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CONGENITAL DOUBLE LIP (A REPORT OF 4 CASES)

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SUMMARY

Four cases of congenital double lip are reported along with discussion on aetiological and epidemiological aspects of this anomaly.

Congenital anomalies which commonly affect the oral commissure and lips are various types of cleft lip and congenital sinuses of the lip, but rarely, one sees abnormalities like microstomas (Basu and Pan, 1976), double mouth (Maisel, 1981), mucous fistulae (Mahler, 1975 and Holbrook, 1970) and various types of double lip. Congenital double lip occurs mostly in the upper lip but sometimes lower lip may be involved. In this deformity, a double vermilion is visible with a transverse groove between the two.

Case Reports

Among the four cases being reported, three were males and one female. Their ages were 15, 16, 18 and 20 years respectively. All the four cases got treatment for cosmetic reasons. No one had any defect in speech, other congenital deformities or family history of similar abnormality. On general physical examination no other abnormality was detected.

Local examination: Upper lip was involved in all the four cases. In all the cases, a shallow transverse sulcus was present on the mucosal surface giving the lip a look of having two vermilions (Fig. 1). Buccal portion of the vermilion was loose, redundant, soft, non-pulsatile and irreducible. Mandible, palate, nose, eyes and lower lip did not reveal any abnormality.

Operation: All the patients were operated under general anaesthesia. The redundant buccal fold was excised by making two transverse

elliptical incisions. The lip was reconstructed by careful approximation of the mucosal edges (Fig. 2). At operation thickness of the lip was found to be due to excessive submucous tissue. Muscles of the lip looked normal. On histological examination, excised tissue consisted of mucous and glandular tissues with variable amount of fibrosis and round cell infiltration. All the cases were satisfied with the treatment.

Discussion

Congenital double lip is a rare anomaly which usually involves the upper lip, but seldom the lower lip (Mahler, 1982) or both lips (Hausamen et al., 1969) may be involved. Occasionally, hemi-lip involvement has been described (Delaire, 1969). This deformity in its simple form is the commonest but rarely it may be associated with other congenital anomalies (Barnett, 1973). Rintala (1981) noticed that some times lower lip sinus syndrome is mistaken as double lip. Most of the acquired forms of double lip belong to Ascher syndrome. In the acquired variety, the swelling may not only be more wide spread and uniform but also it may recur following surgical excision.

Aetiology: In the opinion of Foman (1939) the cause of this deformity is most probably the displacement of the fibres of orbicularis oris muscle associated with pouting of hypertrophied glandular and submucous tissue. Others have suggested that muscle fibres are always normal and the double lip is due entirely to



Fig. 1. Showing complete upper double lip.

over growth of mucous glands. According to Warbrick (1952), the foetal lip during the last weeks of 1st trimester consists of two parts; the inner pars vielosa and the outer pars glabra. These two are separated by a horizontal sulcus and it is the persistence of this sulcus which results in the congenital double lip deformity. Autosomal dominant inheritance (Barnett et al., 1973) endocrine disturbances and allergic reactions (Stehr et al., 1962) have been put forward as other possible aetiological factors.

Epidemiology: Sporadic reports of one or two cases of double lip have appeared in the



Fig. 2. Showing partial upper double lip.

literature at one or the other time, but no definite epidemiological reports have appeared. According to Calman (1953), uptil 1953 only 12 cases were recorded in the literature. Reports of Gorun (1976) and Lebuisson et al. (1978) put this number at more than one hundred.

From the study of literature and our own experience, we are of the opinion that double lip is a rare congenital anomaly, and it usually involves the upper lip. Most of the cases report for treatment in teenage for cosmetic reasons and the condition is easily treatable by surgery.

REFERENCES

- 1. Asher, K. W.: Blepharochalasis mit struma und Doppelippe. Klinische Monatsblatter für Augenheilkunde and für augenarztliche. Fortbildung, 1920; 65: 86.
- 2. BARNETT, M. L., Bosshardt, L. L. and Morgan, A. F.: Double lip and double lip with blepharochalasis (Asher syndrome). Oral surgery, oral medicine and oral Pathology, 1973; 34: 727.
- 3. Basu, B. N. and Pan, M.: Mustafi human embryology. Jyoti Publishers. 9, Gokul Boral Street, Calcutta-12, 1976.
- 4. Calman, J. S.: Congenital double lip. Record of a case with a note on embryology. Brit. J. Plast. Surg., 1953; 5:197.
- 5. Delaire, J., Landais, H., Billet, J. and Lajarte, A. Y.: Les doubles Levers. Actualites odonto stomatologiques (Paris), 1969; 87: 365.
- 6. Foman, S.: Surgery of injury and plastic repair. Baltimore, Williams and Wilkins, 1939.
- 7. GORUN, R. J., PINDBORG, J. J. AND COHEN, M. M.: Syndromes of the head and neck. 2nd edition, New York, McGraw Hill Company, 1976.

- 8. Hausamen, J. E., Solbach, H. G. and Pape, H. D. Klinischer beitrag zum Ascher syndrome. Deutsche Zahnartzliche Zietschriftt (Munich), 1969; 24: 983.
- 9. Holbrook, L. A.: Congenital midline sinus of the upper lip. Brit. J. Plast. Surg., 1970; 23: 155.
- 10. Lebuisson, D., Leroy, L., Aron, J. J., Jeaneau, E. and Quillard, J.: Le syndrome de Laffer-Ascher, Journal Francis d'ophthalmologie, 1978; 12: 750.
- 11. Mahler, D. M.: Lateral congenital sinus of the upper lip. Brit. J. Plast. Surg., 1975; 28: 203.
- 12. Mahler, D. M.: Double lower lip. Ann. Plast. Surg., 1982; 8: 248.
- 13. Maisels, D. O.: Reduplication of the mouth and mandible. Brit. J. Plast. Surg., 1981; 34: 23.
- 14. RINTALA, A. E.: Congenital double lip and Ascher syndrome. Brit. J. Plast. Surg., 1981; 34: 31.
- 15. Sharma R. N. and Tara Chandra: Congenital double lip—A case report. Indian J. Plast. Surg., 1984; 17: 57.
- 16. Stehr, K., Werb, K., and Loblich, H.J.: Pathogenese and Therapie des Ascher syndrome. Deutche Medizinische Wochenschrift, 1962; 87: 1148.
- 17. WARBRICK, J. G., McIntyre, J. R. and Ferguson, A. G.: Remarks on the aetiology of congenital bilateral fistulac of the lower lip. Brit. J. Plast. Surg., 1952; 4: 254.

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