Frontoethmoidal Sinus Mucoceles Report of Two Cases

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Abstract: Mucoceles are chronic and benign lesions caused by accumulation and retention of mucus secretion in a sinus due to partial or complete occlusion of the ostium which causes progressive distention of the bony wall because of increased pressure. They occur most commonly in the frontal and ethmoidal sinuses.

We present two patients with anterior frontoethmoidal mucoceles who were treated surgically in our clinic. The clinical presentation was pain and deformation of the frontal region and medial canthus in both cases. Chronic sinusitis was detected as a predisposing factor in one case. A transfrontal approach and obliteration of the ostium followed by aspiration of the sinus content was the treatment of choice in both cases. No recurrence was observed in a one-year follow-up.

Although mucoceles are often seen in ENT practice, intracranial and/or orbital extension may be seen in neurosurgery. But recurrence may occur despite surgical intervention. Surgery offers the only effective treatment; the type of procedure selected depends on the location and extent of the mucocele and the nature of any existing complication.

Key words: Ethmoid sinus, frontal sinus, mucocele, paranasal sinus

INTRODUCTION

Occlusion of the sinus ostium due to various pathologies can cause slow and progressive accumulation of secretions. The obstruction may be caused primarily by cystic dilatation of the goblet cell gland besides secondary reasons (2, 7). Congenital pathologies such as agenesis of the ostium (8), tumoral pathologies such as osteoma, fibrous dysplasia, bone lipoma, osteoclastoma, haemangioma, epidermoid, metastatic tumours, craniopharyngioma, cyst formation by embryonic pituitary rests and nasal polyps (3, 4, 15) are primary reasons, while secondary reasons are inflammatory diseases such as asthma, allergies and chronic sinusitis, traumatic causes such as displaced fracture fragments and iatrogenic reasons such as surgery (6, 12). For it is possible mucocele to develop with or without occlusion (8). It has been reported that calcifying fibroma, meningioma and acoustic neurinoma may accompany mucoceles (4, 9). Osteoma may be seen in 12.5 - 50 % of cases during surgery (15). A previous history of sinusitis and rhinitis occurs in 35 - 50 % of cases, trauma in 10 - 28 % and allergies in 11 % (4, 5). Although retention cysts resulting from obstruction and dilatation of tubuloacinar glands are used synonymously with mucocele, they are different in origin and clinical progress. Usually asymptomatic, they do not require treatment (16).

CASE REPORTS

Case 1

A 55-year-old woman was admitted with swelling of the medial side of the left orbit and frontal region, epiphora, frontal and periorbital headache. She had been treated for sinusitis for three years prior to admission. Deformation of the orbital region had developed and progressed in the last eight months. On physical examination, inferomedial displacement of left eye and hyposmia in the right side were observed. The nasolacrimal canal was open. Eye movements, visual acuity and fundus were normal and there was no loss of sensation on the face. Skull x-rays demonstrated sclerotic bone lesions in the frontoethmoidal region, irregularity in the superior and medial orbital wall, lateral displacement of the

lamina papyracea and opacification in the left maxillary sinus (Figure 1). Computed tomography (CT) showed an isodense lesion (55 HU) in the left frontal and ethmoidal sinuses 5x5x4 cm in size causing orbital displacement by destruction of the medial orbital wall. The lesion had extended to the superior nasal cavity and anterior cranial fossa with the erosion of the anterior and posterior walls of the frontal sinus and the superior wall of the ethmoidal sinus via cribriform, perpendicular and ethmoidomaxillary plate destruction and invaded the orbit with the erosion of the lamina papyracea and supramedial wall. The lesion had not infiltrated the anatomical structures. There was a thin fat plane between the medial rectus muscle and the lesion (Figure 2). Biochemical examinations were normal.

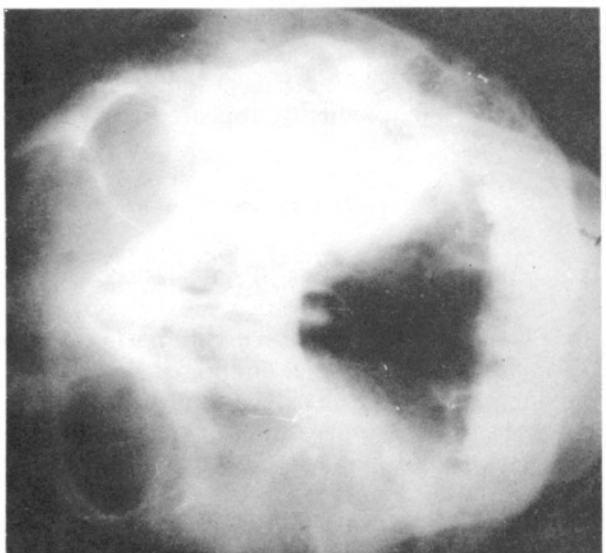

Fig 1: Skull x-ray film showing a lesion in the left frontoethmoidal region occupying and remodelling the sinus and opacification in the left maxillary sinus.

The patient was operated on via a bifrontal incision. Twenty-five ml of green, viscous mucous fluid was aspirated from the cavity. The mucosa was excised and the ostium of the sinus was obliterated by wax. The dura was intact.

In the early postoperative course, the patient's pain disappeared. Pathological examination revealed hyalinized fibrotic wall covered with ciliated pseudostratified cuboidal cells. There were haemorrhages, histiocytes, lymphoplasmocytic cell infiltration, multinuclear giant cells and cholesterol cleft in all layers of the wall. Examination of the mucous material revealed dense nuclear debris and mixed inflammatory cells especially neutrophil leucocytes.

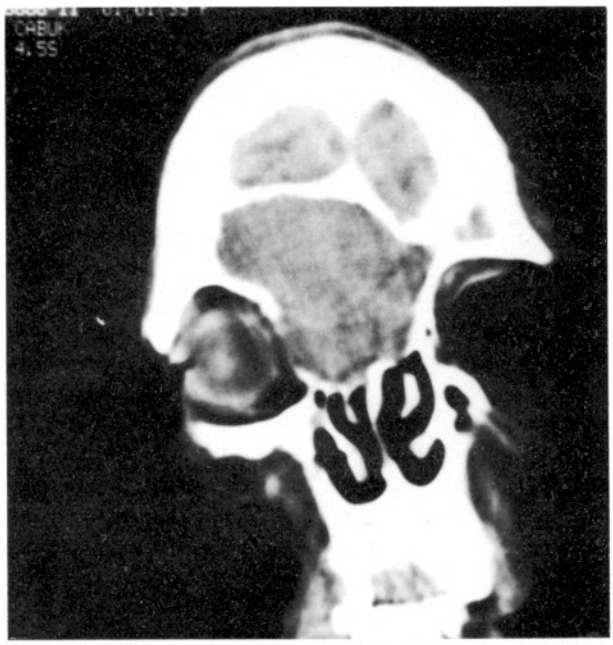

Fig. 2: CT scan showing an isodense mass and lateral displacement of the lamina papyracea and the orbit, erosion of the ethmoidomaxillar plate with extension to the superior nasal cavity in the left frontal and ethmoidal sinus.

Case 2

A 60-year-old man was hospitalized with swelling and pain of the left frontal region that had started five years ago and progressed in the last three months. On physical examination, there were no abnormalities except a fluctuating lesion with cosmetic deformation in the left orbital region. X-rays of the skull showed a marginal sclerosis consistent with an expanding mass of the left frontal sinus. The superior and medial orbital walls were displaced inferiorly and laterally (Figure 3). CT revealed a homogeneous mass lesion (47 HU) 3x4x5 cm in size with slight enhancement in the frontal and anterior ethmoidal sinuses and extending to the orbitofrontal regions (Figure 4). Biochemical tests were normal.

Frontal sinus exploration was performed using a left frontal incision. Brownish, odoriferous, mucous fluid was aspirated that had eroded the anterior, posterior and inferior walls of the sinus. The lesion was removed, and the ostium was obliterated. The dura was intact and pulsatile. Histological examination of the mucous membrane showed pseudostratified ciliary columnar epithelium containing the goblet cells among the condensed mucous fragments. Lymphoplasmocytic cell infiltration, histiocytes, giant cells, hyalinized stroma containing cholesterol cleft were also observed (Figure 5). On microbiological examination, polymorphonuclear leucocytes were seen. Cultures

Fig. 3: Anteroposterior film of the skull showing inferior displacement of the superior and medial walls of the orbit and clear-cut bone destruction (marginal sclerosis) in the left frontal region.

Fig. 4: CT scan reveals a homogeneous mass with destruction of the anterior and posterior walls of the sinus.

of the mucous material were sterile. The case was considered to be a mucocele in the light of these findings. Regarding the slight contrast enhancement, microbiological examination and character of the aspirated fluid, the case was considered to be

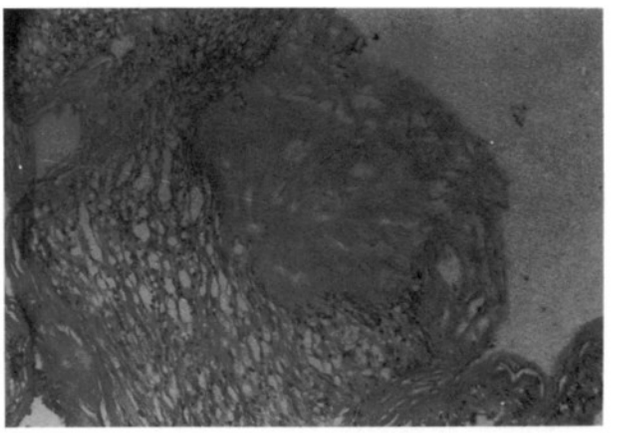

Fig. 5: Photomicrographic appearance of the lesion showing the areas of mononuclear cell infiltration and mucoid lake in the underlying fibrous tissue. (haematoxylin and eosin; original magnification, X 100)

mucopyocele. Following ten days antibiotic therapy, the patient's complaints disappeared and he was discharged.

DISCUSSION

Mucoceles are paranasal sinus lesions usually seen in adults with equal incidence in both sexes (16). Infrequently, they may be seen in children (5, 8). They are common in frontal and ethmoidal sinuses, and rare in sphenoid and maxillary sinuses (5, 11, 18). Atypical localizations like frontal lobe (17), intrasellar and retroorbital regions (1) as well as anterior clinoid processes (4) have also been described. In these cases, the sinus connection has not been shown because of early closure of the path, agenesis of the sinus ostium and ectopic or aberrant sinus without an ostium (5, 8). Three types of ethmoidal mucocele are described as anterior, middle and posterior. Posterior ethmoidal mucoceles are called sphenoethmoidal mucoceles because they originate from the sphenoidal sinus (12, 18). Mucoceles may show extensions to various regions according to their localization. Sphenoid mucoceles may extend to the cavernous sinus, sella turcica, clivus, superior orbital fissure, orbit and ethmoidal sinus. Maxillary mucoceles may narrow the pterygopalatine and nasal fossae, and extend into the ethmoidal sinus and orbit with erosion of the ethmoidomaxillary plate (6, 12). Ethmoidal mucoceles may show extensions into the maxillary sinus and the anterior cranial fossa by destruction of the perpendicular and cribriform plate, into the orbit and optic canal by erosion of the lamina papyracea (9, 11, 18). Frontal mucoceles may extent into the ethmoidal sinus, orbit or anterior cranial

fossa. The medial rectus muscle and globe are displaced anterolaterally or usually inferiorly in frontoethmoidal mucoceles because of erosion of the supramedial and medial wall of the orbit. The nasolacrimal canal and the nasal cavity may be involved. Mucoceles may be attached to the dura if the posterior wall of the frontal sinus is destroyed (9). Delfini eloquently classified paranasal sinus mucoceles according to their localization and intracranial extensions to establish the surgical approach (4). **Type 1:** anterior without extension, **Type 2:** anterior with extension, **Type 3:** posterior without extension, **Type 4:** posterior with extension. The presented cases were evaluated as type 2 mucoceles because of the anterior frontoethmoidal localization and intracranial and orbital extension. Symptoms and signs may show differences according to localization. Mucous and mucopurulant nasal discharge, frontal and periorbital pain radiating to the vertex, temporal and occipital regions, facial deformation, diplopia and loss of smell may be seen. Cliniçal findings are ophthalmoplegia, exophthalmos, epiphora, chemosis, gradual loss of visual acuity, optic atrophy, nasal polyp, spontaneous pneumocephaly, sweating dysfunction with sympatic system involvement. Chemical meningitis may be seen (3, 6, 8, 11, 16). Deformation, pain, globe dislocation and hyposmia were the main symptoms because of the anterior localization of the mucoceles in our cases. Orbital apex syndrome, optic canal syndrome and endocrinopathy may be seen in posterior localization of mucoceles (2, 4, 18). X-rays may provide information about destruction of the inner table of the frontal bone and roof of the orbit, cloudy appearance of the sinus, disappearance and remodelling of the normal sinus walls and marginal sclerosis (8, 16). There may be erosion or displacement in the frontozygomatic arcus, sellar and orbital walls (2, 5, 6). Conventional tomograms and CT have been very helpful in evaluating the character and extent of mucoceles (9). CT has distinct advantages over MRI for assessment of bony erosions and extensions of the lesions as seen in our cases (2, 7). Perugini et al. proposed some criteria for accurate diagnosis of mucoceles (13). a. homogeneous isodense mass occupying the sinus with exophytic alterations, b. clear margins without signs of infiltration of adjacent anatomical structures, c. patchy osteolysis d. no enhancement. Spontaneous hypodense and hyperdense lesions have also been described (4). Differences in the densities of these lesions may be related to the age and consistency of the entrapped secretions (6). Enhancement may be seen if the mucocele is infected (mucopyocele) as in one of our cases (9, 15). There may be ossification and calcification in the osteolytic region. On MR images, the mucocele's contents will have varying signal intensities on T_1 and T_2 weighted images according to the protein concentration and may be misleading (4, 15). The mucosa will enhance at the periphery of the nonenhancing secretions after contrast administration. Angiogram and orbital venogram reveal an avascular mass, displacement of the vascular structures but are not used today (5, 7, 8).

Destruction of the sinus wall forms a communication between the epidural space and the atmosphere which causes intracranial and extracranial complications that include extradural aerocele, CSF fistula, pneumocephalus, orbital cellulitis, meningitis, brain abscess and even sepsis (15, 16, 18).

Pathological examination shows PAS (+), pseudostratified ciliary columnar epithelium containing the goblet cells or small cubic epithelium covering the mucoperiostal wall. It may also show chronic nonspecific inflammatory tissue revealing squamous metaplasia or fibrous tissue consisting of mononuclear cell infiltration (4, 10, 12, 17). Similar findings were observed in our cases. Culture results may be positive, when the mucocele is infected. No organisms were isolated from the mucous content of our second case with mucopyocele.

Management of a mucocele that has extended into the anterior cranial fossa and / or orbit is surgery. The main purpose of surgical treatment is to evacuate the lesion, radically remove the sinus mucosa to prevent relapse and re-establishment of a plane of separation between the extracranial and intracranial space as well as reconstructive surgery to achieve satisfactory cosmetic results. Frontotemporal, frontal, bifrontal craniotomies; transnasal, transethmoidal, transpalatal, transorbital or combined approaches may be used according to localization of the lesion.

Sphenoidotomy, transfrontal sinusotomy and ethmoidectomy may also be used for simple drainage (4, 7, 11, 18). Basically, transcranial or maxillofacial approaches should be used for mucoceles with or without intracranial extensions respectively. Maxillofacial approaches were usually performed in anterior mucoceles (Type 1) and posterior midline mucoceles without intracranial extension (Type 3) in ENT practice (4, 10, 14). Some maxillofacial surgeons prefer two - stage operations, draining the sinus content first followed by removal of the sinus mucosa (5, 12, 15). Removal of the anterior wall of the frontal sinus is generally performed only for osteomyelitis. Cranialization of frontal sinus involves removal of

the posterior table and all sinus contents. The posterior wall of the sinus should be restored with a patch of galea graft because of the risk of infection, rhinorrhea and pneumocephaly. The sinus ostium should be occluded by melted bone wax and muscle grafts to prevent recurrence (4, 8, 15). Haemorrhagic nasal discharge and 4-10 % recurrence may be seen following the operation (2 ,4, 14). In our cases, bifrontal and frontal approaches were used due to frontoethmoidal localization with intracranial extension (Type 2). After excision of the mucocele, the mucosa was completely removed and the ostium was obliterated to prevent recurrence of the mucocele. The anterior and posterior walls of the sinus were left intact because of the absence of osteomyelitis. There was no recurrence in one year follow-up.

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