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Case Report

Phaeoacremonium cinereum: an uncommon microorganism causing rare occurence of isolated right sphenoid fungal rhinosinusitis

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ABSTRACT

Fungal rhinosinusitis (FRS) can be either invasive or non-invasive, with the later characterized by the disease confined to the paranasal sinuses, mucous membrane, and bone wall without fungal invasion, including fungus ball (FB) and allergic fungal rhinosinusitis. Infection involving sphenoid sinus usually provokes headache or involvement of the surrounding cranial nerves due to its unique and posterior location in the nasal cavity. We encountered a case of right isolated sphenoid non-invasive fungal sinusitis caused by rare microorganism namely Phaeoacremonium cinereum. The disease was diagnosed based on clinical, endoscopy, radiological findings as well as specimen culture and sensitivity. Computed tomography (CT) scan provides information regarding the extension of sinusitis to dental, orbit, cranial bones and intracranial region. Once the diagnosis of sphenoid fungal rhinosinusitis is made, endoscopic sphenodotomy is the treatment of choice.

Keywords: Fungal rhinosinusitis, Sphenoid, Phaeoacremonium cinereum, Sphenoidoctomy

INTRODUCTION

European Position Paper on Rhinosinusitus and Nasal Polyps 2012 (EPOS) defines chronic sinusitis as more than two of the clinical symptoms (nasal blockage or discharge, facial pain, hyposmia or anosmia) and supported by the evidence of endoscopic findings (nasal polyp, mucopurulent discharge or edema within the ostemomeatal complex) or imaging evidence (mucosal changes within the ostiomeatal complex and/or sinuses in CT).[1] Sphenoid sinus is situated posteriorly in the apex of the nasal cavity with many vulnerable structures around it such as dura mater, cranial nerves (III, IV, V1, V2, and VI), optic nerve and chiasm, internal carotid artery, cavernous sinus, pituitary gland. In view of this specific anatomical location, patients with sphenoid sinusitis rarely complain of nasal symptoms such as nasal obstruction or rhinorrhea. In turn, most of them complain about headache and symptoms of cranial nerves involved. Mostly, it is the bacteria which causes chronic sphenoid sinusitis, followed by fungal organisms, with Aspergillus species being the most common. Hereby we present a case of right isolated sphenoid non-invasive fungal sinusitis caused by a rare fungus: Phaeoacremonium cinereum and right sphenoidoctomy was proceeded in view of unresponsive to medical treatment.

CASE REPORT

A 77-year-old lady with underlying hypertension, had a history of nasal congestion and non-foul smelling rhinorrhea for the past 20 years. She previously worked as a teacher and has had home gardening experience for 30 years. She came to our Otorhinolaryngology (ORL) department this year in view of her worsening nasal symptoms for the past 3 months and also complaint of bilateral orbital and facial tenderness, occasional headache and foul-smelling nasal discharge from right

nostril associated with post nasal drip. Otherwise, she didn't have fever or recent upper respiratory tract infection symptoms, and could tolerate orally without loss of smell, appetite and weight.

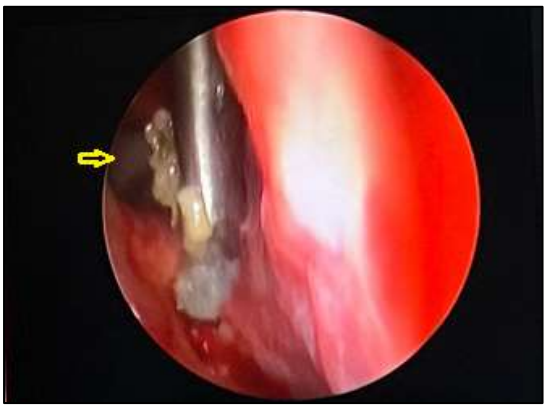

Figure 1: The second pass of the nasoendoscopy showed peanut butter like discharge from the sphenoid sinus ostium (yellow arrow).

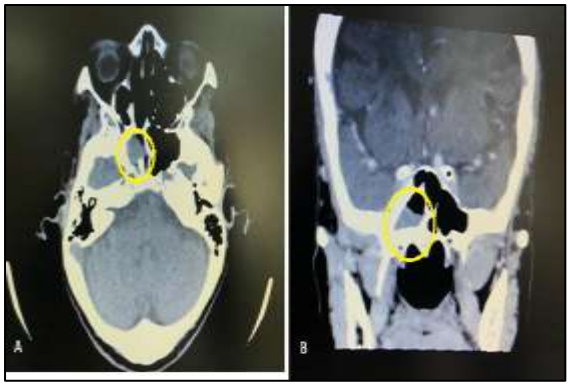

Figure 2: CT paranasal sinus, (A) Axial view, and (B) Coronal view, showed polypoidal mucosal thickening in the right sphenoid sinus (yellow circle).

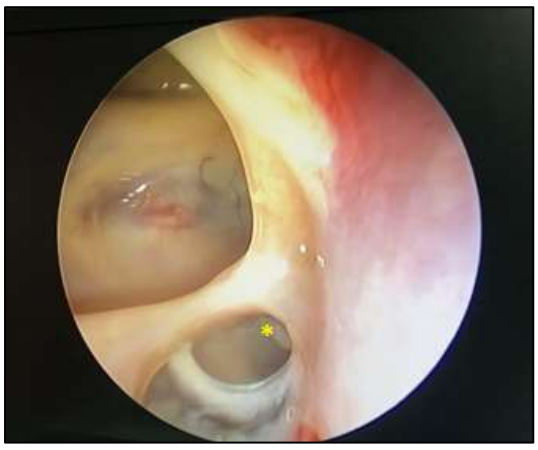

Figure 3: Post sphenoidotomy, the nasoendoscopic view of the right sphenoid cavity was clear (yellow asterisk).

Before she presented to our clinic, she was treated for acute rhinosinusitis and received oral ciprofloxacin and metronidazole for 3 weeks at a private medical center, however her symptoms persisted. Upon examination, she had minimal tenderness over right facial region upon palpation and had no signs and symptoms of neurological or ocular deficit. During the second pass of the nasoendoscopic examination, we noted minimal thick, tenacious and peanut-butter like discharge trickling from the right sphenoid sinus ostium (Figure 1). Other structures were unremarkable and the left nasal cavity was normal. Ear, throat and neck examinations were unremarkable. Thus, our provisional diagnosis was acute on chronic right sphenoid sinusitis, differential with fungal rhinosinusitis.

At the meantime, a computed tomography of paranasal sinus (CT PNS) was arranged and it showed polypoidal mucosal thickening in the right sphenoid sinus (Figure 2) without foci or calcification within and suspicious obliteration of the right frontal ethmoidal recess. There was no evidence of base of skull erosion and other paranasal sinuses were clear from the CT. After thorough discussion with her about the clinical findings and CT findings, she agreed for right endoscopic sinus surgery and disease removal. Right endoscopic sphenodotomy was performed under adequate local anaesthesia; intraoperatively, the thick, tanned and peanut-butter like substance, debris and crust in the right sphenoid sinus were cleared thoroughly. Subsequently the right sphenoid sinus cavity (Figure 3) was irrigated with copious amount of normal saline, and hemostasis was done with bipolar diathermy. The discharge was sent for culture and sensitivity and the fungal culture isolated: Phaeoacremonium cinereum. She was advised to do regular nasal douching at home and comply with nasal spray fluticasone. 2 weeks later, her symptoms improved, and she was well and subsequently discharged.

DISCUSSION

The diagnosis of sphenoid sinusitis might be delayed due to its non-specific and vague symptoms.[2] In view of some important structures (dura mater, cranial nerves (III, IV, V1, V2, and VI), optic nerve and chiasm, internal carotid artery, cavernous sinus, pituitary gland, sphenopalatine artery) around the sinus, these symptoms are often referred to these structures rather than the sinus itself.

In a study conducted by Supranee Fooanant et al, the most common symptom of sphenoid sinusitis is headache, mostly described as hemicranial headache (63.9% of its study population), and followed by visual disturbance or diplopia, cranial nerve deficit.[3,4] Commonly, the headache caused by sphenoid sinus disease is aggravated by head movement, coughing, walking, or bending.[5-7] Posterior nasal discharge is non-specific and commonly experienced by some patients.[8] Furthermore, nasal endoscopy can be unremarkable for

the patient with sphenoid sinusitis.[9] Thus, these often lead to delay of the diagnosis for several years.[10]

Besides that, Friedman et al had conducted a study on 50 patients and concluded that 72% of their cases were due to inflammation with chronic sinusitis 34%, fungal ball 20%, mucocele 12%, acute sinusitis 4%, and chronic invasive fungal sinusitis 2%. Neoplasms were seen in 16% of patients (benign 10%, malignant 6%).[11] In his study, the most common pathogens causing chronic sphenoid sinusitis were bacteria (methicillin-resistant Staphylococcus aureus being the most common, followed by Pseudomonas aeruginosa); followed by fungal organisms, with Aspergillus species being the most common. Other pathogens that have been reported include Streptococcus pneumoniae, aerobic Gram-negative bacilli (Pseudomonas aeruginosa, Klebsiella pneumoniae, Haemophilus influenzae Escherichia coli) and anaerobes (Peptostreptococcus, Fusobacterium, Prevotella and Porphyromonas). As for our patient, she suffered nonspecific headache, facial pain as well as rhinorrhea for two decades, and there was thick, tanned discharge from the right sphenoid ostium during second pass in nasoendoscopy, thus, a provisional of right isolated sphenoid non-invasive fungal sinusitis was made.

Fungal rhinosinusitis (FRS) is generally divided into invasive and non-invasive fungal infection. Non-invasive fungal rhinosinusitis characterized by the disease confined to the paranasal sinuses, mucous membrane, and bone wall without fungal invasion, including fungus ball (FB) and allergic fungal rhinosinusitis.[12,13] FB usually involves the maxillary sinus, followed by the sphenoid sinus.[12,13] Isolated sphenoid sinus FB is rare, especially for immunocompetent patients. Mostly, patients had treatment history with broad-spectrum antibiotics prior to the diagnosis of sphenoid sinus FB, thus; this may lead to the imbalance of the bacterial flora in human body and create an environment to enhance fungal growth.[14-16] Recent studies showed FB predominantly involves elderly female.[17-20] This was similar to our patient who is an immunocompetent, elderly female, and she received multiple courses of antibiotics prior to the diagnosis of right isolated sphenoid sinus FB.

Aspergillus species is the most common organism causing sphenoid FRS, however, the isolation of Phaeoacremonium Cinereum organism from our patient's right sphenoid discharge was indeed rare. Phaeoacremonium species found on diseased grapevine and woody plants, with Phaeoacremonium Cinereum being one of the newest species which was isolated from a young vine in Spain and Iran. Phaeoacremonium cinereum has been proved to have a maximum growth temperature of 37 C in growth temperature studies, suggesting that they can survive at human body temperature.[21] Phaeoacremonium Cinereum infections in human has been noted to increase over the past two decades and they are widely environmentally disseminated inoculum which can be found in soil, dust or indoor water sources.[22] Most of these reported infections involve subcutaneous abscesses, cysts, fungemia and osteoarthritis regardless of immunity state and these are often due to traumatic inoculation.[23] In our case, the patient had a history of home gardening for decades which impose a risk factor of obtaining such microorganism from woody plants or soil. The ability of this bacteria surviving at 37^0C has somemore explained why the bacteria can survive in her sphenoid sinus for months.

Computed tomography of the paranasal sinuses (CT PNS), aids in the diagnosis of FB. The involved sinus may appear opacified in a plain CT and it's usually limited to one sinus as the FB grow within the lumen of a paranasal sinus, and sometimes the 'double density' sign may appear. Once diagnosed sphenoid sinus FB, endoscopic sphenodotomy is the treatment of choice.[24] The time of surgical intervention is suggested to be done as early as possible to prevent the complications of intracranial or orbital involvements. Sphenodotomy, by widening of the sphenoid ostium, facilitates the disease clearance; provides adequate assessment to the sphenoid sinus during regular nasal toileting and improves accessibility for nasal douching. Once disease is thoroughly cleared and the normal sinus drainage system is restored, FB hardly recurs. The relief of symptom is found in about 75% of patients who clear the disease via surgical approach.[25]

CONCLUSION

Fungal rhinosinusitis usually requires endoscopic or open sinus surgery as a definite treatment option. The disease may not be life threatening at the initial stage but has significant morbidity and impact on the quality of life. Image guided surgery is advisable for the more complicated lesions. Usually patients with chronic sinusitis have satisfactory outcomes and relief of symptoms in about 75% of patients after who fail medical treatment and receive functional endoscopic sinus surgery. Rarely, chronic fungal sinusitis can result in orbital and intracranial infections but patient should be warned regarding this complication.

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