Case Report

Unilateral Maxillary Double Lip - A Rare Case Report

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Abstract:
Double upper lip is an infrequent oral anomaly that may be acquired or congenital. It has no gender or racial predilection. It is a deformity that interferes with speech and mastication. It may occur as an isolated case or in association with other lesions. Surgical intervention (simple excision) produces good functional and cosmetic results. In this article, we reported an unusual case of unilateral double lip in the upper lip of an 18-year-old female patient. An overview of the etiology, clinical presentation, histopathologic features and treatment are discussed.
Key words: Double lip, Ascher syndrome, Macrocheilitis.

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INTRODUCTION

Double lip 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] The current incidence of double lip is unknown, although Calnan stated in 1952 that only a dozen are cases reported in the literature. [2] Double lip is an accessory fold of redundant mucous membrane inside the vermilion border. It is caused by hyperplastic tissue of the labial mucosa that becomes more prominent with tension caused by smiling. This congenital or acquired abnormality can interfere with chewing, speaking, and esthetics. Recognition of double lip and appropriate surgical treatment can reduce these potential problems.[3] During smiling the lip is retracted and the mucosa is positioned over the maxillary teeth, resulting in a "cupid's bow" appearance whereas it is not evident when the mouth is closed.[2] In this article, we report the occurrence of unilateral double lip in the upper lip of a 18-year-old female patient.

CASE-REPORT

A 18-year-old female patient reported to Hospital with the chief complaint of unilateral swelling of left side of the upper lip and wanted to get it corrected. (Figure 1)

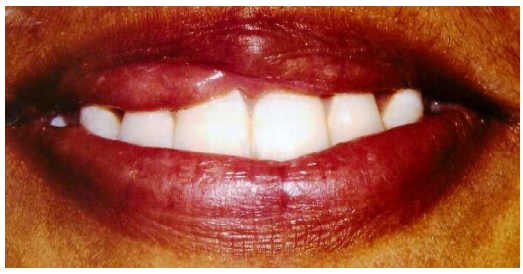

Figure 1: Clinical Photograph showing unilateral double lip in relation to right side of upper lip.

There was no relevant family history and no previous history of trauma. The swelling on the left side was present for a period of 2 years, whereas the right side is normal. Patient gave a history of gradual onset of the swelling with no associated pain. The patient had a history of lip sucking for a period of about 8-10 years. On examination the swelling of the left side measured 0.5x1 cm, oblong in shape and extended 0.7 cm onto the labial mucosa. 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. There was no blepharochalasis and no thyroid gland enlargement. A provisional diagnosis of congenital unilateral upper double lip was established, and surgical excision was suggested to the patient. Bilateral infraorbital nerve blocks were administered so as to avoid distortion of the tissue mass. Hyperplastic upper labial tissue was demarcated, marked and excised by a transverse elliptical incision from the left commissure to the midline. A light compression dressing was applied for 24 hours after the procedure. No postoperative problems were observed, and the cosmetic result was good. On histopathological examination of the excised specimen, the labial mucosa revealed non keratinized stratified squamous epithelium. Underlying connective tissue stroma separated by basement membrane showed densely packed collagen bundles with numerous blood vessels and minor salivary glands. Deep part of section also showed few adipose cells. (Figure 2)

DISCUSSION

Double lip is a rare oral anomaly of congenital or acquired origin that is equally prevalent in both genders and also shows no racial predilection.[4]

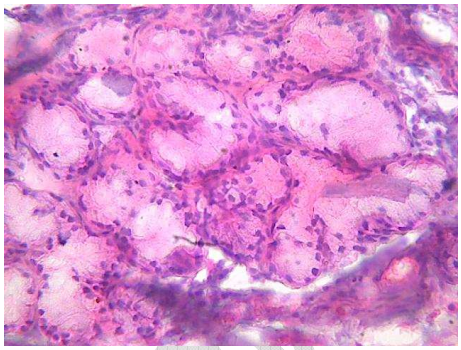

Figure 2: Photomicrograph showing saliva salivary gland acini. (H&E; 40X)

The condition consists of a fold of excess or redundant hypertrophic tissue on the mucosal side of the lip.[5] It occurs most often bilaterally on the upper lip, but may be unilateral; and can affect both the lips.[6] The condition, also referred to as macrocheilitis or hamartoma with no predilection in terms of race or sex.[7] It also forms a part of Ascher's syndrome characterized by double lip, blepharochalasis and non-toxic thyroid enlargement.[1] Adult male patients with prolonged exposure to ultraviolet radiation are more prone to this entity.[8] Since the present case did not reveal any history of thyroid disturbance or blepharochalasis we consider it as Non syndromic type of unilateral double lip.

While congenital cases stem from a developmental anomaly. During development, the upper lip mucosa is made up of two transversal zones: an outer cutaneous zone (pars glabra) and an inner mucosal zone (pars villosa). Although the enlargement of the lip may be present from birth, it can become more apparent after the eruption of the teeth. Moreover, it has been suggested that the original double lip may be enhanced by a reactive process after a "sucking-in" of the tissue between the teeth, or malocclusing dentures.[9] The acquired form of double lip may be secondary to trauma and oral habit, and may develop in

association with Ascher's syndrome which consists of the triad of blepharochalasis, nontoxic thyroid enlargement and double lip.[6,10] In the present case patient had history of oral habit of lip sucking for a period of about 8-10 years. Blepharochalasis is a condition characterized by thinning or atrophy, wrinkling and discoloration of the skin of the eyelids, and a subsequent prolapse of orbital fat and lachrymal glands and the ultimate drooping of the affected eyelid.[11]

The clinical examination of our patient revealed absence of any features suggestive of blepharochalasis. Another uncommon acquired condition is Cheilitis Glandularis, an inflammatory hyperplasia with varying degrees of inflammation of the lower labial salivary glands. The etiology of cheilitis glandularis is unknown, although familial inheritance and congenital predisposition, bacterial infection and irritation from sun, chemicals and tobacco have been observed as causes. The differential diagnosis of cheilitis glandularis and congenital double lip is important, because cheilitis glandularis has been associated with an increased risk of the development of squamous cell carcinoma.[10]

The differential diagnosis should also include vascular tumors, lymphangioma, angioedema, cheilitis granulomatosis, Meischer syndrome, mucocele, salivary gland tumours, inflammatory fibrous hyperplasia, sarcoidosis, and plasma cell cheilitis. Such lesions are frequently associated with a uniformly enlarged lip without a midline constriction dividing the lip. [12, 2]

Reported histological findings include prominent salivary glands and mixed inflammatory cell infiltration.[3] In our patient, we found minor salivary glands without inflammatory cell infiltration. Recurrence of the disease is rarely observed.[13] In the current case, no recurrence was observed over a year of follow up, the patient's prognosis remained good. Treatment is indicated when the condition interferes with speech and chewing, or for cosmetic reasons.[14] Various surgical techniques to correct a double lip have been described, but simple excision through an elliptical incision is usually recommended.[13] In our case, the double lip was corrected surgically through a transverse elliptical incision.

CONCLUSION: Albeit, Double lip is a rare entity but it should not be overlooked by dentist as it can be associated with other syndromic condition. It should be carefully differentiated from other disorders such as mucocele, cheilitis glandularis, sarcoidosis, plasma cell gingivitis etc.

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