Paranasal Sinus Mucoceles and their Ocular Impacts: A case report

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Abstract

INTRODUCTION

Paranasal sinus mucoceles represent a build-up of mucous-containing, epithelium-lined, cystic lesions within air-filled cavities in the bones surrounding the nasal cavity. Sinus cavities may accumulate excessive mucous when the normal drainage pathways are blocked. Paranasal sinus mucoceles may exert pressure on adjacent structures, including the orbit, potentially leading to proptosis, diplopia, or even vision loss. Compression of the optic nerve or globe can cause optic neuropathy, underscoring the importance of early diagnosis and intervention to preserve ocular health.

CASE REPORT

A 56-year-old Hispanic male complained of proptosis, periorbital edema, and pain in the right eye, accompanied by cluster headaches that had persisted for the previous six months. Initial examination demonstrated an uncorrected visual acuity of 20/20 in both eyes at distance and near. Clinical assessments further revealed Hertel exophthalmometry measurements of 21 mm in the right eye and 19 mm in the left (base of 108 mm), along with resistance to manual pressure in the affected right eye. Subsequent computed tomography imaging of the head and orbits identified a sizable, well-defined mass originating in the right nasal cavity, extending into multiple paranasal sinuses, and inducing structural alterations in the medial wall of the maxillary sinus, consistent with paranasal sinus mucoceles.

CONCLUSION

Proptosis and lid edema can arise from various etiologies, including intracranial masses, autoimmune disorders, or impaired venous return. This case report adds another consideration of a diagnosis in a patient with a history of sinus surgery. A comprehensive history and meticulous clinical examination are paramount for discerning pathological manifestations in establishing an accurate diagnosis.

KEYWORDS

Paranasal sinus mucocele, proptosis, periorbital swelling, headache

INTRODUCTION

Periorbital edema and proptosis are typically associated with space-occupying lesions, impaired venous return, autoimmune-related, or inflammatory conditions.[1,2] This case report highlights a patient presenting with proptosis and periorbital edema that was found to be due to a paranasal sinus mucocele. Paranasal sinus mucoceles are cystic lesions filled with mucus and lined with epithelium, which typically occur due to sinus obstruction.[3] Primary causes of mucocele development include ostial blockage, poly degeneration, or cystic dilation of mucosal glands, while secondary causes often result from prior sinus surgery or trauma.[4] The frontal sinus is the most frequent site for mucocele development (60-89%), followed by the ethmoid (8-30%) and maxillary (<5%) sinuses, with a male predominance.[3,4] Common symptoms include headaches, periorbital pain/pressure, proptosis, periorbital swelling, and diplopia.[5]

CASE REPORT

A 56-year-old Hispanic male presented to the clinic in 2024 with a complaint of pain, proptosis, and a swollen right eyelid that had persisted for the previous six months. He reported experiencing frontal cluster headaches 3 - 4 times a week in association with these symptoms. The patient had a history of Laser-Assisted in Situ Keratomileusis and a previous diagnosis of allergic conjunctivitis. His medical history included functional endoscopic sinus surgery in January 2016. No other medical conditions or medications were reported.

Unaided Snellen visual acuity at distance and near were 20/20 in each eye. A cover test revealed orthophoria at distance and 6-prism diopter exophoria at near. Eye movements were concomitant in different gazes and head tilts with no observed diplopia. Ocular motilities tested by Broad H were unrestricted, but the patient reported pain in the right eye upon right gaze. Confrontation field was full to finger count in both the right and left eyes. Pupils were equal, round, and reactive to light without any relative afferent pupillary defect. Intraocular pressures (iCare @ 11:34 AM) were 14 mmHg and 12 mmHg in the right and left eyes, respectively.

Anterior segment examination revealed scattered diffuse superficial punctate keratopathy in the right eye more than the left from incomplete eye closure and mild corkscrew vessels on the temporal bulbar conjunctiva on both eyes. Slit lamp examination revealed no cells and flare within the anterior chamber. Examination of the inferior palpebral conjunctiva revealed grade 1 papillae, grade 1 staining of the bulbar conjunctiva, with trace conjunctivochalasis in both eyes. There was a mild resistance to manual pressure along with periorbital edema in the right eye. Hertel exophthalmometry measured 21 mm OD and 19 mm OS with a base of 108 mm. Contrast sensitivity testing (Pelli-Robson at 1 meter; Lighthouse International) revealed 1.65 in the right, left, and both eyes (meeting age normal monocularly and reduced binocularly).[6] The results of a red cap test and colour vision testing were unremarkable (100% monocularly). A Humphrey Visual Field Analyzer (Zeiss-Humphrey, Liandro, CA; 30-2 SITA Fast) revealed no field defects.

Posterior segment examination testing revealed suspicious blurring of the optic disc border in both eyes. This was noted superior nasal of the right eye and nasal of the left eye. Further examination of the optic nerve head revealed a dilated central retinal vein of the right eye compared to the left, without pallor in either eye. The macular region was evenly pigmented with no abnormalities. Optical coherence tomography was performed with a Heidelberg Spectralis (Heidelberg Engineering, Heidelberg, Germany) and revealed healthy macular regions and pink nerves. Structural parameters of the optic nerve were analyzed using the Nsite axonal application from Heidelberg Spectralis (Nsite Axonal Analytics), which revealed greater nasal thickening in the right eye than in the left.

The patient was assessed to have periorbital edema and proptosis in the right eye with a query of optic nerve elevation in both eyes with an enlarged central retinal vein in the right eye.

The following differential diagnoses were considered:

Conditions that encompass both proptosis and periorbital edema can be broken down into four general categories: autoimmune-related (thyroid eye disease), inflammatory (orbital cellulitis, sarcoidosis, idiopathic orbital inflammation, or granulomatosis with polyangiitis), impaired venous return (carotid cavernous fistula or cavernous sinus thrombosis), and space-occupying lesions (mucoceles or orbital tumor).[1,2] Autoimmune disorders can include thyroid eye disease, orbital cellulitis, sarcoidosis, idiopathic orbital inflammation, and granulomatosis polyangiitis.

1. Graves' disease (thyroid-associated orbitopathy) is an autoimmune disease that is characterized by thyroid dysfunction.[7] It is mostly associated with hyperthyroidism; however, it can be noted in hypothyroid (4%) and euthyroid (6%).[7] The most common signs and symptoms for this condition are upper eyelid traction (91%), proptosis (62%), periorbital edema (32%), restriction of eye muscles (42%) and conjunctival hyperemia (34%).[7] This diagnosis requires blood tests including thyroxine (T4), triiodothyronine (T3), thyroid-stimulating hormone (TSH), and thyroid-stimulating immunoglobulin (TSI).[7] Occasionally, euthyroid patients require computed tomography or magnetic resonance imaging to rule out the diagnosis.[7]

2. Orbital cellulitis is an infection of the soft tissues within the orbit, posterior to the orbital septum.[8,9] This is typically due to a sinus infection, but can also be caused by dental fracture, orbital fracture, or orbital infection (dacryoadenitis).[8,9] Common signs and symptoms include decreased visual acuity, restricted ocular motility, fever, malaise, proptosis, headaches, significant periorbital edema, chemosis, and hyperemia.[8] Orbital cellulitis is an ocular emergency and must be treated with intravenous and oral antibiotics.[8,9]

3. Idiopathic orbital inflammation is a benign inflammatory condition without an identifiable cause.[10] This condition can be categorized into anterior, posterior, diffuse, or apical depending on the location of inflammation.[10] Diffuse idiopathic orbital inflammation can cause proptosis, along with pain and periorbital swelling with limited ocular motility.[10] A diagnosis of idiopathic orbital inflammation is a diagnosis of exclusion, where other etiologies have been ruled out.

4. Granulomatosis with polyangiitis is a rare vasculitis affecting small-medium vessels in the sinus, throat, respiratory tracts, and kidneys.[11,12] Ocular signs and symptoms typically include scleritis, peripheral ulcerative keratitis, uveitis (non-granulomatous anterior), diplopia, proptosis, and potential vision loss.[12] Blood tests, including anti-neutrophilic cytoplasmic antibodies (ANCA), renal function, urinalysis, erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), and further imaging (radiological and computed tomography of the lungs) are required for diagnosis.[11]

5. A carotid cavernous fistula is due to an abnormal shunt from the internal carotid artery to the cavernous sinus, where it affects cranial nerves III, IV, V1, V2, and VI.[13] Typical signs include pulsating proptosis, cranial nerve palsy, corkscrew blood vessels, and conjunctival chemosis.[13] Diagnosis requires neuroimaging (including but not limited to computed tomography and magnetic resonance imaging) of the head and orbits.[13]

6. Orbital tumors or masses can arise from various etiologies and present with a wide range of signs and symptoms. The orbit houses critical structures and tissues within a confined space, making it susceptible to visually significant symptoms and irreversible vision loss when abnormal masses are present. Common presenting signs include proptosis, extraocular muscle limitations, exposure keratopathy, optic neuropathy, and more.[14] Symptoms may involve vision loss, diplopia, pulsatile proptosis, headaches, and irritation.[14] Due to the low incidence of orbital masses, diagnostic imaging, such as computed tomography or magnetic resonance imaging, is often required for an accurate diagnosis.

To further differentiate the etiology underlying the patient's ocular condition, a blood sample for thyroid testing was ordered through the family doctor and computed tomography of the head and orbits was ordered through general ophthalmology.

The results for thyroid stimulating hormone and free thyroxine were within the normal ranges. The computed tomography (with contrast) report from the radiologist revealed a large circumscribed expansile mass, extending from the right nasal cavity into multiple paranasal sinuses. The mass caused remodeling along the medial wall of the right maxillary sinus and right orbit, resulting in a mass effect on the right orbital structures. Notably, the mass has significantly increased in size since the CT in January 2016. A final diagnosis of paranasal sinus mucocele was made, which led to a referral to Otolaryngology for management.

A report back from the ophthalmologist noted successful functional endoscopic sinus surgery where the mucocele was removed without intracranial or intraorbital complications by the otolaryngologist. The three-month post-surgical examination results revealed unaided visual acuities of 20/20 in each eye, as well as Hertel Exophthalmometry measurements of 20 mm and 20 mm (base of 110mm) in each eye, respectively. Note the minor differences in base measurements (from 108 mm to 110 mm). Intraocular pressures of 11 mmHg and 10 mmHg were measured in the eyes. Posterior segment examination revealed a normal central retinal vein caliber in both eyes. The patient was relieved from headaches and facial asymmetry due to proptosis was resolved. He was discharged from care. Continuous care consisting of annual eye examinations has been booked in the Optometry clinic.

DISCUSSION

Proptosis associated with periorbital edema can be attributed to different etiologies, including but not limited to the conditions described above.[1,2] This case report describes the case of a 56-year-old man who had proptosis associated with periorbital edema that was determined to be from a space-occupying lesion. a paranasal sinus mucocele.

Paranasal sinus mucoceles arise from the blockage of sinus ostia, leading to the formation of cysts lined with epithelium and filled with mucus.[3] The exact incidence of paranasal sinus mucoceles is unknown. Devars du Mayne et al. (2012) noted 68 retrospective cases over a span of 16 years (1993-2009) in France, and Scangas et al. (2013) reported 133 cases over a span of 24 years (1987-2011) in Pennsylvania.[5,15] The exact pathophysiology and etiology of paranasal sinus mucoceles remain uncertain. One theory suggests that mucoceles result from the cystic degeneration of sero-

mucinous glands, leading to the formation of retention cysts.[3,16] Primary mucoceles occur in individuals without a history of sinus surgery or identifiable predisposing factors, while secondary mucoceles develop as complications of procedures such as functional endoscopic sinus surgery, the Caldwell-Luc operation, or traumatic events.[4,16,17] Paranasal sinus mucoceles can manifest at any age and in both genders, though they are most commonly observed in individuals aged 40 to 60 years, with a higher incidence among males.[3,16] Recurrence rates of paranasal sinus mucocele range from 10 to 16 % and it can occur from 4 months to 16 years after surgery (averaging around 4 years).[5,15,18,19] A de novo etiology is seen in 25.6% and secondary causes (including trauma) account for around 8.3% of the cases.[5,15,18,19]

The accumulation of mucus within the mucocele promotes the extension of sinus bone walls and enlargement of the mass.[16] Prostaglandins, along with pro-inflammatory cytokines such as interleukin 1 (IL-1) and tumor necrosis factor-alpha (TNF-α), as well as collagenases, contribute to osteolysis, facilitating expansion of the cyst.[16] Lund et al. observed that IL-1 may play a crucial role in the erosion of bone overlaying the expanding mucocele.[20] The expansile nature of the mucocele can lead to intracranial and intraorbital extensions. Studies, such as that by Scangas et al., have reported that approximately 44.9% of mucoceles extend into intraorbital or intracranial spaces, leading to ophthalmologic symptoms due to the compression of adjacent orbital structures.[5,16] Clinical presentations vary and may be rhinological, neurological, or ophthalmological depending on the mucocele's location.[3] Signs and symptoms may include periorbital edema (9.8%) and pain (12.0%), headaches (42.1%), facial pressure (28.6%), toothache, nasal obstruction (26.3%), proptosis (11.3%), reduced extraocular motility, and diplopia (6.0%).[3,5] Interestingly, 8.3% of the patients with paranasal sinus mucoceles are asymptomatic.[5]

Hertel Exophthalmometry is used to provide a quantitative clinical measurement of the anterior position of the globe with respect to the orbital rim, where the distance from the lateral orbital rim to the surface of the cornea is measured. Population studies have shown that the results of Exophthalmometry can vary between 12 and 24 mm depending on race, gender, and age. For example, subjects of Asian descent show measurements between 15 and18mm, Caucasians range between 15 and 21 mm, Hispanics range from 15 to 20 mm, and African Americans could range between 17 and 24 mm. Despite the large range, the two eyes should not differ by more than 2 mm.

Mucoceles have the potential to entrap the optic nerve, leading to blindness. In cases of inflammation (mucopyocele/pyocele), direct compression of the optic nerve can result in reduced vision.[21] Additionally, mucoceles can exert pressure on the lamina papyracea, impinging upon the optic canal and causing visual disturbances.[3,21] Mucopyocele/pyoceles can cause direct compression, ischemia, or ocular neuritis, which results in optic neuropathy.[3,21] The patient in the present case had an asymmetric intraocular pressure reading; although both readings were still within normal limits, the pressure in the right eye was slightly elevated compared to that in the left. In association with the enlarged central retinal vein of the right eye, the asymmetric intraocular pressures could be secondary to the expansion of the mucocele causing a mass effect and compression of the adjacent structures.[22] Prior research has indicated that frontal and anterior ethmoid mucoceles can cause dissimilar intraocular pressures.[22] In contrast, posterior ethmoid and sphenoid mucoceles are closer to the cranial nerves and more frequently linked to visual disturbance and orbital apex syndrome.[22]

Diagnosis typically involves a combination of physical examination and radiological assessments, including computed tomography scans and magnetic resonance imaging.[3,23] Treatment primarily revolves around surgical excision, often with early intervention to prevent visual impairment. Surgical options include external approaches such as Lynch-Howarth incision, Caldwell-Luc surgery (osteoplastic flap), or endoscopic marsupialization.[3,5,16] The choice of surgical method depends on factors such as the lesion's location, size, and extent of expansion.[3] Endoscopic techniques are favored by most surgeons due to superior cosmetic outcomes, reduced morbidity, lower recurrence rates (<10%), and less disruption of the nasal anatomy and physiology.[3,4,16,24,25] However, Caldwell-Luc surgery may be considered in cases of concomitant pathological mucosal disease.[3] More conservative management options may involve intravenous antibiotics, non-steroidal anti-inflammatory drugs, and nasal decongestants to address associated infectious conditions, although these may not fully resolve mucoceles or orbital complications arising from mucopyocele/pyocele.[3,26]

This case report described a 56-year-old Hispanic male who presented with symptoms of pain, proptosis, and headaches for the previous six months. After completing a computed tomography scan, he was diagnosed with paranasal sinus mucocele that was impeding his right orbital structures (including the optic nerve), causing proptosis without any visual complications. Mucoceles are benign, slow-growing lesions in the sinus that can cause visual complications if not detected promptly. They have the potential to enlarge and lead to bone remodeling, which can impact nearby tissues, including the orbit. The mainstay treatment is functional endoscopic sinus surgery which preserves the anatomy of the structures and decreases the rates of morbidity and recurrence. Patients with a history of paranasal sinus mucocele should be fol-

lowed-up continuously after the procedure to prevent any recurrence and changes to the quality of life. This report highlights a potential complication from sinus surgery that could lead to orbital compression, proptosis and eyelid edema. ●

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