# Special Article

# Choanal Atresia

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*Choanal atresia is caused by failure of resorption of the bucco-pharyngeal membrane during embryonic development. The atresia can be membranous or bony in nature, but is usually mixed in most cases. When the atresia is bilateral, newborns can have severe airway distress and cyanosis is alleviated by crying. Bilateral choanal atresia is managed with an oropharyngeal airway. Flexible nasal endoscopy and computed tomography can confirm the diagnosis. Surgery is the definitive treatment with two main approaches, namely transnasal or transpalatal. The transnasal route is currently the preferred procedure and can be performed in a minimally invasive fashion with endoscopic instrumentation. It is a safe and rapid procedure even in very young children, with no complications and a high rate of success. The use of a navigation system for surgical planning and intraoperative guidance and powered instrumentation can improve treatment outcome. The transpalatal approach is more invasive and reserved for failed endoscopic cases.*

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Choanal atresia is the developmental failure of the nasal cavity to communicate with the nasopharynx. Choanal atresia is a relatively rare congenital anomaly and occurs in approximately 1 in 5000 to 8000 live births, with a female to male ratio of 2:1[1]. Choanal atresia can be associated with other congenital anomalies up to 50%, whereas the rest have isolated anomalies. The most common associated congenital anomaly is CHARGE association (C = coloboma, H = heart disease, A = atresia of choanae, R = retarded growth and development, G = genital hypoplasia, E = ear deformities or deafness)[2]. Other anomalies associated with choanal atresia include polydactyly, nasal-auricular and palatal deformities, Crouzon's syndrome, Down syndrome, Treacher-Collins syndrome, DiGeorge syndrome, craniosynostosis, microencephaly, meningocele, meningoencephalocele, facial asymmetry, hypoplasia of the orbit and midface, cleft palate, and hypertelorism[3]. Samadi et al[4] retrospectively reviewed medical comorbid conditions of 78 children with choanal atresia. They found that common medical problems were otitis media with effusion (32%), upper and lower airway diseases (32% and 23%, respectively), cardiac anomalies (19%), and gastrointestinal tract disorders (18%). Statistically significant correlations were found for bilateral choanal atresia and cardiac disorders, CHARGE syndrome, obstructive sleep apnea, hematological problems, and prematurity or failure to thrive.

Generally, 65% to 75% of patients with choanal atresia are unilateral, whereas the rest are bilateral[5]. About 30% are pure bony, whereas 70% are mixed bony-membranous[6]. The atretic plate is usually sited in front of the posterior end of the nasal septum. The anatomic deformities include a narrow nasal cavity, lateral bony obstruction by the lateral pterygoid plate, medial obstruction caused by thickening of the vomer, and membranous obstruction[7]. Acquired posterior choanal atresia rarely occurs. It is usually caused by rhinopharyngeal injury *e.g.* after adenoidectomy; radiotherapy for nasopharyngeal carcinoma[8]; tuberculosis or syphilis of epipharynx[9], or sometimes by unknown causes.

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## Embryogenesis

There have been several theories regarding the embryogenesis of choanal atresia, but it is generally thought to be secondary to persistence of

either the nasobuccal membrane of Hochstetter or the buccopharyngeal membrane from the foregut. This membrane normally ruptures between the fifth and sixth weeks of gestation to produce choanae. Failure of this membrane to rupture causes atresia of choanae[10]. Other theories are abnormal persistence of mesoderm causing adhesions in the region of the nasal-choana or misdirection of mesodermal cell migration secondary to local factors[11]. In addition, a defect in the region of the nasal and palatal processes surrounding the nasobuccal membrane probably plays a role, leading to the associated findings of an accentuated arch of the hard palate, a medial location of the lateral and posterior nasal walls, and thickening of the vomer.

**Clinical manifestation**

Since a newborn child is an obligate nasal breather, respiratory distress occurs with patients with bilateral choanal atresia at or shortly after birth. They present with cyclic cyanosis relieved by crying. Airway obstruction during feeding but relieved by crying demonstrates that the oral airway is intact while the nasal airway is obstructed. The improved respiratory distress after crying may delay the diagnosis. Respiratory collapse may occur, and feeding difficulties may lead to failure to thrive. Most patients with bilateral choanal atresia are detected within the first month of life. However, it can be diagnosed in adults with long-term bilateral nasal obstruction and rhinorrhea[12].

Patients with unilateral choanal atresia rarely present with immediate or severe airway obstruction. They normally present within the first 18 months of life with feeding difficulties and nasal discharge, but may present with unilateral nasal obstruction and discharge in later life (Fig. 1). Obstruction of the contralateral side from infection or adenoid enlargement may precipitate airway symptoms and prompt medical evaluation. On anterior rhinoscopy, the occluded nasal cavity is typically filled with thick, tenacious secretions.

**Diagnosis**

There are numerous ways to diagnose choanal atresia. The simplest method is to pass a soft, red-rubber catheter (no. 8 French catheter) or 2.6-mm feeding tube through the nose into the nasopharynx. If the catheter is visualized in the oral cavity or oropharynx, a nasal airway is present. Inability to pass that catheter to the pharynx raises the possibility of choanal atresia; however, the diagnosis can be missed because of misdirection or coiling of the flexible catheter. The effort to pass a catheter through a patent but narrow airway may cause edema and/or bleeding, resulting in complete obstruction. In addition, observing misting on a metal spatula or a laryngeal mirror or detecting movement of a cotton wisp placed beneath the nose when the mouth is closed or during feeding and auscultation of the nares may be helpful. This documentation of the airflow rules out an atresia, but does not preclude a stenosis. A drop of methylene blue can be placed into the nasal cavity on the suspected side. If the blue dye appears in the oral cavity or oropharynx, atresia is excluded.

Patients may also be examined with a rigid or flexible endoscope, operating microscope, mirror examinations, or digital examination. The use of flexible fiberoptic endoscope is the preferable method of choice because nasal patency can be assessed, and the anatomy (nasal vestibule, nasal septum and lateral nasal wall) can be evaluated. Additionally, other causes of airway obstruction such as congenital nasal septal deviation, piriform aperture stenosis, nasal cavity stenosis, nasolacrimal duct cysts, or other space occupying lesion can be ruled out. A traditional method of diagnosis is radiography using radiopaque contrast material instilled into the nasal cavity with the patient in a supine position. Acoustic rhinometry can be used for the diagnosis, but is especially helpful in the post-operative period to follow patients for restenosis[13].

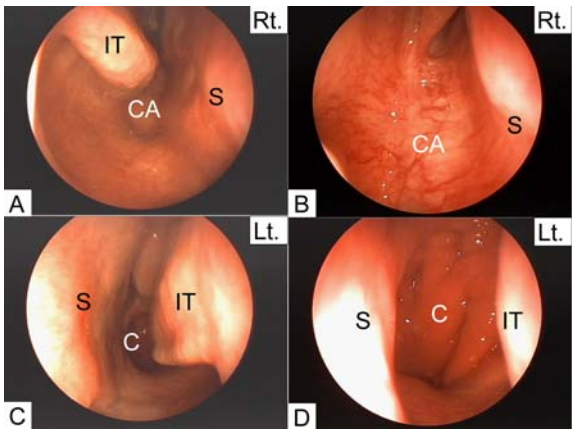


**Fig. 1** A 31-year-old male with a unilateral nasal obstruction and persistent mucoid discharge since childhood. Nasal endoscopy (0 degree telescope) revealed right choanal atresia on the nasal side (A, B) and normal left nasal cavity (C) and choana (D). (IT-inferior turbinate, S-septum, C-choana, CA-choanal atresia)

Computed tomography (CT), especially axial plane, is the radiographic procedure of choice since it can demonstrate the nature (bony or membranous), position, and thickness of the obstructing segment, which helps the surgeon in designing a plan for repair, and other abnormalities, such as dermoids, encephalocele, gliomas, anterior skull base defect[10] can be excluded. CT usually shows a widening of the posterior septum and a dense bony thickening of the lateral buttress. Adequate using vasoconstrictor drops and nasal toilet, sedation or general anesthesia may be essential in infants to obtain good-quality images. With CT scans, normative data are now available for neonates, up to 6 months of age, regarding the size of the nasal cavity[14].

**Management**

Management of these patients varies and depends on age, type of atresia, and general condition of patients. Because infants are obligate nasal breathers, bilateral choanal atresia is a life-threatening situation since, if not promptly recognized, it can lead to severe asphyxia and death immediately after birth. Nasal airway obstruction from bilateral choanal atresia is usually demonstrated immediately after delivery. Suckling or a mouth-closed posture will precipitate a crisis. Immediate management of bilateral choanal atresia involves training the infant to breathe through the mouth with the aid of an indwelling oral appliance such as a McGovern nipple or an oropharyngeal airway. A McGovern nipple is an ordinary nipple with a single enlarged hole and can be used for gavage feeding. It is secured with ties or tape around the ears. The infant can be fed, and the airway is protected while definitive management is delayed to allow time for a complete workup to rule out other anomalies. Endotracheal intubation is usually unnecessary unless the infant requires mechanical ventilation. If there is severe respiratory distress and airway cannot be established by endotracheal intubation, an emergency tracheotomy should be performed until further evaluation and treatment can be established. Nevertheless, surgical correction is usually necessary early in life.

Unilateral atresia is rarely emergent. The repair is generally delayed for at least 1 year, which allows the operative site to enlarge and reduces the risk of postoperative stenosis, unless there are feeding difficulties. Repair of atresia can be performed anytime after the airway has been secured, and other anomalies have been evaluated. Surgical repair and anesthesia in very young infants is technically difficult; however, they usually tolerate stenting better than older infants. Transnasal and transpalatal approaches are common surgical techniques used. Transnasal approach with the use of a rod-lens telescope is the method of choice and has been used successfully in newborns and infants and is suitable for membranous or very thin bony atresia, while transpalatal approach is normally reserved for the older children, thick bone, or case with restenosis. Better instrumentation in endoscopic sinus surgery and advances in CT scans have made the transnasal repair the most popular method. Nasal endoscopy is beneficial in the management of choanal atresia since it helps to confirm the diagnosis, characterize the extent of lateral nasal wall contribution to the stenosis or atresia, evaluate the composition of the atresia (bony and/or membranous), guide surgery, and provide postoperative surveillance.

The transpalatal procedure has a higher success rate (ability to create larger initial opening), enables better visualization (wider surgical exposure and improved access to the posterior vomer) and preservation of mucosal flaps along the newly formed apertures (decreased incidence of postoperative scarring and restenosis), and permits short-term stenting. In addition, there is less chance of disorientation and significant surgical complications intracranially[11]. However, it may lead to a growth disturbance to the hard palate, alveolar arch, and midface, occlusive abnormalities *e.g.* crossbite, palatal fistula, increased operative time and blood loss, risk of damage to the greater palatine neurovascular bundle, and injury of the soft palate resulting in a future problem with rhinophonia.

The main advantage of the transnasal procedure is minimally invasive, quick (avoiding the need for prolonged anesthetic agents), less traumatic with minimal blood loss, and provides excellent visualization and the ability to perform exact surgery on patients of all ages[15]. Its disadvantage is limited field of vision (which risks injury to sphenopalatine artery or skull base), even with a microscope, especially in newborn infants or a case with deviated nasal septum, large turbinates, or small size of the nasal cavity, and the inability to adequately remove enough vomerine septal bone to prevent restenosis. Practically, creation and maintenance of the flaps can be very difficult. Although any surgery on the nose or septum carries a risk of inhibition of facial growth, it has not been reported with transnasal approach. Richardson and Osguthorpe[16] compared transnasal with transpalatal

repair in 37 with congenital atresia or severe stenosis of both posterior nasal choanae. They found that transnasal repair allowed correction with minimal blood loss and without facial growth or occlusal abnormalities.

**Transnasal procedure**

All surgical procedures are performed under general anesthesia, and the airway is prior controlled by endotracheal intubation or rarely tracheostomy. For transnasal approach, the most precise techniques can be done under an operating microscope or telescope[15]. Soft palate retraction enables the visualization of posterior choanae. Gauze packs should be placed in the nasopharynx to protect the nasal surface of the soft palate, confirm the anatomy when the atresia plate is penetrated, and prevent blood from being swallowed or aspirated. The use of vasoconstrictor drops and nasal suction provide a good view of anterior nares. The mucosa over the anterior face of the atretic area, posterior portion of the septum, and lateral nasal wall are injected with a solution of lidocaine with diluted epinephrine under direct visualization, and cooperation with pediatric anesthesiologist is required. The nose is then packed with cottonoid pledgets soaked in a solution of 0.025% oxymetazoline. These packs are left in place for 10 minutes. Excellent vasoconstriction is achieved using this method. The anterior mucosa overlying the atretic plate is incised vertically, and the mucosal flap is elevated medially and laterally by the use of a flap or sickle knife (Fig. 2C). If the atresia is only membranous, the incision may open the choanae. If the bone is present, the atretic plate is perforated using a curved urethral sound starting at the junction of the atretic plate, hard palate, and vomer (inferomedial part of atretic bone), since this junction is usually the thinnest portion of the atresia. It is important to remember that the atretic plate in the newborn is close to the basisphenoid, which separates the nasal cavity from intracranial contents. Care should be taken not to damage the postnasal space since it may result in basisphenoid fracture. Therefore, the nasopharynx should be approached inferiorly. The membrane or bone from the atretic plate, lateral nasal wall (medial aspect of pterygoid plate), and lateral portion of the posterior septum is then removed by curette or burr, exposing the nasopharyngeal region and the previously placed gauze packs, until a size of 18 French-gauge dilator can be introduced with ease. It is essential to adequately resect the abnormal posterior vomer to create a common cavity posteriorly, which provides a larger nasal airway, a lower stenosis rate, and a favorable outcome of the procedure. This can be accomplished using the drill and tissue shaver or a back-biting forceps. After bone removal, the mucosal flaps should be folded posteriorly (medial and lateral flaps) before a stent is placed.

Nevertheless, it is difficult to preserve mucosal flap and completely remove the thick bony plate, which may lead to restenosis. Stamm and Pignatari[17] performed transnasal micro-endoscopic surgical approach in 33 patients with choanal atresia utilizing "nasal septal crossover flap technique" to diminish the incidence of restenosis. Their success rate for unilateral cases was 86%, while for bilateral cases was lower, 72%. Newer endoscopic techniques with powered instrumentation[18,19] or laser[20] and the computer-assisted navigation system[21,22] can enhance the safety and efficacy in the repair of choanal atresia especially in refractory cases.

Recent reports using transnasal endoscopic repair have shown excellent results. Khafagy[18] performed transnasal endoscopic repair in nine infants with bilateral choanal atresia and stented neochoana for 5 to 8 weeks. Five cases remained patent after removal of stenting. Two patients required revision surgery. Pasquini et al[23] also reported the

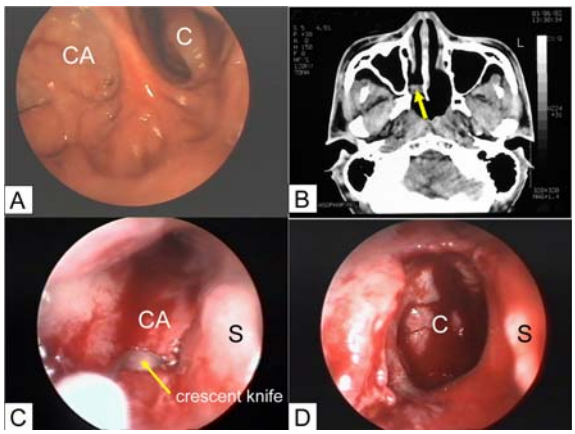


**Fig. 2** The same patient from Fig. 1. The right choana was completely closed as seen by 70 degree telescope on the nasopharyngeal side (A). CT scan showed membranous atresia on the right side (yellow arrow) (B). Transnasal repair was performed using crescent knife to make a curvilinear incision at the inferior border of choana (C). Atretic area was removed until the chaoana was large enough (D). (S-septum, C-choana, CA-choanal atresia)

effectiveness of the transnasal endoscopic approach for the treatment of 14 patients with congenital choanal atresia (three cases were bilateral, while 11 were unilateral). The overall follow-up period ranged from 2 to 64 months (mean 31 months). Only one restenosis was observed.

Possible complications of the transnasal approach include pressure necrosis of anterior nares or columella, plugging and displacement of the indwelling stents, cerebrospinal fluid leaks, meningitis, and granulation tissues around the stents.

**Transpalatal procedure**

For transpalatal approach, the palate is exposed with a mouth gag and injected with lidocaine with epinephrine for hemostasis, which requires cooperation with a pediatric anesthesiologist. Then, a curved or vertical incision is made in the soft and hard palatal mucosa to expose hard palatal bone and the soft palatine muscle. The curved incision starts behind the maxillary tuberosity on one side and is carried medially to the alveolar ridge to the canine region, and the same incision is performed on the other side to create U-shaped palatal flap. The mucoperiosteal flaps are elevated posteriorly to the edge of the hard palate, and care should be taken to preserve the neurovascular bundle from the greater palatine foramen. The soft palate is retracted posteriorly and superiorly to expose the posterior edge of the hard palate and the nasopharynx. Then, the posterior hard palate is removed using a cutting bur or a Kerrison punch to expose the nasal cavity basal mucosa, and the mucosa is reflected as a flap and lifted until it reaches the choana. The posterior part of the septum and lateral superior nasal wall should be removed. A stent is placed, and the nasal mucosal flaps are placed on the inferior aspect of each tube before the palatal flap is replaced and sutured.

Possible complications of the transpalatal approach include pressure necrosis of anterior nares or columella, plugging and displacement of the indwelling stents, palatal flap dehiscence, development of maxillary hypoplasia resulting in midface retrusion and malocclusion, and granulation tissues around the stents.

**Stents**

Stenting is needed to maintain the patency of the new lumen and hold the mucosal flaps in place. A No. 3.5 or 4 endotracheal tube cut to the appropriate length and shape is commonly used as a stent since it is available in the operating room, easily shaped and trimmed. A 3 to 4 mm section of the middle part of the tube is removed to allow non-traumatic positioning across the columella. The opening should be large enough to allow easy passage of suction catheters through the endotracheal tube anteriorly into the nasopharynx. The posterior end of the stent should pass through the posterior choana, but it should not touch the nasopharyngeal wall. Its length should be adequate to support the newly formed choana, but short enough to avoid discomfort or regurgitation of food through the nose. The stent is fixed in place using 2-0 silk transseptal sutures. It should be hidden about 3 to 4 mm behind the alar rim, and should not protrude from the nose to prevent being grasped by the infant or child, and should not be visible externally to prevent social and psychologic trauma in an older child.

Uncommonly, a single stent may be used in case of unilateral atresia. After the operation, the child can breathe through the stent, and meticulous care is needed to prevent plugging of secretion, infection, stent displacement, and columella ulceration. Regular suction should be carried out after instillation of 0.9% normal saline drops, and parents are trained to use suction apparatus and saline irrigation. Care should be taken to the area of fixation of the stents to prevent erosion and permanent scarring. The broad-spectrum oral antibiotics should be prescribed for the whole period of stenting to reduce the risk of purulent rhinorrhea. Complications of stents include granulation tissue formation, nasoseptal perforations, and erosion of the naris of the nose or columella.

A stent should be kept for 2 to 3 months until the choanal operative site is mucosalized since the incidence of restenosis is high. Since maintenance of stents is difficult, educating parents and close follow-up are very essential. Occasionally, nasal dilatation and revision surgery may be indicated after stent removal. They may require multiple procedures to achieve the desirable patency. Samadi et al[4] retrospectively reviewed surgical outcomes of 78 children treated for choanal atresia transnasally with an average follow-up of 35 months. Main outcome measures were type and number of procedures and airway patency. Surgery was effective in establishing a patent airway in all cases. Unilateral choanal atresia required, on average, 2.7 total procedures, including dilation and removal of stents, compared with bilateral choanal atresia, which required 4.9 procedures. They concluded that meticulous postoperative care particularly stent management and routine postoperative revision

endoscopy were crucial for successful treatment of choanal atresia.

Although it has been suggested that a stent should be kept for a long period of time, in some cases, success has been seen with minimal duration of stenting or without stents. Van Den Abbeele et al[24] report the favorable outcome of transnasal endoscopic repair of choanal atresia using powered instrumentation and routine postoperative revision endoscopy in 40 children (unilateral: n = 26, bilateral: n = 14). Nasal tubes in neonates or infants and nasal packing in older children were removed after 2 days. Postoperatively, 32 patients (80%) had normal nasal patency and a satisfactory choanal diameter without prolonged nasal stenting after surgery. In agreement with this study, Rombaux et al[19] also performed the endoscopic endonasal approach using the microdebrider to correct seven children with unilateral choanal atresia. At the end of the procedure, topical application of mitomycin was performed without postoperative nasal stenting. Of the seven patient procedures, six (85.7%) remained patent (follow-up range 12 to 36 months). Holzmann and Ruckstuhl[25] also supported the technique without stenting due to requiring less invasive post-operative care. They found favorable results of the transnasal surgical repair in eight patients with unilateral choanal atresia by which no stents were used. The success of choanal patency was followed by nasal endoscopy with a mean post-operative follow-up time of 1.9 years. In all patients, both choanae remained patent. No further treatment was required. Moreover, Tzifa and Skinner[20] described the endoscopic repair of three patients with unilateral choanal atresia with the KTP laser with no requirement for stenting. Follow-up was between 12 months and four years with all choanae remaining patent, and no dilatation was needed. These data suggest that post-operative stents can be placed for a shorter duration and may not be required in some selected cases.

Avoidance of stents has a great deal of appeal, but may be associated with a higher rate of restenosis. Rahbar et al[26] reported a good result using mitomycin for prevention and treatment of scar formation in five patients with choanal atresia. Prasad et al[27] also found a favorable result (17 patients: 85%) of the use of topical mitomycin as an adjunct to the surgical repair of 20 patients with choanal atresia in improving patency with a decreased need for stenting, dilatations, and revision surgery. Holland and McGuirt[28] evaluated the intraoperative use of mitomycin in eight patients with bony choanal atresia undergoing choanal atresia repair, which were compared with 15 historical controls. All patients in both groups were treated with post-operative stenting. The success rate of the repair of the choanal atresia as determined by the post-operative need for dilation or revision surgical procedures was compared. The post-operative dilations for soft tissue restenosis in children with intra-operative use of mitomycin were significantly less than those in the controls. These results show that mitomycin is an effective and reliable treatment for improving the surgical outcome for choanal atresia repair by reducing post-operative scarring, obviating the need for postoperative dilations and potentially eliminating the need for surgical stenting.

After surgery, patients should be closely followed-up for a long time to check restenosis. If re-stenosis occurs, transnasal revision surgery or dilations can be safely and easily repeated. This will help to break up the forming scar, and eventually leads to better patency. Increasingly larger diameter male urethral sounds are lubricated and gently passed through the nose into the nasopharynx. The stenotic part is enlarged using guarded drills[29,30]. Resection of granulation tissue can be done with the use of $CO_2$ laser. With the use of best possible techniques of surgical correction, medication, and stenting, the authors may cut down on postoperative stenosis formation and the need for dilatation and revision surgery in the future.

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# *รูจมูกด้านหลังตัน*

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*รูจมูกด้านหลังตัน เกิดจากการที่ยังมีเยื่อบุ bucco-pharyngeal อยู่ ไม่สลายหายไปในช่วงของการเจริญเติบโต ซึ่งส่วนที่ยังเหลืออยู่นั้นอาจเป็นเพียงเยื่อบาง ๆ หรือ เป็นกระดูกก็ได้ ซึ่งส่วนใหญ่มักจะเป็นแบบผสม คือ มีทั้งเยื่อบางและส่วนของกระดูกร่วมกัน ผู้ป่วยเด็กแรกเกิดที่มีรูจมูกด้านหลังตันทั้งสองข้าง จะมีปัญหาการอุดกั้น ทางเดินหายใจ อาจทำให้เขียวได้ แต่ในขณะผู้ป่วยร้อง และมีการหายใจทางปาก อาการเขียวจะดีขึ้น การรักษาเบื้องต้นคือให้ใส่ oropharyngeal airway การวินิจฉัยสามารถทำได้โดยใช้กล้องส่องตรวจในโพรงจมูก และเอกซเรย์คอมพิวเตอร์ การรักษาทำได้โดยการผ่าตัดซึ่งมี 2 วิธีคือ ผ่าตัดทางจมูกและทางเพดานปาก วิธีผ่าตัดทางจมูกเป็นวิธีที่นิยมทำโดยใช้เครื่องมือขนาดเล็กและกล้องส่อง เนื่องจากเป็นวิธีที่ปลอดภัย มีอันตรายต่อเนื้อเยื่อ และอวัยวะรอบข้างน้อย และสามารถทำได้เร็วแม้ในผู้ป่วยเด็กที่มีอายุน้อย มีโอกาส ผ่าตัดสำเร็จสูง และมีภาวะแทรกซ้อนต่ำ ปัจจุบันมีการใช้เครื่องนำวิถี โดยใช้คอมพิวเตอร์ช่วย ซึ่งจะทำให้ ผ่าตัดได้ถูกต้องและแม่นยำมากขึ้น และมีเครื่องมือทุ่นแรงที่จะทำให้ผ่าตัดได้ง่ายขึ้น และได้ผลดียิ่งขึ้น ส่วนการผ่าตัดทางเพดานปากนั้น มักจะมีการบอบช้ำของเนื้อเยื่อรอบข้างมากกว่า และมักจะทำในรายที่ล้มเหลวจากการผ่าตัดทางจมูกเท่านั้น*