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Case Report

Simultaneous bilateral choanal and nasopharyngeal stenosis in an adult patient: case report and treatment approach

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ABSTRACT

Choanal and nasopharyngeal stenosis in adults are extremely rare. They usually are detected and treated in pediatric age as they typically result from congenital malformations. In adulthood, they mostly result from acquired causes such as iatrogenic trauma after surgeries, infectious or autoimmune diseases, or radiotherapy. However, sometimes it is not possible to determine the exact etiology. Treatment is challenging, with restenosis being the most feared and frequent complication. With this case report, we aimed to highlight the diagnostic and treatment approaches for choanal and nasopharyngeal stenoses in adults. We present a 61-year-old patient with persistent bilateral nasal obstruction and discharge, snoring, and recurrent frontal headaches for years which turned out to be a bilateral choanal and nasopharyngeal stenosis. Previously, the patient had been diagnosed with chronic rhinosinusitis without any other relevant medical history. Additionally, no previous surgical procedures or radiation therapy were found to be associated with the etiology. The patient was treated with surgery. A combined modality of trans-nasal endoscopic excision of choanal and nasopharynx fibrous tissue using cold cutting instruments and posterior septectomy followed by the topical application of Mitomycin-C, without stenting, was an effective option.

Keywords: Choanal stenosis, Nasopharyngeal stenosis, Adult, Trans-nasal approach, Mitomycin-C

INTRODUCTION

Simultaneous choanal and nasopharyngeal stenosis in adults is extremely rare. Congenital malformations, as newborns are obligatory nasal breathers, are usually detected and treated in pediatric age due to the respiratory distress they cause.[1] The acquired cases mainly result from iatrogenic trauma caused by extensive and aggressive nasal, oropharyngeal and/or nasopharyngeal surgeries or secondary to radiotherapy.[1-4] They may also be related to inflammatory pathology or, more rarely, granulomatous, or autoimmune diseases.[5,6] In extremely rare cases, the cause is uncertain and remains undetermined. We present a rare clinical case of idiopathic bilateral choanal and nasopharyngeal stenosis in an adult patient and its treatment approach.

CASE REPORT

A 61-year-old man presented with persistent bilateral nasal obstruction and discharge, snoring, and recurrent frontal headaches for years. He does not remember if he also had these complaints in childhood or adolescence. He mentioned a previous diagnosis of chronic rhinosinusitis, about ten years ago, treated with cycles of topical corticosteroids in acute phases, but only with partial improvement. Besides that, the patient had no epistaxis, crusting, or ocular symptoms. There were also no ear or throat complaints and no history of neoplastic disease, head trauma, surgical procedures, congenital abnormalities, or autoimmune disease.

On examination with flexible nasoendoscopy, in addition to a nasal septum deviation and nasal polyps confined to

the middle meatus, he presented accumulated mucous rhinorrhea in the nasal cavities and bilateral choanal stenosis (<5 mm), with a smaller choanal opening on the left side (Figure 1). Examination of the nasopharynx through the choanal stenosis revealed a thick fibrous tissue of the posterior and lateral walls, with partial obliteration of its lumen. Oral cavity and oropharynx examination was normal.

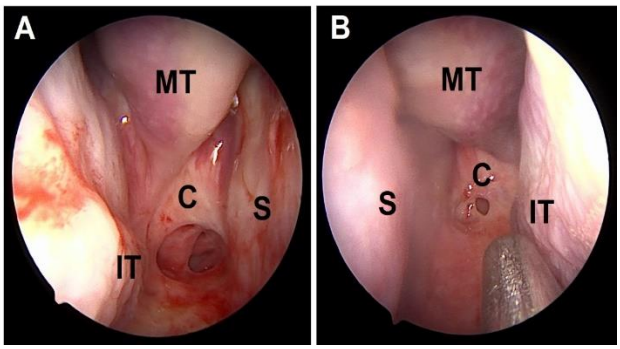

Figure 1: Preoperative endoscopic view of choanal stenosis (A) right nasal cavity, and (B) left nasal cavity.

C=Choana; IT=inferior turbinate; MT=middle turbinate; S=septum.

A computed tomography (CT) scan of the nose and paranasal sinuses was performed which revealed a concentric thickening of the frontal-ethmoidal-maxillary mucosa suggestive of chronic rhinosinusitis, associated with a soft tissue density choanal lumen reduction and partial nasopharyngeal obstruction (Figure 2). Further laboratory tests revealed a C-reactive protein level of 4.10, a sedimentation rate of 11, with no changes in the immunoglobulin study, normal serum concentration of angiotensin-converting enzyme, and negative antinuclear antibody and rheumatoid factor.

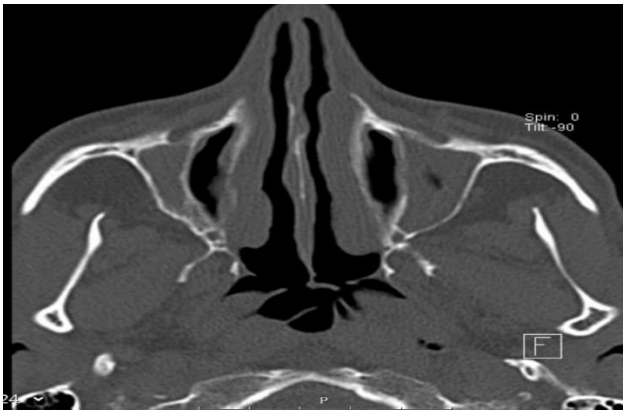

Figure 2: Axial view of CT scan showing a bilateral low density choanal lumen stenosis and partial nasopharyngeal obstruction.

Given the patient's complaints, it was decided to conduct a trans-nasal endoscopic surgery to enlarge the choanal lumen and for assessment of nasopharyngeal stenosis.

Under general anesthesia and endoscopic visualization using a 0º endoscope, we start to decongest the nasal cavity with pledgets soaked in 10% cocaine solution, which were removed after 10 minutes, and additionally injected 1% lidocaine with 1:100,000 epinephrine with a spinal needle around the stenosis and in the posterior septum. The Kerrison punch forceps was used to widen the circumferential opening of the choanal stenosis. Additional fibrotic tissue from the posterolateral wall of the nasopharynx was excised with a microdebrider and Weil-Blakesley through-cutting forceps. To further enlarge the neochoana, posterior septectomy was performed by partial resection of the posterior bony septum using backward biting forceps. We tried to preserve as much as possible the adjacent and overlying normal mucosa. In addition to the choanal and nasopharynx permeabilization procedure, the patient also underwent bilateral uncinectomy, maxillary antrostomy, anterior ethmoidectomies, Draf I procedure, and inferior turbinoplasties for concomitant chronic rhinosinusitis. All excised tissue was sent for histopathological examination. Then, at sites where tissue excision was performed in the choanas, we applied topically cotton pledgets soaked with 1 ml of mitomycin-C at a concentration of 0.4 mg/ml, that were held in place under endoscopic visualization for 5 minutes. After the surgical procedure, septal silastic splints and bilateral nasal packing with Merocel® were placed. No nasal stents were inserted. The patient was discharged the following day on 8 days of oral antibiotic (amoxicillin/clavulanic acid 875 mg/125 mg, bid), 5 days of systemic corticosteroid therapy (deflazacort 30 mg, bid), and regular saline douches. Nasal packing was removed after 8 days.

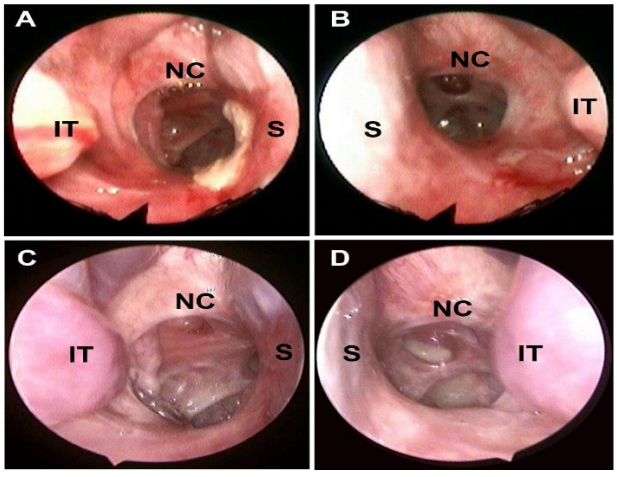

Figure 3: Postoperative endoscopic view (A) right nasal cavity, 1 month follow-up, (B) left nasal cavity, 1 month follow-up, (C) right nasal cavity, 6 months follow-up, and (D) left nasal cavity, 6 months follow-up.

NC=Neochoana; IT=inferior turbinate; S=septum.

Weekly endoscopic evaluation was performed during the first month for cleaning of crusts and for results evaluation. Steroid nasal spray (50 mcg of fluticasone furoate, qd) was

administered after the first postoperative month, which was continued for three months. After the first month and until the sixth month after the surgery, a monthly follow-up visit with an endoscopic evaluation was maintained. Short- and medium-term follow-up (1 and 6 months, respectively) showed good results, with choanal and nasopharyngeal patency and significant improvement in symptoms, without velopharyngeal incompetency (Figures 3 and 4). The histopathological analysis of the resected tissue revealed nasal mucosa tissue with moderate lymphoplasmacytic infiltrate and submucosal hyalinized fibrosis. To date, it has not been possible to determine the exact etiology of the bilateral choanal and nasopharyngeal stenosis of the presented case.

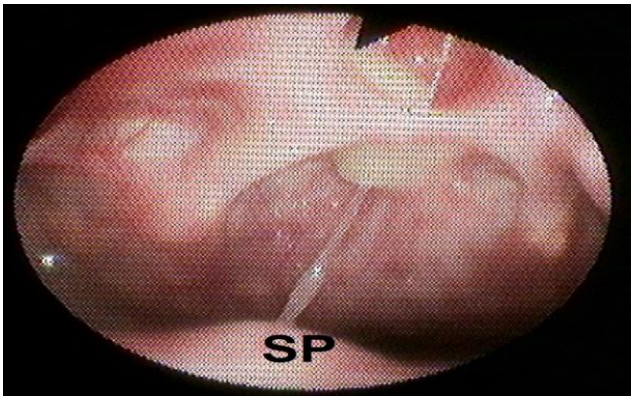

Figure 4: Postoperative endoscopic view of the nasopharynx.

SP=Soft palate.

DISCUSSION

Severe bilateral choanal and nasopharyngeal stenosis are rare entities, especially those diagnosed only in adulthood. Most tend to be congenital, specially choanal stenosis or atresia, and typically present in the first days of life and are treated almost immediately due to the associated breathing difficulties.[7] When these anomalies are acquired, they mainly result from iatrogenic trauma caused by nasal surgeries, oropharyngeal surgeries, and/or nasopharyngeal surgeries.[2] There are several described factors that can precipitate the formation of stenosis after surgical procedures, like patients with a history of keloid formation, excessive postoperative bleeding, postoperative infection, LASER surgery, gastric reflux, excessive resection of posterior tonsillar pillars in adenotonsillectomy, adenoidectomy in conjunction with various palatoplasty techniques, or after excessive electrocautery for bleeding control.[1,4] Among the other causes of acquired choanal and nasopharyngeal stenosis are radiotherapy for nasopharyngeal carcinomas, granulomatous diseases such as sarcoidosis and lupus, infections of the upper respiratory tract, including syphilis, rhinoscleroma, diphtheria, and tuberculosis, especially prior to antibiotics, and autoimmune diseases like Sjögren's Syndrome.[1,5,6,8,9] An accurate diagnosis of the etiology and an appropriate local and systemic treatment approach can only be achieved through a detailed clinical history and an analytical analysis that focuses on these possible secondary causes, as we did in this case.

Since we were unable to identify an etiological factor behind the simultaneous bilateral choanal and nasopharyngeal stenosis, a congenital malformation must be considered, making this case extremely rare.

Nasal endoscopic examination, although very useful and effective in the initial diagnosis of choanal or nasopharyngeal stenosis, does not allow the characterization of the type of stenosis nor does it allow an accurate assessment of the nasopharynx in the event of concomitant choanal stenosis or atresia. In congenital choanal atresia, a review of the anatomical characteristics revealed that approximately 70% are mixed bony-membranous whilst the remainder are pure bony.[10] On the other hand, most acquired choanal atresia is fibrous-membranous.[8] Thus, high-resolution CT scan must be performed to confirm the clinical diagnosis and for accurate characterization of the nature and thickness of the stenosis. In our patient, pure membranous choanal and nasopharyngeal stenosis were demonstrated with the CT scan.

Given the negative consequences associated with simultaneous severe bilateral choanal and nasopharyngeal stenosis like nasal obstruction, chronic sinonasal problems, sleep apnea, swallowing difficulty, and eustachian tube and middle ear problems, treatment is a priority. The type of treatment will depend on the location, etiology, severity, and type of stenosis. The primary goals are to achieve adequate airway patency and prevent recurrences.

In mild stenosis, the use of local injection of triamcinolone acetonide has been described.[11] However, in most cases, it is necessary to perform surgery to remove all fibrous tissue, while preserving nasal mucosal flaps to cover any denuded bone to prevent granulation.[12] There is no universally accepted surgical technique, so several have been described in the literature: tissue excision with microdebrider or cold sharp instruments, excision with carbon dioxide LASER, excision with coblation or temperature-controlled radiofrequency devices, endoscopic balloon dilation, Z palatoplasty or laterally based pharyngeal flap (the latter three techniques are mainly described for nasopharyngeal stenosis and not in isolated choanal stenosis).[5,13,14] For each technique, several approaches can be performed. The trans-nasal endoscopic assisted procedure appears to be the most common, as it provides clear visualization of the surgical field, precise removal of affected tissues, the capability of creating mucosal flaps and posterior septectomy, if necessary, and a short hospital stay.[8,15] However, the trans-septal, trans-palatal, and trans-oral approaches are also valid options, especially in pediatric age where the small nasal cavities preclude the endoscope and instruments introduction. Once again, no single approach is superior in terms of

functional results, but most cases described in the recent literature were treated by an endoscopic trans-nasal approach, as we did in our case, and no technical complications were described.[12]

Nevertheless, surgical treatment of choanal as nasopharynx stenosis is very challenging due to the high risk of recurrence, occurring in up to 36% of cases.[16] Thus, the treatment must include measures to minimize this complication. Topical application of mitomycin-C, at a concentration of 0.2-1 mg/ml, usually 1 ml, for 3-5 min, has been studied as an adjunct to surgical repair. Mitomycin-C is an antibiotic produced by Streptomyces caespitosus, which inhibits fibroblast proliferation and growth when applied topically to control scar tissue formation.[9] Due to its toxic nature, it is used off-label, but no systemic side effects are described in topical applications, and it is already widely used to treat airway stenosis. A retrospective study by Carter et al demonstrated that mitomycin-C was associated with decreased granulation tissue formation and a decreased need for revision surgery.[17]

A second measure described to prevent recurrences is postoperative stenting for maintaining neochoana patency, which can be last between two weeks and two months after surgery. Nasal stenting seems to have an important role after the correction of stenosis secondary to radiotherapy or iatrogenic trauma with LASER surgeries, but overall there seems to be no difference in the rate of restenosis with and without the use of stents.[1,9] Additionally, the disadvantages of its use include longer hospitalizations, the need for antibiotics therapy for a long period of time, the possibility of foreign body reaction and pressure necrosis of the columella, as well as trauma to the nasal mucosa resulting in the formation of granulation tissue.[7] Therefore, we decided not to place a nasal stent after permeabilization surgery in the presented case. These adjunct treatments, whether the use of stents or the topical application of mitomycin-C, are still the subject of some controversy and do not have sufficient scientific evidence to be recommended in clinical practice, especially in the adult population, where studies on the treatment of choanal and nasopharyngeal stenosis and atresia are extremely rare.

Finally, endoscopic posterior septectomy is another approach to further enlarge the neochoana and help in achieving enduring anatomical choanal patency. A case series study and systematic review on this topic performed by Shute et al showed that the resection of up to half of the bony posterior septum is one of the major determinants to avoid revision surgery.[12]

CONCLUSION

We present an extremely rare case of idiopathic simultaneous choanal and nasopharyngeal stenosis in an adult patient. Although the patient's age, a purely fibrous-membranous stenosis, and the simultaneous occurrence of choanal and nasopharyngeal stenosis are more suggestive of an acquired cause, the clinical history and etiological study were negative, so, in this case, the hypothesis of a congenital malformation must be considered. With this case, we intend to review the diagnostic approach and the existing treatment options. We conclude that a high-resolution CT scan and a complete etiological study directed to the possible secondary causes should be performed to guide our treatment approach. Although a variety of surgical techniques are available, none seem to provide completely satisfactory results, so restenosis remains a prevalent and feared problem. The management with a trans-nasal endoscopic approach and surgical excision of the fibrotic tissue with cold cutting instruments is safe and allows a good operative field vision. Posterior septectomy provides an enlargement of the neochoana with less possibility of restenosis. Topical application of mitomycin-C at a concentration of 0.4 mg/ml, without post-operative stenting, seems to be an effective and safe option to ensure good long-term results. Further research, with detailed comparisons of existing surgical techniques and adjunctive therapies to prevent recurrences, should be performed to clarify the optimal treatment approach in choanal and nasopharynx stenoses in adults.

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