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Case reports

LIP PITS ABSCESS: ISOLATED CONGENITAL MIDLINE UPPER LIP SINUS

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Abstract: Congenital midline upper lip sinuses or fistulas are exceptionally uncommon condition following abnormal fusion of embryologic structures. Here, we report a case of congenital upper lip sinus type I presented as upper vestibular fold abscess in a seven year old boy.

Key words: Congenital, midline sinus, upper lip, vestibular abscess.

INTRODUCTION

Congenital midline lip fistulas or sinuses are infrequent malformations. The anomalies are typically found in Van der Woude syndrome, characterized by a pair of lower-lip dimples in relation with cleft palate. The incidence of the congenital lower lip fistula is considered to be 0.001% whereas upper lip sinuses are even more unusual, and they usually present as a dimpling into a blind sinus, that breaches the orbicularis oris muscle ending under neath the mucosal surface of the lip, without communication with the oral cavity (1, 2). Median fistula of the upper lip may be accompanied with other congenital midline abnormalities, such as nasal dermoid cyst, median cleft, sinus of the labial frenulum, midline sinus of the dorsum of the nose, double frenulum or hypertelorism, suggesting a collective embryological developmental fault of midline malformations (3).

CASE REPORT

A7-year-old boy presented to have a midline pit in the upper-lip philtrum since birth. He had recurrent bulge around the frenulum of the upper lip with clear fluid discharge through external orifice of upper lip. There were no other related congenital anomalies and none of his family members had similar symptoms, lip pits, cleft lips or cleft palates. He was treated by paediatric surgical team with a course of antibiotics but to no avail when the swelling recurred multiple times. He was then referred to our department with features of an abscess and pus discharge coming out from the sinus. Clinical examination revealed an upper lip sinus located on the midline of the philtrum just below the base of columella with a small pit of 1.0 mm in diameter (Figure 1). Over the frenulum portion of upper lip vestibule, there was a fluctuant swelling with very thin erythematous mucosal surface (Figure 2). There were no abnormalities noted elsewhere in the body. He was started with antibiotic and posted for surgical exploration and drainage of the abscess. Intraoperatively, a small transverse incision was made over the upper vestibular swelling to drain the pus and a metal probe was inserted into the opening on the cutaneous surface of the up-

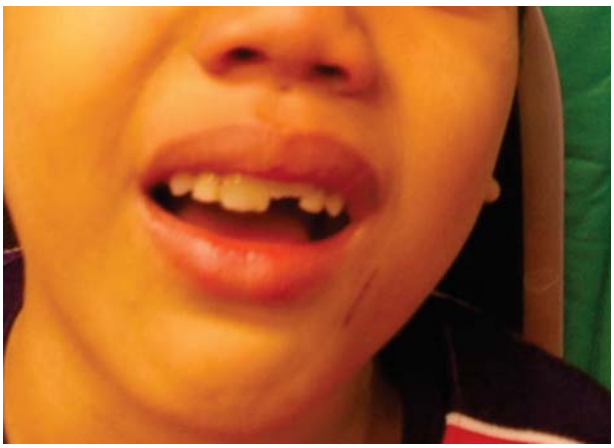

Figure 1. *Small pit in the midline of philtrum just below the base of columella*

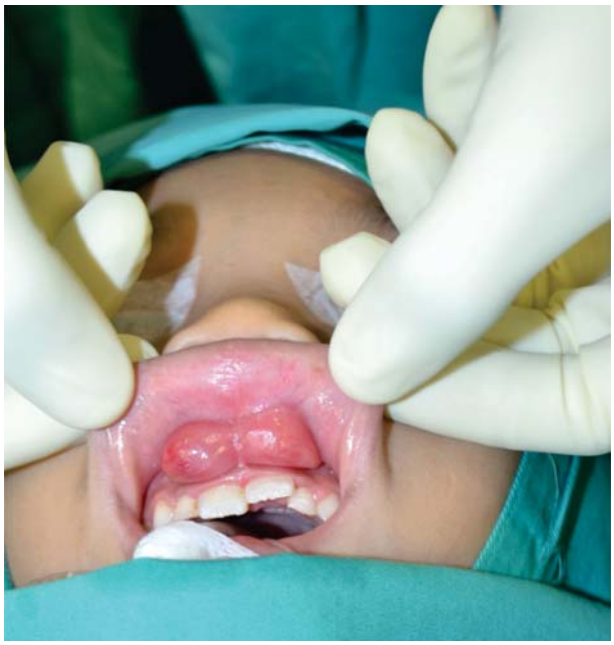

Figure 2. *The upper vestibular fold swelling existed on either side of frenulum*

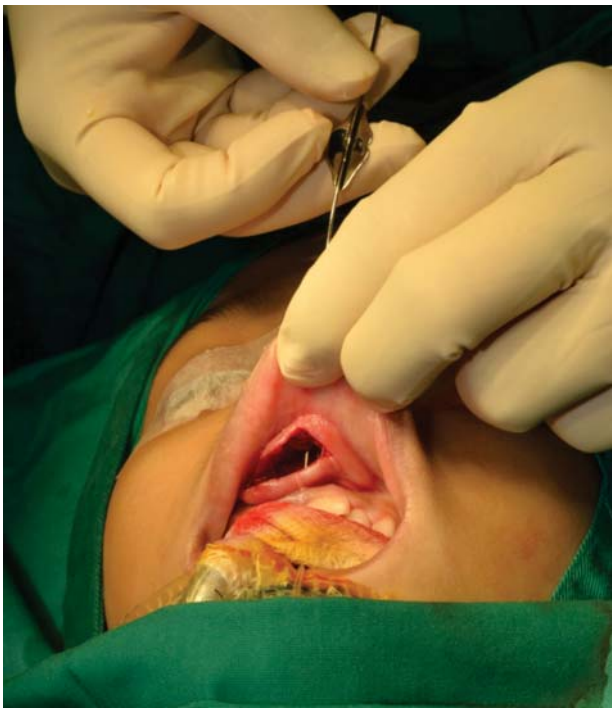

Figure 3. *Communication between the sinus with abscess cavity as shown by the metal probe*

per lip which was found to becomm unicated with the intraoral swelling (Figure 3). Then the entire sinus tract was excised completely, leaving a small ellipse of skin around the defect and the inner surface area was curetted. Layered closure of the skin done while the intraoral wound was packed with saline soaked ribbon gauze dressing for secondary wound healing. Histopathological examination of the specimen showed a tubular structure measuring 7 mm in length and 5 mm in diameter. Cut section shows present of lumen in the centre of the tubular structure. Microscopically, the sinus tract lined by stratified squamous epithelium with underlying stroma show mild lymphocytes and plasma cell infiltrate. The tissue from intraoral mucosa show typical features of an abscess. At one week postoperatively the wound healed well.

DISCUSSION

Congenital upper lip sinuses are so rare and have only been sporadically reported in journals with no consensus on the definitive treatment of choice (2). To date, only several cases of upper lip sinus and fistulas have been reported worldwide with only 31 individual cases comprise of 13 cases of type I with female predilection, 9 cases of type II and 9 cases of type III. Among the 13 cases of type I upper lip sinus, only 1 was a male similar to our case which made it up to only 2 cases have been reported so far over the globe (4). While the mechanisms of the development of congenital upper lip sinuses are still partially understood, there are three leading theories behind their aetiologies; 1) the invagination theory suggests that upper lip sinuses are a result from a failed ectodermal invagination of the nasal placodes during the fronto nasal process development, 2) the merging theory which states that the sinus is due to abnormalities in the normal mesodermal integration development, and 3) the fusion theory proposes an incomplete integration between the fronto nasal and maxillary processes (1). Because this condition is uncommon, much of our understanding is based on details of individual case reports. In 2011 Aoki et al developed a classification system for upper lip sinuses; (a) type I: midline sinus with no associated anomalies; (b) type II: midline sinus with additional anomalies; and (c) type III: the lateral sinus with or without associated anomalies (5).

A high index of suspicion is important in identifying this rare condition especially when patients present with periodic upper lip swelling and discharge. Repeated infections occurred in 25% of the reported cases. A comprehensive history and examination should be conducted, to look properly for congenital pits on the lips and accompanying congenital abnormalities. Van der Woude syndrome is known to be associated with cleft lips, palate and lower lip pits; however, upper lip sinuses are not known to be linked with any specific mode of genetic inheritance (1).

Our case represent type I of midline upper lip sinus, without other accompanying anomalies and the patient's family history also did not reveal any similar conditions, which suggests that the abnormality may be spontaneous rather than inherited and was a rarest of

all forms of classification. Without awareness and concern for underlying sinus, one can never formulate the complete diagnosis and therefore addressing adequately any secondary condition of it e.g. intraoral abscess. Management comprises of complete surgical excision via an intraoral or extra oral approach; whereas incomplete excision leads to recurrences and ultimately cosmetic deformities. Some centres injected 0.5% methylene blue through the opening of fistula to ensure complete excision of the tract and served as indicator of incorrect plane of dissection (3, 5, 6). Different centre may treat the abscess separately whereby the abscess is drained first before an excision of the tract is carried out (5). In this case, the drainage and curettage of the abscess with excision of the sinus tract were done simultaneously to reduce anaesthetic risk to paediatric patient as well as to minimize any psychological trauma to the patient. Nevertheless, longer follow up need to be carried out in order to monitor any recurrence and complication.

In conclusion, recurrent vestibular abscess is one of the complication/sequelae of midline sinus. Complete removal is recommended to render patient free of the disease without recurrence.

DECLARATION OF INTEREST

The authors declare that there are no conflicts of interest.

Sažetak

APSCES NA USNAMA: IZOLOVANI KONGENITALNI SINUS SREDNJE LINIJE GORNJE USNE

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Kongenitalni sinusi ili fistule srednje linije gornje usne su izuzetno retka stanja, koja prate abnormalno spajanje embrioloških struktura. U ovom radu izložićemo prikaz slučaja kongenitalnog sinusa gornje usne tip I, koji se prezentuje kao apsces vestibularnog nabora gornje usne kod sedmogodišnjeg dečaka.

Ključne reči: kongenitalno, sinus srednje linije, gornja usna, vestibularni apsces.

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