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Case Report Open Access

Orbital Pseudotumor Initially Presented as Chronic Sinusitis: A Case Report

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Abstract

Idiopathic orbital pseudotumor, is defined as a benign, non-infective lesion characterized by orbital inflammatory process. A 73-year-old man, with a 1 year history of severe headache and unilateral left-sided chronic pan-sinusitis without orbital involvement underwent functional endoscopic sinus surgery (FESS) surgery due to sinusitis recurrence. Also Dacryocystorhinostomy (DCR) surgery and re-surgery of sphenoid sinus and middle turbinecomy were conducted. About 5 months later, patient presented with ocular pain, diplopia and reduced left eye movements. He underwent surgery and biopsy was conducted for histopathology and immunohistochemistry (IHC) and their reports confirmed the diagnosis of orbital pseudotumor. The initial symptoms did not start from orbit, interestingly. Notably, one of the differential diagnoses of unilateral chronic sinusitis can be orbital pseudotumor.

Keywords: Idiopathic orbital pseudotumor; Chronic sinusitis

Introduction

Idiopathic orbital pseudotumor, is defined as a benign, non-infective and autoimmune disease characterized by orbital inflammatory process with unknown local or systemic etiology [1]. It includes up to 10% of all orbital tumors [2,3]. It can involve any part of the orbit and unilateral presentation is more usual than bilateral one [1,4]. It often involves orbital fat, extraocular muscles and lacrimal glands respectively [3]. Imaging evaluation of orbital pseudotumor commonly involves computed tomography (CT) scan or magnetic resonance imaging (MRI) [5]. CT scan is a preferred imaging modality for evaluating orbital pseudotumor [6]. The first-line treatment for orbital pseudotumor is corticosteroid [7]. This study evaluated a 73-year-old man with unilateral pseudotumor of the left orbit with 1 year history of chronic sinusitis.

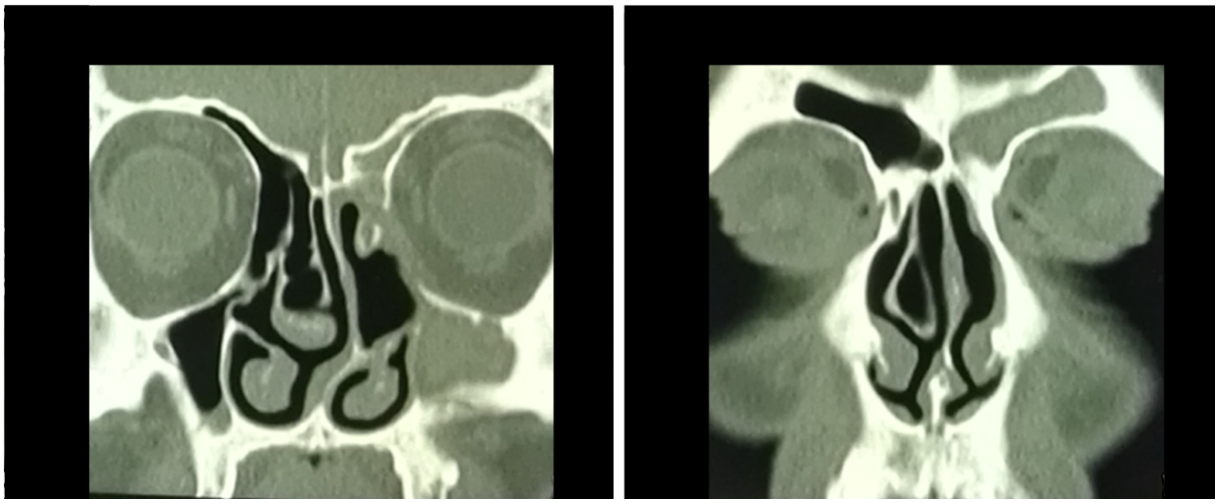

Figure 1: Unilateral left-sided pan-sinusitis.

Case Presentation

The patient was a 73-year-old man with the initial presentation of severe headache resistant to clinical treatment. Patient with the symptom of one-sided headache referred to the neurologist, initially. The imaging demonstrated a unilateral left-sided pan-sinusitis (Figure 1). Patient referred to otorhinolaryngologist. After receiving systemic antibiotics due to no improvement, surgery and sinus drainage was indicated due to the extent of all sinus involvements. Patient underwent functional endoscopic sinus surgery (FESS) of sphenoid and ethmoid sinuses. Five months later, nasolacrimal duct obstruction

occurred due to chronic inflammation of sinus and nasal mucosa. Patient with the symptoms of tears and chronic dacryocystitis underwent Dacryocystorhinostomy (DCR) surgery. About 6 month later re-surgery of sphenoid sinus was done due to the recurrence of sinusitis and patient underwent middle turbinectomy too. Patient referred with clinical manifestations of left ocular pain, diplopia, left lower lid and cheek hypoesthesia, fat atrophy under the left eyelid, left lower and upper eyelid retraction and reduced left eye movements about 5 months after the last surgery (Figure 2). The extraocular muscle examination (Figure 3) showed severe left medial and inferior gaze restriction. Paranasal sinuses spiral CT scan with and without contrast revealed chronic sinusitis, significant inflammatory change and increased density of peri-orbital fat at infero-medial aspect of left eye involving inferior and medial rectus. Retrobulbar region was not involved and there was no significant optic nerve involvement (Figure 4). Dexamethasone, clindamycin and cefazolin were started for the patient according to CT report and diagnosis of orbital cellulitis without clinical response and antibiotics were discontinued. Patient had consulted with the ophthalmologist. Due to the course of the disease, the diagnosis of orbital pseudotumor was suggested and steroid therapy was started and biopsy had been done. Following steroid therapy the dramatic response to pain started within 12 h and histopathology and immunohistochemistry (IHC) reports following biopsy, confirmed the diagnosis of orbital pseudotumor. The patient's symptoms (especially the pain) never improved within one year before pseudotumor treatment. After pseudotumor diagnosis and treatment, there was a substantial improvement of patient's pain and eye movements. The steroid was tapered slowly. After stopping steroids, symptoms were relieved completely.

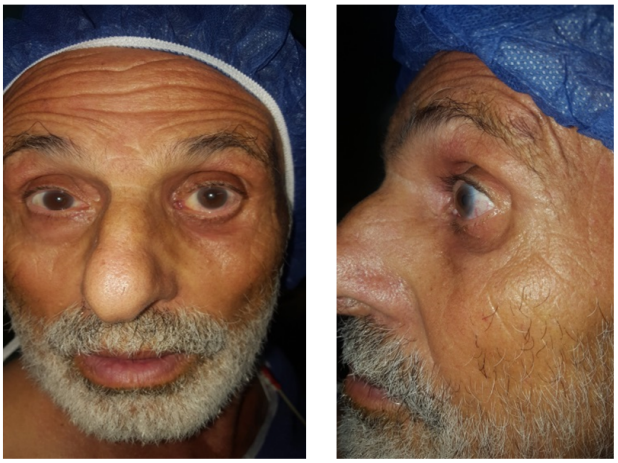

Figure 2: A 73-year-old man presented with ocular pain, left lower and upper eyelid retraction and fat atrophy under the left eyelid.

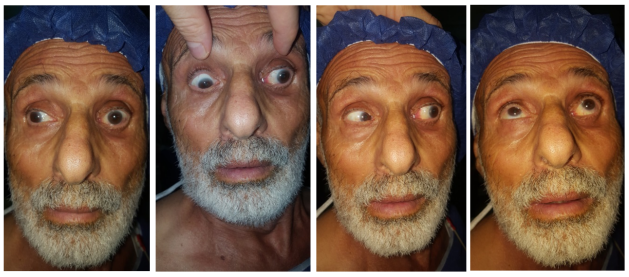

Figure 3: Reduced left eye movements. Severe left medial and inferior gaze restriction.

Discussion

Idiopathic orbital inflammation, also known as orbital inflammatory pseudotumor has no known etiology and pathogenesis at the moment. Infection and immune-associated etiologies may play important roles [5,8]. In our case, the cause of recurrent sinusitis was unknown initially and the orbital involvement which helped us in orbital pseudotumor diagnosis was a late finding. Although the diagnosis of orbital pseudotumor is based on clinical findings, the other inflammatory causes should be excluded [5].

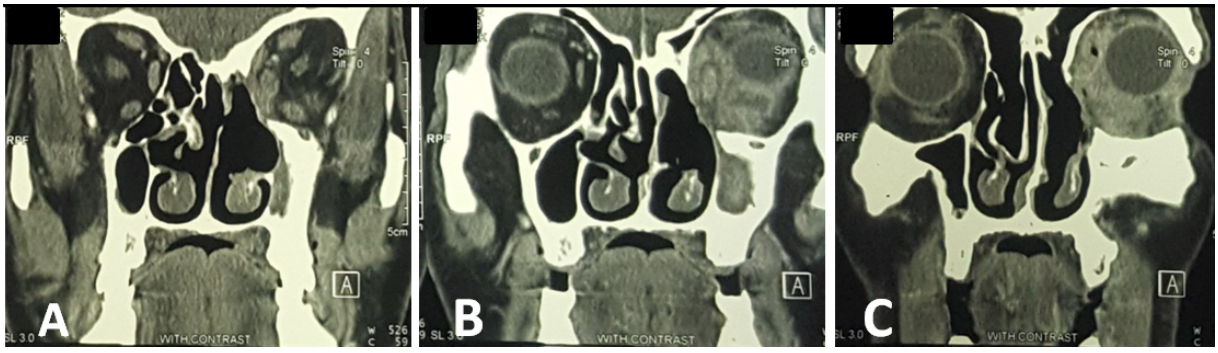

Figure 4: Coronal CT scan of left orbital pseudotumor. Retrobulbar was not involved and there was no significant optic nerve involvement (A). Note the increased density of peri-orbital fat at infero-medial aspect of left eye globe involving inferior and medial rectus caused by the orbital pseudotumor (B, C).

The most important diagnostic criteria for orbital pseudotumor include: clinical course and imaging, dramatic response to steroid therapy and pathological confirmation. The most prevalent symptom in adults is pain. Typical findings of orbital pseudotumor include acute

onset, ocular pain, peri-orbital edema and red eyes [6]. Also diplopia, chemosis, decreased vision and extraocular muscle restriction may be found [7,9]. The initial symptoms did not start from orbit in our case, interestingly. Our patient had symptoms of long-term and recurrent chronic sinusitis and hemi facial pain that never improved during one year and left orbit involved after that. Orbital pseudotumor can be localized or diffuse. If localized, inflammation can often involve extraocular muscles, lacrimal glands and cavernous sinus. If diffuse, it can involve peri-orbital fat and intraorbital soft tissue [10]. Extraocular muscle, peri-orbital fat and intraorbital soft tissue were involved in our case. The severity of orbital pseudotumor involvement may vary in different parts of the orbit [3]. It can involve orbital apex with the extension to the cavernous sinus (Tolosa-Hunt syndrome) [11]. In our patient, the anterior peri-orbital fat was involved substantially without orbital apex involvement. Therefore all patients with suspected orbital pseudotumor need to undergo a full ophthalmic examination that involves eyelid, orbit, extraocular muscles, globe, and optic nerve function evaluations. One of the differential diagnoses of orbital pseudotumor is orbital cellulitis [8]. Our case had 1 year history of unilateral left-sided chronic pan-sinusitis, interestingly and orbital cellulitis was observed on CT scan without clinical response to antibiotics. Following steroid therapy the pain and eye movements' limitation were dramatically improved.

Conclusion

There is probably a common immunopathogenesis in a unilateral non-infectious chronic sinusitis with orbital pseudotumor. This study recommends that, in patients with unilateral non-infectious chronic sinusitis with unknown etiology, biopsy should be done to rule out orbital pseudotumor.

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