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Adult Presentation of Congenital Midline Upper Lip Sinus

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***Abstract*: Midline congenital malformation of the upper lip sinus is an extremely rare presentation. Congenital lower lip sinus reported as 0.001% of the general population, with even rarer sinus presentation over the upper lip. Till date, there was only one case reported in Malaysia. We hereby report the findings and treatment of congenital columella sinus of a 19-year-old gentleman.**

***Keywords*: Adult, Columella, Sinus tract, Upper lip.**

1. Introduction

Congenital columella sinus is an extreme rare disease in Malaysia. First case was reported by Lannelongue et al. as early as 1879. Although the exact pathophysiology of this condition is not well understood, there were three main theories, which are invagination, merging and fusion theories, by Satvinder Singh Bakshi et. al., 2014. A classification of upper lip sinuses which was developed by Aoki et al. in 2011 will be discussed. This case discusses regarding rare case of upper lip sinus and its surgical management.

2. Case Report

A 19 years old gentleman presented with sinus at the base of the columella. Intermittently, he experienced discharge from the sinus in his teenage years. History taking revealed the presence of sinus since birth. He did not have any family history of congenital craniofacial abnormality.

Upon examination, the sinus was approximately 3mm with no intraoral extension. (Fig. 1) No other congenital anomalies were identified. A cone beam CT scan was done with radiopaque cone gutta percha into sinus. (Fig. 2) It demonstrated 1.5cm depth extending to anterior maxilla. Surgical excision was planned for this patient. Intraoperatively, a fistula probe was inserted through the cutaneous sinus at base of columella. Sterile water was flushed through the sinus using 20G branula. However, there was no communication with the oral cavity. (Fig. 3)

An elliptical incision was made over the cutaneous punctum with probe inserted as a guide of the sinus tract. The excision of the tract was performed in both upper buccal sulcus approach and extraoral approach. (Fig. 4) A total sinus tract measuring 15mm length and 5mm diameter was excised and sent for histopathology examination. (Fig. 5)

A. Histopathology Findings

Macroscopic examination showed a gross specimen of grayish tissues measuring in 15mm length and 5mm in diameter. Microscopically, serial sections showed areas of granulation tissue with mild lymphocyte and histiocyte infiltrates. The tissue background consists of dense fibrocollagenous tissue. Few nerve fibres are identified. No epitheliod granuloma or malignancy seen.

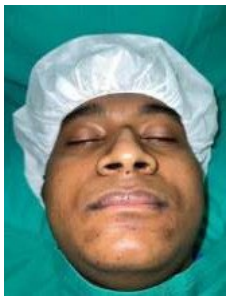

Fig. 1. Sinus at the base of columella

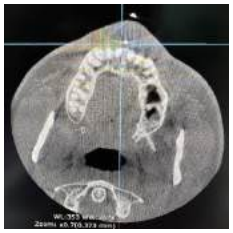

Fig. 2. Cone beam CT scan with radiopaque cone gutta percha into sinus

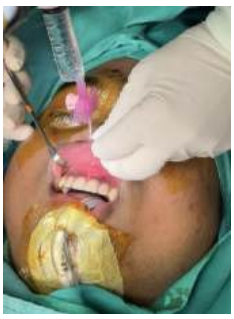

Fig. 3. Flushing of sterile water through sinus to identify communication intraoral

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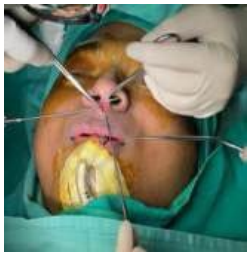
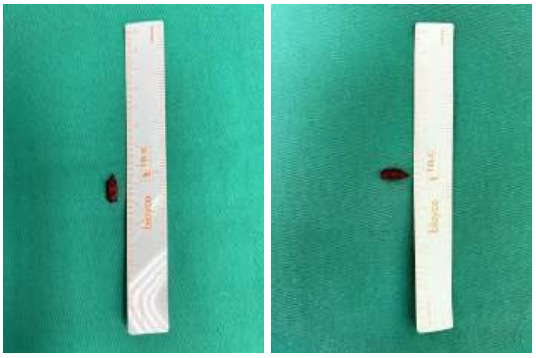


Fig. 4. Elliptical incision made over the cutaneous sinus



A - Length of sinus tract B - Diameter of sinus tract

Fig. 5. Excised sinus tract

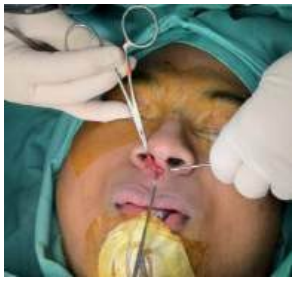


Fig. 6. Excision of the sinus tract with extraoral approach

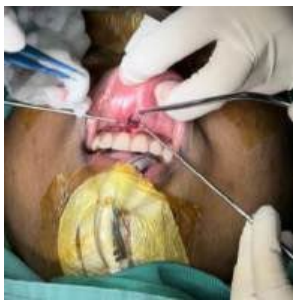


Fig. 7. Elliptical incision over the end point of sinus intraorally

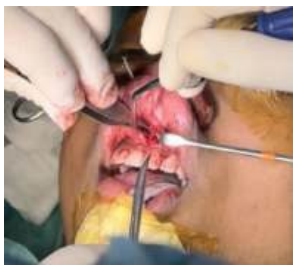
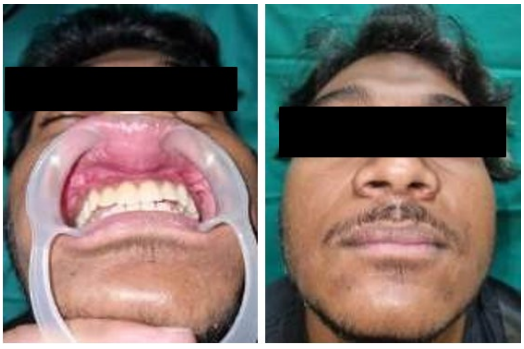


Fig. 8. Whole sinus tr act being identified, with arrow showing the end point of probe

A - Upper buccal B – Extra oral

Fig. 9. Post repair of both incisions made



A - Upper buccal B – Extra oral

Fig. 10. One month post excision

3. Discussion

Ever since congenital columella sinus was first described by Lannelongue et al in 1879, there were only 57 other cases reported in literature worldwide. Asian was the predominant ethnicity of this congenital defect. Nagasao et al., 2014, also reported females are more commonly affected. The only classification available was described by Aoki et al., in 2011, which classified upper lip sinuses into three categories according to its location and presence of other anomalies. There were cases reported that the epithelial anomaly occurred either in isolation or associated with congenital deformities such as cleft lip and palate in Van der Woude Syndrome. This classification is purely descriptive at this stage as no study has looked into its implication towards prognostication and treatment.

a) Type I: Midline sinus without accompanying anomalies
b) Type II: Midline sinus with accompanying anomalies
c) Type III: Lateral sinus with or without accompanying anomalies

In view of the rarity of upper lip sinus, its etiology is poorly understood. The Invagination Theory suggested that the failure of ectodermal invagination of nasal placodes during the formation of frontonasal process to a sinus tract at the upper lip. In the contrary, the merging theory proposed that sinuses are formed as a result of aberrations in the normal mesodermal merging process. Lastly, fusion theory postulates the possibility of a failure of complete fusion between the frontonasal and maxillary process. (Satvinder Singh Bakshi et. al., 2014)

The mainstay treatment of upper lip sinuses is excision of the sinus tract. Upper buccal sulcus excision and incision and drainage are the most commonly described approach by previous case reports. As we were unable to demonstrate an

internal opening in this patient, extraoral approach was used. Extraoral elliptical incision was first made around the cutaneous opening, as shown in Fig. 6. Using a Probe as a guide, the sinus tract was skeletonized from surrounding tissue and extending intraorally. As the sinus abuts shortly before making an internal opening. End point of sinus at the intraoral surface was estimated by palpating for the tip of the probe. An elliptical incision was made around palpated tip of the probe to locate the end point of the sinus. The sinus tract was excised from intraoral approach following the probe. (Fig. 7)

Complete delineation and excision of the whole sinus tract was performed with no remnants left behind. (Fig. 8) Part of the orbicularis oris muscle was cut and repaired with absorbable suture. Skin was closed with non absorbable suture. (Fig. 9). In some of the cases, where the upper lip sinus is complicated with abscess, surgical incision and drainage is preferred. A month after the excision, patient did not have any pus discharge from the base of his columella. Wound at both excision site healed well with minimal scarring. Patient was given subsequent follow up on the scar management. (Fig. 10)

4. Conclusion

A rare case of congenital upper lip sinus without accompanying anomalies was presented in this case report. Despite being rare, a thorough history taking and physical examination is the key in managing an unaccustomed case. Surgical excision remained the mainstay treatment in columella sinus with both intraoral and extraoral surgical approaches can be considered. Complete excision of the sinus is vital to prevent recurrence and optimal cosmetic results.

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