Columellar Sinus: A Rare Congenital Isolated Sinus

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Abstract:
Midline congenital malformation of the nose is a very rare presentation and it’s midline situation seems curious and is very difficult to explain on the basis of the present day conception of the embryology of the nose. The prevalence of lower lip sinuses has been estimated to be about 0.001% of the general population. Upper lip sinuses are even more uncommon. We herein report a case of a 13 years old girl having congenital sinus affecting the upper part of the columella.

Key words: congenital sinus, midline sinus, columellar

Introduction:
Isolated upper lip or columellar sinuses are rare epigenetic events1. They usually present with history of recurrent infection and discharge with no communication with the oral or nasal cavities1. This epithelial anomaly has been reported to occur in both the upper and lower lips, either in isolation or in association with congenital deformities such as a cleft lip and palate.

There are several hypotheses on the etiology of this condition: Ectodermal invagination of the nasal placodes in the frontonasal process (invagination theory)2; Breakdown of the mesenchyme-poor, fused maxillary processes (merging theory)3; Failure of complete fusion of the maxillary processes growing together over the frontonasal process (fusion theory) and failure of involution of supernumery nasal placodes. We further intent to discuss the theories of origin by literature review.

Case report:
A 13 years old girl presented with a pit in the nasal columella since birth. She had recurrent swelling in the floor of the nose and intermittent discharge from the orifice. The location of the sinus was at the base of the columella (fig.1). On probing, length of the sinus was about 2 centimeter and did not communicate either with oral or nasal cavities. No other congenital abnormalities were located in faciomaxillary or cervical region. She appeared well with no other significant medical history and none of her family members had similar symptoms, cleft lips, cleft palletes or lip pits. Sinogram was done (fig 2) which showed a complete delineation of a midline sinus. Based on the clinical and radiological findings a diagnosis of isolated congenital midline upper lip sinus was made. The patient was referred to an ENT specialist for surgical treatment.

Fig-1: Sinus at the base of the columella.
Fig-2: Sinogram showing midline upper lip sinus.
Discussion:

Most sinuses around the mouth are situated in the lower lip and oral commissures. Very few cases have been reported in the midline area of the upper lip\(^4\). Distribution of the midline sinuses were studied. The most common location is the philtrum, followed by the frenulum and a few on the vermilion\(^4\). The prevalence of lower lip sinus has been estimated to be 0.001% in the general population. A review of the English literature revealed no predilection for either sex\(^5\). The main problems associated with this condition are appearance and occasional mucoid discharge; often it is symptomless and is discovered accidentally. In our case intermittent discharge was the main symptom. Etiology concerning sinuses of the upper lip is obscure, but several hypothesis have been proposed. The most accepted explanation is based on the clas-sic fusion theory of the development of the face advanced by Ducty and His\(^6\). A sinus of the upper lip is considered to be formed by the failure of complete fusion of the maxillary processes similar to the formation of other clefts on the face. The case of the lateral fistula that had the vermilion island may support this hypothesis\(^7\). However, the frequency of the median cleft lip is much lower than that of the lateral cleft, whereas a midline sinus of the upper lip is more frequent than a lateral sinus. Furthermore, all of the reported cases of the upper lip sinus including our case had no communication with the oral cavity. If the upper lip sinus is formed by the failure of the fusion, it would be possible for the sinus to go through the lip. Therefore, this hypothesis does not appear to clearly account for the formation of the midline sinuses of the upper lip\(^4\).

Alternative concepts regarding the mechanism of midfacial development have been proposed by Veau and by Meurer and Hoppe\(^6\), which is generally named the merging theory. In this theory, the aggregation of mesodermal cells gives rise to the swellings and furrows; the differential accumulation of these mesodermal cells causes the obliteration of the furrows resulting in the emergence of characteristic facial features\(^8\). One author asserted that a paucity of the mesodermal cellular layer would produce facial clefts\(^9\). The midline deformities of the upper lip can be explained by aberrations in the mesodermal merging process. Embryonic nasal pits are formed by a process of epithelial burrowing and invagination, as proposed by author\(^9\). Since this process of invagination occurs to form the nasal cavities, it can be postulated that this mechanism forms the midline upper lip sinus\(^9\).

The treatment of MULS, which show recurrent inflammation, pus discharge or poor esthetics, involves surgical excision of the sinus tract. The asymptomatic patients can be managed conservatively.

Conclusion:

The midline upper lip sinus (MULS) are rare congenital events. They may exist separately or associated with other malformation like bifid anterior nasal spine and a dermoid. We cannot draw any conclusion about the etiology but this report will help for further understanding of the etiology of upper lip sinuses. These sinuses should be addressed surgically to prevent recurrent infections.

References:


