Bilateral asymmetrical mucoceles of the paranasal sinuses with unilateral orbital complications

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Abstract
We report the rare occurrence of bilateral asymmetrical mucoceles of the paranasal sinuses that resulted in a unilateral orbital complication. The patient was a 47-year-old woman who presented with complaints of diplopia, blurred vision, and protrusion of her right eye that had progressed over a period of several months following an upper respiratory tract infection. Computed tomography detected the presence of two large, asymmetrical mucoceles. The lesion on the right involved the frontal and ethmoid sinuses, and the one on the left involved the ethmoid sinus. The mucoceles were locally expansile and had eroded the surrounding bony structures on the right. The expansile nature of the right-sided mass had displaced the right orbit, which was the cause of the vision deterioration. Transnasal endoscopic surgery was performed to excise and marsupialize the mucoceles. This modality was preferred over conventional open surgery because it affords good visualization, it is safe, and it is a less morbid procedure. The patient's recovery was uneventful, and she was discharged home on the third postoperative day. On continuing follow-up, her vision had improved, her intraocular pressure had returned to normal, and her orbits were in their normal position. Based on our literature search, no case of bilateral frontal and ethmoid sinus mucoceles has been previously reported.

Introduction
Mucoceles of the paranasal sinuses are slowly growing, locally expansive epithelial lesions with osteolytic features that can involve surrounding structures such as the orbit and intracranial cavity. Patients with paranasal sinus mucoceles often present to the ophthalmology clinic for evaluation of eye complications such as diplopia, decreased visual acuity, and orbital globe displacement. Other signs and symptoms can include facial pain and/or swelling, headache, nasal obstruction, cerebral abscesses, pneumoencephaloceles, and even meningitis, depending on the specific area involved.1-3

Most paranasal sinus mucoceles involve the frontal or ethmoid sinus. The diagnosis is established by radiographic imaging; the gold standard in terms of diagnostic precision is computed tomography (CT),4 although magnetic resonance imaging (MRI) is helpful in some cases. The preferred method of treatment is transnasal endoscopic surgery because it is associated with minimal trauma and morbidity.

Bilateral paranasal sinus mucoceles are rare. When they have been reported, almost all were symmetrical and nearly all involved the frontal sinuses.5,6 We present a case of bilateral asymmetrical paranasal sinus mucoceles that involved the ethmoid sinus on the left and the frontal and ethmoid sinuses on the right. To the best of our knowledge, this is the first report in the English-language literature of bilateral asymmetrical mucoceles involving the frontal and ethmoid sinuses on one side and the ethmoid sinus on the other side.

Case report
A 47-year-old woman presented with complaints of diplopia and blurred vision that had progressed over a period of several months. Her symptoms were associated with protrusion of her right eye. About 2 or 3...
months prior to the onset of her eye complaints, she had experienced an upper respiratory tract infection. She denied headaches, facial pain, nasal obstruction, rhinorrhea, and any other nasal symptoms commonly associated with diseases of the paranasal sinuses.

On examination, a non-pulsatile proptosis of the right eye was noted (Hertel values: 22 mm for the right eye and 17 mm for the left eye), and the right globe was displaced inferolaterally. Ocular movements and visual field were restricted in the right eye, especially on elevation. The best-corrected visual acuity was 6/36 on the right and 6/9 on the left. The patient had a grade I relative afferent pupillary defect on the right. Her intraocular pressure was elevated in the right eye (26 mm Hg) and normal in the left (17 mm Hg). The optic cup/disk ratios were 0.5 and 0.3, respectively; both optic disks were pink.

Endoscopic examination of the nasal cavities revealed only a prominent right ethmoid bulla; no significant finding of note was seen in the left nasal cavity. However, CT revealed the presence of two large, bilateral, asymmetrical paranasal sinus mucoceles. The mucocele on the right involved the frontal and ethmoid sinuses, and it had compressed the right orbit and displaced it inferolaterally (figure). Bone was eroded in the postero-superior aspect of the right frontal sinus. The left-sided mucocele occupied the ethmoid sinus. No intracranial extension was noted on either side.

Endoscopic marsupialization of the two mucoceles was performed. Intraoperatively, the mucocele on the right was noted to have herniated into the right ethmoid bulla. Following uncinctomy and removal of the bulla, the mucocele was identified. It was thick-walled, presumably as a result of chronic infection and longstanding fibrosis. Opening of the mucocele released a copious amount of viscous, purulent mucus, which was drained and sent for culture. The culture was negative. The incised opening was then marsupialized. Examination of the mucocele cavity revealed that the mucocele had eroded the lamina papyracea. The entire orbital contents were ballotable; the medial aspect of the right orbit was endoscopically observed to be ballotable with externally applied orbital pressure. Further examination revealed that both the frontal and ethmoid sinuses on the right had been eroded to the point where they formed a single cavity. At the postero-superior aspect of the frontal sinus, the skull base was eroded, and the dura was seen to pulsate through the thinned-out bony remnant. No cerebrospinal fluid leak was observed.

A similar procedure was performed on the left side. The uncinctomy revealed the bulging mucocele in the ethmoid bulla, which was marsupialized. Findings on examination of the mucocele cavity on the left were similar to those on the right except for a lack of bony erosion.

The patient’s recovery was uneventful, and she was discharged home on postoperative day 3. At review 6 weeks later, her vision had improved to 6/9 bilaterally, her intraocular pressure had returned to normal, and her orbits were in their normal position. No change from the status quo was observed during continuing follow-up.
Discussion

The etiology of paranasal sinus mucoceles, while not fully defined, is often associated with ostial obstruction of the affected sinus. This obstruction can occur as a result of chronic processes such as rhinosinusitis (infectious or allergic), sinonasal polyposis, craniofacial trauma, previous surgery, benign tumors (e.g., osteoma or fibrous dysplasia of bone), or malignant neoplasms (primary or metastatic). In some cases, there is histologic evidence of an increase in the number of secretory cells in the lining membrane; hypersecretion of mucus may be a contributing factor. In our patient, however, the development of the mucoceles appears to have occurred secondary to an episode of upper respiratory tract infection that she had experienced several months prior to presentation.

Most paranasal sinus mucoceles arise in the frontal sinus (60% of cases) or the ethmoid sinus (30%); fewer than 10% are localized in the maxillary sinus, and sphenoid sinus involvement is rare. Their prevalence peaks in the third and fourth decades of life, and they have no predisposition for either sex.

Fewer than 10 cases of bilateral paranasal sinus mucoceles have been reported in the English-language literature. Varghese et al reported the first known case of bilateral asymmetrical mucoceles in 2004; those lesions involved the maxillary and ethmoid sinuses on opposite sides. To the best of our knowledge, ours is the first report of bilateral asymmetrical mucoceles involving the frontal and ethmoid sinuses on one side and the ethmoid sinuses on the other.

Mucoceles tend to expand, remodel, and readsorb the bony walls of the affected paranasal sinus, changing their integrity and occasionally affecting neighboring structures such as the orbit and intracranial cavity. The pathophysiology of this phenomenon is believed to involve osteolysis caused by a reduction in the vascularization of bone secondary to compression and/or the action of inflammatory mediators that are abundantly present in the mucus, such as cytokines (interleukins 1 and 6), vascular adhesion molecules, and prostaglandins.

While opacification, bone erosion, and expansion of a mucocele may be seen on simple x-ray, CT is the preferred mode of radiographic examination because it illustrates clearly the bone involvement and the presence of orbital and/or intracranial extensions, and it is therefore very useful in planning surgery. MRI is best reserved for patients whose mucocele formation occurs secondary to a sinonasal tumor because in these patients, only the lining membrane of the mucocele will be enhanced on intravenous contrast. A tissue biopsy is usually not performed because the diagnosis of mucocele is quite apparent on both clinical and radiographic grounds. A biopsy can be carried out if any suspicious lesions are noted during surgery.

Treatment of mucoceles is surgical. At one time, access was generally achieved via the external route (frontoethmoidectomy). However, demonstrations of the safety of the transnasal endoscopic approach has made it today's treatment modality of choice. Transnasal endoscopy is minimally invasive, it preserves the epithelium, and it is associated with low morbidity when used for the marsupialization and drainage of mucoceles.

References