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CASE REPORT

Bilateral Choanal Atresia in an Adolescent Female: A Rare Case Report

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Abstract: Choanal atresia is a rare congenital anomaly of the nasal cavities characterized by lack of patency of the posterior ends of one or both nasal cavities (choanae). It is the most common congenital anomaly of the nasal cavity. Bilateral choanal atresia accounts for a third of the cases and is almost invariably detected in the neonatal age due to respiratory distress. Detection of bilateral choanal atresia in adulthood is extremely rare and has been reported only a few times. We report a case of a teenage girl who was diagnosed with bilateral choanal atresia after presenting with longstanding snoring and intermittent nasal discharge. She was managed with bilateral transnasal endoscopic choanoplasty to restore the choanal patency.

Keywords: choanal atresia, bilateral, choanoplasty

Introduction

Choanal atresia is the lack of patency of one or both choanae.[1] Although generally uncommon, it is the most common congenital anomaly of the nasal cavity. It is seen more commonly in females.[2] Bilaterality is relatively common occurring in up to one third of affected patients.[2] Clinical presentation of patients with choanal atresia depends on whether it is unilateral or bilateral. Unilateral choanal atresia may remain undetected well into adulthood. Bilateral choanal atresia is almost always detected in neonates and infants with signs of upper airway obstruction. Detection of bilateral choanal atresia in adults is extremely rare. As to our search, only 13 cases have been reported to date (Table 1). We present a case of bilateral choanal atresia in a 15-year-old girl who presented with longstanding snoring and intermittent nasal discharge. The diagnosis was ascertained with PNS CT and nasal endoscopy.

Case Report

A 15-year-old girl presented with longstanding snoring and intermittent bilateral nasal discharge. She has been breathing through her mouth for as long as she remembers. Perinatal history was not available. Initial baseline workup was unremarkable. With the suspicion of a sinonasal pathology, non-contrast CT of the paranasal sinuses (Figure 1) was done. It revealed a thickened posterior vomer measuring 7 mm, and medialized pterygoid plates resulting in narrowing of bilateral choanae. The remaining opening was seen to be completely bridged by a membranous structure. An additional incidental finding of a bipartite atlas was also noted. Nasal endoscopy (Figure 2) clearly demonstrated the atretic plate. With the diagnosis of bilateral mixed choanal atresia, transnasal endoscopic choanoplasty (Figure 3) was done on an elective basis. The atretic membranes were first perforated with suction tip. The bony component was cut with rongeur forceps. The vomer was then removed with backbiter forceps. The immediate post operative course was uneventful, and the patient was subsequently discharged. Nasal endoscopy images obtained on her 5th post-operative week (Figure 4) shows patent bilateral choanal openings. The patient also reported significant improvement of her symptoms with complete resolution of the snoring and nasal obstruction.

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Table 1 Previously Reported Cases of Bilateral Choanal Atresia Detected in Adulthood

Authors	Year of Publication	Age/Sex of Patient	Associations Reported	Management
Panda N. et al[12]	2004	22/M	None	Endoscopic choanoplasty
El-Sawy H. et al[11]	2006	24/F	Paranasal sinus hypoplasia	Endoscopic choanoplasty
Yasar H. et al[13]	2007	51/F	None	Endoscopic choanoplasty
Aksoy F. et al[14]	2009	23/F	None	Endoscopic choanoplasty
Tinoco P et al[6]	2009	34/F	None	Endoscopic choanoplasty
Sami A et al[10]	2013	60/F	None	Refused treatment
Sami A et al[10]	2013	24/M	None	Endoscopic choanoplasty
Durmaz C. et al[15]	2016	23/F	Pyknodysostosis	Endoscopic choanoplasty
Roshan K et al[16]	2016	20/F	None	Endoscopic choanoplasty
Anajar S et al[5]	2017	18/M	None	Endoscopic choanoplasty
Mengi E et al[9]	2020	60/M	None	Endoscopic choanoplasty
Kars A et al[17]	2020	18/F	None	Endoscopic choanoplasty
Sutikno B et al[18]	2021	27/F	None	Endoscopic choanoplasty

Discussion

Choanal atresia is the complete blockage of the posterior nasal openings (choanae). It is thought to occur due to failure of the bucconasal membrane to degenerate during the fifth to sixth week of fetal life. It is the most common congenital nasal anomaly; it occurs in 1 in 8000 births. It is seen more commonly in females.[2] Two major types have been described. The most common one is a mixed bony and membranous atresia (70% of the cases), with pure bony atresia accounting for the remaining 30% of the cases. Pure membranous atresia is another type the existence of which is questionable according to current literature.[3,4]

About half of patients with choanal atresia have other associated congenital malformations. Among these, CHARGE syndrome is the most commonly reported association.[4] Other reported associations with choanal atresia include Treacher Collins, Pfeiffer and Crouzon syndromes.[5] Cases of bilateral[6] choanal atresia are more likely to be associated with other anomalies than their unilateral counterparts.[7] In our case, a rare congenital variant of a bipartite (split) atlas was found.

The diagnosis of choanal atresia can be confirmed by nasal endoscopy or cross-sectional imaging. The diagnostic modality of choice in patients with suspected choanal atresia is non-contrast CT of the paranasal sinuses. In addition to making the diagnosis, CT also helps in determining the type of atresia and treatment planning.[6] Suction of nasal secretions and administration of topical vasoconstrictors is recommended prior to scanning.[3]

Bilaterality is relatively common occurring in up to one third of affected patients.[2] Clinical presentation of patients with choanal atresia depends on whether it is unilateral or bilateral. Unilateral choanal atresia may remain undetected well into adulthood; it is usually diagnosed after the patients present with unilateral discharge or obstruction. Bilateral choanal atresia on the other hand is almost always detected in neonates and infants with signs of upper airway obstruction. The typical history is that of a neonate who presents with respiratory distress and cyanosis which worsens upon feeding and is relieved with crying.[8] This is explained by the fact that newborns are obligate nasal breathers; so, diagnosis of bilateral choanal atresia is considered as an emergency requiring measures such as passage of an oral airway.[4] Detection of bilateral choanal atresia in adults is extremely rare. As to our search, only 13 cases have been reported to date. The age of patients previously reported range

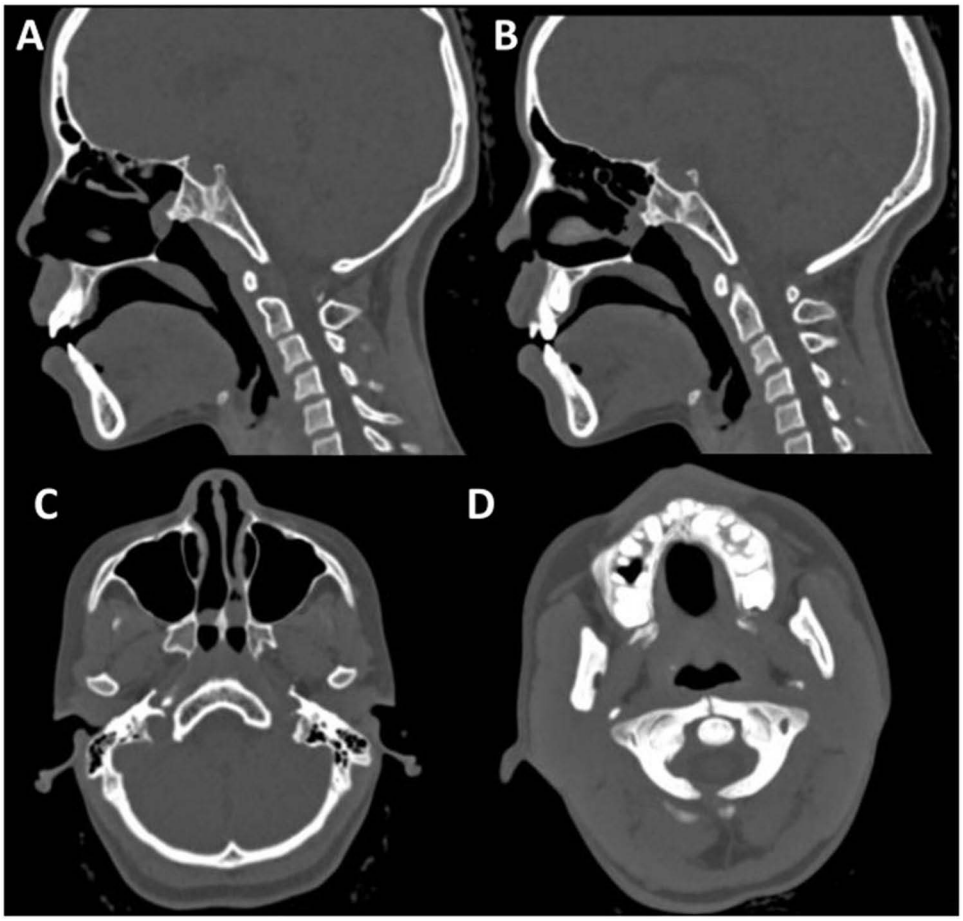

Figure 1 Sagittal (**A** and **B**) and axial (**C**) CT images of the paranasal sinuses in bone window settings demonstrate thickening of the vomer (in **C**), medialization of the pterygoid plates (in **C**) and membranous bridging of the remaining choanal opening. Minimal retained secretions are seen in the nasal cavities. A lower axial section (**D**) shows bipartite atlas, in which both anterior and posterior arches of C1 are unfused.

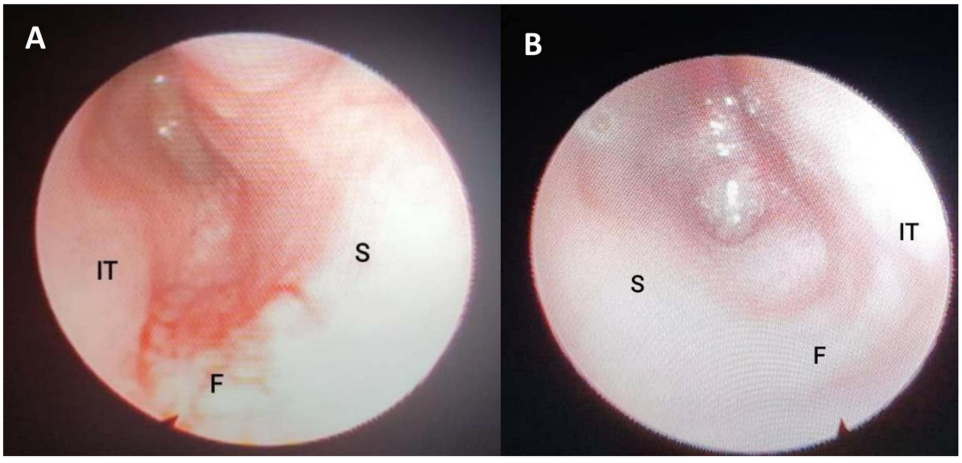

Figure 2 Nasal endoscopic images of the right (**A**) and left (**B**) nasal cavities demonstrate atretic plates in the posterior aspects of the nasal cavities bilaterally.
Abbreviations: IT, Inferior turbinate; S, Septum; F, Floor of nasal cavity.

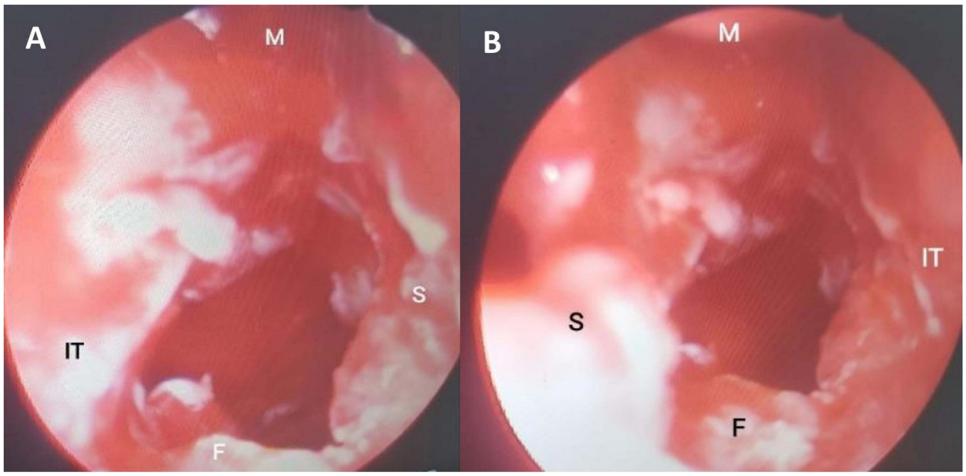

Figure 3 Intraoperative nasal endoscopic images show patency of the right (**A**) and left (**B**) choanae.
Abbreviations: F, Floor of nasal cavity; IT, Inferior turbinate; M, Middle turbinate; S, Septum.

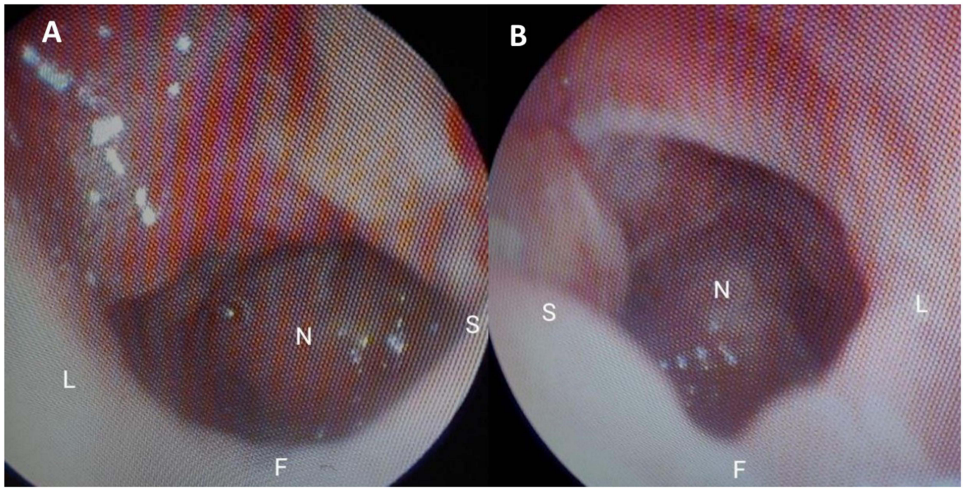

Figure 4 Nasal endoscopy images of the right (**A**) and left (**B**) nasal cavities 5 weeks post-op show patent choanae bilaterally.
Abbreviations: F, floor of nasal cavity; L, lateral wall; S, septum; N, nasopharynx.

from 18 years[5] to 60 years old.[9] Trans-nasal endoscopic surgery was done for all patients except one patient who opted out of surgical management.[10] One case of restenosis after surgery requiring repeat operation has been reported.[11] Our patient underwent bilateral trans-nasal endoscopic surgery. Her post operative course is uneventful. She is currently being followed at the outpatient clinic and is doing well as of the time of writing this report.

Consent Information

Written informed consent was obtained from guardians of the patient to publish this case report. Any personal details and diagnostic images were anonymized to meet the confidentiality requirements. Institutional approval was not required to publish the case details.

Disclosure

The authors report no conflicts of interest in this work.

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