

Congenital Lower Lip Fistula*

(Report of Six Cases)

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THE present report is a short description of six cases of congenital lower lip fistulae, a rare lesion estimated to occur once in 2,00,000 births. The anomaly however would not seem to be that uncommon since we have come across six cases in a brief period of a few months. It has somehow escaped reportage in the Indian literature. Philips (1968) in a review described 11 new cases and worked out that approximately 280 cases have been reported so far. Majority of the cases described have been more or less symmetrically bilateral. Only a few were unilateral sinuses. Of the latter, only four have been noted (Rintala, 1970).

Congenital lower lip fistula is a small mucosal lined tract, which opens just on one side of the midline, mounted on a nipple-like protuberance of the vermillion. Deeper down, it burrows into the orbicularis-oris muscle traversing a length of 10 to 18 mm., going slightly laterally and backwards. Most commonly this condition is bilateral, so that there are two openings, one on either side of the midline of the lower lip, about 10 mm.

apart. The two walls of the tract are close to each other and the fistula is almost collapsed. Still rarer variety of this anomaly is a median lower lip fistula, in which one tract is present going deep down into the orbicularis-oris muscle, with the external opening on the median line of the vermillion of the lower lip.

The etiology is not quite understood. It has been suggested that the sinuses are related to the abnormal persistence of the lateral sulci of the mandibular arch (Warbrick 1952). The occasional familial appearance is explained by a single autosomal dominant gene of variable expressivity (Philip 1968). These theories do not explain or even take into consideration the median fistulae, which have been attributed to a disturbance of the fusion of the primordia of the mandible (Rintala 1970).

The treatment of the condition is very simple. One would, however, wonder whether it is required at all. The inconvenience is minimal and the patient usually too poor to feel concerned. On the other hand the well

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to who would certainly seek correction.

Case Reports :

Case No. 1—A small girl of 11 years was brought to the hospital by her parents in February, 1973 for bilateral cleft lip group IA, with protruding pre-maxilla. Neither the parents nor the girl herself complained anything about the fistula. The obvious deformity and the nipple-like prominence on either side of the median line of the lower lip was quite apparent (Fig. 1). Once one is aware



Fig. 1—Photograph showing congenital lower lip fistula

of this condition it is quite impossible to miss it. If the patient is asked to purse his lips, a minute bead of mucous will show up on the opening. A probe was put in, which went down for 18 mm. The opening easily admitted the probe. The opening could also be stretched to a breadth of 8 mm. There was no history of similar or other congenital defect in the family.

Under oro-tracheal anaesthesia, a probe was passed into the fistulous opening to determine its depth and the posterior extent. It was then taken out and the fistulous tract was packed with fine gauze. An elliptical incision was made a few mm. away from the

fistulous opening and parallel to the vermilion border. The tracts were dissected out together with a circle of the mucous membrane. The nipple like prominence of the mucosa was taken into the excised tissue. The dead space was obliterated by 4/0 chromic catgut and the mucosa stitched by 6/0 atraumatic silk (Fig. 2).



Fig. 2—Photograph taken after surgery

The cut specimen showed the walls of the fistula wide open because of the formaline fixation. The lumen was reduced to a narrow slit at its depth (Fig. 3).

Histologically it is all stratified squamous epithelium continuous with the epithelium of the lower lip. The mucous glands open into the fistulous tract.

Case No. 2—A small boy of seven years age came for a similar complaint as case No. 1, i.e. a cleft lip Group IR. He had a bilateral fistula (Fig. 4). The fistulous openings were 6mm. away from the midline of the vermilion. The opening could be stretched to 8 mm. and the length of the fistula was 16 mm. on either side. No other member of the family had this defect.

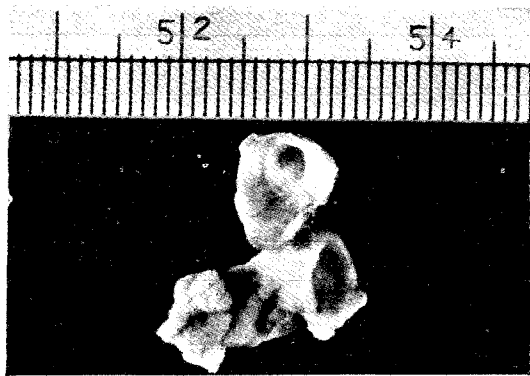


Fig. 3—Dissected specimen of the fistulous tract

Case No. 3—It was a six month old baby which was admitted for cleft lip group IL and also had a bilateral lower lip fistula. The history of similar congenital deformity in the family was missing in this case as well. The opening was 5 mm. from midline and the length was 10 mm.

Case No. 2 & 3, have had their cleft lip repaired but have not yet turned up for treatment of the fistula.



Fig. 4—Photograph showing lower lip fistulae with cleft lip Gr. I A.

Case No. 4, 5 & 6—These three cases belong to one family. The patient a boy of 12 brought to us, had a cleft lip group IA with bilateral fistula. His mother and maternal grand-mother both have similar bilateral fistula. The great grand-mother, now deceased, also had the same defect according to the grand mother. Genetic studies are being carried out in all the living members of the family.

Comments

The main interest lies in the etiology. In three cases this anomaly was an isolated incidence as far as the family was concerned, though combined with lip defect. The other three cases belonged to one family. One would tend to agree that it is a developmental defect and it may only be the failure of the obliteration of the lateral sulci of the lower lip. However, since the two anomalies i.e. congenital fistula and the lip defect are usually associated as in our four cases and also there is the familial occurrence, one would think that the theory of the genetic defect does seem to need further study. This is being explored in the familial group, result of which will be reported.

Summary

Six cases of the congenital lower lip fistula are described. Included in this is a brief summary of three cases all present in one family. The suggested factors in etiology are mentioned.

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