



A rare case report of bilateral choanal atresia in an adult

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ABSTRACT

INTRODUCTION: Adult choanal atresia is a rare congenital malformation of the nasal cavity characterized by the complete obliteration of the posterior choanae, only ninth cases are reported in the literature, we present in our observation the tenth case of adult bilateral choanal atresia.

PRESENTATION OF CASE: A 18-year-old man presented at our department with complaints of nasal obstruction, nasal discharge, snoring, anosmia, and mouth breathing since childhood. Endoscopic examination and paranasal sinus tomography revealed bilateral choanal atresia. An endoscopic choanoplasty was performed. Follow-up evaluation at postoperative 12th month showed that his symptoms improved significantly and, on endoscopic examination, both choanae remained patent.

DISCUSSION AND CONCLUSION: Adult bilateral choanal atresia is a rare entity. The revelation in adulthood of congenital bilateral atresia remains exceptional. Nasal endoscopy and preoperative computed tomography scan help in planning surgery. Endoscopic transnasal choanoplasty is the criterion standard treatment. The exact role of the postoperative stent and use of mitomycin C is controversial.

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1. Introduction

Choanal atresia is a rare congenital malformation of the nasal cavity characterized by the complete obliteration of the posterior choanae. In contrast to the unilateral form, bilateral choanal atresia is a life-threatening condition.

Bilateral disease is rarely seen in adults. An only nine reported cases to date [1,2]. We present a tenth case of congenital bilateral choanal atresia in a 18-year-old man and describe the clinical features, evaluation, and surgical steps.

This work has been written in accordance with the SCARE criteria [3].

2. Case report

A 18-year-old north African man presented at our department with complaints of nasal obstruction, nasal discharge, snoring, anosmia, and mouth breathing since childhood, there was a history of episodes of cyanosis in childhood, with recovery on crying, and frequent hospital admissions for recurrent pneumonia. There was no history of trauma or nasal surgery. Clinical examination revealed prognathism, hypoplasia of the maxilla, malocclusion with Class

III. There were no other congenital anomalies. Endoscopic examination and paranasal sinus tomography revealed bilateral choanal atresia (Figs. 1 and 2)

An endoscopic choanoplasty was performed in which the bony atresia was removed, and drilling of the vomer and medial pterygoid plates was performed. A bilateral nasal pack was kept for 4 days. No stenting was done. The patient was discharged the next day after surgery and recommended to daily irrigate the nasal cavity with saline. Follow-up evaluation at postoperative 12th month showed that her symptoms improved significantly and, on endoscopic examination, both choanae remained patent (Fig. 3).

3. Discussion

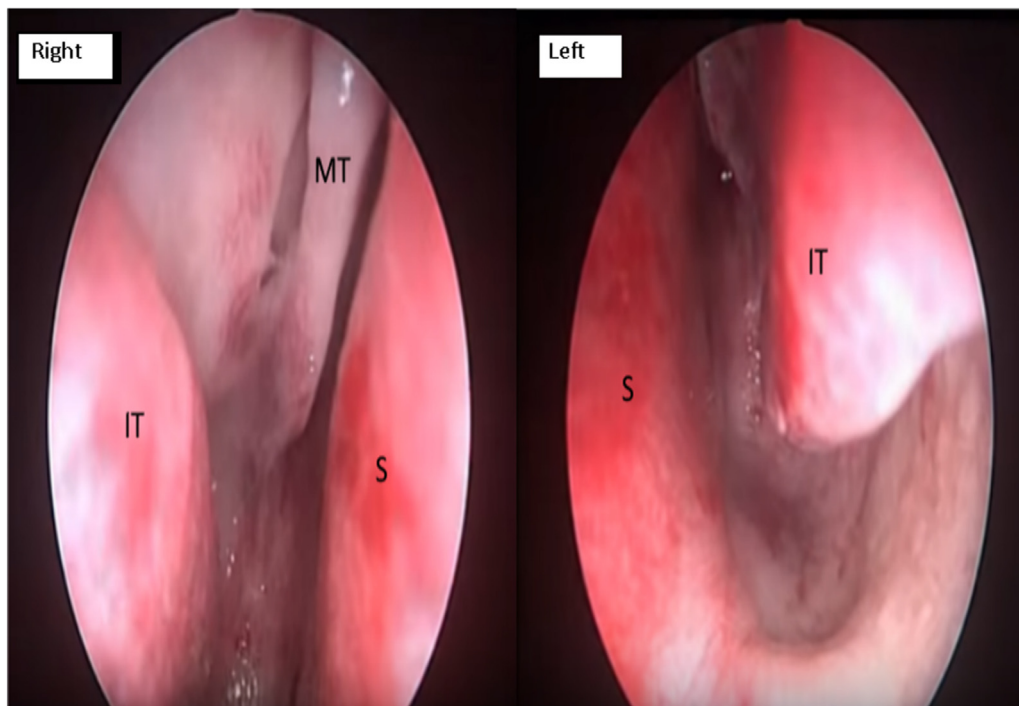
Choanal atresia is a congenital malformation which consists of total or subtotal obstruction of the posterior orifices of the nasal cavity. It is a rare affection, seen in 1 out of 5000–8000 births, it is associated with other congenital abnormalities in 50% of the cases [2,4].

The bilateral form represents a diagnostic and therapeutic neonatal emergency.

Choanal atresia is a frequent component of congenital disorders such as CHARGE, Treacher Collins, Crouzon and Pfeiffer syndromes. However, the detailed cellular and molecular mechanisms underpinning the etiology and pathogenesis of CA remain elusive [5]. Last study discovered that mutations in retinol dehydrogenase 10 (Rdh10), which perturbs Vitamin A metabolism and retinoid signaling, exhibit fully penetrant choanal atresia [5].

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IT, inferior turbinate; MT, middle turbinate; S, septum.

Fig. 1. XXX.

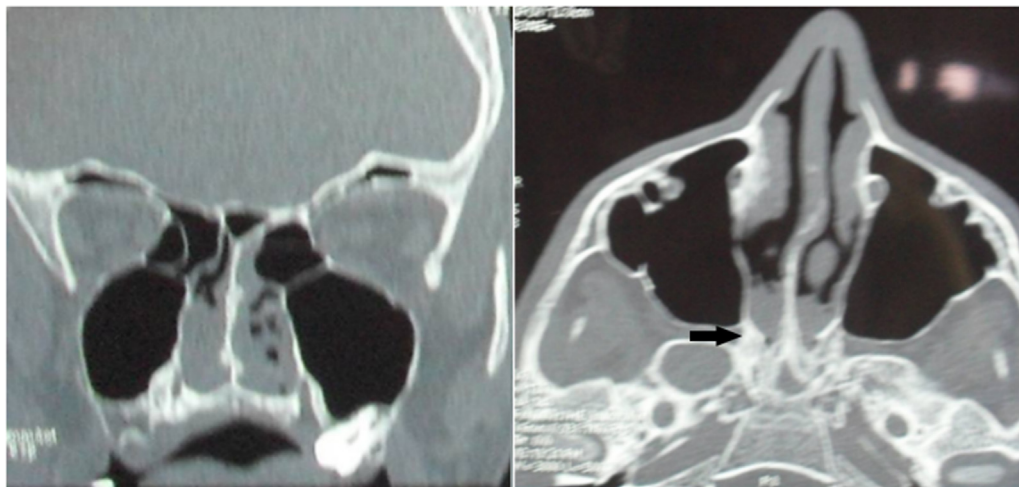


Fig. 2. XXX.

The revelation in adulthood of congenital bilateral atresia remains exceptional in partial or neglected forms in our case we report a bilateral choanal atresia [2].

The atresic plate is located at the palatomaxillary junction in front of the posterior border of the vomer and the palate. The atresic plate has a thickness varying from 1 to 12 mm [4].

The choanal atresia takes various aspects on the anatomical plane: Single or bilateral atresia; bone, membranous or mixed; Symmetrical Shrinkage or not, predominant or not on one wall or the other [6].

Transnasal choanoplasty was performed in our patient. It began with choanal repermeabilization using a microdebrider followed by an enlargement of the posterior part of the nasal cavity interesting the vomer bone whose resection of its posterior border is the most

effective gesture to obtain a large and stable orifice. The mucous plane is reconstituted to prevent restenosis.

Repair of choanal atresia has evolved significantly over the past decades. Open trans-palatal techniques have given way to minimally invasive endoscopic repair [7]. The role of a stent is controversial. Some investigators advocate its supremacy [8]. Novel application of steroid eluting sinus stent in minimizing scarring following endoscopic repair of choanal atresia with a high level of success [7]. Aksoy et al. used mitomycin C after choanoplasty to prevent stenosis [9].

In our case, neither of these adjuvants were used. During the postoperative period, the nasal package was removed after 4 days, and the patient started with a nasal douching for 3 weeks.

Of the nine documented cases of adult bilateral choanal atresia in literature, only one patient had restenosis, requiring revision

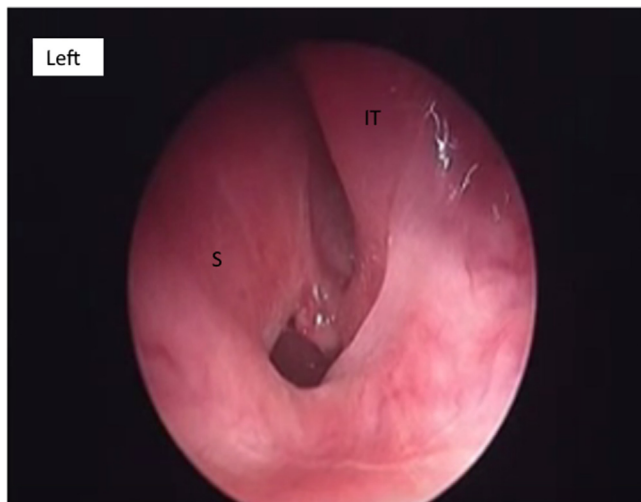


Fig 3. XXX.

surgery [10]. Our patient was followed up for 12 months, and the nasal endoscopy revealed a good evolution with well-patent posterior.

4. Conclusion

The bilateral choanal atresia is considered incompatible with life. Early surgery in the neonatal period is necessary for survival. Adult bilateral choanal atresia is a rare entity. Nasal endoscopy and preoperative computed tomography help to guide the surgical procedure. Endoscopic transnasal choanoplasty is considered the standard therapeutic choice. Postoperative restenosis remains a frequent postoperative complication. The exact role of the postoperative stent and use of mitomycin C is controversial.

Conflicts of interest

The authors declare having no conflicts of interest for this article.

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Ethical approval

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Author contribution

Said Anajar: Corresponding author writing the paper
Jawad Hasnaoui: writing the paper
Reda Abada: study concept
Sami Rouadi: study concept
Mohammed Roubal: correction of the paper
Mohammed Mahtar: correction of the paper

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