

CASE REPORT

Craniofacial/Pediatric

Long-term Results of Three-layered Closure of Oronasal Fistula: A Case Report

Bogna Zielinska-Kazmierska, MD Joanna Grodecka, MD Weronika Lucas Grzelczyk, MD Magdalena Jozefowicz-Korczynska, MD, PhD

Summary: An oronasal fistula is an interconnection between the oral and nasal cavities. It interferes with normal functioning and affects quality of life by impeding eating and speech. The most common management strategy is surgical treatment. The aim of our study was to present the results of a long-term follow-up of effective treatment of a case of oronasal fistula that occurred without any cause and comorbidities. A 46-year-old woman reported the sudden appearance of a painless oronasal fistula. She demonstrated nasal regurgitation-that is, loss of fluids through the nose while drinking-and slurred, nasal speech. Clinical examination revealed a soft tissue defect (diameter: 5 mm) on the left side of the palate. Computed tomography revealed a gap in the hard palate bone, measuring 25×30 mm. No complications were noted following the procedure, nor at 10-year follow-up. It seems that the use of bone substitute and palatal flap for spontaneous oronasal fistula closure was fully justified. The patient may have had an undiagnosed congenital defect of the hard palate, and the deficit may have appeared due to bone loss occurring in her forties. However, no consensus exists regarding the best treatment management for oronasal fistulae located in the palatal region, and further comparative studies between the existing techniques are needed. (Plast Reconstr Surg Glob Open 2021;9:e3964; doi: 10.1097/GOX.000000000003964; Published online 17 December 2021.)

fistula is a pathological, abnormal connection between two or more anatomical spaces or organs. Oronasal fistulae arise as an interconnection between the oral and nasal cavities. They are relatively rare anatomical defects¹⁻⁴ and are believed to be caused by genetic and environmental factors, as well as teratogenic substances. Oronasal fistula is recognized as one of the symptoms of congenital syphilis, and may be attributed to chronic cocaine abuse.^{5,6} It may also be a result of iatrogenic complications such as failure to heal after surgical repair of a cleft palate, alveolar treatment^{3,7,8} removal of rhinoliths, and surgical removal of ectopic teeth. It has also occurred after traffic accidents.9-11 A few cases have arisen as a result of nasal stone removal¹ or vein embolization after epistaxis.⁴ Palatal perforation can also occur after nasal septoplasty.^{12,13} Furthermore, they can develop as a late sequela of osteoradionecrosis of the maxillary

From the Balance Disorders Unit, Department of Otolaryngology, Head and Neck Oncology, Norbert Barlicki Memorial Teaching Hospital No. 1 of the Medical University of Lodz, Lodz, Poland. Received for publication May 29, 2021; accepted October 8, 2021. Copyright © 2021 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000003964 bone with impaired healing, and have been associated with the administration of chemotherapy. $^{\rm l4}$

This interconnection between the oral and nasal cavities reduces quality of life by impeding eating and speech.¹⁵ As such, a great number of surgical techniques have been developed for managing the condition, including obturator prosthesis, autologous and alloplastic grafts, and local, regional, and free nonvascularized or vascularized flaps.^{7,15-18} However, no consensus exists regarding the best treatment type for oronasal fistulae located in the palatal region, and further comparative studies are needed in this regard.¹⁹

Although the literature includes cases of oronasal fistulae occurring as a result of injuries to the maxillofacial region, oncological surgeries, cleft palate, or extraction of teeth retained in the maxilla,^{7,16–18} none describe any cases of spontaneous oronasal fistulae in middle-age patients. Therefore, this study presents the results of a long-term follow-up examination of successful treatment of an oronasal fistula, which occurred without any cause and comorbidities.

CASE DESCRIPTION

The patient, a 46-year-old woman, requested a maxillofacial surgical consultation regarding ailments caused by an oronasal fistula. The consultation took place at Norbert Barlicki Memorial Teaching Hospital in Lodz, Poland, in

Disclosure: The authors have no financial interest to declare in relation to the content of this article. September 2010. The fistula appeared suddenly and painlessly. The subject demonstrated nasal regurgitation—that is, loss of fluids through the nose while drinking—and slurred, nasal speech. Clinical examination identified loss of soft and hard tissue (5 mm diameter) on the left side of the palate (Fig. 1).

The patient consulted with laryngologists, who found a defect in the lower part of the cartilage and bone nasal septum on clinical examination. The patient did not report any nasal symptoms. A few years earlier, during a laryngology consultation, she was informed about a defect in the nasal septum, but as it did not cause any discomfort, the patient did not decide to pursue treatment. A tri-plane computed tomography (CT) revealed a congenital absence of septum on the left side of the hard palate, measuring 25×30 mm, and located in the anterior segment of the nasal septum. These findings confirmed the initial clinical diagnosis (Fig. 2).

The patient was offered prosthetics treatment at the prosthetics department, to which she agreed. A partial cast was made and fitted, with a metal plate covering the hole of the palate. However, this type of treatment was not satisfactory: the prosthetics caused a lot of discomfort, and some liquid still continued to pass through the nose. Based on the clinical examination, analysis of CT scans, and the patient's expectations, a treatment plan was presented. The possible complications were discussed with the patient before the procedure, and she agreed to the treatment. The patient was fit and well with no comorbidities, and the laboratory blood tests were in normal ranges.

Surgery Description

Surgery was performed under general anesthesia with the aid of endotracheal intubation. An incision was made on the palatal side, 5 mm below the gingival margin and continued along the teeth from 17 to 27. A mucosal-periosteal flap was prepared, including palatal arteries on both sides. Then, the mucosal-periosteal flap was removed from the palate, up to the back of the hard palate, to expose the bone defect.

The fistula was extracted and secured with mattress sutures, without tissue stretching. Next, the nasal mucous membrane was attached using mattress sutures, and a collagen bioresorbable membrane (Biotec) was applied. The membrane was then covered with a soft titanium mesh



Fig. 1. The visible oronasal fistula on the palate before surgery.



Fig. 2. A frontal CT scan of the maxilla showing a visible bone defect: hard palate and nasal septum.

and fixed to the surrounding palatal bone. The mesh was finally covered by a mix of two bone substitutes: Cerasorb and Bond Bone. Upon hardening, they formed a smooth and hard surface. Finally, the reconstructed area was covered by the mucoperiosteal palatal flap and sutured tightly using occasional sutures, thus achieving a planned lack of palatal bone cover. A large pressure dressing was applied and secured to the teeth. A feeding tube was inserted through the nose into the stomach. The patient tolerated the surgery very well. On postoperative day 5, due to an increased activation of the vomiting reflex, the dressing was removed. The plate remained stable and the feeding tube was retained. On day 10, after feeding tube removal, the patient was discharged from the clinic. On day 12, the sutures were removed. The surgical wound healed properly. After the operation, the patient remained under the supervision of the Department of Clinical Surgery. After 4 months, a CT scan confirmed the closure of the palate without any pathological effects in the nasal cavity (Fig. 3). The CT examination was performed again after 20 months and no pathological effects were detected (Fig. 4). The patient has been under the supervision of our outpatient clinic for 10 years, and palate closure has been repeatedly confirmed during clinical examinations.

DISCUSSION

Our 10-year follow-up study confirmed effective surgical closure of a spontaneus oronasal fistula without any trauma, infection, or additional comorbidity. This case was complex because there was a significant defect in the bones of the hard palate and the nasal septum, which required correction of the hard tissues. Without this procedure, the small mucosal fistula would certainly regenerate. The lack of a bone base was an indication that it should be reconstructed before palatal mucosa flap plasty.



Fig. 3. Image of the operated area 24 months after the procedure.



Fig. 4. A frontal CT scan of the maxilla 2 months after the procedure.

It is worth emphasizing that surgical treatment of fistulae is often complicated by a high recurrence rate of up to 68% in middle-aged patients.^{3,16} All surgical methods carry a risk of complications such as detachment, bleeding from the flaps, or fistula recurrence.²⁰ A meta-analysis of 11 studies concerning the incidence of oronasal fistula after cleft palate surgery in 2505 children under the age of 4 years found a very low recurrence rate, suggesting that children have different growth potential and better surgical outcomes than adults, and thus, a lower complication rate.²¹ For that reason, our patient was forewarned of the possible complications. Despite this, she was otherwise completely healthy: she worked as a traffic controller at a major train station and had to undergo stringent, periodical medical checkups. We suspect that she likely had an undiagnosed congenital defect of hard palate and nasal septum, but these conditions did not cause her any discomfort before the appearance of the fistula.

The selected method of fistula closure with the use of a collagen membrane, titanium mesh, and bone substitutes seemed the most suitable. While the soft tissue loss was 5 mm, considerably more palate bone was lost (25–30 mm). In our opinion, by passing the oral feeding process is of paramount importance to proper healing of the wound. The insertion of the feeding tube through the nose into the stomach allowed the wound and the dressings to be maintained in pristine cleanliness.

The final result was satisfactory for both us and the patient. Cleft palate and oronasal fistula treatment using collagen membrane has also been reported.³ An untreated oronasal fistula is not a life-threatening condition, but may negatively affect quality of life, leading to development of other comorbidities. In many cases, surgical treatment is the only therapeutic option. In the literature, treatment methods vary from prosthetics to surgery. The closure of palatal fistulas can be achieved using different techniques, depending on the size and the experience of the surgeon.²² Although there is a continuous search for new solutions, few articles provide a detailed method that seems to be both simple and effective.^{22,23}

The use of flaps (or local, regional, and distant grafts of mucosa, fat, cartilage, bone, fascia, muscle, or dermis) has been widely described. Additionally, nonresorbable, synthetic materials such as acellular dermal matrix²⁴ and Poly-D with L-lactic acid¹⁰ are becoming more popular, as are more recent innovative techniques that alter resorption periods in resorbable membranes.^{22,24} In all cases, the specialist must perform a multilayer and tension-free reconstruction where possible.

This is the first time we have encountered such a case of oronasal fistula in adulthood. Our approach seems to be adequate and the follow-up indicates good effectiveness. The main limitation is the lack of more patients who would undergo this type of surgery.

CONCLUSIONS

The procedure was successful, and no complications were observed during the 10-year follow-up. Therefore, it seems that the employed bone substitute and palatal flap procedure is suitable for the closure of spontaneous oronasal fistulae.

> Weronika Lucas Grzelczyk, MD Medical University of Lodz Lodz, Poland E-mail: weronika.lucas@gmail.com

ACKNOWLEDGMENTS

This study was supported by a grant from Medical University of Lodz (No 503/1-036-02/503-21-002-18). The authors express their sincerest gratitude to Bogumila Bruc, MA from the Information and Library Center of the Medical University of Lodz for her assistance with the literature search.

REFERENCES

 Igoumenakis D, Athanasiou S, Mezitis M. A bizarre cause of extensive oronasal fistula. J Craniofac Surg. 2013;24:e483–e484.

- Arora S, Garg LN, Julaha M, et al. Naso-oral fistula due to rhinolithiasis: a rare presentation. J Oral Sci. 2009;51:481–483.
- **3**. Sader R, Seitz O, Kuttenberger J. Resorbable collagen membrane in surgical repair of fistula following palatoplasty in nonsyndromic cleft palate. *Int J Oral Maxillofac Surg.* 2010;39:497–499.
- 4. Ntomouchtsis A, Venetis G, Zouloumis L, et al. Ischemic necrosis of nose and palate after embolization for epistaxis. A case report. *Oral Maxillofac Surg.* 2010;14:123–127.
- Rilo B, Fernández-Formoso N, DaSilva L, et al. Prosthetic management for a palatal perforation from cocaine abuse. J Prosthodont. 2015;24:586–588.
- 6. Rampi A, Vinciguerra A, Bondi S, et al. Cocaine-induced midline destructive lesions: a real challenge in oral rehabilitation. *Int J Environ Res Public Health.* 2021;18:3219.
- Denadai R, Seo HJ, Lo LJ. Persistent symptomatic anterior oronasal fistulae in patients with Veau type III and IV clefts: a therapeutic protocol and outcomes. *J Plast Reconstr Aesthet Surg.* 2020;73:126–133.
- 8. Stein MJ, Zhang Z, Fell M, et al. Determining postoperative outcomes after cleft palate repair: a systematic review and metaanalysis. *Plast Recontr Aesthet Surg.* 2019;72:85–91.
- 9. Dhingra S, Gulati A. Teeth in rare locations with rare complications: an overview. *Indian J Otolaryngol Head Neck Surg.* 2015;67:438–443.
- Stalder MW, Wise MW, Dupin CL, et al. Versatility of subscapular chimeric free flaps in the secondary reconstruction of composite posttraumatic defects of the upper face. *Craniomaxillofac Trauma Reconstr.* 2015;8:42–49.
- Koçak HE, Özdamar K, Bilgi B, et al. A rare cause of intranasal mass: bilateral ectopic nasal teeth. *Iran J Otorhinolaryngol.* 2017;29:287–289.
- Tilaveridis I, Kalaitsidou I, Kyrgidis A, et al. Palatal perforation as a rare complication of nasal septoplasty. *Aesthetic Plast Surg.* 2016;40:850–853.
- Alhedaithy RA, Alsaleh SA. Oronasal fistula post-septoplasty. Saudi Med J. 2017;38:310–313.

- 14. Bhattacharyya T, Koto M, Ikawa H, et al. Assessment of risk factors associated with development of oronasal fistula as a late complication after carbon-ion radiotherapy for head and neck cancer. *Radiother Oncol.* 2020;144:53–58.
- 15. Torroni A, Longo G, Marianetti TM, et al. The use of a superiorly based melolabial interpolated flap for reconstruction of anterior oronasal fistulas: an easy and practical solution. *Ann Plast Surg.* 2015;75:163–169.
- Anani RA, Aly AM. Closure of palatal fistula with local doublebreasted mucoperiosteal flaps. J Plast Reconstr Aesthet Surg. 2012;65:e237–e240.
- Lee JY, Alizadeh K. Spacer facial artery musculomucosal flap: simultaneous closure of oronasal fistulas and palatal lengthening. *Plast Reconstr Surg.* 2016;137:240–243.
- Jeong EC, Yoon S, Jung YH. Radial forearm osteocutaneous free flap for recontruction of hard palate with alveolar defect. J Craniof Surg. 2017;28:e438–e440.
- Miranda BL, Júnior JLA, Paiva MAF, et al. Management of oronasal fistulas in patients with cleft lip and palate. *J Craniofac Surg.* 2020;31:1526–1528.
- Eunsum S, Yun HJ, Karm MH, et al: Airway management in pediatric flap division for oronasal fistulaclosure: a case report. *J Dent Anesth Pain Med.* 2018;18:309–313.
- Bykowski MR, Naran S, Winger DG, et al. The rate of oronasal fistula following primary cleft palate surgery: a meta-analysis. *Cleft Palate Craniofac J.* 2015;52:e81–e87.
- 22. Alonso V, Abuin AS, Duran C, et al. Three-layered repair with a collagen membrane and a mucosal rotational flap reinforced with fibrine for palatal fistula closure in children. *Int J Pediatr Otorhinolaryngol.* 2019;127:109679.
- 23. de Souza Lopes PH, Sampaio Dde O, de Souza Menezes BL, et al. Combined palatal flap and titanium mesh for oroantral fistula closure. *Ann Maxillofac Surg.* 2015;5:89–92.
- 24. Agir H, Eren GG, Yasar EK. Acellular dermal matrix use in cleft palate and palatal fistula repair: a potential benefit? *J Craniofac Surg*. 2015;26:1517–1522.