Case Report

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Surgical Correction of Congenital Double Lip - A Case Report

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Abstract

Lip provides an attractive and pleasing appearance to the face. Lip anomaly is characterised by the presence of a excess fold or redundant hypertrophic or hyperplastic tissue on mucosal side of the lip , which is referred as double lip. It can be either congenital or acquired and unilateral or bilateral. Such rare entity can affect either upper lip or lower lip or both as seen in Aschers syndrome which can be easily diagnosed clinically and rarely reports were published on congenital double lip. During growth and development, the upper lip possesses an outer cutaneous zone often referred as pars glabra and an inner mucosal zone which is known as pars villosa which are the two transverse zones. The non-inflammatory labial mucous gland hyperplasia of the pars villosa and unreasonable abundant areolar tissue results in double lip. Treatment should be carried out by an intraoral excision of the mucosa and sub mucosal tissue, without involvement of the underlying muscle. This article is a report of a 10 years old boy with this deformity who presented with the complaint of huge lips causing unaesthetic appearance of face and was surgically managed improving the appearance of the face aesthetically acceptable which in turn improves the confidence level of an individual.

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INTRODUCTION

Double lip might be either inborn (Congenital form) or obtained (Acquired form) and is one of the rarest types of an oral anomaly. Generally, alluded to as macrocheilia or hamartoma. Double lip is an extraordinary oral abnormality, the upper lip is more ordinarily influenced than the lower lip, and once in a while, both the lips are affected (Martins *et al.*, 2004; Palma and Taub, 2009). It is described by abundance tissue mass or embellishment overlay of excess mucosal tissue present inside the vermilion fringe. The surgical management for this congenital lip anomaly is outlined in this case report.

CASE REPORT

A 10-year-old kid reported with the presence of huge upper lip since birth, giving an unaesthetic appearance of the face, and was referred to the Department of maxillofacial surgery, Saveetha dental college for surgical correction. Additionally, trouble in speech and mastication caused by the presence of overabundant tissue mass. There was a prominent midline constriction band between the bulges of the labial mucosa due to large frenal attachment in upper labium. The patient did

not report any relevant past clinical history, family ancestry of a double lip or parafunctional oral propensities, and there was no history of injury to the region. The finding of Ascher's disorder was precluded from the diagnosis as there was no thyroid gland enlargement or blepharochalasis. On clinical examination, there was an excessive mucosal tissue bulge evident few distances from the commissure on either side. Only when the patient grinned or talked, the bulges, i.e. the excess tissue were obvious and was not noticeable at rest (Figure 1). Upper labial frenectomy was planned as the patient was additionally having large frenal attachment which was obstructing and producing discomfort during brushing. Complete blood investigation was within normal limits, and the patient was prepared for surgical treatment under general Anaesthesia. Incision marking was made, and then local anaesthetic nerve block and infiltration were given to minimise the damage of tissue mass and for hemostasis.

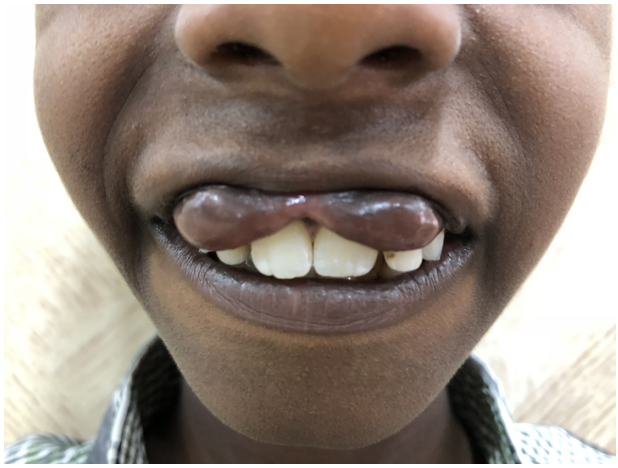

Figure 1: Double Lip Evident During Smile

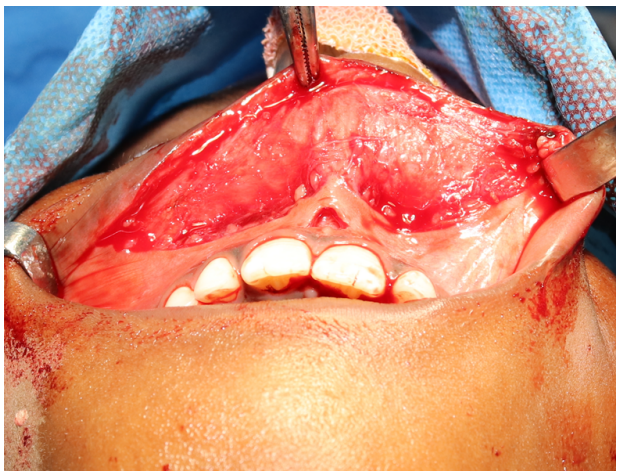

Figure 2: Surgical excision of the redundant labial mucosa

After placing an elliptical incision, surgical excision of hypertrophic tissue was done using a scalpel. (Figure 2) Demarcation of upper lip crest from the excessive mucosal bulges was done by placing free sutures using 3-0 silk, and tissue mass were held using Ellie's tissue forceps, approximately few millimetres away from the vermilion border of the upper lip. Then the tissue was stretched, creating tension so that surgical visualisation of scalpel placement is precise. After surgical excision of excessive mucosal tissue, suturing was done using 5-0 resorbable suture material. (Figure 3) After a periodic review and follow up, remaining unreserved sutures were removed, and the surgical site was debrided at around the tenth day after surgery. Promising aesthetic and functional results were obtained. Wound irrigation using 0.1% povidone-iodine had been done. Oral hygiene instructions were reinforced to the patient. Non keratinised stratified squamous epithelium with numerous collagen fibres, narrow papilla, fat cells, blood and numerous mixed minor salivary glands was revealed on histopathological examination. Few muscle fibres were also evident in the specimen. Review and recall were done at the end of 3 and 6 months and one year.

Figure 3: Suturing done using 5-0 vicryl suture

DISCUSSION

During development and advancement, the upper lip comprises of two transverse zones: an external cutaneous zone frequently alluded as standards glabra and an internal mucosal zone which is known as standards villosa. The non-provocative labial mucous organ hyperplasia of the standards villosa and unreasonable areolar tissue brings about double lip (Costa-Hanemann *et al.*, 2004). A double lip can be either inherent or obtained of which innate structure happens during the second or third month of development because of diligence of the sulcus

between the standards glabrosa and the standards villosa of the upper lip and may not be typically visible until the emission of permanent teeth (Eski *et al.*, 2007). Congenital kind of double lip influences both the sexes similarly and as a rule, happens on upper lip reciprocally. Male sexual orientation inclination of 7:1 was accounted for by Palma and Taub (2009). In upper lip is influenced, the hyperplastic tissue generally extends past the vermilion of the lip and appear as a "cupid's bow" when the upper lip is strained, and when lower lip is included, respective bulbous masses are obvious which extends intraorally. The obtained type of double lip may happen uni-along the side and ordinarily happen auxiliary to oral propensities, injury or may create in relationship with Ascher's syndrome (Alkan and Metin, 2001; Reddy and Rao, 1989; Converse *et al.*, 1978). Patients have a respectively double upper lip with large frenal attachment since birth. Double lip has additionally been accounted for in a relationship with lower double lip and different variations from the norm, for example, bifid uvula, congenital fissure, chellitis glandularis and hemangiomas (Parmar and Muranjan, 2004; Costa-Hanemann *et al.*, 2004). Buccal part appeared as two mucosal lumps on either side with a focal tightening which was a steady component for a situation arrangement of the innate double upper lip by Reddy and Rao.8 However there was a moderately focal narrowing in the current case. The mucosal lump is made out of hyper-trophied sub mucosal organs. Thus there is no side effect when the upper lip is very still, yet when grinning or talking the repetitive tissue gets apparent and shows up past the vermeil-particle outskirt of the lip (Wood and Goaz, 1997; Barnett *et al.*, 1972; Cohen *et al.*, 1988). There are no muscle strands found in the double lip, and the life systems of the orbicularis oris muscle are normal. (Converse *et al.*, 1978). A double lip (which is the consequence of hypertrophy of the standards villosa) is thought to result from an overstated flat sulcus between the standards gla-brosa and the standards villosa during the advancement of the lips (Kalkur *et al.*, 2019; Ali, 2007; Rintala, 1981). Treatment ought to be vehicle ried out by an intraoral extraction of the mucosa and submucosal tissue, without the association of the hidden muscle. Different careful strategies have been depicted to fix double lip: W-plasty, electrosurgical extraction, triangular extraction and transverse circular extraction.

Double lip is a rare anomaly of the lip, clinical separation from different sorts of incessant enlargements of lip must be made (Daniels, 2010; Parmar and Muranjan, 2004). During growth and development, the upper lip possesses an outer cutaneous zone often referred to as pars glabra and an inner mucosal zone which is known as pars villosa which are the two transverse zones. The non-inflammatory labial mucous gland hyperplasia of the pars villosa and unreasonable abundant areolar tissue results in double lip (Costa-Hanemann *et al.*, 2004). Double lip can be either inherent (congenital form) or obtained (acquired form) of which congenital form occur during the second or third month of gestation due to diligence or persistence of the sulcus between the pars glabrosa and the pars villosa of the upper lip and may not be typically noticeable until the growth of permanent teeth (Eski *et al.*, 2007). Congenital type of double lip affects both genders equally and usually occurs on upper lip bilaterally. Palma and Taub (Palma and Taub, 2009) reported male gender predilection of 7:1. In upper lip is affected, the hyperplastic tissue often projects beyond the vermilion of the lip and take the form of a "cupid's bow" when the upper lip is tensed, and when lower lip is involved, bilateral bulbous masses are evident which projects intraorally. The basic form of the double lip may occur unilaterally and usually occur secondary to oral habits, trauma or may develop in association with Ascher's syndrome (Alkan and Metin, 2001; Reddy and Rao, 1989; Converse *et al.*, 1978). Patient has a bilaterally upper double lip with large frenal attachment since birth in our case, and such entity has been reported rarely in the literature. Double lip has also additionally been reported in association with lower double lip and other abnormalities such as bifid uvula, cleft palate, chellitis glandularis and hemangiomas (Parmar and Muranjan, 2004; Costa-Hanemann *et al.*, 2004). Buccal portion appeared as two mucosal bulges on either side with a central constriction which was a constant feature in a case series of the congenital double upper lip by Reddy and Rao (1989). However there was a moderately focal narrowing in the current case. The mucosal bulge is composed of hypertrophied submucosal glands. So there is no symptom when the upper lip is at very still, but when smiling or talking the redundant excess mucosal tissue mass becomes evident and appears beyond the vermilion border of the lip (Wood and Goaz, 1997; Barnett *et al.*, 1972; Cohen *et al.*, 1988). There are no muscle fibres or filaments seen in the double lip and the anatomy of the oral musculature is normal (Converse *et al.*, 1978). A double lip (which is the result of hypertrophy or hyperplasia of the pars villosa) is thought to result from an exaggerated horizontal sulcus between the pars glabrosa and the pars villosa during the development of the lips (Kalkur *et al.*, 2019; Ali, 2007; Rintala, 1981). Treatment should be carried out

by an intraoral excision of the mucosa and submucosal tissue, without the involvement of the underlying muscle. Numerous surgical techniques like W-plasty, electrosurgical excision, triangular excision and transverse elliptical excision have been illustrated in various works of literature to repair double lip.

CONCLUSION

Double lip ought to consistently be assessed completely to preclude Ascher syndrome. The causes of acquired double lip, for instance, trauma because of different causes (past orthodontic treatments, parafunctional oral habits, ill fitting prosthesis etc) ought to be assessed and evaluated, if present, it should be addressed and treated accordingly. A psychological appraisal of the individual might be prudent and psychological guiding of the patient, before any medical procedure, is of most extreme importance for understanding the patient's desires from the surgical intervention, as also to insist on proper follow-up. The treatment of inherent double lip is necessary for aesthetic concern as in our case or when redundant tissue meddles with mastication or speech or when leads to several other parafunctional oral habits resulting in biting of the excess mucosal tissue. Surgical management of such rare entity must be accomplished by excision of the mucosa and sub mucosal tissues, without distortion of surrounding tissues and involvement of the underlying muscle fiberes.

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Conflict of Interest

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