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청소년기에 발견된 상구순 누공의 치험례

정한주 · 강석주 · 김진우 · 선 욱
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Congenital Upper Lip Sinus Found in Adolescent Patient: A Case Report

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Purpose: Congenital sinus of the upper lip is extremely rare and only 3 cases have been reported domestically. We report a case of congenital sinus of midline upper lip, which was found in an adolescent patient.
Methods: A 14-year-old girl presented with a small pit on midline of the upper lip, which was visible at birth. The patient had never been treated for the congenital sinus because it was asymptomatic. Surgical excision under local anesthesia was performed.
Results: The sinus had a tract extending into 5 mm posteroinferior and had not penetrated the oral cavity. Histological examination showed a fistulous tract lined by keratinized squamous epithelium. After complete excision, there was no recurrence and we obtained a satisfactory cosmetic result.
Conclusion: Congenital sinus of the midline upper lip is extremely rare. This is a special case that is reported because it did not cause symptoms for the patient until she reached adolescence.

Keywords: Congenital sinus, Upper lip

Case Report

Introduction

Congenital sinus occurring in the lips is so rare that its incidence rate is below 0.001%.[1] Midline upper lip sinus is much rarer than that on the lower lip, and only one case has been reported in Korea by Lee et al.[2] Most patients with congenital sinus of the lip are diagnosed during their infancy or early childhood and treated immediately through resection.

The authors found a very rare clinical case of an adolescent patient showing the characteristics of upper lip sinus that was treated successfully.

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Case Report

A 14-year-old girl visited our hospital with a deep hollow lesion in the middle of the philtrum. The lesion had been noted at birth, but was not treated because there were no symptoms of pain, swelling, discharge, or inflammation. There was a small oval hole about 1 mm in diameter around 5 mm below the columella base on the midline of the philtrum. When a probe was inserted, the sinus was directed toward the posterior side, and its depth was around 6 mm (Fig. 1).

The authors thought that it might be the result of an injury during childhood that had not been treated properly and, as a result, the injury had left a spot-size lesion which had been epithelialized, but the patient denied any history of facial injury or any other congenital deformity. The patient did not have family history of facial sinus. The purpose of visiting the hospital was only to remove the small spot for cosmetic reasons.

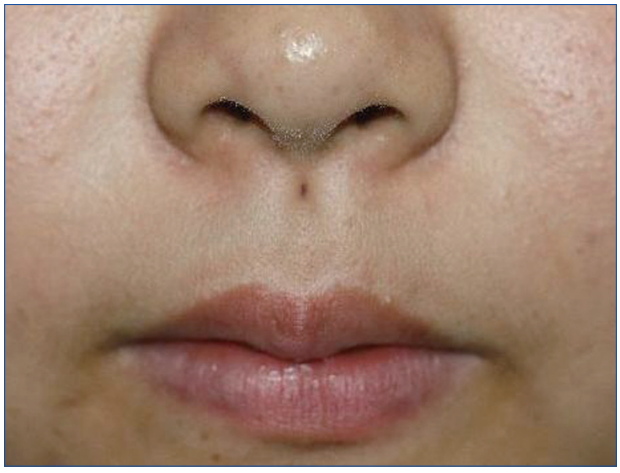

Fig. 1. Preoperative view: 0.5×1 mm sized small pit is visible in midline of the philtrum.

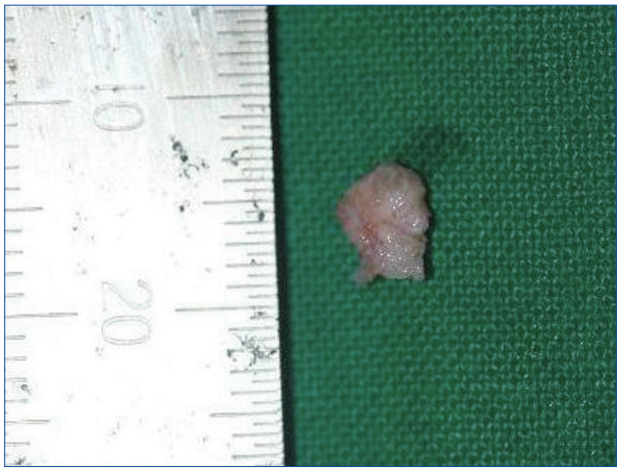

Fig. 2. Excised mass from the philtrum contains 4×5×7 mm sized mass, which contains a fistulous tract. The fistulous tract was observed in the center of this excised sample.

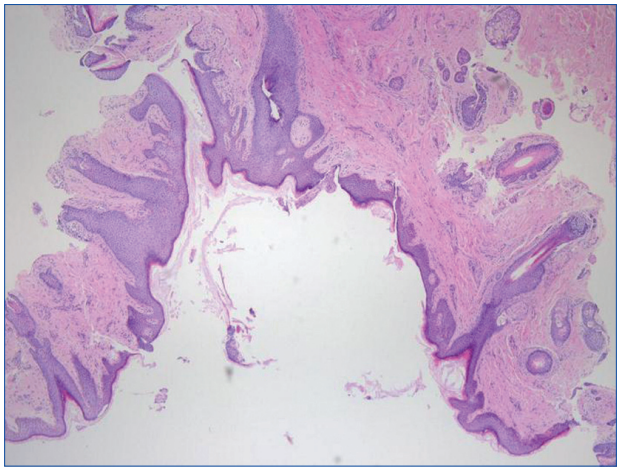

Fig. 3. Histologic finding showed fistulous tract lined by keratiniazed squamous epithelium. Sweat glands and hair follicles were around the tract, but heterotopic tissue was not found (H&E, ×40).

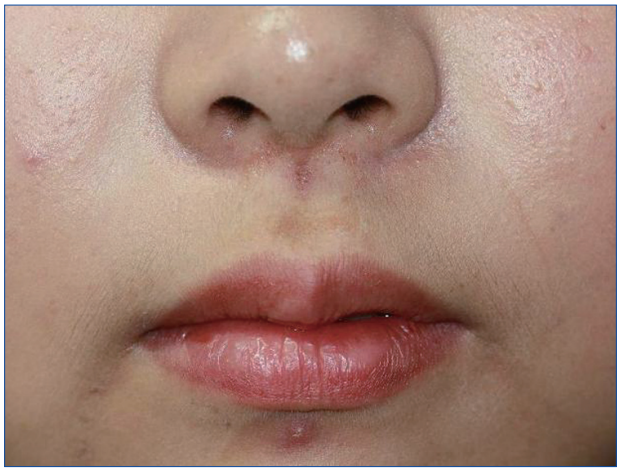

Fig. 4. Postoperative view 1 year after the complete excision was performed.

The fistulous tract was removed under local anesthesia. The lesion ended just beneath the mucosal surface of the upper lip and did not communicate with the oral or nasal cavities. The size of the specimen was $4 \times 7\ mm^2$ (Fig. 2). Microscopic examination revealed that it was lined by cornified squamous epithelium with sebaceous glands and hair follicles surrounding the end of the tract (Fig. 3). Ectopic tissue was not found. No postoperative recurrence was noted on one-year follow-up (Fig. 4).

Discussion

Congenital sinus in the upper lip is such a rare disease in which only 40 cases have been reported. The first reported midline sinuses of the upper lip were reported by Lannelongue and Menard in 1891 and Clavet in 1899. Among them, 26 occurred on the midline, 14 occurred laterally, and 3 occurred bilaterally.[3]

Congenital sinus that occurred on the midline was more frequent in women, and half of the cases were associated with congenital anormalies, such as cleft lip, lingual deformity, and

fistula of the nose.[3] Around 20 cases of congenital midline sinus have been reported in Bonn since the first case was reported by Tange in 1965. In Korea, 1 case of midline upper lip sinus reported by Lee et al.[2] and 2 cases of congenital lateral sinus of upper lip have been by Lee et al.[4]

The size of opening has varied from between 1–2 mm and the depth between 5 to 30 mm, but there has been no case in which the opening was connected to the oral cavity.[5] Accompanying deformities include cleft lip, abnormalities of the maxillary labial frenulum, dental alveolus fissure, and bifid uvula, and some cases were found together with accompanying Pierre-Robin's syndrome or idiopathic precocious puberty.[6]

Histologically, the sinus tract is covered with squamous epithelial cells, and in some reported cases, it was accompanied by sebaceous gland, salivary gland, hair follicles, hyaline cartilage, or mucous gland.

Differential diagnoses include tichofolliculoma, a cartilage-shaped chondroid syringoma in the upper lip. Histologically, tichofolliculoma is characterized by a number of other hair follicles formed radially centering on a hair follicle, and cartilage-shaped chondroid syringoma is characterized by the tubular structure of epithelial cells with 2 or more layers and homogenous basophilic cells filling the tubular lumen.

The pathogenesis of congenital sinus of the lip has not been explained clearly, but is explained by 3 theories. First, the fusion theory hypothesizes that sinus in the upper lip occurs due to failure in the complete fusion of the maxillary processes in birth. The immersion theory assumes the insufficient inflow of mesodermal cells into the ectodermal structure on the midline of the lip.[1,3,5,7] These 2 theories are similar to the theories on cleft face. Another hypothesis is the invagination theory in which upper lip sinus is formed by processes with invaginated epithelial cells as in the formation of nasal pit during the developmental stage.[8] If upper lip sinus occurs due to failure in the inflow of mesodermal cells or in the fusion of the maxillary processes, there should have been cases of upper lip congenital sinus connected to the oral cavity or the nasal cavity, but such a case has not been found. Thus, the invagination theory seems to be more persuasive. However, the exact etiology and genetic course of congenital upper lip sinus remain obscure.

Most of upper lip midline sinus patients are usually detected before their early childhood due to recurrent inflammation. Thus, in general the disease is treated surgically for preventing infection when it is found. It is treated with surgical resection that includes the sinus and the sinus tract, and postoperative prognosis has been favorable and cosmetically satisfactory.

Our case showed the typical histological characteristics of very rare upper lip midline sinus without accompanying congenital deformities. On the other hand it displayed an atypical clinical pattern: a long-term asymptomatic state, which was detected at the oldest age among cases reported in Korea. Therefore, we report the additional case of congenital upper lip sinus and hope this report will help further the understanding of the upper lip sinus.

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