HEAD AND BRAIN SCAN FINDINGS IN RHINOCEREBRAL MUCORMYCOSIS: CASE REPORT

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Brain and bone scan findings in two patients suffering from rhinocerebral mucormycosis following kidney transplantation are presented.

Two patients who had had kidney transplants and were sustained for over a month on immunosuppressive drugs developed a rare type of opportunistic fungal infection—mucormycosis. They were examined in various stages of their disease. Special attention was paid to the scintillagraphic findings.

In the fungi of the order Phycomycetes, family Mycoraceae, the genus Mucor (Micheli, 1729; Lichtheimia of Villemin, 1904) is a widespread saprophyte and may become pathogenic in humans with inadequate resistance (*1*). The resistance is lessened in uncontrolled diabetes, leukemia, severe burns, liver cirrhosis, and especially in patients receiving immunosuppressive treatments. Mucor penetrate the organs through orbital, oral, nasal, or skin pathways and produce granulomatous, vascular, thrombotic, exudative, and necrotic lesions while the hyphae penetrate all normal structures.

When nasosinusal penetration is followed by invasion of the meningoencephalon, rhinocerebral mucormycosis results and may be fatal. With the increasing number of organ transplant procedures and concurrent immunosuppressive treatment, mucormycosis is more frequently encountered. Two cases of this infection were referred recently to our Institute for radioisotopic studies.

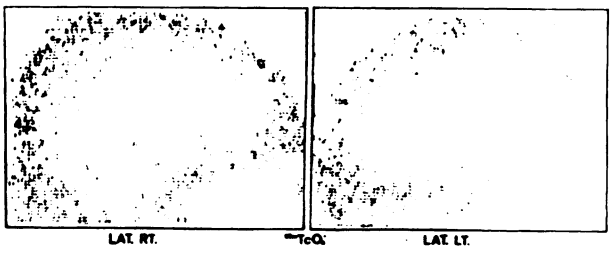

FIG. 1. Right lateral view: frontobasal peripheral concentration of sodium radiopertechnetate. Left lateral view: smaller insignificant concentration of radiopharmaceutical that can be considered normal.

CASE REPORTS

Case 1. A 44-year-old man with bilateral adult polycystic kidneys underwent transplant of one kidney in August 1972. He was maintained on a regimen of immunosuppressive drugs.

Two months after transplantation, the patient developed paresthesia on the right side of his face and paranasal swelling on the right. Nasal cavity examination showed necrotic bone of the middle concha; cultures of bone and nasal smears revealed mucormycosis. Subsequently, the swelling increased and peripheral facial nerve palsy appeared. Immunosuppressive therapy was discontinued and antifungal treatment was initiated (amphotericin B and 5-fluorocytosine). Radiologic examination of the sinuses was normal. Neurologic re-examination 6 weeks later still showed right Bell's palsy with blepharoptosis.

At that time, 9 months after the onset of the mucormycosis, kidney rejection occurred and the patient was referred for brain scanning.

The brain scan (Fig. 1) showed a frontal-basal, supraorbital abnormal concentration of activity adjacent to the calvaria, extending towards the upper border of the frontal region.

The patient died 1 week later. At autopsy the right sphenoethmoidal sinuses were filled with mucoid material. Histologic examination demonstrated ulceration of the mucosa and bone necrosis, hyperemia, and thrombosis of vessels. Similar hyperemic–

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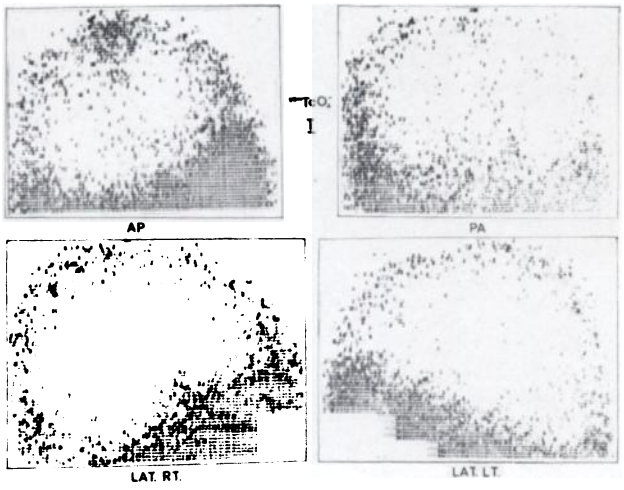

FIG. 2. No lesion in brain hemisphere area; large homogenous triangular concentration in left naso-orbital-calvarian region on anterior and left lateral views.

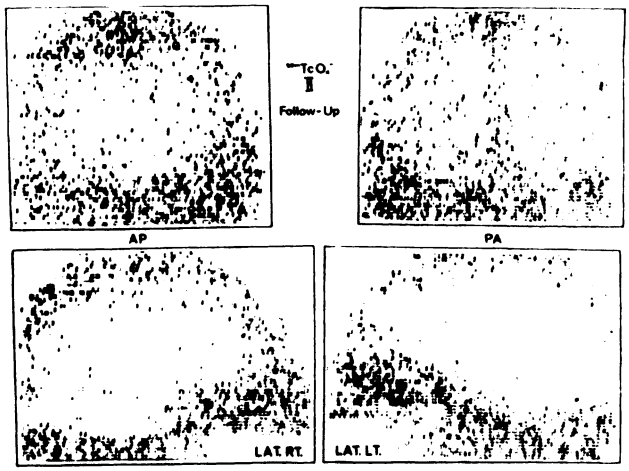

FIG. 3. Followup: sodium radiopertechnetate brain scan showing regression in concentration of activity and homogeneity of pathologic triangle.

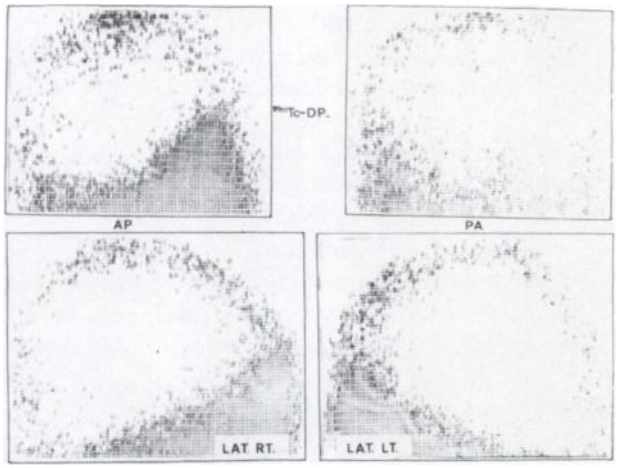

FIG. 4. Technetium-99m diphosphonate bone scan showing large triangular area of activity in left naso-orbital-calvarian region. Area is larger than one in Fig. 3.

thrombotic findings were found in the adjacent fronto-orbital walls on the right. Mucor and penicillum hyphae were identified microscopically. The brain was normal.

Case 2. A 24-year-old man with chronic end-stage glomerulonephritis had been on hemodialysis since 1971. He had bilateral nephrectomy in September 1972 and a kidney transplantation in March 1973. One month later he showed signs of acute rejection; the immunosuppressive treatment was increased with a parallel improvement in the transplant function. A few days later the patient complained of painful teeth in the left maxilla. Two days later this was followed by facial swelling and blood-tinged secretions in the left nasal region.

Two days later as swelling of the left side of the face spread towards the orbital region, blepharoptosis and palsy of the trigeminal and facial nerves also appeared. Necrosis of the bony middle and lower left nasal conchae was found. The nasal secretion cultures grew mucormycosis fungi after 48 hr.

Immunosuppressive therapy was discontinued and antifungal therapy was initiated (amphotericin B and 5-Fluorocytosine). Gradual improvement of the mucormycotic condition occurred but the mandibular nerve-branch palsy persisted. The renal transplant had to be removed in May 1973 because of acute rejection. In spite of discontinuing the immunosuppression drugs and proceeding with the antifungal therapy, the mucormycosis produced osteomyelitis of the maxilla and hard palate. Because of root infection, the upper left teeth were extracted. Radiologic examination of the sinuses showed thickening of the left maxillary mucosa.

Brain imaging was performed about 3 weeks after the onset of the mucormycosis, following appearance of left facial nerve palsy, including paresis of the right forehead and eyelids, with residual swelling in the left side of the face (Fig. 2). A large, triangular area of radioactivity was observed in the left naso-orbital-calvarian region. These findings can be explained on the basis of hypervascularization in the edematous, granulomatous tissue because of mucormycotic invasion of the left rhino-orbital-skull region (mucosa, bones, meninges).

After 3 weeks of intensive antifungal therapy, the patient's symptoms were somewhat alleviated but he still had residual facial mandibular palsy and maxillary osteomyelitis. At that time, brain imaging demonstrated regression in concentration and homogeneity of the pathologic region (Fig. 3) that may have been due to diminished vascularity and increased bone necrosis. The last assumption was verified by a ^{99m}Tc-diphosphonate bone scan performed 4 days later (Fig. 4) that revealed intensive osteogenic ac-

tivity of the sequestrated structures invaded by the fungi. This involved area appeared larger than that presented by the hypervascularization and granulation changes (Fig. 2).

DISCUSSION

There are several reports concerning the subject of rhinocerebral mucormycosis. A few were associated with renal (*2,3*) and cardiac transplantation (*4*) and some were not associated with organ transplantation (*5*). Up to the present, there has been no mention of an in vivo demonstration by isotopic methods of organs involved in mucormycosis.

The two cases presented demonstrate the possibilities of radionuclide brain imaging in the visualization of the shape and extent of local mucormycotic invasion. This adds valuable information to the clinical picture and effectiveness of the antifungal treatment.

The two cases of rhinocerebral mucormycosis demonstrated the following two features: (A) mass lesion was not detected in the brain tissue, and (B) abnormal accumulation of radioactivity by brain and bone imaging was found in the rhino-orbital fronto-calvarian region according to the localization of the mucormycosis.

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