Case Report

Rhino-orbital-mucormycosis as a presenting manifestation of gestational diabetes mellitus

Mohd Hayat, Syed Mushtaq, Sameena Saba, Riyaz Saif

Department of Medicine, SMHS Hospital Srinagar, J&K, India

ABSTRACT

Rhino-orbital mucormycosis is an uncommon and aggressive, angioinvasive fungal infection that occurs in immunocompromised states like diabetes mellitus, chronic renal failure, hematological malignancies and deferroxamine therapy. We report a patient who presented with rhino-orbital mucormycosis at six months of gestation and was incidentally detected to have diabetes. She was successfully treated with amphotericin B and appropriate surgery. To the best of our knowledge, there is no such report in the literature.

Key words: Gestational diabetes mellitus, mucormycosis, India

Introduction

Rhino-orbital-mucormycosis is an aggressive, angioinvasive fungal infection seen in immunocompromised hosts. The risk factors are poorly controlled diabetes mellitus, neutropenia, hematological malignancies, long-term deferroxamine therapy, intravenous drug abuse and cytotoxic therapy.[1] Patients with diabetes constitute the single largest category of patients with mucormycosis (60-81%).[2] Rhino-orbital- mucormycosis (ROM) is seen as a presenting feature of diabetes in one fourth of patients.[3] However , there is only a single case report of rhino-orbital-mucormycosis in pregestational diabetes.[4] We present a patient who presented with ROM and was diagnosed to have gestational diabetes mellitus.

Case Report

A 24-year-old female who was six months pregnant complained of pain and swelling in the right eye of five days duration. She developed right nasal obstruction and right facial weakness three days later. She had no associated visual loss or diplopia. Subsequently, she was admitted with osmotic symptoms, recurrent vomiting and altered sensorium. She neither had any history of fetal wastage nor any past history of glucose intolerance. She had no family history of diabetes. Clinical examination revealed that patient was drowsy, pulse 110/min regular and BP 120/75 mmHg. The right eye was proptosed with periorbital swelling and she had right seventh nerve palsy. There was bloody nasal discharge with septal perforation of nose. Throat examination was normal. Investigations revealed random venous plasma glucose of 484 mg/dl with normal arterial blood gases and electrolytes. Hemogram showed hemoglobin of 7.6 g/l, total leucocyte count 16000/cmm; differential leucocyte count of N80 L12M5. HbA_{1C} was 8 %. CT of the brain with orbital sections [Figure 1] revealed an abnormal heterogeneous soft tissue swelling with post-contrast enhancement involving the right ethmoid gallery with extra- and intraconal orbital extension and associated right cavernous sinus thrombosis. Fungal smear showed aseptate right angled hyphae on 10 % KOH mount. Growth on Sabouraud's dextrose agar was identified as Rhizopus oryzae. Histopathology was suggestive of mucormycosis. The patient was started on intravenous insulin infusion, amphotericin B (liposomal) at 0.3 mg/kg/day and gradually increased to 1 mg/kg/day with monitoring of serum

Access this article online	
Quick Response Code:	Website: www.ijem.in
	DOI: 10.4103/2230-8210.83055

Corresponding Author: Dr. Syed Mushtaq, Department of Medicine, SMHS Hospital Srinagar J&K, India. E-mail: dr_mushtaq012@rediffmail.com

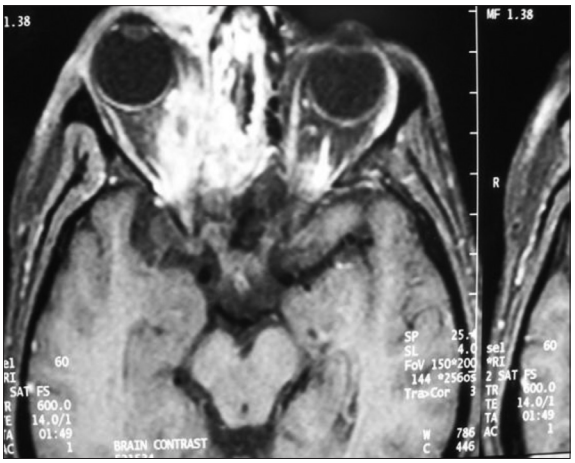

Figure 1: CT of the brain with orbital section showing an abnormal heterogeneous soft tissue swelling with post-contrast enhancement involving the right ethmoid gallery with extra- and intraconal orbital extension

electrolytes and renal functions (total cumulative dose 2.5 g). Surgical debridement in the form of right pansinusectomy and rhinotomy was done. Although patient had a fetal wastage, she recovered fully and is presently well controlled on two doses of premixed insulin.

Discussion

Rhino-orbital- mucormycosis (ROM) is a rare disease with overall prevalence of diabetes in 0.15%. However, rhino-orbital-cerebral mucormycosis as a presenting manifestation of diabetes is rare. In a previous series of 35 patients of diabetes, we noted ROM as a presenting feature in one fourth of patients.[3] Trivedi *et al*.,[4] reported a woman with pregestational diabetes mellitus who presented as ROM during gestation. The patient had a classical syndrome of diabetic ketoacidosis, visual loss, ophthalmoplegia and sinusitis. Our case was not a known case of diabetes. She presented with ROM at six months of gestation and was incidentally found to have diabetes. She presented with periorbital swelling, proptosis, facial weakness, septal perforation and pansinusitis. The organisms causing mucormycosis are Rhizopus, Mucor, Absidia, appearing in decaying vegetation and dung. Infection occurs when airborne hyphae are inhaled and deposited in upper or lower respiratory tract. The fungi penetrate arterial walls producing ischemia, thrombosis and infarction. Patients usually present with low-grade fever, dull sinus pain, nasal congestion, bloody nasal discharge and diplopia. Spread occurs via vessels to paranasal sinuses and to central nervous system via orbit and cribiform plate. Cavernous sinus thrombosis may occur due to orbital invasion. The diagnosis of mucormycosis was based on direct microscopy from nasal aspirate and on histopathology examination. Our patient was successfully treated with intravenous liposomal amphotericin B, surgical debridement and supportive care. The use of amphotericin B is associated with toxicity in form of renal failure, hypokalemia, hepatic impairment, fever and chills. Liposomal delivery allows the drug to be both less toxic and more effective.[5]

The prognosis of mucormycosis has markedly improved over past 30 years with 90% survival rate. In our previous series,[3] we noted a 68% survival rate. The factors related to poor survival were delay in diagnosis and treatment, facial and eyelid gangrene, hemiplegia, cerebral invasion by mucorales and treatment with amphotericin B alone. Our patient had a fetal wastage at six months which could be related to hyperglycemia and her concomitant illness. In fact, studies have shown that well regulated glycemia and intensive pregnancy follow-up of diabetic women reduces stillbirths, neonatal complications and neonatal macrosomia incidence.[6,7]

Conclusion

This case is unusual as it is the first report of a patient with gestational diabetes mellitus presenting as rhino-orbital mucormycosis. The present case also highlights the importance of early establishment of diagnosis, prompt initiation of amphotericin B therapy with surgical debridement and supportive care in successful management of rhino-orbital mucormycosis.

References

1. Yohai RA, Bullock JD, Aziz AA, Markert RJ. Survival factors in rhino-orbital-cerebral mucormycosis. Surv Ophthalmol 1994;39:3-22.
2. Ferry AP, Abedi S. Diagnosis and management of rhino-orbitocerebral mucormycosis (phycomycosis): A report of 16 personally observed cases. Ophthalmology 1983;90:1096-104.
3. Bhansali A, Bhadada S, Sharma A, Suresh V, Gupta A, Singh P, *et al*. Presentation and outcome of rhino-orbital-cerebral mucormycosis in patients with diabetes. Postgrad Med J 2004;80:670-4.
4. Trivedi TH, Jadhav DL, Yeolekar ME, Shejale SB, Pereira OT, Varaiya AY, *et al*. Rhinocerebral mucormycosis in a pregnant woman with diabetic ketoacidosis. J Assoc Physicians India 2002;50:592-4.
5. Fisher EW, Toma A, Fisher PH, Cheesman AD. Rhinocerebral mucormycosis: Use of liposomal amphotericin B. J Laryngol Otol 1991;105:575-7.
6. Djelmis J, Blajic J, Bukovic D, Pfeifer D, Ivanisevic M, Kendic S, *et al*. Glycosylated hemoglobin and fetal growth in normal, gestational and insulin dependent diabetes mellitus pregnancies. Coll Antropol 1997;21:621-9.
7. Ivanisevic M, Bukovic D, Starcevic V, Djelmis J, Pfeifer D. Influence of hyperglycemia on early embryonal growth in IDDM pregnant women. Coll Antropol 1999;23:183-8.

Cite this article as: Hayat M, Mushtaq S, Saba S, Saif R. Rhino-orbital-mucormycosis as a presenting manifestation of gestational diabetes mellitus. Indian J Endocr Metab 2011;15:S65-6.

Source of Support: Nil, **Conflict of Interest:** None declared.