CASE REPORT

Isolated Orbital Aspergillosis in an Immunocompetent Patient

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Abstract

A 30-year-old immunocompetent male presented with progressive painless proptosis with double vision and no visual loss. The patient had a mass lesion in the lateral part of the orbit infiltrating the lateral rectus on computed tomography. Lateral orbitotomy was done and the mass was biopsied revealing it to be aspergillosis. The patient was put on intravenous amphotericin B but did not tolerate it and was therefore started on voriconazole. There was a complete resolution of the symptoms with voriconazole.

Keywords: Orbital, aspergillosis, voriconazole, immunocompetent.

INTRODUCTION

Isolated invasive orbital aspergillosis in immunocompetent healthy individuals is an uncommon entity with no cases reported in the English literature so far. Most of the reported cases are secondary to the involvement of the paranasal sinuses.[1-3] The commonest presenting symptom is painless progressive proptosis and is associated with high degree of morbidity and mortality. We here present one such case that presented to us with isolated orbital invasive aspergillosis in our institute.

CASE REPORT

A 30-years-old male presented to the out patient department of Fortis Hospital, Mohali, Punjab, India with the chief complaints of painless and progressive forward protrusion of the right eyeball of 8 months duration. The protrusion of the eyeball was associated with double vision of 2 months duration in lateral gaze. There was no visual deterioration. There were no associated nasal complaints in the form of obstruction, epistaxis, nasal discharge, and anosmia. The examination revealed mild deviation of the nasal septum towards right side with normal appearing middle meatal mucosa. There was axial proptosis with restricted extraocular movements in the lateral direction. The visual acuity was normal. Fundus examination was normal. The patient was subjected to contrast enhanced computed tomography (CECT) of the paranasal sinuses which showed minimally enhancing lesion in the intraconal portion of the right orbit lateral to the optic nerve and infiltrating lateral rectus. It was extending to the superior orbital fissure and the orbital apex region with no intracranial extension (Fig. 1). The patient was subjected to biopsy by lateral orbitotomy approach as there was no sinus component of the lesion. The histopathology revealed it to be invasive aspergillosis. The patient was subjected to a battery of tests for his immunological status. He was negative for viral markers for HIV, HBsAg, HCV and was not found to be diabetic. Nitrozolium blue test was negative. His CD4 count was detected to be on the lower side of normal but was found to

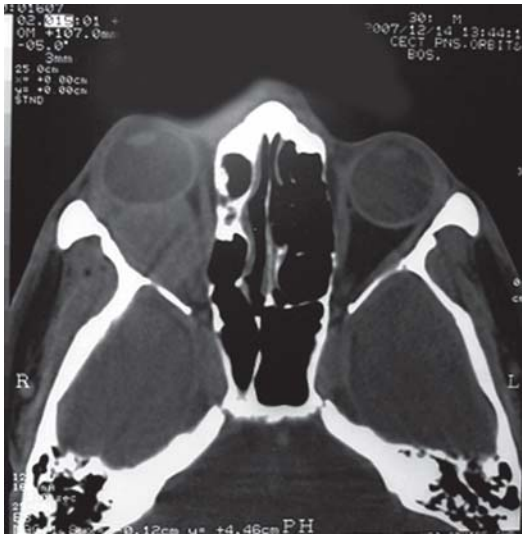

Figure 1: Axial section of the orbit showing lesion in the lateral part of the orbit displacing optic nerve and infiltrating lateral rectus

be idiopathic. Fungal serology was positive for Aspergillus fumigatus. The patient had a normal vision and there was no intracranial extension of the lesion hence was subjected to intravenous conventional amphotericin in a dosage of 1 mg/kg/day. The proptosis started to respond but the patient started having derangements of the renal parameters so he could only be given a cumulative dosage of 1 gm. He was evaluated with the help of radiology (CECT) showing good response (Fig. 2). There was total resolution of diplopia and marked improvement in proptosis. Then he was subjected to oral voriconazole in a dosage of 6 to 8 mg/kg/day for a period of 6 months and there was total resolution of proptosis. There were no major adverse effects of oral voriconazole. Radiology revealed lesion in the same location

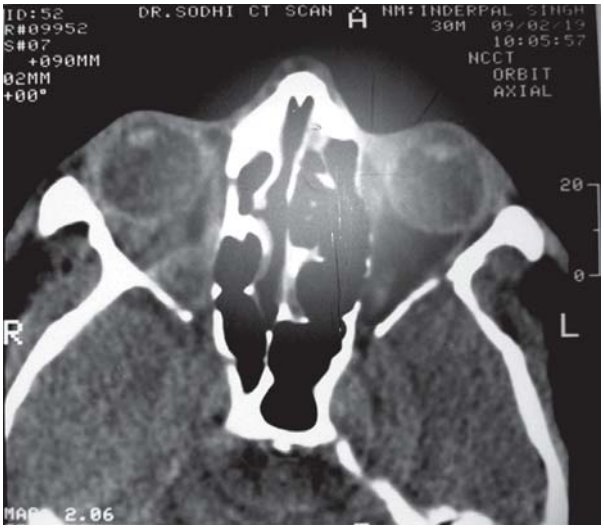

Figure 2: Axial section of the orbit showing lesion in the lateral part of the orbit displacing optic nerve and infiltrating lateral rectus with reduction in the lesion following amphotericin B

though markedly reduced in the size (Fig. 3). The fungal serology was repeated that showed reduction in the value by more than four fold. The patient is still on follow-up and after two years of therapy is disease free.

DISCUSSION

Invasive aspergillosis is a well-known entity in immunocompromised patients, with the primary risk factors being neutrophil defects and corticosteroid use.[4]

Other predisposing factors include HIV infection, diabetes mellitus, use of prosthetic devices or trauma, hematological malignancies.[4] Rarely has invasive aspergillus infection been described in immunocompetent patients.[5,6]

The diagnosis is established by histopathology. Mauriello et al,[7] Austin et al,[8] and Heier et al[9] reported patients that required a second biopsy, and there are cases where the diagnosis was made at autopsy.[4,10,11] If diagnosis is not made on first biopsy and suspicion is high for the fungal pathology, a second biopsy should be performed. The diagnosis was established after biopsy in our patient.

Fungal serology is a useful aid in establishing the diagnosis but it is positive in only 60% of the cases. It was found to be positive in our case. The main role lies in the follow-up of these cases when the biopsy and the radiology may be inconclusive but a raised titer four times or more makes it strongly suspicious for either the residual or the recurrent lesion. Radiological findings were typical for the invasive fungal sinusitis.

The disease spreads to adjacent structures like cranial cavity through focal bony erosion or even through vessel walls, causing compromise of the function in the form of double vision or loss of vision as was seen in our case and

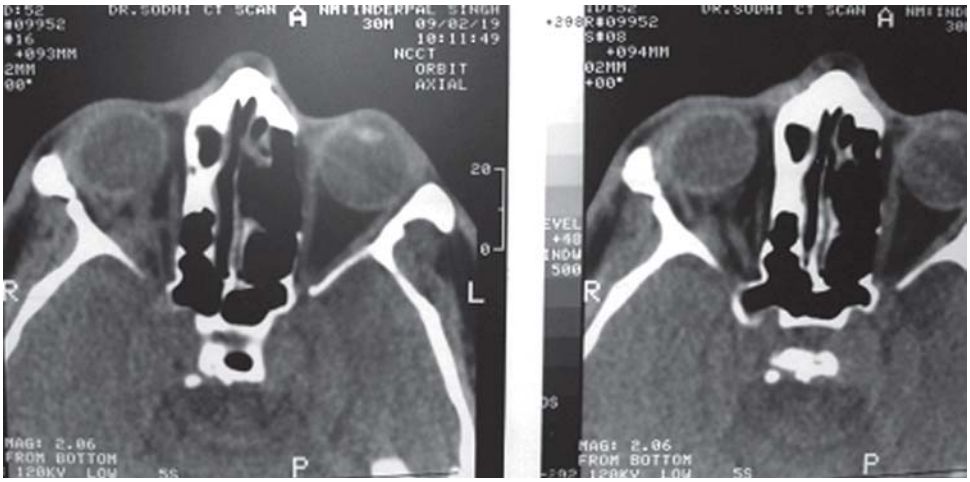

Figure 3: Axial section of the orbit showing lesion in the lateral part of the orbit displacing optic nerve and infiltrating lateral rectus with marked reduction following voriconazole

is in accordance with the literature. The prognosis of invasive orbital aspergillosis is significantly worse than the noninvasive forms of sinus aspergillosis,[12,13] likely because of penetration of bone and blood vessel walls, which often cannot be eradicated by surgery given the anatomic location where drug penetration may also be worse. So far the literature mentions a survival rate of around 30%.

The treatment modalities for invasive fungal sinusitis have been desribed by many authors.[13-18] There is no definitive treatment protocol described for such clinical entity. We, in view of the normal vision and no intracranial extension considered a conservative management in the form of intravenous amphotericin followed by oral voriconazole in this case and had total clinical response suggested by resolution of the symptoms and radiological reduction in the lesion size with more than four fold decrease in the values of fungal serology. This case also highlights the efficacy of oral voriconazole in the cases of invasive aspergillosis.

REFERENCES

1. Bradley SF, McGuire NM, Kauffman CA. Sino-orbital and cerebral aspergillosis: Cure with medical therapy. Mykosen 1987;30:379-85.
2. Green WR, Font RL, Zimmerman LE. Aspergillosis of the orbit. Arch Ophthalmol 1969;82:302-13.
3. Yu VL, Wagner GE, Shadomy S. Sino-orbital aspergillosis treated with combination antifungal therapy. Successful therapy after failure with amphotericin B and surgery. JAMA 1980;244: 814-15.
4. Levin LA, Avery R, Shore J, et al. The spectrum of orbital aspergillosis: A clinicopathological review. Surv Ophthalmol 1996;41:142-54.
5. Gupta AK, Ghosh S, Gupta AK. Sinonasal aspergillosis in immunocompetent Indian children: An eight-year experience. Mycoses 2003;46(11-12):455-61.
6. Gupta AK, Gupta AK. Postgraduate institute management protocol for the management of aspergillus flavus sinusitis: Is it effective? Int J Inf Dis 2009;13(2):134-39.
7. Mauriello JA, Yepez N, Mostafavi R, et al. Invasive rhinosino-orbital aspergillosis with precipitous visual loss. Can J Ophthalmol 1995;30:124-30.
8. Austin P, Dekker A, Kennerdell JS. Orbital aspergillosis. Report of a case diagnosed by fine needle aspiration biopsy. Acta Cytol 1983;27:166-69.
9. Heier JS, Gardner TA, Hawes MJ, et al. Proptosis as the initial presentation of fungal sinusitis in immunocompetent patients. Ophthalmology 1995;102:713-17.
10. Lowe J, Bradley J. Cerebral and orbital aspergillus infection due to invasive aspergillosis of ethmoid sinus. J Clin Pathol 1986;39:774-78.
11. Suzuki K, Iwabuchi N, Kuramochi S, et al. Aspergillus aneurysm of the middle cerebral artery causing a fatal subarachnoid hemorrhage. Intern Med 1995;34:550-53.
12. Denning DW, Hanson LH, Perlman AM, et al. In vitro susceptibility and synergy studies of aspergillus species to conventional and new agents. Diagn Microbiol Infect Dis 1992;15:21-34.
13. Denning DW. Aspergillus species. In: Mandell GL, Bennett JE, Dolin R (Eds). Principles and Practice of Infectious Diseases (5th ed). Philadelphia: Churchill Livingstone 2000;2:2674-85.
14. Massry GG, Hornblass A, Harrison W. Itraconazole in the treatment of orbital aspergillosis. Ophthalmology 1996; 103:1467-70.
15. Streppel M, Bachmann G, Damm M, et al. Successful treatment of an invasive aspergillosis of the skull base and paranasal sinuses with liposomal amphotericin B and itraconazole. Ann Otol Rhinol Laryngol 1999;108:205-07.
16. Schneemann M, Schaffner A. Host defense mechanisms in aspergillus fumigatus infections. Contrib Microbiol 1999;2: 57-68.
17. Weber RS, Lopez-Berestein G. Treatment of invasive Aspergillus sinusitis with liposomal-amphotericin B. Laryngoscope 1987;97:937-41.
18. Stevens DA, Kan VL, Judson MA. Practice guidelines for disease caused by aspergillus. Clin Infect Dis 2000;30:696-709.