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CASE REPORT

CONGENITAL MIDLINE SINUS OF THE UPPER LIP

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Congenital sinuses and fistulae of the lip are rare, and those on the midline of the upper lip are extremely rare. We found only 8 reported cases in the literature.[1-7] In this paper a report of our case and some considerations on the etiology are presented.

CASE REPORT

A 3-year-old boy came to our clinic on February 1, 1980, requesting the removal of a midline sinus of his upper lip. He was the product of a normal pregnancy and uncomplicated delivery. There was no family history of any congenital deformities or consunguinity. He was born with a pit in his upper lip and had a history of inflammation and swelling around the sinus which subsided after drainage. The orifice of the sinus was about 1 mm in diameter, which was located on the midline at the junction of the upper and the middle third of the philtrum (Fig. 1). The general appearance of the lip and the nose were quite normal and symmetrical.

Under general anesthesia we examined the sinus before starting the surgery. A blunt probe was passed into the sinus for a distance of about 12 mm, backwards and upwards toward the anterior nasal spine. There was no communication to the mouth or nasal cavity. The sinus was then dissected out via a small elliptical incision after injecting gentian violet to detect any branching of the sinus. It extended through the orbicularis oris muscle down to the anterior nasal spine where it ended blind.

The histopathologic examination revealed that the tract was lined with stratified squamous epithelium (Fig. 2). The sinus was filled with keratin and hair, and there were sebaceous glands, hair follicles and sweat glands, adjacent to this sinus.

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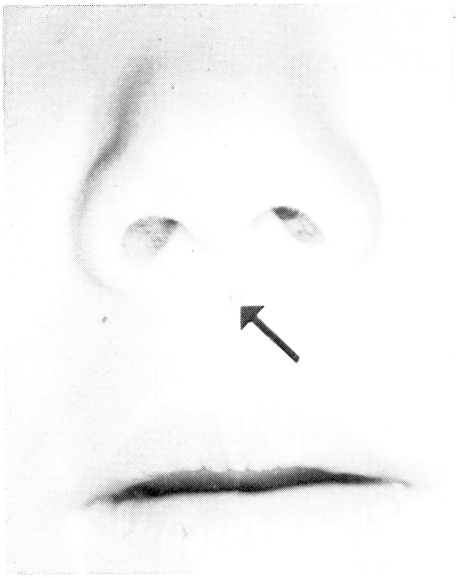

Fig. 1. Midline sinus of the upper lip.

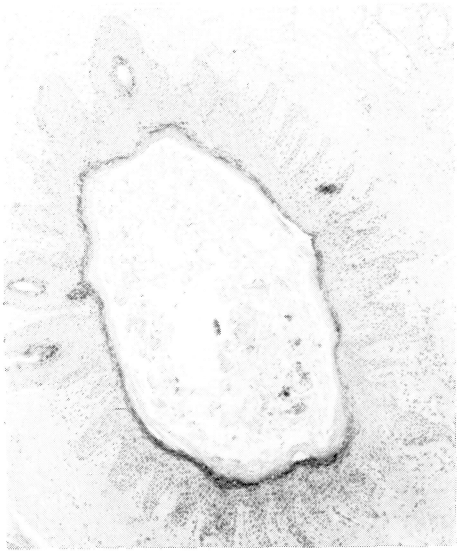

Fig. 2. Microscopic section of the sinus, lined with stratified squamous epithelium and containing keratin material (H&E, 10×.)

DISCUSSION

The gross findings in all the 8 previously reported cases were similar. Seven cases had an orifice in the center of the philtrum, and only Bartels'[6] case had an opening in the vermilion. In all the 8 cases, the sinus was blind and ended near the nasal spine, as our case. It is interesting that the Holbrook's[5] case had an opening 2 mm in diameter in the center of the philtrum and surrounded by a broader ring of thin red epithelium, similar to the vermilion of the lip, but not confluent with it.

Microscopically, the sinuses were lined with squamous epithelium, and hair follicles and sebaceous glands were observed in six cases. In Holbrook's[5] case, the tract was closely associated with a structure of hyaline cartilage, and in Miller's[7] case no epidermal appendages or glands were found.

In 6 cases, there were no associated abnormalities. Mackenzie's[4] case had a cyst and fistula on the dorsum of the nose. Bartels'[6] case had hypertelorism.

Some knowledge about the embryonic development of the upper lip is required to have a discussion about this congenital lesion. The embryonic development of the face takes place between the fourth and eighth weeks of gestation. The nasal placodes arise from either side of the frontonasal prominence just above the stomodeum. The limbs of the nasal placode become the medial and lateral nasal processes. The medial nasal processes coalesce in the midline during the sixth week, forming globular process. Globular process in turn forms the tip of the nose, the columella, the philtrum, the frenulum and the entire primary palate.

There are two leading theories about the formation of the upper lip. The first is the classic fusion theory proposed by Dursy[8] and His[9], and the second is the mesodermal migration theory represented by Stark[10]. The classic theory persists the pre-existence of clefts of the lip and Stark's challenge is the view that clefts are due to arrested development, for the etiology of the cleft lip. The mesodermal migration theory is appreciated generally for it can explain not only the embryological mechanism of the cleft lip but also that of rare craniofacial clefts.

But as Stark[11] himself said, the concept of His' seems to be correct as far as the central portion of the face is concerned. The existence of the congenital midline sinus of the upper lip could support the idea that the fusion really occurs in the midline.

There are several another explanations about the etiology of the congenital midline sinus of the upper lip. Holbrook[5] presented a hypothesis to explain the cartilagenous structure observed in his case, that the lesion arose as an exaggerated deepening of the philtral depression which occurs naturally as a

mechanism of forming philtrum in the eighteenth week of the fetal development.

Miller[7] presented the epidermal invagination theory in which the sinus is thought to be formed by the similar mechanism that naturally forms the nasal cavities.

Taking the variety of the location of the sinuses into consideration, we think the most plausible etiology for the midline sinus of the upper lip is that the sinuses are the remnants of the fusion of the medial processes.

SUMMARY

An extremely rare case of congenital midline sinus of the upper lip is presented and the theories of its etiology are discussed.

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