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CASE REPORT

ACQUIRED NON-SYNDROMIC MAXILLARY DOUBLE LIP: A CASE REPORT

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ABSTRACT

Double lip is an uncommon facial anomaly which maybe either congenital or acquired. Most often it is non syndromic, however it is also known to be associated with syndromes involving systemic disorders. Following is a report on an acquired case of double lip which was not found to be associated with any syndrome.

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INTRODUCTION

Double lip or macrochelia is an anomaly affecting the lips. The prevalence of double lip affecting the upper lip is greater than that of the lower lip (Kenny *et al.,* 1990; Peterson, 1972). A recent report by Palma and Taub has suggested a male predilection of 7:1 (Palma and Taub, 2009). During foetal 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. Double lip is a hypertrophy of the pars villosa thought to arise during the 2nd and 3rd month of gestation. Persistence of exaggerated horizontal sulcus between the pars glabrosa and the pars villosa gives rise to the congenital type of double lip (Rintala, 1981). When the lip is tensed, pars villosa sags below the pars glabrosa, resulting in "cupid's bow" appearance (Martins *et al.,* 2004). It often takes the form of two masses of hyperplastic tissue on either side of the midline (Barnett *et al.,* 1972).

CASE REPORT

A 65 year old male from Madhikrishnapuram, Tamil Nadu, reported with a chief complaint of enlarged lip from childhood and difficulty in speech due to the enlarged lip. Patient is known diabetic and hypertensive and is under medication for the past 2 years. No relevant personal history was obtained. However, history of trauma was given by the patient during childhood. Intra oral examination showed swollen and oedematous maxillary labial mucosa with two masses of hyperplastic tissue, on either side of the midline. On palpation it was soft in consistency, non-tender and had attachments to the labial frenum. Depth of vestibule in the maxillary anterior region was 4mm; within normal limits (3.5 to 4.5mm). No midline diastema was seen and Class I molar relation was noted. Generalised attrition was observed. Extra oral examination did not reveal drooping of eyelids or any thyroid enlargement.

DISCUSSION

Diagnosis of double lip is purely clinical. Clinical features of double lip become distinct only after the eruption of the permanent teeth (Desai *et al.,* 2015). It often occurs bilaterally on the upper lip or may occur unilaterally and can affect both lips (Barnett *et al.,* 1972). It is observed that the upper double lip is not appreciated at rest but during smiling or laughing when the lip is tensed. The reason for this being the contraction of muscle orbicularis oris exaggerating the horizontal sulcus which in turn retracts the lip and places the mucosa over the maxillary teeth giving an appearance of double lip (Lamster, 1983). Double lip has been shown to be associated with some syndromes. Laffer in 1909 (Gorlin *et al.,* 1976) described double lip associated with blepherochalasis. Ascher in 1920 described a triad of double upper lip, blepherochalasis (inflammation of eyelids) and non-toxic thyroid enlargement (Ali, 2007).

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Figure 1. Patient reported with a chief complaint of enlarged lip

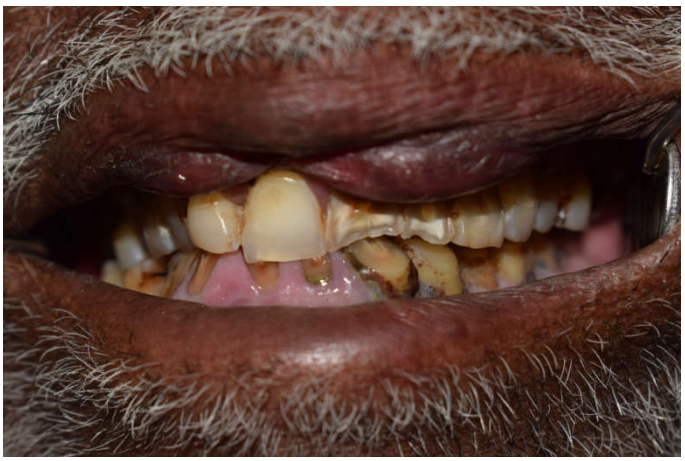

Figure 2. Examination showed two masses of hyperplastic tissue, on either side of the midline

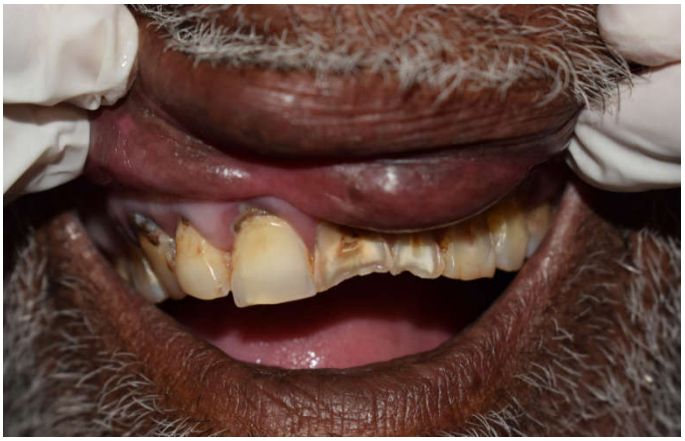

Figure 3. On smiling, contraction of orbicularis oris retracts lip giving appearance of double lip

Ascher's syndrome is an autosomal dominant disorder. The syndrome was ruled out in the above case as patient did not present with any clinical features corresponding to that of Ascher's syndrome. Congenital double lip can occur in isolation or as part of a syndrome. Paramar and Muranjan reported on a possible new syndrome compromising of double upper and lower lip, unilateral ptosis, hypertelorism, blepharochalasis, broad nose and bilateral 3rd finger clinodactyl (Parmar and Muranjan, 2003).

Costa-Hanemann et al reported double upper lip associated with hemangiomas (Costa-Hanemann *et al.,* 2004). Reports of double lip associated with bifid uvula, cheilitis glandularis and cleft palate have also been recorded (Barnett *et al.,* 1972; Cohen *et al.,* 1988; Calnan, 1952).

Conclusion

Double lip maybe congenital or acquired. Although esthetics is the main concern, function is also affected in a few cases. Surgical management provides an excellent prognosis with minimal recurrence rate (Peterson, 1972). Identification of double lip and knowledge of its association with various syndromes is vital for proper diagnosis and prevention of systemic complications, if any.

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