

## BILATERAL LOWER LIP SINUSES ASSOCIATED WITH BILATERAL CLEFT LIP

MASOOD H. KHAN AND ARSHAD H. KHAN

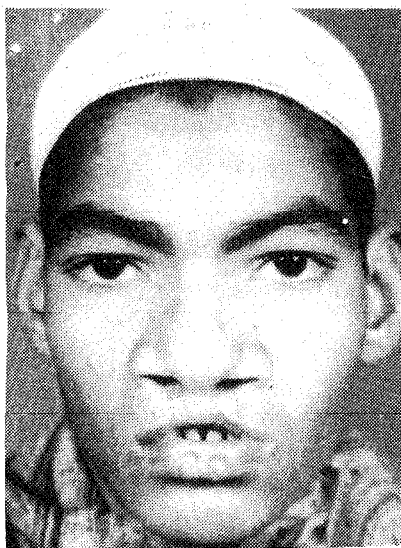
### SUMMARY

*A case of bilateral lower lip sinuses associated with bilateral cleft of the lip is being presented.*

Lower lip sinuses are among the rarest congenital anomalies recorded with an estimated incidence in the general population of 1 : 75,000—1 : 100,000 (Cervenka et al., 1967; Coccia and Bixler, 1967). Usually they are noted as a feature of the Vander Woude Syndrome (1954) and have a bilateral and symmetrical distribution about the midline of the lower lip (Rintala et al., 1970; Gorlin et al., 1976).

### Case Report

A fourteen year old male presented with a symmetrically placed pair of circular dimples on the vermilion



border of the lower lip, one on either side of the midline. He had associated bilateral cleft lip which was repaired earlier. Besides the discharge of mucous secretion from the openings, the sinuses were asymptomatic and caused no pain. There was no other associated congenital abnormality. No history of such a congenital anomaly in other siblings could be elicited. The maternal history was normal and there was no history

of teratogen exposure.

### Discussion

Congenital sinuses of the lower lip were first described by Demarquay (1845). They usually occur bilaterally and symmetrically on the vermilion border of the lower lip and extend as blind ending sacs for a variable distance into the orbicularis oris muscle (Watanabe et al., 1951; Gorlin et al., 1976). Atypically placed sinuses have also been reported and these may be unilateral, in the midline of the lower lip (Phillips, 1968), on the upper lip (Hosokawa et al., 1983; Grenman et al., 1985) or at the commissures of the mouth (Baker, 1966).

Various microforms of lower lip sinuses have also been described and these include microsinuses, and unilateral or bilateral conical elevations of the lower lip and mucosa. Careful examination may be necessary to identify these cases (Rintala and Ranta, 1981).

The etiology of congenital lip sinuses is not yet known. However, hereditary transmission has been noted. Baker (1964) reported one family of three generations with eight cases of lip sinuses. Converse et al. (1956) encountered a family in which congenital lip sinuses were present in 18 members spread over four generations. These cases represent an autosomal dominant condition which has varying degrees of expression and penetrance (Cervenka et al., 1967; Schinzel and Klausler, 1986).

The treatment of congenital lip sinuses is by surgical excision, care being taken not to disrupt the integrity and function of the orbicularis oris muscle (Watanabe et al., 1951; Wang and Macomber, 1956). Occasionally, the sinuses may contain heterotopic salivary gland tissue and so adequate excision is essential since

mucous retention cysts may occur if excision is incomplete (Ratcliffe and Milling, 1989). Other surgical procedures that have been tried include, an intraoral window (Rose, 1968) electrocoagulation (Baxter, 1939; Khanna, 1970) and vertical wedge excision (Hoppman, 1971).

#### REFERENCES

1. BAKER, B.R. : Pits of the lip commissures in Caucasoid males. *Oral Surgery, Oral Medicine, Oral Pathology*. 1966; 21 : 56.
2. CERVANKA, GORLIN, R.J. AND ANDERSON, V.E. : The syndrome of pits in the lower and cleft lip and/or palate. *Genetic considerations American Journal of Human Genetics*. 1967; 19 : 416.
3. COCCIA, C.T. AND BIXLER, D. : Cleft lip, cleft palate and congenital fistulas of the lower lip. Report of a familial occurrence. *Oral Surgery, Oral Medicine, Oral Pathology*. 1967; 24 : 246.
4. DEMARQUAY, J.N. : Quelques considerations sur le bec-de-lievre. *Gazette Medicale de Paris*. 1845; 13 : 52.
5. GORLIN, R.J., PINDBORG, J.J. AND COHEN, JR., M.M. : Syndrome of the Head and Neck. Second Edition. New York : McGraw-Hill Book Company. 1976.
6. GRENMAN, R., SALO, H. AND RINTALA, A. : Midline sinuses of the upper lip : case report. *Scandinavian Journal of Plastic and Reconstructive Surgery*. 1985; 19 : 215.
7. HOFFMAN, S. : Congenital lip sinuses; hereditary aspects and their relationships to cleft lip and palate. *British Journal of Plastic Surgery*. 1971; 24 : 241.
8. HOSOKAWA, K., SUSUKI, T., KIKUI, T.A. AND TAHARA, S. : A congenital lateral sinus in the upper lip. *Annals of Plastic Surgery*. 1983; 11 : 69.
9. KHANNA, N.W. : Congenital lip sinuses. *Australian and New Zealand J. of Surgery*. 1970; 40 : 184.
10. PHILLIPS, R.M. : Congenital fistulas of the lower lip : report of a case. *Journal of Oral Surgery*. 1968; 26 : 606.
11. RATCLIFFE, R.J. AND MILLING, A.P. : Unilateral right lower lip sinus associated with an ipsilateral cleft of the lip and palate. *British Journal of Plastic Surgery*. 1989; 42 : 92-94.
12. RINTALA, A.E., LAHTI, A.Y. AND GULLING, U.S. : Congenital sinuses of the lower lip in connection with cleft lip and palate. *Cleft Palate Journal*. 1970; 7 : 336.
13. RINTALA, A.E. AND RANTA, A. : Lower lip sinuses : 1. Epidemiology, microforms and transverse sulci. *British Journal of Plastic Surgery*. 1981; 34 : 26.
14. SCHINZEL, A. AND KLAUSLER, M. : The Van der Woude syndrome. (Dominantly inherited lip pits and clefts). *Journal of Medical Genetics*. 1986; 23 : 291.
15. VAN DER WOUDE, A. : Fistula labii inferioris congenita and its association with cleft lip and palate. *American journal of Human Genetics*. 1954; 6 : 244.
16. WANG, M.K.H. AND MACOMBER, W.B. : Congenital lip sinuses. *Plastic and Reconstructive Surgery*. 1956; 18 : 319.
17. WATANABE, Y., IGAKU-HAKUSHI, Otake, M. TOMIDA, K. : Congenital fistulas of the lower lip : report of five cases with special reference to the etiology. *Oral Surgery, Oral Medicine, Oral Pathology*. 1951; 4 : 709.

#### The Authors

MASOOD H. KHAN, M.S., M.S., *Professor and Chairman*, Deptt. of Surgery, J.N. Medical College, A.M.U., Aligarh.

ARSHAD H. KHAN, M.S., *Lecturer*, Deptt. of Gen. Surgery, J.N. Medical College, A.M.U., Aligarh.

#### Request for Reprints

PROF. M.H. KHAN, M.S., M.S., *Prof. & Chairman*, Deptt. of Surgery, J.N. Medical College, Aligarh.