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Case Report

Giant mucocele secondary to cocaine abuse

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ABSTRACT

Mucoceles are benign lesions originating in the paranasal sinuses. Presented here is a novel case of a giant frontoethmoidal mucocele extending into the orbit and causing proptosis. The etiology was chronic transnasal cocaine abuse leading to extensive scarring and distortion of the nasal anatomy.

Introduction

Mucoceles are epithelial lined mucus-containing benign lesions which originate in the paranasal sinuses. They arise as a sequela of obstruction of the sinus drainage pathways due to chronic inflammation, trauma, repeated infections or prior surgery [1]. In this report, we describe a unique case of a giant frontoethmoidal mucocele with destructive changes of the nasal septum secondary to chronic cocaine abuse. Incidence of cocaine-induced midline destructive disease is 4.8% among cocaine abusers [2]. Commonly described manifestations of intranasal cocaine abuse include epistaxis, crust formation, nasal septal perforation, saddle nose or alar deformities, hard palate destruction in advanced diseases, and erosion of the internal nasal and sinus structures [3–5].

Case report

A 50-year-old woman presented to the emergency room with proptosis and restricted movements of her right eye. She had history of intermittent eye pain, nasal obstruction and frontal headache since one and a half years. She was unable to blow her nose, as it put pressure on her eyeball. There was no prior history of sinus surgery or history of tumor.

Patient was a past cocaine abuser for many years. Physical examination was significant for a marked inferolateral proptosis of the right eye. Fundic examination was unremarkable. No afferent pupillary defect was noted on ophthalmic examination. An anterior nasal septal perforation was present. The nasal cavity was obstructed by a large soft tissue mass. No cervical lymphadenopathy was present. Ears and oral cavity were unremarkable. Blood workup was normal.

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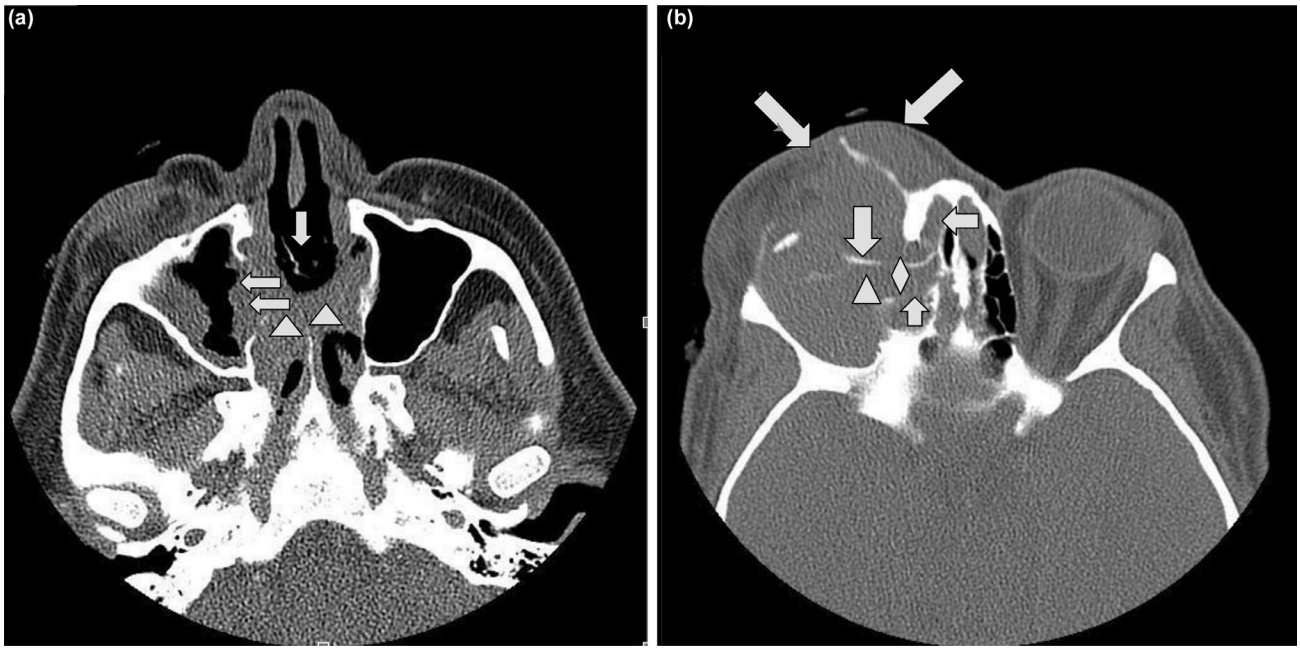

Fig. 1 – (A) Axial nonenhanced CT scan at the level of the maxillary sinuses reveals irregular soft tissue in the nasal cavity which crosses the midline with destruction of the nasal septum (triangles) and associated anterior septal defect (downward arrow). Irregular soft tissue extends into the right maxillary sinus through a destroyed medial wall of the maxillary sinus (leftward pointing arrows). (B) A more superior scan demonstrates a soft tissue mass in the anterior nasal cavity (leftward arrow) and right anterior ethmoid region with destruction of the ethmoid septae (upward arrow) and destruction of the lamina papyracea (diamond shape). Expansile mass extends into the orbit (triangle) and contains thin bony septations (downward arrow). In addition, it protrudes into the subcutaneous tissues anterior to the orbit (oblique arrows).

CT imaging of the brain and sinuses was obtained and showed nasal septal perforation (Fig. 1A). A large soft tissue mass occupied the nasal cavity and right ethmoid air cells with extension to the orbit (Fig. 1B). Bony destruction of the medial orbital wall was seen (Fig. 1B). The expanding mass in the orbit measured 5.6 × 4.6 × 4.3 cm and demonstrated thin bony septations (Fig. 1B). Contrast enhanced MRI of the brain and orbits demonstrated rim enhancement of the sinonasal soft tissues consistent with inflammatory changes (Fig. 2C). Centrally in the nasal cavity, a persistently hypointense focus was seen which indicated presence of fibrotic/scar tissue ruling out the possibility of a granuloma or tumor. No intranasal mass or tumor was seen. Proteinaceous fluid was present in the maxillary sinus (Fig. 2A-C). The orbital mass demonstrated variable low signal with mild peripheral enhancement consistent with a mucocele (Fig. 2C). Bony septations were seen within this orbital mucocele (Fig. 1B).

The patient underwent right orbitotomy where a whitish mass was found. Biopsy of the lining of this orbital mucocele revealed fibrocollagenous tissue, with no evidence of tumor. Subsequently the patient had a resection of the frontoethmoidal mucocele via a bicoronal flap approach. Operative findings confirmed thickening and remodeling of several septations of the frontal and ethmoid sinuses due to the pressure exerted by the large mucocele. Aspirate from the mucocele revealed mucinous material with cellular debri, ciliated columnar cells and foamy macrophages. Histopathology of the cyst wall showed mucosal tissue with chronic infection and a focal histiocytic aggregate consistent with mucocele. Nasal endoscopy revealed extensive scarring with distortion of the nasal anatomy. A large fibrotic scar was seen extending from the septum to the lateral nasal wall. A large anterior and posterior intranasal perforation involving the bony and cartilaginous part of the nasal septum was found. Since there was obstruction to the ostia and outflow tracts of the ethmoid, maxillary and frontal sinuses, bilateral ethmoidectomy, right frontal sinusectomy, and endoscopic maxillary antrostomy were done.

Biopsy of the nasal scar tissue revealed fragments of fibrocollagenous tissue, focally vascularized fibroblastic tissue with organizing fibrin consistent with a reparative process, with no presence of granulomas or tumor.

Discussion

Cocaine is a stimulant alkaloid that occurs within the leaves of the shrub Erythroxylon coca [5]. The leaves can be chewed directly or the drug can be prepared in more concentrated forms. On the street, it is often diluted with adulterants like talc, procaine, sugar, amphetamine, or lidocaine. This mixture can then be snorted or applied to mucous membranes where it is absorbed into the blood stream [5,6]. Its pharmacological effects are manifold.

Chronic intranasal administration of cocaine causes irritation and atrophy of the nasal mucosa. In addition cocaine induced intense vasoconstriction of the small blood

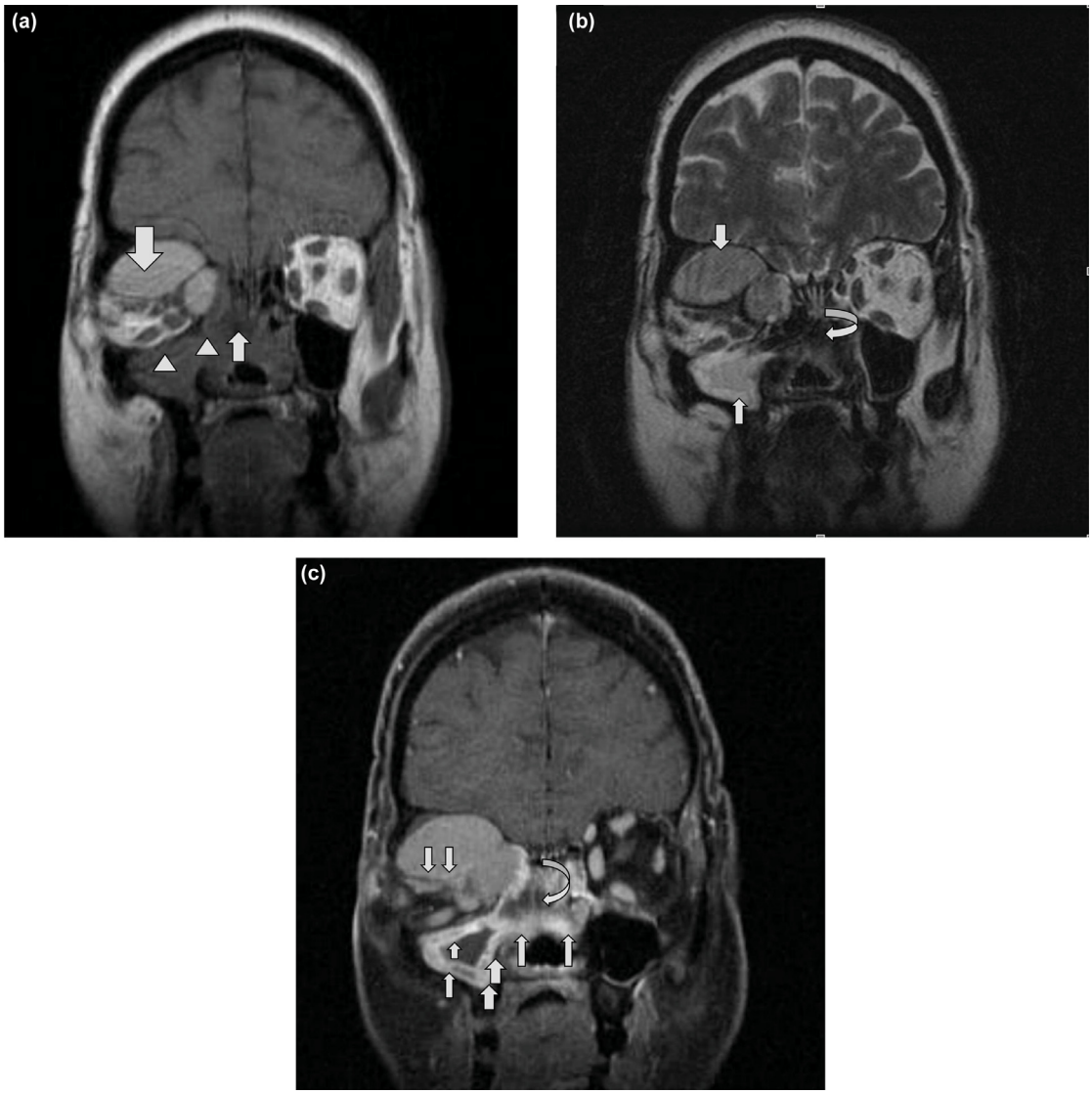

Fig. 2 – (A) Coronal T1 Weighted Image reveals confluent sinonasal soft tissue (triangles) isointense to the brain parenchyma that contains a low signal area in the midnasal cavity (upward arrow). The orbital mass is hyperintense due to proteinaceous, mucoid material in the lesion (downward arrow). (B) Coronal T2 Weighted Image shows the mass in the middle nasal cavity to be of low signal (curved arrow) related to scar tissue. The maxillary sinus is hyperintense (upward arrow) related to chronic sinusitis. The orbital lesion appears hypointense (downward arrow). (C) Contrast Enhanced Fat Suppressed T1 Weighted Image at the same level demonstrates rim enhancement of the confluent sinonasal soft tissue (upward arrows). The lesion in the central nasal area shows no enhancement (curved arrow). Faint rim enhancement of the inferior wall of the orbital mass is noted (downward arrows).

vessels, decreases nutrient delivery to the underlying cartilage leading to necrosis of the mucoperichondrium and eventually causing septal perforation [3,5,6]. Furthermore, cocaine also impairs the body's defense mechanism by slowing nasal mucociliary transport [5]. Nasal septal perforation is the best known complication of nasal cocaine consumption, found in approximately 5% of cocaine snorters [7].

Extensive osteocartilaginous necrosis is caused by a combination of the ischemic effect of cocaine or the topical decongestant and the caustic effect of adulterants. It is further potentiated by the local anesthetic effects which result in trauma or thermal injury leading to secondary infections [2,6,8,9]. Commonly described manifestations of intranasal cocaine abuse include epistaxis, crust formation, nasal septal perforation, saddle nose or alar deformities, hard palate destruction in advanced diseases, and erosion of the internal nasal and sinus structures [3–5]. Unique in our patient is the manifestation of a giant mucocele formed secondary to chronic cocaine abuse. Mucoceles are epithelial lined mucus containing benign lesions which originate in the paranasal sinuses. They are the most common expansile lesions of

the paranasal sinuses. A mucocele develops when there is obstruction of a sinus ostium or a compartment of a septated sinus by chronic sinusitis, polyp, tumors, prior surgery, or trauma [1,10].

The sinus cavity expands with the trapped secretions and is eventually completely mucus-filled and develops into a mucocele. The entrapped secretions cause pressure necrosis and slow erosion of the inner sinus bony wall, soon areas of deossification set in when the body's repair mechanisms cannot keep up with the extensive erosion [10].

Inflammatory mediators like prostaglandins, IL-1,TNF have also been proposed to play a role in the bone resorption and expansion of the mucoceles [11]. The expanding mucocele takes the path of least resistance.

Mucocoles have been described as a sequela of trauma. In one study, mucocele was the most frequent soft tissue lesion associated with trauma [12]. Sporadic cases of giant frontal mucocele have been described with manifestations ranging from extra-axial proptosis to complete loss of vision [13–15]. The majority of mucoceles occur in the frontal sinus with the ethmoid sinuses representing next most common location [16]. Mucoceles can be seen in association with cystic fibrosis [17]. MRI appearances vary based on the relative proportions of water and protein and enhancement is confined to the periphery of the lesion [16,18]. Invasive fungal infections can be considered in the differential diagnosis, as they have a propensity for orbital invasion [19]. Five cases of osteocartilaginous necrosis of the sinonasal tract as a sequel of cocaine abuse were reported in 1989 and those authors proposed including destructive midface lesions such as Wegener's granulomatosis and polymorphic reticulosis in the differential diagnosis [20].

The patient had a large nasal septal perforation with extensive scar tissue formation and chronic sinusitis related to prolonged cocaine abuse. The blockage of the sinus drainage pathways led to the development of this giant frontoethmoidal mucocele, which subsequently led to destruction and remodeling of the sinus wall with bony spicules in the mucocele. Although CT scan better delineated the bony details of the sinonasal disease, MRI accurately depicted the different components of the intranasal soft tissues and characteristics of the mucocele.

Extensive distortion and destruction of anatomy caused by intranasal scarring led to a giant mucocele which extended into the orbit. In summary, a unique case of a giant frontoethmoidal mucocele is presented which was secondary to cocaine induced inflammatory and destructive changes. MRI was invaluable in depicting the extent of this lesion. Cocaine abuse should be considered as a possible cause of frontoethmoidal mucocele.

Patient consent statement

No identifiable information revealing or alluding to the identity of the patient is contained in the manuscript or figures.

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