

Localized osseous cryptococcal infection

Report of 2 cases

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Cryptococcal infection in man is widely disseminated and commonly involves the central nervous system. Primary bone involvement is rare. We report on 2 cases of localized cryptococcal osseous lesions treated successfully by surgery.

Case reports

Case 1

A 5-year-old girl was referred with a 2-month history of pain and swelling over the left distal thigh. When initially seen at the referring clinic, a diagnosis of acute osteomyelitis was made and the child was treated with oral antibiotics and an above-knee plaster cast.

The child was afebrile, and the thigh was minimally indurated and bony tenderness was not present. The knee joint was normal and the rest of the physical examination was noncontributory. Apart from an ESR of 51 mm, the rest of the hematologic examinations were normal. The cerebrospinal fluid for cryptococcal antigen was negative. Scintimetry revealed an increased uptake in the left distal femur. The chest radiograph was normal, and the left distal femur showed a lytic lesion over the posteromedial aspect with surrounding periosteal reaction (Figure 1).

The area of induration was approached from the medial aspect. The periosteum was thickened, and there was no pus in the soft tissues. A window was made over the posteromedial cortex, and thick pus was evacuated. The medullary cavity was curetted and lavaged with saline. The wound healed uneventfully. Culture revealed *Cryptococcus neoformans*.

The child was immobilized in an above-knee plaster cast for 6 weeks; in view of the single nature of the lesion without any pulmonary or meningitic involvement, she was not treated with any antifungal agents. At 18 months the child was asymptomatic, and the ser-

um and the cerebrospinal fluid for cryptococcal antigen tests were negative. Radiographs of the left femur showed satisfactory healing.

Case 2

A 29-year-old female presented with a 3-month history of vague pain over the right gluteal region. There was minimal tenderness over the right ilium and no soft-tissue swelling. The right hip was normal. The hematologic examinations were normal apart from an ESR of 60 mm. Scintimetry revealed a localized area of increased uptake over the right ilium. The chest radiographs were normal. A radiograph of the pelvis showed a lytic lesion over the right ilium, and a CT scan showed two well-circumscribed lytic lesions with the outer cortex broken in one area (Figure 2).

At surgery a small amount of thick pus had broken through the outer wall of the ilium. The entire area of bony involvement was excised. *Cryptococcus neoformans* was isolated from this specimen. In view of the soft-tissue contamination, the patient was treated with 5-fluorochromes and Amfotericin B. Two days after therapy, the patient refused further medical treatment because of severe vomiting and rigors. She has been followed up for 18 months and has remained asymptomatic.

Discussion

The pigeon appears to be the chief vector for the distribution and maintenance of *Cryptococcus neoformans*. The organism is usually found in debris of pigeon roosts.

There are essentially two types of cryptococcal in-

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Figure 1A

Figure 1B



Figure 2

Figure 1. A 5-year-old girl with a cryptococcal infection in the left femur.

A. At diagnosis after 2 months of pain and swelling.
 B. Healing at 18 months.

Figure 2. A 29-year-old woman with a cryptococcal infection in the right ilium.

fections, but the manifestation depends on the host response rather than the strain of the organism. In the normal patient, infection following inhalation of the organism is usually rapidly resolved with minimal symptoms and the disease is usually subclinical; growth-inhibiting substances are normally present in the body fluids (Igel and Bolande 1966). If the number of organisms inhaled is considerable, an infection may be initiated in the lungs with occasional transitory foci of infection in other sites. Even though the tissue reaction and cellular defenses are evoked slowly, they are usually adequate to contain the infection. Infrequently in normal patients, a chronic infection is established

that gives rise to flare-ups of systemic, cutaneous, or meningitic involvement. Such patients require treatment.

The second type of disease is usually associated with neoplasms, debilitating disease, and in compromised hosts is usually a result of drug therapy. Due to the poor immune response in these patients, the infection spreads rapidly to involve almost all the organs, especially the central nervous system (Collins et al. 1951).

Involvement of bone as part of a systemic infection is said to occur in 10 percent of the cases (Chleboun and Nade 1977). Primary bone involvement is rare; the most common areas of involvement are the pelvis and the spine, although occult disseminated disease may

become manifest many years after the appearance of an isolated lesion (Morris and Wolinsky 1965).

Localized osseous cryptococcal infection can usually be successfully treated without the use of antifungal agents (Chleboun and Nade 1977). Amphotericin B and 5-fluorochromes have proved effective in the treatment of osseous *Cryptococcus* with associated systemic involvement. Due to poor absorption after oral administration, the drug is given intravenously. The dosage is then gradually increased, and the renal function is carefully monitored. The smaller dose of Amphotericin B, apart from reducing nephrotoxicity, increases the penetration of 5-fluorochromes over the cytoplasmic membrane of a fungal cell (Utz et al. 1975).

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