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Malignant epithelioid hemangioendothelioma at the site of a hip prosthesis

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We report a case of malignant epithelioid hemangioendothelioma in the acetabulum and ipsilateral femur 11 years after a total hip prosthesis.

The development of a sarcoma at the site of a total hip prosthesis is rare. Up to now, there are approximately 2 million total hip prostheses inserted around the world. In the Anglo-Saxon orthopedic literature, 4 cases of sarcoma at the site of a total hip prosthesis have been reported (Penman and Ring 1984, Swann 1984, Bagó-Granell et al. 1984, Ryu et al. 1987). Two patients had a malignant fibrous histiocytoma, 1 patient had an undifferentiated osteosarcoma, and 1 patient had an undifferentiated sarcoma. We report on a patient who presented with a malignant epithelioid hemangioendothelioma. This is a vascular tumor sometimes described as histiocytoid hemangioma (Rosai 1979), with a behavior intermediate between a hemangioma and a conventional angiosarcoma (Enzinger 1983).

Case report

A 72-year-old woman with arthrosis secondary to congenital hip dysplasia underwent a bilateral total hip replacement in 1974. The prostheses were of the Charnley-Müller type (Sulzer, Winterthur), a polyethylene cup and a Co-Cr-Mo alloy (Protasul®) stem fixed with polymethylmethacrylate (Palacos®) cement. She had a revision with a long-stem Charnley-Müller prosthesis on the left side due to aseptic loosening 3 years later. She was seen annually; and in July 1985, 11 years after the primary total hip prosthesis was inserted, she complained of progressive pain in the right hip and leg. There was a 4-cm shortening of the right leg with a very small range of painful movement. The ESR was 28 mm. Radiographs revealed a severe osteolysis around the cup and a subsidence and varus tilt of the femoral component (Figure 1).

She had a revision operation of the right hip performed in August 1985. After removal of the cup and cement, large masses of bleeding granulation tissue were found with a large defect in the medial wall of the acetabulum. An autografting of bone was done (Slooff et al. 1984), and a Müller prosthesis was inserted and fixed with antibiotic-loaded bone cement (Palacos®). The femoral component was also loose and exchanged. Postoperative care included anticoagulants, systemic antibiotics, and indomethacin to prevent periarticular ossifications.

In January 1986, she was seen at the outpatient department with malaise and progressive pain in the right hip region radiating to the knee. There was a tender fluctuating swelling in the right groin. The ESR was 120 mm/h, Hb 4.5 mmo/L, calcium 3.50 mmol/L, and phosphate 0.66 mmol/L. The radiographs showed osteolysis of the medial acetabulum wall, ramus superior ossis pubis, and os ischii (Figure 1). There was also an osteolysis in the ipsilateral femur 8 cm under the tip of the prosthesis. No other pathologic features were found in an extensive screening. An open biopsy was performed from both osteolytic tumors. The histopathologic diagnosis for both tumors was epithelioid hemangioendothelioma (Figure 2).

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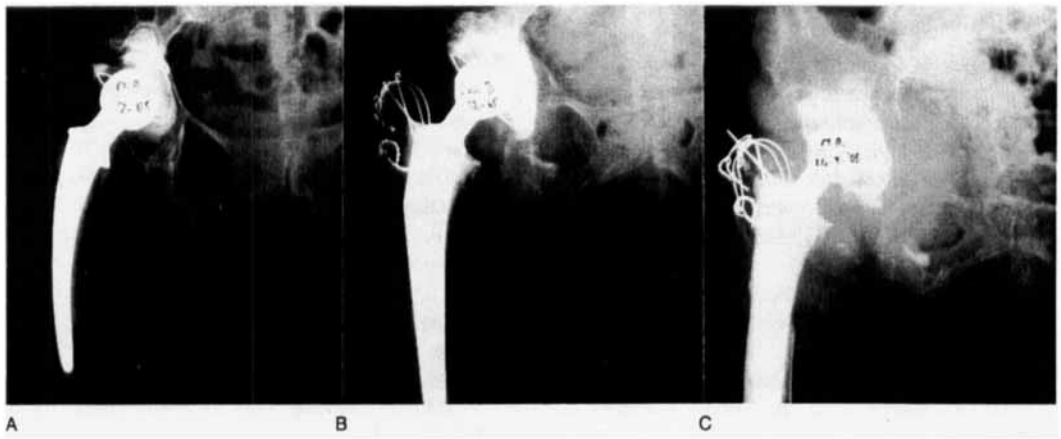

Figure 1. A 72-year-old woman with coxarthrosis underwent total hip arthroplasty in 1974. A progressive osteolysis around both components was first seen in 1977.
A. Status 1985.
B. Four months after revision.
C. Eight months after revision; massive osteolytic tumor in the pelvis and proximal femur.

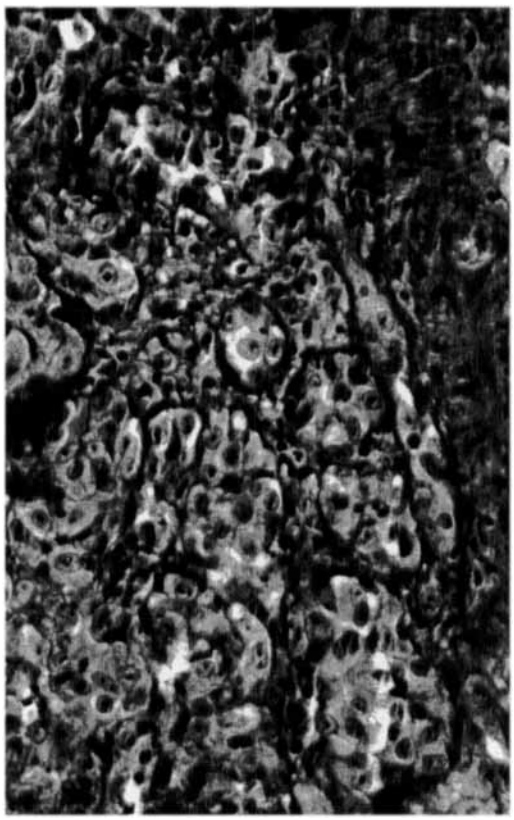

Figure 2. Epithelioid hemangioendothelioma. A proliferation of endothelial cells resembling histiocytes or even epithelial cells are arranged in small nests. Within some of these, a beginning of a central lumen formation can be seen. The tumor cells showed intracytoplasmic staining for Factor VIII-related antigen, a marker for endothelial cells. Elastic van Gieson, ×200.

Discussion. An epithelioid hemangioendothelioma is a vascular tumor, presenting in this case as an angiosarcoma. Angiosarcoma of the bone is rare, representing less than 1 percent of primary malignant bone tumors (Netherlands Committee on Bone Tumours 1966). An angiosarcoma has not been previously described in connection with a total hip replacement, although this tumor is found more frequently at the site of a surgical implant than can be expected (Enneking 1983).

Enneking (1983) reported the unique feature of angiosarcoma in its ability to spread along the medullary canal or across joints into the next bone. In our case, there was probably tumor spillage during the revision operation, because it is likely that the tumor was already in the acetabulum at the time of the operation. The development of a sarcoma in connection with cemented total hip replacement, as in this case, can be coincidental. On the other hand, one should be aware of the possibility in young patients in whom the prosthesis will be in situ over an extended period of time.

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