SMJ Saratov Medical Journal
ISSN 2712-8253, Saratov Medical Journal
2024. Volume 5. Issue 3. Article CID e0301
DOI: 10.15275/sarmj.2024.0301

Original article

Reprint

Evaluating the efficacy of surgical treatment in infants with choanal atresia

Olga A. Breeva[1] ✉, Mihail M. Polunin [1,2], Alan I. Asmanov [1]
✉ breevaluka77@mail.ru

[1]Russian National Research Medical University, Moscow, Russia
[2]Morozovsky Children's Clinical Hospital, Moscow, Russia

Received 29 January, 2024, Accepted 9 July 2024

Abstract:
Objective: to evaluate the efficacy of stentless endoscopic surgery for congenital choanal atresia (CCA) in infants.
Materials and Methods. Our study involved 51 infants under 1 year of age who underwent stentless choanoplasty. Main group: infants operated for the first time (n=27). Comparison group: infants with relapse after traditional choanoplasty with stents (n=24). Endoscopic examination and saturation levels were employed to evaluate treatment results. Quality of life was assessed using la Qualité de Vie du Nourrisson questionnaire for children from 3 months of age to 1-3 years of age. Parents of 45 infants participated in the survey: 25 infants were from the main group and 20 infants represented the comparison group.
Results. No signs of restenosis were detected during endoscopic examination of the nasal cavity and nasopharynx in 50 patients during follow-up observation (1-2 years). Saturation after surgery increased from 98.4±2.4% to 99.4±1.2% (p=0.001) in patients of the main group and from 93.5±4.7% to 99.0±1.0% (p<0.001) in the comparison group. Assessment of quality-of-life values after surgical treatment by the stentless method showed an increase from 14.4±0.4 to 15.3±1.8 (p=0.056) in the main group and from 14.0±0.4 pts to 15.6±1.0 pts (p<0.001) in the comparison group.
Conclusion. Endoscopic endonasal stentless method of surgical treatment of CCA in infants is characterized by higher efficacy vs. traditional choanoplasty with stenting.

Keywords: choanal atresia, choanoplasty, infants, restenosis, fibrin glue, la Qualité de Vie du Nourrisson questionnaire

Cite as: Breeva OA, Polunin MM, Asmanov AI. Evaluating the efficacy of surgical treatment in infants with choanal atresia. *Saratov Medical Journal* 2024; 5(3): e0301. https://doi.org/10.15275/sarmj.2024.0301

Introduction

The absence of communication between the nasal cavity and nasopharynx characterizes congenital choanal atresia (CCA), which is a developmental anomaly in which the posterior sections of the nasal cavity are completely or partially blocked by a bone/membranous bone plate. One in 5-8 thousand newborns has a developmental anomaly, which mainly manifests itself as choanal atresia caused by a bony tissue (up to 89% of cases), but can sometimes occur in the form of mixed or membranous forms. Unilateral lesions occur twice as often. In girls, choanal atresia develops twice as often as in boys. CHARGE and other syndromes occur in over 45% of cases with bilateral choanal atresia and other variants of craniofacial anomalies [1]. It is important to keep in mind that most newborns cannot breathe through their mouth due to a number of anatomical and functional features. E.g., their laryngopharynx is located higher than in adults. When the infant swallows, the larynx rises and closes the passage between the soft palate and the nasopharynx. The newborn can breathe through the mouth only during crying [2]. Crying immediately eliminates the signs of respiratory failure, causing the cyanosis to disappear, but when the crying stops, the mouth closes again, and the entire cycle repeats [3]. Thus, the clinical manifestations of CCA can vary from difficult nasal breathing on the affected side to asphyxia of the newborn. In the latter case, the child needs resuscitation, the delay of which can lead to death. The main goal of surgical treatment of this pathology is the complete restoration of nasal breathing by eliminating the pathological anatomical obstacle between the nasal cavity and the nasopharynx. However, there is still no reliable surgical method that completely excludes the possibility of recurrent choanal atresia [4]. The main traditional method of CCA surgery is endoscopic endonasal choanotomy using stents in the form of tubes, in which a stent tube is installed in the wound channel after resection of excess mucosa, bone and membranous structures to prevent restenosis. This method is recommended by most authors who offer various types of protectors and different periods of their fixation in the operated choana [5], which in some cases allows achieving a slight improvement in nasal breathing. However, wearing a silicone stent (from 2 to 6 months) requires constant care (up to 6 times a day depending on the diameter of the tubes), significantly reducing the quality of life (QoL) in infants, as well as their parents or legal representatives. Besides that, long-term wearing of the stent causes inflammation of the surrounding mucosa, trauma to the mucosa of the nasal cavity and nasopharynx, increased formation of granulation tissue, formation of bedsores, and purulent deterioration of cartilaginous and bone structures, thereby requiring constant monitoring by an inpatient otolaryngologist with frequent revisions of the nasal cavity under anesthesia with a change of stents and removal of granulation tissue. Despite all efforts

of doctors and parents of the child, a removal of the stent tube triggers a high risk of restenosis [6].

The goal of our study was to evaluate the efficacy of a stentless endoscopic surgery for CCA in infants.

We proposed and tested a stentless method of choanoplasty: the flaps were fixed with fibrin glue (patent for invention No. 2789967 of 14 February 2023) [7] (*Figure 1*). A distinctive feature of this surgical technique is the formation of a single neochoana via the resection of the posterior portion of the vomer, and part of the perpendicular plate of the ethmoid bone and quadrangular cartilage. Then, upper and lower septal flaps are formed with endoscopic scissors and a sickle knife from the excess mucous membrane; they are fixed to the wound surfaces with fibrin glue. Hence, there is no need to implant various stents and tampons in the nasal cavity postoperatively because infants can breathe through their nose on their own in the first hours after the operation.

Materials and Methods

The study protocol was approved by the local ethics committee. Parents of each patient signed an agreement confirming their consent to participate. We performed over 100 surgeries for CCA in the Clinic of Otorhinolaryngology, Veltischev Research and Clinical Institute for Pediatrics and Pediatric Surgery, from 2019 through 2023. The study included 51 infants aged from birth to 1 year of age (mean age: 7.3 months), with difficulty breathing through the nose of varying severity. Of these, 30 girls (58.8%) and 21 boys (41.2%) were operated on. Bilateral choanoplasty was observed in 30 infants (58.8%), while unilateral choanoplasty was observed in 21 infants (41.2%), including right-sided choanoplasty in 13 infants (61.9%) and left-sided choanoplasty in 8 infants (38.1%). To compare the characteristics of the two surgical methods, the patients were distributed among two groups: the main group consisted of patients admitted to our clinic for the first time (n=27, 52.9%) while the comparison group encompassed patients admitted to the clinic after the traditional choanoplasty with stenting (n=24, 47.1%). All infants were operated on using a stentless surgery with posterior septal flaps fixed with fibrin glue. The efficacy of treatment was assessed primarily by the clinical need for reoperation in the follow-up period from 6 to 12 months.

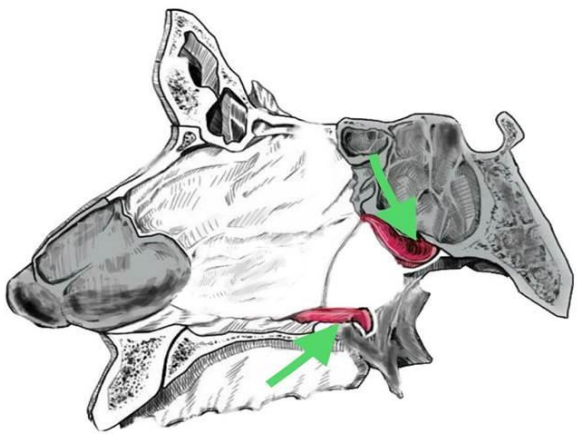

Figure 1. Sites of application of fibrin glue for fixation of upper and lower posterior septal flaps

Distribution of the examined infants by age and gender at the time of initial admission showed that CCA was more common in girls than in boys, albeit no statistical difference was found between the groups (p=0.1). Patients in the main group were slightly older at the time of admission, their mean age was 7.7±3.6 months, while in the comparison group it was 7.1±3.9 months (p=0.55). Clinical and demographic parameters of study subjects did not differ statistically significantly between groups.

To objectify the study results in infants, we employed pulse oximetry to measure the oxygenation of arterial blood in their lungs. The study was conducted in 50 infants aged from 1 month to 1 year, while 1 child at the age of 20 days was on mechanical ventilation. To assess the severity of respiratory disorders during sleep, mean saturation values were recorded every 15 minutes. The Mindray iMEC 8 patient monitor was used to monitor, display, review, store and transmit various physiological parameters. All infants underwent a repeat sleep breathing test 12 months after the operation.

To assess QoL preoperatively and 12 months after the surgery, parents were offered to complete a validated la Qualité de Vie du Nourrisson (QUALIN) questionnaire for infants aged 3 months to 1-3 years [8]. Parents of 45 infants participated in the survey: 25 (92.6%) in the main group and 20 (83.3%) in the comparison group.

We conducted a comprehensive analysis of physical, psychological and social functioning in children of this age. The Russian version of the QUALIN questionnaire includes only the parent form, which assessed four main aspects of the infant's functioning: behavior and communication (B&C: 13 questions), ability to stay alone (ASA: 5 questions), family environment (FE: 4 questions), neuropsychic development and physical health (NPD&PH: 11 questions). To determine the patient's QoL, we took into account the total score assessed on a five-point scale. The higher this indicator, the better the infant's QoL [9].

After a standard otolaryngological and endoscopic examination, most infants underwent an assessment of the preoperative computed tomography results. If concomitant pathology was detected, infants were consulted by a neonatologist, geneticist, ophthalmologist, neurologist, urologist and other specialists, and a questionnaire was administered.

Statistical processing of the obtained data was performed using the IBM SPSS version 26.0 software. Descriptive statistics of the study results were presented as counts and frequencies (expressed as percentages) for qualitative variables. For normally distributed quantitative characteristics, arithmetic means (M) and standard deviations (SD) were used. For non-normally distributed quantitative characteristics, we presented the median (Me) and interquartile range (Q25, Q75). The distribution pattern of quantitative variables in the observation groups was verified by the Shapiro-Wilk test.

When confirming the normal distribution of quantitative variables, the statistical significance of differences for multiple comparisons was tested using the analysis of variance. For paired comparisons, we employed the parametric Student's t-test for independent samples.

In the absence of a normal distribution of variables, the nonparametric Kruskal-Wallis test was used for multiple

comparisons of groups, while the Mann-Whitney U test was performed for paired comparisons. We used the nonparametric Wilcoxon test for dynamic comparisons of variables. Differences between parameters were considered statistically significant at p<0.05.

Results

The outcome of surgical treatment was assessed by analyzing the results of endoscopic examination, which allowed visualizing the choanae. All infants underwent endoscopic examination of the nasal cavity on postoperative days 7-10 prior to their discharge; in the follow-up, all children were also examined after 1, 6, and 12 months. The main group included infants admitted for the first time, n=27 (52.9%). Patients in the comparison group were admitted with a relapse after traditional choanoplasty using stenting, of which 8 (33.3%) infants had tube stents and 16 (66.7%) had none.

The largest number of patients were admitted with complete right-sided choanal atresia, while the smallest number of infants had partial right-sided choanal atresia (*Table 1*). All patients were hospitalized due to poor nasal breathing and with signs of respiratory failure.

The mean period of wearing the stent was 3.8 months. Of the patients in the comparison group, 15 (62.5%) infants underwent a single operation, 9 (37.5%) infants underwent multiple surgeries. The mean surgery duration was 73.8±32.4 min and did not differ statistically significantly between the two groups, amounting to 68.3±9.7 min in the main group and 80.4±34.4 min in the comparison group, respectively (p=0.193).

Mean saturation levels before the operation were patients, 98.4±2.4% in main group and 93.5±4.7% in the comparison group. Saturation was higher in primary patients than in infants admitted with a relapse after the traditional surgery (p<0.001).

Mean saturation in patients after endonasal endoscopic stentless surgery was as follows: 99.4±1.2% in the main group and 99.0±1.0% in the comparison group. In both groups, saturation after the stentless choanoplasty performed to correct the CCA was statistically significantly higher than before surgery: in patients of the main group, it increased by 1.0±1.2% (p=0.001); in those of the comparison group, by 5.5±3.7% (p<0.001).

Table 1. Frequency of occurrence of patients with different types of congenital choanal atresia and with different sides of its localization

Type of atresia		Group, count (%)			*p*
By side of its localization	By extent	Main, *n*=27	Comparison, *n*=24	Total, *n*=51	
Right-sided	Complete	20 (74.1)	22 (91.7)	42 (82.4)	0.100
	Partial	3 (11.1)	0 (0.0)	3 (5.9)	0.092
Left-sided	Complete	10 (37.0)	21 (87.5)	31 (60.8)	<0.001
	Partial	4 (14.8)	0 (0.0)	4 (7.8)	0.050

The preoperative QUALIN questionnaire scores (*Figure 2*) were higher in the main group than in the comparison group, albeit there was no statistically significant difference between the study groups (p>0.05), implying similar levels of QoL in patients of both groups. Comparison of scores of QoL indicators before and after surgical treatment demonstrated that they increased in the B&C questionnaire section in the main group to 3.6±0.3 pts and to 3.6±0.2 pts in the comparison group, which was statistically significant (p<0.001), thereby confirming better treatment results after surgery. The scores of QoL indicators before and after surgical treatment increased in the ASA questionnaire section to 3.7±0.5 pts in the main group and to 3.9±0.2 pts in the comparison group, which was also statistically significant (p<0.001), thereby implying better postoperative treatment outcomes (p<0.001). Comparison of the FE section scores revealed an increase to 3.7±0.4 pts in the main group and to 3.7±0.3 pts in the comparison group, which was not statistically significant in terms of a difference between groups (p=0.020), implying a slight improvement in the QoL of patients in this questionnaire section both before and after surgery. The QoL scores in the NPD&PH section increased to 4.3±0.6 pts in the main group points and to 4.4±0.32 pts in the comparison group, thereby revealing statistically significant differences indicating better postoperative patient treatment results (p<0.001).

The size of the choana when the newly formed neochoana was less than 50% of the norm was considered a relapse of the disease. To compare the outcomes of postoperative treatment between the traditional and stentless surgery methods, we identified a group of infants after the traditional surgery (n=24) and compared treatment results in it with those of infants after the stentless surgery in both main and comparison groups.

The greatest number of postoperative complications occurred after the traditional method of surgery with stenting in patients with CCA (*Table 2*). Comparison of the incidences of postoperative complications after the stentless surgery with those after the traditional surgery revealed statistically significant differences in almost all main parameters, confirming more favorable outcomes of surgical treatment in the main group. However, the data on the frequency of occurrence of infants with rare postoperative complications and synechiae, although more common after the traditional method of surgery, did not differ statistically significantly between the groups (p>0.05).

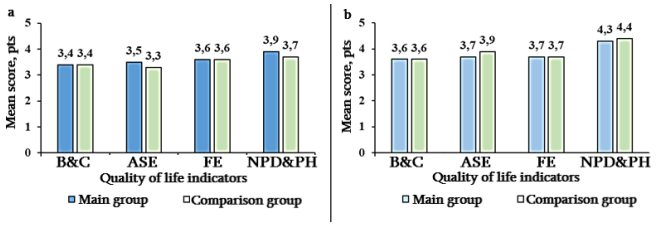

Figure 2. Questionnaire results (scores) by sections (scales) determining the quality of life in patients before (a) and after (b) the stentless surgery: B&C – behavior and communication; ASA – ability to stay alone; FE – family environment; NPD&PH – neuropsychic development and physical health

Results of postoperative treatment based on endoscopic examination of the nasal cavity and nasopharynx 12 months after surgery in patient groups

The following were considered rare complications in the traditional surgery group after long-term wearing of the stent tube (over 6 months): perforation of the nasal septum in the posterior sections, rupture of the columella after a purulent infection with long-term wearing of the stent fixed in the anterior sections of the nasal cavity. In 1 patient from the main group, the postoperative period was complicated by a secondary infection after the stentless surgery. The child was diagnosed with osteomyelitis of the left knee joint, a puncture was performed, and microbiological examination of the puncture revealed the growth of Candida albicans. The diagnosis of osteomyelitis of the left knee joint of fungal etiology was confirmed. The child was transferred to a specialized department with correction of therapy in the form of antifungal and immune drugs, and was discharged with positive dynamics. In 49 (98.04%) patients, nasal breathing was completely restored within 1 month after the operation. The wound surfaces were entirely epithelialized, and the formed flaps completely covered the bone structures of the posterior parts of the nasal cavity, and the size of the neochoana was more than 1 cm (*Figure 3*). Relapse of the disease in the form of stenosis was detected only in 2 patients (comparison group), which was most likely associated with an individual feature of the body, prone to excessive growth of scar tissue.

Table 2. Frequency of occurrence of postoperative complications

Postoperative complications	Group, count (%)			*p*
	Main, *n*=27	Comparison, *n*=24	Total, *n*=51	
Nasal synechiae	2 (7.4)	5 (20.8)	7 (13.7)	0.164
Relapse	0 (0.0)	16 (66.7)	16 (31.4)	<0.001
Granulations		8 (33.3)	8 (15.7)	0.001
Purulent complications		12 (50.0)	12 (23.5)	<0.001
Rare complications		2 (8.3)	2 (3.9)	0.126
Displacement/occlusion of stents		2 (9.1)	2 (3.9)	<0.001

Figure 3. The ultimate appearance of the neochoana 6 months after the stentless choanoplasty with fixation of flaps using fibrin glue

Discussion

With the development of endoscopic surgery in recent decades, the use of endoscopic endonasal choanoplasty in infants has become relevant. The traditional choanoplasty with stenting is widely used by pediatric otolaryngology surgeons immediately after the diagnosis of CCA [1, 10]. This method allows avoiding severe consequences of respiratory failure and tracheostomy in emergency situations. However, prolonged use of stent tubes (from 2 to 6 months and longer) substantially deteriorates the QoL in patients and increases the likelihood of complications such as relapses, granulation formation, purulent complications, bleeding, etc. [5, 11]. The results of our study unambiguously suggested that the proposed surgical treatment method based on the creation of a single postoperative neochoana with the formation of lower and upper flaps of the atretic mucous membrane and their fixation with fibrin glue allows minimizing and virtually eliminating the risk of restenosis and other postoperative complications associated with wearing a stent tube.

The presented data demonstrate the prospects for using this surgical method in the rehabilitation of patients with CCA in the postoperative period. This method significantly reduces the rehabilitation time (from 3-8 months to several days) and improves the infant's postoperative QoL vs. the traditional surgery. Parents can perform simple procedures for caring for their child at home in the postoperative period and do not need frequent revisions of the nasal cavity in a hospital setting.

Conclusion

The results of our study confirm that the method of surgical treatment of CCA without stenting is more effective than the traditional surgical technique with the use of stents. Our method allows performing surgical interventions in the earliest stages of birth, since there is no need to install stents or tamponade of the nasal cavity after surgery, which reduces the rehabilitation period of infants. In addition, according to our observations, the risk of restenosis is virtually absent, which is extremely important for newborns.

Author contributions: all authors contributed equally to the preparation of the manuscript.

Conflict of interest: none declared by the authors.

References

1. Koppen T, Bartmann D, Jakob M, et al. Diagnostics and therapy of bilateral choanal atresia in association with CHARGE syndrome. *J Neonatal Perinatal Med*. 2021; 14 (1): 67-74. https://www.doi.org/10.3233/NPM-200450
2. Zaitseva OV. Normalization of nasal breathing function in infants. New approaches. *Attending Physician* 2010; (3): 61. (In Russ.).
3. Tomoum MO, Askar MH, Mandour MF, et al. 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*. 2018; 132 (4): 329-35. https://www.doi.org/10.1017/S0022215117002614
4. Strychowsky JE, Kawai K, Moritz E, et al. To stent or not to stent? A meta-analysis of endonasal congenital bilateral choanal atresia repair. *Laryngoscope* 2016; 126 (1): 218-27. https://www.doi.org/10.1002/lary.25393

5. Wang PP, Tang LX, Zhang J, et al. Combination of the endoscopic septonasal flap technique and bioabsorbable steroid-eluting stents for repair of congenital choanal atresia in neonates and infants: A retrospective study. *J Otolaryngol Head Neck Surg*. 2021; 50 (1): 62. https://www.doi.org/10.1186/s40463-021-00547-5.
6. Ferlito S, Maniaci A, Dragonetti AG, et al. Endoscopic endonasal repair of congenital choanal atresia: Predictive factors of surgical stability and healing outcomes. *Int J Environ Res Public Health* 2022; 19 (15): 9084. https://www.doi.org/10.3390/ijerph19159084
7. Breeva OA, Asmanov AI, Bogomilsky MR. Patent #2789967 C1 of the Russian Federation, MPK A61B 17/00, A61B 17/24, A61F 2/18. Method of adhesive fixation of septal mucosal flaps in the choana in patients undergoing choanoplasty: #2021133852: submitted on 22 November 2021: published on 14 February 2023. EDN YKSJLJ. (In Russ.).
8. Baranov AA, Albitsky VYu, Vinyarskaya IV. Study of quality of life in medicine and pediatrics. *Issues of Modern Pediatrics* 2005; 4 (2): 7-12. (In Russ.).
9. Manificat S, Dazord A, Langue J, et al. Evaluation de la qualité de vie du nourrisson et du très jeune enfant: Validation d'un questionnaire. Etude multicentrique européenne Evaluation of the quality of life of infants and very young children: validation of a questionnaire. Multicenter European study. *Arch Pediatr*. 2000; 7 (6): 605-14. (In French). https://www.doi.org/10.1016/s0929-693x(00)80127-x
10. Bartel R, Levorato M, Adroher M, et al. Performance of endoscopic repair with endonasal flaps for congenital choanal atresia. A systematic review. *Acta Otorrinolaringol Esp (Engl. Ed.)*. 2021; 72 (1): 51-6. https://www.doi.org/10.1016/j.otorri.2020.01.002
11. Kotova EN, Vyazmenov EO, Bogomilsky MR. New ways to solve the problem of restenosis in surgical treatment of congenital choanal atresia in children. Bulletin of Otorhinolaryngology. 2020; 85 (1): 45-7. (In Russ.).

Authors:
Olga A. Breeva – Otorhinolaryngologist, Clinic of Otorhinolaryngology, Veltischev Research and Clinical Institute for Pediatrics and Pediatric Surgery, https://orcid.org/0009-0007-5147-5750;
Mihail M. Polunin – DSc, Associate Professor, Head of the Department of Otorhinolaryngology, School of Pediatrics, Russian National Research Medical University, https://orcid.org/0000-0002-5929-1469;
Alan I. Asmanov – Instructor, Department of Otorhinolaryngology, School of Pediatrics, Russian National Research Medical University; Head of the Department of Acute and Chronic Pathology of the Ear, Throat and Nose, Veltischev Research and Clinical Institute for Pediatrics and Pediatric Surgery, https://orcid.org/0000-0002-3116-6447.