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MR Imaging Findings of Orbitofacial Infarction Secondary to Rhinoorbital Mucormycosis : A Case Report[1]

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Rhino-orbital mucormycosis is the most frequently fatal fungal infection commonly occurring in patients with poorly controlled diabetes mellitus and and those who are immunocompromised, and requires prompt treatment. We describe a case of rhino-orbital mucormycosis with orbital cellulitis and paranasal sinusitis, as seen on initial MR images, which on follow-up images had evolved to orbitofacial infarction. MR imaging was useful for the demonstration of orbitofacial infarction, seen as areas of lack of enhancement and thus suggesting vascular involvement by mucor hyphae.

Index words : Orbit, MR
Orbit, inflammation
Mucormycosis

Rhino-orbital mucormycosis is a relatively rare fungal infection that commonly occurs in diabetic patients and for which prompt surgery and medical therapy is required if its high morbidity is to be reduced. Black necrosis of the nasal turbinate is a characteristic but late feature (1). Rarely, mucormycosis can cause a serious condition known as orbital infarction, defined by Borruat et al. (2) as an ischemia of all intraorbital and intraocular structures. We describe one case of orbitofacial infarction complicated by rhino-orbital mucormycosis. To our knowlege, it is the first reported case of orbitofacial infarction revealed by MR imaging.

Case report

A 61-year-old diabetic woman visited our ophthalmology clinic with a three- day history of frontal headache and right ocular pain, periorbital swelling and sudden visual loss. She had been taking insulin irregularly for one year. The right periorbital region was edematous, with associated ocular chemosis and proptosis. There was no light perception in the right eye and the pupil was fixed. MR imaging of the orbit demonstrated periorbital swelling, with streaky and reticular enhanced lesions in the right periorbital and intraorbital region, suggesting orbital cellulitis (Fig. 1A). Enhanced minimal mucosal thickening was seen in the right ethmoid and maxillary sinuses; cavernous sinuses, however, were symmetrical without abnormal signal intensity. The initial diagnosis was orbital cellulitis complicated by paranasal sinusitis. Nasal examination revealed erythematous nasal mucosa without purulent discharge or necrosis. CT scans obtained on the same day showed a polypoid soft tissue lesion in the right ostiomeatal unit and associated sinusitis in the ipsilateral maxillary and ethmoid sinuses (Fig. 1B). Endoscopic intranasal right frontoethmoidectomy and middle meatal antrostomy were performed; histologic examination revealed only chronic inflammation. A course of intravenous broad-spectrum antibiotics was begun, but one day after surgery, right periorbital swelling progressed to the

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cheek and forehead. The ethmoidectomized cavity was filled with blood clots. The next day, the right periorbital swelling worsened and ischemic lesion was noted around the right cheek. The patient's temperature had risen to 38.3°C, white blood cell counts were 18,000 /mm^3, and the sodium/potassium level was 129/3.4 mEq/L. Because fungal infection was suspected, treatment with systemic amphotericin B was started empirically. On the third postoperative day, the patient became lethargic. Skin necrosis was found on the right eyelid, inner canthus, and cheek, and left periorbital swelling was also noted. Follow-up MR imaging of the orbit showed a lack of enhancement of right orbital structures, the right inner canthus, right nasal mucosa, and the nasal septum, and on postcontrast enhanced T1 weighted images, infiltration of left orbital fat was apparent (Fig. 1C). On the fourth postoperative day, the patient became drowsy, and the left eye progressed also became blind, with no light perception. The right facial necrosis and the right nasal cavity was covered with black crust. Biopsy was obtained from the necrotic lesion of the nasal cavity, and pathologic examination revealed clusters of hyphae of mucoraceous zygomycetes (Fig. 1D).

Discussion

Mucormycosis infection of the sinonasal cavities and orbital strucures is a serious condition that requires prompt diagnosis and treatment in order to minimize its high mortality rate. Characteristic black necrosis of the nasal turbinate may not appear until late in the course of the disease.

The fungi responsible for mucormycosis have a propensity for blood vessel invasion, producing arteritis, thrombosis, and secondary ischemia and tissue necrosis

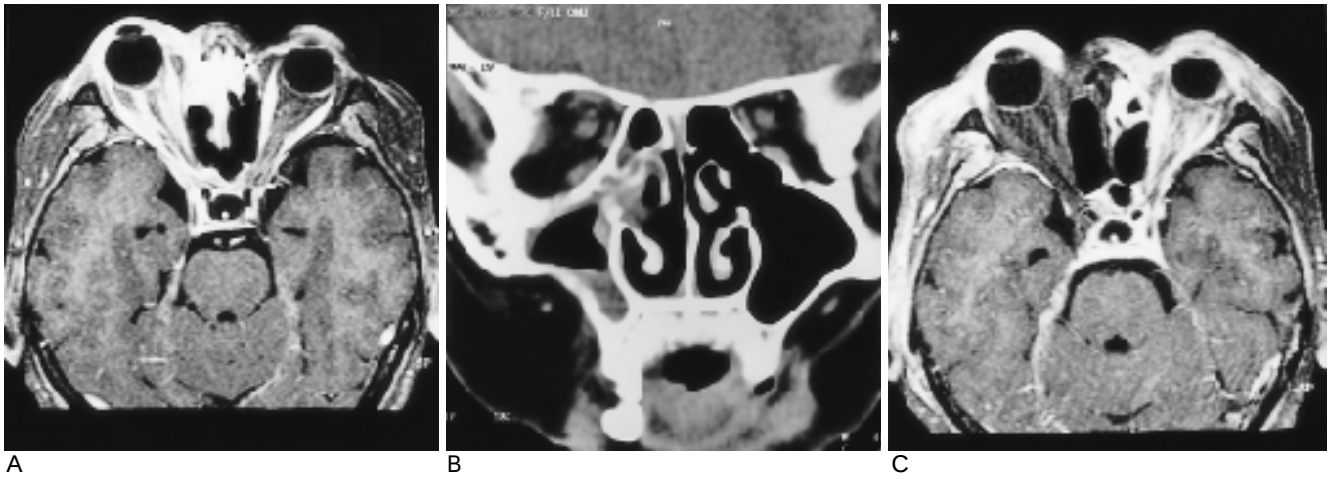

Fig. 1. Orbitofacial infarction complicating rhinoorbital mucormycosis in a 61-year-old woman.
A. Initial contrast enhanced axial T1-weighted MR image with fat saturation shows enhancement of the eyelid and retroocular fat in the right. Note enhancing mucosal lesion in the right ethmoid sinus.
B. Contrast enhanced coronal CT of the paranasal sinuses obtained at the same day reveals opacification of the right ethmoid and maxillary sinuses associated with polypoid soft tissue lesion obliterating the ostiomeatal unit. Fluid level is noted in the right maxillary sinus.
C. Follow-up contrast enhanced axial T1-weighted MR image with fat saturation obtained four days after surgery shows lack of enhancement in the extraocular muscles, the inner canthus, the anterior ethmoid sinus, and the nasal septum in the right. Right cavernous sinus also shows no enhancement. Note enhancement of the eyelid and retrocular fat in the left associated with left ethmoid sinusitis.
D. Photomicrograph of the necrotic specimen shows clusters of hyphae of mucoraceous zygomycetes with characteristic broad, pleomorphic, irregular and nonparallel contours(Methenamine silver stain ×400). Branches arise haphazardly, often at right angles to the parent hyphae.

(1-5). MR imaging findings of rhinocerebral mucormycosis, including acute cerebritis and cerebral infarcts due to arterial thrombosis, have been described in several articles (6-9). MR images obtained during the early course of the disease may, however, show only mild inflammatory changes including mild periorbital soft tissue swelling and reticular orbital infiltrations suggestive of orbital cellulitis. In retrospect, blindness might in our case be a valuable sign. The clinical presentation of orbital ischemia in mucormycosis may include proptosis, total external and internal ophthalmoplegia, and early blindness. In general, acute blindness is uncommon in orbital cellulitis secondary to bacterial sinusitis (1). Blindness may reflect involvement of the central retinal and/ or ophthalmic artery, the former being one of the smallest but most important branches of the latter. Because the retinal artery is essentially an end artery, its obstruction by an embolus or thrombosis leads to instant and total blindness (10). Borruat et al. (3) reported three cases of orbital infarction syndrome, one of which was caused by mucormycosis, defining orbital infarction as ischemia of all intraorbital and intraocular structures. The involvement of the central retinal and/or ophthalmic artery may produce blindness; short posterior ciliary artery involvement may result in anterior ischemic optic neuropathy and/or choroidal ischemia, while involvement of the arterial supply to the extraocular muscles may lead to ophthalmoplegia. Due to rich anastomoses between the internal carotid artery(ophthalmic artery and its branches) and the external carotid artery(maxillary, facial, superficial temporal arteries, and their branches), global orbital infarction is a rare disorder. In our case, in addition to global orbital infarction, ipsilateral facial necrosis also occurred reflecting the simultaneous involvement of the branches of the internal and external carotid arteries. Fungi might be propargated from orbital vasculatures to facial vessles through rich anastomoses. Orbitofacial infarction by mucormycosis can be diagnosed on the basis of no enhancement of the right ocular and orbital structures, nose, cheek, or temporal and infratemporal structures, as seen on contrast enhanced MR imaging, as well as clinical signs of necrosis.

In summary, we have described an unusual case of orbitofacial infarction complicated by rhinoorbital mucormycosis. MR imaging was useful for the demonstration of orbitofacial infarction as an area of lack of enhancement, probably caused by the involvement of mucormycosis in various arteries.

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