Choanal atresia: the 13 years' experience of our department

Original Article

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Article received on July 13, 2023.
Accepted for publication on January 29, 2024.

Abstract

Objective: To audit our experience with Choanal Atresia (CA).
Study design: Observational retrospective study.
Material/Methods: Clinical data of all CA patients followed from 2003 to 2020 at Hospital Central do Funchal were analysed.
Results: Eleven patients were included and 63,64% were females. They were divided into two groups: Unilateral (63,64%) and bilateral atresia (36,36%). In both groups, mixed type atresia was the most common. Regarding unilateral group, endoscopic repair was performed in 85,71% of cases (71,4% with stents) at a median age of 15 years old. Median number of surgeries was 1,29. Bilateral group had more congenital malformations (50%) and had more reinterventions. All patients underwent an endoscopic approach at a median age of 8 days old.
Conclusions: Associated malformations, younger age at first surgery and higher number of surgeries were more associated to bilateral CA.

Keywords: Choanal atresia; Congenital abnormalities; Surgery

Introduction

Choanal atresia (CA) is a rare congenital malformation caused by the persistence of the buccopharyngeal membrane. Its incidence ranges from 1:5,000 to 1:8,000 live births,[1,2] and it is more often unilateral than bilateral (60% vs. 40%) and affects twice more females than males.[2] Unilateral CA occurs more frequently on the right side and may not be diagnosed in the neonatal period, sometimes remaining undiagnosed even in adulthood.[2,3] This type of atresia may require surgery, depending on the severity of symptoms such as persistent unilateral rhinorrhea, which results from the blockade of the normal nasal mucociliary clearance route toward the nasopharynx.[3] Bilateral CA is a life-threatening condition in newborns, who are

obligate nose breathers, and requires urgent surgical action once diagnosed.[3,4] CA can be clinically confirmed by the inability to pass a 6F plastic catheter through the nose to the nasopharynx (a distance of approximately 32 mm). Nasal endoscopy is important for direct visualization and for distinguishing between atresia and stenosis. CA is defined as a narrow patent choana measuring less than 6 mm in diameter.[5] Several techniques can be used to remove the atretic plates, often depending on the surgeon's preference because the best technique has not been established yet in the literature.[2–4] The objective of this study was to describe the treatment of CA at the otorhinolaryngology department of Hospital Dr. Nélio Mendonça, Funchal, Madeira Island, Portugal.

Materials and methods

This retrospective observational study analyzed the clinical data of all patients who were diagnosed with CA and followed up for at least 1 year between 2003–2020 at Hospital Dr. Nélio Mendonça in Funchal, capital of Madeira Island, Portugal, which has a population of approximately 250,700 inhabitants.[6]

Results

The study sample included eleven pediatric patients diagnosed with CA and operated by the same surgeon, comprising 64% females and 36% males. The sample was divided into two groups: unilateral CA (64%, n = 7) and bilateral CA (36%, n = 4). In the unilateral group, 57.4% of the patients had mixed osseous and membranous CA (described as the most common in the literature),[2] 29% had purely membranous CA, and 14% had purely osseous CA. One patient had an associated cardiac malformation (persistent ductus arteriosus), while the others had no associated malformations. Most patients (85.71%, n = 6) underwent transnasal endoscopic correction, but the transpalatal approach was used in the case of a 26-year-old patient with mixed CA.

The unilateral group had a median age of 15 years (interquartile range [IQR] 25 years, 1 month–42 years) at the time of the surgery. Nelaton© probes were used as stents in 80% of the cases, and a Foley© silicone catheter was used in a 42-year-old patient (20%). In this group, one non-cooperative pediatric patient required surgical reintervention under general anesthesia in the operating room to remove the stent with safety and comfort. In the bilateral group, three patients (75%) had mixed CA and one (25%) had purely osseous CA. Associated malformations were observed in 50% of the patients with bilateral CA (one case of persistent ductus arteriosus and one case of CHARGE syndrome, which stands for coloboma, heart defects, atresia, retardation, genital disorders, and ear disorders).

All patients underwent transnasal endoscopic correction. The median age at first surgical procedure was 8 days (IQR 4.5 years, 3–20 days).

Table 1
Age (months) of the patients with unilateral choanal atresia (CA) at the first surgical procedure

Case	Age (months)
A	8
B	180
C	312
D	1
E	300
F	504
G	5

All patients underwent transnasal endoscopic surgery, and the osseous portion of the atretic plate was removed using a Kerrison micro-punch, except in one patient, who required a diamond drill and excision of the posterior part of the vomer. A Nelaton© probe was used for 6–8 weeks, requiring a mean of three surgical reinterventions (2–5). Thus, the bilateral group required a greater number of surgical reinterventions than the unilateral group (1.29 vs. 3, p = 0.024). This can be explained by the need for surgical revision and stent replacement by a larger one or definite removal.

Figure 1
Patients with bilateral choanal atresia (CA) distributed by the presence or absence of comorbidities

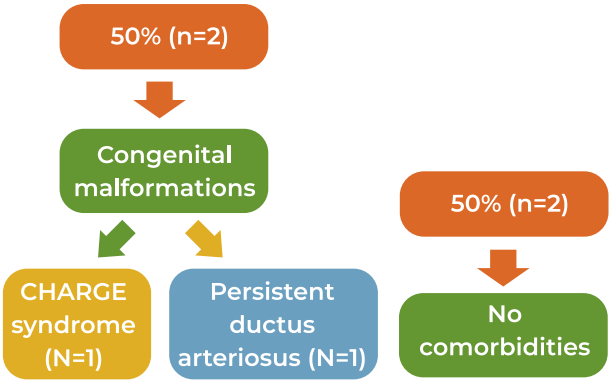

Table 2
Age (days) of patients with bilateral choanal atresia (CA) at the first surgical procedure

Case	Age (days)
H	20
I	6
J	6
K	3

Additionally, a patient with bilateral CA required balloon dilation of the new choanae after stent removal. There were no cases of recurrence during follow-up.

Discussion

The major limitation of this study was the small sample size (11 cases), although it is worth considering that it was carried out on a small island with a population of 250,000 inhabitants.[6] However, our results corroborate those of other studies, such as a German study by Van Schaik et al. and a Canadian multicenter study by Paradis et al., which analyzed the largest sample to date (215 patients).[2,3] These studies also reported a higher incidence in females (63.6 % in our study vs. 59.6%–65.6% in the above two studies), and the percentage of unilateral CA in our study is consistent with the percentage reported in the German study (63.54% vs. 58%).[3] Compared to the Canadian study, there was a slightly lower percentage of mixed CA (63.6% vs. 73.22%), and a relatively higher proportion of purely membranous CA (18% vs. 10.5%) in our study. In our study, 91% of the patients underwent transnasal endoscopic surgery, contrasting with 32% in the Canadian multicenter study.[2] No surgical treatment has been considered as the gold standard to date, with several techniques described in the literature.[4,7] Transnasal surgery was selected for the majority of the cases because of the lower risk of iatrogenic injury leading to postoperative fibrosis and restenosis compared to the transpalatal approach.[4] Other relevant factors are the advances in endoscopic instrumentation and lower risk of dental and facial growth changes reported in the literature.[8]

The stent usage rate was also similar between our study and the aforementioned literature (81% vs. 70%).[2,3] Although these devices reduce the risk of restenosis, their use remains controversial because they can lead to infection, scarring disorders (especially in the septum, alar cartilages, and columella), and granulation with possible restenosis,[4] with a higher risk in children, especially newborns, and in cases of bilateral CA.[4] A higher proportion of stents was used in our study than in the study by Paradis et al., but there was less need for surgical reintervention (45% vs. 49%).[2] Although stents are suitable for bilateral CA, some studies showed better treatment results in cases of unilateral CA with the transnasal approach without stents, which resulted in less trauma and easier postoperative care.[9–12] However, they were used in 71% of the cases with unilateral CA in our study, with good results. No specific stents have been recommended, as they are measured and adapted intraoperatively.[4] They should be made of a soft material and maintained in situ for a short period of time.[13] The most well-known and frequently used materials are:

- Polyvinyl chloride (Portex©): It is the most common, recommended by surgeons because of good results and low complication rates;[14,15] it has the disadvantage of yielding

to the internal nasal temperature, becoming obstructed under ambient pressure in some cases, which leads to restenosis.[16]

- Endotracheal tube: It reduces the risk of restenosis by enabling re-epithelization of the new choanae.[16]
- Polyethylene tracheal tube: It showed no significant differences between uni- and bilateral CA.[17]
- Silicone material: It can be maintained for shorter periods in unilateral atresia and for longer periods in osseous atresia; it can be perforated and maintained in a "U" shape on the back of the nose in bilateral cases.[18] An example is the Foley© catheter, which may be better tolerated, and is easy to introduce, fix, and remove. This material also has a lower rate of complications such as infection or septal and/or columellar necrosis, and may be comfortably used in unilateral CA, with lower restenosis and complication rates.[19]
- Silastic© tubing: It can be maintained for 12–16 weeks postoperatively but has a restenosis rate of 36%.[20]
- Teflon© stent: It does not seem to affect the mucociliary clearance.[21]
- Metal reinforced silicone rubber tube (described in a case).[22]
- Nelaton© probe: It has broad availability and low cost.[23]

Some studies have reported that the use of stents increases the risk of complications; however, their appropriate use, when accompanied with clear instructions to the patient and family members on how to clean the secretions, reduces the risk of complications and restenosis.[4]

A few studies have reported the excision of the posterior part of the vomer in cases of bilateral atresia, and studies by El-Ahl et al. and El-Anwar et al. demonstrated that this resection could eliminate the need for stents.[24,25]

The present study showed that the bilateral CA group had a higher prevalence of associated malformations, experienced first surgical intervention at a younger age, and underwent a greater number of surgical procedures, suggesting a higher rate of restenosis. These data corroborate the findings of previous studies.[2–4] A German study has also reported a greater number of interventions in the bilateral CA group.[2]

Conclusion

CA has a higher prevalence in females, and the bilateral group showed a higher prevalence of associated malformations, first surgical intervention at a younger age, and a greater number of surgical procedures, indicating a higher rate of restenosis.

While the use of stents demonstrated good results, it is crucial to provide clear instructions to patients and family members about how to maintain these devices.

Conflicts of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Data Confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

Funding Sources

This work did not receive any contribution, funding or scholarship.

Availability of scientific data

There are no datasets available, publicly related to this work.

Bibliographic references

1. Assanasen P, Metheetrairut C. Choanal atresia. J Med Assoc Thai. 2009 May;92(5):699-706.
2. Paradis J, Dzioba A, El-Hakim H, Hong P, Kozak FK, Nguyen LHP. et al. A national study of choanal atresia in tertiary care centers in Canada - part II: clinical management. J Otolaryngol Head Neck Surg. 2021 Jul

13;50(1):46. doi: 10.1186/s40463-021-00503-3.
3. van Schaik CGR, Paasch S, Albrecht T, Becker S. Treatment of choanal atresia in a cohort of 29 patients: determinants for success or failure. Int J Pediatr Otorhinolaryngol. 2022 Sep;160:111240. doi: 10.1016/j.ijporl.2022.111240.
4. Alsubaie HM, Almosa WH, Al-Qahtani AS, Margalani O. Choanal atresia repair with stents and flaps: a systematic review article. Allergy Rhinol (Providence). 2021 Dec 17;12:21526567211058052. doi: 10.1177/21526567211058052.
5. Flint P, Haughey B, Lund V, Robbins K, Thomas JR, Lesperance M. et al. Cummings Otolaryngology Head and Neck Surgery, E-Book, 3-Volume Set, 7th ed. Philadelphia: Elsevier; 2020. 3568 p.
6. DREM. Direção Regional de Estatística da Madeira. Resultados definitivos dos CENSOS 2021. Em foco [Internet] 2023 Jan 12; Disponível em: https://estatistica.madeira.gov.pt/download-now/social/popcondsoc-pt/popcondsoc-censos-pt/popcondsoc-censos-emfoco-pt/send/47-censos-emfoco/15577-em-foco-censos-2021-resultados-definitivos-1.html
7. Saraniti C, Santangelo M, Salvago P. Surgical treatment of choanal atresia with transnasal endoscopic approach with stent- less single side-hinged flap technique: a 5-year retrospective analysis. Braz J Otorhinolaryngol. 2017 Mar-Apr;83(2):183-189. doi: 10.1016/j.bjorl.2016.03.009.
8. Saraniti C, Santangelo M, Salvago P. Surgical treatment of choanal atresia with transnasal endoscopic approach with stentless single side-hinged flap technique: 5 year retrospective analysis. Braz J Otorhinolaryngol. 2017 Mar-Apr;83(2):183-189. doi: 10.1016/j.bjorl.2016.03.009.
9. Holzmann D, Ruckstuhl M. Unilateral choanal atresia: surgical technique and long-term results. J Laryngol Otol. 2002 Aug;116(8):601-4. doi: 10.1258/00222150260171588.
10. Grundfast KM, Thomsen JR, Barber GS. An improved stent method for choanal atresia repair. Laryngoscope. 1990 Oct;100(10 Pt 1):1132-3. doi: 10.1288/00005537-199010000-00020.
11. Brown OE, Smith P, Armstrong E, Grundfast KM. The evaluation of choanal atresia by computed tomography. Int J Pediatr Otorhinolaryngol. 1986 Nov;12(1):85-98. doi: 10.1016/s0165-5876(86)80061-1.
12. Lantz HJ, Brick HC. Surgical correction of choanal atresia in the neonate. Laryngoscope. 1981 Oct;91(10):1629-34. doi: 10.1288/00005537-198110000-00007.
13. Velegrakis S, Mantsopoulos K, Iro H, Zenk J. Long-term out- comes of endonasal surgery for choanal atresia: 28 years' experience in an academic medical centre. Eur Arch Otorhinolaryngol. 2013 Jan;270(1):113-6. doi: 10.1007/s00405-012-1982-y.
14. Lazar RH, Younis RT. Transnasal repair of choanal atresia using telescopes. Arch Otolaryngol Head Neck Surg. 1995 May;121(5):517-20. doi: 10.1001/archotol.1995.01890050015003.
15. Nithyasundar A, Narayanan DS. Choanal atresia: experience with transnasal endoscopic technique. J Pharm Sci Res. 2016;8(2):86–87.
16. Sadek SA. Congenital bilateral choanal atresia: a novel stenting technique in neonates. Rev Laryngol Otol Rhinol (Bord). 2000;121(1):49-51.
17. Van Den Abbeele T, Francois M, Narcy P. Transnasal endo- scopic treatment of choanal atresia without prolonged stenting. Arch Otolaryngol Head Neck Surg. 2002 Aug;128(8):936-40. doi: 10.1001/archotol.128.8.936.
18. Rodríguez H, Cuestas G, Passali D. A 20-year experience in microsurgical treatment of choanal atresia. Acta Otorrinolaringol Esp. 2014 Mar-Apr;65(2):85-92. doi: 10.1016/j.otorri.2013.09.005.
19. Bartal N. An improved stent for use in the surgical management of congenital posterior choanal atresia. J Laryngol Otol. 1988 Feb;102(2):146-7. doi: 10.1017/s0022215100104360.
20. Richardson MA, Osguthorpe JD. Surgical management of choanal atresia. Laryngoscope. 1988 Sep;98(9):915-8. doi: 10.1288/00005537-198809000-00002.
21. Mantovani M, Mosca F, Laguardia M, Di Cicco M, Pignataro L. A new dynamic endonasal stent for bilateral congenital choanal atresia. Acta Otorhinolaryngol Ital. 2009 Aug;29(4):209-12.
22. Al-Qahtani AS, Messahel FM. Choanal atresia repair. The use of reinforced silicone tube to prevent restenosis. Rhinology. 2003 Mar;41(1):54-5.
23. Ceylan K, Emir H, Kizilkaye Z, Samim E. Bilateral congenital choanal atresia in a 7-day-old patient: transnasal endoscopic repair with stent. Eur Arch Otorhinolaryngol. 2007 Jul;264(7):837-40. doi: 10.1007/s00405-007-0265-5
24. El-Ahl MA, El-Anwar MW. Stentless endoscopic transnasal repair of bilateral choanal atresia starting with resection of vomer. Int J Pediatr Otorhinolaryngol. 2012 Jul;76(7):1002-6. doi: 10.1016/j.ijporl.2012.03.019.
25. El-Anwar MW, Nofal AA, El-Ahl MA. Endoscopic repair of bilateral choanal atresia, starting with vomer resection: evaluation study. Am J Rhinol Allergy. 2016 May;30(3):95-9. doi: 10.2500/ajra.2016.30.4321.