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Case Report

Mucocele of Frontal Sinus: 20 Years Post-Injury; A Case Report

Sumit Vishwakarma[1], Arul Selvi Subramaniam[1]*, Sachin A. Borkar[2], and Deepak Agrawal[2]

[1]*Department of Laboratory Medicine, Jai Prakash Narayan Apex Trauma Centre, AIIMS, India*

[2]*Department of Neurosurgery, Jai Prakash Narayan Apex Trauma Centre, AIIMS, India*

***Corresponding author**

Arul Selvi Subramaniam, Department of Laboratory Medicine, Jai Prakash Narayan Apex Trauma Centre, AIIMS, India, Email: arulselvi.ipnatc@gmail.com

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Abstract

Frontal sinus mucocele may present years after trauma. The interval between the initial injury and the presenting signs and symptoms may range from 1 to 35 years. Trauma accounts for 9–28% of the mucocele formation.The aim of this article is to present a rare case of mucocele that developed in a 35-year-old Indian man after having a frontal sinus fracture approximately 20 years back. This case also brings insight onproper diagnostic assessment, managementand needs of long term follow-up of frontal sinus fractures.A review of the literature was performed which showed only a few documented cases of mucocele formation after trauma. We therefore advocate long term follow-up of at-risk-patients to properly investigate them for development of mucocele.

KEY MESSAGES

Mucoceles are rare complications that can develop many years after trauma, thus necessitating a virtually life-long follow-up. Histopathology plays an important role in confirming the diagnosis of mucocele alongwith radiological and clinical correlation.

INTRODUCTION

The mucocele of the frontal sinus is a benign entity that develops from accumulation of mucoid secretions in a cyst lined by muco-periosteum and respiratory epithelium [1]. The histological features of mucocele are quite nonspecific soit requiresa clinical, radiologic, and pathologic correlation to confirm the diagnosis. We present case of a 35-year-old man who developed a mucocele of the frontal sinus 20 years after the initial trauma. Review of literature shows giant frontal mucoceles are an uncommon clinical entity and they can lead to orbital, extracranial and anterior cranial fossa extension.

CASE PRESENTATION

A 35-year-old male, presented with a history of rhinorrhoea and anosmia since last two weeks. He had history of road traffic accident and sustained head injury approximately 20 years back for which he received symptomatic care. The patient confirmed that he was symptom free before the accident. He had a surgery (Craniotomy) 10 years after the initial trauma after which he had H/o an episode of meningitis which was managed with antibiotics and supportive measures. Since then he had repeated attacks of rhinorrhoea and anosmia three to four times in a year. On present examination, his nasal cavity was normal on anterior rhinoscopy. His eye movements and vision were normal. His MRI scan revealed a homogenous mass in left frontal sinus so a left frontal craniotomy and evacuation of mucocele (type 5A) with repair of defect under general anesthesia was done. During surgery the posterior and inferior walls of the frontal sinus were found to be destroyed and approximately 2.5 X 2.5 cm, translucent, faint greenish-grey, encapsulated soft tissue completely filling the left frontal anterior fossa was resected and sent for histopathological examination. Frontal mucosa was completely removed. The sinus was irrigated with saline.Standard rehabilitation measures was taken. The anterior bony defect was repaired with a metallic mesh. On gross, we received the above described tissue which grossly revealed greenish-grey content with smooth wall. Microscopic examination revealed a cyst wall focally lined by pseudo stratified, ciliated columnar epithelial lining (Figure 1) with foci's of mucinous content, granulation tissue and areas of hemorrhage (Figure 2). A diagnosis of a post-traumatic mucocele was made after correlation with clinical and radiological findings. The postoperative course was uneventful with no feature of recurrence since then.

Review of literature

We gathered information published on the subject of mucocele with history of trauma. EuDaly and Kraus [2] reported a case of frontal mucocele following previous facial trauma with hardware reconstruction. Yi-Chun et al. [3], reported a case of a nasal mucocele originating from complex facial fractures. Koudstaal et al. [4] reported three cases occurring 13–35 years after trauma. Weitzel et al. [5], and Mcgrawwall [6] reported few cases of mucocele including cases 16 years after initial trauma presenting with CSF leak and 23 years after the initial

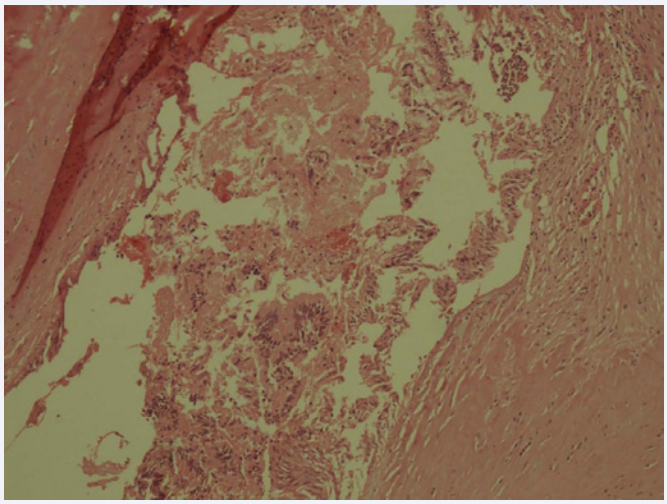

Figure 1 (H&E, 20 x), Cyst wall with focal respiratory epithelium.

trauma with forehead swelling respectively.Sara Weidmayer [7] presented a case of frontal mucocele with intracranial extension causing frontal lobe syndrome. Steinberg et al. [7], presented a casewith proptosis and diplopia 15 years after sustaining a trauma to the frontal sinus.The second case was presentedwith recurrent frontal headaches of five years' duration 33 years after the initial trauma.In the literature there is no consensus regarding the follow up, because of the long period after trauma until the mucocele develops [8]. Some adviced lifetime follow up for these patients and some suggest a follow-up period of 5 or 7 years [9].

DISCUSSION

Mucocele of the frontal sinus was first described by Dezeimeris in 1725 and the term mucocele was coined by Rollet. A frontal mucocele may be fronto-ethmoidal or strictly frontal. The male: female ratio varies a lot in the literature from 7:1 to 1:1. The patients present mostly between the second and the seventh decade with an average of 50 years [10]. Mucocele that result from trauma are mainly due to compromised ventilation, and can occur between 1 and 35 years later. Trauma accounts for 9–28% of the mucocele formation [11].They are not common in the pediatric age group [12]. 65% to 98% of the mucocele appear in the frontal sinus and 8–25% in the ethmoidal sinus. [12] Maxillary sinus mucocele is rare and represents less than 10% of all paranasal sinus mucoceles and are more common in Asian population [13]. They are considered to be caused by obstruction of drainage ostium of affected paranasal sinus due to chronic rhino sinusitis (infectious or allergic), nasosinusal polyposis, craniofacial trauma, previous surgery, benign tumors (osteomas, bone fibrous dysplasia), or malignant neoplasms (primary or metastatic).Pathophysiology of the mechanism of bone reabsorption due to mucoceles is still not clear but it is believed that osteolysis is produced by devascularization of the bone because of compression and/or by the action of inflammation involving cytokines (IL1, IL6), vascular adhesion molecules and prostaglandins [14].

The mucoceles are slow-growing and are associated with symptoms depending upon their location and extentsuch asfrontal headache, proptosis, diplopia, nasal congestion, visual changes, fluid leakage from the nose and swelling over the forehead. When a mucocele erodes the posterior wall of the frontal sinus because posterior wall is notably thin, it may cause meningitis, meningoencephalitis, pneumocephalus, brain abscess, seizures or CSF fistulas [14-16].

Differential diagnosis of mucocele includes cholesterol granuloma, epidermoid cyst, meningioma, encephaloceles, neurofibroma, salivary adenoma, paraganglioma, nasoangiofibroma, malignant neoplasms and Space occupying lesions seen on CT such as schwannoma, chordoma, ethmoidal chondromyxoma, cystic hypophyseal adenoma and retrobulbar cysts [17].

Based on the extent of frontal mucoceles, it is classified in various groups by Har-EI [18] as given below-

Type 1: Mucocele limited to frontal sinus only, with or without orbital extension.

Type 2: Mucocele involving frontal and ethmoids, with or without orbital extension.

Type 3A: Mucocele erodes the posterior wall of the frontal sinus with minimal or no intracranial involvement.

Type 3B: Mucocele erodes the posterior wall of frontal sinus with major intracranial extension.

Type 4: Mucocele erodes the anterior wall of the frontal sinus.

Type 5A: Both anterior and posterior walls of frontal sinus are involved with minimal or no intracranial extension.

Type 5B: Both anterior and posterior walls of frontal sinus are involved with major intracranial extension.

Accordingly, the case describedhere can be regarded as a 3A-type mucopyocoele. Our patient had erosion of the posterior table of the frontal sinus and extension into the extradural space as detected only by CT scan. When a mucocele become infected, it converts into pyocele and giving rise to the symptoms and signs of inflammation.

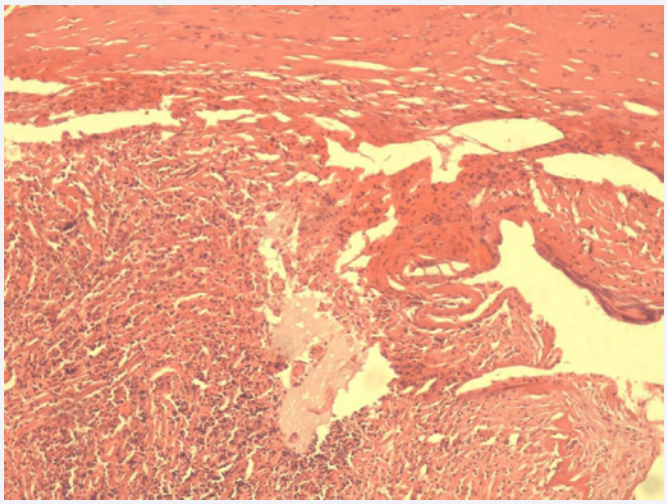

Figure 2 (H&E, 40x) Cyst wall with chronic inflammation. Mucinous material also seen.

The histologic appearance of mucocele can be very subtle, showing strips of flattened, pseudo stratified, ciliated, columnar epithelium. Metaplastic changes of the epithelium may beseen in long-standing cases. The ciliated epithelium is quite remarkable (Figure 2). Intenseintralesional pressure results in bone remodeling and destruction. There may be reactive bone formation immediately adjacent to the epithelium. Mucinous material may be present and is usually associated chronic inflammation. Rupture and spillage of mucoid content may result in fibrosis, granulation tissue, hemorrhage, and cholesterol cleft formation or cholesterol granuloma [19].

Both CT and MRI imaging are important to the workup of mucoceles. A CT scan is the best modality to demonstrate a homogenous mass and is helpful to demonstrate the extent of bone expansion and/or erosion [20]. MRI helps to in the approximity of mucocele to brain, orbit, and the overlying soft tissues [21]. The hyperintense T2-weighted signal of the mucocele secretion on MRI make itpossible to differentiate them from the non-ossified matrix of fibro osseous lesions [22]. MRI with contrast enhancement is useful to differentiate mucoceles from neoplasms [23].

Latest view on management of mucocele is to conduct functional, least invasive and low morbidity procedure with nasosinus endoscopic surgery, with marsupialization and abundant drainage of the lesion with preservation of the epithelium [24]. The recurrence rate of 7% is noted [25]. Frontal sinus fracture are potentially life threatening complication and are divided in an early, withinsix weeks and late, after six weeks group. Early complications are wound infection, cerebro-spinal fluid leakage, paresthesia, meningitis, cavernous sinus thrombosis, brain abscess and headache. Late complications include cosmetic problems, brain abscess, osteomyelitis and pyomucocele formation [26].

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