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Unusual Presentation of Allergic Fungal Sinusitis Sparing Maxillary Sinus

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1. Introduction

In the last decade the understanding of fungal sinusitis classification well developed and recognized. It's basically classified into two main classification invasive fungal sinusitis and noninvasive fungal sinusitis. Invasive fungal sinusitis is sub-divided into divided into chronic invasive and acute fulminant invasive categories. While the non-invasive type is also sub-divided into two types, fungal ball and allergic fungal sinusitis (AFS) [1]. Allergic fungal sinusitis (AFS) is a benign, non-invasive, extra-mucosal fungal sinus disease representing an hypersensitivity/allergic reaction to extra-mucosal fungus in the sinus which is similar to allergic bronchopulmonary aspergillosis (ABPA) [2]. Millar et al. was the first who report the clinical features of AFS in the 1980s [3]. Pathologically, AFS is defined by the presence of fungal mucin, which is a tenacious, thick, eosinophilic discharge with characteristic histologic findings[4]. Many literatures have studied the clinical presentation, diagnosis and management of AFS. However, there are few articles focusing on the uncommon clinical presentation of AFS. This article showed rare presentation of AFS which present with heterogenicity of sphenoid sinus and right ethmoid sinus, but sparing maxillary sinus. It was diagnosed and managed at King Fahad Specialist Hospital (KFSHD), a tertiary care hospital in Al-Dammam, Saudi Arabia.

Case Report

54 years old Saudi gentleman known case diabetes mellitus type 2, hypothyroidism, complain of headache, nasal obstruction, hyposmia, nasal discharge (thick brown) for 2 years. His family and psychological history were insignificant. Flexible endoscopic nasal examination showed multiple Rt sided grade 3 polyps, thick brown secretion, deviated nasal septum to left. Eye, ear, throat, head and neck examination were clear. CT sinuses showed a heterogeneous opacity of sphenoid sinus and right ethmoid sinus but sparing maxillary sinus. The patient was counseled about the nature of the disease, the surgical management, treatment plans and recurrence. The patient underwent endoscopic sinus surgery and removal of polyps, thick allergic mucin and fungal mud performed by A.A. (rhinologist). Post operation oral steroid course for 2 weeks and nasal steroid spray and nasal washes. Culture was positive for asperigillus. Histopathology of nasal polyp reported as inflammatory (allergic IgE). Patient is following in the clinic and doing fine for the moment without signs and symptoms of recurrence.

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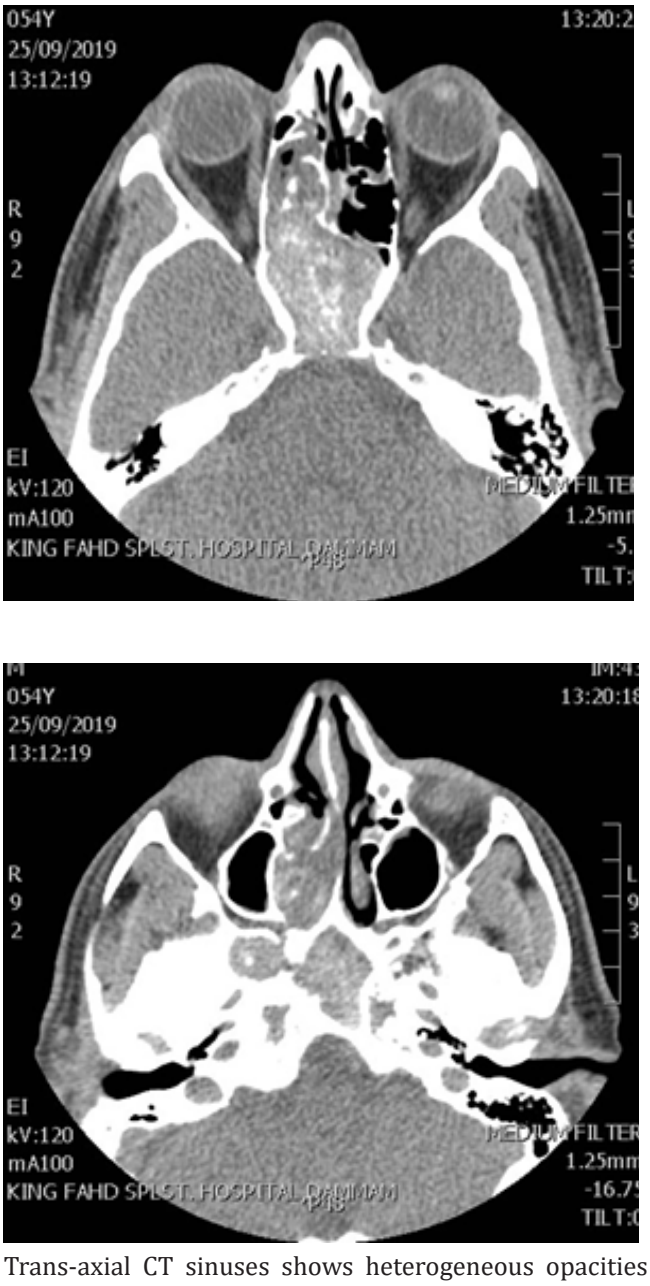

Trans-axial CT sinuses shows heterogeneous opacities filling the sphenoid and right ethmoid sinus but sparing maxillary sinus

Discussion

Allergic fungal sinusitis (AFS) is a benign, non-invasive, extra-mucosal fungal sinus disease representing an hypersensitivity/allergic reaction to extra-mucosal fungus in the sinus which is similar to allergic bronchopulmonary aspergillosis (ABPA). Robson et al. were the first who introduce the term allergic fungal sinusitis in 1989 [5]. They have identified several fungi causing similar clinical features. The most common fungi responsible of AFS are Dematiaceous fungi including; Bipolaris spicifera, Alternaria, Curvularia lunata, Helminthosporium, Chrysosporium, Drechslera, Fusarium and Rhizopus [6]. They account for 84% of the cases, followed by Aspergillus species (13%) and other rare species (<1%) [7]. Kuhn and Bent, have described 5 major and multiple minor criteria for diagnosis of allergic fungal sinusitis, which have been slightly modified over time. Major criteria include (1) nasal polyposis; (2) evidence of type I, IgE-mediated hypersensitivity confirmed by history, serology, or skin tests; (3) eosinophilic mucus without fungal invasion into sinus tissue; (4) positive fungal smear or culture; and (5) characteristic CT findings. Minor criteria include (1) unilateral predominance, (2) history of asthma, (3) CharcotLayden crystals, (4) peripheral eosinophilia, and (5) radiographic bone erosion. Elevated serum precipitin (IgG) antibodies against fungal antigens and IgE levels were not included in the criteria [8]. However, some patients didn't fulfil this criteria, but have been treated for AFS successfully. Computed tomography scans showed soft tissue mass with heterogeneous density in the sinuses with patchs of calcifications inside [9]. A wide consensus on the appropriate treatment of AFS has not been reached [10], and both medical and surgical procedures are used. Long-term medical therapy includes oral or topical corticosteroids, immunotherapy or/and antifungal agents. The use of steroids is helpful to reduce mucosal edema, polyps and inflammation [7]. Many authors suggested the use of antibiotics with steroids preoperativly to reduce the chances of superadded postoperative bacterial sinusitis and edema respectively [7,10]. The use of antifungal agents is controversial because it showed variable results. Endoscopy remaining the standard therapeutic step for both. Fungal debris removal from the involved sinuses, normal mucociliary drainage pathways establishment, and debulking of diseased tissue, are the main goals. In the post-operative period, oral steroids, nasal saline irrigation, and nasal corticosteroid sprays are administrated to avoid recurrence [11]. Post-operative follow-up is mandatory for all patient to identity and treat early recurrences [12-16].

Conclusion

This case report will enhance more understanding of such rare clinical presentation of allergic fungal sinusitis. AFS should be in the differential diagnosis even when heterogeneous opacification sparing maxillary sinus which is very rare.

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