Outcomes of Choanal Atresia Transnasal Repair: Factors Related to Restenosis

Résultats de la chirurgie endonasale de l'atrésie choanale: Facteurs influençant le resténose

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ABSTRACT

Aim: Choanal atresia is an uncommon nasal malformation, that it could be fatal when it is bilateral. Since its first description, diagnosis means and treatment's approaches have been evolved. We aim through our study to discuss the outcomes of the transnasal approach in the management of choanal atresia and assess factors related to restenosis.
Methods: We performed a retrospective charts' review of 29 patients data with the diagnosis of choanal atresia, during 14-years long period (from January 2002 to December 2015).
Results: The mean age of patient was 10 years. We observed a female predominance. Neonatal respiratory distress revealed the choanal obliteration in four cases. The choanal atresia was unilateral complete in 21 patients (72%), incomplete in two patients (6%) and bilateral in six patients (20.6%). Choanal atresia was osseous in four cases (17.3%), membranous in three cases (10.3%) and mixed in 16 cases (55.1%). The overall success rate after a first intervention was 62%; it reached 85% after a second intervention. The analytical study showed that the age under 3 years was significantly correlated with a risk of restenosis (p=10-3). There was a tendency for recurrence in patients with associated malformations with no significant risk (p=0.16). No significant relationship was found between stenting and success rate (p=0.12).
Conclusion: The success of CA repair depends significantly on the age of the child. Other factors such as bilateral form, postoperative stenting and associated malformations seem to upper the risk of restenosis.
Key words: choanal atresia, CT scan, endoscopy, nasal obstruction.

RESUMÉ

But: L'atrésie choanale est une malformation peu fréquente. Bilatérale, elle peut mettre en jeu le pronostic vital. Depuis sa première description, les moyens diagnostiques et les approches thérapeutiques ont évolués. Notre objectif est de discuter les résultats du traitement endonasal de l'atrésie choanale et de déterminer les facteurs en relation avec le risque de resténose.
Méthodes: Nous avons réalisé une étude rétrospective ayant colligé 29 dossiers de patients pris en charge dans notre service pour atrésie choanale durant une période de 14 ans (janvier 2012-décembre 2015)
Résultats: L'âge moyen était de 10 ans. Nous avons noté une prédominance féminine. Une détresse respiratoire néonatale a été observée chez quatre patients. L'atrésie était unilatérale complète chez 21 patients (72%), incomplète chez deux patients (6%) et bilatérale chez six patients (20,6%). Elle était de type osseuse dans quatre cas (17,3%), membraneuse dans trois cas (10,3%) et mixte dans 16 cas (55,1%). Le taux de succès global une première chirurgie était de 62%; il a atteint 85% après une seconde chirurgie. L'analyse statistique a montré que seul l'âge inferieur à trois ans avait une relation statistiquement significative avec le risque de resténose (p=10-3). On a constaté plus de cas de resténose chez les patients porteurs de malformations associés mais la relation n'était pas statistiquement significative (p=0,16). De même on n'a pas trouvé de relation significative entre le calibrage et le taux de succès (p=0,12).
Conclusion: Le succès de la chirurgie de l'atrésie choanale dépend de façon significative de l'âge de l'enfant. D'autres facteurs semblent majorer le risque de resténose tels que l'atteinte bilatérale, les malformations associées et le calibrage postopératoire.
Mots-clés: atrésie choanale, tomodensitométrie, endoscopie, obstruction nasale

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INTRODUCTION

Choanal atresia (CA) is defined by an obliteration or a narrowing of the posterior part of the nasal cavity, due to failed recanalization of the nasal fossae during fetal development and persistence of abnormal bony or soft tissue [1,2]. It was first described by Roederer more than 250 years ago [3]. CA was widely studied among the pediatric and the adult population, its incidence is between 1:5000 and 1:10.000 live births [1,4]. It can be unilateral or bilateral; early reports suggested that 90% are pure bone atresia and 10% are pure membranous [4]. Recent studies reported that CA is mixed in 70% and pure bony in 30% [5]¬¬¬¬. Its ethiopathogenis remains unclear. Bilateral obstruction in newborns leads habitually to respiratory distress. In those patients, treatment is usually needed in the first few days of life, while unilateral choanal atresia may be better tolerated and treatment may be delayed [1,6. Endoscopic surgery and CT scan have considerably improved the diagnosis of CA [6]. The first international guidelines of pediatric choanal atresia were recently published in the purpose of improving the management of such disease [1].

MATERIAL AND METHODS

We report a retrospective study of 29 charts' data of patients treated for choanal atresia in our department during a period of 14 years (from January 2002 to December 2015). Catheters were used to confirm nasal obstruction in newborns. Four millimeters 0° rigid endoscopes were used to highlight the atretic plate in older patients. Maxillofacial CT scan was performed in all patients to confirm the diagnosis and assess the atresic plate structure. All patients underwent transnasal surgery under general anesthesia after an oral intubation and topic decongestion of nasal cavities. Two techniques were used: nasal puncture in four newborns and endoscopic approach using drill, cold instruments and contact laser diode in the others patients. The endotracheal tubes were used for stenting. All patients underwent postoperative follow-up with nasal endoscopy. The overall follow-up period ranged from one to 5 years. We consider as a surgery failure the choanal restenosis. We studied the predicting factors of choanal restenosis using a univariate analysis conducted by the SPSS 20 software. The threshold of statistical significance was set at 5%.

RESULTS

The mean age was of 10 years [one day-30 years]. Four patients were newborns. CA occurred more in female than in male (sex ratio: 2). Neonatal respiratory distress revealed the choanal obliteration in four cases. Nasal obstruction was the main symptom in the other patients. Non-syndromic malformations were associated in six patients (20.6%) (Table I).

Table I: Associated malformations

Case	Age of patient at diagnosis	Laterality	Associated malformations
1	17 months	Unilateral	Feet deformities
2	10 years	Unilateral	Bifid uvula
3	30 years	Unilateral	Umbilical hernia
4	New born	Bilateral	Single umbillical artery/ Facial dysmorphology/ Interventricular communication
5	New born	Bilateral	Renal cystic dysplasia
6	20 years	Bilateral	Psychomotor retardation /bilateral neurosensorial hearing loss

We had no case of CHARGE syndrome. Nasal endoscopy and CT scan attested that CA was unilateral in 23 patients and bilateral in six patients (Table II, Figure 1).

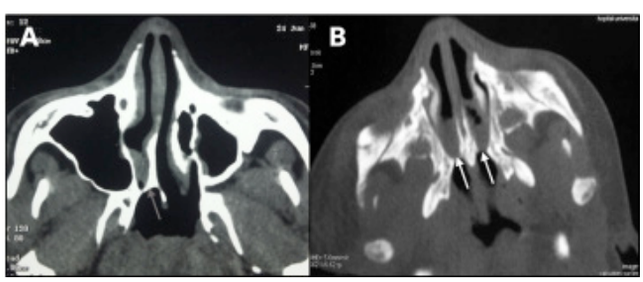

Figure 1: CT scan showing unilateral osseous right side choanal atresia (A) (grey arrow) and bilateral mixed choanal atresia (B) (white arrows)

Table II: Nasal endoscopy and CT scan data

Nasal endoscope feature	CA structure on CT scan			Total of patients
	Osseous	Mixed	Membranous	
Unilateral complete CA	4	16	1	23
Unilateral incomplete CA	0	0	2	
Bilateral complete CA	0	5	1	6
Bilateral incomplete CA	0	0	0	

Four newborns with bilateral CA were treated by transnasal puncture, using a hemostasis forceps, with a bilateral stenting during eight weeks. Twenty-five patients were treated by endoscopic approach; contact laser diode was used in three cases. Stents were used in 17 cases including all cases treated with laser. Complications such us synechiae occurred in four patients, infection in five patients and fall of the stent in one case. The overall success rate was of 62%, this rate reached 72% (table III) if we consider only the endoscopic approach results. Restenosis occurred one month after stenting removal in all newborns treated by nasal puncture. It interested one side in three cases and both sides in one case. Restenosis occurred averagely six months after surgery in eight patients treated by an endoscopic approach (29%) (Table III, Figure 2).

	Table III: CA results of the endoscopic approach				
	Without stenting		With stenting		Total
Laterality of CA	Success	Failure	Success	Failure	
Bilateral	-	-	2	-	2
Unilateral	9	2	7	5	23
Total	9	2	9	5	25

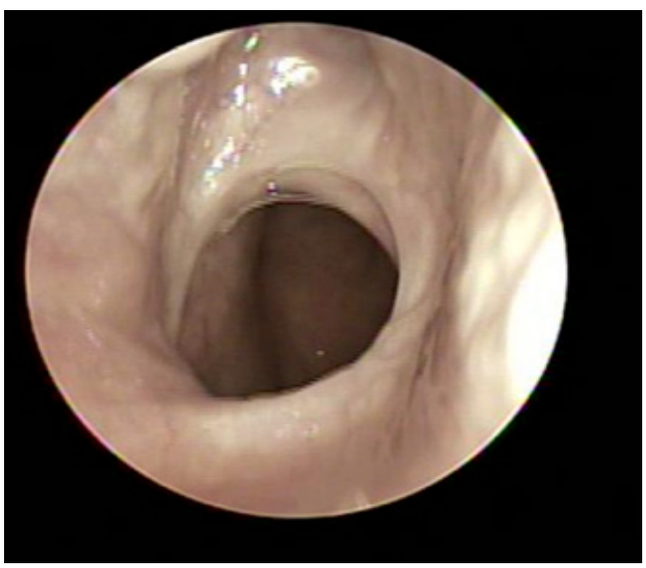

Figure 2: Postoperative feature of a right choana

All cases of restenosis were managed by an endoscopic approach using cold instruments with stenting. The success rate reached 86% after the second intervention. The univariate study showed that an age under three years was the only significantly predicting factor of restenosis (p=10-3) (Table IV).

Table IV: Univariate comparative study of restenosis predicting factors after choanal atresia surgery

Studied factors		Restenosis		P
		N*	%	
Age	<3 years	9	88.9	10-3
	>3 years	20	15	
Laterality	Unilateral	23	30.4	0,16
	Bilateral	6	66.7	
Associated malformations		6	66.7	0,16

studied factors			Restenosis			P
			n**	%		
Atretic plate structure	Bony		4		50	0,62
	Membranous		5	20		
	Mixed		26	38.5		
Stenting		Yes	24		50	0,12
	No		11	18.2		

*: number of patients
**: number of choana

DISCUSSION

Choanal atresia (CA) is relatively a rare disease [1,2]. Some studies claim that CA is two or three times more frequent in females [7]. In our study, we found that CA is more frequent in females. CA can be uni or bilateral. Unilateral forms affect the right side twice as much as the left [2,7]. The characteristics of CA follow a “2–1” rule, namely the ratio of unilateral to bilateral CA, female to male and the right sided to the left sided CA [7,8]. Previously, 90% of CA reports have indicated that 90% of CA are bony, while they are membranous in only 10% [9]. However, with the improvement of CT-scan analysis of CA structure, recent reviews suggest that CA are mixed in more than 70% of cases, and that all membranous forms have some bony component [8]. Congenital anomalies can be associated to CA in up to 50% of cases [8,9]. The most common malformation is CHARGE syndrome (Coloboma, Heart abnormalities, choanal Atresia, growth or mental Retardation, Genitourinary anomalies, and Ear abnormalities) [8,9,10]. The most common clinical presentation is a chronic unilateral nasal obstruction, persistent mucoid rhinorrhea, or a history of chronic sinusitis. However, bilateral CA can be diagnosed in newborns with cyclical periods of respiratory distress relieved by crying or mouth breathing [7,9,11].

Clinical diagnosis of CA is habitually easy by seeing the atretic plate in nasal endoscopy or objectifying the nasal cavity impermeability by inserting a suction tube [12]. Computed tomography is necessary to the diagnosis by confirming the diagnosis of CA, study the structure, and though plan surgery [1,13]. Temporal bone CT scan are also recommended in order to detect eventual associated ear malformations particularly of the semi-circular canal especially in patients with CHARGE syndrome [14].

Several surgical techniques have been reported in the literature since the first description of the CA entity. The transpalatal procedure was the most used approach till the 90’s, when endoscopic CA repair was described by Stankiewicz [15]. Nowadays, it becomes the most common used procedure [1]; in fact, it allows an excellent visualization to remove the atresic plate with minimal bleeding [15]. Transpalatal approach could be used in cases of local anatomical difficulties [1]. According to some authors, unilateral CA surgery should be reported at least to the age of 6 months, ideally to one or two years [1]. For bilateral forms, which is a life threating situation, transnasal puncture, using a hemostasis forceps was used to manage with those cases in Tunisia before the evolving of the endoscopic surgery such in our cases [10,16]

However most of surgeons delay surgery after assessment of general anesthesia risk factors mainly cardiac abnormalities [1].

The CA repair technique depends on surgeon preference. Drilling with atretic plate microdebridement is the most used, balloon dilation and cold instruments are also used but less frequently. The laser as CO2,

Nd-YAG, holmium-YAG, potassium titanyl phosphate (KTP), and contact diode (CDL) have been described for CA repair [17,18]. However, the lack of significant advantage compared to conventional instruments, increased fire risks, and reported deaths from air embolism related to Nd-YAG laser limits the use of such technique [7,19]. As the technology continues to evolve and future research provides more outcome data, lasers may play a further role in the CA repair.

In recent years, several studies have suggested that the use of stents is controversial, it is not always recommended after endoscopic surgery because of the risk of stent-related injuries, local infections, inflammation and necrosis, potentially resulting in permanent septal perforation or cosmetic deformity and ulcerations [10]. The preferred stenting material is an endotracheal tube [1].

Success rate in the literature ranges from 45.3% to 92.85% [11,20,21]. It reaches 100% after a second intervention in the study of Zainine et al [10]. In our study the success rate was of 62% after the first intervention and reaches 86% after the second one.

In their meta-analysis summarizing data of 238 patients with CA, Durmaz et al [22] found a transnasal endoscopic repair success rate of 85.3%. In our study, it was of 68%. This variability can be explained by the heterogeneity of surgical techniques and the exact definition of surgery success or failure. Most of studies demonstrated that surgery failure was associated with an early age of surgery of CA, in the bilateral forms, bony structure and associated malformations [20]. According to Teissier el al [23], predictive factors of restenosis are an associated gastro-esophageal reflux disease and patient age younger than 10 days. However, previous surgery and associated malformations are not predictive of restenosis. Uzomefuna et al [20], claimed that the increasing age and avoidance of nasal stenting may improve the surgical endoscopic transnasal approach outcome. According to some authors, surgeon's experience should be considered in analyzing the endoscopic approach results [23]. In our study, we found that only an age younger than three years was significantly associated with a high risk of restenosis.

CONCLUSION

The success of CA repair depends significantly on the age of the child; however, it seems that bilateral CA, postoperative stenting and associated malformations upper the risk of restenosis. Actually, there is a consensus that aim to homogenize pediatric CA management. We could extrapolate recommendations to deal with adults' forms considering obviously the different surgeons' experience.

Compliance with ethical standards

Conflict of interest:

The authors stated that there is no conflict of interest.

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