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Case Series

Invasive fungal granuloma: a diagnostic challenge

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ABSTRACT

Invasive fungal rhinosinusitis is a rare and life-threatening with high mortality and morbidity. Granulomatous invasive fungal rhinosinusitis has a time course exceeding 12 weeks. This disease has been reported primarily in south of Asia, however, it is very rare. When the invasive fungal sinus infection presents in a form of a granuloma with unusual presentation then they pose a diagnostic challenge to medical professionals. We present a case series of 3 cases of invasive fungal granuloma with varied presentations, which posed as a challenge in diagnosing them. Chronic invasive fungal granuloma (CIFG) of the paranasal sinuses is seen in immunocompetent hosts, especially those that are in the 3rd and 4th decades of their lives. Involvement of maxillary sinus and orbit is common than the sphenoid sinus involvement. Routine H and E staining may prove inadequate. Special stains such as the GMS stain should be employed in the slightest doubt of a fungal aetiology. A team approach towards patients is paramount for early diagnosis and timely medical and surgical intervention.

Keywords: Fungal granuloma, Invasive fungal sinusitis, Sphenoid granuloma

INTRODUCTION

Fungal rhinosinusitis can be classified as invasive and non-invasive on the basis of the histopathological findings of the invaded tissue. Invasive fungal rhinosinusitis is further classified into 3 groups: acute invasive (fulminant) fungal rhinosinusitis, granulomatous invasive fungal rhinosinusitis, and chronic invasive fungal rhinosinusitis.[1,2] Chronic non-invasive fungal sinusitis may be defined as the presence for months or years of mycelial mass which remains confined to the sinus cavity. Fungi don't destroy the local architecture. In contrast invasive fungal sinusitis is characterised by insidious extension across the mucosa into bone and other contiguous structures such as the orbit and brain.

Invasive fungal infections are seen both in immunosuppressed and immunocompetent hosts, but chronic invasive fungal granuloma (CIFG) usually presents in immunocompetent patients. Granuloma formation is caused by an inherited disorder of the immune system, affecting a specific part of the immune system, which results in the inability to produce the group of chemicals (reactive oxygen species) used by the immune system to kill invading microorganisms such as *Aspergillus*.[3] The granuloma is the last resort response, where the immune system tries to localize and prevent an infective or irritant stimulus by walling it off with a compact aggregate of histiocytes. As a result, microorganisms tend to persist and form a granuloma as the other cells of the immune system accumulate around the microorganism, preventing its spread in the body.

CASE SERIES

Case 1

40 years old female, farmer by occupation, presented with complaints of decreased sensation of left side of face and tingling sensation on left side of face, otherwise the patient had no significant past history, her general physical examination and ENT examination was normal ,except for wasting of muscles on left temporal region (Figure 1). Patient was subjected to CT scan of brain and paranasal sinuses (Figure 1). CT scan showed opacification of left sphenoid sinus with erosion of lateral wall of left sphenoid sinus and mass abutting the left internal carotid artery.

Biopsy was taken from the left sphenoid sinus under endoscopic guidance under local anaesthesia. Histopathological examination revealed chronic granulomatous lesion. On special staining with Gomori silver methamine (GMS) staining, non-segmented filamentous fungus probably of family mucoraceae was isolated and then diagnosed as invasive fungal granuloma, negative for AFB stain (Figure 1). Later patient underwent endoscopic sinus surgery and granuloma was removed in bits and pieces, she was on injection liposomal amphotericin-B 3mg/kg 5 days in a week for 6 weeks, with monitoring of serum electrolytes and she is on follow up and doing well.

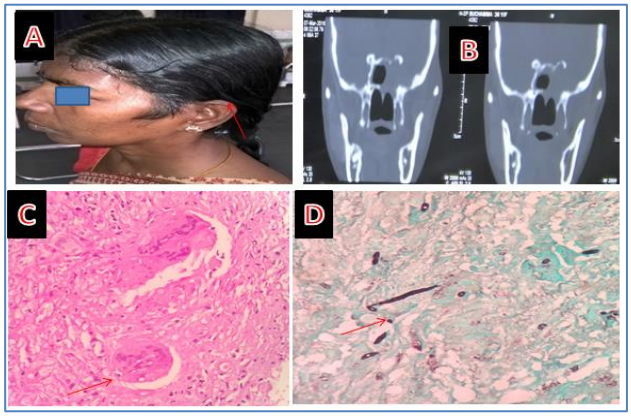

Figure 1: (A) Wasting of left temporal region, (B) CT scan of brain and paranasal sinuses was taken, showing opacification of left sphenoid sinus with erosion of lateral wall of left sphenoid sinus and abutting the left internal carotid artery, (C) H and E staining showed granuloma, and (D) Gomori silver methamine shows multiple filamentous fungi.

Case 2

35 years old female, farmer. Reported to ophthalmology clinic with complaints of protrusion of left eye which was insidious in onset and slowly progressive in nature associated with decreased vision and progressed to complete loss of vision. No other significant history. Examination revealed proptosis of left eye, no perception of light in the left eye, absent movements of the eye ball, ENT examination was normal. Patient was subjected to CT scan of orbits and paranasal sinuses. Endoscopic guided biopsy taken from left nasal cavity. Histopathological examination revealed chronic granulomatous lesion. On special staining with Gomori silver methamine (GMS) staining, non-segmented filamentous fungus probably of family mucoraceae was isolated and then diagnosed as invasive fungal granuloma, negative for AFB stain was suggestive of chronic granulomatous lesion and was subjected to GMS staining and fungus was isolated and then diagnosed as invasive fungal granuloma.

Later patient underwent endoscopic sinus surgery and granuloma was removed in bits and pieces, she was on injection liposomal amphotericin B 3mg/kg 5 days in a week for 6 weeks, with monitoring of serum electrolytes, liver function tests, she developed asymptomatic hypokalaemia for which she was treated with oral syrup of potassium chloride. Patient did not consent for orbital excentration. She is on follow up. Patient is left with no vision and no movements of left eye ball.

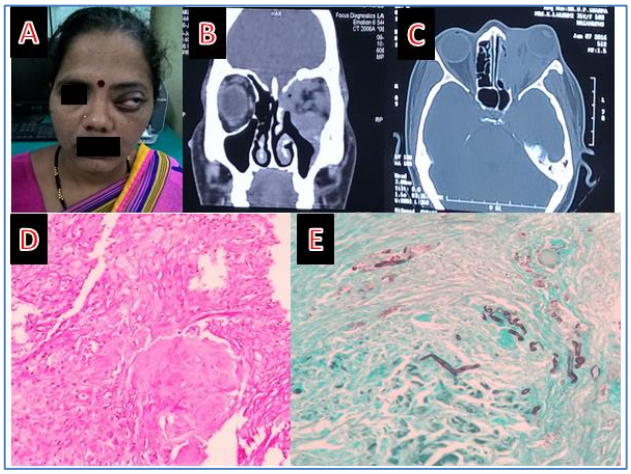

Figure 2: (A) Absent left eye ball movements, (B and C) CT scan of orbits and paranasal was taken, suggestive of opacification of left anterior and posterior ethmoid sinuses, heterogeneous densities in the left orbit with erosion of medial wall and floor of the orbit, and (D and E) H and E staining showing granuloma, GMS staining shows filamentous fungi with septations.

Case 3

55 years old male patient came with complaints of left nasal discharge, protrusion of left eye, no vision in the left eye, he was known case of allergic fungal polyposis. He had history of endoscopic sinus surgery 10 years back. Examination revealed multiple polyps in the right nasal cavity, mucoid discharge in left nasal cavity, no polypoidal changes seen. Eye examination was suggestive of protrusion, chemosis and no perception of

light in the left eye. Patient was subjected to CT orbits and para nasal sinuses. CT shows opacification of right anterior and posterior ethmoiditis, heterogeneous opacification of left anterior and posterior ethmoids, left orbit and erosion of floor, medial and lateral wall of the orbit.

Endoscopic guided biopsy was taken from right and left nasal cavity. Histopathology was suggestive of inflammatory polyposis of right nasal cavity and chronic granulomatous lesion with fungal elements on left side, then diagnosed as invasive fungal granuloma on left side and allergic fungal polyposis on the right side.

Later patient underwent endoscopic sinus surgery and granuloma was removed in bits and pieces, he was on injection liposomal amphotericin B 3mg/kg 5 days in a week for 6 weeks, with monitoring of serum electrolytes, liver function tests. Patient developed complete prolapse of the left eye, orbital excentration was done. Now patient is on follow up.

Figure 3: (A) Left eye proptosis and (B) suggestive of opacification of right anterior and posterior ethmoiditis, heterogeneous opacification of left anterior and posterior ethmoids, left orbit and erosion of floor, medial and lateral wall of the orbit.

DISCUSSION

The idea of reporting these cases are varied presentation of chronic invasive fungal sinusitis, in immunocompetent hosts which needs early diagnosis and prolonged treatment and follow up. Patients often present with nonspecific symptoms and have an indolent clinical course. Therefore, they are usually associated with delayed diagnosis, which may increase the morbidity as well as mortality.[4] In contrast the presentation of acute invasive fungal sinusitis is seen in immunocompromised hosts and diagnosis and treatment is usually straight forward even though the prognosis is poor.

Chronic fungal sinusitis has been rendered difficult by the infrequency of the infection, the prolonged follow up necessary to understand the natural history of the disease. Although many features about prognosis and management of chronic invasive fungal sinusitis currently available but pathogenesis of invasive fungal sinusitis in immunocompetent patient is poorly understood.[5]

Regarding our first case, which is diagnosed as invasive fungal granuloma of left sphenoid sinus with maxillary nerve involvement in otherwise healthy middle-aged female, isolated sphenoid sinus involvement of invasive fungal granuloma is not reported in the literature. It was a diagnostic challenge as the clinical presentation of the patient was only numbness of the left half of face, surgical challenge was that the granuloma involved the lateral wall of the sphenoid, given its proximity to carotid and optic nerve complete clearance of the disease was not possible. However, she showed improvement with injection amphotericin-B.

2nd case the presentation was late after decreased vision in the left eye, it was purely lack of clinical knowledge regarding the presentation of the chronic invasive fungal sinusitis as eye pain being the initial symptom in this case. Early diagnosis would have saved the vision in this patient.

3rd case was interesting case with recurrent allergic fungal polyposis on one side and chronic invasive fungal granuloma on the other side in both the diseases the treatment modalities are different where in allergic fungal polyposis treatment of choice is surgery and steroids, where as in invasive fungal granuloma dual modality approach with systemic antifungals and surgery is treatment of choice. Early diagnosis would have saved the vision in this patient prevented the morbidity.

CONCLUSION

Histopathology is the gold standard for diagnosis of invasive fungal granuloma. Though routine H and E staining may prove inadequate, special stains such as the GMS should be employed in the slightest doubt of a fungal aetiology. CT and MRI assist in diagnosis by outlining the disease extent. Rhinologists should be aware of varied presentations of the CIFG in immunocompetent patients, early diagnosis and multidisciplinary approach is the key towards the disease outcome.

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REFERENCES

1. Shazo RD, Chapin K, Swain RE. Fungal sinusitis. New England J Med. 1997;337(4):254-9.
2. Chakrabarti, Sharma SC, Chander J. Epidemiology and pathogenesis of paranasal sinus mycoses. Otolazyngol Head Neck Surg. 1992;107:745-50.
3. Chakrabarti A, Denning DW, Ferguson BJ, Ponikau J, Buzina W, Kita H, et al. Fungal rhinosinusitis: a categorization and definitional schema addressing

current controversies. Laryngoscope. 2009;119(9):1809-18.
4. Chang C, Gershwin ME, Thompson GR. Fungal disease of the nose and sinuses: an updated overview. Current Allergy Asthma Reports. 2013;13(2):152-61.
5. Bashburn RG, Kennedy DW, Begley MG, Henderson DK, Bennett JE. Chronic fungal sinusitis in apparently normal hosts. Medicine. 1988;67:231-47.

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