Management of Maxillary Aspergillosis in a Patient with Diabetic Mellitus Followed by Prosthetic Rehabilitation

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Abstract

Background Mycotic infection of the paranasal sinuses and Maxilla are on the rise globally. Since 1968 the number of reported cases has increased three fold of all the fungal infections. Aspergillosis is one of the most rapidly progressing and lethal form of fungal infection. Among all fungal infections, Aspergillosis is the commonest fungal infection in human beings especially in orofacial region. Its early medical and surgical treatment can improve survival. We report a case of invasive Aspergillosis of maxilla in a patient with diabetes mellitus who was managed by systemic as well local antifungal therapy, along with surgical debridement and followed by prosthetic rehabilitation.

Keywords Fungal infection · Diabetes · Paranasal sinuses · Invasive Aspergillosis

Introduction

Aspergillosis is one of the most rapidly progressing and lethal form of fungal infection. This fungus invades the

arteries, forms thrombi within the blood vessels that reduces the blood supply and causes necrosis of hard and soft tissues. Thrombosis of the internal maxillary artery or descending palatine artery from a fungal infection could result in necrosis of a portion of the maxilla [3]. Once entered in arteries, fungus can spread to orbital and intracranial structures [4]. In the head and neck, involvement of the sinuses and orbits is common, often in association with fever, cellulitis, serosanguinous drainage, and proptosis [5]. In its invasive form, Aspergillosis spreads by osseous erosion or vascular invasion. Conditions that favour fungal infections are diabetes, long term antibiotic and steroid treatment, radio, chemotherapy, immunosuppressive treatments, and immunodeficient diseases [6]. Many authors suggest that mycotic infections are found more commonly in apparently healthy patients. We classify this case as a jaw cavity mycetoma which is the most frequent form of rhino nasal mycosis. Here we report a case of invasive Aspergillosis of facial region with necrosed maxilla in a patient with diabetes mellitus who was managed by systemic as well local antifungal therapy, along with surgical debridement.

Case Report

A 70-year-old male known case of diabetes, reported to Department of Oral and Maxillofacial Surgery, Rural Dental College, Loni for evaluation of pain in left maxillary posterior region since last 6 months, patient also complains of nasal congestion, headache and numbness on upper lip region since last 2 months. In significant history patient under went extraction of upper and lower teeth before 1.5 years. After that the patient remained edentulous without any prosthesis.

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Fig. 1 Preoperative view showing necrotic maxilla and palate

Intraoral examination showed edentulous necrotic bone involving hard palate and upper alveolus completely on left side and up to premolar region on right side measuring approximately 8 cm in length. Even some perforations were also present in palate (Fig. 1). Radiologic examination revealed obliteration of left maxillary sinus showing involvement of left pre-maxilla region with extension into left maxillary sinus and left Zygomatic region. Lateral wall of left side maxillary sinus was completely eroded. The radiologic appearance of the paranasal sinus is a focal radio dense shadow in the sinus (Fig. 2) [7]. In the lights of CT above findings were confirmed. In addition to paranasal sinuses, other sites of infection with in head and neck region includes the ear and orbit but rarely, in our case also ear and orbit was unaffected.

Hard tissue specimen along with adjacent soft tissue was incised under local anesthesia and sent for histopathology. The clinical and radiological diagnosis was invasive fungal infection and the same was confirmed as Aspergillosis in histology (Fig. 3). Aspergillus is recognized by its septate hyphae, which branches at 45 degree angle. The invasiveness of fungi becomes most lethal once the hyphae entered blood vessels, where thrombi are formed [7, 8]. Same fungal branching and septate hyphae with branching at an acute angle were noticed in our case.

Patient sugar levels were controlled and the patient was operated for surgical debridement and curettage of necrotic bone. Whole of the necrotic palate was detached from surrounding attachments and removed. Curettage was done up to left Zygomatic arch region, lining of the sinus was removed completely, even some part of

Fig. 2 PNS showing obliteration of left maxillary sinus

Fig. 3 Histology examination shows fungal hyphae are noticed at acute angles

Zygomatic bone was curetted out (Fig. 4). The operated area was irrigated with combination of Amphotericin powder and normal saline. Patient was kept on injection Amphotericin B 25 mg in 500 cc of saline twice along with antibiotics for next 5 days and the blood urea and creatinine levels were monitored regularly. From 6th post

Fig. 4 Immediate post operative view after removal of necrotic bone

Fig. 6 Obturator cum prosthesis

Fig. 5 Follow up picture showing wound dehiscence

Fig. 7 Patient rehabilitated with obturator cum prosthesis

operative day patient was shifted to tablet Fluconazole 150 mg OD. At the same time operated site was irrigated twice daily with normal saline mixed with Amphotericin powder.

In the Post operative period, dehiscence was noticed because of that a communication was formed between oral cavity and maxillary sinus (Fig. 5). Patient was kept on

regular follow up; dressing was carried out regularly by Neosporin packs in the defect for next 3 months. After 4 months the biopsy from the previously operated site was taken to find out any remnants or reoccurrence of disease.

After 6 months oral rehabilitation was done with obturator cum complete denture prosthesis to improve quality of patient's life (Figs. 6, 7).

Discussion

Priest botanist Micheli first described Aspergillosis in 1729 [9]. In 1847 Sluyter first identified Aspergillus in a woman dying of a pulmonary infection. Zarinko, in 1885 described Aspergillus infection of the maxillary sinus. First report of Aspergillosis involving maxilla was by Morrel Mackenzie in 1893. Hora differentiated Aspergillus as infiltrating and non-infiltrating with incidence of bone invasion as the primary difference [10]. The invasiveness of fungi becomes most lethal once the hyphae enter blood vessels, where thrombi are formed [11, 12]. Aspergillus is a fungus that is commonly found in the atmosphere and is often the blue-green mold found on bread [13]. Inhalation of the Aspergillus spores can lead to colonization in upper and lower respiratory tracts, allergic response or invasiveness destructiveness. There are several species of the fungi but, Aspergillus fumigatus and Aspergillus flavus being most dominant [8]. Aspergillus flavus is considered to be the most destructive in the paranasal sinuses and oral cavity because of its potent toxin producing abilities [14]. Local factors, such as sinus obstruction, may promote growth of Aspergillus species, also favoured by hypoxia and anaerobic conditions. Beck-Mannagetta et al. indicated that maxillary sinus Aspergillosis can be induced by the presence of endodontic filling material like zinc oxide. The fungus itself does not contain any chlorophyll and therefore does require any light for growth. It does, however need a host supplying it with nutrient such as glucose, nitrogen, sulphur, phosphorous, potassium, calcium, magnesium and iron. The pattern in a culture medium is always circular because of centrifugal linear growth unless inhibited by natural or artificial barriers. This is why Aspergillus in the paranasal sinus eventually develops into a ball-shaped mass [14]. The centre of the mass contains calcium phosphate and calcium phosphate and therefore mimics a foreign body on radiography [7]. In our case dense radio opaque mass was evident in radiographs.

The fungal infection is more common in immunocompromised patients, patients on steroid therapy, leukemic and lymphoma patients, patients with chronic sinusitis, prosthetic devices, and chronic drinkers. Literature shows patients suffering from long standing diabetes mellitus are the main victims [15]. Our patients was also suffering from diabetes mellitus for a long period. Uncontrolled diabetes mellitus can alter the normal immunologic response of patients to infections. Such patients have decreased granulocyte phagocytic ability with altered polymorphonuclear leucocyte response [4].

As the fungal infection is very rapidly spreading disease, so it's early and proper management is must. Initial improvements with medical therapy not obviate the need for surgical care. Surgical management along with

systemic antifungal therapy is the choice of treatment mentioned in most of the literature. But in our case along with the above mentioned methods we also used antifungal drugs locally which proved to be beneficial.

Comments

Successful treatment of Aspergillosis consists of aggressive surgical debridement of necrotic tissue, systemic antifungal therapy and control of any underlying disease process. The same criterion was followed by us in our case report. First uncontrolled diabetes was managed, followed by surgical debridement of affected area along with systemic antifungal therapy. In our case irrigation of operated area was done with amphotericin mixed with normal saline which we thought was very helpful in recovery.

Since last 1 year patient is on regular follow up with controlled diabetes. Our patient was also rehabilitated with prosthesis 6 months after the operative procedures which provided him a good quality of life.

References

1. Mc Gill TJ, Simpson G, Healy GB (1980) *Fulminant Aspergillosis* of the nose and paranasal sinuses: a new clinical entity. *Laryngoscope* 90:748–754
2. Matjaz R, Jernej P, Mirela K-R (2004) Sinus maxillaris mycetomas of odontogenic origin: case report. *Braz Den J* 15(3):248–250
3. Pogrel MA, Miller CE (2003) A case of maxillary necrosis. *J Oral Maxillofac Surg* 61:489–493
4. Auluck A (2007) Maxillary necrosis by mucormycosis. A case report and literature review. *Med Oral Patol Oral Cir Bucal* 12:E360–E364
5. Tugsel Z, Sezer B, Akalin T (2004) Facial swelling and palatal ulceration in a diabetic patient. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 98:630–636
6. Ribeiro NFF, Cousin GCS, Wilson GE, Butterworth DM, Woodward RTM (2001) Lethal invasive mucormycosis: case report and recommendations for treatment. *Int J Oral Maxillofac Surg* 30:156–159
7. Ogata Y, Okinaka Y, Takahashi M (1997) Antrolith associated with Aspergillosis of the maxillary sinus: report of a case. *J Oral Maxillofac Surg* 55:1339–1341
8. Napoli JA, Oliver J (1991) Aspergillosis and necrosis of the maxilla: a case report. *J Oral Maxillofac Surg* 49:532–534
9. Robb PJ (1986) Aspergillosis of the paranasal sinuses: a case report and historical perspective. *J Laryngol Otol* 100:1071–1077
10. Titche LL (1978) Aspergillosis of the maxillary sinus. *Ear Nose Throat J* 57:62–66
11. Singer MI, House JL (1979) Invasive mycotic infections in the immunocompromised child. *Otolaryngol Head Neck Surg* 87:32–34
12. Erlichman Medical Center, Trieger N (1978) Aspergillus infection in a patient receiving immunosuppressive drugs. *J Oral Surg* 36:978–981

13. Shanon MT, Sclaroff A, Colm SJ (1990) Invasive Aspergillosis of the maxilla in an immunocompromised patient. *Oral Surg Oral Med Oral Pathol* 79:425–427
14. Rossouw DP, Swart JG (1988) *Aspergillus fumigatus* infection of the maxillary sinus. *S Afr Med J* 73:47–78
15. Sharda DM, Arunkumar G, Vandana KE, Rao PS (2006) Sino-orbital Aspergillosis in a diabetic patient. *Indian J Med Microbiol* 24:4–138