Cheilitis Glandularis: Report of a Case Affecting the Upper Lip

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Abstract: Cheilitis glandularis is a rare disorder characterized by swelling of the lip with hyperplasia of labial salivary glands, typically in the lower lip of adult males. A definitive cause and treatment for this disorder have not yet been established. Herein is reported a case of cheilitis glandularis affecting the upper lip with nodules, treated by surgical excision with good post-surgical results.

1. Introduction

The term cheilitis glandularis (CG) was first used by Volkman1 to describe the affliction of five patients with deep, suppurative inflammation of the lower lip. These patients exhibited impaired lower lip movement due to enlargement of the minor salivary glands, enlargement and ectropion of the lower lip, and mucopurulent discharge from dilated ducts. Volkman1 also observed ducts with many small red macules caused by inflammation, and extensive catarrhal inflammation of the oral and pharyngeal mucosa.

Since Volkman's initial findings, occasional cases of CG have been reported, such as those recently described by Oliver et al.,2 Stuller et al.,3 and Rada et al.4 The reported cases include symptoms of enlarged lower lip and pimple-like or punctate lesions with mucous exudation.

Cases of CG reported until now have mostly been found in the lower lip of adult males. In 1890, Unna5 reported a case in which CG appeared on the upper lip, but as such cases are rare only a few have been documented in detail.6,7

2. Case Report

1) Past history
A 63-year-old male patient presented himself at our hospital with the complaint of swelling of the upper lip. The patient had visited another hospital 6–7 years previously when he had noticed swelling of the upper lip with induration. A biopsy was done at that time. Since the result was benign the lesion was not treated. However 6 months prior to coming to us swelling of the lip worsened, prompting another visit to the hospital. After the inflammation had subsided, three salivary stone-like objects of approximately several millimeters in diameter were surgically removed. Four and a half months later the patient called on a prosthodontist at our hospital to have a denture made. Since swelling of the upper lip was still observed, he was referred to our Oral Surgery Department.

2) Clinical findings
Slight diffused swelling and erythema of the upper lip were observed (Fig. 1). Palpation revealed soft nodules and hard objects approximately several millimeters in diameter distributed over the entire upper lip. Minor salivary duct orifices on the labial mucosa were found to be papulomatous. When pressure was applied on the upper lip, a translucent mucous fluid and yellowish tinted pus were released.

No other member of the patient's family had any history of similar symptoms. There was no abnormality in general and laboratory examination and Wassermann reaction results were negative.
3) Surgical procedure
Surgical removal of nodules from the upper lip was performed. A horizontal incision was made on the labial mucosa. The nodules with labial salivary ducts were separated from submucosal tissues and removed. The excised nodules ranged from 2 to 10 mm in diameter with a soft elastic consistency (Fig. 2A). Some nodules contained hard rubbery granules (Fig. 2B). Altogether 13 such granules were found, yellowish white in color and 1 to 3 mm in diameter. The wound was closed with silk interrupted sutures and healed uneventfully. The upper lip has healed well without recurrence in 2 years of follow-up.

4) Histological findings
Histologically the removed material was found to have caused dilation of the secretory ducts...
with some ducts having a cystoid structure (Fig. 3). Interstitially, minor salivary glands were found to be infiltrated by round cells, consisting mainly of lymphocytes and plasmacytes. The lumen of the cyst-like structures consisted of light eosin-staining mucous substances and dark eosin-staining spherical nonstructured granules. These granules were thought to be the yellowish white granules extracted surgically and shown in Fig. 2B. Figure 4 is a higher-power view of the cystic space. Minor salivary glands were enlarged and in the interstitium infiltration of the round cells and some neutrophils were observed. As indicated at the upper left, the lining consisted of pseudostratified ciliated epithelium with glandular and eosinophilic cells. The left side of Fig. 5 is the histological view of a section stained with PAS. The granules shown in Fig. 2B reacted slightly to PAS, while surrounding mucous substances reacted strongly to PAS. PAS positive cells were also found sporadically among epithelial cells of the cystic wall. The histological view on the right side of Fig. 5 shows a section stained with Alcian blue, in which the granules were nonreactive, while the peripheral mucous substances were reactive to the stain. As with PAS, some cells in the cystic wall epithelium reacted positively to Alcian blue. Figure 6 is a scanning electron microphotograph of one of the granules shown in Fig. 2B. X-ray microanalysis of this sample revealed similar findings at the center and the periphery: a high peak of sulfur and no peaks of calcium and phosphorus (Fig. 7).

From these findings, this case was diagnosed as a cheilitis glandularis purulenta superficialis.
3. Discussion

The nature of CG is not clear and it is sometime confused with a labial lymphangioma or desquamative cheilitis. CG is typically characterized by hyperplasia of labial glands, enlargement of the lip with swelling, erosion and protrusion, mucous secretion by a large number of ductal orifices in the labial mucosa, dilation of ducts and cystic formation.

Schuermann\(^9\) classified cheilitis glandularis into three types by pathologic findings: (1) cheilitis glandularis simplex (simple type) which has no inflammation, (2) cheilitis glandularis purulenta superficialis (superficial suppurative type) which has chronic inflammation caused by infection, and (3) cheilitis glandularis apostematosa (deep suppurative type) which involves a deep seated infection with abscess formation and fistulous tracts. This classification is currently the most commonly accepted.

Almost all reported cases of CG have been in the lower lip of an adult. Weir et al.\(^9\) reported a case in a five-year-old boy (the youngest case yet to be reported), but CG in the upper lip is very rare. Unna\(^5\) reported the first case of CG in the upper lip in 1890 and later Winchester et al.\(^7\) reported a case affecting the upper lip. In Japan only one case of cheilitis glandularis purulenta superficialis with salivary stone has been reported previously, by Nagayama et al.\(^1\)

The histological characteristics of CG have not been uniformly defined, but Weir et al.\(^9\) summarized their findings as follows; irregular acanthosis, prominent dilation of the mucous ducts in the submucosa, hyperplasia of mucous glands, and chronic inflammation involving the stroma of the mucous glands. Doku et al.\(^11\) reported that CG manifested as hyperplasia of labial mucous glands with superimposed bacterial infection of long duration. It is commonly thought that labial duct hyperplasia, or enlargement or dilation of the ducts, is observed in CG. However, Swerlick's patho-histological study\(^6\) on a wedge-shaped sample from a resected block showed histologically that no size difference existed between CG-affected and normal labial ducts, nor were hyperplasia or hypertrophy found. In cheilitis glandularis simplex no inflammation occurs, but in cheilitis glandularis purulenta superficialis and in cheilitis glandularis apostematosa, submucosa under the epithelium and the interstitium of the ducts are thought to have definite infiltration of inflammatory cells. In the case we are reporting, dilation of secretory ducts and cystoid structures were observed, and an eosin blue-staining mucous substance was found in the cystic space. Infiltration of round cells was found in the interstitium. These features are consistent with cheilitis glandularis purulenta superficialis. Thirteen yellowish granules were enclosed in the lesions. X-ray microanalysis showed these granules to have uniform composition throughout, with a high concentration of sulfur and almost no concentration of calcium and phosphorus. These granules also reacted slightly positively to PAS and negatively to Alcian blue. From these results we deduce these granules to be composed of neutral glycoprotein. It could be speculated that the salivary stone-like substance might be formed by the deposition of calcium and phosphorus on these granules.

Although many speculations have been offered on the causes of CG, a definitive causes has not yet been established. CG has been suggested to be caused by actinic damage,\(^2,9,12\) smoking,\(^9,13\) syphilis,\(^1\) poor oral hygiene,\(^9\) gingivitis or periodontitis,\(^9\) other bacterial infection,\(^11\) and congenital conditions.\(^9,11,14\) When CG is found among family members, a hereditary atopy or Melkerson-Rosenthal syndrome are suggested as causes.\(^4\) CG may also be caused by emotional upsets.\(^15\) Some researchers\(^5,6\) explain the tendency of CG occurring in the lower lip by the greater possibility of chronic irritation, trauma and ectropion to the lower lip. Pain is not necessarily present with CG and when present, is believed to be caused by bacterial sialoadenitis and mucositis.\(^5\)

Most cases of CG are treated surgically.\(^2-4,9,11,14\) Especially, a “wedge-shaped” excision of the lesion is recommended.\(^3,4,11\) Hyperplasia of salivary glands, accelerated inflammation and spread of infection have occurred in radical procedures, causing a preference for the conservative surgical procedure. Some researchers\(^6,8,12\) believe CG to be a precarcinomatous change. Indeed, in 1962 Michalowski\(^15\) reported six cases of CG with squamous cell carcinoma and pointed out the possibility of CG degenerating into squamous cell carcinoma. Schuermann\(^9\) reported a frequency of
malignant transformation to be 12–33%. Therefore, the vermilionectomy is believed to be especially beneficial in removing possibly precarcinomatous tissue, while also being functionally and esthetically useful in simultaneously treating ectropion and extrusion of the lip.4)

In our case, we obtained good results by attention to the mucosal flap and removal of the nodules from the upper lip. Although this method is not commonly seen in other reports, we chose it because the occurrence of lip cancer in Japanese is extremely rare. This method is appropriate in that it is possible to curette a large area including ducts and cysts without much risk, and also fulfills esthetic objectives.

References

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