A rare case of orbital mucormycosis with gas gangrene panophthalmitis

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Mucormycosis is a disease caused by a fungus from the Phycomycetes class and the Mucorales order. Phycomycetes are ubiquitous fungi occurring in soil, air, skin, body orifices, manure, and food. They are commonly seen in diabetics, particularly in patients with ketoacidosis.¹⁻⁶ Numerous other predisposing conditions have been reported.^{2,3,6}

Gas gangrene refers to a necrotising infection of the soft tissue caused by a *Clostridium* species. *Clostridium perfringens* is recovered in about 80% of cases. Other *Clostridium* species which commonly cause gas gangrene in humans are *novyi*, *septicum*, and *sordellii*.⁷

Case report

A 76-year-old Saudi Arabian man was admitted to the coronary care unit of the Security Forces Hospital with acute myocardial infarction. The patient had been taking oral hypoglycaemic

drugs for diabetes mellitus for the past 12 years; on admission this was changed to insulin. He was also on oral steroids for his bronchial asthma. The left eye had been totally blind for the past 10 years and he had previously had glaucoma surgery. His right eye was pseudophakic.

On the fifth day of admission the patient suddenly developed painless left proptosis and lid swelling. There was no history of trauma but mild epistaxis on the third day of admission had been noted. On initial ocular examination, there was severe panophthalmitis, orbital cellulitis, and proptosis with lid oedema (Fig 1). Foul smelling brownish pus with gas bubbles was exuding from the cornea and conjunctiva. Ocular motility was absent. An urgent computed tomography (CT) scan of the orbit and brain showed the orbit to be full of gas, with loss of definition of orbital contents, and the left eye ball was pushed forward (Fig 2).

Within hours, the patient also became febrile with marked periorbital pain. Gas gangrene of the orbit was considered. A Gram stain of orbital contents aspirated with an 18 gauge needle showed numerous long Gram positive bacilli. No fungi were seen. On the same day, under general anaesthesia, a left orbital exenteration was performed. Gas bubbles were coming out of the necrosed orbital tissues accompanied by a very foul smell and the surgery was almost bloodless. The orbital walls were intact and seemed healthy. The orbital contents were cultured and cytological and histological examination carried out.

The antibiotics were changed to piperacillin 2 g intravenously at 6 hourly intervals and penicillin G 3 mega units intravenously, also every 6 hours. The patient became afebrile and generally looked better.

Histological examination showed numerous broad non-septate, irregularly branching, thick walled hyphae in the necrotic tissue (Fig 3), the optic nerve sheath, and also within the vascular lamina. The hyphae were consistent with zygomycetes (*Phycomycetes mucor* species). A Gram stain of the tissue revealed numerous Gram positive bacilli (Fig 4), both in the orbit and ocular tissue. However, the fungi were only seen in the orbit. The culture on neomycin sheep blood agar plates, incubated in an anaerobic chamber, revealed heavy growth of *Clostridium sordellii*. Further identification of *Clostridium* species was done by using an Anident system kit. No other bacteria had grown within the culture. Urine and blood cultures were negative for *Clostridium* and other bacteria. Clinically, gas gangrene or mucormycosis was not seen in other parts of the body.

Soon after the histology revealed mucormycosis the patient was put on amphotericin B 60 mg

Figure 1 Left orbital cellulitis with marked proptosis.

Figure 2 Computed tomography scan showing left orbit full of gas with loss of definition of orbital contents and left atrophic eyeball pushed forward.

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Figure 3 Mucormycosis in orbital tissue. Broad thick walled, branching non-septate hyphae (haematoxylin and eosin stain $\times 140$).

intravenously at 6 hourly intervals. Contents cultured on dextrose agar plates were negative for fungi. Forty eight hours after surgery, the patient developed a fever, confusion, and right sided hemiparesis. A CT scan of the brain showed infarction of the left frontal lobe and internal capsule. Examination of the throat showed an extension of the infection to the hard palate. Unfortunately, the patient's condition deteriorated and he developed respiratory arrest 6 days after the operation. The patient was put on mechanical ventilation. He had a second cardiac arrest on the ninth postoperative day but did not respond to cardiopulmonary resuscitation and died.

Comment

Mucormycosis is notable for its high morbidity and mortality rates. The disease typically manifests itself as unilateral facial pain, headache, internal and external ophthalmoplegia, blepharoptosis, and severe visual loss.¹⁻⁶

The nose and the paranasal sinuses are usually the initial sites of infection. Orbital involvement can occur by a contiguous spread through the normal foramina in the medial wall or by invading the blood vessels. Most patients with rhino-orbital mucormycosis have a predisposing

systemic disease. Mucormycosis has been reported occasionally in otherwise healthy individuals.⁸⁻⁹ Multiple risk factors were present in this case including diabetes mellitus, systemic corticosteroids, and perhaps recent acute myocardial infarction.

Management of orbital mucormycosis depends on early diagnosis, control of any underlying disease, systemic antifungal therapy, and aggressive surgical debridement.^{1,3,5,6,8-11} Amphotericin B was first introduced into clinical use in phycomycosis in 1955 by Harris¹² and still seems to be the drug of choice.

Aggressive surgical debridement including exenteration is necessary in most cases. However, Bullock *et al*³ reported two cases of rhino-orbital mucormycosis successfully treated without exenteration with the sparing of vision in one case. Kohn and Hepler⁶ reported eight cases of rhino-orbital mucormycosis managed successfully without exenteration. In 1989, Luo *et al*¹⁰ reported a case of orbital mucormycosis with retinal and ciliary artery occlusion. It was treated successfully without exenteration.

Gas gangrene panophthalmitis usually follows after penetrating trauma, with or without a retained intraocular foreign body.¹³⁻¹⁷ The classic cause of this infection was nicely summarised by Duke-Elder.¹⁴ Leavelle¹³ in 1955 reviewed 53 previously reported cases and also added three cases of his own. In 1974, Frantz *et al*¹⁸ reported the only known case of *Clostridium perfringens* endophthalmitis produced by endogenous spread. So far, almost all the cases were treated by enucleation or evisceration. Crock *et al*,¹⁹ in 1985, reviewed the literature up to 1983 and added a case of their own. They described a patient with a retained globe and useful visual acuity of 20/60 after an ocular infection with *Clostridium perfringens*. This patient had bilateral ocular trauma. The right eye was enucleated and an aqueous sample from the left eye showed *Clostridium perfringens*. Wiles and Ide²⁰ reported a case of *Clostridium perfringens* endophthalmitis following penetrating trauma: lensectomy and subsequently vitrectomy improved vision to 20/40. Kelly and Steahly²¹ reported a case of successful prophylaxis of *Clostridium perfringens* endophthalmitis. This patient had received a penetrating eye injury with a nail contaminated with manure. After lensectomy and vitrectomy the patient was given an intravitreal antibiotic injection and his final corrected vision was 20/25. These are the only three cases of treated *Clostridium perfringens* endophthalmitis with retained useful vision.

It seems that our patient may be the only known case of *Clostridium sordellii* panophthalmitis with orbital cellulitis. In the spore form, *Clostridium* is resistant to many disinfectant agents. We believe this patient had intraocular *Clostridium* spores from previous surgery or, possibly, an old injury. Mucormycosis is known to invade the walls of the blood vessels and to cause occlusion, thrombosis, and infarction. *Clostridium* is known to thrive in tissues with low oxygen tension. In this case, recent mucormycosis vascular occlusion caused a tissue hypoxia which helped in fulminating *Clostridium* growth and spread.

Figure 4 Numerous large Gram positive bacilli, *Clostridium*, seen in ocular and orbital tissue (Gram staining).

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