Cryptococcic (Torula) Granuloma of the Skull*

Case Report

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Cryptococcosis, or torula infection, is recognized as an uncommon disease caused by a yeast-like parasitic fungus having a special predilection for the central nervous system and the lungs. More recently Cryptococcus neoformans has come into rather general use for the pathogenic organism of this disease. Among others, Muchmore, et al.,23 have found that bird droppings are the major source of human infection, with the respiratory tract as the usual point of entrance in the body.

The salient peculiarities of this fungal infection have been described comprehensively in several publications,1,3,4,9-11,14,16,18,19,21,25,26,28,29 Not uncommonly the protean nature of its clinical manifestations has caused confusion with tuberculous meningitis, coccidiodial meningitis, epidemic encephalitis, and tumor or abscess of the brain. Frequently, moreover, the etiology has been revealed only at the time of necropsy.

Before the introduction of amphotericin B in 1956,13 cryptococcal meningitis was almost always fatal, three quarters of the patients dying during the first year of illness. Freeman10 pointed out the variable virulence of cryptococcosis, and there have been well-documented cases of patients surviving many years. Becson,2 for example, reported the case of a woman finally succumbing to a chronic form of cryptococcal meningitis after 16 years. Nevertheless, survival for longer than 3 years is unusual. Since the original report of Appelbaum and Shtokalko1 on the effectiveness of amphotericin B in the treatment of cryptococcal meningitis, a number of reports have confirmed and extended this observation,3,4,8,15,27 Survival rates have ranged from 60 to 83%, a distinct contrast to reports before its introduction.

In their review of 178 cases of cryptococcosis with central nervous system involvement, Carton and Mount11 in 1951 found 42 cases in which neurosurgical procedures were performed; nine of these had single or multiple granulomatous masses in the brain or spinal cord, and two were discovered at autopsy.

Ley, et al.,17 reported a case of cryptococcal granuloma of the cervical cord in 1951. The nature of the tumor was not recognized until completion of the microscopic studies. Remarkably in their case, this child of 8 years, who had a quadripareisis before operation, recovered completely.

Liu20 found only five cases in which a sizable granuloma had been surgically removed from the brain. The first patient, reported by Gáspár12 in 1929, had a 6-cm mass in the left parietal region and died 3 weeks after operation. The second case was one of a cryptococcal granuloma in the left cerebellar hemisphere. The patient was discharged against advice 1 month after surgery with evidence of generalized infection. In the third case,29 a granulomatosus mass was completely removed from the right cerebellar hemisphere. The patient showed evidence of recurrence 4 months later and died 5 days after a second operation. The fourth case,15 was operated upon for a large cystic mass in the left subfrontal region. Although the patient had evidence of systemic cryptococcosis, he was living and well 11 months postoperatively.

In the fifth case,6 there was a mass in the left cerebellar hemisphere containing cryptococcal bodies in the paraffin sections. The patient died 5 months postoperatively. In his case,20 Liu removed a calcified cryptococcal granuloma from the right frontal lobe. His patient was well 12 months postoperatively. Manguenllo and Nichols22 removed an intraventricular cryptococcal granuloma from the right lateral ventricle. Death occurred 10 days postoperatively.

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Since the introduction of amphotericin B such surgical excisions of cryptocococ granulomas have obviously had a more promising prognosis.

Case Report

In January, 1957, the patient, a 55-year-old petroleum engineer, developed right-sided headaches which he thought the result of an upper respiratory infection. During the latter part of April he noticed a small lump developing in the right parietal scalp. Radiographs obtained May 15 revealed an osteolytic defect in the right posterior parietal region of the skull underlying the scalp swelling. His earlier history had been uneventful.

Examination. The salient features in the examination included that of a slight swelling in the right parietal area, which was not fluctuant and only slightly tender to palpation. He was mentally alert and cooperative. The general neurological and physical examinations were unrevealing. Radiographs of the chest, including anteroposterior and lateral views, showed no indication of abnormality.

Operation and Findings. On May 23, 1957, the small subcutaneous tumor was excised. The underlying osteolytic defect was enlarged slightly and a yellowish epidermal granuloma removed. The frozen section did not suggest a tumor, and for this reason cultures were obtained. Subsequent microsections were described as showing a thickened granulomatous process with proliferating fibroblasts, large mononuclear cells, and newly formed capillaries. Throughout the granuloma there were frequently fairly large collections of various-sized yeast cells, the largest being the size of an erythrocyte. A few of these showed budding; some contained dark blue endospores. Wet mounts of the material stained with India ink showed capsules. Cryptococcus neoformans was cultured from this material.

A spinal puncture after the operation revealed only one cell and no growth on culture; the spinal fluid protein was 34 mg%, and the serology was negative. Cryptococcus neoformans was cultured from sputum obtained May 25.

Postoperative Course. When seen June 8 he felt weak and tired, but the discomfort in the right parietal region had disappeared. He was readmitted to the Santa Barbara Cottage Hospital on June 19 for the administration of amphotericin B, which was obtained prior to its general release through the courtesy of E. R. Squibb and Sons. He was given intravenously a total of 1500 mg of this antifungal preparation.

On June 10, 1957, the California State Laboratory confirmed the diagnosis of Cryptococcosis neoformans from the skull biopsy of May 23, 1957, and also confirmed the presence of the same fungus obtained from the sputum May 25.

The patient was reexamined August 12, 1957. At this time he looked well, was eating better, and had more strength. He had no headache, and there was no soreness over the operated area. Thereafter the patient has been followed at intervals and was asymptomatic at the most recent examination in December, 1966 (Dr. Charles A. Smolt), 9½ years postoperatively.

Discussion

Localized cryptocococ granuloma can occur without other evidence of central nervous system disease. This rather unusual case simulated that of an osteolytic lesion of the skull associated with an overlying tumor of the scalp suggesting a malignant metastatic lesion. Laboratory studies established the diagnosis of Cryptococcosis neoformans.

The importance of prompt administration of amphotericin B, as a specific antifungal antibiotic, as an adjunct to surgery is readily apparent.

Summary

We have reported the occurrence of a cryptocococ osteolytic lesion of the skull with an overlying granuloma, successfully treated by surgical excision and amphotericin B. The patient has remained well for 9½ years. We have reviewed other related reports and have discussed the nature of this infection and its treatment.

Bibliography