

Complications of Endoscopic Surgery of Congenital Choanal Atresia: How to Avoid It?

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ABSTRACT

Background: Choanal atresia is developmental failure of nasal cavity to communicate with nasopharynx causing complete obstruction of the nasal cavity.

Objective: This study was aimed to review the trans-nasal endoscopic approach systematically as a minimally invasive surgery to treat unilateral or bilateral choanal atresia, to analyze the complications of this procedure and how to avoid it.

Patients and Methods: This retrospective study included twenty-five patients, with congenital choanal Atresia (CCA) who underwent primary transnasal endoscopic repair. The data was collected from patients records at Zagazig University Hospital.

Results: The common complication in the study was restenosis, bilateral choanal atresia (BCA) cases were operated at an early age had high rate of recurrence while unilateral choanal atresia (UCA) case has a lower recurrence rate. The current study showed statistically significant relation between restenosis post endoscopy surgery of CCA and age at time of operation, site of atresia, except cases operated using surgical technique iii that started with vomer resection and technique vii with preservation of mucosa using fibrin glue. It noticeable that restenosis commonly occurred among young age exposed to endoscopic surgery and who had bilateral stenosis. Surgical bony resection is usually limited in cases of BCA because both pterygoid processes are medialized and the obtained choana is therefore narrower, whereas, by definition, one pterygoid processes is in normal position in UCA and is covered by a perfect mucosa lining.

Conclusions: It could be concluded that the endoscopic transnasal approach is an effective, safe and minimal invasive surgery to treat unilateral or bilateral CCA. Starting with vomer resection give good result regarding patency of neochoana with low complications rate compared with other endoscopic techniques.

Keywords: Choanal atresia, Complications, Bilateral choanal atresia, Congenital choanal atresia.

INTRODUCTION

Choanal atresia (CA) is a life-threatening, but relatively uncommon, anomaly involving the congenital obliteration of the posterior nasal choana, due to the blocked oronasal membrane in children ⁽¹⁾. The incidence of CA is one in 5000 to 9000 live births ⁽²⁾.

The nasal obstruction in CA can be either bony, membranous, or mixed, recent studies have indicated 70% mixed bony and membranous and 30% pure bony ⁽³⁾. CA can also be categorized as unilateral, bilateral, or due to other craniofacial abnormalities. 20%-50% of patients with CA, particularly those affected by BCA, suffer also from other genetic malformation. CHARGE syndrome is the most common associated congenital anomalies ⁽⁴⁾. Unilateral CA affects only one nostril, usually the right, side in the ratio of 2:1, the child is often present late with nasal obstruction and unilateral rhinorrhea ⁽¹⁾.

In contrast, bilateral CA can be identified early after birth in most cases, as infants preferably breathe from the nose until four months of age due to the high cervical location of the larynx; therefore, bilateral CA in neonates is considered medical emergency causing respiratory distress demanding immediate intervention ⁽⁵⁾. Currently, nasal endoscopy and multi-slice CT scan are the procedure of choice for the diagnosis of CCA ⁽⁶⁾.

The definitive treatment of CA is surgical, various surgical procedures have been described, and the success is primarily measured based on the "patency"

of the choana and absence of symptoms. Although the treatment techniques for CA have been extensively reported, there is no general consensus about the optimal surgical technique. The various surgical procedures to treat CA include transnasal puncture, transpalatal resection, and endoscopic transnasal repair, which can be supported with or without stents ⁽⁷⁾.

Traditionally, stenting of the opened choana has played a key role in the postoperative management of CA.

Park et al. ⁽⁸⁾ reported that 92 of 95 associates of the American Society of Pediatric Otolaryngology consistently deployed stents.

However, in recent years, several studies have suggested that the use of stents is not always required after endoscopic surgery, and their use is still controversial, as cases of stent-related injuries, local infections, inflammation, and necrosis, potentially resulting in permanent septal perforation or cosmetic deformity and ulcerations have been reported ⁽⁹⁾.

However, the potential advantages of using stents include the avoidance of early restenosis, assisting the healing of mucosal flaps, and the prevention of post-surgery edema ⁽¹⁰⁾.

This study was aimed to review the trans-nasal endoscopic approach systematically as a minimally invasive surgery to treat unilateral or bilateral choanal

atresia, and to analyze the complications of this procedure and how to avoid it.

PATIENTS AND METHODS

This retrospective study included a total of twenty-five patients with congenital CA. They represented all patients with congenital CCA underwent primary transnasal endoscopic repair at Zagazig University Hospitals during a period of 5 years.

Medical records were analyzed for diagnostic modality, clinical symptoms, age, sex, laterality, time of surgery, age at surgery, surgical technique, and postoperative complications.

Methods of diagnosis: All patients in the study received a full history, clinical examination review and underwent fiberoptic nasal endoscopic examination and an axial CT scan of the head to confirm the diagnosis.

Surgical techniques reported were aiming to improve surgical outcome:

- 1) Stentless transnasal endoscopic repair⁽¹¹⁾.
- 2) Transnasal endoscopic repair with stentless mucosal flap technique⁽¹²⁾.
- 3) Stentless transnasal endoscopic repair starting with vomer resection ending by no stent nor packing⁽¹³⁾.
- 4) Transnasal endoscopic repair with fenestrated endotracheal tube stent⁽¹¹⁾.
- 5) Endoscopic repair with topical intra-operative mitomycin application⁽¹⁴⁾.
- 6) Endoscopic repair using post-operative parasseptal silicone sheet⁽¹⁴⁾.
- 7) Transnasal endoscopic repair with preservation of mucosa using fibrin glue technique⁽¹⁵⁾.
- 8) Endoscopic laser-assisted CA repair⁽¹⁶⁾.

Ethical Consideration:

An approval of the study was obtained from Zagazig University Academic and Ethical Committee. This work has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

Statistic analysis

The collected data were coded, processed and analyzed using the SPSS (Statistical Package for Social Sciences) version 22 for Windows® (IBM SPSS Inc, Chicago, IL, USA). Data were tested for normal distribution using the Shapiro Walk test. Qualitative data were represented as frequencies and relative percentages. Chi square test (χ^2) to calculate difference between two or more groups of qualitative variables. Quantitative data were expressed as mean \pm SD (Standard deviation). Independent samples t-test was used to compare between two independent groups of

normally distributed variables (parametric data). P value < 0.05 was considered significant.

RESULTS

17 patients (68%) were females, and 8 patients (32%) were males. The median age for the patients was 12 days, ranged from 3 days to 10 years. The main symptoms for bilateral choanal atresia cases were respiratory distress, in contrary nasal discharge and nasal obstruction was the main presenting symptoms in unilateral cases. Two patients had associated anomalies, one patient had cardiac anomalies and the other patient had CHARGE syndrome. The median age at time of surgery was 6 days for bilateral choanal atresia cases and 4 years for unilateral choanal atresia cases (table 1).

Table (1): Demographic data of studied Patients (n=25).

	Number of patients	%
Sex		
- Females	17	68.0
- Males	8	32.0
Age per years	12 days (3days	
- Median (Range)	-10 years)	
Symptoms		
-Respiratory distress	14	56.0
-Nasal obstruction & Nasal discharge	11	44.0
Medical history		
-No associated abnormalities	23	92.0
-Congenital heart lesion	1	4.0
-Charge syndrome	1	4.0
Frequency of side of congenital choanal atresia among studied children site of lesion		
-Bilateral	14	56.0
-Unilateral	11	44.0

Figure (1) Shows that early complications of endoscopic surgery for congenital choanal atresia among studied children, 16.0% had edema and airway obstruction, frequently with technique i, ii, iv. While 16.0% had Septal injury frequently with technique i, ii, iv, v and 16% had turbinate injury, frequently with technique v, vi, vii, viii also 16% developed anterior nasal injury, frequently with technique i, ii, iv, v, and use of powered instruments, 12.0% were suffering from bleeding, frequently due to excessive drilling of anterior or lateral wall of sphenoid sinus and middle turbinate mucosal damage and 4.0% of the children were suffering from Skull base Injury due to superior drilling, or feeding difficulty due to stents use, or palate injury due to excessive inferior drilling.

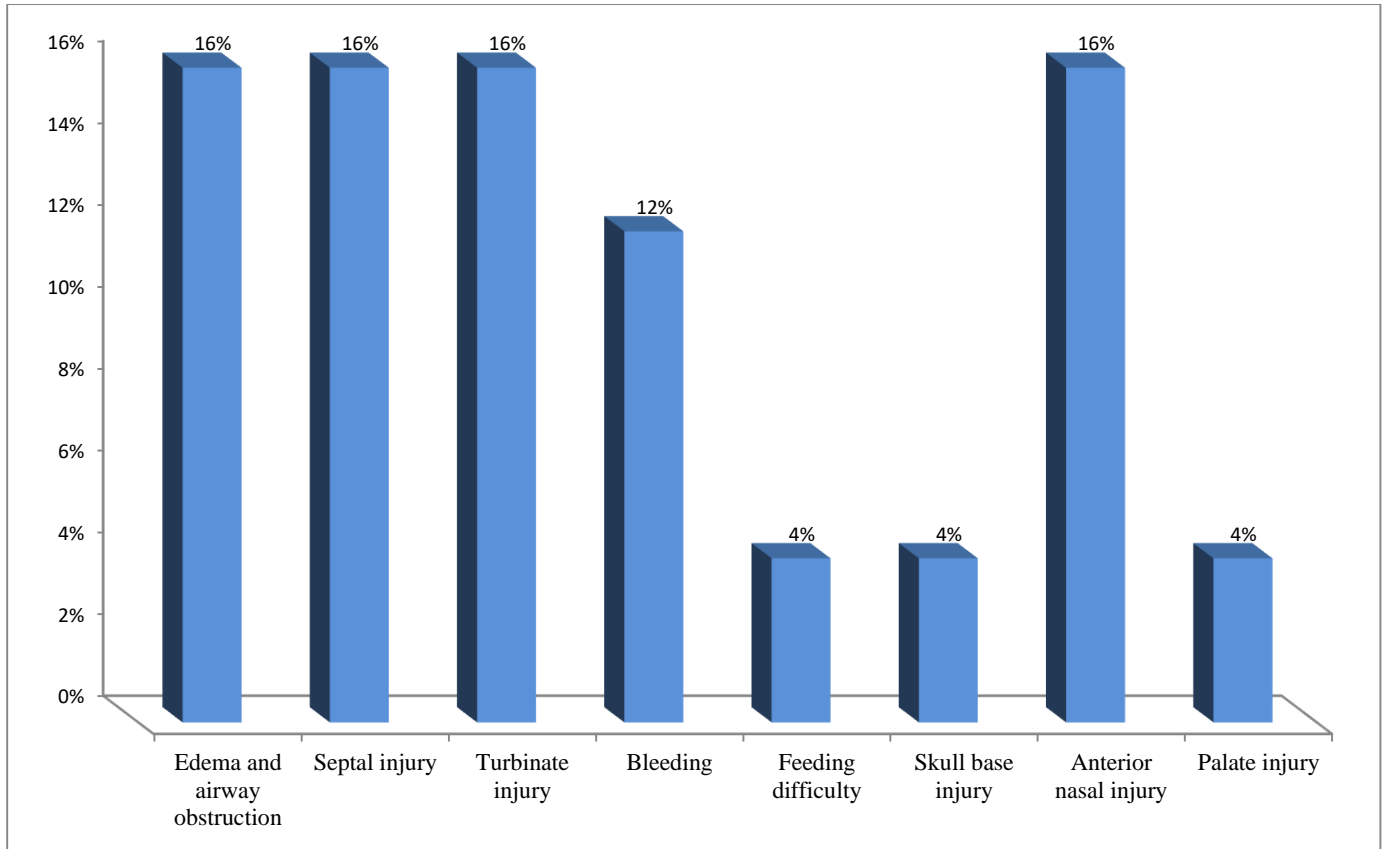


Figure (1): Percentage of early complications of endoscopic surgery for congenital choanal atresia among studied children.

Table (2) shows that one patient (4.0%) with BCA had nasal infection due to use of stent, 5 patients (20.0%) had granulation tissue formation, 4 had bilateral CA and one unilateral CA, its due to use of stents and keeping bare bone exposed, also 2 patients (8.0%) had crust formation observed with stents use, one patient had bilateral CA formation, other one unilateral CA.

In addition, 6 patients (24.0%) had post endoscopic adhesion 3 of them had bilateral CA, others unilateral CA, commonly due to use of stents.

Finally, 48% of patients had post endoscopic restenosis two thirds of them had BCA while one third had UCA, restenosis commonly observed with techniques i, ii, iv, v, vi, viii. one patient had both granulation tissue formation and nasal infection included in both groups.

Table (2): Frequency and percentage distribution of late complications of post endoscopic surgery for congenital choanal atresia children (N. 25).

Late complications	Yes N. (%)	Site	Technique
Nasal infection	1 (4.0%)	bilateral	Stents
granulation tissue formation	5 (20.0%)	(4 patients) bilateral (1 patient) unilateral	Bare bone stents
Crust formation	2 (8.0%)	(1 patient) bilateral (1 patient) unilateral	Stents
Adhesion	6 (24.0%)	(3 patients) bilateral (3 patients) unilateral	Non sheet stent
Restenosis	12 (48.0%)	(8 patients) bilateral (4 patients) unilateral	i,ii,iv,v,v i,viii

Table (3) shows that median of duration per months lapse for restenosis was 7.5 months with range from two months to 5 years among studied patients.

Table (3): Mean and standard deviation of duration per months lapse for restenosis (N. 12).

Duration lapse for restenosis per months	
Mean ± SD	19.5±30.6
Median(range)	7.5(2-96)

Table (4) shows statistically significant relation between restenosis post endoscopic surgery of congenital choanal atresia and age at operation p=0.038, site of choanal atresia p=0.008. It noticeable that restenosis commonly occurred among young age exposed endoscopic surgery and who had bilateral stenosis. This developed with all surgical techniques but it's less in techniques iii & vii.

Table (4): Relation between restenosis post endoscopic surgery of congenital choanal atresia and patient characteristics (N=25).

	Restenosis				N.	χ^2	p-value
	Yes (N.12)		No (N.13)				
	No.	%	No.	%			
Age at operation							
Median (range)	6 days (3 days-9 yrs.).		4years (3 days-10 years).			U=2.1	0.038 (S)
Sex							
Females	10	58.8	7	41.2	17	f	0.202
Males	2	25.0	6	75.0	8		
Side of choanal atresia							
Bilateral	10	71.4	4	28.6	14	6.9	0.008 (S)
Unilateral	2	18.2	9	81.8	11		
Associated congenital anomalies							
No	12	52.2	11	47.8	23	f	0.99
Yes	1	50.0	1	50.0	2		

χ^2 Chi square test f= fisher exact test u= Mann Whitney u test. (S) significant p<0.05

DISCUSSION

The current study included 14 patients (56%) with bilateral choanal atresia and 11 patients (44%) with unilateral choanal atresia. 17 patients (68%) were females, and 8 patients (32%) were males. The median age for the patients was 12 days, ranged from 3 days to 10 years. Two patients (8%) had associated anomalies, one patient had cardiac anomalies and the other patient had CHARGE syndrome. The median age at time of surgery for BCA was 6 days, while the median age at time of surgery for UCA was 4 years.

The obtained data were tabulated, and the obtained results were compared with the results of other studies in literatures regarding postoperative complications. BCA cases commonly present with respiratory distress, cyclic cyanosis, feeding difficulty, Chest infection was one of presenting symptoms in 6

patients with BCA Cases of UCA were presented by persistent unilateral rhinorrhea and unilateral nasal obstruction.

Regarding early postoperative complications, the following points were recorded:

4 patients (16%) developed edema and airways obstruction necessitating prolonged intubation and ICU monitoring, it would be due to associated airway anomalies, mucosal injury and excessive drilling during surgery. To avoid this complication, the presence of synchronous airway anomalies as well as the risks associated with intubation and ventilation difficulties needed to be considered to decrease risk of anesthetic related complications as laryngospasm, bronchospasm, oxygen desaturation, reintubation, difficult extubation and cardiac arrest (17). Patient with bilateral CA especially if surgical procedure has been lengthy, airway edema is present or patient is hemodynamic unstable, should remain intubated in NICU under monitor until the issue have been resolved and the patient is stabilized (18). Powered instrumentation with soft tissue shavers and small drills are believed to be less traumatic to nasal mucosa, allow better healing of the mucosa which decrease risk of mucosal edema (19).

Regarding bleeding, 3 patients (12%) developed bleeding which would be caused by excessive mucosal damage , excessive drilling to anterior wall of sphenoid sinus during surgery and injury to sphenopalatine artery and its septal branch or vidian artery, its usually controlled by backing or cauterization. **Khafagy** (20) reported one case died from profuse IO bleeding, presumed hemorrhagic disease of newborn while **Nour et al.** (21) reported one case of bleeding that require endoscopic control. This hemorrhagic complication can be avoided by backing the nasopharynx to protect adjacent structures, keeping the surgical field clean of blood and debris by continues suction, prevention of excessive damage to mucosa, and avoid excessive drilling to anterior wall of sphenoid sinus, technique iii help this more than other by creating a wide surgical field and working through both nasal cavities. The resection of atretic plate usually started at junction between vomer and hard palate at the inferomedial direction of atretic plate which represent the safest portion of blocking membrane to avoid hemorrhagic complications (Sphenopalatine & Vidian artery injury). The use of soft tissue shaver and small drill is less traumatic to nasal mucosa which decrease risk of bleeding (22).

Regarding mucosal injury, 4 patients (16%) developed septal injury, other 4 patients (16%) developed turbinate injury. These iatrogenic injuries may be due to small nasal anatomy in neonates that restrict the size of instrumentations used during surgery, unclear surgical field, excessive drilling, and associated anatomic anomalies. The mucosal complications can be avoided by careful preoperative assessment of nasal anatomy, using nasal decongestant to diminish volume of nasal turbinate, endoscopic nasal examination before

starting surgery, keeping surgical field clean by continuous suction and avoid excessive drilling. Anatomic landmarks have to be respected and basic surgical guideline should be followed **Teissier et al.** (23) technique iii starting with resection of the vomer and use 2 nostril for instruments will help to avoid this complication.

As regard skull base injury one patient (4%) developed skull base injury which may be caused by misdirection of perforation of atretic plate, failure to back the nasopharynx, working in unclear surgical field and excessive drilling. This injury can be avoided by backing the nasopharynx to protect adjacent structures, directing the puncture of atretic plate inferiomedially to avoid skull base and keeping the surgical field clean by continues suction, technique iii help vision by using two nasal sides simultaneously which provide clear anatomic orientation and keeping inferior away from skull base.

Regarding nasal skin injury 4 patient (16%) developed nostril injury that may be caused by thermal damage as result of excessive use of power instruments and drilling. **Mohamed et al.** (11) reported one case of palatal fistula, one case of alar cartilage injury and a case of septal thermal injury during intranasal drilling of atretic plate and suffered alar collapse later, as an intraoperative complication following trans nasal endoscopic CA repair.

This injury is usually avoided by careful use of power instruments, keeping it away from nasal skin and nasal saline irrigation, use of technique iii help avoiding aggressive use of power instruments.

One patient (4%) developed soft palate injury that healed conservatively, this consistent to studies of **El-Ahl et al.** (13), **De Freitas et al.** (24) and **Ibrahim et al.** (25). Keeping surgical field clean and avoid excessive inferior drilling will help to reduce risk of palate injury.

One patient (4%) developed feeding difficulties that may related to mucosal edema, use of stents and inadequate vomer resection which can avoided by careful use of power instruments, the use of soft tissue shaver, small drill which is less traumatic to mucosa allowing better mucosal healing with early resumption of oral feeding, and creating wide cavity posteriorly that provide a large nasal airway so the child can feed and breathe without any difficulty, technique iii& vii avoid these.

Most of the patients resume oral feeding on the evenings of operation day and discharged on next day, this is in contrast to trans palatal approach where convalescence is prolonged due to palatal incisions and associated edema that will delay resumption of oral feeding and discharge from hospital.

The late complications reported in the current study were nasal infection, granulation tissue formation, intranasal crust, Synechia and restenosis

The common late complications reported in current study was restenosis (48%). BCA repair is associated with an increased risk of restenosis as

reported by **Teissier et al.** (23), **Chia et al.** (26) and **Mohamed et al.** (11), this tendency was significantly reported in the current study. The relatively high recurrence rate in the current study could be attributed to the high proportion of bilateral case, that need surgery in early neonatal period. **Friedman et al.** (27) reported that children with low weight at time of surgery, restenosis is more frequent than in higher weight children. **El-Ahl et al.** (13). Technique vii also showed low rate of restenosis.

Restenosis can be avoided by creation a widely patent posterior nasal choana sufficient for normal bilateral nasal breathing, removal of adequate portion of posterior part of the vomer which is essential step to get good result as failure commonly attributed to inadequate vomer resection, minimizing mucosal injury is also important step to avoid endonasal scar tissue formation. Efficient postoperative nasal saline irrigation is extremely beneficial for surgical outcome, and teaching patients or their parents how to do it properly at home is highly recommended to avoid secretion accumulation, crusting, fibrotic scar and restenosis, moreover periodic postoperative endoscopic surveillance or second look procedure to clean the neochoana of secretions, clots, crust, will help to avoid inflammatory process, infection, granulation tissue formation and restenosis **Kwong**(28) and **Saraniti et al**(29). The sequence of steps of endoscopic technique that begin with vomer resection is an important element to prevent restenosis and improve the outcome (13,30).

Regarding granulation tissue formation and crusting 5 patients had granulation tissue formation and 2 patients had crust formation, frequently due to use of stents and keeping bare bone exposed. **Saafan et al.** (31) reported in their study that 5 patients represent 50% of studied group using stents suffered postoperative granulation tissue formation as compared to 2 case 20% of non-stent group, while **Josephson et al.** (32) reported one case who developed granulation tissue formation a week after stent removal. Granulation tissue formation and crust can be avoided by prevention of excessive damage or removal of mucosa, use of mucosal flap to cover the raw areas, avoid excessive drilling of the bone and use of stents as possible, frequent nasal saline irrigations and periodic endoscopic surveillance.

Regarding post-operative adhesion (Synechia) 6 patients (24%) developed adhesion in current study **Mohamed et al.** (11) reported one case of mild nasal synechia with mild nasal obstruction and nasal discharge that improve after operative separation, while **Ibrahim et al.** (25) reported 3 infants developed postoperative nasal synechiae with no further treatment required. Postoperative synechiae can be avoided by preventing damage to nasal mucosa, avoid excessive drilling, covering raw surface, keeping the new choana clean of secretion, debris, and clot.

Regarding nasal infection one patient (4%) had nasal infection due to use of stent. One should avoid use

of stents as possible, and patient should receive prophylactic antibiotic to overcome this complication.

No growth disturbance of the palate, alveolar arch, and mid face, nor occlusive abnormality were recorded, post transnasal endoscopy as compared to transpalatal approaches.

CONCLUSION

It could be concluded that the endoscopic transnasal approach is an effective, safe, and minimal invasive surgery to treat unilateral or bilateral CCA, offering direct approach to the atretic plate, restore normal nasal passage, avoid damage to any growing structures, short surgical time, hospitalization stay with minimal morbidity and mortality. Starting repair with vomer resection (technique iii), give good result regarding patency of neochoana with low complications rate compared to other endoscopic techniques, no age or site limit for the procedure.

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