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CASE SERIES

Endoscopic endonasal repair of neonatal bilateral choanal atresia: a case series

António Andrade[1,2], *Pedro Barros*[1,2], *Cecília Pereira*[3], *Gil Coutinho*[1,2], *Ricardo Vaz*[1,4,5,6], *Jorge Spratley*[1,2,5], *and Carla Pinto Moura*[1,7,8,9]

[1]Department of Otorhinolaryngology, Unidade Local de Saúde São João, Porto; [2]Unit of Otorhinolaryngology, Department of Surgery and Physiology, Faculty of Medicine, University of Porto, Porto; [3]Department of Otorhinolaryngology, Unidade Local de Saúde do Alto Minho, Viana do Castelo; [4]Unit of Anatomy, Department of Biomedicine, Faculty of Medicine, University of Porto, Porto; [5]CINTESIS@RISE, Faculty of Medicine, University of Porto, Porto; [6]NeuroGen Research Group, Center for Health Technology and Services Research (CINTESIS), Porto; [7]Department of Human Genetics, São João Local Health Unit, Porto; [8]Department of Genetics, Faculty of Medicine, University of Porto, Porto; [9]RISE-Health, Department of Pathology, Faculty of Medicine, University of Porto, Porto. Portugal

Abstract

Introduction and objectives: Bilateral choanal atresia (CA) is a rare neonatal diagnosis that may cause immediate airway compromise and is frequently impacted by associated malformations. We aimed to describe the surgical management and outcomes of infants with bilateral CA treated with an endoscopic endonasal approach. **Methods:** A retrospective case series included consecutive infants with bilateral CA managed surgically between January 2008 and December 2024. Diagnosis was endoscopically confirmed and imaging was used for operative planning. Surgery followed a standardized endonasal sequence including septal mucosal incision, posterior septectomy, bilateral choanal opening, and mucosal flap repositioning to cover exposed bone. Stents were reserved for significant septal deviation, with planned removal after two to three weeks. Postoperative surveillance included scheduled endoscopic assessments, and balloon dilatation was performed when clinically significant restenosis was detected. **Results:** Six infants were included (four of whom were female). The age at surgery ranged from five to 107 days (mean 29.5 days). Three patients had a syndromic diagnosis (two with CHARGE syndrome and one with Treacher Collins syndrome). Two infants had esophageal atresia with tracheoesophageal fistula, one requiring jejunostomy. Temporary nasal stenting was used in two cases due to septal deviation. In one infant, an additional posterior membranous obstruction was identified intraoperatively. During follow-up, three patients required balloon dilatation for restenosis. One patient underwent an elective tracheostomy for planned craniofacial management. **Discussion:** Endoscopic endonasal repair allows airway patency; however, postoperative surveillance is required, together with the management and control of co-existing comorbid conditions. Where syndromes are involved, bilateral CA may include unexpected anatomic abnormalities that should be anticipated during planning.

Keywords: Choanal atresia. Nasal surgical procedures. Infant. Newborn. CHARGE Syndrome. Mandibulofacial dysostosis.

***Correspondence:**
António Andrade
E-mail: antoniolfandrade@gmail.com

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Cirurgia endonasal endoscópica e abordagem multidisciplinar na atresia coanal bilateral neonatal: uma série de casos

Resumo

Introdução e objetivos: A atresia coanal bilateral é um diagnóstico neonatal raro, podendo causar compromisso imediato da via aérea e sendo frequentemente condicionada por malformações associadas. Pretende-se descrever a abordagem cirúrgica e os resultados de recém-nascidos com atresia coanal bilateral tratados por via endoscópica endonasal. **Métodos:** Série retrospetiva de casos recém-nascidos com atresia coanal bilateral submetidos a cirurgia entre janeiro de 2008 e dezembro de 2024. O diagnóstico foi confirmado por endoscopia e a imagiologia foi utilizada para planeamento operatório. A cirurgia seguiu uma sequência endonasal padronizada, descrita por incisão na mucosa do septo, septectomia posterior, abertura coanal bilateral e reposicionamento de retalhos mucosos para cobertura de osso exposto. A colocação de stents foi reservada para desvio septal significativo, com remoção planeada às 2 – 3 semanas. A vigilância pós-operatória incluiu avaliações endoscópicas programadas, e foi realizada dilatação com balão perante re-estenose clinicamente significativa. **Resultados:** Foram incluídos seis recém-nascidos, quatro do sexo feminino. A idade à data da cirurgia variou entre 5 e 107 dias (média 29,5 dias). Três doentes apresentavam diagnóstico sindrómico (dois com síndrome CHARGE e um com síndrome de Treacher Collins). Dois recém-nascidos apresentavam atrésia esofágica com fístula traqueo-esofágica, um necessitando de jejunostomia. Devido à presença de desvio septal, foi necessária a colocação temporária de *stent* nasal. Num caso foi identificada intra-operatoriamente uma obstrução membranosa posterior adicional. No seguimento, três doentes necessitaram de dilatação com balão por re-estenose. Um doente realizou traqueostomia eletiva para posterior cirurgia craniofacial. **Discussão:** A cirurgia endoscópica endonasal permite a permeabilização da via aérea, mas é necessária vigilância pós-operatória, em conjunto com abordagem e controlo das comorbilidades coexistentes. Em contexto sindrómico, podem existir alterações anatómicas inesperadas que devem ser antecipadas no planeamento.

Palavras-chave: Atresia coanal. Procedimentos cirúrgicos nasais. Recém-nascido. Síndrome CHARGE. Disostose mandibulofacial.

Key points

What is known
- Bilateral choanal atresia causes neonatal airway obstruction and frequently coexists with syndromic or multisystem anomalies.
- Endoscopic endonasal repair is well-established, yet optimal adjuncts such as stenting and restenosis prevention remain debated.
- Restenosis after repair often requires close endoscopic follow-up and occasional dilatation or revision.

What is added
- Standardized endoscopic sequence with mucosal flap coverage achieved patency with selective, short-term stenting reserved only for septal deviation.
- Rare cases of additional posterior membranous obstruction may occur, requiring intraoperative recognition and tailored surgical management.
- Multidisciplinary cooperation is crucial to prioritize the timing of choanal repair alongside other surgeries in medically complex infants.

Introduction

Choanal atresia (CA) is a rare congenital malformation characterized by obstruction of the nasal choanae, resulting in discontinuity between the nasal cavity and the nasopharynx.[1–3] While unilateral forms may remain asymptomatic or present later in childhood with chronic nasal obstruction or rhinorrhea, bilateral CA is a neonatal emergency, as these neonates typically present shortly after birth with respiratory distress, feeding difficulties, and cyclical cyanosis that improves with crying.[1–3]

Several embryologic theories have been proposed to explain the pathogenesis of choanal atresia, including persistence of the buccopharyngeal membrane, persistence of the nasobuccal membrane of Hochstetter, failure of mesodermal resorption in the nasochoanal region, and disruption of neural crest cell migration.[4] These developmental abnormalities may occur alongside other congenital anomalies. Choanal atresia frequently coexists with craniofacial and multisystem anomalies and is often encountered alongside syndromes, particularly CHARGE syndrome.[1,5] It may also be associated with cardiovascular, gastrointestinal, and auditory abnormalities, which can impact management and outcomes.[3] Diagnosis is initially suspected clinically and by the inability to pass a suction catheter through

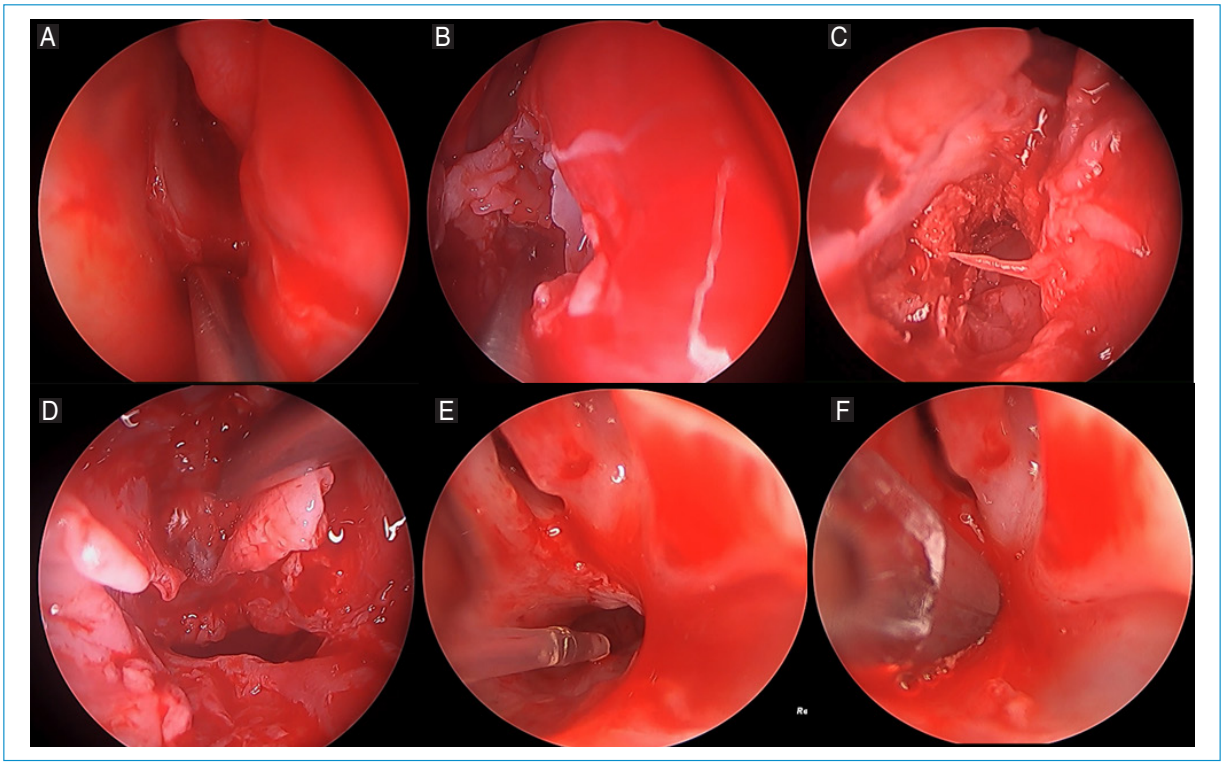

Figure 1. Endoscopic endonasal repair of bilateral choanal atresia. The surgery begins with a septal mucosal incision, followed by posterior septectomy **(A and B)** and bilateral choanal opening **(C)**. In this case, an additional posterior membranous obstruction was identified (black star) and subsequently excised. Mucosal flaps were repositioned to cover exposed bone, maintaining patency of the newly created neochoana **(D)**. Temporary stents were used selectively in cases requiring the closed reduction of septal deviation and were removed after two to three weeks. During follow-up, when choanal restenosis was detected, balloon dilatation was performed **(E and F)**.

the nasal cavity.[6] Nasal endoscopy confirms the site of obstruction, and computed tomography characterizes the atretic plate and guides surgical planning.[1]

Surgical repair is the definitive treatment for CA, and over recent decades, endoscopic endonasal approaches have largely replaced traditional transpalatal or transeptal techniques, offering improved visualization and reduced morbidity.[1,7,8] Postoperative care and serial follow-up are essential, as restenosis may develop and compromise airway patency.

Given the rarity of bilateral CA and the heterogeneity of clinical cases and comorbidities, published evidence largely consists of small retrospective series. Therefore, our goal is to evaluate infants diagnosed with bilateral CA that underwent endoscopic endonasal surgery, focusing on associated comorbidities and their management and outcomes.

Methods

This retrospective case series included consecutive infants with bilateral congenital choanal atresia managed at a tertiary university hospital between January 2008 and December 2024, identified through the electronic medical record. Exclusion criteria were unilateral CA and incomplete operative or follow-up data.

Diagnosis followed routine clinical practice for neonatal nasal obstruction, including the endoscopic confirmation of posterior nasal obstruction. Computed tomography was obtained for further evaluation and operative planning.

Data was abstracted from clinical records, including sex, age at surgery (days), stent placement, postoperative balloon dilatation (BD), and status at last follow-up at December 2024. Comorbidities, syndromic diagnoses, and related surgeries were documented. Ethics committee approval was obtained.

Operative management followed a standardized endonasal sequence (Fig. 1). After the induction of general anesthesia, a septal mucosal incision was made and a posterior septectomy performed to create a common posterior corridor. The atretic plate was first approached at the hard palate–vomer junction, where

Table 1. Patients' characteristics

Sex	Age at surgery (days)	Associated comorbidities and syndromes	Stenting	Balloon dilatation	Follow-up
Female	7	CHARGE: right ear malformation, coloboma, microphthalmia, inner ear malformation	✓		Uneventful
Male	5	Treacher Collins syndrome, renal atrophy		✓	Tracheostomy
Male	107	Esophageal atresia with tracheoesophageal fistula, severe gastroesophageal reflux, multicystic kidney, bronchopulmonary dysplasia		✓	Death
Female	12	None			Uneventful
Female	40	CHARGE: coloboma, esophageal atresia with tracheoesophageal fistula; growth hormone deficiency, patent ductus arteriosus		✓	Uneventful
Female	6	None	✓		Uneventful

it was gently perforated and then progressively widened. The plate was then opened bilaterally to create a symmetric neochoana. Wherever feasible, mucosal flaps were mobilized to cover exposed bone along the vomer and lateral nasal wall to limit granulation and reduce the risk of restenosis. Stenting was not routine. Stents were reserved for cases requiring the closed reduction of a significant septal deviation, with planned removal at two to three weeks.

In postoperative care and surveillance, infants were followed by the otorhinolaryngology team with scheduled endoscopic assessments. When clinically significant restenosis was observed on follow-up, these cases were referred to endoscopic BD.

Data was analyzed with IBM SPSS Statistics, version 27. Continuous variables were summarized using mean with standard deviation and median with interquartile range, and categorical variables were summarized as absolute frequencies and percentages.

Results

Six infants with bilateral osteo-membranous CA were included, and outcomes were available for all six patients through to the end of follow-up. Patient characteristics are summarized in table 1. Four patients were female (66.7%), and the age at surgery ranged from five to 107 days (mean 29.5 days). A syndromic diagnosis was recorded in three out of six cases (50%), with CHARGE syndrome (n = 2) and Treacher Collins syndrome (n = 1). Two infants had esophageal atresia with tracheoesophageal fistula, treated surgically in the first days of life. One of these required a jejunostomy.

All patients underwent endonasal endoscopic repair, as illustrated in figure 1. Temporary stenting was used selectively in two out of six cases (33.3%) in the presence of septal deviation, and stents were removed after two to three weeks. One neonate with Treacher Collins syndrome underwent an elective tracheostomy to facilitate planned orthognathic procedures, including mandibular distraction. In this same patient, after completion of the standard choanal opening, an additional posterior membranous obstruction was identified intraoperatively, which had not been described preoperatively. This finding was considered consistent with a possible remnant of the embryologic nasobuccal membrane, creating a second obstructing plane posterior to the atretic plate at the level of the nasopharyngeal inlet above the soft palate (Fig. 2).

During follow-up, three out of six infants (50%) required revision with BD due to choanal restenosis (Fig. 1E and F). One patient, born with esophageal atresia, presented severe gastroesophageal reflux (GER) that remained difficult to control despite medical therapy with a proton pump inhibitor and a histamine-2 receptor antagonist. In this case, choanal restenosis appeared to coincide with episodes of reflux exacerbation. By the end of follow-up, one death had occurred (one out of six, 16.7%).

Discussion

Bilateral CA is an uncommon but clinically significant neonatal diagnosis, as airway compromise may be immediate and the clinical course is often strongly impacted by the overall medical complexity. It should therefore be understood not as one operative event, but as a longitudinal process in which perioperative decision-making, careful postoperative surveillance,

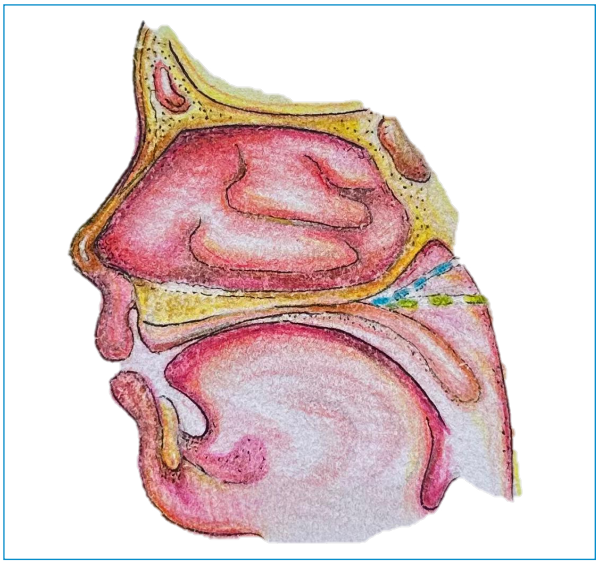

Figure 2. Posterior membranous remnant. Schematic sagittal view of the nasal cavity and nasopharynx illustrating an additional posterior membranous remnant at the nasopharyngeal inlet, creating a second obstructing plane posterior to the choanal atresia. The blue and green markings indicate two plausible intraoperative configurations.

and timely conservative revisions together determine durable patency.

Additionally, bilateral CA frequently lies within a broader malformation context. Extensive reviews describe substantial rates of associated airway and craniofacial abnormalities, including CHARGE and Treacher Collins syndrome, and emphasize that these infants may present with multilayer airway vulnerability and multiorgan involvement.[9,10] This multidisciplinary framing is not merely theoretical: comorbidity-directed priorities can directly impact the timing and sequencing of interventions. This case series illustrates this principle through infants requiring the early correction of esophageal atresia with tracheoesophageal fistula and the use of elective tracheostomy as part of a planned craniofacial pathway. These scenarios reinforce that, although CA repair is performed within pediatric otolaryngology, its management is intrinsically multidisciplinary.

At the anatomical level, bilateral CA is rarely a simple "plate" obstruction. Endoscopic and imaging typically demonstrate a distorted posterior nasal framework, classically including thickening of the anterior portion of the pterygoid plates and enlargement of the posterior vomer, with a variable membranous component.[1] Even with careful imaging and endoscopic assessment, malformation contexts should prime teams to anticipate atypical findings. As illustrated in this case series, an additional posterior membranous obstruction plane was found in one infant after completing the standard neochoana. This can be interpreted within embryologic theories described for CA, including abnormal persistence of primordial membranes.[11] Therefore, anticipating possible additional malformations is important in order to plan appropriate interventions.

Restenosis and the need for dilatation were observed primarily in syndromic cases in our series, as all the infants who required dilatation came from that context. Although exacerbation of GER was associated with CA restenosis in one patient, identifying just one predictor of restenosis is challenging, as outcomes likely reflect the combined effect of the syndromic background, age at repair, baseline anatomic constraints (bony/membranous components), and postoperative inflammatory response rather than any isolated factor.[12,13]

As described by Stamm and Pignatari, the endoscopic flap-based technique aims to preserve and reposition mucosal flaps to facilitate re-epithelialization, reduce exposed bone, and enable close postoperative endoscopic surveillance.[7,8]

The role of stents remains controversial regarding improved long-term patency and is applied inconsistently across series.[8,14] In our experience, stents were used only to optimize the correction of septal deviation rather than to maintain the patency of the neochoana.

Balloon dilatation has emerged as a minimally invasive adjunct in the management of choanal atresia and was first reported for choanal atresia/stenosis in 2011.[15] The concept was extrapolated from its prior use in subglottic and tracheal stenosis. In contemporary practice, BD is performed transnasally under endoscopic guidance. A balloon catheter is positioned across the stenotic neochoana and inflated to deliver controlled radial force, thereby enlarging the lumen while minimizing shear trauma to the surrounding mucosa. Reported advantages include reduced mucosal injury, shorter operative time, and the potential to avoid prolonged stenting.[15,16] Available retrospective studies suggest that BD may improve airway patency, particularly in cases of mild to moderate restenosis, although repeat dilatations may be required.[16] In the present series, BD proved to be a useful and reproducible option for the treatment of postoperative restenosis. Despite these encouraging reports, the evidence remains heterogeneous and predominantly retrospective. Prospective controlled studies are needed to establish optimal patient selection criteria, standardize dilatation parameters, and evaluate long-term efficacy compared with alternative strategies, including stenting and other adjunctive pharmacotherapy.

Topical mitomycin C (MMC) was not used in our series. Although MMC has been described as an adjunct in choanal atresia surgery, the available evidence for benefit in bilateral choanal atresia is weak and largely retrospective, and a meta-analysis concluded that there is insufficient data to determine whether MMC is a useful adjunct.[14,17] While MMC appears to be generally safe, current evidence does not support its routine use in this setting.[14,17]

This study has important limitations. It is a retrospective observational case series with a small sample size, as expected for a rare neonatal condition, which limits external generalizability. Syndromic heterogeneity and variable comorbidity profiles further constrain risk factor analysis and reduce the generalizability of the findings.

In conclusion, endoscopic endonasal repair with mucosal flap coverage is an effective technique for the management of bilateral choanal atresia, with selective short-term stenting reserved mainly for cases with septal deviation. Syndromic disease and major comorbidities require a multidisciplinary framework and careful operative planning, as they may also be associated with unanticipated intraoperative anatomical findings that should be considered during preoperative assessment and counselling. Given the ongoing risk of restenosis and other complications, structured postoperative endoscopic surveillance is essential to detect early narrowing and to determine the need for conservative revision or reintervention.

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Ethical considerations

Protection of human subjects and animals. The authors declare that no experiments on humans or animals were performed for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from all patients, and secured approval from the Ethics Committee. SAGER guidelines have been followed as applicable to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing or creation of the content of this manuscript.

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