

## Non-Syndromic Congenital Maxillary Double Lip: A Rare Case

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### ABSTRACT

A double lip is a rare anomaly characterized by a horizontal fold of redundant mucosal tissue that is situated proximal to the vermilion border. It may be either congenital or acquired and has no gender or race predilection. It occurs most often in the upper lip, although both upper and lower lips are occasionally involved. Surgical intervention (simple excision) produces good functional and cosmetic results. In this report, a case of a non – syndromic congenital maxillary double upper lip and a new way to treat this anomaly is described. Double lip is of special interest in dental profession as a dental surgeon is normally the first one to diagnose this rare and uncommon condition.

**Key Words:** Double upper lip, Congenital anomaly, Lip surgery

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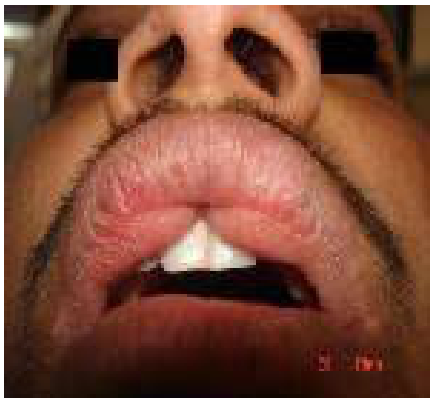
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Double upper lip is an infrequent oral anomaly that may be acquired or congenital. It has no oral or racial predilection. The current incidence of double lip is unknown, although Calnan stated in 1952 that there were only a dozen cases reported in the literature.(1) It more commonly affects the upper lip and often takes the form of 2 masses of hyperplastic tissue on either side of the midline.(2) Double lip is caused by excessive areolar tissue and non - inflammatory labial mucosal gland hyperplasia of the pars villosa.(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.(4) Histopathological examination of most of the cases in the

literature reveals squamous epithelium, hyperplastic mucous glands and no evidence of muscle tissue in the surgical specimen. Double lip may require surgical correction for esthetic reasons. Treatment should be accomplished by excision of the mucosa and sub mucosal tissues, without involvement of the underlying muscle (5).

### Case Report

A healthy 16 years old male patient presented to department of oral and maxillofacial surgery at ITS Centre for Dental Studies and Research, Murad Nagar, Ghaziabad, U.P, India, complaining of a lip defect. The lip defect he stated was present since childhood. He denied any complaints other than cosmetic



**Fig 1: Pre-Operative Intra-Oral Photo Showing Double Lip**



**Fig 2: Showing Stay Sutures**



**Fig 3: Intra-Operative Transverse Elliptical Incision**

ones. During the course of the examination, a bilateral extra fold of tissue with a midline constriction in the form of a Cupid's bow was noted in the inner surface of the upper lip (Fig 1). The overlying mucosal tissue appeared intact, smooth, with no palpable masses or surface changes. There were no other associated congenital abnormalities. The patient denied any other significant medical history.

A provisional diagnosis of congenital bilateral 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. Stay sutures were used to fix the extra redundant mucosal mass so as to define an outline on the innermost aspect of the upper lip, as the margin was ill defined there (Fig 2). The redundant mass was excised by transverse elliptical incisions (Fig 3) using a Servotome and the defect was closed in layers. A light compression dressing was applied for 24 hours after the procedure. No postoperative problems were observed, and the cosmetic result was good (Fig 4).

Histologic examination (Fig 5) of the excised material revealed sections of soft tissue covered by stratified squamous epithelium with parakeratosis. Numerous minor salivary glands with moderate lymphocytic infiltration were present in the underlying connective tissue. No muscle fibres were present in the specimen.

## Discussion

Double lip consists of a fold of excess or redundant hypertrophic tissue on the mucosal side of the lip. 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, has no predilection in terms of race or sex (7,8). The congenital form of double lip is thought to arise during the second or third month of gestation from a persistence of the sulcus between the Pars Glabrosa and the Pars Villosa of the lip. During fetal development, the upper lip mucosa consists of two transverse zones, an outer zone, which is smooth and similar to skin, the Pars Glabrosa and the inner zone, which is villous and similar to the oral mucosa, the Pars Villosa (9). The furrow dividing the double lip represents the exaggerated boundary line between the



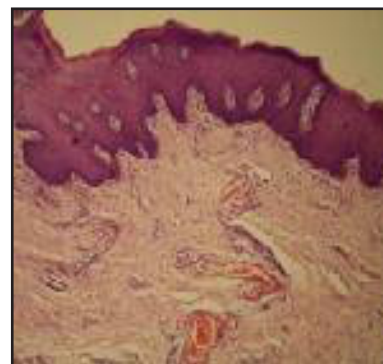
**Fig 4: Post-Operative Intra-Oral Photo Showing Corrected Double Lip**

two zones. In the double lip the buccal villous part becomes hypertrophic. In some patients, the central constriction is apparently due to the attachment of the upper frenulum. Although present at birth, the congenital condition may become apparent only after eruption of the teeth. Treatment of double lip is surgical and becomes necessary for cosmetic reasons, as in the present case, or if it interferes with speech or mastication. Recurrence has not been observed(10).

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.(6,11). The first case of double lip and blepharochalasis was reported in 1909, and the association of these findings with thyroid enlargement was noted by Ascher(3,6). It is not clear whether thyroid enlargement is a consistent or necessary feature of the syndrome. The lip becomes enlarged in a manner suggestive of angioneurotic edema and, over time, this swelling partially resolves. The association of congenital double lip with other abnormalities as bifid uvula and cleft palate has been described(12).

Another uncommon acquired condition is Cheilitis Glandularis, an inflammatory hyperplasia with varying degrees of inflammation of the lower labial salivary glands(11). 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(11). The differential diagnosis should also include vascular tumors, lymphangioma, angioedema, cheilitis granulomatosa, 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).



**Fig 5: Histopathological Picture Of The Double Lip**

Several surgical techniques have been described to repair double lip: W - plasty, electrosurgical excision, and triangular excision. In the current case good results were obtained with transverse elliptical excision using a Servotome.

In conclusion, treatment of congenital double lip is indicated when the excess tissue interferes with mastication, speech or leads to habits such as sucking or biting of the redundant tissue or is of esthetic concern to the patient.

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