Introduction

A mucocele is a benign and relatively rare lesion consisting of an epithelial wall characterized by slow expansive growth in relation to a dynamic process of resorption, erosion, and bone remodelling, and it can result in new cystic formation with a typically sterile mucoid content. The aetiology of mucocele development is not well known, but they are traditionally differentiated into primary forms originated from cystic degeneration of sinus mucosa, and secondary forms originated from obstruction of the paranasal sinus ostium and resulting in mucus accumulation. Such obstruction can be due to anatomical abnormalities of the ostium, sinonasal inflammation, polyps, trauma, previous surgery, and rarely tumours, such as osteomas. In the post-surgical forms, the pathogenetic mechanism is the formation of fibrotic scar bands that obstruct normal sinus drainage.

Paranasal mucoceles develop predominantly in the frontal sinuses (60%), followed in frequency by the ethmoidal (20-30%), maxillary (10%), and sphenoid (2-3%) sinuses. They are usually asymptomatic until they invade the adjacent structures. A mucocele of the nasal septum is a very rare occurrence, with the literature including reports of only four cases in adults and one in a child. Although previous studies have used imaging techniques, such as CT and MRI, for nasal septal mucocele diagnosis, none have used cone beam CT (CBCT). Here we describe the use of MRI and CBCT to investigate the case of a nasal septum mucocele that was discovered 23 years after endoscopic nasal surgery.

Case report

Our patient was a 49-year-old man who had complained for a long time of a modest bilateral nasal obstruction, predominantly on the left, and occasional episodes of headache. Twenty-three years earlier, the patient had undergone removal of a left frontal mucocele, performed with an open approach using the Ogston Luc technique. Endoscopy showed bilateral swelling of the nasal septum covered by normal mucosa with a smooth surface and soft consistency, which considerably reduced the respiratory space (Figure 1). The swelling was in contact with the lateralized left middle turbinate, preventing a complete view of the middle meatus, which appeared devoid of

Financial support: This research received no specific grant from any funding agency of the commercial or non-profit sector.

Conflict(s) of Interest: None.
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non-enhancing content was observed (Figure 2). A mucocele of the nasal septum was diagnosed.

To evaluate the status of subtle bony structures of the midface and to map the ostiomeatal complex prior to endoscopic surgery, we then performed CBCT (Scanora 3D, Soredex, Tuusula, Finland) with 90 kVp, 12.5 mA, 20 s rotation time, FOV 13×14.5 cm, and 0.25×0.25×0.25 mm voxel size. CBCT located the lesion in the posterior portion of the septum, involving the perpendicular plate of the ethmoid and destroying the anterior ethmoid cells on the left side, but sparing the left lamina papyracea. It also caused nasal obstruction on both sides, causing severe narrowing of the nasofrontal duct on both sides and blocking the left infundibulum (Figure 3). The patient underwent endoscopic marsupialization of the lesion, maintaining the right side of the mucosa. A mucinous yellowish secretion was drained. Complete erosion of the perpendicular plate of ethmoidal bone was discovered. The postoperative course was uncomplicated and the patient was free of lesions at follow-up one month later.

Discussion

Secondary mucoceles originate from sinus ostium obstruction and the subsequent retention of mucous secretion inside the cavity. In iatrogenic forms, the obstruction is usually linked to the formation of scarring branches that obstruct paranasal sinus pathological secretions. MRI examination performed on a 1.5-T superconducting MR device (GE Medical Systems, Milwaukee, WI, USA) revealed an ovalar lesion with a regular low-intensity rim and a high-intensity content on both T1 and T2 sequences. After i.v. administration of gadolinium (0.1 mL/kg body weight; Multihance, Bracco, Milano, Italy), a peripheral enhancing rim with non-enhancing content was observed (Figure 2). A mucocele of the nasal septum was diagnosed.

MR study, A: FSE T1 sagittal image showing an ovalar lesion with a regular low-intensity rim and high-intensity content (asterisk) located in the posterior part of the nasal septum. B: FSE T2 coronal image also showing the lesion with high-intensity content (asterisk). C: FSE T1 fat saturated axial image after i.v. administration of paramagnetic contrast medium, showing a peripheral enhancing rim (arrowheads) with non-enhancing content.
Secondary septal mucocele

The innervated area of the trigeminal branches, or nasal obstruction. The most frequent complication of mucoceles is infection with the formation of a pyocele, which leads to the appearance of algic symptoms. In advanced stages, the symptoms depend on the site of involvement. Frontoethmoidal mucoceles (70-80%) may lead to face swelling, especially in the periorbital region, sometimes severe enough to cause exophthalmos and diplopia. Pain is usually absent, but there may be tension in the orbital region and sometimes disorders of ocular motility. The less common maxillary mucoceles may result in swelling of the maxillary region, nasal obstruction, problems with dental arches, and sometimes diplopia the orbital floor is involved. Sphenoidal mucoceles can cause headaches and neuroendocrine or visual symptoms, related to their extension to the floor or saddle apex.

Figure 3
CBCT study. MPR on the coronal (A), sagittal (B), and axial (C) planes, as well as a 3D reformation (D). The lesion is located in the posterior portion of the septum, involving the perpendicular plate of the ethmoid, and destroying the anterior ethmoid cells on the left side but sparing the left lamina papryracea (arrowheads). It caused nasal obstruction on both sides with severe bilateral narrowing of the nasofrontal duct, and blocked the left infundibulum.
of the orbit. The rare mucoceles of the nasal septum remain asymptomatic until their size causes nasal airway obstruction or hyposmia.

In the literature, only five cases of mucocele of the nasal septum are reported. Three were primary mucoceles, one occurring in a 13-year-old child and the other two in adult patients. The two other cases were likely secondary to previous endonasal surgery. Regarding the localization of these five mucoceles, two were localized at the level of the anterior-superior portion of the septum, one at the level of the median portion, and two at the level of the postero-superior portion of the nasal septum. In our present case, the mucocele was secondary to an Ogston Luc surgery that was performed 23 years earlier. This is the first reported case of a secondary mucocele of the nasal septum arising this long after surgery. Although the lesion presented a very slow growth, over time it became large enough to occupy a great part of both nasal cavities, resulting in almost complete destruction and remodelling of the postero-superior portion of the perpendicular plate of the ethmoid bone and causing non-specific obstructive symptoms.

Differential diagnoses in this case included meningocele, encephalocele, neurofibroma, angiofibroma, lymphoma, inverted papilloma, adenocystic carcinoma, plasmacytoma, dermoid cyst, intraseptal abscess, infections, and nasal foreign bodies. Imaging techniques are an affordable method for diagnosing mucocele of the nasal septum. CT is used to evaluate acquired lesions of the nasal septum, to detect calcifications and assess cartilage, to define the type of pathologic process (destructive vs. expansile bone involvement) and lesion extension (nasal cavities, sinuses, orbits, and/or cranium), and to identify lymph node involvement. MRI is indicated to define lesion margins and intracranial extension, to discriminate between a tumour and postobstructive sinusitis, and also to detect lymph node involvement. In MRI, contrast medium administration is used to assess lesion vascularity, and to identify the intracranial dural and perineural extension of a tumour. MRI findings indicating a mucocele include the observation of a cystic lesion with a variable signal intensity of the content. A low intensity on T1 and a high intensity on T2 typically indicate a liquid-watery content; however, the degree of hydration or presence of blood products will affect MR signal variability, causing a different degree of increase on T1 – as in the presently described case – and a lower intensity on T2 with respect to pure water (Figure 2). Administration of i.v. gadolinium allows exclusion of vascularization of the content – in our case, showing rim enhancement of the wall (Figure 2) – and can reveal the underlying cause if a secondary mucocele is suspected.

This is the first reported case in which a mucocele of the nasal septum was studied by CBCT. CBCT has superior spatial resolution with respect to multidetector CT, producing high-quality subtle bony definition, and thus is an effective alternative to multidetector CT for sinus imaging with the advantage of a lower radiation dose. In our case, CBCT was performed after MRI and allowed a correct definition of the subtle bony anatomy of the nasal cavity and paranasal sinuses as part of the preoperative planning before functional endoscopic sinus surgery. CBCT was useful for determining that the lesion was located in the posterior portion of the septum, involved the perpendicular plate of the ethmoid, and had destroyed the anterior ethmoid cells on the left side but spared the left lamina papyracea. CBCT also demonstrated a severe narrowing of the nasofrontal duct on both sides and blocking of the left infundibulum (Figure 3).

Conclusion

Nasal endoscopy and imaging are essential diagnostic tools and provide important anatomical information for use in preoperative planning. Once the nature of a lesion has been well clarified, endoscopic nasal surgery is the gold standard treatment. It is therefore advisable to proceed with mucocele endoscopic drainage and marsupialization, with an abundant resection of one of the two mucosal sides, in order to avoid recurrence.

References

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