cheilitis glandularis is a disease of unknown origin, however many etiological factors have been postulated over the years. The lack of sufficient data makes the diagnosis and treatment of the disease cumbersome. However, due to its malignancy potential it cannot be ignored, as it still remains an eminent threat. Hence, further awareness should be created about and precautionary measures should be taken in order to avoid the disease.

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