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Mucormycosis and Ameloblastoma of the Upper Jaw

Summary: We report a case of mucormycotic infection complicating a clinically silent ameloblastoma in an aged diabetic woman. Diagnosis was made by culture and biopsy of the affected tissue, where foreign-body granulomas were seen, an unusual inflammatory response for mucormycosis. A good outcome was achieved on the basis of radical surgery and amphotericin B infusion.


Introduction

A large number of predisposing factors to mucor infection have been reported, but solid neoplasias are exceptional (1). Diagnosis requires biopsy of the affected tissue where foreign-body type granulomas are rarely observed (2). The high mortality may be decreased by prompt medical therapy and radical surgery (3). We report on a patient with ameloblastoma of the maxilla and rhinofacial granulomatous mucormycosis occurring at the same place. To our knowledge, the presence of a tumor as a local factor favouring mucor infection has not yet been considered.

Case report

An 80-year-old woman was admitted for severe polydipsia and polyuria lasting five days. She had type II diabetes mellitus for five years. Two months before entry, she was operated on her left eye for cataracts. A few days later she developed acute dacryocystitis and was started on intramuscular penicillin and local chloramphenicol. In spite of initial improvement, the disease slowly became suppurative and, on the day of admission, a rapidly expanding inflammatory process with development of a necrotic scar was noted.

On examination the patient appeared dehydrated with low grade fever and alert. There was a suppurative-necrotic ulcer in the left ala of her nose, involving the medial canthus of her left eye. There was scleral injection, fixed and dilated pupil and complete ophthalmoplegia (Figure 1). Sensation was impaired with an absence of corneal reflex. A palatal ulcer beside the left alveolus was discovered.

The urine gave a +++ test for glucose and a + test for ketones. ESR was 142 mm/h, normal leukocyte count, creatinine 1.3 mg/dl, urea nitrogen 60 mg/dl and glucose 678 mg/dl. Arterial blood gases and pH were normal.

The patient was started on insulin, intravenous fluids and antibiotics. A chest X-ray film was normal. Computed tomography of paranasal sinuses showed clouding of the left maxillary antrum, partial opacity of the left ethmoid sinus and a high density matter that seemed to erode the bone walls filling the left antrum. The treatment included intravenous amphotericin B.

The patient improved on therapy and the ulcer healed. The necrotic scar was removed surgically, and the patient made a good recovery. A biopsy of the scar tissues revealed no evidence of further infection.

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trum. Thickening of the left medial rectus muscle and proptosis of the left eye were also observed. Several blood and urine cultures were negative, while cultures from the ulcer yielded polymicrobial flora and *Rhizopus* sp. Speciation of the fungi was not done. Scraping of the left maxillary antrum was performed for pathologic investigation that revealed necrotic tissue areas with severe inflammatory changes and several foreign-body granulomas with giant cells and occasional central necrosis. In the center of some granulomas there were broad, nonseptate hyphae suggestive of mucormycosis (Figure 2). The rest of the tissue showed typical histological features of an ameloblastoma (Figure 3). The patient was started on intravenous amphotericin B for two months with a cumulative dose of 2.5 g. After having destroyed the left eye, maxillary sinus, left hard palate and nasal cavity, the infection stopped progression. Then it was decided to perform plastic surgery following resection of all devitalized and necrotic tissue. Hyphae and ameloblastoma cells could not be identified. At the last control, eight months later, the patient was in good general condition. No signs of relapse were noted.

**Discussion**

Mucormycosis is a relatively uncommon fungal infection caused by saprophytic fungi of the class *Zygomycetes*. Its main clinical picture and management have been well discussed in many other papers (1–7). We only want to focus attention on several features that make this case a singular one.

First, it is striking to find these two mutilant diseases, mucormycosis and ameloblastoma, at the same place, especially because they are rare and differential diagnosis between them may be required: clinically, they both can lead to facial deformity, local pain, epistaxis, nasal obstruction and palatal fistula (8, 9). However, other symptoms easily raise the suspicion of mucormycotic infection, as did in our patient: quick spread of the lesion, eye involvement, necrotic scar, etc. Moreover, it has been stated that mucormycosis does not directly complicate solid neoplasias (1, 10) unless immunodepressed conditions are present (11). To our knowledge, this case shows an association not previously reported.

Computed tomography played a role in accurate diagnosis, showing characteristic pictures for mucormycosis (12) and clarifying the presence of an antrum mass. In some way, a silent ameloblastoma in that site could have favoured fungal growth and extension through the antrum, as could have been done by other known predisposing factors such as previous operation, suppurrative infection and diabetes mellitus, which were also present.

Another unusual feature observed in this case is the type of inflammatory response for mucormycosis in the form of foreign-body granulomas, an event rarely described (2, 13). Perhaps the disease is usually so aggressive or the host is in such an immunocompromised condition that this type of defense is also impaired and has no chance to be effective.

Finally, we have to mention the successful treatment of this aged patient, one of the oldest we have observed. Many physicians would have refused to perform radical surgery in an 80-year-old diabetic woman, a recognized, critical, main step in the treatment of mucormycosis (1, 4, 6) and ameloblastoma (8, 14). Again age is not a factor limiting survival (3). At the present time, amphotericin B remains the drug of choice in the management of mucormycosis, until other substances are provided that can be more easily handled, are more effective and less toxic (15) than the ones we dispose of nowadays.

**Literature**