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An Interesting Case on Rhino-cerebral Mucormycosis

Sehra D*[1], Relia P[1] and Gupta B[1]

[1]Institution Maharaja Agrasen Hospital, New Delhi, India

***Corresponding author:** Dr. Devindra Sehra MD, DTCD. Institution Maharaja Agrasen Hospital, Punjabi Bagh, New Delhi, India, Email: sehradev@yahoo.com

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Abstract

Rhinocerebral Mucormycosis (RCM) is a rapidly progressing infection seen frequently in patients with malnutrition, uncontrolled diabetes, or in patients with immunocompromised status like hematological malignancies or who are on immunosuppressive or cytotoxic therapy [1]. The disease manifests initially as sinusitis, and is therefore diagnosed in an advanced stage [2]. We hereby report a rare case of RCM with Candidiasis in a 40 years old female from an urban background who presented with the complaints of unilateral facial pain, orbital swelling, headache and right sided nasal blockage with deviation of mouth towards left side. The biopsy report of the affected lesion revealed mucormycosis with fungal hyphae. This report is to emphasize that in patients who present with unilateral headache, rhinorrhea, rigors, and symptoms suggestive of sinusitis, RCM should be foremost in the differential diagnosis, especially if diabetes exists as a comorbid condition.

Keywords: Rhinocerebral Mucormycosis; Candidiasis; Immunosuppressive; Unilateral facial pain; Orbital swelling; Headache; Right sided nasal blockage

Abbreviations: RCM: Rhinocerebral Mucormycosis.

Introduction

Mucormycosis is a rare and a lethal infection caused by a fungus from the order Mucorales (Mucor, Rhizomucor, Rhizopus, Absidia species) found commonly in soil, vegetation, decaying food and organic matter [3]. The ubiquitous fungus becomes life threatening especially in the immune compromised patients and in those with uncontrolled blood sugar levels. Depending on the tissues involved, mucormycosis has been categorised in several clinical forms such as rhinocerebral (further characterized as rhino-orbital, rhinomaxillary,and rhino-orbitocerebral), pulmonary, cutaneous, gastrointestinal and disseminated [4]. Rhinoorbitocerebral is the most common form seen in diabetic patients [5]. Usually patients present with complaints suggestive of sinusitis and are initially treated with antibiotics; but they deteriorate rapidly and generally do not respond to the conventional treatment [2]. RCM is diagnosed by clinical presentation, signs, histological examination and culture reports. We hereby report a case of 40 years uncontrolled diabetic patient who presented with the complaints of unilateral facial pain, orbital swelling, headache and right sided nasal blockage with deviation of mouth towards left side, who was promptly diagnosed but could not be saved inspite of best efforts due to late presentation to the hospital.

Case Report

A 40 years housewife from an urban background presented to the emergency department of MAHARAJA AGRASEN HOSPITAL, NEW DELHI with the complaints of headache for 14 days, deviation of mouth towards left and drooling of saliva from mouth. She complained of right sided facial pain with orbital swelling from 12 days and difficulty in speech for 1day. The patient also complained

of severe pain behind the right eyeball with proptosis. There was no history of fever, trauma, vomiting, or loss of consciousness. She gave history of diabetes mellitus for 7-8 years for which she was on insulin therapy. She also gave history of hypertension for which she was on erratic treatment. No other significant history was elicitable apart from a recent operation on left eye for cataract.

On examination, the patient was conscious, well oriented to time, place and person. She was a febrile; Pulse rate was 110/minute. Blood pressure was 110/80 mmhg, and HbA1c was 12.1. Her forehead creases were decreased on the right side with complete ptosis. Right eyelid was edematous and right pupil was fixed and non-reacting to light. No movement of external occular muscles was seen. No perception of light was noted in the affected eye (Figure 1). Left eye was normal.

Right side of the face had decreased sensation to light touch; Drooling of saliva was present from the left angle of mouth. No signs of meningeal irritation were noticed. Right nostril was blocked and when the patient was asked to blow the nose, scaly debris was noticed. On the basis of history and general examination patient was initialy diagnosed as a case of right sided Bell's palsy with sinusitis and orbital cellulitis and she was started on intravenous antibiotics therapy. Patient deteriorated and black eschar formed over the right side of the face with involvement of right orbit. MRI of the face showed inflammation involving subcutaneous tissue of right side of face extending upto right parapharyngeal space and right maxillary sinusitis (Figure 2). Biopsy was taken from the lateral wall of right nasal cavity lesion which showed ribbon like aseptate hyphae suggestive of mucor mycosis and associated candidiasis (Figure 3a,3b,3c). On the basis of the biopsy report, systemic antifungal therapy was started in the form of liposomal amphotericin-B. In view of progressive nature of eschar, surgical debridement and fasicotomy was done and her blood sugar was controlled on insulin therapy. Maxillotomy with partial mandibulectomy of right side of the face was done. Wound pus culture showed the growth of Pseudomonas aeruginosa. In view of culture reports antibiotics were modified. But patient's condition deteriorated and she was put on mechanical ventilation. However inspite of all efforts patients couldn't be revived.

Discussion

Mucormycosis is a potentially fatal infection with a mortality rate of 50-100 % [6]. RCM is most common presentation of the disease. Amongst rhinocerebral mucormycosis 70% of the patients have diabetes mellitus, whereas about 40% of patients of mucormycosis in general are diabetic [7].

After infecting the nasal mucosa or palate, the fungus spreads by direct extension from the sinuses into the rhino-orbital region and by the hematological spread to the angular, lacrimal and ethmoid vessels. The hematological spread of the fungal hyphae and dissemination to cerebrum and lungs can be fatal in immunocompromised hosts. Mucor hyphae form thrombi within the blood vessels causing necrosis of the tissues due to reduced vascularity [8]. Functioning of the cranial nerves II, III, IV and VI is affected and this may result in proptosis, periorbital cellulitis and total visual loss. An irregular black eschar on the palate or nasal mucosa and presence of facial pain is a consistent finding and has been observed in this reported patient also [9].

Any diabetic patient who presents in diabetic ketoacidosis, and exhibits signs suggestive of sinusitis should be assessed for mucormycosis and suspected to have this infection until proved otherwise [10]. This is because the early diagnosis and treatment of the patients is extremely important for the cure of ailment.

In normal hosts macrophages prevent infection by phagocytosis and oxidative killing of spores. In diabetic patients these cells are dysfunctional, have an impaired glutathione pathway and have decreased phagocytic activity. A substance called rhizoferrin is produced by the fungal hyphae. In the serum rhizoferrin binds with the iron at low pH, and an iron-rhizoferrin complex forms which promotes the fungal growth [11]. As ketoacidosis results in release of iron in serum from proteins, the iron-rhizoferrin complex formation is enhanced with subsequent accelerated fungal growth.

Treatment include systemic antifungal medications, radical surgery and treatment of the predisposing diseases. Liposomal amphotericin-B (amphotericin-B colloidal dispersion) is the preferred medication as it is less nephrotoxic. As it is difficult for the antifungal medications to penetrate the affected area adequately, surgical treatment is an additional adjunct. Liposomal amphotericin-B, irrigation, nasal and orbital packing with surgical debridement form the mainstay of the treatment [12].

Conclusion

RCM should be included in the differential diagnosis of all diabetic patients who present in diabetic ketoacidosis with features of sinusitis. It is a rapidly progressing disease with fulminant course. Prognosis depends upon early diagnosis and a multidisciplinary team approach which includes ENT surgeon, diabetologist, ophthalmologist, neurologist and a dental surgeon. Absences of intracranial and orbital extension are the

indicators of good prognosis [2]. Meticulous control of blood sugar, surgical debridement and high dose antifungals are the important factors which leads to decrease in the morbidity and mortility.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing Interests

The authors declare that they have no competing interests.

Figures

Figure 1: Black Eschar formation over the right side of face with the involvement of right orbit.

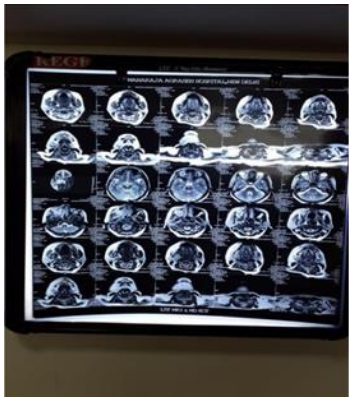

Figure 2: MRI of the face, which showed area of soft tissue inflammation involving subcutaneous tissue of right side of face extending upto right parapharyngeal space and right maxillary sinusitis.

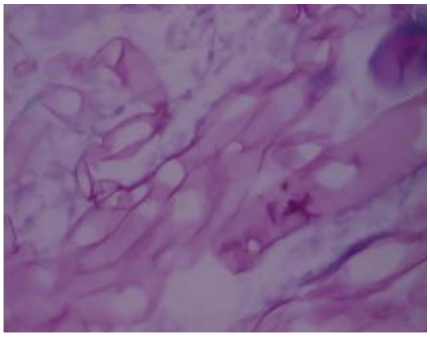

Figure 3a: Ribbon like aseptate hyphae suggestive of mucormycosis

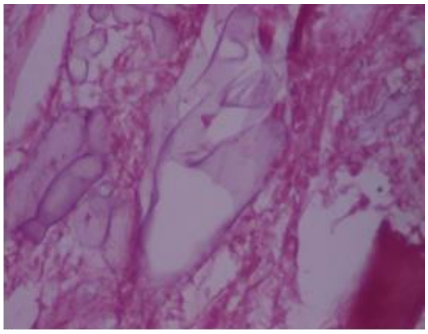

Figure 3b: Ribbon like aseptate hyphae suggestive of mucormycosis.

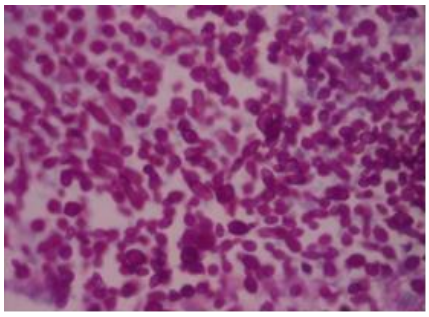

Figure 3c: With associated Candidiasis.

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