INVASIVE ASPERGILLOSIS INVOLVING MULTIPLE PARANASAL SINUSES – A CASE REPORT

*S Agarwal, A Kanga, V Sharma, DR Sharma, ML Sharma

Abstract

A case of invasive multiple paranasal sinus aspergillosis with bony involvement is reported. A young immunocompetent lady presented with bilateral nasal obstruction due to polyps. Radiologically and histopathologically a fungal cause was kept a possibility, and the diagnosis of Aspergillus fumigatus was established by demonstration of acute angle branching septate hyphae on direct wet mount and repeated isolation in culture. Patient responded favourably to surgical excision of polyps and oral itraconazole post operatively.

Key words: Aspergillus, aspergillosis, paranasal sinuses

Aspergillus spp. rank closely behind Candida spp. in causing invasive fungal infections in humans. Primary lesions can be localized in the eyes, paranasal sinuses, external ears and larynx in apparently healthy individuals.1 There are more than 185 species of Aspergillus and over 95% of all infections are caused by A. fumigatus, A. flavus and A. niger. A. fumigatus alone accounts for the large majority of both invasive and noninvasive aspergillosis.2 Paranasal sinus aspergillosis includes a spectrum of disease classically described to be four types: allergic, noninvasive, invasive and fulminant.3 Exposure to Aspergillus spp. is universal but clinical syndrome and pathologic spectrum depends on underlying lung architecture, host’s immune response and degree of inoculum. The nose and paranasal sinuses have local factors which promote fungal infection including nasal polyps, recurrent bacterial infections, neutropenia and chronic rhinitis with stagnation of nasal secretions. Other underlying factors include prolonged antibiotic therapy, immunosuppressive drugs and corticosteroids.2 Acute Aspergillus sinusitis is a life threatening condition encountered in immunocompromised patients. Though decreased immune response has been described as of paramount importance in aspergillosis, review of literature shows that cases of invasive and semi-invasive aspergillosis in non-neutropenic patients have occurred.4 Any type of paranasal aspergillosis may progress to or be associated with more aggressive disease illustrating the importance of early recognition of this increasingly encountered disease.

Case Report

A 28 year old female presented with two month history of nasal obstruction and bilateral swelling medial to medial canthus of the eyes in August 2004 in the department of Otorhinolaryngology. She had complaints of repeated episodes of nasal obstruction for past many years with regression of symptoms temporarily. Onset was insidious starting on right side and gradual progression to bilateral total nasal blockside. Patient subsequently developed breathing through mouth and bilateral watering of eyes more on the right side. She complained of scanty seropurulent, odourless nasal discharge. She denied epistaxis, excessive sneezing, headache, paranasal discharge or trauma to nose. History of recurrent upper respiratory infections including fever, cough, sputum production, ear discharge, earache, tinnitus, vertigo, ataxia and asymmetry of face or facial pain were negative. She had no history of tuberculosis, diabetes mellitus asthma, use of corticosteroids, other immunosuppressants or prolonged antibiotic therapy.

On general physical examination, she was afebrile with a blood pressure of 124/80 mm of Hg and pulse rate of 95 per minute. All other vital parameters were normal. Anterior rhinoscopy of revealed bilateral pale white nasal polyps. Polyps were non-tender and probe could be moved all around the growth. Nasal mucosa was normal with no bleed on touch. Bilateral nares blockage was assessed by patency test. There was no abnormal finding on posterior rhinoscopy. Irregular, tender swellings were palpable on both sides medial to medial canthus of eyes with upward extension on the upper eyelids. Ethmoidal sinus was also tender. Clinical examination of ears and throat was normal.

Laboratory values were notable for haemoglobin of 12 gm%. The white blood cell count was 10.8 x 10^9/L with 69% segmented neutrophils, 25% lymphocytes, 4% monocytes, 2% eosinphils and no basophils. Random blood glucose of 107 mg% and fasting level of 59 mg% were recorded. An HIV test was done as there was no clinical suspicion that the patient was immunocompromised.

*Corresponding author
Department of Microbiology (SA, AK, VS) and Department of Otorhinolaryngology (DRS, MLS), Indira Gandhi Medical College, Shimla, Himachal Pradesh - 171 001, India.
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Plain, contrast and high resolution CT scan for nasal sinus showed soft tissue density masses in maxillary, ethmoid and sphenoid sinuses. There was bony destruction of medial wall of right maxillary antrum with high density probably calcareous deposition. Attenuation of nasal septum, ethmoid trabeculae and uncinate process were observed. Identification of infundibulum, laminae papyraceae and right orbit were also present.

 Provisional diagnosis of sinonasal polyposis possibly due to fungal cause with infiltration into nasal cavity and right orbit was made. Patient was started on roxithromycin 300mg per day and advised “functional endoscopic sinus surgery”. During surgery, multiple nasal polyps coming out of right maxillary sinus opening and middle meatus on the left side were excised. The lesion yielded a characteristic caseous cheesy material coming from the maxillary sinus which was subsequently curedtted. She was started on gatifloxacin. Excised tissue was received for wet mount preparation and culture in sterile saline. Microscopic examination of the wet mount preparation revealed necrotic tissue and fungal hyphae measuring 3mm to 6mm in diameter. Hyphae were septate, hyaline and showing branching at acute angles.

Culture on Sabouraud dextrose agar (SDA) with chloramphenicol were incubated at 37 °C and 25 °C. On day three of incubation, few white fluffy colonies with yellow to green centre were seen on the obverse with a yellow pigment on the reverse. Colonies turned grayish green by day ten. Lactophenol cotton blue wet mount prepared from the fungal growth demonstrated septate hyphae from which nonseptate short smooth conidiophores terminating in vesicle. The features were consistent with the diagnosis of Aspergillus fumigatus.5

Histopathological examination of the fibrinopurulent mass revealed abundant septate fungal hyphae. Three days hence patient was kept for endoscopic examination and biopsy was taken. Subsequent culture grew A. fumigatus on day eighth of incubation. Histopathological examination showed necrosis, acute inflammatory cell infiltration and inflammatory cell infiltration and inflammatory granulation tissue. Fungal hyphae were not seen on routine haematoxylin and eosin staining and Grocott silver staining could not be performed. Patient was started on oral itraconazole 400 mg/day. She was reviewed for endoscopic examination after one month. Assessment revealed few bilateral small nasal polyps which were removed from ethmoid gallery and sent for...
histopathological examination. Antifungal therapy was continued and patient showed dramatic improvement.

Discussion

Since first reported by Katzenstein and her colleagues in 1983 there have been few further reports of allergic Aspergillus sinusitis (AAS). The condition is still poorly recognized by both clinicians and pathologists with diagnosis and characterization continuing to be controversial. AAS is considered essentially a saprophytic condition with a benign course. Interestingly, however, Dunlop and Bilson described a case that resulted in visual failure due to orbital involvement. Thus a spectrum of disease severity beginning with benign AAS can be demonstrated. Despite such a capacity for destruction, tissue invasion by Aspergillus has not yet been described in this condition. Sinusitis due to Aspergillus may present in two different forms. The acute life threatening form is seen in immunocompromised patients. Paranasal sinus granuloma formation is a slowly progressive condition as seen in the present case. The typical patient of allergic fungal sinusitis is an immunocompetent, atopic, young adult with a long standing history of allergic rhinitis, nasal congestion, headache, polyposis, asthma and/or recurrent sinusitis. The mean age group of 34.6 +/- 14 years has been reported by Chakrabarti et al from Chandigarh. There is a male preponderance with male to female ration ranging from 1.5:1 to 3:1. On examination, polyps may be evident with associated mucopurulent discharge, enlarged turbinates, hyperplastic nasal mucosa and occasional facial pain.

CT scan has a role in demonstrating extent of lesion and bone destruction. Usually, there is involvement of posterior sphenoid, ethmoid septa, laminarPapyracea and medial antral wall. Bony involvement is reported in 30 to 50% cases. at surgery apart from the polyps, the most striking feature is the thick green- brown to yellow inspissated necrotic or gelatinous wall. Bony involvement is reported in 30 to 50% cases.2 1.5:1 to 3:1.2,6 On examination, polyps may be evident with associated mucopurulent discharge, enlarged turbinates, hyperplastic nasal mucosa and occasional facial pain.

The usual fungus isolated is Aspergillus; other cause being phaeoid fungi like Bipolaris, Exserohilum, Curvularia and Alternaria. Demonstration of Aspergillus spp. by both culture and microscopic examination of tissue provides the most firm diagnosis. The appearance of fungal hyphae on histopathological examination, though highly suggestive, is not always seen on routine haematoxylin and eosin stains. The positive results of wet mounts and repeated isolation on culture confirms the diagnosis of Aspergillus beyond doubt in this case. Reports of histopathology are supportive evidence. The particular Aspergillus spp. causing infection may depend on geographical location. Case reports of paranasal sinus mycoses from India and Sudan have reported Aspergillus flavus as the cause of majority of cases.6-8 The isolate in our case is A. fumigatus which is rarer. The differential diagnosis of allergic bronchopulmonary aspergillosis could be set aside by the absence of pyrexia, recurrent wheezing bronchitis, sputum production and blood eosinophilia. Our patient was clinically reviewed by endoscopic visualization and continued on oral itraconazole 400mg/day for ten days after which the patient became symptom free. In conclusion, we emphasize that awareness regarding the possibility of invasive paranasal sinus aspergillosis is required when examining patients with nasal polyposis in healthy immunocompetent young adults.

References