Choanal Atresia Repair in Pediatric Patients: Are Use of Stents Recommended or Not

Abdullah Albdah[1], Maram Alanbari[2] and Fahad Alwadi[3]

[1]Department of ORL, King Salman Hospital, Saudi Arabia

[2]Department of ORL, General Directorate of Health Affairs, Saudi Arabia

[3]King Salman Hospital, Ministry of Health, Riyadh, Saudi Arabia

Submission: January 16, 2019; **Published:** January 23, 2019

*Corresponding author: Abdullah Albdah, Department of ORL, King Salman Hospital, Saudi Arabia

Abstract

Choanal atresia, the obliteration or narrowing or nasal choana, is widely studied across pediatric and adult population. While unilateral choanal atresia can remain unidentified for several years, bilateral choanal atresia requires immediate intervention as children are primarily nasal breathers till initial 4 months of age. Several surgical methods are reported for repairing choanal atresia in children, and the choice of postoperative management with or without stent is still controversial. In this review we analysed several recent studies in which the surgery for choanal atresia repair was followed by stent-assisted and/or stentless management. The results of this study are likely to pave the way for further understanding of the choice of methods to increase patency and reduce the possible complications.

Introduction and Background

Choanal atresia (CA) is a life-threatening, but relatively uncommon, anomaly involving the congenital obliteration or narrowing of the posterior nasal choana, resulting due to the blocked oronasal membrane in children [1]. The incidence of CA is one in five to nine thousand live births [2]. The nasal obstruction in CA can be either bony or membranous; while early reports suggested bony and membranous obstruction as 90% and 10%, respectively [3], more recent studies have indicated 70% mixed bony and membranous and 30% pure bony [4]. CA can also be categorized as unilateral, bilateral, or due to other craniofacial abnormalities. Unilateral CA affects only one nostril, usually the right nostril more, in the ratio of 2:1 [1]. In such cases, the child is often undiagnosed, and the requirement for treatment is realized only when nasal obstruction and unilateral rhinorrhoea is observed. In contrast, bilateral CA can be identified right after birth in most cases as infants preferably breathe from the nose until four months of age due to a high cervical location of the larynx [5]; therefore, bilateral CA in neonates is considered lethal. Children with craniofacial abnormalities form a very small subgroup of the population and have thick pterygoid lamina [6].

Surgically, CA can be managed with various procedures, and the success is primarily measured based on the "patency" of the choana, although several studies have also considered a number of operations, duration of stenting, etc. among other measures. Although the treatment techniques for CA have been extensively reported, there is no consensus on the choice of treatment as the outcomes are affected by several other conditions, such as feeding patterns and global progress, which in turn affects the overall development of the patients. Nevertheless, the various surgical strategies to treat CA include transnasal puncture, transpalatal resection, and endoscopic resection, which can be supported with or without stents [6]. Traditionally, stenting of the opened choanahas played a key role in the postoperative management of CA. Park et al. [7] reported that ninety-two of ninety-five 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 [8,9]. However, the potential advantages of using stents include avoidance of early restenosis, assisting the healing of mucosal flaps, and prevention of post-surgery edema [10]. The present review provides a comprehensive study of the management of CA with or without stent in the pediatric population based on the literature and identifies the potential areas of future studies.

Review Methods

Papers relating to CA repair in pediatric patients with or without the use stent of were identified from Google Scholar, PubMed (up to 2019), and World Wide Web. The relevant titles and abstracts were searched using the following key terms "Choanal atresia," "stent," "stentless," "pediatric." Studies pertaining to "adults," "craniofacial abnormalities," and use of "mitomycin C" were excluded. Furthermore, literature references

were thoroughly scanned for additional papers, and relevant articles were also studied. The principal search criterion for every article was CA-related outcomes with or without stents in pediatric populations, and the relevant articles have been included in the review.

Results

Several studies have compared the patient outcomes pertaining to repair of CA in the pediatric population with or without the use of stents. The relevant papers were analyzed and included in the study.

Stent-Assisted Repair of Choanal Atresia

Several studies have reported the success of transnasal endoscopic CA repair with the use of stents. The favorable outcomes largely depend on multiple factors such as the material of stent, the positioning of stent, the duration of stenting, and the type of stent. One such study by Nithyasundar & Narayanan [11] described the role of the material of stent in the success of the surgical outcome. Stents made of soft polyvinyl chloride (Portex) were used in patients aged six days to two years, of whom two had bony CA, one had bony membranous, and one had membranous CA. Also, two patients had bilateral CA, and two had unilateral CA. Stents were used in all cases, and the patients were followed up postoperatively for one to two years. The surgery outcomes showed 100% success, and revision surgery was not required in any case. The authors concluded that several factors played the key role in improving the success rate, such as the use of Portex stents, broad-spectrum antibiotics throughout the stenting period, and minimum four weeks of stenting, and thereafter gentle removal of the stent to prevent trauma and bleeding. Another study by Rodriguez et al. [12] retrospectively evaluated forty-nine pediatric patients of age three days to thirteen years with CA, who were treated with the transnasal microscopic method, and silicone endonasal stent was placed for one to twelve weeks. Of these, thirty-five patients required revised surgery, and nine had complications. Overall, the study found stent-assisted repair to be successful and effective in the pediatric population.

The positive effect of duration of stenting on surgical outcome was demonstrated in a study by Freitas & Berkowitz [13], who reported stent-assisted transnasal endoscopic repair of CA in twenty-three neonates of age zero to thirty-two days. All patients were stented for an average of seventy-five days, of which twenty were sufficiently followed up. Of these, six required revision surgery, three had one revision procedure, and three had no revision procedure. The study concluded that stenting for a period of three months decreased the chances of early stenosis. In another interesting case, Gupta & Kaur [14] presented the case study of a newborn baby with bilateral CA, who was treated with transnasal endoscopy with stents. The stents were kept for four weeks, and the child was followed up for one year with no complications. Similar results were reported by Riepl et al. [15] who treated six patients of bilateral

CA of age three days to two months. Transnasal endoscopy was performed, and stents were placed in all the cases for at least six weeks. The study suggested that bilateral stents should be used, particularly in very young patients to avoid early restenosis. Comparable to the abovementioned study, Romeh & Albirmawy [16] also retrospectively analyzed cases which performed the stent-assisted transnasal endoscopic repair for management of CA. In total, the cases of fifty-four children of age seven days to fourteen years were analyzed, the duration of stenting was five to seven days to avoid any complications. Thus, the shortened period of stenting was reported to be one of the factors resulting in successful patient outcomes.

Standard endoscopic techniques are typically deployed to manage CA. However, Jones et al. [2] reported an exceptional case of bilateral CA an infant with CHARGE syndrome (coloboma, heart defects, CA, retardation of growth, genital abnormalities, and ear abnormalities) and difficult anatomy of the skull base. A curved mastoid burr was utilized; stents were placed on day one post operation and were removed after one week. Breathing and feeding were found normal. After one month, the patient reported increased work of breathing and nasal congestion, which was treated well. After three months, revised choana displayed atretic bone; therefore, treatment with the curved burr was repeated, and the child was successfully treated. Recently, advancements in the type of stents used have shown favorable outcomes and reduced the otherwise commonly occurring side-effects of using traditional stents. For example, steroid eluting stents have shown promising results in postsurgical maintenance of sinus ostia patency and synechiae prevention in adults [17,18].

Bangiyev et al. [19] utilized mometasone furoate steroid eluting stents (Propel, Intersect ENT, Menlo Park, California, USA) to treat CA in three pediatric patients of age two years, one day, and sixteen years hoping to avoid postsurgical stenosis. The first two cases were identified with bilateral CA, whereas the third case exhibited unilateral CA. Case 1 had membranous atresia, case 2 had bony atresia, whereas case 3 had mixed type atresia. The stent was successfully deployed in all the cases, and it was found safe and effective with no restenosis in any patient with twelve months follow up. Interestingly, the steroid stent was used "off-label" as it has been Food and Drug Administration (FDA) approved only for the adult population. As a topical nasal spray, the steroid has been FDA approved at 100 μg per day dosage for children of age more than two years; however, it is presently not approved for children of age less than two years. Therefore, the authors recommended further studies on safety profiling and analyzing long-term effect and efficacy in the pediatric population.

Stentless Repair of Choanal Atresia

Although numerous studies have reported the success of using stents post-surgery in the repair of CA, several studies have also reported favorable outcomes without the use of stents. In a

retrospective study, Brihaye et al. [20] comprehensively analyzed the outcomes of surgery for CA without the use of the stent in thirty-six children from 1999 to 2015, with 50% unilateral and 50% bilateral cases. Fibrin glue was used to attach the mucosal flaps, and the patients were followed up for six years on average. The authors concluded that surgery was safe for newborns and that restenosis could be minimized by not using the stents and following proper management procedures. Similarly, another successful surgery was documented by Saitabau [21], who presented the case of a sixteen year old girl with bilateral congenital CA of a mixed type who was treated at the Muhimbili National Hospital, Tanzania. Atretic choana was endoscopically treated without stenting under general anesthesia.

The repair of CA by transnasal endoscopy has been widely accepted. Schoem [9] retrospectively reviewed the outcomes without stent in thirteen children aged two to thirteen days with unilateral and bilateral CA. All the patients were given combinations of oral antibiotics, oral steroids, and topical nasal steroids. The study concluded that repair of CA by transnasal endoscopy is safe and effective without the use of stent. El-Ahl & El-Anwar [22] also evaluated stentless repair of CA by transnasal endoscopic approach. The study involved seven patients of age three–fifteen days with bilateral CA. The surgery avoided stent-related complications, and the study concluded that the technique was effective with good patency and faster recovery of the patients. Similarly, El-Anwar et al. [23] also presented a study of 25 patients of age range three to fifteen years, in whom bilateral CA was repaired via stentless transnasal endoscopic approach. During follow-up, wide choana was seen in eighteen patients, narrow choana in six, and restenosis in one patient. Overall the results were found satisfactory without using stents. The transseptal approach to repair CA without the use of stents was reported by Wormald et al. [24] in sixteen pediatric patients and one adult patient. In total, seven cases were bilateral, and ten were unilateral. The study reported the requirement of post operation transfusion due to intraoperative bleeding in two neonates and postoperative respiratory complications in two patients. The study recommends following stentless approach for lasting patency.

Comparative Studies of CA Repair with and Without Stent

Several studies have comprehensively evaluated the effect of stent-assisted and stentless postoperative management by comparing the two in the same patient population. While many studies have reported similar success rate with or without the use of stents, others have reported the advantage of stentless method over stent-assisted approach. Wolf et al. [10] retrospectively reviewed the cases of pediatric patients of age less than eighteen years, who underwent endoscopic surgery with or without stents from 2001 to 2012 at their department. The pediatric population treated comprised of both unilateral and bilateral CA of mixed and bony type, and a 100% patency rate was reported in both the groups with no significant differences. The study

concluded that outstanding postoperative results could be accomplished both with and without stents. Also, stents were recommended in newborns to prevent restenosis and following lethal complications. However, in children of age more than one year with mild symptoms, stents were not recommended due to the possibility of complications. Similar results were also reported by Moreddu [25] in a review in which stenting was performed in eighty-five of one hundred fourteen patients from November 1986 to November 2016. The study concluded that no stent was equally safe as stent as no significant difference was seen in terms of the number of procedures or patency. However, stent duration (mean 27.7 days) was negatively related with the long-term analysis. In another retrospective study, Kim et al. [26] evaluated the factors influencing the outcomes of surgery on patients aged six days to twenty-eight years.

No significant differences in surgical outcomes were seen with or without the use of stent as the rate of restenosis was 42.9% with stent and 47.4% without the stent. Tomoum et al. [27] reported a comparative evaluation of endoscopic repair of CA in seventy-two newborns using mirrored L-shaped flap without stenting versus stenting without a flap. Patient follow up revealed higher granulation tissue in patients with stent (53.3%) than without (28.6%) and 33.33% stenosis in patients with stent than 21.40% in without stent. The study concluded that stentless endoscopic repair with a flap was safe and effective. Another study by Newman et al. [28] compared stent-assisted and stentless management of CA in forty-two children of age three days to fifteen years. Stents, when used, were kept for 15 days or more; however, the differences were not significant between the groups. However, the study also suggests the basis of results to be a potential bias as stents were deployed based on the severity of the disease and therefore recommends that stents should be used or not based on the specific case.

The advantage of stentless method over stent-based method was reported by Saafan [29] in twenty children, ten with and ten without stents. The stent was kept for four weeks and follow up for one to two years revealed that choanal narrowing, stenosis, and granular tissue formation was significantly higher in the stent-assisted group, whereas no significant differences were observed in terms of closure. Thus, the study concluded that the use of stents might not be necessary for the repair of bilateral CA. Similar results were reported by Uzomefuna et al. [30] for stent-assisted repair of CA via transnasal endoscopy in thirty-one children of age one day–fifteen years. The incidence of restenosis was higher in cases with stents;80% for age less than ten months and 57% for stents for four to six weeks, in comparison to those with no stents showing 33.3% restenosis, thus concluding that restenosis cases were higher with the use of intranasal stents. Furthermore, Eladl & Khafagy [31] presented a retrospective study of one hundred twelve infants of age one to twenty-eight days who underwent endoscopic treatment for the repair of bilateral CA with or without stent. Restenosis was observed in 42% patients, and 74.5% patients in the stent group

required second evaluation in comparison with 20.6% on the non-stent group. The patients had stents was nearly two to six weeks. The study reported that higher success rate was achieved without the use of stents.

Discussion

The treatment strategies for CA have been described since the mid-19th century; however, the choice of ideal technique is still debated. The present review focused on stent-assisted and stentless repair of CA in the pediatric population. A wide variation in the techniques and outcomes was seen, and several factors were observed to contribute to the success of the surgery. Ideally, the success of the surgery is measured in terms of the higher patency and lower incidence of stenosis, hospital stay, morbidity, and mortality [32]. Several techniques have been reported for the treatment of CA, of which the most commonly used are endoscopic transnasal, trans septal, and trans palatal. The success of CA depends on the surgery as well as the minimal or absence of postoperative complications. The choice of using stent or not has been widely discussed in the literature, and although there appears to be no consensus, certain factors are known to increase the chances of success with minimal complications, such as the choice of the material of stent, the duration of stenting, patient age. The most commonly observed complications following the surgical correction of CA are stenosis and granulation tissue. While these have been commonly reported with the use of stents, some studies have suggested that the use of soft material stents than traditional stents could reduce the complications. For example, Lazar and Younis [33] and Nithyasundar & Narayanan [11] reported exceptional results with the use of Portexpolyvinyl chloride stents, whereas Rodriguez et al. [12] reported the use of silicone stents, and Bartel [34] suggested the use of Foley catheter to avoid stent-related complications. In addition, the "off-label" use of drug-eluting stents also gave promising results, though further studies are required to confirm the same [19].

Stent duration and outcome of the duration of stenting was found to be another factor which affected the successful repair of CA in pediatric patients.

The studies' results were found controversial. Most of the studies reported successful outcomes with longer periods of stenting; Nithyasundar & Narayanan [11] recommended minimum four-weeks of stenting, and longer duration of stenting with no complication was seen in studies by Rodriguez et al., Freitas & Berkowitz, Gupta & Kaur, and Riepl et. al., who deployed the stents for one–twelve weeks, eleven weeks, four weeks, and six weeks, respectively [12-15]. However, some studies also reported the shorter duration of stenting as the possible reason for the successful outcome. The study by Romeh & Albirmawy [16] deployed stents for five-seven days and found favorable outcomes in the patient population. However, some studies have recommended three and six weeks of stenting for unilateral and bilateral CA, respectively [34], although other studies suggest

that the duration of stenting should be individualized till the development of mucosa [33].

The stent-associated success of CA repair was also found to vary depending on the age of the patient at the time of surgery. Wolfet al. [10] reported that the use of stents is the ideal treatment of choice for newborns to avoid restenosis and other complications and also for sufficient weight gain before the surgery and that in children more than one-year-old with mild symptoms, stents should be avoided due to possible complications. Thus, while several studies have shown superior results with stentless surgery in comparison with stent-assisted surgery, the reason could be due to the age of the pediatric population studied. This could be the explanation for the observations by Saafan [29], where the use of stent in children aged 1-5 years showed significantly higher complications. However, no such direct evidence exists confirming the proportional risk of stent-assisted surgery with age, and therefore, the choice of postoperative management for CA repair often depends on the surgeons' preference [35]. This review had a few limitations. First, we excluded stentless surgery with mitomycin treatment, which could affect the possible success rate observed with stentless management. Second, these observations are not applicable to every case of postoperative management of CA repair due to the possible effect of other previously mentioned factors. Third, some articles could have been missed and not included in the study. Therefore, further studies are required to establish a standard procedure to be followed for the postoperative management of CA repair.

Conclusion

We believe that the surgical outcomes of CA repair without stents reduced showed fewer complications, thus decreasing the intensity of postoperative management. The stent-assisted procedure could be preferred for younger patients with no complications; however, the use of the stent should be evaluated for every case as several factors affect the patency. However, further high-level studies are required to draw clear conclusions.

References

1. Anajar S, Hassnaoui J, Rouadi S, Abada R, Roubal M, et al. (2017) A rare case report of bilateral choanal atresia in an adult. Int J Surg Case Rep 37: 127-129.

2. Jones DJ, Vandjelovic ND, Gonik NJ (2018) Novel use of a curved mastoid burr in the management of a difficult case of choanal atresia. Otolaryngology Case Reports.

3. Fraser JS (1910) Congenital atresia of the choanae. Br Med J 2: 1698-1701.

4. Brown OE, Pownell P, Manning SC (1996) Choanal atresia: a new anatomic classification and clinical management applications. The laryngoscope 106 (1 Pt 01): 97-101.

5. Voegels RL, Chung D, Lessa MM, Lorenzetti FT, Goto EY, et al. (2002) Bilateral congenital choanal atresia in a 13-year-old patient. Int J Pediatr Otorhinolaryngol 65(1): 53-57.

6. Ramsden JD, Campisi P, Forte V (2009) Choanal atresia and choanal stenosis. Otolaryngol Clin North Am 42(2): 339-352.

7. Park AH, Brockenbrough J, Stankiewicz J (2000) Endoscopic versus traditional approaches to choanal atresia. Otolaryngol Clin North Am 33(1): 77-90.

8. Van Den Abbeele T, François M, Narcy P (2002) Transnasal endoscopic treatment of choanal atresia without prolonged stenting. Arch Otolaryngol Head Neck Surg 128(8): 936-940.

9. Schoem SR (2004) Transnasal endoscopic repair of choanal atresia: Why stent? Otolaryngol-Head and Neck Surg 131(4): 362-366.

10. Wolf A, Lang-Loidolt D, Koele W, Wolf G, Thurnher D, et al. (2016) Are stents beneficial in endoscopic choanal atresia repair of newborns and children? Case series of 11 patients. Clin Otolaryngol 41(6): 821-825.

11. Nithyasundar A, Narayanan DS (2016) Choanal Atresia: Experience with Transnasal Endoscopic Technique. J Pharm Sci Res 8: 86.

12. Rodríguez H, Cuestas G, Passali D (2014) A 20-year experience in microsurgical treatment of choanal atresia. Acta Otorrinolaringologica English Edition 65(2): 85-92.

13. De Freitas RP, Berkowitz RG (2012) Bilateral choanal atresia repair in neonates-a single surgeon experience. Int J Pediatr Otorhinolaryngol 76(6): 873-878.

14. Gupta M, Kour C (2017) Congenital Bilateral Choanal Atresia. A Rare Case. J Rare Disor Diagn Therap 3: 9.

15. Riepl R, Scheithauer M, Hoffmann TK, Rotter N (2014) Transnasal endoscopic treatment of bilateral choanal atresia in newborns using balloon dilatation: Own results and review of literature. Int J Pediatr Otorhinolaryngol 78(3): 459-464.

16. Romeh HE, Albirmawy OA (2010) A 13-year experience and predictors for success in transnasal endoscopic repair of congenital choanal obliteration. Int J Pediatr Otorhinolaryngol 74(7): 737-742.

17. Forwith KD, Chandra RK, Yun PT, Miller SK, Jampel HD (2011) ADVANCE: a multisite trial of bioabsorbable steroid-eluting sinus implants. The Laryngoscope 121(11): 2473-2480.

18. Murr AH, Smith TL, Hwang PH, Bhattacharyya N, Lanier BJ, et al. (2011) Safety and efficacy of a novel bioabsorbable, steroid-eluting sinus stent. Int Forum Allergy Rhinol 1(1): 23-32.

19. Bangiyev JN, Govil N, Sheyn A, Haupert M, Thottam PJ (2017) Novel application of steroid eluting stents in choanal atresia repair: a case series. Ann Otol Rhinol Laryngol 126(1): 79-82.

20. Brihaye P, Delpierre I, De Villé A, Johansson AB, Biarent D, Mansbach AL (2017) Comprehensive management of congenital choanal atresia. Int J Pediatr Otorhinolaryngol 98: 9-18.

21. Saitabau Z, Elimath M, Moshi N, Richard E, Ntunaguzi D (2018) Bilateral congenital choanal atresia in a 16-year old girl at Muhimbili National Hospital, Tanzania. Tanzania J Health Res 20: 1-6.

22. El-Ahl MA, El-Anwar MW (2012) Stentless endoscopic transnasal repair of bilateral choanal atresia starting with resection of vomer. Int. J. Pediatr. Otorhinolaryngol 76(7): 1002-1006.

23. El-Anwar MW, Nofal AA, El-Ahl MA (2016) Endoscopic repair of bilateral choanal atresia, starting with vomer resection: Evaluation study. Am J Rhinol Allergy 30(3): 95-99.

24. Wormald PJ, Zhao YC, Valdes CJ (2016) The endoscopic transseptal approach for choanal atresia repair. Int Forum Allergy Rhinol 6(6): 654-660.

25. Moreddu E, Rossi ME, Nicollas R, Triglia JM (2018) Prognostic Factors and Management of Patients with Choanal Atresia. J Pediatr 204: 234-239.

26. Kim H, Park JH, Chung H, Han DH, Kim DY, et al. (2012) Clinical features and surgical outcomes of congenital choanal atresia: factors influencing success from 20-year review in an institute. Am J Otolaryngol 33(3): 308-312.

27. Tomoum MO, Askar MH, Mandour MF, Amer MA, Saafan ME (2018) Stentless mirrored L-shaped septonasal flap versus stented flapless technique for endoscopic endonasal repair of bilateral congenital choanal atresia: a prospective randomised controlled study. J Laryngol Otol 132(4): 329-335.

28. Newman JR, Harmon P, Shirley WP, Hill JS, Woolley AL, et al. (2013) Operative management of choanal atresia: a 15-year experience. JAMA Otolaryngol Head Neck Surg 139(1): 71-75.

29. Saafan ME (2013) Endoscopic management of congenital bilateral posterior choanal atresia: value of using stents. Eur Arch Otorhinolaryngol 270(1): 129-134.

30. Uzomefuna V, Glynn F, Al-Omari B, Hone S, Russell J (2012) Transnasal endoscopic repair of choanal atresia in a tertiary care centre: a review of outcomes. Int J Pediatr Otorhinolaryngol 76(5): 613-617.

31. Eladl HM, Khafagy YW (2016) Endoscopic bilateral congenital choanal atresia repair of 112 cases, evolving concept and technical experience. Int J Pediatr Otorhinolaryngol 85: 40-45.

32. Pirsig W (1986) Surgery of choanal atresia in infants and children: Historical notes and updated review. Int J Pediatr Otorhinolaryngol 11(2): 153-170.

33. Lazar RH, Younis RT (1995) Transnasal Repair of Choanal Atresia Using Telescopes. Arch Otolaryngol Head Neck Surg 121(5): 517-20.

34. Bartal M (1988) Improvement stent for use in management of congenital choanal atresia. J Laryngol Otol 102(2): 146-147.

35. Josephson GD, Vickery CL, Giles WC, Gross CW (1998) Transnasal endoscopic repair of congenital choanal atresia: long-term results. Arch Otolaryngol Head Neck Surg 124(5): 537-540.