Congenital Maxillary Double Lip: Case Report

Konjenital Maksiller Çift Dudak

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ABSTRACT Double lip is a rare oral anomaly caused by hyperplastic tissue of the labial mucosa that becomes more outstanding with tension caused by smiling or mouth opening. A double-lip is an anomaly which may be either congenital or acquired. It occurs most often in the upper lip, although both upper and lower lips are occasionally involved. The surgical treatment options including various excision techniques may be indicated for cosmetic reasons. In the case presented, satisfactory function and aesthetic results were achieved via conventional transverse elliptical incision. Additionally, the development, clinical appearances, and the associated anomalies of this rare entity are briefly discussed.

Key Words: Maxilla; congenital abnormalities; ablation techniques

ÖZET Çift dudak, gülümseme ya da ağız açma fonksiyonu sonucunda ortaya çıkan gerilme ile birlikte daha görünür hale gelen, labial mukozanın hiperplazik dokusundan kaynaklanan ve ender olarak görülen bir anomalidir. Çift dudak, doğumsal ya da edinsel olarak ortaya çıkabilir. Daha çok üst dudakta gözlenmesine karşın, ender olarak alt ve üst dudakların her ikisinin birden etkilenmesi de söz konusudur. Kozmetik nedenlerden dolayı bu anomalinin cerrahi tedavi seçenekleri birçok farklı eksizyon tekniklerinin kullanımını gerektirebilmektedir. Çalışmamızda sunumu yapılan olguda, konvansiyonel transvers eliptik insizyon ile tatmin edici düzeyde işlevsel ve estetik sonuçlar elde edilmiştir. Ayrıca, bu nadir oluşumun gelişimi, klinik görünümü ve ilişkili olduğu anomaliler kısaca tartışılmıştır.

Anahtar Kelimeler: Maksilla; konjenital anomaliler; ablasyon teknikleri

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Double lip is a term used to describe an infrequent deformity of the upper or lower lip, which consists of an accessory fold of redundant mucous membrane inside the vermilion border.[1] It is an unusual oral anomaly that may be acquired or congenital. The upper lip is involved more often than the lower, although on occasion both may be involved.[2] Double lip is caused by excessive areolar tissue and non-inflammatory labial mucosa gland hyperplasia of the pars villosa. The condition may require surgical correction due to the aesthetic expectations of the patient and can be corrected by surgical removal of the hypertrophied glands and labial mucosa. A non-syndromic case of a maxillary double lip in a 42 ye-

ar-old man and the surgical treatment procedure of the condition are here described.

CASE REPORT

A healthy 42 year-old man was seen at the Oral and Maxillofacial Department of the Gülhane Military Medical Academy, Ankara, with the complaint of a disfigured upper lip. Unilateral extra fold of tissue was visible during physical examination (Figure 1). The overlying mucosa was intact and appeared normal. There were no other associated congenital abnormalities and no history of trauma. The patient stated that he occasionally "sucked in" the extra tissue during times of stress. A provisional diagnosis of congenital unilateral upper double lip was made, and surgical excision was suggested to the patient. Removal of the accessory lip was performed via a transverse elliptical incision under infraorbital and ring block. After removal of the accessory fold, the labial mucosa was dissected from the underlying tissues to prevent postoperative contraction (Figures 2, 3). Closure was by running 4/0 polyglactin 910 (Vycril) suture (Figure 4). Postoperative recovery was uneventful. Histological examination of the excised material revealed sections of soft tissue covered by stratified squamous epithelium. Numerous minor salivary glands, with moderate lymphocytic infiltration, were present in the underlying connective tissue. A few muscle fibers were also present in the specimen. After three years, an ideal lip occlusion is still present (Figure 5).

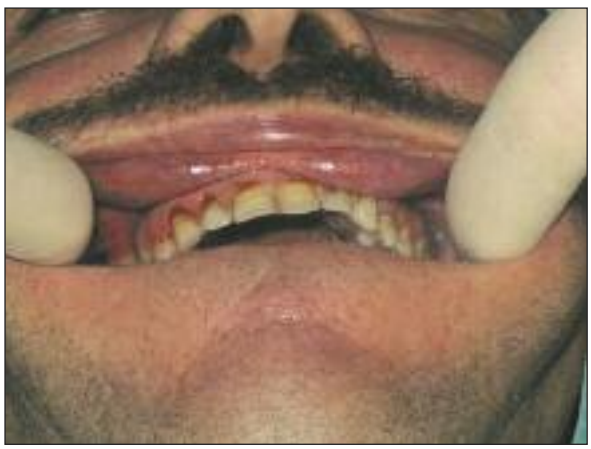

FIGURE 1: Unilateral extra fold of tissue during physical examination.

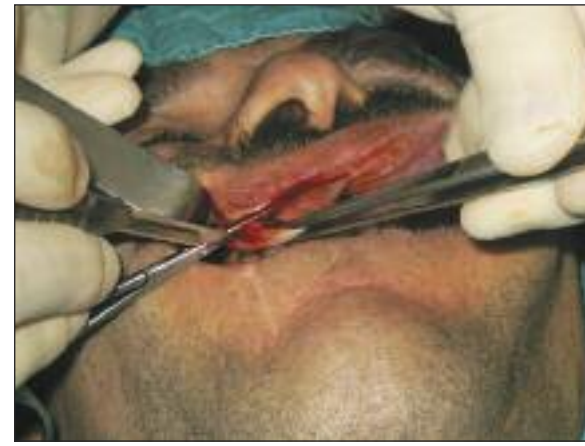

FIGURE 2: Transverse elliptical incision.

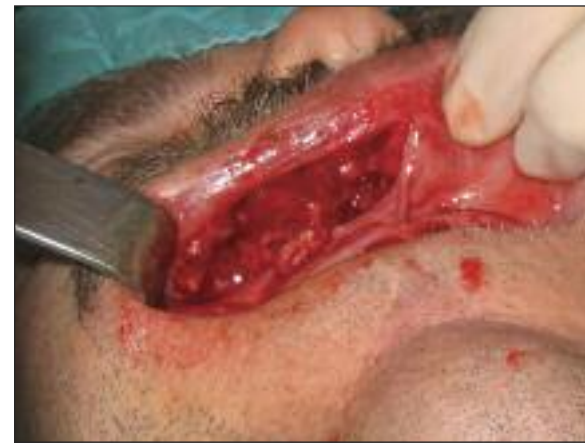

FIGURE 3: Intra-operative view.

DISCUSSION

Double lip is a rare oral anomaly of congenital or acquired origin that is equally prevalent in both genders.[3] In acquired cases, the condition usually result from trauma and may be enhanced by a reactive process after a "sucking-in" of the tissue between the teeth similar to our case.[4,5] The condition can also occur as a component of syndromes e.g. Laffer-Ascher's syndrome and inflammatory diseases e.g. cheilitis glandularis.[6]

Laffer-Ascher's syndrome is a rare disease first described in 1920 by Ascher, and characterized by a double upper lip, blepharochalasis, and nontoxic thyroid enlargement.[3,7,8] Angioneurotic edema on both eyelids as well as on the upper lip constitutes the main early clinical feature of this syndrome.[9]

FIGURE 4: Closure was by running 4/0 polyglactin suture.

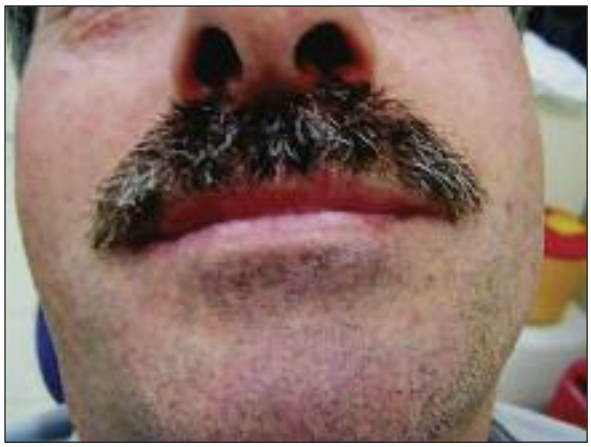

FIGURE 5: The postoperative frontal view after 3 years.

Cheilitis glandularis is a clinically descriptive diagnosis that refers to an uncommon, poorly understood, and fundamentally benign inflammatory disorder of the submucosal glands in the lower lip.[10] The condition is an inflammatory hyperplasia characterized by varying degrees of inflammation of the lower labial salivary glands. 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.[6] The presence of chelitis glandularis should be investigated for the presence of neoplasia, immunosuppression, or inflammatory diseases related to extremely poor oral hygiene.[11] In a case reported by Leao[12] and coauthors, cheilitis glandularis was the presenting clinical finding in a patient later discovered to have undiagnosed HIV-infection.

The congenital cases stem from a developmental anomaly and usually involve the upper lip, but it may also affect the lower lip. The congenital form usually presents at birth and becomes apparent with the eruption of the permanent teeth. During development, the upper lip mucosa is made up of two transversal zones: an outer skin-like cutaneous zone (pars glabra) and an inner villous mucosal zone (pars villosa).[13-15] A double lip occurs as a result of the hypertrophy of the pars villosa and derives from an exaggerated horizontal sulcus between the pars glabra and the pars villosa during the second or third month of gestation.[13]

Treatment is indicated when the condition interferes with speech and chewing, when the excess tissue interferes with leads to such habits as sucking or biting the redundant tissue or for cosmetic reasons.[14] Various surgical techniques to correct the double lip have been described: elliptical incision,[13] W-plasty,[15] triangular incision,[16] midmoon incision,[2] electrosurgical excisioni[17] elliptical incision on each side, combined with a vertical midline z-plasty[18] and laser surgery[19] to release the lip's central constriction.

Obtaining well positioned and symmetric postoperative scars is the main problem of the W-plasty technique. In comparison with the elliptical incision, triangular and mid-moon incisions are more difficult to perform. Electrosurgical incision can result in postoperative contraction due to the scar tissue formation which may hinders obtaining an ideal lip occlusion. For the treatment of the condition, an excision through a transverse elliptical incision is usually recommended.[14]

Functional disabilities and aesthetic expectations necessitate the surgical correction of the double lip. In the current case, the double lip was corrected surgically through a transverse elliptical incision. Considering the simplicity of the technique, a transverse elliptical excision is a choice of treatment.

REFERENCES

1. Greenfield MF, Icochea R, Hoffman C, Gropper C. Double lip: an unusual presentation. Cutis 2000;66(4):253-6.
2. Cortés-Aroche S. [Double lip treated with "midmoon" incision. Report of a case]. Rev Med Inst Mex Seguro Soc 2007;45(3):277-80.
3. Santos PP, Alves PM, Freitas VS, Souza LB. Double lip surgical correction in Ascher's syndrome: diagnosis and treatment of a rare condition. Clinics (Sao Paulo) 2008;63(5):709-12.
4. Hanemann JA, Oliveira DT, Gomes MF, dos Anjos MJ, Sant'ana E. Congenital double lip associated to hemangiomas: report of a case. Med Oral 2004;9(2):156-8, 155-6.
5. Alkan A, Metin M. Maxillary double lip: report of two cases. J Oral Sci 2001;43(1):69-72.
6. Cohen DM, Green JG, Diekmann SL. Concurrent anomalies: cheilitis glandularis and double lip. Report of a case. Oral Surg Oral Med Oral Pathol 1988;66(3):397-9.
7. Ali K. Ascher syndrome: a case report and review of the literature. Oral Surg Oral Med Oral Pathol 2007;103(2):26-8.
8. Halling F, Sandrock D, Merten HA, Hönig JF. [Ascher's syndrome]. Dtsch Z Mund Kiefer Gesichtschir 1991;15(6):440-4.
9. Dalmau J, Puig L, Roé E, Peramiquel L, Pimentel CL, Alomar A. Blepharochalasia and double lip: diagnosis and treatment of Ascher's syndrome. J Eur Acad Dermatol Venereol 2006;20(10):1390-1.
10. Neville BW, Damm DD, Allen CA, Bouquot JE. Salivary gland pathology. In: Neville BW, Damm DD, Allen CA, Bouquot JE, eds. Oral and Maxillofacial Pathology. 3rd ed. St Louis: Saunders Elsevier; 2009. p.462-3.
11. Carrington PR, Horn TD. Cheilitis glandularis: a clinical marker for both malignancy and/or severe inflammatory disease of the oral cavity. J Am Acad Dermatol 2006;54(2):336-7.
12. Leao JC, Ferreira AM, Martins S, Jardim ML, Barrett AW, Scully C, et al. Cheilitis glandularis: An unusual presentation in a patient with HIV infection. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003;95(2):142-4.
13. Reddy KA, Roa AK. Congenital double lip: a review of seven cases. Plast Reconstr Surg 1989;84(3):420–3.
14. Benmeir P, Weinberg A, Neuman A, Eldad A, Lusthaus S, Rotem M, et al. Congenital double lip: report of five cases and a review of the literature. Ann Plast Surg 1992;28(2):80-2.
15. Guerrero-Santos J, Altamirano JT. The use of W-plasty for the correction of double lip deformity. Plast Reconstr Surg 1967;39(5):478-81.
16. Lamster IB. Mucosal reduction for correction of a maxillary double lip. Oral Surg Oral Med Oral Pathol 1983;55(5):457–8.
17. Peterson A. Electrosurgical correction of maxillary double lip. Den Dig 1972;78(4):182-8.
18. Eski M, Nisanci M, Aktas A, Sengezer M. Congenital double lip: review of 5 cases. Br J Oral Maxillofac Surg 2007;45(1):68-70.
19. Özcanlı Ç, Başak PY. [Laser and use in dermatology]. Turkiye Klinikleri J Med Sci 2002; 22(6):620-9.