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Case Report
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Double Frontal Sinus Mucocele with Fungal Ball

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Abstract

Frontal sinus mucocele is commonly seen, but double frontal sinus mucocele is rare. Furthermore double frontal sinus mucocele with fungal ball is extremely rare. Recently we experienced an 80 years old female patient had double frontal sinus mucocele with fungal ball presented with pressure sense over frontal and which was successfully treated with endoscopic sinus surgery. To the best of our knowledge, this is the first case of double frontal sinus mucocele with fungal ball and we report this case with a review of related literatures.

Keywords: Fungus; Mucocele; Frontal sinus

Introduction

Mucoceles are benign, expansile, cyst-like lesion of the paranasal sinuses [1]. The mucoid secretions of mucoceles are usually sterile. However, secondary infections, mostly bacterial, may lead to the development of pyoceles. In contrast, mucoceles with superimposed fungal infections are extremely rare [2]. Only six cases have been reported in the literature, of which, two were allergic fungal sinusitis and the other four were fungus ball within a mucocele of the sphenoid sinus or ethmoid sinus [3-7]. In this article, clinical and radiological findings of an 80 years old female patient with isolated frontal sinus fungus ball within a double mucocele presented with head ache were discussed with recent literature.

In this case, there was fungus ball within a mucocele in upper compartment and only mucocele in lower compartment but which one was the first originated compartment was questionable. To the best of our knowledge, this is the first case of fungus ball within a double mucocele of the frontal sinus.

Case Report

An 80 years old female patient with a benign medical history, including no history of allergies, was referred for evaluation of a mass lesion in the left frontal sinus. The patient presented with 3 years history of pressure sensation over the left frontal with intermittent pain. There was no history of nasal surgery and head trauma history and she did not have any systemic medical disease. The patient did not complain of sinus symptom, patient's visual acuity and fields were bilaterally normal. All routine laboratory investigations including hematological, serological and allergy test were within normal limits. Nasal endoscopic examination revealed no significant abnormal findings in nasal cavities.

Coronal section of Computed Tomography (CT) scanning revealed an expansile, dumbell shape lesion which contained irregular high-density, calcified lesion in the left frontal sinus (Figure 1A). Axial section of CT images also showed expansile lesion with calcification and bony remodeling and sclerotic change of the surrounding bone in left frontal sinus and frontal recess, but no evidence of intracranial invasion or bony destruction of posterior wall of frontal sinus and skull base (Figure 1B).

Axial T1-weighted Magnetic Resonance Imaging (MRI) showed expansile, high signal intensity frontal sinus mass and there was hypointense signal lesion suggesting fungus ball (Figure 2A). Coronal and sagittal T2-weighted scan showed expanded frontal sinus mass and it seemed to be divided into two layers; double mucocele, and there was hypointense signal within the upper mucocele. Also the hypointensity signal mass seemed to be fungus ball (Figure 2B and 2C). The pre-operative diagnosis was double mucoceles with fungus ball in upper compartment.

The patient was underwent endoscopic sinus surgery under general anesthesia. Frontal recess was dissected and the floor of lower mucocele was detected. When the lower mucocele was opened, large amount of dark brown-colored thick mucoid secretion was drained and floor of the mucocele

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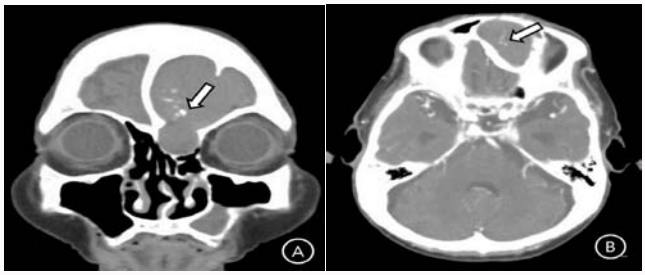

Figure 1: Coronal (A) and axial (B) enhanced computed tomography scan showing a large, expansile, heterogenous mass and the typical hyper attenuating fungus ball with calcific foci (arrow) in the left frontal sinus.

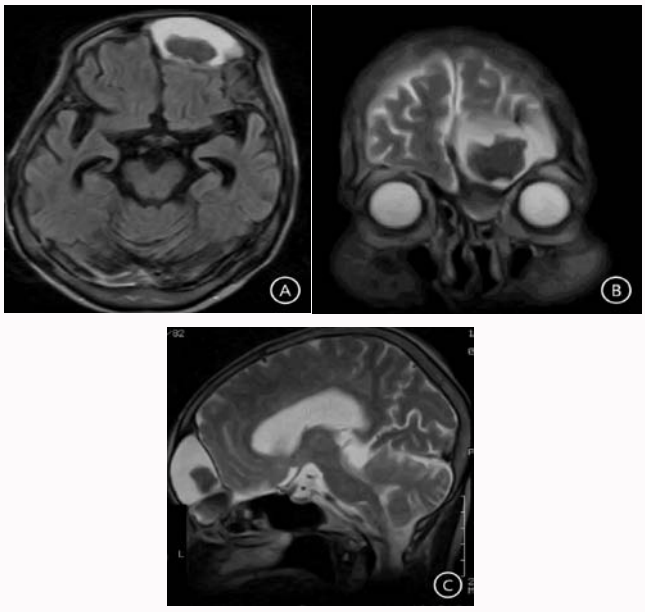

Figure 2: Axial T1-weighted (A) and coronal T2-weighted (B) magnetic resonance imaging scans shows expansile, none homogenous frontal sinus mass. Sagittal T2-weighted (C) magnetic resonance imaging scan shows expanded frontal sinus mass and frontal recess mass seemed to be divided into two layers.

was further widened. The floor of upper mucocele was intact and very thin, and the wall was pulsating. When the upper mucocele was opened, brown-colored mucoid discharge was drained and mucopurulent, cheesy or clay like material suggesting fungus ball. The fungus ball and mucous secretion were successfully removed and the inferior wall of the upper mucocele was widened enough to maintain patency. The post-operative course was uneventful.

Histopathological examination of the cheesy, clay like material was consistent with an aspergilloma. Microscopic examination demonstrated septae and 45°, dichotomous, branching hyphae, establishing a diagnosis of aspergillus (Figure 3). At 24 months follow up, the patient was diseased free without any sequelae.

Discussion

A mucocele is an expansile lesion of the paranasal sinuses [1]. Mucoceles with a superimposed fungus ball are extremely rare. It does not ever reported before that fungus ball within a mucocele of the frontal sinus. Besides the mucocele was double mucoceles in this case. The coexistence of a fungus ball within a mucocele can be explained by the hypothesis that the fungus ball within the sinus ostium and the development of a mucocele [2].

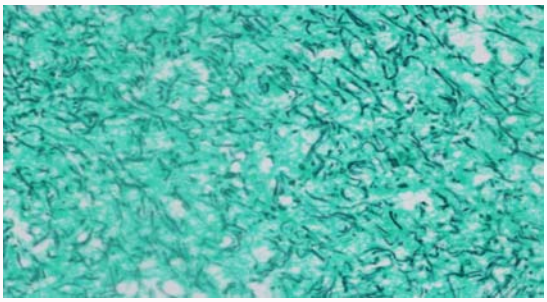

Figure 3: Histopathological finding of fungal hyphae showing acute angled branching and septated (Grocott's methenamine silver stain, ×200) (B).

Fungal ball tends to occur in older individuals with an apparent female predilection. Afflicted individuals are usually immune competent. Similarly to this case, individuals commonly described a chronic pressure sensation involving one of the paranasal sinuses [8]. But usually many of patients are either asymptomatic or have minimal symptoms, one of the things that plays an important role in the differential diagnosis of mucocele and fungal sinusitis is radiologic examination. Especially CT and MRI scans are useful [4]. Mucoceles and fungal sinusitis often present mass like lesion in the paranasal sinuses on CT scan. Mucoceles present as homogenous opacification like brain parenchyma [9]. In contrast, fungal often present high density material containing calcification. Therefore calcified features are not always presented in fungal sinusitis, it is difficult to distinguish between two disease by calcification [10,11]. Mucoceles often present as hypointense lesion on T1 weighted MRI, and hyperintense lesion on T2 weighted MRI. In contrast-enhanced MRI images, the sinus mucosa enhances as a thin line surrounding the mucocele. However, the MRI appearance of mucoceles varies depending on their protein concentrations, which change over time [12]. If the mucocele has more than 25 percent of protein, MRI showed a low signal intensity lesion on both T1W and T2W images. Besides, fungal sinusitis is seen with low signal intensity on both T1W and T2W images [13]. If the mucoceles are filled with a high viscosity of mucus concentration, it will be difficult to differentiate mucocele from fungal ball using radiologic findings. Therefore, it can be confirmed by postoperative histologic patterns and the mucus to see during surgery.

The only curative treatment for fungus ball within mucocele is surgery, which allows removal of fungal debris from the affected sinus and re establishes its proper ventilation and drainage. Antifungal management is generally unnecessary and should be reserved for the immune compromised patient [14]. In our patients, frontal sinusotomy enabled successful endoscopic drainage of the fungus ball and purulent secretions, without administration of systemic antifungal agents.

As we described above, we had to think about which mucocele was formed in advance. One theory is that the frontal sinus was obstructed by mucocele at entrance of frontal recess and then frontal sinus mucocele was formed and superimposed with fungus ball. The other theory is that fungus ball within a mucocele was formed in advance and then the other mucocele was formed by spontaneous inflammation just below the frontal sinus mucocele. It is questionable but we think the former theory is more reasonable.

Frontal sinus fungus ball within a mucocle is extremely rare. Although the advance of the diagnostic tools and examinations of radiological image, diagnosis of this disease still remains a challenge.

Understanding the clinical presentation and knowing its characteristic radiologic features are needed to the clinician to use appropriate diagnostic techniques for confirmation. Prompt diagnosis and initiation of appropriate treatment are essential to avoid a protracted of serious outcome.

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