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Case Report

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A CASE REPORT ON A RARE CASE OF RHINOCEREBRAL MUCORMYCOSIS IN A FEMALE DIABETIC PATIENT IN A TERTIARY CARE HOSPITAL, KANCHIPURAM

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ABSTRACT

Background: Rhinocerebral mucormycosis is a fatal rapidly progressing fungal infection caused by members of order Mucorales such as Rhizopus, Absidia and Mucor. The disease manifestations sinusitis, oedema of eyelids, consist proptosis, ptosis, ophthalmoplegia, cranial nerve palsy and with the cerebral involvement invariably fatal. Treatment with antifungal agents can only prolong the life expectancy but not curative. In our present case per nasal swab was sent for fungal culture. It was inoculated into SDA. After 24 hours of incubation abundant cottony growth was observed. Microscopically cellophane preparation showed rhizoids, sporangiophore, vesicle,, sporangium and sporangiospores strongly

suggesting RhizopusThe case was clinically followed. Clinical history was that of a female patient 50 years old attending diabetic OP with pain in the left side of face 1 week duration, sudden inability to open the left eyelid, pain around eye associated with double vision more on leftside. One episode of dark coloured nasal discharge 1 week ago. MRI scan report showed space occupying lesion. By correlating clinical, MRI scan findings and microbiological report the case was diagnosed as Rhinocerebralmucor mycosis. The prognosis of rhinocerebral mucormycosis is poor. Present case as it has invaded through paranasal sinuses its course is invariably fatal. This emphasizes effective interaction among clinicians and diagnostic team essentially needed for proper early diagnosis of cases.

KEYWORDS: Mucorales, Rhizopus, Ptosis, **O**phthalmoplegia,, MRI, Rhinocerebral mucormycosis, LAMP

INTRODUCTION

Mucormycosis. [1] is a relatively rare fulminant opportunistic infection caused by widespread, saprophytic thermotolerent fungi notably species Rhizopus, Mucor and Absidia. Humans get infected through air born spores. [2] The incidence is approximately 1.7 per 1,000,000 per year. [3] The fungus has tendency to invade blood vessel wall after successful primary infection. Further results in dissemination of mycotic fungi and formation of metastatic foci. [4,5] Mortality rate is 50% in systemic disease while with cerebral involment it reaches upto 80%. [6] If clinical manifestations are not familiar it may lead to diagnosing and treatment dilemma.^[7] The clinical manifestations begin as sinusitis, progress to local invasion resulting in edema of eyelids, proptosis, malar anaesthesia,, internal, external ophthalmoplegia finally fatal outcome.[8]

CASE REPORT

A female patient, aged 50 years was admitted into our hospital with a history of multiple cranial nerve palsy (3rd, 4th, 5th, 6th, 7th cranial nerve), type2 diabetes mellitus, systemic hypertension. The patient complained of pain over left side of the face followed by dental extraction 20 days back. This was associated with pain in the left ear and difficulty in swallowing and chewing. There was facial puffiness, left eye ptosis and chemosis. Eye abduction restricted, left side LMN type of facial palsy noticed. There was history of nasal discharge from the left nostril, it was brownish in colour, non-fowl smelling. There was history of anosmia. No limb weakness. The patient was drowsy, oriented, afebrile, CVS and RS normal. Bloodpressure -180/100, Pulse rate – 84/minute. Temperature 98°C, Random blood sugar - 536 mg/dl. CNS examination showed following features- speech normal, conjunctival reflex absent on the left side, allodynia on the left side of face, muscles of mastication were weak, uvula central,, pupils dilated, left side ptosis, Rhomberg's sway on closing the eyes observed. Fundus normal, muscle tone normal, power5/5, Deep tendon reflexes positive.

Biochemical investigations

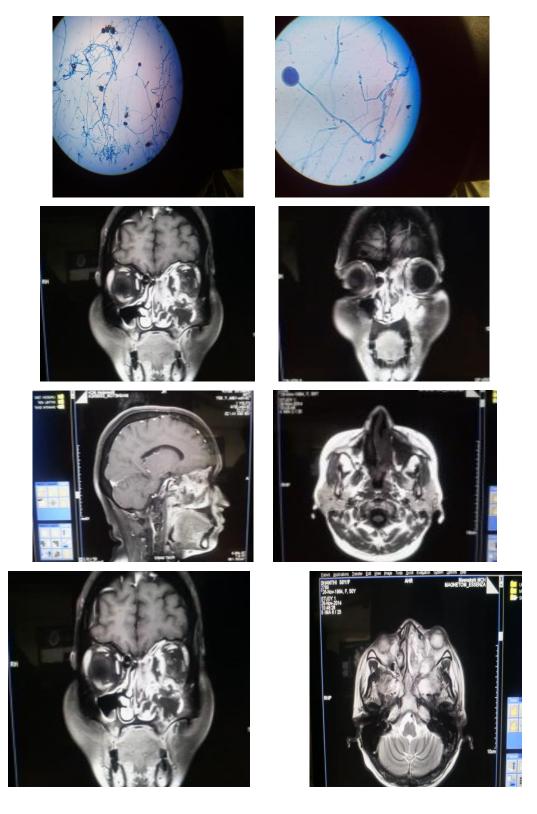
Hb	11.2
PCV	34
PC	6.24
MCV	82
MCH	27
MCHC	33
TotalRBC	4.13
ESR	119/140
Total WBC	13200
Neutrophil	77
Lymphocytes	23

Neutrophil	77
Lymphocytes	23
B.Urea	70
B.creat	1.2
Na+	123
K+	58
Cl-	84
HCO3	20

Based on clinical examination Multiple cranial nerve palsy /Mucormycosis/Tolosa Hunt syndrome was given as presumptive diagnosis.

Urine examination		
Albumin	trace,	
sugar	+++,	
pus cell	3-5	
epithelial cells	1-2,	
Ketone bodies	present	

Biochemical investigation showed the present of sugar in urine & Ketone bodies. Complete Blood count was within normal limit.



The patient was sent for MRI scan, neurological opinion, ophthalmology opinion, Diabetic OP, otolaryngology department to exclude sinonasal polyposis and to know the need for biopsy. Pernasal swab was sent for fungal culture to microbiology department.

The patient was givenInj. Human Actrapid10.0 IV Start, Inj Tramadol2ccIV Start, T. Amlong 5 mg 0-01, Inj insulin10 units. T. Prolomet- 25mg.followed by Nifedifine – 10mg1-0-1 BP chart and Blood Glucose chart is maintained.

MRI scan T1, T2, Axial and sagittal planes showed hyperintense mass lesion involving maxillary sinus and nasl cavity, microbiology fungal culture report showed typical growth of Rhizopus. Correlating all the three MRI Scan, microbiology report, clinical findings the case was diagnosed as Rhinocerebral mucormycosis. The patient was adviced for Liposomal AmphotericinB 5mg/kg/day iv after nephrology opinion. As the patient was not affordable for treatment, she was discharged.

DISCUSSION

Mucormycosis is usually common in immunocompromised patients with the history of Diabetes mellitus esp during periods of ketoacidosis, neutropenia, immunosuppressive therapy, long term antibiotic usage, trauma, surgical wounds, burns. [9] Other risk factors include haematological malignancy, bone marrow or stem cell transplantation, dexoferritin therapy. Rhizopus species have an active ketone reductase system and thrive in high glucose and acidotic condition. Diabetic patients will have decreased phagocytic activity because of impaired glutathione pathway. [12] As the fungi in turn need decreased levels of neutrophils for their optimal growth in humans (13), thus diabetics favours their rapid growth. [13] These factors support the diagnosis of present case as mucormycosis. Mucormycosis presents in rhinocerebral, cutaneous. [14] pulmonary, gastrointestinal, disseminated forms. [15] Rhinocerebral mucormycosis is predominantly associated with poorly controlled Diabetes and Diabetic Ketoacidosis. [16] The germination of spores is faster in carbohydrate rich environment.^[17] by low oxygen, high glucose, acidic medium and high iron levels. [18] Rhino cerebral mucormycosis patients present commonly with history of orbital and facial pain, nasal discharge or stiffness, sinusitis, alteration in vision. Physical examination shows periorbital and facial edema, orbital involvement may lead to loss or impairment of function of the cranial nerves II, III, IV and VI with resultant proptosis, ptosis, papillary dilatation, partial or total ophthalmoplegia & visual loss. Black eschar is usually seen over septum, palate or nasal turbinates.^[19] MRI Scan is useful for detecting cranial extension. Definitive diagnosis needs histological as well as isolation by using fungal culture media. [20] Culturing per nasal swab by using media like SDA results in growth by 48-72 hours. Many species have been reported. The most common being Rhizopus

orizae.^[21] The surface of the petridish is covered with wooly mycelium (lid lifter). Microscopic morphology shows broad, irregular width, ribbon like, aseptate hyphae, the spores are produced in sac like asexual sporangia, dense white cottony growth first becomes grey later, sporangiospores are globose or ovoid.^[4] In our case all the above mentioned clinical features, MRI and microbiological findings are significantly correlating to be diagnosed as Rhinocerebral mucor mycosis.

The treatment in present case consisted of treating underlying predisposing condition i.e., Diabetes mellitus, treatment with LAMP liposomal Amphotericin B with continuous monitoring of renal function as the drug is nephrotoxic. [22] along with hyperbaric oxygen as high pressure oxygen destroys fungal spores. In polyene resistant cases treatment with Posoconazole and iron chelation therapy indicated. [23]

CONCLUSION

As the treatment of Rhinocerebralmucormycosis is a costeffective as well as associated with sideeffects such as nephrotoxicity, the awareness about the possible consequences of uncontrolled Diabetes due to irregular medication and diet should be evaluated seriously and corrected to certain extent through health education. The disease has rapid fatal outcome relatively difficult to predict, thereby the clinicians awareness, early diagnosis, initial treatment and if needed surgical intervention is essentially needed for successful management. Moreover effective interaction among clinicians and diagnostic team i.e., a multidisciplinary approach consisting of physician, dental specialists, ENT surgeon, ophthalmologist, neurologist, a microbiologist pathologist and radiologist is utmost needed.

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