American Journal of Medical Case Reports, 2014, Vol. 2, No. 3, 52-54
Available online at http://pubs.sciepub.com/ajmcr/2/3/3

DOI:10.12691/ajmcr-2-3-3

Supernumerary Nostril Associated with Dermoid Cyst: A Rare Case Report and Review of Literature

Sah BP*, Bhandary S, Pokharel A, Shilpakar SL, Chettri ST, Paudel D

Department of Otolaryngology and Head and Neck Surgery, B.P. Koirala Institute of Health Sciences, Dharan, Sunsari, Nepal
*Corresponding author: bpshahent@gmail.com

Received March 03, 2014; Revised April 04, 2014; Accepted April 07, 2014

Abstract Supernumerary nostril is an extremely rare congenital entity which results from aberrant embryological development. The review of the literature reveals that only 31 cases of supernumerary nostril have been reported. They can be associated with other congenital anomalies. The accessory nostril may or may not communicate with the ipsilateral nasal cavity, probably depending on the degree of embryological progression of the anomaly. A case of supernumerary left nostril with no nasal cavity communication and with dermoid cyst over the right nasomaxillary groove is presented. The surgical treatment is described and the different speculative theories related to the embryogenesis of supernumerary nostrils are also reviewed.

Keywords: *supernumerary nostrils, accessory nostril, dermoid cyst*

Cite This Article: Sah BP*, Bhandary S, Pokharel A, Shilpakar SL, Chettri ST, and Paudel D, "Supernumerary Nostril Associated with Dermoid Cyst: A Rare Case Report and Review of Literature." *American Journal of Medical Case Reports*, vol. 2, no. 3 (2014): 52-54. doi: 10.12691/ajmcr-2-3-3.

1. Introduction

Supernumerary nostril is a very rare type of congenital anomaly. The first reported case was published in 1906 by Linsday, who described a patient with bilateral supernumerary nostrils. In that case, the external openings of the supernumerary nostrils were situated above the normal nostrils, and the accessory nasal cavities communicated with the ipsilateral nasal cavities. In his report, Lindsay proposed the theory of dichotomy by atavism or parallel evolution. [1] In 1920, Tawse reported a patient with a unilateral supernumerary nostril that communicated with the nasal cavity. [2] In 1987, Reddy and Rao reported a case of a third nostril that was situated below the left nostril; they hypothesized that the extra nostril arose as a result of an accessory placode or pit. An accessory nasal placode may be present either above or below the normal nasal placode [3].

In this article, we describe a case in which the supernumerary nostril with a small accessory nasal cavity, which did not communicate with the normal nasal cavity on the same side, appeared in a 15 year old boy who also had a dermoid cyst first noticed by the the patient 3 years back.

2. Case Report

A 15 year old boy was brought to the ENT Department for evaluation of an opening above the left nostril since birth and a swelling along the left nasomaxillary groove noticed since last 3 years. The patient didn't give any history of discharge from the accessory opening. According to the history provided by the mother, her pregnancy had been uneventful and the patient's birth was normal. The patient had reached normal milestones of motor and intellectual development. No family history of such an anomaly was reported.

Physical examination revealed the presence of a small cavity 3 mm above the left nostril. The cavity was lined with hair follicles. Nasal endoscopy of the accessory nasal cavity revealed that it was small and did not communicate with the ipsilateral normal nasal cavity, a finding that was later confirmed by CT Scan. No anomalies were seen in the normal nasal cavities. Examination also revealed a cystic lesion of size 2.5cm x 1.5cm involving the nasofacial groove and adjacent soft tissue of the face on right side. FNAC from the swelling showed features of cystic lesion. Careful examination of other systems did not detect any other abnormality.

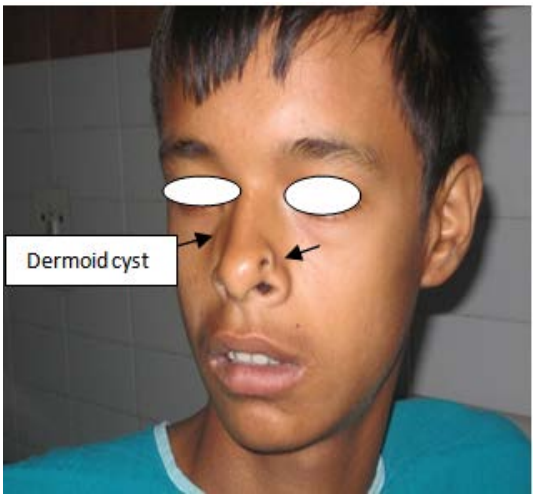

Figure 1. Left Accessory Nostril and Right Dermoid

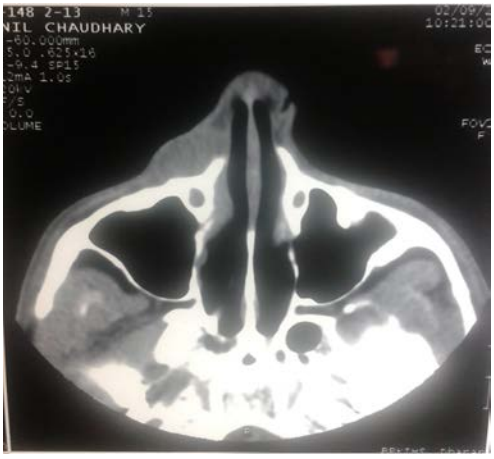

Figure 2. Axial CT Scan of Nose and PNS showing accessory nostril not communicating with nasal cavity

The patient underwent surgical excision of the dermoid cyst and accessory nostril followed by reconstruction of the defect. With the patient under general anesthesia, a elliptical incision was made incorporating the margin of the accessory nasal cavity. The mucous membrane and an almost equally thick section of submucous tissue in the accessory nasal cavity were then excised. Extreme care was taken to avoid damage to the cartilaginous frame of the accessory nasal cavity and the nasal cavity on the left side. The raw roof and the raw floor of the accessory nasal cavity were sutured together to obliterate the cavity, and the skin incision was closed with 4-0 polypropylene suture material. Similarly an incision was given along the right nasofacial groove and dermoid cyst was excised. The skin incision was closed with 4-0 polypropylene suture material. The patient recovered uneventfully.

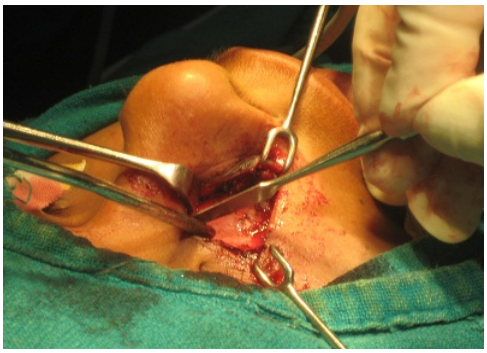

Figure 3. Removal of Dermoid cyst

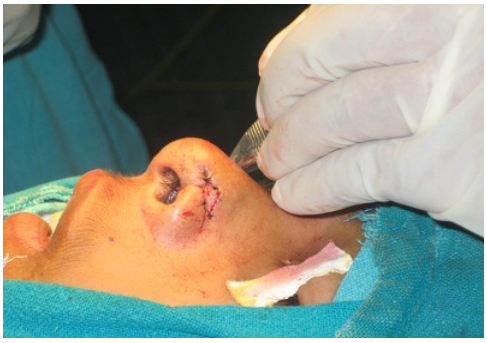

Figure 4. Removal of accessory nostril

Histopathological examination of the excised tissues confirmed the diagnosis of accessory nostril and dermoid cyst respectively. At the 2 week follow-up examination, the patient was doing well. The functional outcome was excellent and the cosmetic result was satisfactory.

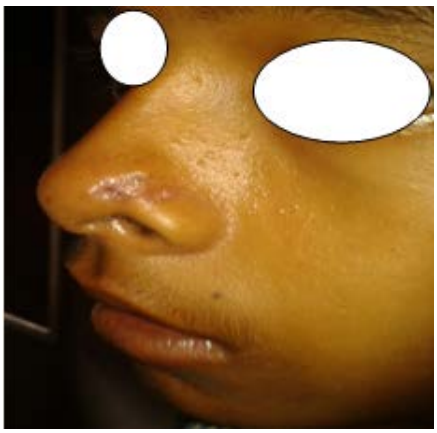

Figure 5. Healed Repaired accessory nostril

3. Discussion

The face develops from five facial primordia appearing as prominences around the stomodeum or primitive mouth. There is a single median frontonasal prominence and paired maxillary and mandibular prominences. These prominences are produced by the neural crest cells migrating into the branchial arches during the fourth week of gestation. Facial development occurs mainly between the fourth and the eighth week. By the end of the fourth week, bilateral thickenings develop on the ventrolateral ectoderm of the frontonasal prominence, called nasal placodes.

The margins of the placodes proliferate into medial and lateral nasal prominences, resulting in the placodes lying in depressions called nasal pits. The medial nasal prominences merge together and with the maxillary and lateral nasal prominences, resulting in separation of the nasal pits from the stomodeum. The nasal pits become nasal sacs and then develop into nostrils and nasal cavities [4].

Supernumerary nostrils are exceedingly rare congenital anomalies of unclear etiology. In 1962, Erich reported a case of double nose. He also supported Linsday's theory of dichotomy by atavism or parallel evolution, and he further speculated that if the accessory nasal pit is located too laterally, the fusion of the lamina is not affected, which leads to the formation of a supernumerary nostril. [5] In 1972, Onizuka and Tai reported the case of a single accessory nostril that had developed above the nasal ala. [6] In 1987, Nakamura and Onizuka reported a similar case, and they hypothesized that the cause was probably a localized defect in the lateral nasal process. [7] In 1992, Chen and Yeong described a case of bilateral supernumerary nostrils that were situated below the normal nasal openings, and they proposed treating such anomalies by staged corrective surgery. [8] In 2001, Hallak et al reported a case of supernumerary nostril in which a blind cavity was present in a normally developed nose. They advocated that corrective surgery be performed at an early age to prevent any possible alar deformity. [9] The present case is the 32nd case of supernumerary nostril being reported. Aslanabadi et al. reviewed 31 cases of supernumerary nostril that had been reported worldwide (Table 1) [10].

Table 1. Cases of supernumerary nostril reported in the literature. [10]

No.	Reported by	Year reported	Side of nose	Upper/Lower/ Same level	Medial/Lateral	Isolated or associated anomalies
1.	Lindsay	1906	Bilateral	Upper	No data	Isolated
2.	Tawse	1920	Right	Upper	Lateral	Isolated
3.	Simonetta	1936	Left	Upper	No data	Isolated
4.	Holmes	1950	Left	Upper	No data	No data
5.	Onizuka and Tai	1972	Left	Upper	No data	Isolated
6.	Get'man	1973	No data	No data	No data	No data
7.	Sharma	1975	Nose tip	Upper	Medial	Isolated
8	Rawat and Gupta	1975	Right	Same level	Medial	Congenital double columella
9.	Nakamura and Onizuka	1987	Right	Same level	Medial	Isolated
10.	Reddy and Rao	1987	Left	Lower	No data	Isolated
11.	Coessens et al	1992	Right	Upper	Lateral	Hypoplastic heminose
12.	Coessens et al	1992	Right	Upper	Lateral	Hypoplastic heminose
13.	Chen and Yeong	1992	Bilateral	Lower	No data	Isolated
14.	Chen and Chen	1994	Left	Upper	No data	Isolated
15.	Deshpande	1995	Right	Upper	No data	Isolated
16.	Jian et al.	1995	Left	Upper	Lateral	Cleft lip and palate
17.	Umeda et al	1996	Bilateral	Upper	No data	Cleft lip and palate
18.	Tambwekar et al	1997	Right	Upper	No data	Incomplete naso-ocular cleft
19.	Williams et al	1998	Right	Same level	Lateral	Isolated
20.	Ramachandraiah and Chakravarthi	1999	Columella	Same level	No data	Isolated
21.	Hallak et al	2001	Left	Upper	Lateral	Isolated
22.	Zbar et al	2003	Left	Upper	Medial	Isolated
23.	Jagannathan et al	2003	Right	Upper	Medial	Contralateral naso-ocular cleft
24.	Cuervo de la Calle et al	2004	Columella	Same level	Medial	Congenital auricular hypoplasia
25.	Guion-Almeida et al	2004	Left	Upper	Lateral	Unilateral cleft lip palate
26.	Guion-Almeida et al	2004	Bilateral	Upper	Medial	Frontonasal dysplasia
27.	Sinha et al	2005	Left	Upper	No data	Microcornea and congenital cataract
28.	Numanoglu et al	2007	Bilateral	Upper	Lateral	Esophageal atresia and patent ductus areteriosus
29.	Powar et al	2007	Right	Same level	Lateral	Esophageal atresia and patent ductus areteriosus
30.	Franco et al	2008	Left	Upper	Lateral	Isolated
31.	Aslanabadi et al	2009	Left	Same level	Lateral	Esophageal atresia, imperforate anus and patent ductus areteriosus
32.	Sah BP et al	present case	Left	Upper	Lateral	Dermoid cyst

Most reported cases of supernumerary nostrils have been unilateral, and most were associated with other craniofacial malformations, such as a facial cleft. A supernumerary nostril may or may not communicate with the ipsilateral normal nasal cavity, depending on the extent of the anomaly's embryologic progression. [10] Our case of supernumerary nostril is not communicating with nasal cavity and is associated with dermoid cyst. This association was not reported previously.

4. Conclusion

Because of the extreme rarity of this congenital anomaly, its cause and development remain largely hypothetical. One common observation noted is that all authors advise early surgery, including excision of the fistulous or blind tract, or performing a fistulorhinostomy when the proximal portion is not accessible. Surgery at an early age is recommended; it will prevent possible subsequent alar deformity and will provide a more normal appearance, essential for normal psychosocial development [11].

References

[1] Linsday, B., A nose with supernumerary nostrils, *Transactions of the Pathology Society of London*, 57, 329, 1906.

[2] Tawse, H.B., Supernumerary nostril and cavity, *Proc R Soc Lond*, 13, 28, 1920.

[3] Reddy, K.A., Rao, A.K., Triple nostrils: A case report and review, *Br J Plast Surg*, 40, 651-2, 1987.

[4] Moore, K.L, *The developing human: Clinically oriented embryology*, WB Saunders, Philadelphia, 1988, 207.

[5] Erich, J.B., Nasal duplication. Report of case of patient with two noses, *Plast Reconstr Surg*, 29, 159-66, 1962.

[6] Onizuka, T., Tai, Y., Supernumerary nostril. Case report, *Plast Reconstr Surg*, 50, 403-5, 1972.

[7] Nakamura, K., Onizuka, T., A case of supernumerary nostril, *Plast Reconstr Surg*, 80, 436-41, 1987.

[8] Chen, M.T., Yeong, E.K., Supernumerary nostrils, *Br J Plast Surg*, 45, 557-8, 1992.

[9] Hallak, A., Jamjoom, H., Hosseinzadeh, T., Supernumerary nostrils: A case report and review, *Aesthetic Plast Surg*, 25, 241-3, 2001.

[10] Aslanabadi, S., Djalilian, H., Zarrintan, S., Supernumerary nostril together with esophageal atresia, imperforate anus and patent ductus arteriosus: a case report and review of the literature, *Pediatr Surg Int*, 25, 433-36, 2009.

[11] Chen, C.T., Chen, Y.R., A case report of triple nostrils, ChangGung Med J, 17, 280, 1994.