



Endoscopic Choanal Atresia Repair with Posterior Septectomy and Widening Toward Pterygoid Plate to Create a Wide Single Nechoana, Case Series

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ARTICLE INFO

Article history:

Received: Sept 26, 2023

Accepted: Mar 11, 2024

Published: April, 2024

KEYWORDS

1. Choanal Atresia
2. Single Nechoana
3. Endoscopic Atresia Repair

ABSTRACT

Objective: This study focuses on surgical technique, assessment of the outcomes and the recurrence rate of the disease. **Patients and methods:** This retrospective study was done on 18 patients with congenital choanal atresia at Azal specialized hospital and Al Kuwait University hospital, Sana'a, Yemen between 2014 and 2022. All patients underwent endoscopic repair with posterior septectomy, removal of atretic plate, lateral widening toward the pterygoid plate and creation of wide single nechoana with nasal stenting.

Results: Of 18 cases, 2 cases were excluded because of short period of follow up. Of the 16 remaining cases, 13 cases (81.25%) showed good results during the period of follow up (6 months to 9 years) from the initial surgery. Restenosis occurred in 3 (18.75%) bilateral choanal atresia cases which underwent revision surgery with good results. **Conclusion:** Endoscopic creation of wide single nechoana with stenting followed by close follow up and family education give good success rate.

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1. Introduction:

Choanal atresia (CA) is defined as a complete obstruction of one or both sides of the posterior nasal openings which leads to failure of communication between the posterior nasal cavity and nasopharynx. Choanal atresia, first described by Roederer in 1755. It is a rare congenital disorder with an estimated incidence

varies between 1:5000 to 1:9000 live births [1]. There are still controversies on the exact pathogenesis of CA [2]. Four embryological theories exist: (1) the persistence of the buccopharyngeal membrane from the foregut; (2) failure of perforation of the nasobuccal membrane of Hochstetter; (3) abnormal persistence or location of mesoderm forming

adhesions in the naschoanal region; (4) misdirection of neural crest cell migration [3].

Twenty to 50 % of all patients have other congenital anomalies, Coloboma, Heart disease, Choanal atresia, Mental and growth retardation, Genital hypoplasia, Ear deformities (CHARGE syndrome) [4]. Study by Brown's and associates (1996) revealed :70 % mixed (membranous and bony) and 30 % pure bony. No patients were found to have a purely membranous type. Both genders are affected, with a male to female ratio of 1:2 [5]. Anatomically, the boundaries of the posterior choanae include the undersurface of the body of the sphenoid bone superiorly, the medial pterygoid lamina laterally, the vomer medially, and the horizontal portion of the palatal bone inferiorly [1,2]. The anatomic features that characterize CA include a narrowing of the nasal cavity, thickening and impingement of abnormal pterygoid bone at the posterior choana, thickening of the posterior bony vomer, and membranous obstruction [5].

Bilateral choanal atresia (BCA) presents as a medical emergency at birth associated with respiratory distress [2]. In contrast, unilateral choanal atresia (UCA) presents later in life with unilateral nasal obstruction associated with unilateral persistent, profuse and thick nasal discharge [1]. Diagnosis of CA can be suspected by the inability to pass nasal catheter, confirmed by endoscopic examination and CT scan of the nose [2]. The definitive treatment of CA is by

surgical repair. Studies showed that, restenosis is the most common complication of surgical repair [2].

2. Patients and methods:

This study focuses on our experience in the management of congenital CA using transnasal endoscopic repair to evaluate our technique and how it affects the recurrence rate. The study obtained the approval of the ethical committee of our institution on 20/12/2022. This study was done by retrospective review of the files of 18 patients with congenital CA at Azal specialized hospital and Al Kuwait University hospital, Sana'a, Yemen between March 2014 and September 2022. All the patients who were included in the study gave oral consent by one of their parents for approval to use the patients' data for research purposes and publication. The patients' personal data were not identified on the study sheets after data collection. We excluded cases of acquired BCA and UCA post radiotherapy as well as cases of congenital nasal aperture stenosis that were misdiagnosed as BCA. All patients underwent full clinical evaluation, routine preoperative investigations and CT scan of the nose and nasopharynx for confirmation of the diagnosis (fig 1 A and B). All pediatric patients underwent cardiological evaluation and fitness. Informed consent was taken regarding the nature of operation, risks, the need for revision surgeries, possible outcomes and complications.

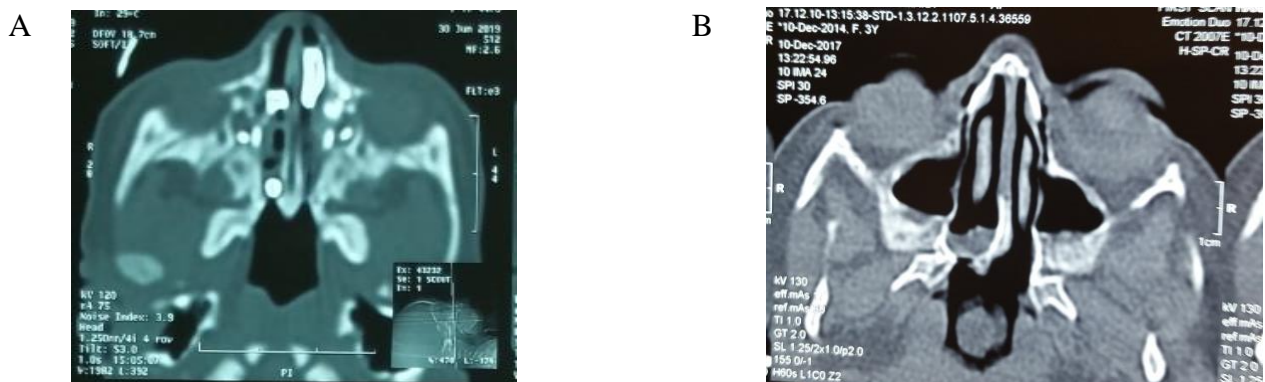


Figure 1. PNS CT scan, bone window, axial cut demonstrating (A) BCA of mixed type (bony and membranous), thick posterior part of vomer and membranous atretic plate. Note: injected contrast in nasal cavity by radiologist did not pass to the nasopharynx. (B) UCA (right side) of bony type, thick and lateralized posterior part of vomer; thick and medialized pterygoid plate and bony atretic plate.

All patients underwent endoscopic CA repair with posterior septectomy, removal of atretic plate, lateral widening toward pterygoid plate to create a wide single neochoana with stenting followed by close follow up and education of the parents on nursing of nostrils to avoid local complications of stents. Nursing of the nostrils was done through saline irrigation and application of local antibiotic ointment around the stents. The stents were removed after 4 weeks. Follow up was assessed by using endoscopic examination and by asking about parent's satisfaction regarding child nasal breathing. The periods of follow up ranged from 6 months to 9 years. The follow up was done weekly in the first month during stenting to avoid local complications and monthly for three months after removal of stents to assure the patency and absence of granulation tissue formation in the neochoana. In case of granulation tissue formation, follow up was done weekly to remove it by endoscope at outpatient department. Then, intermittent follow up was done according to the family need for consultation and preference of the surgeon.

Instruments: 30 or 0 degree of 4mm sinuscope, pediatric instruments of functional endoscopic sinus surgery (sickle knife, Cottle dissector, straight Blakesley forceps and cutting forceps, micro-scissor, micro-suction, backbiting forceps and Kerrison) and micro-ear instruments (micro-

suction, cup and crocodile forceps). A drill and micro-debrider were used as needed.

Steps of operation include: Application of local decongestant (xylometazoline nasal drops) 30 minutes preoperatively (figure 2). The surgical procedure, under general anesthesia starts with local infiltration of posteroinferior part of the septum by normal saline. An important landmark is the posterior end of the middle turbinate, the superior limit of surgery is below this level to avoid bleeding and skull base injury. Incision and elevation of the septal flap at posteroinferior part of the septum and atretic plate (figure 3 and 4). Perforating the atretic plate at its thinnest inferomedial part by small suction or Cottle dissector (figure 5). Removal of posteroinferior part of the vomer (figure 6) and widening the choana laterally toward the pterygoid plate by curate in neonates or by microdebrider and drill in older children and adults. Creating a wide single neochoana (figure 7) with trial of preserving the mucosa as much as possible. Resurfacing the exposed bone with flap as much as possible. Insertion and fixation of the stents using endotracheal tube size 3-3.5 in neonate with BCA (figure 8) and size 4 – 4.5 for older children with UCA. In BCA we used 2 pieces, one in each nostril with a small piece between them to protect columella of the nose from pressure necrosis. Fixation of the stents was done using a small suture and needle (vicryl 4/0).

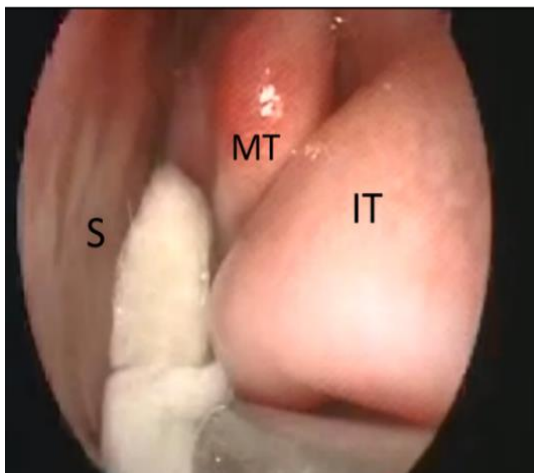


Figure 2. Application of local decongestant.
IT: inferior turbinate, S: septum, MT: middle turbinate.

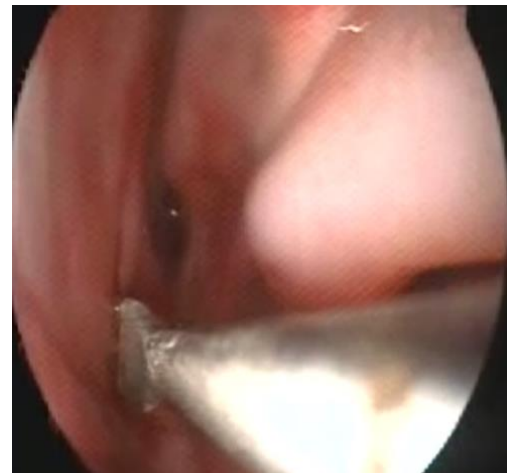


Figure 3. Incision



Figure 4. Elevation of the septal flap at posteroinferior part of the septum



Figure 5. Perforating the atretic plate at its inferomedial part.

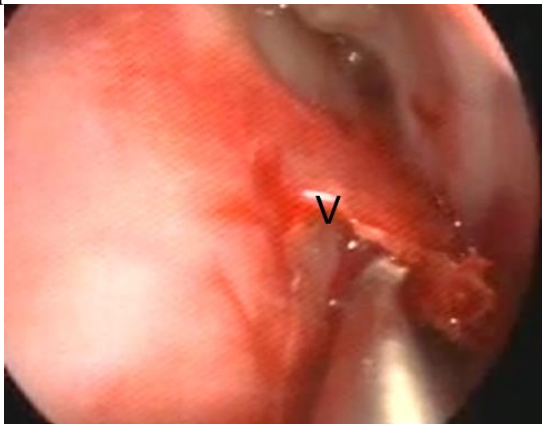


Figure 6. Removal of posteroinferior part of the vomer (V).

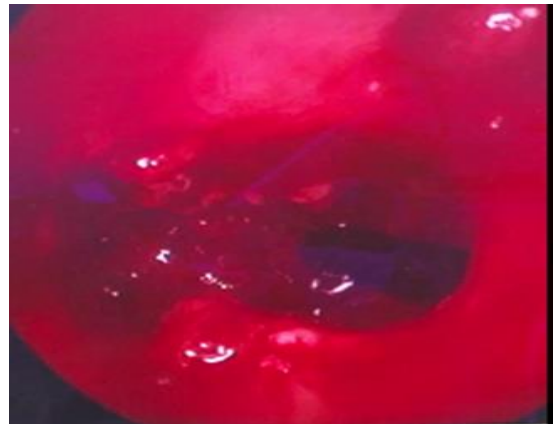


Figure 7. Creation of a wide single neochoana



Figure 8. Insertion and fixation of the stents.

3. Results:

Eighteen patients were analyzed, 13 females (72.2%) and 5 males (27.8%), age varying from 2 days up to 18 years old. Thirteen of the 18 patients were with BCA (72.2%), twelve of them (92.3%) were of mixed type (membranous and

bony) atresia and one (7.7%) was pure bony atresia. Five of the 18 patients were with UCA (27.8%), 4 of them in the right side and 1 in the left side, 3 of them of mixed type (60%) and 2 of bony type (40%). Duration of admission ranged from 0 to 5 days except 2 cases (case No 11, 13, table I) was 26 days and 14 days respectively,

because of congenital cardiac malformation and chest infection. Normal lactation was started at the second day of operation in most of the cases of BCA, except 2 cases (case No 11 and 13, table I) because of aspiration. All cases of UCA started normal lactation and/or oral feeding in the same day of operation. No significant intraoperative complications were encountered except for one case (case No 11, table I) which developed immediate post-operative bleeding from the lateral wall of the neochoana that underwent hemostasis by coagulation and received 50 ml of blood transfusion. Unfortunately, we have two cases of mortality (case 11 and 13, table I), case 11 died after 2 months of operation outside the hospital due to congenital cardiological anomalies, the other case (case 13) died in the hospital after 2 weeks of operation due to chest infection. These 2 cases were excluded from follow up results.

The complications encountered during stenting were obstruction of the stents in case No 2 (table I) one week after the operation and was replaced under general anesthesia (GA). Case No 8 (table I) developed superficial reddish hue of the nares due to large size of the stents after two days of operation which was replaced with smaller ones under GA. After removal of stents, granulation tissue formation was encountered in three cases

(2 cases of BCA, case No 8,10, table I and one case of UCA, case No 4, table II). The granulation tissue was removed at outpatient department using 0-degree endoscope of 4 mm diameter and micro-ear instruments (crocodile, cup micro-forceps and micro-suction). Intranasal synechia (figure 9) in case No 5 (table I) of BCA was found between inferior turbinate and septum which was released during revision surgery. The rate of formation of granulation tissue and synechia was (18.75% and 6.25% respectively). Significant restenosis of the neochoana (figure 10) was found in case No 5 (table I) after 1 year of operation and underwent widening using microdebrider with good result post-revision surgery. This case was associated with adenoidectomy. Complete obstruction of the neochoana was encountered in case No 3 and 4 (table I) after 1 and 2 months respectively and underwent revision endoscopic repair and stenting with good result post-revision surgery. Case No 3 was a case of BCA with right accessory large pseudo nostril and very small right proper nostril (proboscis lateralis) associated with narrow nasal cavity (figure 11) which is a very rare congenital malformation. In case No 4 the removal of stents was done after 3 days of operation; the parents did not return for follow up until 2 months later when the patient was presented with complete obstruction.

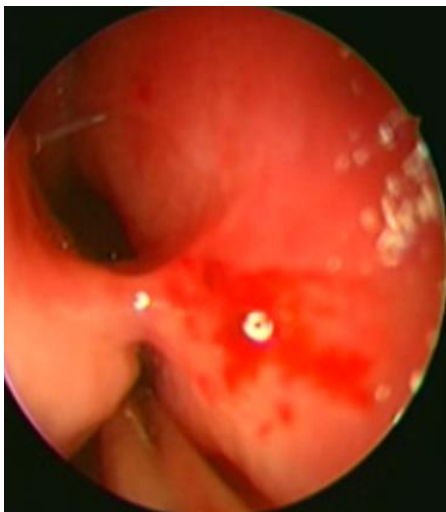


Figure 9. Synechia between inferior turbinate and septum.



Figure 10. Significant neochoanal restenosis after one year of operation in case of BCA.

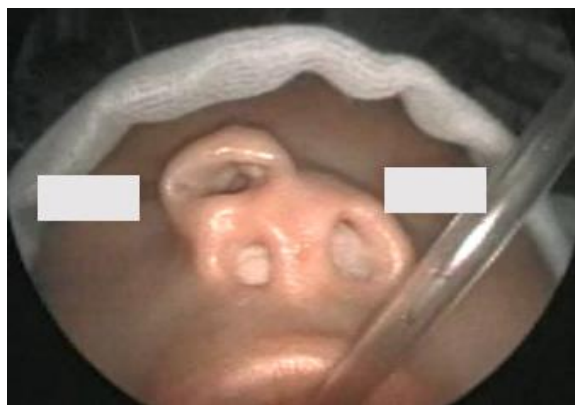


Figure 11. A case of BCA associated with right large pseudo nostril and very narrow right proper nostril (proboscis lateralis).

Table 1. Clinical characteristics and outcomes in cases of BCA

No/Sex /Age	CT scan	Associated malformation	outcomes	Revision surgery	Duration of admission
1/F/13years	mixed	Severe DNS	patent	No	2 days
2/F/3days	mixed	Nil	patent	No	3 days
3/F/7days	mixed	Proboscis lateralis, ASD, VSD and club foot.	Complete obstruction	Yes, after 1month	5 days 1day
4/F/4days	bony	Nil	Complete obstruction	Yes, after 2 months	3 days 1day
5/F/23months	mixed	Dextrocardia Palatal fistula, previous CA surgery*	Stenosis Synechia.	Yes, after 1 year.	2 days 1day
6/F/2weeks	mixed	Nil	patent	No	2 days
7/F/7days	mixed	Nil	patent	No	3days
8/F/4days	mixed	Nil	patent	No	3 days
9/F/28days	mixed	Nil	patent	No	3 days
10/F/7days	mixed	Nil	patent	No	4 days
11/M/2days	mixed	CHARGE S. +Left facial nerve palsy+ Craniosynostosis	Patent	No	26 days
12/M/45days	mixed	No	patent	No	2days
13/M/2days	mixed	No	Patent	No	14 days

DNS: deviated nasal septum. ASD: atrial septal defect. VSD: ventricular septal defect. CA: choanal atresia, *: no details, CHARGE syndrome (coloboma,

ASD, VSD, BCA, undescended testis, left auricular deformity). **Table 2.** Clinical characteristics and outcomes in cases of UCA.

No/Sex/Age	Side	CT scan	Associated malformations	outcomes	Revision surgery	Duration of admission
1/F/3 years	RT	bony	Nil	patent	NO	2days
2/F/17 years	RT	Mixed	Sever DNS	patent	NO	1 day
3/M/4 years	RT	Mixed	Nil	patent	NO	2 days
4/M/20 years	LT	bony	Severe DNS	patent	NO	0
5/F/18 years	RT	Mixed	Sever DNS	patent	NO	1 day

The success rate in cases of BCA was 72.7% (8 of 11 cases) (Success was defined as no

significant stenosis that requires another intervention and the parents' satisfaction about

child nasal breathing). The rate of significant restenosis and complete obstruction in cases of BCA after initial operation was 27.3% (3 of 11 cases of BCA). All of them underwent successful revision endoscopic repair and now are doing well with good nasal breathing. In contrast, no case of UCA developed significant restenosis or complete obstruction (zero%). The overall success rate in all cases was 81.25% (13 of 16 cases). The overall restenosis rate was 18.75% (3 of 16 cases).

4. Discussion

Bilateral choanal atresia presents as an airway emergency at birth because newborns are obligate nose breathers until mouth breathing is established with the descent of the larynx approximately 4–6 weeks of life. The treatment of BCA should be started by adequate oral airway, the preferred oral airway is McGovern nipple associated with small feeding tube to provide feeding needs [2]. In this study, 2 cases of BCA (case No 9 and 12, table I) were referred with oral airway from other locality to our hospital after 28 and 45 days of birth respectively. In some cases of BCA, endotracheal intubation (ETI) and tracheostomy may be required [2]. In this study case No 11 and 13 presented with ETI in the neonate unit because of significant respiratory distress not improved by oral airway. Asher et al. (1990) recommended to do tracheotomy for the patients with BCA associated with CHARGE syndrome due to high failure of repair because of narrower posterior choanal region than those with isolated CA [6,7]. None of the cases of BCA in our study underwent tracheotomy before endoscopic repair. Eladl and Khafagy (2016) recommend that the preoperative investigations for BCA should include echocardiography [8]^[8]. In this study, all cases of neonates with BCA underwent echocardiography to exclude congenital heart diseases. Unilateral choanal atresia does not present as an emergency, and it is diagnosed and treated more often in childhood than in infancy [9,10]. In our study, cases of UCA were diagnosed at 3, 4, 17, 18 and 20 years old.

Surgical repair of CA.

There are multiple surgical techniques to correct CA: Transnasal puncture, transpalatal and transnasal endoscopic technique. Transnasal puncture technique is a blind maneuver that needs multiple revisions and dilatations; and associated with risk of CSF leakage and meningitis [2,11]. Thus, we think it is contraindicated in the era of endoscope. The transpalatal approach is more destructive and carries risk of bleeding, airway obstruction due to edema, fistulas, necrosis of palatal mucosa, velopharyngeal insufficiency, growth defects of palatal bone and crossbite [2,5]. Some authors recommend it in children above 6 years old or with recurrent cases [2,10]. We think it has no indication in the era of endoscope because endoscopic repair showed good results even in recurrent cases. Transnasal endoscopic repair of CA was first demonstrated by Stankiewicz in 1990 and has become the primary procedure by most surgeons [11]^[11]. Kamel (1994) reported that it is the most direct approach to the posterior nasal choana and is performed completely under excellent visualization [12]. It is safe, effective with minimal complications, helps to preserve the mucosa and avoid injury to nearby important structures [8,12,13]. Bleeding can be minimized by avoiding working laterally and unlike transpalatal repair there is no risk of abnormal midfacial growth or occlusal abnormalities [14,15]. In our study all cases underwent transnasal endoscopic repair of CA. None of the cases in this study developed midfacial growth or occlusal abnormalities.

Ramseden et al. (2009) reported that the lateral wall is the principle challenge of choanal atresia surgery, as most surgical corrections tend to address the septum and atretic plate only [1]^[1]. In this study all cases underwent removal of posteroinferior part of bony septum (vomer), atretic plate and lateral widening toward pterygoid plate as much as possible. According to Teissier et al. (2008), to evaluate the choanal patency, they recommend passing a flexible, 4-mm, fiberoptic endoscope non traumatically

through the passage. If no rubbing occurred on the edges of the choanae during this passage, the child was considered asymptomatic [16]. In our study, we depended on the family opinion about the adequate improvement of symptoms such as nasal breathing, suckling and the absence of nasal discharge which are confirmed by nasal endoscopic examination.

The success and restenosis rate in CA repair

The success and restenosis rate of CA surgery can be very variable [17, 18]. Durmaz et al. (2008) presented a study with meta-analysis and found that the postoperative success rate in a total of 238 patients with CA was 85.3 % and the rate of restenosis was 14.7% [19]. Several studies reported that the restenosis rate is higher in cases of BCA ranging from 14 to 45% while in UCA cases ranging from 0 to 20% [16,18 ,19,20,21]. Attia et al. (2021) presented a case series of 42 patients where the restenosis rate was 72%. They stated that this variation is probably caused by the difference in the study population size, disease characteristics and surgical techniques [17]^[17]. In our study, the overall success rate in all cases from the initial operation was 81.25% (13 of 16 cases). The overall restenosis rate was 18.75% (3 of 16 cases). The rate of significant restenosis in cases of BCA after initial operation was 27.3% (3 of 11 cases of BCA), while no case of UCA developed significant restenosis (zero%). We noted that restenosis in our study occurred in cases of BCA between 2014 and 2017. This may reflect the effect of the age factor and our limited experience at that time, which agrees with Tiessier et al. (2008) when they reported that the surgeon's learning curve and the age factor are important elements that seem to influence surgical outcome [16].

Timing of operation

Limited studies recommend that the preferred time of BCA repair is between the 10th to 13th days of age even in premature newborns [22]. However, Murray et al. (2019) stated that in addition to definitive relief of respiratory distress, immediate surgical intervention for

BCA offers a lower treatment failure rate as compared to delayed surgical intervention [23]^[23]. We noted that the repair of BCA in neonates at 10 days of age and above may give better access to the choana and facilitates handling of instruments. On the other hand, the delay of repair exposes the neonates to hazards of respiratory distress, difficulty in feeding and prolongs the period of admission in neonate unit. Moreddu et al. (2019) recommend delaying surgery for UCA after age 6 months and/or weight >5 kg when possible [10]^[10]. Murray et al. (2019) concluded that there was no difference in outcomes for patients with UCA treated surgically in the immediate or delayed setting, after careful discussion of surgical risks and benefits, timing of surgery should be guided by patient goals and symptoms [23]^[23]. We think the delay of repair in UCA is better, to avoid anesthetic risks, enhance the visualization of the choana and ease the follow up.

Intranasal stenting

Intranasal stenting is an issue of controversy, several systematic reviews with meta- analysis [17,24 ,25] concluded no significant decrease of restenosis for CA repaired with and without the use of nasal stents. Strychowsky et al. (2016) conducted a systematic review and meta-analysis on the efficacy and safety of stenting in the endoscopic repair of congenital BCA. They found that stenting may be associated with more complications, but they concluded that the use of stenting should be considered in the context of the individual patient and surgeon preference [25]. Eladl and khafagy (2016) recommend using stenting in some selected cases such as: narrow choana after drilling, difficult follow up situations and patients with associated congenital anomalies [8]. Attia et al. (2021) recommend routine stenting for neonates with BCA for 4–6 weeks [17], while Urbancic et al. (2023) recommend the use of stent for 7 days [22]^[22]. In this study we used intranasal stents for 4 weeks to avoid sudden obstruction, respiratory distress, and prolonged admission in the neonate unit as

well as difficulty in endoscopic follow up in the first month of postoperative period.

Flap preservation

Another controversial issue is the preservation of the flap. There are different described techniques for incision and creation of flaps for resurfacing the surgical defect. These include: four flaps with cruciate incisions [12], side-hinged double flap, single side hinged flap, upper hinged flap [21,26], mirrored L- shaped septonasal flap [27]^[27] and multiple flaps secured with fibrin glue. None of these techniques has become standard. Durmaz et al. (2008) and El-Ahl et al. (2012) reported that resurfacing the surgical defect by mucosal flap does not seem to significantly influence the surgical outcome [19, 28]. Romeh and Albirmawy et al. (2010) noted that even in revision surgery of the relapsed cases using the same technique without mucosal flap was successful 100% [20]. In our study we found difficulty in fixing the flap in the defect especially in cases of neonates with BCA, so we depended on creating a single wide neochoana with minimizing the mucosal destruction to prevent restenosis.

The application of mitomycin C in the neochoana does not prevent restenosis as stated by many systematic reviews and meta- analysis [17,24,25], so it was not used in this study.

Postoperative complications

Strychowsky et al. (2016) reported that restenosis is the most frequent complication of CA repair regardless of the surgical technique used. However, the etiopathology of restenosis is incompletely understood [25], multiple studies stated that an increased rate of restenosis was found in pure bony type of CA, patients with BCA, association of GERD, inadequate bone resection and early developing experience of the surgeons so they recommend counselling the parents regarding the likelihood of revision surgery [9,17,20]. In our study restenosis occurred in BCA only (3 cases). Although the number of cases in our study is small, multiple

studies are in consistent with our results [17,18,19,20,21]. We believe that the rate of restenosis in cases of BCA is higher than those of UCA because the created neochoana has raw area all around, whereas the raw area in UCA present only in one side. In this study, minor complications were found during follow up including granulation tissue (in 3 cases, 18.75%), and partial synechiae (in one case, 6.25%) between nasal septum and inferior turbinate. According to multiple studies, the rate of granulation tissue formation and synechia is very variable regardless of the surgical techniques used and ranges from 7 % to 53 % [19, 20, 27]. This may confirm that the stentless technique and using of specific flap do not prevent granulation tissue formation and intranasal synechia. Therefore, good post-operative follow up is the key to improve the outcome of CA surgery. In our study, no serious complications were encountered as CSF rhinorrhea or significant bleeding during operation.

Postoperative admission period

Kamel (1994) reported that transnasal endoscopic repair is performed as a same-day surgery [12]^[12]. His study was undertaken on 6 patients with UCA aged between 4 and 18 years which is not applicable on neonates and infants with BCA, thus the period of admission depend on the clinical characteristics of each patient. Cases of BCA associated with congenital cardiological and craniofacial malformations may need prolonged admission. In this study, all cases of BCA were admitted in neonate unit for 2 to 5 days except those with other comorbidities that were admitted for 5 to 26 days (case 3, 11 and 13, table I), whereas cases of UCA were admitted for 0 to 2 days (table II). So, we cannot expect period of admission and this should be explained to the family.

Postoperative follow up and family education

Follow up and family education have major impact on the surgical outcome. After surgery, endoscopic follow-up helps detect and treat any mucosal adhesions, granulations, and/or crusting

to ensure choanal patency [12]. Others recommend revision endoscopy to remove crusts 1 week after surgery [16]. In our study, we relied on family education by teaching the parents how to take care of the neonate through observation of skin color of nostril to avoid pressure necrosis, application of local antibiotic ointment around the stents as well as saline irrigation inside and around the stents with gentle suctioning inside the stents only to avoid crust formation and to keep the nose patent. Nasal irrigation should be continued even after removal of stents, as studies prove that abundant nasal washing is one of the keys to successful management of congenital CA [16]. In this study, follow up was done weekly for the first month to avoid complications of the stents; and monthly after removal of the stents for 3 months by endoscopic examination to remove any granulation tissue.

5. Conclusion:

Endoscopic CA repair is safe, effective with less morbidity. A wide single neochoana with removal of posterior part of the vomer, atretic plate and lateral widening toward the pterygoid plate followed by stenting with close follow up give good success rate. The main factors that may affect outcomes are age and laterality as neonate cases of BCA are more prone to restenosis and complete obstruction that need revision surgery, this should be explained to the family. The increase of experience of surgeon affects positively the result of CA repair. Stenting in our study gave good results and there were no significant complications specially with close follow up and family education.

6. Recommendations:

- 1- The challenge in surgery is in BCA cases especially in neonates and infants so we recommend that the results should be presented separately in each type of CA studies.
- 2- Close follow up and family education about the care of stents are essential to avoid complications of the stents and improve the results.

- 3- Preservation of the flap, resurfacing and fixing it on the defect of the created neochoana need more studies to evaluate its outcome.

Funding

Data collection and writing of manuscript done by authors with no payment received from any source.

Conflicts of interest

The authors declare no conflicts of interest.

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