CASE REPORT

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Maxillary double lip and cheilitis glandularis: An unusual occurence

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Abstract

Double lip is an uncommon oral anomaly, which occurs mostly in the upper lip. It may be congenital or acquired and occurs as an isolated case or in association with other lesions. Cheilitis glandularis is a rare inflammatory disease that affects minor salivary glands and their ducts, predominantly those of the lower lip. In this article, we report a case of double lip and cheilitis glandularis in the upper lip of a 14-year-old female patient. An overview of the etiology, clinical presentation, histopathologic features and treatment are discussed.

Keywords: **Ascher's syndrome, cheilitis glandularis, double lip**

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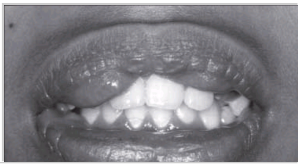

Introduction

Double lip (DL) is a rare congenital or acquired oral anomaly more often affecting the upper lip and may occur as an isolated lesion or in association with other oral anomalies. [1] It usually manifests bilaterally in the upper lip though there are earlier reports of unilateral occurrence in one or both the lips. [2] It also forms a part of Ascher's syndrome characterized by double lip, blepharochalasis and non-toxic thyroid enlargement. [2] Cheilitis glandularis (CG) is a chronic inflammatory disease of salivary acini and ducts, usually of the lower lip. [3] Adult male patients with prolonged exposure to ultraviolet radiation are more prone to this entity. [4] It can occur as a separate lesion or in association with immunosuppresion and malignancy. [3] In this article, we report the unusual occurrence of double lip and associated cheilitis glandularis in an uncommon site, the upper lip of a 14-year-old female patient.

Case Report

A 14-year-old female patient reported to Ragas Dental College and Hospital with the chief complaint of swelling of both the sides of the upper lip. The swelling on the left side was present for a period of 18 months, whereas the swelling on the right side was of six months duration. Patient gave a history of slow growth of the swellings and there was no history of associated pain. The patient had a history of lip sucking for a period of about six years and did not present with functional abnormalities of speech or mastication. On examination the swelling on the right side measured 1x1.5 cm, oblong in shape and extended 0.5 cm onto the labial mucosa and the swelling on the left side measured 1x1 cm and extended upto the labial mucosa intraorally [Figure - 1]. The swellings were of normal mucosal color and were visible only when the lips were stretched or when the patient smiled. They had a pebbly, grainy appearance with a few pinhead-sized red spots. On palpation the swellings on the upper lip were soft in consistency, mobile and fluctuant. As double lip could be associated with blepharochalasis and non-toxic thyroid enlargement, the same was ruled out by a complete clinical examination. In view of the patient's requirement of cosmetic management, a transverse elliptical incision was made and surgical stripping was done under general anesthesia.

On histopathological examination of the excised specimen, the labial mucosa revealed a parakeratinized stratified squamous epithelium in association with a fibrovascular connective tissue [Figure - 2]. The connective tissue exhibited aggregates of mucous acini with areas of degeneration and a dense infiltration of chronic inflammatory cells [Figure - 3]. Ductal proliferation with squamous metaplasia was also seen. Cystic spaces and foci of dilated ducts filled with mucin were also seen [Figure - 4]. The histology of the associated lesion was in favor of cheilitis glandularis and confirmed our final diagnosis of double lip coexisting with cheilitis glandularis in our patient.

Discussion

Double lip is a rare oral anomaly, which has also been described in association with other anomalies like bifid uvula, cleft lip, hemangiomas and cheilitis glandularis. [1],[5],[6] Double lip of both the upper and lower lips associated with hypertelorism, unilateral ptosis, blepharophimosis, broad nose with broad nasal tip, high arched palate and bilateral third finger clinodactyly has been reported in a 21 year old male patient. [7] It is also seen in patients with Ascher's syndrome along with blepharochalasis and goitre of the thyroid gland. In the present case, an ophthalmic examination and ultrasonograph of the neck did not reveal blepharochalasis and thyroid enlargement.

Double lip commonly occurs in the upper lip bilaterally, although few reports of unilateral double lip and occurrence in the lower lip have also been documented. [1],[5] It usually occurs as a redundant fold of tissue on the mucosal part of the lip and may be present either at birth or acquired. During the development of the mucosa, the upper lip consists of two transverse zones viz, an outer zone, which is smooth and similar to the skin called the pars glabrosa and the inner zone, which is villous and similar to the oral mucosa, termed as the pars villosa. [8] The DL develops during the second or third intrauterine month as a result of the persistence of the horizontal sulcus between the pars glabrosa and the pars villosa. [1],[5] In few cases, congenital DL becomes more apparent after the eruption of the teeth. The acquired form of DL is probably a manifestation resulting due to "sucking-in" of the tissue between the teeth or maloccluding dentures. [5] There is a documented report of double lip in a 30-year-old female with the history of lip sucking habit. In our case, the patient had a history of lip sucking for six years during her childhood and this habit could have most probably contributed to the formation of double lip. [2]

Differential diagnosis of double lip include hemangioma, lymphangioma, angioedema, cheilitis glandularis and cheilitis granulomatosis. [1] This condition is treated surgically with W-plasty or transverse elliptical incision to excise the hypertrophic mucosa. [5],[8] Other techniques used are triangular incision and electrosurgical excision. [2] In our patient, the excision of the redundant tissue was performed using a transverse elliptical incision.

CG is a rare chronic inflammatory condition of the minor salivary gland and ducts, usually of the lower lip. [3],[4],[9] The etiology of cheilitis glandularis is not clearly known. Various causative factors like genetic predilection, bacterial infection, poor oral hygiene, chronic irritation due to sun exposure, tobacco and chemicals have been suggested. [2],[4]

CG has been classified into three types; simple, superficial suppurative (Baelz disease) and deep suppurative [Cheilitis glandularis apostematosa]. [3],[4],[9],[10] Simple CG occurs usually in the lower lip and is clinically manifested as an enlarged everted lip with dilated openings in the lip exuding clear dew-like mucous secretions. [3],[4],[9] Purpuric pinhead-sized papules are also seen in simple CG. Bacterial superinfection can result in conversion of simple form into suppurative CG. In suppurative CG, the lip is covered with seropurulent and hemorrhagic crusts with enlarged and tender minor salivary glands. [9] Deep suppurative type is commonly seen in immunosuppressed patients with opportunistic infections. [3], [11] Chronic abscess and fistula formation can also be seen in deep suppurative CG. [9],[12]

Differential diagnoses of CG should include lymphangioma, angioedema, vascular tumours, salivary gland tumours, mucocele, Meischer syndrome, sarcoidosis, plasma cell cheilitis and cheilitis granulomatosis. [2],[3] The diagnosis of CG can be confirmed by histopathology.

The histopathologic features of CG are hyperplastic minor salivary glands with chronic sialadenitis along with dilated ducts containing mucin. In some cases, ductal ectasia, loss of acinar architecture and areas of fibrosis have also been reported. [4] Our case exhibited features of chronic sialadenitis and ductal dilatation and squamous metaplasia of the dilated ducts. Cystic spaces and foci of dilated ducts filled with mucin were also seen.

The diagnosis of CG can be confirmed by histopathology. Treatment of CG is by vermilionectomy followed by antibiotic therapy for suppurative disease. Medical management of CG includes antibiotics, steroids and antihistamine therapy. Cryosurgery and labial mucosal stripping are other surgical techniques in the treatment of CG. [4]

We would like to conclude by stating that double lip is a rare oral anomaly, which a dental practitioner could encounter. Its presence should alert the dentist to carry out appropriate investigations to rule out blepharochalasis and non-toxic thyroid enlargement, which are seen in Ascher's syndrome, which also includes the occurrence of double lip. Given the fact that Cheilitis glandularis is known to coexist with other

malignancies and immunosuppression or that the deep suppurative variant may progress to malignancy, careful evaluation and follow-up is critical in the management of these patients.

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Figures

[Figure - 1], [Figure - 2], [Figure - 3], [Figure - 4]