Use of Massive Allografts to Manage Hydatid Bone Disease of the Femur

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abstract

Hydatid disease is caused by the parasitic tapeworm *Echinococcus granulosus*. Osseous involvement accounts for 0.5% to 4% of cases in humans. Patients usually are from endemic zones and are initially asymptomatic, presenting with pain and edema at a later stage of disease. However, large lesions may present initially as pathologic fractures. Standard radiographs usually show expansive osteolytic lesions associated with initial cortical thinning, with compromise of the metaphysis or epiphysis, and may involve the diaphysis. The finding of periosteal reaction, osteocondensation, calcification, and clear delimitation of the lesions excludes the diagnosis of osseous hydatidosis. However, there are no specific radiographic signs in the affected bone. There is no generally accepted treatment algorithm for osseous hydatid disease. The usual treatment is surgical resection of the affected bone, followed by antihelmintic therapy. Some patients can be treated with intrallesional procedures, such as curettage and allograft or polymethyl methacrylate cement. However, in some advanced cases, such as those with pathologic fractures or recurrences, wide resection may be needed. Several reconstructive alternatives have been reported, such as megaprosthesys, massive allograft, or allogrosthesis composite. The authors retrospectively reviewed 2 cases of femoral hydatidosis treated with wide resection and reconstruction with massive bone allograft. One patient had 5 years of follow-up, and the other had 9 years. No signs of relapse or complications were recorded, and functional outcomes evaluated with the Musculoskeletal Tumor Society scale showed excellent results. These 2 cases support the use of a massive bone allograft as a valid alternative to reconstruction after extensive bone resection for hydatidic disease. [Orthopedics. 2015; 38(10):e943-e946.]

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Drs Muscolo, Zaidenberg, Farfalli, and Ayerza have no relevant financial relationships to disclose.

Dr Aponte-Tinao is a paid consultant for Stryker.

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Received: August 15, 2014; Accepted: February 23, 2015.

doi: 10.3928/01477447-20151002-92
Hydatid bone disease is caused by the parasitic tapeworm Echinococcus granulosus. Osseous involvement accounts for 0.5% to 4% of cases in humans. The spine (vertebral bodies) (35%) and the pelvis (21%) are the most commonly involved skeletal sites, followed by the long bones, especially the femur (16%).

Patients usually are from endemic areas such as the Middle East, Central Asia, South America, East Africa, and Turkey.

Clinical presentation is characterized by pain and edema. However, large lesions may present initially as pathologic fractures. Results of serologic and immunologic tests generally are positive during the early stages of bone hydatidosis and in the presence of cyst complications (rupture and infection or abscess), whereas these results are negative in advanced disease, when the cyst is aging or calcified.

Standard radiographs usually show expansive osteolytic lesions associated with initial cortical thinning, with compromise of the metaphysis or epiphysis that can involve the diaphysis at later stages. The presence of periosteal reaction, osteosclerosis, calcification, and clear delimitation of the lesions excludes the diagnosis of osseous hydatidosis. However, there are no specific radiographic signs in the affected bone. The differential diagnosis includes chronic osteomyelitis, fibrous dysplasia of bone, osteosarcoma, and benign cystic lesions.

There is no generally accepted treatment algorithm for osseous hydatid disease. However, the usual treatment is surgical resection of the affected bone, followed by antihelmintic therapy. Most patients can be treated with intralesimal procedures, such as curettage and allograft or polymethyl methacrylate (PMMA) cement. However, in some advanced cases, such as those with pathologic fractures or recurrences, wide resection may be needed, and several reconstructive alternatives have been reported, including megaprosthesis, massive allograft, and alloprosthesis composite. Despite these treatment options, there is no consensus on the best alternative for reconstruction.

This study evaluated the long-term results of 2 cases of hydatidosis of the femur treated with wide resection and femoral allograft reconstruction.

**Case Reports**

**Patient 1**

A 65-year-old woman presented to the authors’ institution with a hydatid cyst in the bone involving the right femur. Radiographs of the femur showed multiloculated osteolytic lesions affecting the femoral diaphysis. Magnetic resonance imaging (MRI) scan showed expansion and thinning of the cortices. Results of an indirect hemagglutination assay for Echinococcus were positive. A definitive diagnosis was made previously at another institution, based on the findings of needle biopsy and histopathologic examination.

Wide resection was planned, adding 2 cm to the MRI bone compromise of the femoral diaphysis. The defect was reconstructed with an intercalary bone allograft. Fixation was achieved with a locked intramedullary nail and 4 cancellous bone screws at the distal osteotomy. Wide resection was planned, adding 2 cm to the MRI bone compromise of the femoral diaphysis. The defect was reconstructed with an intercalary bone allograft. Fixation was achieved with a locked intramedullary nail and 4 cancellous bone screws at the distal osteotomy (Figures 2-3). At 9-year follow-up, the patient was treated with wide resection and femoral allograft reconstruction. Transplant fixation was obtained with a plate with screws (dynamic compression plate) (Figure 4B). The patient was also treated with albendazole (15 mg/kg/d orally) preoperatively for 1 month and postoperatively for 6 months as adjuvant therapy. The patient had a functional score of 29 of 30 points (97%), indicating an excellent result according to the MSTS scale. The patient was followed for 5 years without any complications.

**Patient 2**

A 33-year-old woman presented to the authors’ institution with a pathologic fracture of the proximal right femur and multiple cysts in the metaphyseal region. The MRI scan also showed high signal intensity in the femoral head and extensive lateral soft tissue compromise.

The patient was treated with wide resection of the proximal femur, with no intralesimal margins. To repair the extensive osteoarticular defect, a proximal femoral allograft prosthesis composite was performed for reconstruction. Transplant fixation was obtained with a plate with screws (dynamic compression plate) (Figure 4B). The patient was also treated with albendazole (15 mg/kg/d orally) preoperatively for 1 month and postoperatively for 6 months as adjuvant therapy. The patient had a functional score of 29 of 30 points (97%), indicating an excellent result according to the MSTS scale. The patient was followed for 5 years without any complications.

**Discussion**

Bone hydatid disease is often asymptomatic for a long time because of its low growth rate, so it is usually detected at an advanced stage, with large osteolytic lesions.

Early diagnosis is uncommon and is primarily based on radiographic findings that show irregular destruction of the
bone, with multiloculated lesions. However, there are no specific radiographic signs in the affected bone.

The most helpful imaging method for diagnosing hydatid bone disease is MRI. Typical MRI findings are a high signal in the T2-weighted image sequence and the shape of a rose or a wheel as a result of spaces or septi of the daughter cyst. The multilocular appearance and medullar involvement are other characteristics seen on MRI scans and are especially important for planning surgical bone resection.

These 2 patients were initially seen at two different institutions, and biopsy and histopathologic examination showed the parasite replacing the bone tissue. The authors recognize the importance of preoperative serologic tests in the differential diagnosis. Specific antibody tests, such as the Casoni test, indirect hemagglutination assay, counterimmunoelectrophoresis, enzyme-linked immunosorbent assay, and gold-labeled antibody, are used to confirm the diagnosis. Baveja et al\textsuperscript{11} reported that the 8 test of immunodiagnosis provides the most sensitive and specific serologic test, with up to 92.6% sensitivity and 91.9% specificity.

Most authors recommend chemotherapy with albendazole or mebendazole in combination with surgery to decrease recurrence rates.\textsuperscript{3,4} Antihelmintic therapy alone has proved ineffective in 25% to 30% of cases.\textsuperscript{10} Patient 1 did not receive antihelmintic adjuvant therapy, but Patient 2 received albendazole both pre- and postoperatively. The patients showed no signs of relapse at 5 years and 9 years of follow-up, respectively. This study included only 2 patients; therefore, it is difficult to determine the role of antihelmintic therapy in the final results.

Surgery is usually the first choice of treatment. In 2001, Yildiz et al\textsuperscript{12} reported 10 patients treated with curettage and repair of the defect with PMMA. They noted that curettage and PMMA may not eradicate bone hydatidosis, but the use of PMMA reduced the rate of recurrence and gave the best outcome in the treatment of bone hydatidosis. However, on long-term follow-up, 3 of the 7 patients had recurrent bone disease.

Wide resection is essential to achieve local control and reduce the recurrence rate, and the massive defect presents a significant challenge to the reconstructive orthopedic surgeon.\textsuperscript{13} Several reconstructive techniques used in orthopedic oncology for the treatment of primary osseous sarcomas are applicable. These include endoprosthetic metallic reconstruction, massive allograft transplantation, and combinations of these.

Natarajan et al\textsuperscript{7} reported the use of a custom megaprosthesis for reconstruction of bone defects secondary to hydatid bone resection in 3 patients, with excellent results and no recurrences. However, this study had a short follow-up, underestimating the probability of recurrent infection or loosening of the implant complicating the prosthetic reconstruction.

There are only 3 published reports of the use of massive allograft reconstruction for significant bone defects after extensive resection as a result of hydatidic disease of the femur. The first case was reported by Capurro and Pedemonte\textsuperscript{14} in 1953. The authors used a complete femur transplant, with follow-up of 10 months. A second similar case was reported by Ottolenghi\textsuperscript{15} in 1966, with follow-up of 36 years.\textsuperscript{16} The last and most recent case was a femoral allograft reconstruction, with follow-up of 26 months.\textsuperscript{17} The current authors reported 2 additional cases treated at their institution, with 5 years and 9 years of follow-up, respectively.
Although good results have been reported with this biologic reconstruction technique, Mankin et al\(^\text{18}\) reported a series of 945 patients with tumor lesions treated with massive bone allografts, with a primary infection rate of 7.9% (not related to reoperation for nonunion or fracture of the graft).

These 2 cases support the use of a massive bone allograft as a valid alternative to reconstruction after extensive bone resection as a result of hydatidic disease.

REFERENCES