Case Report

Acquired maxillary double lip

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Abstract

Double lip is a rare deformity of the face, which can occur as congenital and acquired anomaly. It most often appears as syndromic variety along with systemic manifestations. It is a rare occurrence with slight male predilection. This article is a report of a 20-year-old female presented with the acquired double upper lip for 1 year developed after orthodontic treatment.

Keywords: Ascher's syndrome, double lip, macrocheilia

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INTRODUCTION

Double lip is one of the rarest deformities of the lips.[1] Double lip is also called as "Macrocheilia" or hamartoma, which occurs more commonly in upper lip than lower lip and rarely both the lips are affected.[2] It consists of a fold of excess or redundant hypertrophic tissue on the mucosal side of the lip and is caused by excessive areolar tissue and noninflammatory labial mucous gland hyperplasia of the pars villosa.[3] Double lip usually manifests as two masses of hyperplastic tissue on either side of the midline. However, a unilateral double lip has also been reported. Sometimes, such bilateral hyperplastic tissues are asymmetrical, with one side being bigger than the other.[3]

Double lip can occur as congenital and acquired types. The congenital double lip is due to developmental anomaly. During the fetal period, the mucosa of the upper lip is divided into two transverse zones pars glabrosa and pars villosa. Pars glabrosa is the outer smooth zone close to the skin. Pars villosa is the inner zone similar to the mucosa of the oral cavity. Pars villosa is thought to arise during the 2nd and 3rd months of gestation. Persistence of exaggerated horizontal sulcus between the pars glabrosa and the pars villosa gives rise to the congenital type of double lip. It is commonly seen in the upper lip but may also involve the lower lip.[3-5]

CASE REPORT

A 20-year-old female patient was reported with a chief complaint of poor esthetics due to enlarged upper lip for 1 year. The patient did not mention any other significant medical history. Past dental history revealed that patient underwent orthodontic treatment 2 years back. Clinical examination revealed a mouth opening within normal limits and with no facial asymmetry. Intraoral examination reveals missing all first premolars due to orthodontic treatment. Upper lip shows prominent excess mucosal fold, and it became more prominent on smiling. There were no pain or parafunctional oral habits [Figure 1]. There were not any other associated congenital abnormalities. The patient was aware of double lip; however, she was not bothered

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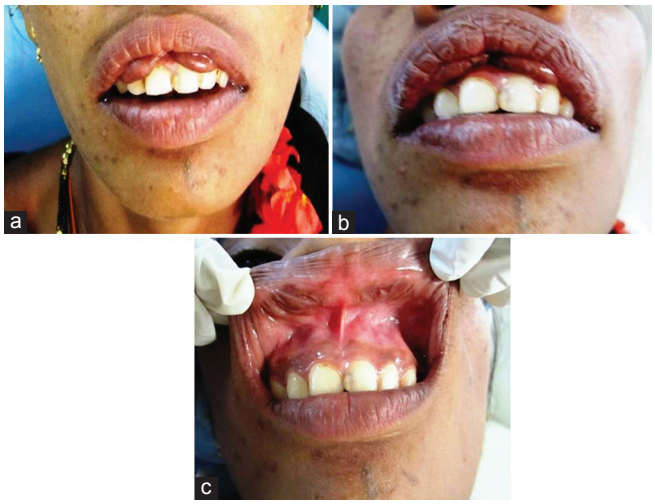

Figure 1: (a and b) Prominent mucosal folds in upper lip. (c) Mucosal fold shows notch in the midline near frenum

enough for surgical correction. A provisional diagnosis of the acquired double upper lip was made. Our patient was referred to the surgery clinic for evaluation regarding surgical correction of the upper lip.

DISCUSSION

Acquired double lip may be secondary to trauma, or oral habits such as sucking the lip between diastema, or between ill-fitting dentures.[4,6,7] Congenital double lip can occur in isolation or as a part of Ascher's syndrome which is also associated with euthyroid goiter and blepharochalasis.[8]

There are several studies mentioned about double lip. There is no race or gender predilection for the occurrence of double lip. Palma and Taub, in 2009, suggested a male predilection of 7:1. Laffer, in 1909, described double lip associated with blepharochalasis.[8]

Costa-Hanemann *et al*. described a case of double upper lip associated with hemangiomas and enlargement of thyroid gland. Congenital double lip has also been associated with bifid uvula, cheilitis glandularis, and cleft palate.[3]

The diagnosis of the double lip is commonly based on clinical findings. The differential diagnosis should include other forms of chronic enlargement of the lip such as hemangioma, lymphangioma, angioedema, cheilitis glandularis, and cheilitis granulomatosis.[9]

Hemangioma is a benign proliferation of blood vessels, and lymphangioma is due to the developmental defect of lymph channels. Both normally appear bluish to pink in color. Angioedema is the sudden diffused swelling of the lips due to allergic reactions to food and drugs. Cheilitis glandularis is due to chronic exposure to the sun, or the elements induce the inflammation of labial salivary glands. Cheilitis granulomatosa is a diffuse soft swelling, commonly seen in lower lip associated with facial paralysis.[10] These conditions, however, do not present with central constriction of the lip which is seen in acquired double lip.[9]

Histopathological examination may be done for academic purpose. Histopathological report shows hyperplastic mucous glands, loose areolar tissue, numerous blood-filled capillaries, and perivascular infiltration with plasma cells and lymphocytes.[9]

Management is aimed at both the functional and esthetic improvements. Surgery and laser are the common ways out to treat the condition.[8]

Surgery includes reduction in the height of the lip by excising the mucosal and submucosal tissues. Transverse elliptical incision and double elliptical incision combined with "Z"-plasty and "W"-plasty are the preferred incision techniques. Local anesthesia leads to tissue distortion and false exaggerated double lip appearances; hence, infraorbital nerve block is preferred.[8,11]

The choice of surgical approach will depend on preference and experience of the surgeons. Transverse elliptical incision is given in those cases without central constriction of the lip.

Double ellipse incision with Z-plasty is used in short midline constriction.[3]

Laser gives very good access to intraoperative field with minimal blood and better postoperative healing, and the procedure is less time-consuming compared to surgery.[12]

Other techniques include electrosurgical excision and triangular excision.[3]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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