Congenital Midline Upper Lip Sinus

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The congenital upper lip sinuses are rare and they have previously been reported only in 40 cases. We have presented a case report of congenital midline upper lip sinus in an elderly age. Although the treatment modality is very easy and successful in any cases, the etiology of this rare congenital situation has been obscure and further studies should be done to find out.

Key Words: Columella, sinus, congenital

Although congenital lower lip sinuses were reported in about 0.001% of the population, the congenital upper lip sinuses are rare and they have previously been reported only in 40 cases.1 The first reported midline sinuses of the upper lip were reported by Lannelongue and Menard in 1891 and Clavet in 1899.2 The midline upper lip sinuses penetrate into the orbicularis oris muscle and end just beneath the mucosal surface, without connection to the oral cavity. There have been some etiologies postulated but the exact reason is still unclear.3 We have presented a case report of congenital midline upper lip sinus in an elderly age.

CLINICAL REPORT

A 47-year-old female with an upper lip fistula at the columellarabial junction was consulted in the outpatient clinic with the complaint of recurrent swelling, pain, discharge and erythema. Patient stated that it was present since her childhood and causing recurrent infections. On physical examination a 1 mm sinus orifice was detected at the junction of philtrum and columella (Figure 1) which was indurated, discharging and edematous. She had no other congenital anomaly or special medical history. The patient was applied systemic antibiotics for 15 days to control the infection and then the patient was operated. The sinus tract was cannulated and found out to extend down to the frenulum through orbicularis oris muscle without any connection to the oral cavity ending beneath the oral mucosa (Fig 2). The sinus tract and the orifice were removed. There were no complications in the follow-up period of 12 months (Fig 3).

DISCUSSION

Congenital lower lip sinuses have been reported in about 0.001% of the population, although, upper lip sinuses are much more rare. At present, there are 40 cases in the literature about congenital upper lip sinus. Among them, 26 sinuses arose at the midline and 14 at paramedian including three cases of bilateral pits.1,4,8 The female predilection especially in the midline sinuses were reported and half of the cases was associated with congenital defects such as upper cleft lip, lower lip pits, fistulae of the dorsum of nose and anomaly of lingual frenulum.3
Grenman et al.² had presented a case report of congenital upper lip sinus with Pierre Robin syndrome and Sumitomo et al. had indicated a case report of congenital sinus of the upper lip with idiopathic precocious puberty³ resulting in that the congenital upper lip sinuses were related with developmental disturbances. The histopathology of the sinuses revealed the squamous epithelial lining and occasionally, salivary glands, mucous glands, sebaceous gland as well as hair follicles. The female patient presented here had no associated congenital malformations or diseases and she had not been treated until the age of 47.

The etiology of this congenital malformation was presented to be inadequate fusion of facial processes, aberrations in the normal mesodermal merging process or abnormal epithelial invagination.¹⁻⁹ Al-Qattan³ proposed the invagination theory, because there were no reports of upper lip sinus associated with oral or nasal cavity that was also the same situation in the patient presented here. Although the treatment modality is very easy and successful in any cases,¹⁰⁻¹² the etiology of this rare congenital situation has been obscure and further studies should be done to find out.

REFERENCES