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CASE REPORT

Congenital double upper lip: review of literature and report of a case

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Abstract

Congenital maxillary double lip is a rare anomaly in which a fold of redundant labial tissue becomes apparent at rest or while smiling. This might pose cosmetic or functional problems, as well as serious impact on psychological well-being, especially when it affects young children or adolescents. The treatment of choice is surgical excision. This paper presents a successfully managed case of maxillary double lip using elliptical incision under local anesthesia in a 15-year-old female patient.

Introduction

Double lip is a rare anomaly of the upper or lower lip, in which a fold of excess or redundant labial tissue is apparent at rest or while smiling.[1–5] This could either be congenital or acquired, affecting the upper lip more frequently than the lower with no sex predilection.[2,3,6] Some authors have described the appearance of a double lip as a ''Cupid's bow'' when it affects the upper lip.[1,6,7] When it affects the lower lip, it appear as a bilateral mass projecting intraorally.[1,8] Very rarely, both upper and lower lips can be affected.[1,9]

Double lip might be present either as an isolated condition or as a component of Ascher's syndrome, which consists of the triad of blepharochalasis, non-toxic thyroid enlargement, and double lip.[1,10,11] It has also been reported that this could present in association with other oral anomalies, such as, bifid uvula,[1] cleft palate,[12] hemangiomas,[13] and cheilitis glandularis.[9] Further, double lip can occur after an injury or as a result of habitual pulling of the mucosa through median diastima.[1] Double lip might pose cosmetic problems, as it is especially visible during smiling, leading to an unsightly appearance. This might affect the psychological well-being of a growing child. This paper describes a case of double lip affecting the upper lip that has been corrected surgically without recurrence.

Case report

A 15-year-old female patient was brought to the Oral Medicine Clinic, Dental Hospital (Teaching), Peradeniya, Sri Lanka with a complaint of a swelling in the upper lip. Unsightly appearance while smiling was the main concern of the patient and the parents. Her parents revealed that the patient covered her mouth whenever she smiled so that her abnormality could not be seen by others. They recognized this abnormality when she was approximately 7 years old, although it was present since her birth. Furthermore, the parents noted that there was a gradual enlargement of the lesion over the years. According to the patient, there were no functional disturbances, such as difficulty in mastication or speech associated with the lesion. There were no other concurrent medical problems.

Clinical examination revealed extra folds of tissue bilaterally on the inner aspects of the upper lip. This did not

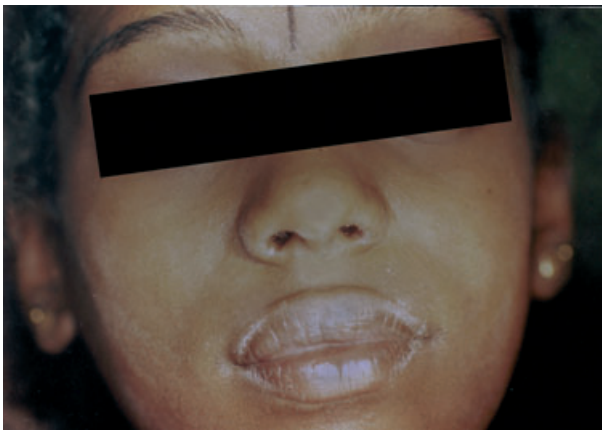

Figure 1. Lips together; no double lip deformity is visible.

Figure 2. Double lip deformity becomes visible when the patient attempts to smile.

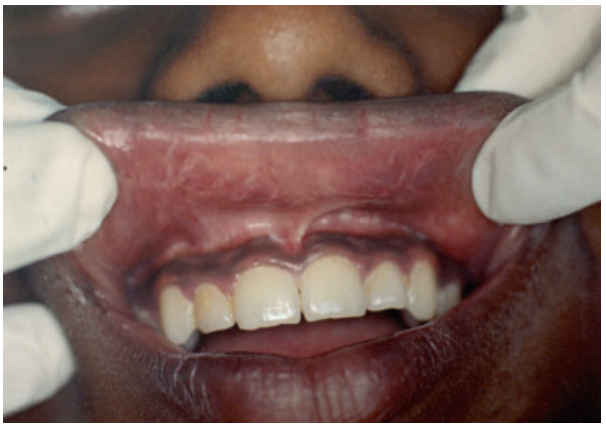

Figure 3. Three months' postoperative, without features of recurrence.

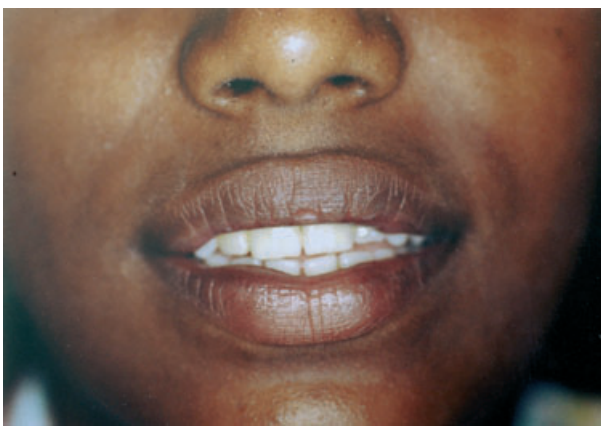

Figure 4. Nine months' postoperative, with successful results.

appear when the patient kept the lips closed (Figure 1). It was clearly visible when the patient kept the lips apart, and became more prominent while smiling (Figure 2). The overlying mucosa of these folds appeared normal. There was no tenderness or any discharge. No other extraoral or intraoral abnormalities were noted.

Based on the clinical findings, the diagnosis of congenital double lip was made. As the patient's main concern was the appearance, the excess tissue (bilateral) was excised under local anesthesia. Bilateral infraorbital block injections (2% lignocaine with 1:100 000 adrenaline) were given to anesthetize the upper lip. Elliptical incision lines were marked around the excess tissue using methyline blue. Excess tissue on both sides was removed by simple excision. The primary closure of the wound was carried out using 4/0 silk. Histological examination was not carried out, as no pathology was detected during the clinical examination and the inspection of the gross specimen. The patient was reviewed 1 week and 3 months after the operation, and no surgical complications or recurrence were noted (Figure 3). At the follow-up visit, 9 months, postoperatively, no recurrence was observed (Figure 4). The patient and her parents were very happy, as the child had no longer covered her mouth while smiling.

Discussion

The exact etiology, pathogenesis, and epidemiology of double lip are yet to be determined.[14] The congenital form of double lip is considered to develop during the 8th and 12th weeks of intrauterine life as a persistence of the sulcus between the pars glabrosa and pars villosa of the lip.[15] Although double lip might be present at birth, it becomes apparent subsequent to the eruption of teeth.[16] Constant suction by the teeth might also lead to gradual enlargement of this hyperplastic tissue.

The deformity does not appear when the patient is at rest, but becomes prominent while smiling.[17] Although there are no functional disturbances, some patients might be psychologically disturbed due to the appearance, especially, when smiling. In the present case, the parents revealed the child's attempt to cover her mouth while smiling. This might have a serious impact on the psychological development of a child.

A differential diagnosis of double lip should include hemangioma, lymphangioma, angioedema, cheilitis glandularis, cheilitis granulomatosa, and other chronic enlargements of the lips.[7,10,15,16] However, congenital double lip can be distinguished from these conditions due to its ''Cupid's bow'' appearance or midline constriction.

Ascher's syndrome is another important differential diagnosis that is characterized by the presence of double lip, blepharochalasis, and non-toxic thyroid enlargement.[15,18] However, Barnett *et al.* argued that thyroid abnormality should not be considered as an essential component in Ascher's syndrome.[1] In the present case, the possibility of Ascher's syndrome was clinically excluded based on the absence of thyroid enlargement or blepharochalasis.

Cheilitis glandularis is a very important differential diagnosis, especially considering its malignant potential. Clinicians should be careful, as the lesion is suggestive of an acquired problem. Cohen *et al.*[9] reported a case of concurrent double lip and cheilitis glandularis and emphasized the need for early treatment.

Costa-Haneman reported a case of concurrent head and neck hemangiomas and a double lip affecting the upper lip. Interestingly, this patient had slight enlargement of the thyroid gland, as well as a normal hormonal assessment.[13]

Surgical correction is the treatment of choice for double lip deformity.[9] In the present case, this was carried out purely because of cosmetic reasons. Surgical procedure can be carried out either under local or general anesthesia. Infraorbital block injection is preferable to infiltration anesthesia, as it prevents the deformation of the labial tissue that is to be excised.[2,3,16,17]

Different surgical techniques have been described in the literature to remove the redundant labial tissue. Simple elliptical incision was the most commonly-used technique described in the published reports.[2–5,10,12–17] However, some authors believed that this surgical method might lead to thinning and deformed lips, as it might cause removal of more tissue than necessary. Thus, in view of reducing such problems, W-plasty[19,20] and Z-plasty[5] have been used as alternative techniques.

Eski *et al.*[5] treated one out of five of their cases using Z-plasty. This technique was chosen to release the small midline constriction and achieved satisfactory results. Guerrero-Santos and Altamirano[19] and Benmeir *et al.*[20] used W-plasty for their patients, and highlighted the usefulness of the method in preventing deformity and avoiding possible ''dog ears''. According to the available literature, highly satisfactory results were achieved by the authors who advocated the most commonly-used, simple elliptical incisions.[2–5,10,12–17] Therefore, in the absence of literature based on comparator studies to assess the outcome of different surgical techniques, the choice of the technique will depend on the preference and the experience of the clinician.

No recurrence of double lip deformity was observed during the follow-up period after successful surgical correction.[2–5,10,12–17] However, Palma and Taub[17] reported a case of double lip in a 70-year-old female and claimed that it was a recurrence, based on the patient's history of a ''surgical procedure carried out over 30 years ago to remove some tissue from upper lip''.

There has been no well-documented, specific protocol or regimen for postoperative follow up.[17] In a recent report, Palma and Taub proposed that the patient can be discharged from follow up if no recurrence is observed for a period of 6 months. In agreement with this suggestion, the present case also showed no recurrence during the follow-up period of 9 months.

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