



Nasal septum mucocele in a patient with two other mucoceles of the paranasal sinuses: late complications of maxillofacial trauma*

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Abstract

Introduction: Mucoceles are pseudocystic lesions that, despite being benign, may cause diverse symptoms related to its expansive nature. Although their most common location in the nose is the frontoethmoidal sinuses, they can occur in other sites. The authors present a case of a patient with a nasal septum mucocele besides mucoceles in other locations, reviewing the literature regarding these entities.

Case presentation: The patient, who had a history of a previous major maxillofacial trauma, was referred to Otorhinolaryngology evaluation because of a long-standing history of nasal obstruction. On nasal endoscopy, a bilateral posterior nasal septum bulging at the choanae level was observed; an irregularity on the external wall of the maxillary sinus was noted in the palpation of the midface. Computed tomography (CT) showed a posterior nasal septum hypodense lesion besides two other lesions with similar features, one in the maxillary sinus, and another in the anterior ethmoid. A magnetic resonance imaging followed, and confirmed the lesions, that were hyperintense in T1 and did not show enhancement with gadolinium. A surgical approach, namely marsupialization through endoscopic sinus surgery, was performed. Histopathology confirmed the three lesions as mucoceles. The patient reported nasal obstruction complaints resolution; he has been in follow-up with no signs of relapse.

Discussion and conclusion: Despite being extremely rare, nasal septum mucoceles should be remembered when evaluating midline nasal lesions, and included in the differential diagnosis. When evaluating patients with a history of major maxillofacial trauma other atypical findings may be present.

Key words: mucocele, nasal septum mucocele, midline nasal lesion, nasal obstruction

Introduction

Paranasal sinus mucoceles are relatively common lesions. Several factors may predispose to its development, of which previous nasal surgery and trauma are the most frequent, but also radiotherapy, rhinosinusitis, and cystic fibrosis⁽¹⁾. Although their most common location in the nose is the frontalethmoidal sinuses, they can occur in other parts of the nasal cavities or paranasal sinuses.

The authors report a case of a patient with multiple mucoceles, one with an extremely rare location, the nasal septum (NS).

Case presentation

A 49-year-old male presented for Otorhinolaryngology evaluation complaining of bilateral chronic nasal obstruction, that had begun several years before and did not improve with local medical therapy. He had a history of major maxillofacial trauma in a car accident 24 years before, with multiple fractures that needed surgical correction, besides right eye enucleation; he had a complete visual loss of the left eye. He had multiple scars on the skin of the nose and malar region. On nasal endoscopy, there was a posterior bilateral bulging of the NS, covered by normal mucosa, that reached the level of the choanae (Figure 1).



Figure 1. Nasal endoscopy on the right nostril showing obliteration of the choanae (IT: inferior turbinate; MT: middle turbinate; NS: nasal septum; SM: septal mucocele; C: choanae; F: floor of the nose).



Figure 2. Computed tomography in the coronal (A and B) and sagittal planes (C) showing the following hypodense lesions: nasal septum lesion (A and C); maxillary lesion (B); anterior ethmoid lesion (C).

This midline lesion caused significant impairment of nasal permeability. There was also a deformity of the middle meatus on the right side. Irregularity of the lateral wall of the right maxillary sinus was perceptible in the palpation of the midface. The patient underwent CT scan (Figure 2) which revealed signs of previous major maxillofacial trauma and reconstructive surgery, with prosthetic material in both lateral walls of the orbit, and phthisis bulbi of the left eye; besides, a hypodense septal lesion was observed, which deformed and eroded antero-inferiorly the sphenoid sinus, namely the sphenoid's rostrum, and partially obstructed choanae bilaterally; another lesion with similar features was found on the right maxillary sinus, which eroded the lateral and posterior wall of the maxillary sinus and protruded to the infratemporal fossa; another analogous lesion was found in the right ethmoid, namely in the agger nasi cell. The CT scan was followed by a magnetic resonance imaging (MRI). The MRI confirmed those lesions, which were hyperintense in T1 and did show enhancement with gadolinium, and were suggestive of mucoceles (Figure 3).

He underwent endoscopic sinus surgery with marsupialization of all the three mucoceles with drainage of mucoid content from all of them. The patient's nose was packed with absorbable gelfoam in both nasal cavities. The postoperative period was uneventful. Histopathologic analysis of the capsules of the different lesions was compatible with respiratory epithelium, which led to the diagnosis of mucoceles. In the follow-up consultations, the



Figure 3. Magnetic resonance imaging in T1 in axial (A), sagittal (B) and coronal (C) planes showing hyperintense lesions: nasal septum lesion (A and C); maxillary lesion (A and B); anterior ethmoid (C).

Table 1	. Previously	reported se	ptal mucoce	eles in the	literature	(yo: year	old)
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	Gender	Age	Risk Factors	Symptoms at presentation	Treatment
Gall and Witterick ${}^{\scriptscriptstyle (4)}$	Male	62 yo	Nasal surgery (septoplasty)	Nasal obstruction	Endoscopic Marsupialization
Hermann and Jahnke ⁽²⁾	Male	54 yo	Nasal surgery (FESS)	Facial pain located in the infraorbital region	Endoscopic Marsupialization
Lei et al. (11)	Male	37 уо	Ø	Enlarging painful nasal mass	Endoscopic marsupialization
Taskin et al. (3)	Male	45 yo	Ø	Bilateral nasal obstruction	Endoscopic complete excision
Aynali et al. (14)	Male	34 yo	Nasal surgery (rhinoplasty) 15 years before	Bilateral nasal obstruction	Endoscopic complete excision
Lo Casto et al. (5)	Male	49 yo	Nasal surgery (FESS for frontal mucocele excision)	Bilateral nasal obstruction	Endoscopic marsupialization
Akidil et al. ⁽⁹⁾	Male	13 yo	Ø	Nasal obstruction	Endoscopic complete excision
Lee et al. (6)	Female	52 уо	Ø	Headache	Endoscopic marsupialization
Hong et al. (7)	Male	52 уо	Maxillofacial trauma 35 years before	Bilateral nasal obstruction	Endoscopic marsupialization
Ghorbani et al. (10)	Male	27 уо	Nasal surgery (septoplasty and turbinoplasty) 5 years before	Nasal obstruction and frontal headache	Endoscopic marsupialization
Rawl et al(8)	Male	57 yo	Nasal surgery (3 times) after nasal trauma 35 years before	Nasal obstruction	External approach (modified Killian incision) with complete excision

patient reported resolution of all complaints, and no relapse was diagnosed.

Discussion

NS mucoceles are extremely rare entities and, in the literature, there are only 11 other reported cases (Table 1). As in the case presented, most of the reports refer to middle-aged male patients⁽²⁻⁸⁾. Nonetheless, there are some younger patients, including one in the pediatric age^(9,10).

While some cases had no previous relevant risk factors, most of the patients had undergone nasal surgery in the past, and one

had a history of maxillofacial trauma several years before, as in the case reported in this paper.

There are two main explanations to mucocele formation of the NS: the first is related to the rare occurrence of a pneumatized NS⁽¹¹⁾. The NS, that extends anteriorly from the columella and posteriorly to the rostrum of the sphenoid sinus, includes three components, the membranous septum, the cartilaginous septum, and the bony septum, the latter consisting of the perpendicular plate of the ethmoid and the vomer⁽¹²⁾. Septal pneumatization, more common in the perpendicular plate of the ethmoid, has a variable reported prevalence, ranging from as low as 0,5 to

	Gender	Age	Risk factors	Symptoms at presentation	Mucocele location	Treatment
Price et al. (17)	Female	56 yo	Maxillofacial trauma	Proptosis and diplopia	Left frontal sinus, ethmoid bilaterally, sphenoid sinus, maxillary sinus	Not described
Sadoff et al. (15)	Female	21 уо	Ø	Cellulitis of the eye and malar region	Maxillary sinus bilaterally	External approach (anterior antrostomy)
Thomé et al. (18)	Male	10 months	Cystic fibrosis	Nasal obstruction	Ethmoid bilaterally	Endoscopic marsupialization
Varghese et al. (16)	Female	65 yo	Ø	Proptosis and swelling of the middle canthus	Anterior ethmoid (right) and maxillary sinus (left)	Endoscopic marsupialization
Berlucchi and Reda- elli de zinis ⁽¹⁾	Female	3 уо	Ø	Proptosis	Maxillary bilaterally and ethmoid bilaterally	Endoscopic marsupialization

Table 2. Previously reported patients with multiple mucoceles in the literature (yo: year old).

as high as 18% of the population⁽¹⁰⁾. Some authors advocate that the obstruction of this pneumatized septum is usually a consequence of trauma or inflammation and that this is the primary event that leads to mucocele formation. The second hypothesis refers to the entrapment of mucosa in the septum after surgery of trauma^(4,10). The latter hypothesis is more controversial given how rare it is, for example, after septal surgery⁽¹⁰⁾. In the case presented it is not easy to determine the precise pathophysiology of the lesion, specifically the location where the mucocele started developing. Still, given its NS postero-inferior location, the authors hypothesize it probably initiated at the level of the vomer and expanded anteriorly and posteriorly, caused the observed bulging of the NS that consequently led to the nasal obstruction complaints, and partially eroded antero-inferiorly the sphenoid sinus, namely its anterior wall and rostrum. Another possible explanation would be that the mucocele started at the rostrum/anterior wall of the sphenoid sinus itself, and grew and eroded the vomer anteriorly, causing the findings described. The reported patient presented with bilateral nasal obstruction as the main symptom, which was related to the septal mucocele that obstructed the choanaes. There are important differences between the clinical presentation of NS mucoceles and mucoceles in different locations: NS mucoceles usually present earlier than in other locations, given the common impairment they cause in nasal permeability, producing nasal obstruction complaints. Nasal obstruction is, according to our review, the most common form of presentation of NS mucoceles^(3,5-7,10,13,14). On the other hand, it is not a common symptom at presentation amongst patients with mucoceles in other locations⁽¹⁰⁾. The differential diagnosis of septal mucoceles depends on its exact location and includes midline nasal masses, such as nasal gliomas, nasal encephaloceles, neuroblastoma, nasal dermoid cysts, epidermoid cyst, hemangioma, lymphangiomas, angiofibromas, and also septal abscess and foreign bodies⁽⁸⁾. The reported case is unique for another reason: while there are several cases in the literature describing mucoceles which

individually are responsible for the erosion of multiple paranasal sinuses, literature is scarce in what refers to multiple individual mucoceles in the same patient. There are only other five cases described of multiple mucoceles (Table 2). Most of the patients reported are female, with no identified risk factors for mucoceles^(1,15,16). There are exceptions: one newborn with cystic fibrosis (CF), and one patient with previous major maxillofacial trauma, as in our case^(17,18). The clinical presentation is variable according to the locations of the mucoceles; nevertheless, given the ethmoid is affected in the majority of cases, ophthalmologic symptoms are common^(1,16,17). Differential diagnosis of multiple mucoceles includes mucous retention cysts, polyostotic fibrous dysplasia, multiple myeloma, and aneurysmal bone cyst⁽¹⁵⁾. In the evaluation of nasal lesions, CT scan, and MRI both have an important role⁽¹⁹⁾. Mucoceles are usually round hypodense lesions in CT scan commonly associated with sinus wall expansion and erosion^(8,16,18). Nonetheless, mucocele may have a similar appearance to other benign lesions with CT scans. MRI, on the other hand, can help differentiate mucoceles from other lesions. assess intracranial and intraorbital extension and involvement. The intensity in MRI is variable according to the protein concentration in the secretions and presence of infection and can vary over time⁽¹⁹⁾. With time, protein concentration tends to increase; a recent mucocele may be hypointense in T1 and hyperintense in T2, while a long-evolving mucocele may be hyperintense in T1 and hypointense in T2⁽¹⁹⁾. Besides, using contrast (such as gadolinium) will only enhance the periphery of the lesion and not its content.

Mucocele treatment has been changing throughout the years⁽²⁰⁾. Marsupialization through endoscopic surgery has gained acceptance with time: it creates a perfectly aerated sinus, easy to examine in the postoperative period, and reduces the need for exams. It is suitable for almost all mucocele locations, and, in our case, we were able to treat effectively all of them. External approaches are still valuable in some situations, such as revision cases, laterally located frontal mucoceles, extensive erosion of

the external plate of the frontal sinus, and intracranial complications (for example, subdural empyemas)⁽²⁰⁾. Regardless of the chosen approach, given mucoceles' tendency for relapse, a long follow-up is desirable.

Conclusions

Although extremely rare, NS mucocele should be considered in the differential diagnosis of midline nasal masses, especially if there is a history of maxillofacial trauma or septal surgery. Other atypical findings may emerge as a consequence of complex maxillofacial trauma. In our case, besides the unusual finding of a NS mucocele, other mucoceles were present, giving physical examination and imaging a relevant role in the evaluation of these patients.

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Authorship contribution

AFL wrote the manuscript. FCM, CA, and SV critically revised the manuscript. All authors read and approved the final manuscript.

Conflict of interest

The authors declare that there is no conflict of interest.

Ethics approval

Not applicable.

Consent for publication

Written informed consent for publication of clinical details and clinical images was obtained from the patient in question.

Availability of data and materials

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