

Choanal atresia: Diagnosis and management

Atrésie des choanes: diagnostic et prise en charge thérapeutique

I. Achour ^{1,2}, I. Kharrat ^{1,2}, M. Ben Ayed ^{1,2}, W. Thabet ^{1,2}, A. Bouraoui ^{2,3}, M. Mnejja ^{1,2}, B. Hammami ^{1,2},
A. Chakroun ^{1,2}, I. Charfeddine ^{1,2}

1 Department of Otorhinolaryngology-Head and Neck Surgery, Habib Bourguiba University Hospital, Sfax, Tunisia

2 Sfax Medical School, University of Sfax, Sfax, Tunisia

3 Department neonatology, Hedi Chaker University Hospital, Sfax, Tunisia

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ABSTRACT

Objective: Choanal atresia (CA) is a rare congenital malformation caused by the obliteration of the posterior choanae by an atretic plate. The aim of our study is to describe the diagnosis and management modalities of CA and to determine the factors associated with recurrence.

Materials and methods: This is a retrospective study based on the medical records of patients with CA managed in our department in the period between 2002 and 2021. We studied the clinical features and management modalities of each patient. For patients who developed a recurrence, we determined the factors associated with recurrence based on a bivariate analysis.

Results: We studied the medical records of 26 patients with either a bilateral (n=8) or a unilateral (n=16) form of CA. The median age at surgery was two days for bilateral forms and 5 years and 4 months for unilateral forms. At computed tomography scan, CA was mixed (n=20), bony (n=4) or membranous (n=2). All patients underwent intranasal endoscopic surgical treatment using cold instruments alone in membranous forms and combined to the drilling of the atretic plate in bony and mixed forms. The surgical management included the resection of the posterior part of the vomer bone and the placement of nasal stents in 10 and 16 patients respectively. We recorded 6 cases of recurrence requiring a surgical re-intervention. The presence of associated cranio-facial malformations was the only factor associated with recurrence (p=0,001).

Conclusion: Choanal atresia diagnosis was based on nasal endoscopy and CT scan. Surgical treatment using transnasal endoscopic approach was an effective and safe technique. Associated local malformations was a factor associated with re-stenosis.

Key words : Choanal atresia, management, endoscopic surgery, recurrence

RÉSUMÉ

Objectifs: L'atrésie des choanes est une malformation congénitale causée par l'oblitération des choanes postérieures par une plaque atrétique. Les buts de notre étude sont de décrire les modalités du diagnostic et de la prise en charge thérapeutiques de l'atrésie des choanes et de déterminer les facteurs associés à la récurrence.

Matériels et méthodes: Il s'agit d'une étude rétrospective qui s'est basée sur les données des patients ayant un diagnostic confirmé d'une atrésie des choanes et qui ont été pris en charge dans notre service dans la période entre 2002 et 2021. Nous avons déterminé les présentations cliniques et les modalités de prise en charge pour chaque patient. Dans les cas de re-sténose, nous avons déterminé les facteurs associés à la récurrence par une étude bivariée.

Résultats: Nous avons étudié les dossiers de 26 patients ayant une atrésie des choanes soit bilatérale (n=8) soit unilatérale (n)=18. L'âge médian lors de la chirurgie était de deux jours pour les formes bilatérales et de 5 ans et 4 mois pour les formes unilatérales. A la tomodensitométrie, l'atrésie était mixte (n=20), osseuse (n=4) ou membranaire (n=2). Tous les patients étaient traités par voie endonasale sous guidage endoscopique en utilisant soit les instruments froids seuls dans les formes membraneuses, soit en association au fraisage de la plaque atrétique dans les formes mixtes et osseuses. La chirurgie a inclus également la résection de la partie postérieure de l'os vomer et la mise en place de sonde de calibre intra nasale chez 6 et 16 patients respectivement. Nous avons noté 6 cas de récurrence ayant tous nécessité une ré-intervention. La présence d'autres malformations crânio-faciales était le seul facteur associé au risque de récurrence (p=0,001)

Conclusion: Le diagnostic de l'atrésie des choanes est basé sur l'endoscopie nasale et la tomodensitométrie. La chirurgie par voie endonasale était un traitement efficace. La présence d'autres malformations était le seul facteur associé à un risque de récurrence dans notre étude.



INTRODUCTION

Choanal atresia (CA) is a congenital obstruction of the posterior choanae by the presence of an atretic plate totally blocking the communication between the nasal cavity and the nasopharynx due to the persistence of the bucco-pharyngeal membrane [1,2]. CA has an estimated incidence that varies between 1/5000 and 1/9000 live births in the world [3,4]. CA can be either unilateral or bilateral. The obstruction can be mixed, bony or rarely membranous. CA is frequently associated to other syndromic or non-syndromic malformations. Isolated forms of CA are less common but could reach 50 % in some series [5,6]. Infants, with bilateral atresia, present with a neonatal respiratory distress; bilateral forms are considered as diagnostic and therapeutic emergencies. Unilateral forms are more tolerated; they present with delayed unspecific symptoms and could be diagnosed at a late age [1]. Nasal endoscopy confirms the presence of choanae obstruction. CT confirms CA diagnosis, determines the composition of the atretic plate, confirms the presence of associated malformations and rules out differential diagnosis [7,8]. CA management includes securing upper airways, screening for the presence of other syndromic and non-syndromic malformation as well as surgically restoring nasal air flow. The management of CA is based on different approaches: mainly transnasal or less commonly transpalatine, transantral, or transeptal [4,9]. One of the most frequent concerns following CA surgery is re-stenosis that could indicate a revision surgery.

The aim of our study is to describe and analyze clinical and imaging characteristics of CA, as well as different surgical treatment results of patients with CA treated in our department. We also aim to determine the different factors that could influence surgical results.

Materials and methods:

1. Patients and study design

This is a retrospective study based on the analysis of medical records of patients with a choanal atresia managed between 2002 and 2021. We included patients with a confirmed CA diagnosis based on clinical and radiological findings and who underwent initial surgical treatment in our department.

For each patient, we recorded age at diagnosis, sex, detailed medical record and clinical manifestations. A nasal catheter insertion was performed followed by a nasal endoscopic examination. A facial CT scan was performed for all patients with axial, coronal and sagittal reconstructions. It determined the imperforation nature, either bony, membranous or mixed as well as the unilateral or bilateral involvement. We identified associated cranio facial malformations and anatomical elements taking part in the atresia: Nasal cavity width, atretic plate thickness, medialization of the pterygoid process and the perpendicular plate of the palatine bone, the vomer thickness and the nasopharynx depth. Cardiac and abdominal ultrasounds were performed prior to any surgical treatment to screen for associated congenital abnormalities.

2. Surgical technique

Surgery was performed under general anesthesia with an oro-tracheal intubation. Nasal packing with adrenaline solution was performed to insure nasal mucosa retraction. Vasoconstrictors were not used for patients under 15 years-old. We used 30-degree and 0-degree endoscopes with diameters from 2.7 to 4 mm. A perforation was performed directly through the atretic plate using laser or cold instruments using endonasal suction tube starting in the inferior and medial part. It allowed a control of nasopharyngeal structures serving as landmarks for choanal enlargement. The atretic plate was then drilled; starting at the inferior and medial portion, reaching the roof of the nasopharynx and its lateral wall. Drilling of the lateral part was carefully done as it can cause a lesion of the sphenopalatine artery. The posterior part of the vomer bone was then removed using either drilling or a retrograde nasal cutting forceps. Nasal stenting was performed using intubation tube; its diameter corresponded to that of an intubation tube adapted to the patient's age. Mucosa was regularized using cold instruments in order to prevent post-operative synechia. Post-operative treatment included repetitive saline solution irrigation, antibiotics and local corticosteroids.

3. Follow up, recurrence and revision surgery

Our patients underwent monthly follow up in order to detect complications, granulation tissue and re-stenosis. The duration of stenting and endoscopic examination findings were noted. A re-stenosis was defined by the recurrence of symptoms as well as the atretic place at the endoscopic examination [10]. If a recurrence was confirmed, the complete or incomplete character, the timing and the type were recorded. We also performed a bivariate analysis in order to determine factors associated with an increased risk of recurrence following the surgical management of CA. For patients who underwent a revision surgery, timing of surgery, operative technique, follow up duration and results were noted.

4. Statistical analysis

Descriptive and analytical statistical analysis were performed using the SPSS 24.0 software for Windows. For qualitative variables we used either Chi-Square test or Fisher's exact test in the case or more than one cell had an expected count of less than 5%. On the hand, for qualitative and quantitative variables we used either Student T test for normally distributed variables and Mann-Whitney U test in the remaining cases. A p value of < 0.05 was considered as statistically significant.

RESULTS:

We identified a total of 26 patients with CA with a median age of management of 2 days [1-90 days] for bilateral forms compared to 5 years and 4 months for unilateral forms [3months- 39years]. CA was diagnosed at birth in all bilateral forms and in 50 % of unilateral forms (nine patients) while 9 patients presented delayed symptoms ranging between 1 month and 39 years. We noted a

male-to-female ratio of 1. All patients suffering from a bilateral choanal atresia (eight cases) presented with a neonatal respiratory distress syndrome and were all managed with the placement of a Guedel cannula. The most reported symptom was rhinorrhea and unilateral nasal obstruction (88.7%) followed by snoring (44.4 %). Flexible nasal endoscopy examination revealed a unilateral choanal obliteration in 18 patients with a right-side predominance (83.3%) and a bilateral form in 8 patients.

We recorded the presence of cranio-facial malformations in six patients (Table I). There was not a significant association between the presence of local malformations and the unilateral/bilateral form of the CA. (p=0.135)

CT scan was performed in all cases. It confirmed the CA diagnosis, the affected side and the imperforation nature in all cases. Mixed atresia was the most frequent type (n=20) followed by pure bony (n=4) and membranous imperforations (n=2). (figure 1) CT showed other associated abnormalities: a hypoplasia of the maxillary sinus and the inferior turbinate (n=1), a hypertrophied contralateral inferior turbinate (n=4) and a deviated septum (n=1).



Figure 1: Facial CT scan , axial bone window, right mixed choanal atresia with lateral deviation and thickness of the posterior part of the vomer bone (thick arrow), medialization of the pterygoid process (thin arrow) , a central membranous part (asterisk)

All patients underwent a surgical treatment using an intranasal endoscopic approach. Surgical techniques depended on the atresia type; using either cold instruments alone or associated to drilling; with/without the resection of the posterior part of the vomer bone (Table II).

Table II: Surgical techniques depending on the atresia type

| | Local malformations | | |
|-------------------------|---------------------|---------------|--|
| | Number | Type | |
| Unilateral forms (N=18) | N=5/18 | Syndromic | CHARGE syndrome (n=1) Pierre Robin syndrome (n=1) |
| | | Non-syndromic | Pseudohermaphroditism Ogival palate |
| Bilateral forms (N=8) | N=1/8 | Non-syndromic | Cleft lip |

Table II: Surgical techniques depending on the atresia type

| | Bony | Membranous | Mixed | Total | Nasal stents |
|---|------|------------|-------|-------|--------------|
| Cold instruments | 0 | 2 | 0 | 2 | 2 |
| Drilling | 3 | 0 | 11 | 14 | 14 |
| Drilling with resection of the posterior part of the vomer bone | 1 | 0 | 9 | 10 | 0 |

In order to avoid restenosis, at the end of the intervention, nasal stents were placed in 16 patients using intubation tubes adapted to the patient's age and the choanal dimensions. The mean duration of nasal stenting for bilateral and unilateral forms was respectively 142 [30-253 days] and 61 days [21-121 days].

The mean length of hospitalization was two days. All patients received post-operative antibiotics with nasal irrigations. Anti-inflammatory drugs were prescribed for seven patients. We noted two cases of epistaxis and one patient died due to neurological anoxia complications.

The mean duration of follow-up was 11 months. Two patients developed a post-operative turberinoseptal synechia while we noted an infection of the nasal fossa in 2 patients with nasal stents and a case of a moderate asymptomatic restenosis. During the follow up, after a mean period of 7.5 months [1-28 months], we recorded six cases of recurrence (23.08 %) that were suspected due to respiratory signs. Restenosis was confirmed by a flexible fibroscopic nasal examination. Recurrences were more frequent in the mixed (n=3) and bony types (n=2). Its percentage varied depending on the surgical technique as detailed in (table III). All 6 patients required a re-operative management that was successful in 83.33 % of cases. We noted 3 episodes of recurrence in one case that required surgical management twice before losing follow up. Global success rate of endonasal endoscopic surgery in our series was 73.07 %, reaching 92.31 % after reoperations.

We studied the factors associated with re-stenosis excluding the patient who died in the immediate post-operative period. We noted that the presence of malformations was associated with recurrence. However, the use of nasal stents as well as the duration of their placement, and the used surgical technique were not associated with recurrence. (Table III).

Table III: Factors associated with recurrence

| Factor | Percentage of recurrence | P value |
|--|--------------------------|---------|
| Choanal atresia type | | |
| - Bony | 2/4 (50%) | 0.234 |
| - Membranous | 1/2 (50%) | 0.430 |
| - Mixed | 3/19 (15.78%) | 0.125 |
| CA laterality | | |
| - Unilateral | 5/17 (29.41%) | 0.624 |
| - Bilateral | 1/8 (12.5%) | 0.624 |
| Surgical technique | | |
| - Drilling and the resection of the posterior part of the vomer bone | 4/10 (40 %) | 0.175 |
| - Cold instruments alone | 2/10 (50 %) | 0.430 |
| - Drilling without the resection of the posterior part of the vomer bone | 1/14 (7.14%) | 0.073 |
| Malformations | 5/6 (83.33%) | 0.001 |
| Nasal stents | 4/16 (25%) | 0.545 |

DISCUSSION:

Choanal atresia is a congenital obstruction of the posterior choanae of the nasal cavity totally blocking communication between the nasal cavity and the nasopharynx [1]. Its incidence varies between 1/5000 and 1/9000 living births in the world [3,4] .



Mean age is widely variable in literature; it depends on the CA type [11]. We reported a median age of 2 days for bilateral forms; one patient was only treated at the age 90 days due to the presence of associated malformations and complications making general anesthesia non possible. Unilateral forms are more common than bilateral ones with a reported percentage of unilateral forms of 60% [12,13]. CA, could be associated to other syndromic or non-syndromic abnormalities in nearly 50 % of cases; predominantly in bilateral forms. [5,7] CHARGE syndrome is the most commonly associated condition with a percentage that varied between 4% and 29 % [5–7,14,20]; it was noted once in our study. Pleiffer, Crouzon and Treacher Collins syndromes are less commonly reported [6–8].

Clinical manifestations and their onset timing depend on the CA type. Bilateral forms manifest early in the neonatal period immediately after birth or at the first post-natal hours, with cyclic respiratory distress and cyanosis. As newborns are obligate nasal breathers, respiratory distress characteristically worsens at feeding and decreases during crying [4,17]. If not treated, evolution could be fatal in few hours requiring oro-tracheal intubation or even tracheostomy. Bilateral forms with a late presentation are exceptional and limited to case reports in literature [18]. All bilateral forms in our series developed a neonatal respiratory distress. Unilateral CA diagnosis is delayed as they could remain asymptomatic and be diagnosed at a later age even during adulthood. They manifest with unspecific signs mainly chronic unilateral nasal obstruction and mucoid rhinorrhea. They could also be suspected at birth via the nasal catheter test [4]. Unilateral rhinorrhea, snoring and nasal speech dominated unilateral CA symptoms in our series.

Choanae permeability can be evaluated in newborns using the nasal catheter test [4,19]. However, there were reported cases of false negatives mainly to the nasal tube winding in the nasal cavity or false positives due to an error in the insertion technique [20].

Flexible nasofibroscope or nasal rigid endoscopy allows a direct visualization of the atretic plate to confirm CA diagnosis and rule out other possible causes of nasal obstruction [17,20]. It confirmed CA diagnosis in all our patients as well as its laterality.

CT scan is indicated in all patients for definitive CA diagnosis. CT determines the obstruction type: bony, membranous or mixed, which helps to prepare for the adequate surgical management. It also rules out the presence of associated cranio-facial malformations [17,20]. CT typically reveals: a narrowing of nasal cavity, thickening of the posterior portion of the vomer bone and its inclination towards the nasal cavity, thickening of the medial plate of pterygoid bone, and a medialization of the lateral nasal wall, with or without a membranous central connection [7,21,22]. CT confirmed the CA diagnosis in all our patients and CA type; with a mixed form predominance (77.27%) followed by bony forms (13.63 %). Similarly to our findings, latest studies reported mixed forms predominance followed by bony CA. Membranous obstruction is rare [8,14,23].

CT also helps determine CA consequences on adjacent structures. CA is believed to cause maxillary sinus hypoplasia and a decreased nasal complex development [14,21]. Other studies showed that there is not an association between the absence of a nasal air flow and the development of para nasal sinuses [24]. CT revealed a hypoplasia of maxillary sinus and inferior turbinate in one of our patients.

Initial management of CA especially in bilateral forms with a respiratory distress first includes securing upper airways via the oral route. It requires Guedel cannula and a nasogastric tube to provide feeding. Oro-tracheal intubation or tracheostomy could be used in cases of persistence of the respiratory distress or the presence of associated malformations and comorbidities [4,9,17].

Associated malformations are known to worsen CA prognosis and could be life threatening with general anesthesia; screening for associated malformations is important prior to any surgical treatment to improve outcomes [20,25,26]. The international Pediatric Otolaryngology Group (IPOG) recommends a CHARGE syndrome evaluation depending on risk factors and clinical findings; cardiac ultrasound indicated prior to surgical treatment while audiological testing is recommended for all CA cases [20].

Surgical treatment aims to restore normal nasal air flow and normal growth of adjacent structures [19]. Timing of surgery is still controverted in literature; it depends on patients' symptoms, CA type and associated abnormalities [23]. Murray et al. [11] concluded in their meta-analysis that immediate surgery insures an immediate relief of respiratory distress and is associated with increased restenosis rate but not with increased second surgery rates. Delayed surgery could be suggested in cases of asymptomatic CA [11]. For bilateral forms, the IPOG recommends surgery whether shortly after diagnosis, or after screening for cardiac abnormalities [20]. The treatment of unilateral forms can be delayed; the IPOG recommends to delay surgery after the age of 6 months; it is also recommended to manage CA before the age of 5 years-old [20]. In our study, bilateral forms were treated after a mean period of 32 days while it varied from 3 months to 39 years in unilateral forms depending on clinical manifestations.

Various surgical approaches were reported. Transnasal endoscopic approach is the gold-standard for definitive repair of CA [4,23,27]. This technique allows an access via the natural orifice, a direct visualization of the obliteration, a precise excision of the atretic plate, without an excision of the palatal fibromucosa [28]. The surgical technique can be based either on the atretic plate drilling, mucosal microdebridement, or less commonly balloon dilation, or laser [20]. Transnasal resection of the atretic plate is based on: drilling of the atretic plate, resection of the posterior part of the vomer and covering all raw surfaces with mucosal flaps in order to avoid re-stenosis [15,27,29]. Other authors recommended a single perforation followed by dilatation and stenting [30]. Laser was also reported in previous studies, whether in membranous forms or in



cases of a re-stenosis following the surgical treatment of a bony CA that included the resection of the atretic plate [9]. Transnasal approach is associated with a shorter operating time, less palatal pedicle lesions and faster wound healing; it allows oral alimentation shortly following surgery [23]. In their meta-analysis, Durmaz et al [27] reported a success rate of 85.3 % in a total of 238 patients; other studies reported percentages between 68 % and 93% [31–33]. All our patients underwent an endoscopically controlled endonasal surgery with a global success of 76.2 % after initial surgery and 95 % after reoperative surgery.

However, this technique can be challenging due to the anatomical dimensions especially in neonates and the possible associated cranio facial abnormalities [11,34]. Other approaches such as transpalatal are more aggressive and less commonly used. It is indicated in cases where the transnasal method is impossible to realize mainly due to anatomical limitations [4,20].

The use of nasal stents is still debated, there is no consensus regarding their role in CA management [19]. The majority of IPOG respondents (60.8%) reported that it is less frequently used [20]. Moreddu et al. [23] recommended nasal stenting in bilateral CA when surgery is performed at a young age and a low weight. They concluded that, in unilateral CA, stenting was as safe as non-stenting. De Freitas et al. [31] suggested that stenting for 3 months reduced recurrence rates. Other studies did not recommend stenting following endonasal repair of CA and reported no benefits in placing nasal stents to avoid recurrences [13]. Some authors reported that the nasal stents cause local infection and the formation of granulation tissue increasing the risk of re-stenosis [29]. In their meta-analysis, Stryshowsk et al. [35] concluded that stents had no effect on success rates and that they resulted in more severe complications. In our study, nasal stents were placed in 16 patients using intubation tubes. It is the preferable stenting material in literature [20]. We concluded that neither stent placement nor its duration influenced recurrence rates. Duration of stenting varied between 3 and 12 months; it was consistent with other studies which also recommended long stenting duration [30,31,33].

Mitomycin C was not used in any of our patients. It is reported to decrease recurrence rates and reduce granulation; however, its role is still controverted in literature. Some authors reported that the molecule prevents stenosis [36]. While others reported that Mitomycin had no significant effect on preventing re-stenosis [19,32]. Attya et al [13] declared higher re-restenosis rates following mitomycin use.

As for post-operative treatment, nasal washes with saline serum, intranasal corticosteroids and protein pump inhibitors (PPIs) are recommended while the use of antibiotics is still debated. Follow up using endoscopic control is recommended 2 weeks following surgery [20]. Nasal saline irrigation and periodic endoscopic control are reported to reduce re-stenosis rates and improve success rates of the initial surgery [29,32]. The IPOG recommends an endoscopic control under general anesthesia after

initial surgical treatment for bilateral CA, CHARGE syndrome, young age, low weight [20].

Restenosis is the most common concern following surgical treatment of CA [4]. In our study, 23.08% of patients presented a re-stenosis and underwent a second surgery via an endonasal endoscopic approach. It is the recommended approach by the IPOG in cases of revision surgery [20].

Re-stenosis rate varied from 7% to 89% in previous series [13,32,37]. Factors influencing re-stenosis rates are: associated malformations, undergoing first surgery at a younger age, stent placement, bony atretic plate and gastropharyngeal reflux [23,25,34,35]. Contrarily to our study, in literature, recurrences are more frequent in bilateral forms [10]. In our study, we noted that bilateral form was not associated with higher re-stenosis rates, similarly to the findings of Abbelee et al. [38] Pure bony CA are more susceptible of undergoing revision surgery in comparison with mixed and membranous forms [10,39]. Age and weight are reported as factors for restenosis [33,39]. Wider patient's size is associated with larger choanal arches opening and then with less recurrences [11,12,23]. Newborns operated at an early age for bilateral CA are then associated with higher recurrence rates. Early age at initial surgery did not influence re-stenosis rates in our study.

The presence of associated malformations is reported to increase recurrence rates [40]. Local malformations were significantly associated with a higher restenosis rate in our study as well. Other authors rather concluded that there was not an association between recurrences and malformations [23,39].

Our study has several limitations. It's a retrospective study and data were retrieved out of old and incomplete medical records in some cases which could have biased results. Besides, the small size of our sample which is due to the rarity of this disease, is insufficient to make precise epidemiological measurements, definitive conclusions regarding the optimal surgical technique as well as solid statistical measurements of factors predicting restenosis. A multi centric and prospective study is then necessary for a better evaluation and more solid statistical measurements.

CONCLUSION:

Transnasal endoscopic surgical approach is the technique of reference in CA treatment. The development of novel endoscopic techniques with a precise CT guidance of anatomical malformations has minimized restenosis percentages in our study as well as in literature. We concluded that the presence of associated malformations is a factor associated with higher recurrence rates while the sex, age, nasal stents and laterality didn't influence recurrence. The age and the weight at initial surgery, the use of nasal stents and mitomycin and the unilateral or bilateral form are still controverted. This is maybe due to the variability of clinical and radiological features of CA between different populations.

**Compliance with ethical standards**

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