Case report

Transnasal endoscopic neochoanal technique: An effective procedure for bilateral choanal atresia in adult female

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ARTICLE INFO

Keywords:
Choanal atresia in adult
Human and disease
Stenosis
Transnasal endoscopic neochoanal technique

ABSTRACT

Background: Surgery on bilateral choanal atresia surgery is challenging and this case is rare in adults. In addition, stenosis issue postoperatively is commonly seen.

Case presentation: Transnasal endoscopic neochoanal technique was performed on a 27 years old female with the detailed surgical procedure was described. Neither stent nor nasal tamponade was applied. Ten months follow-up revealed a patent-wide neochoane.

Discussion: Transnasal endoscopic neochoanal technique without stent and nasal tamponade can be considered the procedure of choice in the management of CA cases.

Conclusions: Transnasal endoscopic neochoanal technique is considered an effective surgical procedure on bilateral choanal atresia.

1. Introduction

Choanal atresia (CA) is a rare congenital anomaly that fails in communication between the nasal cavity and nasopharynx. It occurs in 1:8000 of live births, more prevalent in women than men, 2:1 respectively. Choanal atresia is classified based on unilateral (41%) and bilateral (59%), bony type (90%), and membranous type (10%) with various degrees of stenosis. Bony type of CA is due to thickened vomer and medialization of the pterygoid plate of the sphenoid bone. Around 41–72% of CA cases are a part of CHARGE syndrome consisting of coloboma, heart defects, choanal atresia, retarded growth and development, genital hypoplasia, ear anomalies, and deafness [1].

The majority of CA cases are found in infants [2], where as adults case is very rare [3,4]. New CA cases found in adults are related to socioeconomic factors in the family which are often found in low resource settings [3-5]. The transnasal endoscopic neochoanal technique is an effective technique for the management of CA cases [6,7]. Based on the Surgical Case REport (SCARE) 2020 Guidelines [8], we are interested in reporting cases of CA in adults who underwent a modified transnasal endoscopic neochoanal technique.

2. Case presentation

A 27-year-old female complained of both nasal blockages since birth. Smell impairment, snoring, and mouth breathing accompanied the main symptom. There was no history of breathing support whilst delivered but encountered exhaustion especially on breastfeeding episodes relieved with crying. A saddle nose was noticed on examination. A preoperative nasoendoscopy revealed a different level of nasal floors (higher level of the right nasal floor), blocked choanes with spacious, normal inferior turbinates, and no deviated nasal septal (Fig. 1). Preoperative paranasal sinus CT scan findings supported a bilateral bony type of CA with some degree of stenosis (Fig. 2).

Surgery was performed under general anesthesia. The patient was positioned supine and reverse Trendelenburg 20 degrees. Had the disinfection done, the nasal cavity was prepared with gauze soaked in oxymetazoline 0.05% and lidocaine 1%. The procedure began with an incision on the anterior mucosal surface of the left atretic area up to the vomer. A mucosal flap was created following the incision line to cover the incision wound and the exposed bone in the end of procedure. The atretic bone removal was performed up to vomer (posterior septectomy) by Kerrison ronger and osteotome. The same procedure was performed on the contralateral side. A wide neochoaneae was created (Fig. 3). Surgicel® was applied to ensure hemostasis. No nasal tamponade nor...
A paranasal sinus CT scan revealed a bony type bilateral choanal atresia with some degree of stenosis (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Fig. 1. Right (A, B) and left (C) nasal cavities showed bilateral choanal atresia. IC (inferior concha), Sp (septum), Ap (atretic plate).

Fig. 2. A paranasal sinus CT scan revealed a bony type bilateral choanal atresia with some degree of stenosis (yellow arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

stent was applied. The patient was discharged on the following day. Post-operative nasal saline irrigation was delivered as frequently as twice daily for four weeks.

One month postoperative follow-up revealed a patent neochoanae with a good healing process (Fig. 4). The patient began to be able to identify some odorants. Ten months post-operative follow-up revealed no complaint of breathing problem, smell function was back to normal.

3. Discussion

Bilateral CA is rarely seen in adults, with only 10 cases reported in the literature [4,5,9,10]. Surgical management of CA consists of transseptal, transpalatal, and transnasal approaches [11]. The first successful surgery on a bony type bilateral CA was carried out by Carl Emmert in 1851 on 7 years old boy through a blind bilateral transnasal puncture with progressive dilatation. This blind technique potentially injured the
skull base. In 1908, Uffenorder introduced a transseptal approach through a submucous resection to reach the vomer and removed it along with atretic plate removal to create a large uni-neochoanae. This was the first step toward modern CA surgery. Large resection of the growing septum may result in significant developmental anomalies of the face and nose. This old transseptal approach was lack of good visibility since it was not performed endoscopically. The transpalatal approach was proposed in 1937, entailed large exposure of the choanal region, enabling drilling away of the medial pterygoid plates, enlarging the uni-neochoanae. This technique might lead to serious complications such as abundant blood loss which was critical in newborns. Other disadvantages were its effect on maxillary growth causing cross-bite and high palatal arch deformity, palatal fissure, and velopharyngeal insufficiency [12]. The endoscopic technique is currently used widely. Transnasal endoscopic procedure for CA provides excellent visualization and enables accurate surgery [6,7,11].

The role of the stent is controversial [4,7]. Stentless endoscopic repair reduces the need for frequent postoperative care and also avoids stent-related complications including patient discomfort, a potential nidus for a bacterial colonization that triggers persistent inflammation and fibrosis, and pressure. The recent literature has suggested the benefit of the stent are dubious and the endoscopic procedure for CA can be performed successfully without stenting. Nasal douching during the postoperative period is more helpful for preventing restenosis [13]. Novel application of steroid eluting sinus stent in minimizing scarring following endoscopic procedure for CA show a high level of success. Mitomycin C after choanaloplasty is used to prevent stenosis. Of the ten documented cases of adult bilateral CA in literature, only one patient had restenosis and thus requiring a revision surgery [4].

The current surgical approaches for congenital CA should fulfill the requirement of minimally invasive surgery. Five main surgical objectives should be achieved to minimize the tendency for restenosis such as the construction of an as large as possible uni-neochoanae by safely removing the posterior part of the vomer (posterior septectomy) and by drilling away the medial pterygoid plate. In the case of rhinopharyngeal stenosis, part of the endochondral clival bone should be resected, all raw surfaces should be covered by multiple mucosal flaps secured with fibrin, no stent required, and appropriate postoperative care [10].

In communities with low socioeconomic status living in rural areas and far from sufficient health facilities, ante and postnatal history are unidentified. Meanwhile, choanal atresia is a congenital abnormality that should be identified during newborns [14,15]. In addition, in low socioeconomic communities, nutritional intake during pregnancy tends to be poor, which is a risk factor for choanal atresia [16]. Lack of awareness on the signs and symptoms of choanal atresia that appears such as respiratory distress relieved with crying and recurrent upper respiratory infection also plays a role in the delay of diagnosis [17,18].
4. Conclusion

The transnasal endoscopic neochoanal technique is considered an effective procedure to manage bilateral choanal atresia. Low socioeconomic status causes delay in the management of choanal atresia.

Consent

We have requested the patient’s consent to publish this case report for educational purposes.

Guarantor

Budi Sutikno.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Funding

None.

Supplement

https://www.youtube.com/watch?v=6nbb1zzclnQ&t=23s

Ethical approval

We have conducted an ethical approval base on Declaration of Helsinki at Ethical Committee in Dr. Soetomo General Academic Hospital, Surabaya, Indonesia.

Credit authorship contribution statement

All authors contributed toward data analysis, drafting and revising the paper, gave final approval of the version to be published and agree to be accountable for all aspects of the work.

Declaration of competing interest

The authors declare that they have no conflict of interest.

Acknowledgement

We would like to thank Fis Citra Ariyanto for assisting in editing and proofing.

References

[1] B. Läktor, L.V. Cookonal, J. Gerlinger, A new endoscopic surgical method for unilateral choanal atresia, Laryngoscope 111 (2) (2001) 364–366, https://doi.org/10.1097/00005537-200102000-00033.
[2] S.H. Abdul Cader, F.A. Shah, N. Rghunandanand, Clinical retrospective analysis of 15 cases of choanal atresia - our experience, World J. Otorhinolaryngol. Head Neck Surg. 5 (4) (2019) 188–192, https://doi.org/10.1016/j.wjhn.2019.03.003.
[3] C.D. Durmaz, V. Tas, P. Kocay, O.S. Fitzor, H. Onay, S. Beton, et al., Bilateral choanal atresia in an adult woman with pycnodysostosis, Congenit. Anomalies 57 (3) (2017) 91–92, https://doi.org/10.1111/cps.12264.
[4] S. Anajar, J. Hasnaoui, S. Rouadi, R. Abada, M. Roubal, M. Mahtar, A rare case report of bilateral choanal atresia in an adult, Int. J. Surg. Case Rep. 37 (2017) 127–129, https://doi.org/10.1016/j.jsrcr.2017.05.002.
[5] R.K. Verma, P. Lothen, N.K. Panda, Congenital bilateral adult choanal atresia undiagnosed until the second decade: how we did it, Allergy Rhinol. 7 (2) (2016) 82–84, https://doi.org/10.2500/arr.2016.7.0155.
[6] R. Riepl, M. Scheithauer, T.K. Hoffmann, N. Rotter, Transnasal endoscopic treatment of bilateral choanal atresia in newborns using balloon dilatation: own results and review of literature, Int. J. Pediatr. Otorhinolaryngol. 78 (3) (2014) 459–464, https://doi.org/10.1016/j.ijpedit.2013.12.017.
[7] G.C. De Vincentitiis, M.L. Panatta, C. Scoleri, G. Mathew, A. Kerwan, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Pediatr. Otorhinolaryngol. 118 (2019) 185–194, https://doi.org/10.1016/j.ijpedit.2019.10.034.
[8] A. Kars, F. Bingol, F. Atalay, A rare case report: bilateral choanal atresia in an adult patient, Eur. J. Rhinol. Allergy 3 (1) (2020) 26–28, https://doi.org/10.1016/j.ejral.2020.03.015.
[9] E. Tatar, A. Ozdek, F. Akcan, H. Korkmaz, Bilateral congenital choanal atresia encountered in late adulthood, J. Laryngol. Otol. 126 (9) (2012) 949–951, https://doi.org/10.1016/j.jlarytol.2012.03.009.
[10] M.W. El-Anwar, A.A. Nofal, M.A. El-Ahl, Endoscopic repair of bilateral choanal atresia, starting with vomer resection: evaluation study, Am. J. Rhinol. Allergy 30 (3) (2016) 95–99, https://doi.org/10.2500/arr.2016.30.4321.
[11] P. Brihaye, I. Delpierre, A. De Villé, A.B. Johansson, D. Biarent, A. Lenz, Comprehensive management of congenital choanal atresia, Int. J. Pediatr. Otorhinolaryngol. 98 (2017) 9–18, https://doi.org/10.1016/j.ijpedit.2017.04.022.
[12] E. Tatar, B. Ocal, E. Dogan, O. Bayir, C. Saka, A. Ozdek, et al., Stentless endoscopic repair of congenital choanal atresia: is it enough for maintaining choanal patency? Eur. Arch. Oto-rhino-laryngol. 274 (10) (2017) 3673–3678, https://doi.org/10.1007/s00405-017-4702-9.
[13] V.A. Patel, J. Ramadan, M.M. Carr, Congenital choanal atresia repair: an analysis of adverse perioperative events, Otolaryngol. Head Neck Surg. 159 (5) (2018) 920–926, https://doi.org/10.1177/0194599818797237.
[14] J.B. Meleca, S. Anne, B. Hopkins, Reducing the need for general anesthesia in the repair of choanal atresia with steroid-eluting stents: a case series, Int. J. Pediatr. Otorhinolaryngol. 118 (2019) 185–187, https://doi.org/10.1016/j.ijpedit.2019.01.004.
[15] V. Kancherla, P.A. Rommitt, I. Sun, J.C. Carey, T.L. Burns, A.M. Siega-Riz, et al., Descriptive and risk factor analysis for choanal atresia: the National Birth Defects Prevention Study, 1997–2007, Eur. J. Med. Genet. 57 (5) (2014) 220–229, https://doi.org/10.1016/j.ejmg.2014.02.010.
[16] A. Merrow, S. Harharaen, Imaging in pediatrics, 1st ed., Elsevier, 2017.
[17] E. Vercelli, M.E. Rossi, R. Nicollas, J.M. Triglia, Prognostic factors and management of patients with choanal atresia, J. Pediatr. 204 (2019) 234–239.e1, https://doi.org/10.1016/j.jpeds.2018.08.074.