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Letter to the Editor

Congenital philtral fistula revealing epidermoid cyst: An unusual case

1. Case report

A 10-year-old girl attended the ENT outpatients clinic with a punctate lesion of the philtrum (Fig. 1A) observed by her parents at birth, with a history of intermittent purulent exudate. No similar cases had been observed in the family.

Clinical examination demonstrated a midline punctate orifice of the philtrum with no expulsion of liquid on pressure. Palpation did not reveal any mass or cystic formation and no abnormality was detected on intraoral examination, especially no signs of cleft palate or fistula.

Computed tomography of the face demonstrated a small linear punctate superficial low density formation in contact with the skin, with no erosion of the adjacent bone, surrounded by minor infiltration of the midline fat with no detectable collection.

Ultrasound examination of the upper lip showed minor soft tissue infiltration over the maxillary bone, but did not reveal any fistula track or collection.

In the light of these clinical and imaging findings, the patient was scheduled for surgical resection. Complete en bloc resection of the fistula was performed under general anaesthesia, via an elliptic skin incision around the fistula (Fig. 1B).

Histological examination was suggestive of epidermoid cyst, revealing a non-inflammatory cystic formation lined by epidermal type epithelium, containing a granular layer and surrounding abundant keratinized lamellar material.

Six months after the operation, the patient presented a faint scar in the middle of the philtrum with no signs of recurrence.

2. Discussion

Fistulas of the lips are rare. Fistulas of the lower lip affect 0.001% of the population, while fistulas of the upper lip are even rarer, as fewer than 30 cases have been reported since 1970 [1]; these fistulas are usually isolated [2].

When the fistula is observed in the context of a multiple malformation syndrome, it is commonly associated with midline cleft lip and palate, bifid frenulum, nasal dermoid cyst or hypertelorism [2].

Midline fistulas of the upper lip associated with epidermoid cyst are even more exceptional in this site.

The incidence of these fistulas is underestimated, because they often remain asymptomatic, making them difficult to diagnose. The main presenting complaint is an acute infectious episode [3].

Epidermoid cysts are rarely observed in the head and neck (7%), with 1.6% located in the oral cavity, while the upper lip constitutes an exceptional site [4].

The only curative treatment remains complete surgical resection of the cyst or fistula. The main complication is recurrence in the case of incomplete resection. Complete excision of the fistula can be facilitated by locating the fistula track by using a stylet or methylene blue [5].

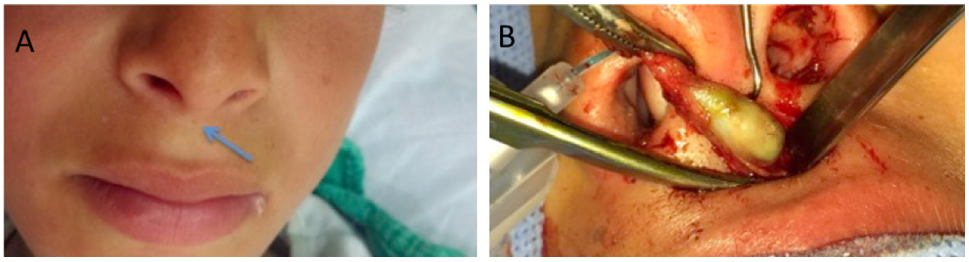

Fig. 1. A. Upper lip skin incision over the midline fistula of the philtrum – blue arrow. B. Dissection of the upper lip fistula via a skin incision.

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Disclosure of interest

The authors declare that they have no competing interest.

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