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Case report ■

Frontal Recess Mucocele Associated With Osteoma: Case Report

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SUMMARY

The paranasal sinus mucocele is an epithelized cystic lesion containing fluid and is characterized by non-neoplastic expansion of the paranasal sinus due to its capacity to erode the overlying bone. Etiology of these lesions is still under debate. In this report, we described a case of a patient with frontal recess mucocele, associated with contralateral frontal recess osteoma. A 55-year-old man was admitted to the Department of Otorhinolaryngology of our hospital with the complaints of a headache affecting the fronto-ethmoidal region, left-sided nasal obstruction, and postnasal discharge. Computed tomography (CT) scan of the paranasal sinuses showed a dense bony lesion in the left and a cystic hypodense mass in the right-sided frontal recess. The patient was treated surgically, with a combined endoscopic-external approach, bilaterally. Histopathological examination showed that the wall of the cyst was mucocele, and the bony formation was osteoma. The frontal recess osteoma is regarded as the primary condition. Thus, better ventilation and drainage of the frontal sinus were reestablished.

Key words: mucocele osteoma, frontal recess, endoscopic sinus surgery, external approach surgery

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INTRODUCTION

Paranasal sinus mucoceles are non-neoplastic benign lesions characterized by cystic expansion of the sinuses by retained mucoid secretions (1). The wall of the mucocele is represented by the mucosal, submucosal and osseous components of the affected pneumatized space (1). In a retrospective study of 112 patients with mucoceles, Natvig and Larsen (2) reported that the most frequently involved sinuses are the frontal (77%), frontal and ethmoid combined (16%), ethmoid (7%), and maxillary (3%). The pathogenesis of this condition is still under debate and a number of hypotheses including pressure-induced osteolysis have been proposed. Mucoceles are created by the occlusion of the sinus ostia; continued mucus production accumulates within a sinus with partial or complete lack of drainage, causing secondary dilatation of the sinus (1). In rare cases, mucoceles have extremely rare location: sphenoid sinus, orbitoethmoid (Haller) cells, concha bullosa, and pneumatized uncinat process (3-6).

The frontal recess is defined as the lower portion of the hourglass-shaped space above the level of the ethmoid infundibulum that gives access to the frontal sinus (7). It is, therefore, defined by its surrounding structures, namely, the middle turbinate, lamina papyracea, ethmoid bulla, and agger nasi cells (7). Normally, the frontal sinus opens into the frontal recess through a channel that is less than 3 mm long, called the frontal ostium (7). Obstruction of this recess results in insufficient ventilation of the frontal sinus. When the drainage route for this cavity is blocked, the entrapped oxygen is absorbed by the mucosa and causes negative pressure inside the sinus which produces pain (7). In this report, we describe the case of a patient with the right-sided frontal recess mucocele, caused by contralateral frontal recess osteoma.

CASE REPORT

A 55-year-old man was referred to our ENT department suffering from headaches affecting the fronto-ethmoidal region, left-sided nasal obstruction, and postnasal discharge. He had had the intermittent headaches for about two years. The patient had no previous paranasal sinus surgery or trauma. There were no disorders at ophthalmological and neurological examinations. Rhinological examination showed a left-sided septal deviation, and postnasal mucous discharge. Mucosa of the nasal cavity seemed healthy. Coronal (Figure 1) and sagittal (Figure 2) computed tomography (CT) scans of the paranasal sinuses revealed a dense, well circumscribed osseous lesion that filled completely the left frontal recess from the anterior to posterior wall, measuring 15x10x7 mm.

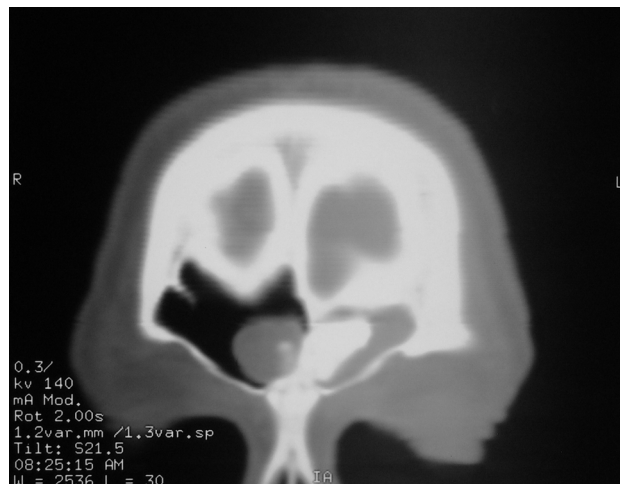


Figure 1. Coronal CT-scan of the paranasal sinuses showing a cystic lesion of the right-sided and bony tumour of the left-sided frontal recess

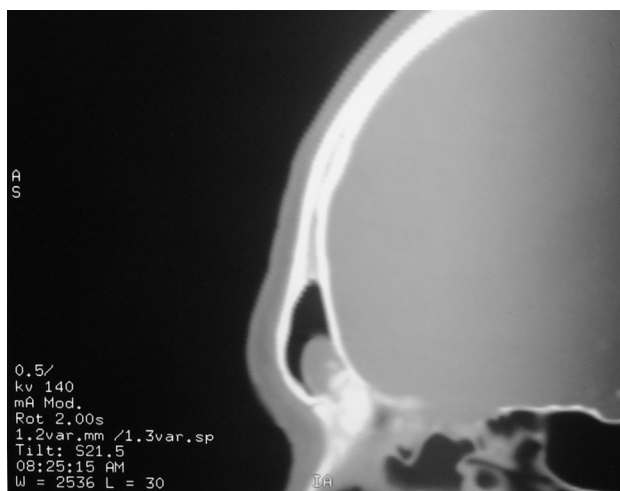


Figure 2. Sagittal CT scan showing the frontal recess tumour and a cyst behind

The inferior portion of the right-sided frontal recess was narrowed, whereas the superior part of this frontal recess was filled by cystic, hypodense, nonenhancing mass with 17x15x10 mm dimension.

The patient underwent combined endoscopic approach and supratrochlear external frontal sinus approach, bilaterally, for excision of these lesions. The surgery was performed under general anesthesia. Nasal mucosal decongestion was bilaterally obtained by applying small cotton gauzes soaked in a solution of cocaine 5% with adrenaline 1:5,000. After middle concha medialization, the middle meatal structures were identified using a 0° endoscope. After uncinat process incision and resection using Blakesley-type forceps, we performed an infundibulotomy, as previously described (8). The ethmoid bulla was opened and completely resected bilaterally using a small forceps. The right frontal recess was filled by cystic lesion with thin bony wall and yellow mucinous fluid, and the left recess by solid osseous

formation. After that, the supratrochlear skin incision was made, bilaterally. After the dissection of the connective tissue and the frontal muscle, the frontal area was fully exposed. A frontal table osteotomy was performed. The frontal osteoplastic flap was based inferiorly in order to expose the right and left frontal sinus cavity (Figure 3).

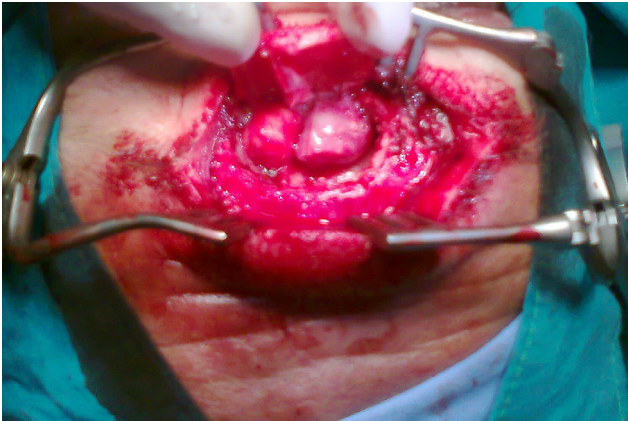


Figure 3. External surgical approach: after frontal sinus trepanation, a soft-tissue mass in the right and bony tumour in the left frontal recess were found

The cystic formation and bony tumour were drilled out using the cavitation technique and by removing the bone boundaries of the lesions. Sinus mucosa was spared whenever possible (Figure 4).

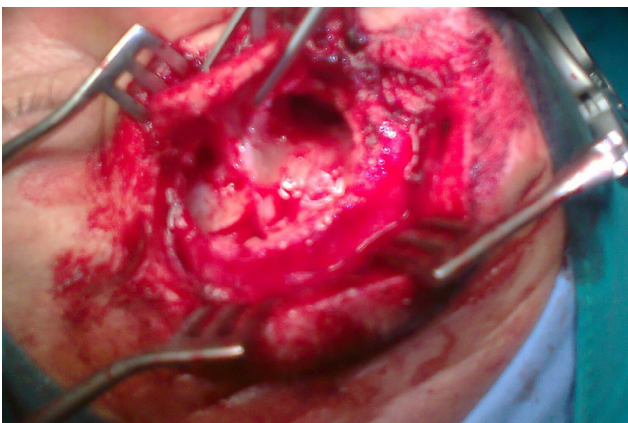


Figure 4. External surgical approach: the status after excision of lesions

No fat obliteration was performed. At the end of the procedure, the bone plate was replaced and anchored. Finally, a septoplasty according to the Cottle procedure was performed. The cystic lesion with yellow mucinous material (Figure 5) and bony tumour were referred to histopathological examination.

Microscopically, the cyst wall fragments showed a lining composed of respiratory epithelium, with variable flattening and attenuation of the pseudostratified columnar epithelium. Furthermore, subepithelial fibrosis with

an eosinophilic, lymphocytic and plasmocytic infiltration was found (Figure 6).



Figure 5. Excised mucocele: a cystic formation with yellow mucous fluid

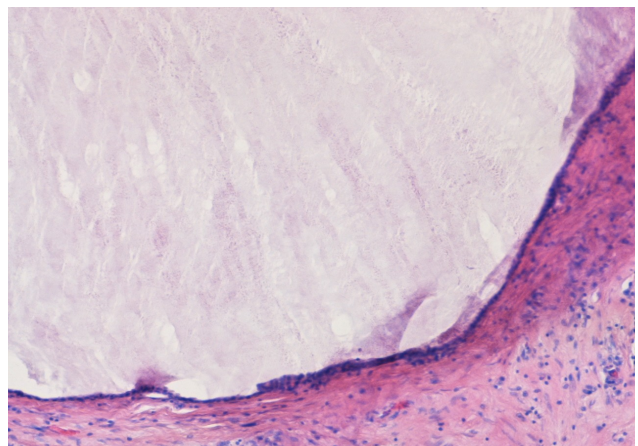


Figure 6. Histopathological finding confirmed the diagnosis of mucocele. A lining composed of pseudostratified respiratory epithelium, with variable flattening and attenuation and subepithelial inflammatory infiltrate. (Haematoxylin & Eosin staining x 100 magnification)

Bacteriological finding of postnasal discharge was positive for *Staphylococcus aureus* culture. However, bacterial mucocele fluid culture was negative. After the surgical treatment, the patient was treated by amoxicillin-clavulanate (in a dose of 1000 mg, two times per day), for seven days, and by saline nasal spray in each nostril for one month. The patient's frontal headache disappeared completely in the first postoperative week. At 12 months post-surgery, the patient remained free of symptoms.

DISCUSSION

Mucoceles and osteomas are isolated lesions in the paranasal sinuses. In case of the association of paranasal sinus mucocele and osteoma, osteoma is considered to cause the mucocele by occlusion of the sinus ostium. Fenton et al. (9) induced mucocele formation in a feline model in experimental conditions. Maturo et al. (10) demonstrated the frontal sinus mucocele formation in a goat model, induced by a long-term frontal re-

cess obstruction by frontalis muscle plugging. However, Hassab and Kennedy (11) performed an ostial obstruction of the rabbit maxillary sinus by histoacryl for 24 weeks. Despite this sustained airtight ostial obstruction, the maxillary sinus showed only an evidence of infection and deranged mucociliary clearance, but no mucocoele formation. The authors concluded that long-term ostial obstruction plays a role in the pathogenesis of chronic sinusitis, but it does not induce mucocoele formation (11). Another theory proposes that mucocoeles are the result of the sinus ostium obstruction and chronic inflammation (12). It has been postulated that the following obstruction of the sinus secondary bacterial infection leads to chronic inflammation mediated by bacterial antigens. Continued stimulation of macrophages, monocytes and lymphocytes by bacterial antigens leads to the synthesis of cytokines, which enhance collagenase and prostaglandin production by osteoclasts, fibroblasts and granulocytes in the mucocoele wall lamina propria (12). Immunohistochemical study conducted by Lund et al. (12) showed that mucocoele mucosa had the high level of immunoreactivity for osteolytic cytokines interleukin-1 alpha (IL-1 α), IL-1 β , tumour necrosis factor-alpha (TNF- α), and IL-6. These factors stimulate the osteoclastic activity and bone resorption and, then, lead to mucocoele expansion (12). On the other hand, IL-1 β and TNF- α are the main pro-inflammatory cytokines of the local Th1 immune response, very important in the first step of antibacterial defence. TNF- α stimulates the cascade of the cytokine production and activation and increases the permeability of the blood vessels, which leads to extravasation of monocytes, granulocytes and macrophages in the place of infection (13).

In our case, mucocoele of the right frontal recess is associated with osteoma of the left frontal recess. The contralateral osteoma represents a mechanical obstruction that forces mucocoele to expand within the frontal sinus cavity. For decades, osteomas of these regions have been treated with open surgical approaches only. External approaches, including lateral rhinotomy and direct anterior surgical exposure of the frontal sinus, have been used to access large osteomas of the fronto-ethmoidal region. However, many forms of paranasal sinus lesions including osteomas may be treated by endoscopic methods, which allowed us to visualize the lesion and surrounding area well during

the procedure. Endoscopic sinus surgery (ESS) is a minimally invasive procedure that offers low morbidity, immediate patient mobilization and good cosmetic results. Patient selection for endoscopic sinus surgery is very important in cases of paranasal sinus osteoma. According to Schick et al. (14), intracranial osteoma extension, antero-posterior diameter of the frontal sinus less than 10 mm, lateral extension behind a virtual plane through the lamina papyracea and erosion of the anterior or posterior wall of the frontal sinus are exclusion criteria for an endoscopic approach. Therefore, endoscopic resection of sphenoid sinus osteomas gives a potential risk of intracranial complications, including a defect in the dura with consequent meningitis, carotid artery rupture, or cavernous sinus damage (14). Endonasal endoscopic resection is possible when osteomas are located medial to the virtual sagittal plane through the lamina papyracea and are attached in the lower portion of the posterior wall of the frontal sinus (15). We performed combined endoscopic and external approach in the surgical treatment. We did not perform the frontal sinus ostium obliteration in order to preserve normal function of the mucosa. The combined external and endoscopic approach was used due to the size of the osteoma and because frontal recess was completely filled by the tumor. ESS is based on the theory that the natural paranasal sinus ostium is the most important area in the pathogenesis of inflammatory disease (16). Based on this opinion, endoscopic surgical opening of the ostium and improved ventilation of the frontal sinus should restore the normal mucosal function. Normal ventilation of the sinus cavity has an anti-inflammatory effect, diminishing the sinus disease symptoms.

CONCLUSION

The association between frontal recess osteoma and contralateral frontal recess mucocoele is a rare finding. In this case, the osteoma is regarded as the primary pathology. We performed a combined endoscopic-external approach in the treatment of our patient for reestablishing the ventilation and drainage of the frontal sinus. Open surgical approach provides better view in cases of lesions, especially if they are situated in narrow pneumatized spaces, such as the frontal recess.

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MUKOKELA FRONTALNOG RECESUSA UDRUŽENA SA OSTEOMOM: PRIKAZ SLUČAJA

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Sažetak

Mukokela paranazalnih sinusa je cistična lezija, obložena epitelom i ispunjena tečnošću, karakterisana netumorskim širenjem šupljine sinusa usled njene sposobnosti da razloži zahvaćenu kost. Uzrok ovih promena je i dalje predmet rasprave. U ovom prikazu smo opisali slučaj 55-godišnjaka, pregledanog u Klinici za otorinolaringologiju naše ustanove, sa glavoboljama fronto-etmoidalnog područja, otežanim disanjem na levu stranu nosa, kao i sekrecijom iz nosa. Kompjuterizovana tomografija (KT) paranazalnih sinusa je pokazala koštanu leziju u levom i cističnu promenu u desnom frontalnom recesusu. Bolesnika smo lečili hirurški, obostrano primenivši kombinovani, endoskopski i spoljni pristup. Histopatološki pregled je pokazao da zid ciste odgovara mukokeli, a da je koštana lezija osteom. Osteom frontalnog recesususa je shvaćen kao primarna lezija. Ovom operacijom smo uspostavili bolju ventilaciju i drenažu frontalnog sinusa.

Ključne reči: mukokela; osteom; frontalni recesus; endoskopska sinusna hirurgija; hirurgija spoljnim pristupom