International Journal of Otolaryngology and Head & Neck Surgery, 2024, 13, 187-193
https://www.scirp.org/journal/ijohns
ISSN Online: 2168-5460
ISSN Print: 2168-5452

Unilateral Choanal Atresia in Adults: A Case Series

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How to cite this paper: Kadasah, S., Qahtani, A.A., Al-Sayed, G., Helali, A.A., Aldhabaan, S., Musleh, A., Asiry, A., Asiri, M., Dlboh, S., Al-Malki, A., Alqhtani, A., Al-khulban, M. and Al-Otaibi, S. (2024) Unilateral Choanal Atresia in Adults: A Case Series. *International Journal of Otolaryngology and Head & Neck Surgery*, **13**, 187-193.
https://doi.org/10.4236/ijohns.2024.133018

Received: March 7, 2024
Accepted: May 28, 2024
Published: May 31, 2024

Abstract

Choanal atresia (CA) is a rare occlusion of the posterior choanae. Unilateral cases have been reported more than bilaterally, and it's more often right-sided in those patients. According to the literature, mixed bony-membranous atresia is the most common type. There is a high incidence of craniofacial and visceral anomalies associated with congenital choanal atresia. Therefore, investigation for associated congenital anomalies is an important step before the surgery. We report 2 cases of incidental finding of unilateral choanal atresia in a 21- and 17-year-old with nasal discharge being the only complaint in the former and nasal obstruction with headache in the latter. The patients were then scheduled for day-surgery as a case of choanal atresia for transnasal, endoscopic repair and posterior septectomy. The patients were discharged home on the same day with the absence of restenosis or other complications.

Keywords

Choanal Atresia, Nasal Obstruction, Congenital Anomaly

1. Introduction

Choanal atresia (CA) is a rare occlusion of the posterior choanae [1] [2].

It could be congenital or acquired [2]. It occurs in 1:7000 - 8000 births worldwide with females being more affected than males (2:1) [1] [3]. Unilateral cases have been reported more than bilateral ones and it's more often right-sided in

those patients [1] [3]. Anatomically, it results from a medialized lateral pterygoid plate and an enlarged vomer [4] and regarding the nature of the occluding structure in CA, it has been reported that mixed bony-membraneous is the most common type followed by pure bony structure, while pure membranous atresia was extremely rare [1] [5]. Occurrence could be partial or complete occlusion, in one choana (unilateral CA) or both choanae (bilateral CA) [1].

A high incidence of craniofacial and visceral anomalies is associated with congenital choanal atresia [1] [2]. Therefore, investigation for associated congenital anomalies is an important step before the surgery [6]. This case series aims to illustrate two cases of unilateral CA in adults presenting with rhinorrhea and nasal obstruction; denoting CA as a potential differential diagnosis to be considered in such cases.

2. Case 1 Presentation

A 21-year-old female presented to our clinic with a history of chronic left nasal obstruction with nasal discharge as the only rhinological symptom. Upon examination with zero-degree rhinoscopy, thick mucus secretion and hypertrophied turbinates were revealed in the left nasal cavity. After careful suctioning, the left choana appeared to be obstructed while the right was patent (**Figure 1**). A non-contrasted CT scan of the paranasal sinuses was done denoting left choanal atresia, with mixed bony and membranous components (**Figure 2**). The patient was then scheduled for day-surgery as a case of choanal atresia repair and posterior septectomy. Utilizing zero-degree rhinoscopy, nasal packing with decongestant followed by choanal dilating puncture was done. Using a debrider, soft tissue components were removed. Backbiting forceps were used to remove the posterior part of the nasal septum allowing a bigger window for drilling the bony portion of the choana until both posterior choanae are connected. Lateral drilling of the pterygoid plates was commenced until both Eustachian tubes could be visualized by zero-degree rhinoscopy from the side of the nasal cavities. The patient was discharged home on the same day. Follow-up at the clinic 2 weeks post-operative revealed minimal crustation, patent choanae with no stenosis and the patient was satisfied regarding their breathing. Another follow-up 1-month post-operative revealed the same findings with patent choanae and absence of stenosis.

3. Case 2 Presentation

A medically free 17-year-old came to our clinic complaining of nasal obstruction which was mainly left-sided and associated with a headache. Examination revealed left deviation of the nasal septum with mixed choanal atresia ipsilaterally which was further confirmed by imaging (**Figure 3**, **Figure 4**). The patient was subsequently booked for choanal opening and widening with posterior septectomy utilizing a diamond drill (**Figure 5**). The left choana was opened and widened until the medial pterygoid was followed by posterior septectomy. Then, left hemi

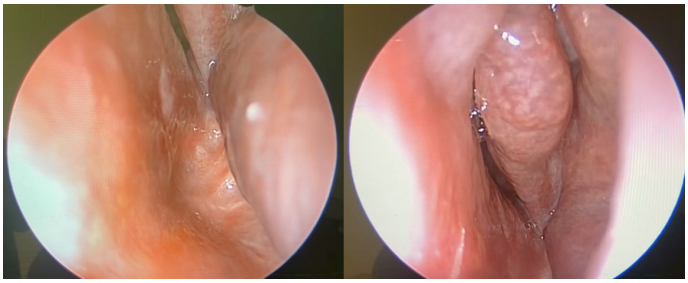

Figure 1. Zero-degree rigid rhinoscopy showing partial, left choanal atresia.

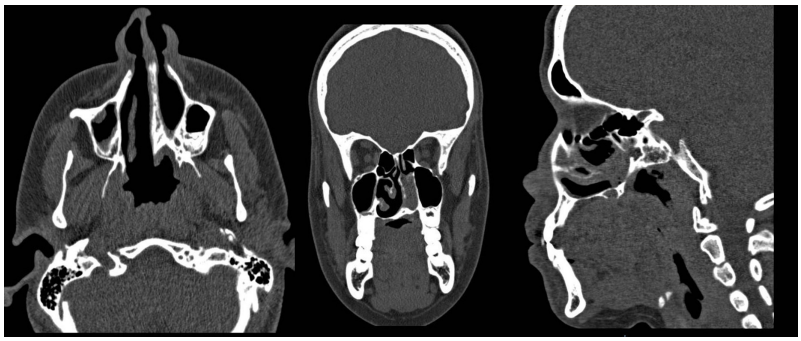

Figure 2. Non-contrast CT PNS denoting left choanal atresia of mixed, bony and membranous components. The nasal septum is slightly deviated to the left. The left bony choana appears narrow measuring approximately 0.3 cm in the axial view at the level of the pterygoid plates, with thickening and leftward bowing of the vomer. Related left nasal bubbly effusions/polyposis are detected.

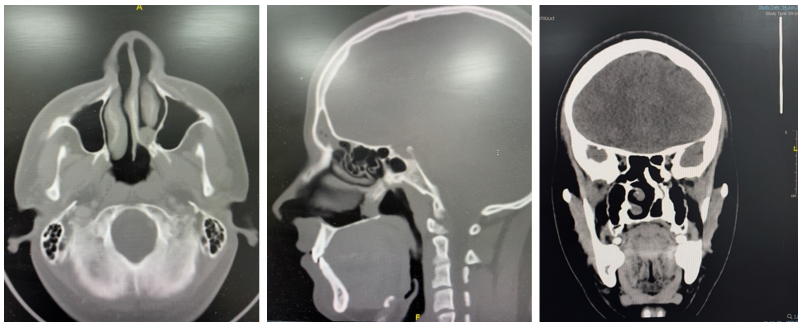

Figure 3. CT facial bones with IV contrast showed a deviated nasal septum with a bony spur on the left side. Attenuated left side of the nasal cavity with markedly attenuated left choana and hypertrophied inferior nasal turbinates with mucosal thickening/polyp on the left side of the nasal cavity.

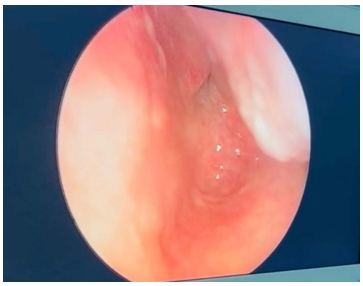

Figure 4. Intraoperative zero-degree rigid rhinoscopy showing left choanal atresia.

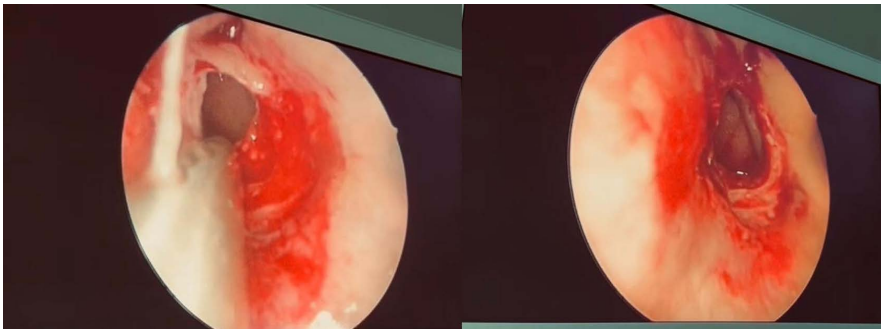

Figure 5. Opening and widening of the choana using a drill.

transfixion incision was done; the flap elevated and deviated septum was removed bilaterally, and the incision closed. A splint was internally fixed, and the patient was instructed to keep it for 10 days. Upon follow-up, the splint was removed, and no nasal obstruction or septal perforation was revealed and the patient reported full satisfaction with patent nasal passages. Both patients are undergoing regular follow-up every 3 - 6 months; assessing for restenosis by direct endoscopy for up to 1-year post-operative.

4. Discussion

Congenital choanal atresia (CA) is an uncommon craniofacial anomaly characterized by obliterated posterior choanae leading to upper airway obstruction [3] [7] [8] [9]. It may be unilateral or bilateral [6] [7] [8] [9]. The exact embryologic explanation of CA is unclear until now. [3] There are 4 theories that were described to explain the cause of CA. The first theory is imperforation of the buccopharyngeal membrane; the second theory is the abnormal resorption of the nasobuccal membrane of Hochstetter; the third is abnormal congenital adhesions of the mesoderm at the choanal site; and the final theory, abnormal mesodermal flow caused by embryologic factors [1] [10]. A study was done and reported unilateral and bilateral CA in babies for women treated with methimazole for hyperthyroidism. Thus, teratogenicity has become a possible cause of CA. [11]

Clinical presentation varies depending on whether it's unilateral or bilateral CA. Patients with bilateral CA present with immediate significant upper airway obstruction and respiratory distress manifested as a drop in oxygen saturation and cyanosis improved upon crying, and aspiration following feeding [1] [2] [3]. Patients with unilateral CA may present in late childhood or even adulthood with minimal symptoms including nasal obstruction, excessive thick mucous discharge, paranasal rash, and less frequently, smell blindness [1] [2] [3].

On examination of CA patients, a catheter inserted in the nose will bend over never reaching the nasopharynx [1] [2] and occlusion seen during mirror examination is confirmatory [1].

Methods of investigation include applying a decongestant followed by nasal endoscopy, as well as an axial view CT scan for paranasal sinuses [2].

Neonates are obligated to breathe through the nose. From this point of view, bilateral choanal atresia is an emergency that needs immediate intervention to avoid unnecessary tracheostomy and even death [1] [2] [3] [6] [12]. Surgical repair for unilateral CA can be delayed to give time for the baby to grow unless there is malnutrition or breathing difficulty [6].

There is a high incidence of craniofacial and visceral anomalies associated with congenital choanal atresia, especially bilateral type [1] [2] [12] [13]. The most common congenital anomalies diagnosed are CHARGE syndrome (coloboma, heart anomalies, choanal atresia, growth retardation, genitourinary abnormality, and ear anomalies) [6]. Therefore, investigating for associated congenital anomalies is an important step before surgery [6].

According to our literature search, reports of unilateral choanal atresia in adults have not been reported often, which could be attributed to the commonly asymptomatic nature of the condition. However unilateral choanal atresia have been reported as acquired cases post-radiotherapy for nasopharyngeal carcinoma. [14]

Surgical repair of CA can be done in many methods chosen depending on many factors including patient age and type of occluding structure. These surgical methods include transnasal, transpalatal and these two methods are more common in recent years [1] [2] [3] [6] Transseptal, transantral, sublabial-transnasal, and open rhinoplasty are also surgical techniques that can be done [2] [3] [6]. Although transnasal endoscopic is the ideal approach in recent years, some studies [3] [4] do not recommend it for neonates due to their narrow nasal cavities.

Postoperative minor complications include bleeding, perforation of the nasal septum, adhesions, restenosis, and the need for revision surgery [2]. Patients who underwent endoscopic transnasal repair are less likely to need revision surgery compared to other methods [2]. Surgical outcomes are not affected by the type of occluded structure (bony, soft tissue, or mixed), mucosal flap use, or whether a stent was applied [2]. Postoperative serious complications can happen such as excessive bleeding as well, skull-base perforation, cerebrospinal fluid fistula, and meningitis [2].

5. Conclusion

Choanal atresia is an important differential diagnosis to be considered in unilateral nasal blockage or rhinorrhea in an adult. Careful nasal endoscopic examination is crucial prior to any rhinological surgery for another cause to ensure bilateral choanal patency. Transnasal endoscopic approach appears to be the most commonly used approach with the least reported complications. Investigations for other associated congenital anomalies is an important step in case of confirmed choanal atresia. Important post-operative follow-up and assessment is imperative to look out for restenosis, septal perforation or skull-base perforations and subsequent CNS infections.

Consent

Written consent was given by the patients to use their pictures and publish this case report.

Conflicts of Interest

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

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