Bilateral Involvement of Frontoethmoidal Sinuses with Orbital Extension; an Extremely Rare Manifestation of Mucocele

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Abstract

Bilateral involvement of frontoethmoidal sinuses with orbital extension; An extremely rare manifestation of mucocele: case report. Objectives: Bilateral frontoethmoidal mucocele with orbital extension is a rare lesion which can imitate bilateral medial canthus masses. Its timely diagnosis and surgical interventions prevent further complications such as intracranial and nasal extensions. Case report: A 32 year-old male was referred to our clinic with complaint of epiphora and continuous headache lasting one year. CT scan depicted bilateral hypodense masses in frontoethmoidal sinuses and bone erosion on medial wall of right orbital cavity. In his surgical history, there was an endoscopic sinus surgery for chronic sinusitis 4 years ago. This document as one of the predisposing factors of mucocele led us to consider mucocele as the most likely diagnosis. Therefore, he underwent endoscopic sinus surgery at doctor's discretion. Preoperative problems disappeared 3 months after surgery. And no recurrence has been observed up to now. Conclusion: Although bilateral frontoethmoidal sinus mucoceles are rare, however, they should be considered in such similar cases and immediate interventions should be made.

Key words: bilateral mucocele, frontoethmoidal mucocele, mucocele, intraorbital extension

Introduction

Paranasal mucocele is a benign cystic lesions covered by epithelium layer. It is caused by mucus secretion and its aggregation in sinus cavities. It is probable that it accompanies by infection then it would be defined as mucopyocele. (Bahadir et al., 2011) Paranasal mucoceles are commonly observed. The major percentage of them consists of frontal mucoceles (60%) and ethmoidal (30%) while a small percentage is dedicated to maxillary (3-10%) and sphenoid (1%) mucoceles. Although frontoethmoidal mucoceles are common, bilateral involvement of these sinuses simultaneously is very rare and a limited number of cases have been reported. (Darouassi et al., 2005; Belli and Oktay, Bilateral Frontal Sinus Mucocele; Sharouny and Narayanan, 2015; Chandra et al., 2007) Mucocele expands gradually without any significant symptom in most cases until its pressure on surrounding constructs and bone erosion occur. (Strek et al., 2007) In frontoethmoidal mucocele various symptoms is observed depending on its location and expansion. However, the most prevalent reported symptoms are frontal headache, proptosis, diplopia which is result of infrolateral globes displacement. (Jacobson et al., 1982) Computed tomography (CT) is appropriate for mucocele diagnosis and investigation of its extension and expansion to surrounding constructs while magnetic resonance imaging (MRI) is applicable to investigating lesion connection with around soft tissues and differentiating it from neoplasms. (Topdag et al., 2015) Since mucoceles intrinsically expand, they should be treated before causing serious complication. There are two approaches for mucocele treatment; external surgery (osteoplastic flap technique with or without destroying sinuses) and endonasal endoscopic sinus surgery. (Beigi et al., 2016) In this study a rare case of bilateral and multiple involvement of frontal and ethmoidal sinuses by mucocele is discussed.

Case presentation:

Patient is a 32 years old male who presented to ophthalmology clinic with complaint of permanent excessive tearing and eyes swelling from one year ago. The symptoms were relieved by prescribed drugs. However, after a while, he presented to ophthalmology clinic again with severe eye swelling. He was diagnosed with lacrimal duct obstruction and suggested surgery. However, the patient refused the surgery. After a while, he presented again to ophthalmology clinic and was referred to our otolaryngology clinic at the ophthalmologist discretion. The symptoms such as periorbital swelling, proptosis in both eyes which is more distinct in the right eye than the left one, and a mild globe displacement in the right eye were observed from the patient's appearance. Physical examination depicted bilateral medial

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canthus palpable and nontender masses.

Furthermore, preoperative vision tests demonstrated normal visual field and the visual acuity of 20/20 in both eyes. Pupils' reflection to the light was normal. In addition, no eye movement restriction was observed in the patient while he complained from pain in medial canthus region by lateral and medial movement. Additionally, he stated that he suffered from continuous progressive headache. His surgical history showed that he underwent an endoscopic sinus surgery for chronic sinusitis about 4 years ago. The patient denied any drug administration or any other diseases in his history. Furthermore, he stated that there was not any similar case in his family.

Therefore, after examination, the radiological investigation was requested and CT scan results depicted bilateral hypodense masses which opacified frontal and ethmoidal sinuses with intraorbital extension. Furthermore, medial wall erosion was distinct in the right orbital cavity. (figure 1)

According to the patient history, physical examination, and CT scan, the patient with the likeliest diagnosis of bilateral mucocele underwent endoscopic sinus surgery under general anaesthesia. The drained sinus content was sent to pathology to further investigation. The pathological investigation results verified the initial diagnosis of bilateral mucocele.

Finally, after two days with successful recovery the patient was discharge and suggested coming back to our clinic for further investigation.

Post-operative CT scan shows satisfactory outcome after two months.

Post-operatively blurred vision, proptosis, and eyes asymmetry and alignment were eliminated.

Furthermore, the periorbital swelling disappeared gradually after operation. Moreover, no alteration in visual acuity and field was observed. In addition, there was no recurrence of the lesion and symptoms in one-year follow-up.



Figure 1: preoperative CT scan revealed hypodense density opacified frontal and ethmoidal sinuses with intraorbital extension

Discussion

Mucoceles are benign lesions which are caused by sinuses' outflow obstruction and accumulation of mucus inside sinuses. The obstruction may be the result of previous sinus surgery scar, sinus inflammation, chronic sinusitis, sinunasal polyps, trauma, fracture in face and sinuses, congenital anomalies, and obstruction of masses such as osteoma, papilloma, and etc.

With the expansion of mucoceles inside the sinuses, sinuses' wall's bones erosion and compressive effects on surrounding structures may occurred. (Khong et al., 2004) Paranasal sinus mucoceles are very common, but bilateral and multiple involvement of sinuses are infrequent and to the best of our knowledge less than 10 cases were reported in litreture. (Belli and Oktay, Bilateral Frontal Sinus Mucocele; Sharouny and Narayanan, 2015; Varghese et al., 2004)

Mucoceles in frontal sinus are more prevalent and observed in all ages while they are more common between the ages of 40 and 60 without sex predilection. (Belli and Oktay, Bilateral Frontal Sinus Mucocele) Mucoceles have various symptoms according to their

location and expansion. For instance, the intraorbital extension produces pain, epiphora, exophthalmos, decrease in visual acuity, globe displacement, ocular movement restriction, and periorbital swelling. While, intracranial extension brings out symptoms like meningitis, headache, cranial nerve paralysis, and epidural abscess and nasal extension causes nasal obstruction, anosmia, post-nasal drip, and rhinorrhea. (Strek et al., 2007)

The main complaints of patients with frontoethmoidal mucoceles are throbbing pain in frontal and occipital region, blurred vision, diplopia, and aesthetic problems such as eyes asymmetry and proptosis. (Belli and Oktay, Bilateral Frontal Sinus Mucocele; Jacobson et al., 1982; Shah et al., 2007)

Differential diagnosis which can cause medial canthus masses with symptom of epiphora and nasal congestion are involvement of lacrimal drainage apparatus such as abnormality in structure and size, infection lesion or malignant tumors such as squamous cell carcinoma (SCC), melanoma, transitional cell, or paranasal sinus involvement such as etmoidal mucocele, and or pranasal sinus neoplasm such as papilloma or SCC, orbital myositis and orbital dermoids, hemangioma, lymphangioma, lymphoma, sarcoidosis. (Friedman et al., 1993)

In diagnosis of frontoethmoidal mucoceles, CT scan and MRI are applied with physical examination and patient history. Mucocele is shown in CT scan as a homogenous hypodense expansile mass. This mass is the substitute of the air inside the sinuses. Furthermore, CT scan depicts surrounding bone erosion and mucocele extension clearly. While MRI may present different image of mucocele depends on the ratio between its water and protein, usually the images are hypointense on T1-weighted images and hyperintense on T2-weighted images. (Topdag et al., 2015; Van Tassel et al., 1989)

Among two approaches of external surgery and endoscopic sinus surgery, the latter is more acceptable since external methods may cause cutaneous nerves damage, scar, and aesthetic problems. (Beigi et al., 2016)

The histopathology of mucocele shows that huge cysts filled by mucus with granulation tissue margin. Furthermore, the infiltration of inflammatory cells which mainly are histocyts, is observed. (Batsakis et al., 1980)

Finally, it is possible to avoid further complications by timely diagnosis and immediate treatment. (Topdag et al., 2015) After operation, the patients' follow-up shows no high recurrence albeit a mucocele needs many years to be created and being symptomatic, therefore, long-term follow-up is required to obtain accurate data.

Conclusion:

The result of discussed case show that although bilateral mucoceles are rare, they should be considered in patient with bilateral medial canthus mass with orbital symptoms.

CT scan and MRI are utilized to diagnose mucoceles and endoscopic sinus surgery is the best choice of treatment.

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