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# Декануляция грудных детей с трахеостомой после хоанопластики

О.А. Бреева[1], М.М. Полунин[1, 2], А.И. Асманов[1]

[1] Научно-исследовательский клинический институт педиатрии и детской хирургии имени академика Ю.Е. Вельтищева, Российский национальный исследовательский медицинский университет им. Н. И. Пирогова, Москва, Россия;
[2] Морозовская детская городская клиническая больница Департамента здравоохранения города Москвы, Москва, Россия

## АННОТАЦИЯ

***Обоснование.*** В работе представлены этапы декануляции грудных детей с трахеостомой с врожденной атрезией хоан после применения бесстентового метода с использованием верхних септальных лоскутов и фиксацией их фибриновым клеем.

***Цель*** — оптимизация, подготовка, сроки и прогностические факторы успешной декануляции у грудных детей после хоанопластики.

***Материалы и методы.*** Объект исследования — 11 детей в возрасте до 1 года с трахеостомой и врожденной атрезией хоан. Основная группа: дети с трахеостомой после традиционной хоанопластики с помощью стентов ($n$ = 5); группа сравнения — дети с трахеостомой, поступившие первично для проведения операции ($n$ = 6). Для оценки результатов лечения выполняли эндоскопическое исследование, трахеобронхоскопию, определяли уровень сатурации при закрытой трахеостоме.

***Результаты.*** При эндоскопическом исследовании полости носа и носоглотки у пациентов первой группы декануляция невозможна из-за малого размера неохоан (мене 50 % нормы), они нуждались в повторной реоперации с применением бесстентового метода. У пациентов второй группы, прооперированных с использованием бесстентового метода хоанопластики, при катамнестическом наблюдении (3–6 мес.) признаков рестенозирования не обнаружено, им произведена успешная декануляция в разные периоды после операции. При катамнестическом наблюдении (1 год) признаков рестенозирования не отмечено у пациентов обеих групп. Удачная декануляция проведена 9 пациентам в разные сроки после операции. У 2 пациентов декануляция отложена на поздние сроки.

***Заключение.*** Длительное нахождение ребенка на искусственной вентиляции легких при двусторонней атрезии хоан недопустимо, и нередко принимают решение о наложении трахеостомы с целью разрешения дыхательной недостаточности. Однако трахеостомия может приводить к развитию большого количества ранних и поздних послеоперационных осложнений, поэтому декануляцию следует осуществлять в самые ранние сроки после восстановления носового дыхания. Предложенный нами эндоскопический доступ с помощью васкуляризованного слизистого лоскута и фибринового клея представляется методом выбора при хоанопластике у детей с трахеостомой. Процесс декануляции у детей даже с восстановленным носовым дыханием после реконструкции хоаны крайне сложен и несет много рисков.

**Ключевые слова:** хоанопластика; атрезия хоан; рубцовый стеноз; дети; трахеостомия.

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# Decannulation of tracheostomized infants after choanoplasty

Olga A. Breeva[1], Mihail M. Polunin[1, 2], Alan I. Asmanov[1]

[1] Veltischev Research and Clinical Institute for Pediatrics and Pediatric Surgery of the Pirogov Russian National Research Medical University, Moscow, Russia;
[2] Morozov Children's Municipal Clinical Hospital of the Moscow City Health Department, Moscow, Russia

## ABSTRACT

***BACKGROUND:*** The paper outlines stages of decannulation in tracheostomized infants with congenital choanal atresia following a stentless surgery using upper septal flaps fixed with fibrin glue.

***AIM:*** The aim of the study was to optimize preparation, timing, and prognostic factors for successful decannulation in infants after choanoplasty.

***MATERIALS AND METHODS:*** The study included 11 tracheostomized children under 1 year of age with congenital choanal atresia. The treatment group included tracheostomized children who undergone standard choanoplasty with stents ($n$ = 5). The comparison group included tracheostomized children admitted for primary surgery ($n$ = 6). Endoscopic examination and tracheobronchoscopy were performed to evaluate treatment outcomes, and saturation was measured with a closed tracheostomy.

***RESULTS:*** In group 1, endoscopic examination of the nasal cavity and nasopharynx revealed that neochoanae were too small (less than 50% of the norm) for decannulation; revision surgery was required using a stentless technique. In group 2, patients after the stentless choanoplasty showed no signs of re-stenosis during the follow-up period (3–6 months). They underwent successful decannulation at different time points after surgery. Over the one-year follow-up, no re-stenosis was observed in either group. In 9 patients, successful decannulation was performed at various time points after surgery. In 2 patients, decannulation was delayed.

***CONCLUSIONS:*** Long-term mechanical ventilation is unsuitable in patients with bilateral choanal atresia, and tracheostomy is a common solution for respiratory failure. However, tracheostomy can have many early and late postoperative complications, so decannulation should be performed as soon as possible after nasal breathing is restored. The proposed endoscopic approach using a vascularized mucosal flap and fibrin glue appears to be the method of choice for choanoplasty in tracheostomized children. The process of decannulation in children, even with restored nasal breathing after choanaplasty, is extremely challenging and is associated with many risks.

**Keywords:** choanoplasty; choanal atresia; cicatricial stenosis; children; tracheostomy.

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## BACKGROUND

Surgical treatment of congenital choanal atresia is one of the most common surgical procedures for congenital malformations of the face and skull in children. It is characterized by complete or partial closure of the posterior parts of the nasal cavity, resulting in the absence of communication between the nasal cavity and the nasopharynx. Congenital bilateral choanal atresia can cause respiratory distress syndrome and asphyxia immediately after birth. In such cases, urgent measures such as intubation, tracheostomy or choanoplasty are required. If treatment is delayed, healthy full-term infants can develop severe hypoxic damage to the central nervous system and even die. Respiratory distress syndrome is characterized by crying, tachypnea (more than 60 breaths per minute) [1], periods of apnea [2], cyclic cyanosis, stertorous breathing [3], and retraction of the muscles of the neck, chest, sternum, and abdomen due to inadequate lung mobility or high airway resistance.

Bilateral congenital choanal atresia in 34% of cases is associated with other upper respiratory tract disorders, leading to respiratory distress syndrome (subglottic stenosis, laryngomalacia, tracheomalacia, etc.). In addition, in 21% of cases, other congenital mandibular anomalies accompany bilateral choanal atresia [4]. Therefore, due to challenging management of congenital malformations in newborns, a tracheostomy may be required as a life-supporting procedure for long-term lung ventilation in order to relieve upper respiratory tract obstruction. Despite the life-supporting purpose of this procedure, it can cause many early and late postoperative complications. Early complications (3%–9%) in pediatric patients include subcutaneous emphysema, pneumothorax or pneumomediastinitis, hemorrhage, recurrent laryngeal and esophageal nerve injury [5], respiratory arrest during tracheostomy [6], accidental decannulation, tracheostomy obstruction with a mucus plug. Late complications include granulation tissue formation (30%–40%) [7, 8], tracheocutaneous fistula [9], tracheoesophageal fistula [10], tracheomalacia [11], subglottic stenosis [6, 12], dysphagia and high risk of respiratory tract infection [13]. To prevent these complications, decannulation should be performed as early as possible [14]. The literature addresses the issue of developing pediatric decannulation protocols, but actions proposed are largely based on the personal experience of healthcare providers and institutions rather than on research [15, 16]. In addition, the literature emphasizes the critical role of a multidisciplinary approach to developing these protocols.

After tracheostomy, with normalization of vital signs and stabilization of the condition, patients with bilateral choanal atresia need choanoplasty to restore communication between both parts of the nose and the nasopharynx. Currently, a conventional technique is used with perforation/resection of the bone and membranous structures of the atretic membrane with placement of a stent (protector or tube) [17]. This approach may improve breathing slightly, but does not prepare the patient for early decannulation because the risk of failure remains. Conventional choanoplasty with stents is associated with a high incidence of restenosis (up to 40%) in infants, which may also require re-positioning of the cannula in the trachea after stent removal from the newly formed choanus [18]. Therefore, prevalence of neonatal tracheostomy remains high despite improved clinical diagnostic criteria, management algorithms for neonatal respiratory distress syndrome due to nasal obstruction, and care of premature and medically challenging neonates. However, several issues remain unresolved, including preparation, timing, and indications for decannulation of patients after conventional choanoplasty with or without stents.

**The aim of the study** was to optimize preparation, timing, and predictors for successful decannulation in infants after choanoplasty.

## MATERIALS AND METHODS

In 2021–2023, our research team at the Otolaryngology Department of the Veltischev Research Clinical Institute of Pediatrics and Pediatric Surgery consulted more than 50 children under one year of age with congenital choanal atresia. The study included 11 children aged 19 days to 1 year who were admitted for tracheostomy for bilateral choanal atresia between 2021 and 2023 (mean age was 5.3 months). The sex distribution was as follows: 4 girls (36.4%) and 7 boys (63.6%). All children received community-based emergency tracheostomy in the first days of life. Among them, 5 patients (45.5%) were admitted primarily for surgical treatment of choanoplasty, 6 patients (54.5%) presented with non-functional choana after conventional choanoplasty with stents (Figure 1).

All patients with tracheostomy were admitted for surgical treatment with subsequent decannulation. For early rehabilitation of these patients, we developed and implemented an innovative technique of choana repair with posterior septal flap formation and fibrin glue fixation. Patients were divided into two groups:

- The first group included 5 patients (45.5%) with tracheostomy after primary choanotomy with ineffective nasal breathing,
- The second group included 6 patients (54,5%) with tracheostomy who were admitted for primary surgery.

Primary surgery was performed in the first month of life in 1 patient (20%), up to 3 months of age in 1 patient (20%),

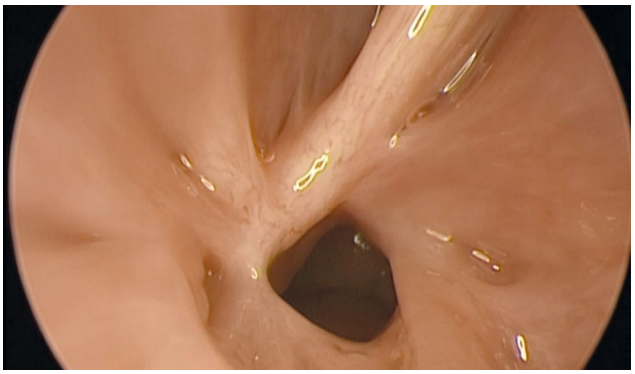

**Fig. 1.** Non-functional choana after previous tube-stent choanolasty
**Рис. 1.** Нефункциональная хоана после предыдущей хоанопластики с применением трубки-стента

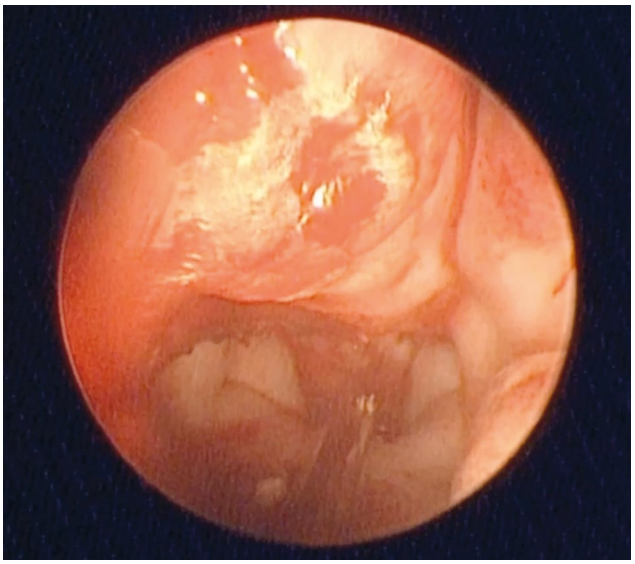

**Fig. 2.** Formed junctional neochoana using fibrin glue immediately after surger
**Рис. 2.** Сформированная функциональна неохоана с применением фибринового клея сразу после операции

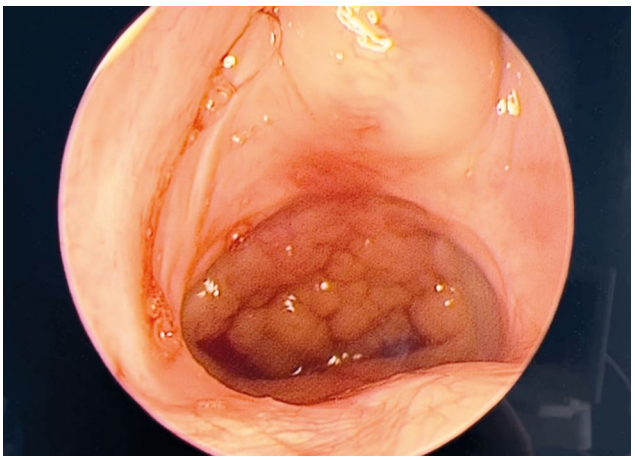

**Fig. 3.** Functional neohoana 1 month after surgery
**Рис. 3.** Функциональная неохоана через месяц после операции

up to 6 months of age in 2 patients (40%), at the age of more than 6 months in 1 patient (20.0%).

Before surgery, all patients underwent routine otolaryngologic and pediatric examination, endoscopic examination of the nasal cavity, computed tomography of the nasal cavity and paranasal sinuses, chest computed tomography as indicated, microlaryngoscopy, and bronchoscopy. All children received endoscopic endonasal choanoplasty with posterior septal flap formation without use of stents and packing and with fibrin glue fixation (patent No. 2789967; Figure 2) [19].

## RESULTS AND DISCUSSION

In the intensive care unit, the tracheostomy cannula was closed on the first postoperative day with a gradual increase in time (starting at 5–10 minutes) under saturation control. On day 3–4, the clinical picture slightly worsened, which was associated with reactive phenomena in the postoperative area. The tracheostomy port was opened or the tracheostomy cuff deflated when saturation levels decreased below 85%–90%. After resolution of reactive phenomena on days 7–10, patients were discharged and followed up by community-based pediatricians and otolaryngologists. Parents were instructed on the gradual closure of the cannula, and some severely ill patients were provided with a personal pulse oximeter. One month after the surgery, the children were admitted for tracheobronchoscopy to detect complications at the site of the tracheostomy (removal of granulations, detection of stenosis and tracheomalacia, etc.) and to replace the tracheostomy tube with a smaller one. Parents were instructed to close the tracheostomy during the day and then at night if the saturation parameters persisted.

Endoscopic examination of the nasal cavity at 1, 3 and 6 months showed complete epithelialization of the wound surfaces at the site of the neochoana. The formed flaps completely covered the bone structures of the posterior parts of the nose and all the wound surfaces of the neochoana, the size of which exceeded 75%. From months 3–6 after surgery, the children were readmitted at various time points for repeat tracheobronchoscopy and an attempt of decannulation. All children underwent overnight pulse oximetry with a closed tracheostomy prior to decannulation; mean saturation ($SpO_2$) was 97.2% (range 95%–98%). Granulations found around the tracheostomy in 4 patients (36.4%) during laryngobronchoscopy were removed and the tracheostomy tube was replaced with a smaller one. Tracheomalacia was observed in 2 patients (18.2%), and their tracheostomy tubes were replaced with smaller ones. Five patients (45.5%) had no visible respiratory tract abnormalities. In 9 patients (81.8%), successful decannulation was performed at different time points without the tracheostomy opening

being sutured. In 2 patients (18.2%), decannulation was delayed due to the risk of asphyxia caused by tracheomalacia during bronchoscopy (Figure 3).

## CONCLUSION

Success of decannulation depends on factors such as comorbidities, physical status, condition of the trachea, area around the tracheostomy tube, and respiratory tracts, as well as neurological status, and age of the patient [20].

Since it is impossible to keep a child with bilateral congenital choanal atresia on mechanical ventilation for an extended period of time, tracheostomy is sometimes performed to improve respiratory failure. Tracheostomy in children often causes serious complications such as pneumothorax, spontaneous decannulation, bleeding, which can lead to asphyxia and other complications. Late complications that may complicate cannula removal should also be considered. They include laryngeal chondroperichondritis, tracheomalacia, granulation tissue growth, and deformation of the tracheal wall at the level of the tracheostomy [21].

Even after restoration of nasal breathing with endoscopic choanoplasty, decannulation in children is a challenging and high-risk procedure. Since children are deprived of normal breathing and natural feeding from the first days of life (due to the lack of nasal breathing, the child cannot suck and is therefore tube-fed), low tone of the laryngopharyngeal and facial muscles can be observed, leading to neurogenic dysfunction of the central nervous system [15]. Based on our data, stent removal most often leads to restenosis, which ultimately prevents the patient from being ready for decannulation and requires repeat surgery. In premature infants, primary conventional choanoplasty with stents may be an alternative to tracheostomy as an emergency procedure in extremely rare cases. The endoscopic approach we propose, using a vascularized mucosal flap without use of stent and fibrin glue, seems to be the technique of choice for choanoplasty in young children with tracheostomy, because it has the lowest risk of recurrence and allows the patient to be prepared for decannulation as soon as possible after surgery (in the intensive care unit).

Length and characteristics of decannulation vary from patient to patient. Microlaryngoscopy and bronchoscopy are important tools to assess decannulation readiness [16]. Overnight pulse oximetry with a closed tracheostomy is an additional tool to evaluate condition of children with a tracheostomy. Our results show that certain pulse oximetry parameters such as oxygen saturation ($SpO_2$), pulse rate, predict success of decannulation in tracheostomized children. These patients require a personalized approach, multidisciplinary management, and a thorough evaluation of their condition prior to decannulation.

## ADDITIONAL INFORMATION

**Author contribution.** All the authors made a significant contribution to the development of the concept, research and preparation of the article, read and approved the final version before publication.

Contribution of each author: *O.A. Breeva* — data analysis, writing the main part of the text, collecting and preparation of samples, writing the main part of the text; *M.M. Polunin, A.I. Asmanov* — experimental design, collecting and preparation of samples, writing the main part of the text, making final edits.

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**Competing interests.** The authors declare that they have no competing interests.

**Ethics approval.** The protocol of the study was approved by the local Ethics Committee of the Children Surgery of N.I. Pirogov Russian National Research Medical University (No. 237 dated 19.02.2024).

**Consent for publication.** Written consent was obtained from the patients for publication of relevant medical information and all accompanying images within the manuscript.

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## AUTHORS INFO

*** Olga A. Breeva**, MD;
address: 2 Taldomskaya St., Moscow, 125412, Russia;
ORCID: 0009-0007-5147-5750;
eLibrary SPIN: 4120-7902;
e-mail: breevaluka77@mail.ru

**Mihail M. Polunin**, MD, Dr. Sci. (Medicine);
ORCID: 0000-0002-5929-1469;
eLibrary SPIN: 1199-4723;
e-mail: mmpolunin@gmail.com

**Alan I. Asmanov**, MD, Cand. Sci. (Medicine);
ORCID: 0000-0002-3116-6447;
eLibrary SPIN: 1650-4608;
e-mail: alan-asmanov@yandex.ru

## ОБ АВТОРАХ

*** Ольга Александровна Бреева**;
адрес: Россия, 125412, Москва, ул. Талдомская, д. 2;
ORCID: 0009-0007-5147-5750;
eLibrary SPIN: 4120-7902;
e-mail: breevaluka77@mail.ru

**Михаил Михайлович Полунин**, д-р мед. наук;
ORCID: 0000-0002-5929-1469;
eLibrary SPIN: 1199-4723;
e-mail: mmpolunin@gmail.com

**Алан Исмаилович Асманов**, канд. мед. наук;
ORCID: 0000-0002-3116-6447;
eLibrary SPIN: 1650-4608;
e-mail: alan-asmanov@yandex.ru

* Corresponding author / Автор, ответственный за переписку