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A combination of unusual abnormalities of the lachrymal apparatus and nostrils in a case of bilateral cleft with choanal atresia

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Summary—An unusual congenital anomaly of a bilateral complete cleft of the primary palate, a complete cleft of the left secondary palate with abnormal nostrils without external openings and right choanal atresia is presented. The child also has a lachrymal fistula below the right lower eyelid and a coloboma of the same eyelid medially. Neither lachrymal system drained into the nose.

Case report

A female neonate born of non-consanguineous parents was seen immediately after birth, following a full-term normal pregnancy and delivery, for a deformity of the face. In addition to the complete cleft of the primary palate on both sides and a complete cleft of the secondary palate on the left side, several additional abnormalities were noticed. The right side of the nose had no external opening. Instead, a very fine opening was noticed on the dome of the deformed nose on the right side which drained serous fluid when the child strained or cried. The left nostril also was abnormal. Most of the opening of this nostril was covered by skin except for a tiny opening which did not drain any fluid (Fig. 1). The right lower eyelid had a coloboma at its medial end and below it there was a swelling which in its centre had a papery-thin area which appeared to be dried crust (Fig. 2). There was a suggestion of epiphora from the right eye. There was no other abnormality on clinical examination. There was no relevant family history or history of ingestion of any teratogenous agents during pregnancy.

On the seventh day a feeding plate made of acrylic was fitted into the palatal cleft which the child tolerated well.

Fig. 1

Fig. 2

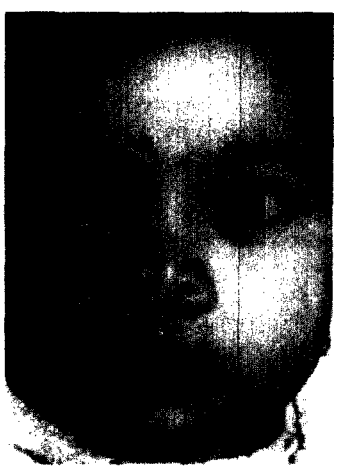

Fig. 3

Figure 1—"Worm's-eye" view of the deformity. The small opening (black arrow) in the left nasal pit. The right nasal pit is not canalised. Instead through a fine opening on the dome of the nasal pyramid a drop of fluid emerges as the child cries (white arrow). The bilateral cleft in front and a cleft of the left secondary palate behind are quite evident. Figure 2—Front view. The coloboma of the right lower eyelid and the lachrymal fistula (split arrow). Figure 3—Postoperative view. The area of the lachrymal fistula on the right side is quiescent.

Four months after birth, during which the child thrived splendidly, surgery was performed under general anaesthesia with an oral endotracheal Oxford tube.

The right lower lachrymal apparatus was first syringed through the lower lid punctum which was located lateral to the coloboma. After some resistance the fluid gushed out from the swelling of the right eyelid, displacing the papery-thin crust. Syringing on the left side was not possible. Increased pressure while syringing on the left side produced a swelling near the medial canthal area but there was no evidence that the fluid had a free passage. On withdrawing the syringe the canthal swelling collapsed and the fluid returned from the lower lid punctum.

Next, an incision was made in the skin which covered the right nostril. About 2.5 cc of white, thick, opalescent fluid gushed out. The fluid was sent for culture and antibiotic sensitivity. The interior of the nasal cavity appeared adequate in its dimensions and was covered by pink mucous membrane. A similar procedure done on the left nostril floor to increase the already existing opening did not release any fluid (as expected) because of the complete cleft of the palate on the left side. The nostril was roomy and had normal mucosa.

The bilateral lip cleft was closed in "one go" with secure muscle apposition under the philtrum (Millard, 1977). Excess vermilion was criss-crossed under the prolabium. No surgical effort was made to shape the nose. Skin was not available for banking flaps for future use (Fig. 3).

A stiff catheter introduced into the right nostril to suck out fluid failed to appear in the throat. A fine curved urethral bougie also failed to appear and was obstructed at 3 cm from the nostril. The anaesthetist was then consulted, who felt that postoperative airway obstruction was not anticipated. He was, however, categorical that any procedure done to open the right posterior choana might cause problems if bleeding appeared in the postoperative period. No surgical procedure was therefore done for the right choanal pathology.

There were no postoperative problems. Two months after surgery mild epithora still occurs from the right eye if the child cries, but the lachrymal fistula has shown quiescence and the swelling around it has reduced. The nares are being kept patent with daily dilatations by plastic spigots. The mother is adept at this, but the right nostril opening has shrunk compared to the left. The opening on the dome has not discharged since the surgery.

Discussion

The abnormality of lachrymal apparatus on both sides, the coloboma in the right lower eyelid and the choanal atresia, seen in this case, have all been described by Tessier (1976) in his type 4 south bound orbito-maxillary central cleft. There is however no other overt soft tissue abnormality in the nasomaxillary region in this case and the cleft in the lip appears to be immediately lateral to the philtral area as in a type 3 cleft (Tessier, 1976) rather than as in the type 4 cleft, more towards the angle of the mouth.

The absence of any external opening in the nose on the right side and a very small opening on the left side are the unusual features of this case. Tessier describes hypoplasia of the nose in similar cases but there appears to be no mention of a similar deformity of the nostril. The alar and nostril deformity in fact, in this case, have an uncanny resemblance to an early embryological structure, the nasal placode with its pit.

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