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# Supernumerary Nostril with Ipsilateral Maxillary Sinus Hypoplasia: Surgical Reconstruction in an Adolescent

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## Abstract

Supernumerary nostril, otherwise known as triple nostril or accessory nostril, is a rare congenital deformity resulting from aberrant embryological development. It is often found associated with other deformities of face such as cleft lip, microcornea and congenital cataract. In the following case report, we describe the case of a 14-year-old boy presenting with a unilateral supernumerary nostril in conjunction with congenital underdevelopment of right maxillary sinus. CT scan showed a deformed right nasal cavity with non-visualization of nasal turbinates and meati and an undeveloped right maxillary sinus. Following reconstructive surgery, the supernumerary nostril was merged with the right nostril to give rise to a single right nasal cavity. A second-stage surgery on the lines of rhinoplasty is planned to further improve the shape of the nose.

## Keywords

Supernumerary Nostril, Maxillary Sinus Hypoplasia, Accessory Nostril, Congenital Deformity, Surgical Reconstruction, Duplication Anomalies, Triple Nostril, Rhinoplasty, Maxillary Sinus Aplasia

## 1. Introduction

Since the first case reported by Lindsay in 1906, fewer than 50 cases of supernumerary nostril have been reported in the literature [1]. While most cases are diagnosed within the first couple of years of life, some minor deformities continue to be undiagnosed till the second decade of life, being misdiagnosed as skin pit mal-

formations [2]. Supernumerary nostril is often confused with polyrhinia (double nose), both of which constitute duplication anomalies of nose. Differential diagnoses of these anomalies include glioma, encephalocele, nasal dermoid, nasolacrimal duct duplication, mid-facial cleft and proboscis lateralis [3].

Accessory nostril is often found in conjecture with other congenital deformities, in this case maxillary sinus aplasia of the same side, 1 similar case been reported previously. Majority cases have no symptoms due to this defect. However some patients may present with headaches, facial pain and voice problems.

## 2. Case Report

A 14-year-old boy hailing from a rural setting presented to the outpatient department with an accessory nostril situated above the right nostril since birth (**Figure 1**). There was no passage of air or complaints of nasal discharge from the accessory nostril. Antenatal history and the child's birth was uneventful. All developmental milestones were achieved on time and the child is intellectually and physically at par with his peers. On examination, an opening of approximately 0.5 cm in diameter was present just lateral to the right nostril. The anterior nasal opening on the right side was smaller in diameter when compared to the left side. On nasal endoscopy, the accessory nostril was lined with mucous membrane and ended as a blind sac. The patient underwent Non-Contrast Computed Tomography of nose and paranasal sinuses (**Figure 2**), which revealed the following:

- Undeveloped right maxillary sinus
- Deformed right nasal cavity and non-visualization of the nasal turbinates and meati
- Nasal septum deviated towards the right

**Figure 1.** Preoperative clinical photograph showing unilateral supernumerary nostril.

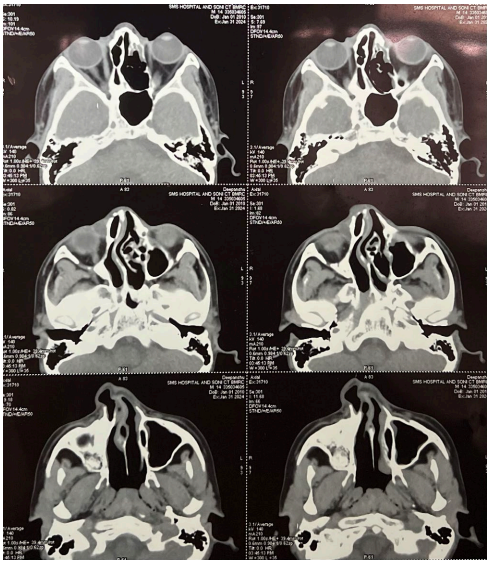

**Figure 2.** Axial computed tomography of nose and paranasal sinus.

Blockage of left OMC along with pansinusitis and obstruction of right nasolacrimal duct were also visualized on imaging which were incidental findings. Thereafter, the patient was planned for reconstruction under general anesthesia. An incision was made over the partition between the accessory and right nostril and dissection was done (**Figure 3**). The partition, which was mostly membranous, was removed and a single cavity was made out of the accessory and right nasal cavity. Inward suturing was done. This reconstructive choice directly addressed the need to improve nasal airflow and symmetry rather than just excising the extra tissue. A triangular wedge was resected from the alar rim of right nostril and suturing was done (**Figure 4**). Patient was followed up on post-operative day 5 and 10, with no fresh complaints a healthy suture line, clear right sided air flow and cosmetic symmetry. The family was also advised a second stage rhinoplasty surgery to attenuate the excess alar cartilage after 6 months.

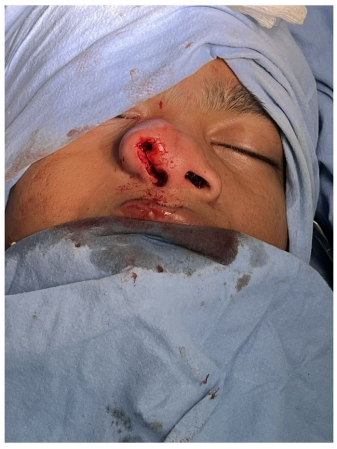

**Figure 3.** Intra-operative image showing incision over the partition between accessory and right nostril.

**Figure 4.** Immediate post-operative photograph.

## 3. Discussion

The combination of nasal endoscopy and NCCT imaging was crucial in ruling out the primary differential diagnoses. Endoscopic visualization of the accessory nostril as a blind sac without any deep-seated tract or discharge made a nasolacrimal duct duplication highly unlikely. Furthermore, the NCCT scan confirmed an intact anterior skull base with no bony defects or intracranial communication, effectively excluding neurogenic anomalies such as encephalocele, nasal glioma, and nasal dermoid cysts. Finally, the absence of a characteristic trunk-like appendage at the medial canthus and the lack of maxillary or palatal clefting on imaging dismissed the possibilities of proboscis lateralis and mid-facial clefts.

Embryology literature states that the mesenchyme covering the caudal surface of the forebrain proliferates with surface ectoderm to form the frontonasal process and the two ectodermal thickenings (nasal placodes) arise on each side of the dependent part of the frontonasal process [4]. Margins proliferate, causing the placodes to lie in depressions known as nasal or olfactory pits. Medial nasal prominences merge together and with the maxillary and lateral nasal prominences, resulting in the separation of the nasal pits from the stomodeum. Nasal pits develop into nostrils and nasal cavities

In 1906, Lindsay put forward a theory which suggested that a laterally located accessory nasal pit leads to formation of supernumerary nose, leaving nasal laminae undisturbed [5].

Nakamura and Onizuka stated that this deformity could be due to random fissuring of the lateral nasal process during fetal development, yielding a unilateral asymmetric deformity. They hypothesized that during the proliferation of mesenchymal cells in the lateral nasal process, a concavity or fissure appears in this area accidentally and, thus, this lateral nasal process is divided into two segments, resulting in two nostrils and two alae on one side. This hypothesis can extrapolate the appearance of accessory nostril either above or lateral (as in this case) to the natural nostril or medially, depending on the position of change in the lateral na-

sal process [6].

Sinha *et al.* reported a case of supernumerary nostril with microcornea and congenital cataract and speculated that anomaly in development of the nasal placodes could be the underlying cause [7].

Erich reported a case of double nose in 1962, supporting Lindsay's theory of dichotomy by atavism or parallel evolution. He further speculated that supernumerary nostril is formed when the accessory nasal pit is located so laterally to the nasal lamina that the accessory nostrils are formed above the natural nostril; do not disturb the fusion of the nasal lam [8].

D.K. Singh *et al.* [9] reported the case of a 7-month-old infant presenting with an isolated supernumerary nostril in which the tract was completely excised and the left nasal ala reconstructed with layered primary closure. Another article reported a 6-day-old neonate with unilateral nasal agenesis along with ipsilateral supernumerary nostril wherein the supernumerary tract was removed and the lateral wall of the nasal cavity reconstructed [10].

This case also highlighted the absence of a well-developed maxillary sinus on the side of the accessory nostril, which could be attributed to two reasons in this patient-intra-uterine developmental abnormalities or due to reduction of nasal ventilation in the first year of life. [11]

Conventional treatment of supernumerary nostril is to remove the nasal tract and correct the ala. The challenge of reconstruction is to restore nasal airflow while attempting to create nasal symmetry.

## 4. Conclusion

Due to its rare occurrence, the exact etiology and development, despite multiple hypotheses, remains largely uncertain. One common strategy all authors advise is early surgical intervention. This is essential in order to prevent the possibility of subsequent alar deformity and physiological limitations. Reconstruction at an early age is also necessary for normal psychosocial development.

## Compliance with Ethical Standards

The authors declare no competing interests. No funding was received for this work. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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