Angiosarcoma around Hip Joint Prosthesis

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Angiosarcoma, a rare and malignant tumor, is known to be associated with prosthesis and trauma. Up to now, only a few cases related to knee prosthesis and fewer cases related to hip prosthesis have been reported.^[1]

A patient visited our clinic in August 2015 presenting with continuous distending pain in his left lateral thigh. The patient underwent left total hip arthroplasty in 2001 due to bone giant cell tumor, which was $1.0 \text{ cm} \times 0.4 \text{ cm} \times 1.2$ cm restricted to the left femoral head without metastasis. and the implant was provided by polyethylene to titanium alloy, Zimmer Company, Warsaw Indiana, USA. Physical examination revealed an 8 cm × 10 cm fluctuating mass in the proximal end of the thigh. To relieve the pain and swelling, a fine-needle puncture was performed and about 430 ml bloody liquid was drained on September 8, 2015. However, 2 weeks later, the mass recurred and it was bigger than before. A computed tomography scan of the left femur showed an irregular heterogeneous density shadow around the middle and upper area of the left thigh muscle. Coagulation function was normal. Another 800 ml bloody liquid was drained on September 20, 2015.

In October 2015, the hematoma of the left thigh recurred for the third time. A magnetic resonance imaging scan showed soft tissue swelling on the anterior lateral part of the left hip joint [Figure 1], and the bone imaging of ^{99m}Tc-methylene diphosphonate presented the high radioactive uptake on the left femoral head edge, proximal femur, and adjacent soft tissue. The lesion was suspected to conform to operation indication and therefore, the mass was resected and the local structure was reconstructed with a prosthesis with bone cement on November 10, 2015. The pathology revealed inflammatory necrosis and chronic inflammation of fibrous tissue; bone tissue existed in proximal femur; necrosis and



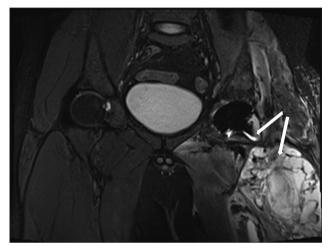


Figure 1: The magnetic resonance imaging of the patient with angiosarcoma. The arrows indicated angiosarcoma around hip prosthesis.

granulation tissue around left hip prosthesis; degeneration and necrosis in the left thigh mass.

Two weeks after the operation, however, an even larger hematoma recurred again in his left thigh, and the patient was unable to maintain in standing position because of myasthenia. The patient complained of numbness on the

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On January 12, 2016, several skin ulcerations emerged with dark, bloody effusions. There was no obvious coagulation dysfunction. Angiography revealed rupture of the left femoral deep artery and treated with arterial embolization. Two days later, the patient spiked a fever with T_{max} of 38.5°C.

A week later, the patient complained of hypesthesia in the left lower limb and decreased muscle strength. The ulcers still existed with a foul odor. Ultrasound revealed a 4.1 cm × 1.0 cm popliteal vein thrombosis. Vancomycin (0.5 g, Q12h) and piperacillin/sulbactam sodium (5 g, Q12h) were applied to this patient.

The features of this patient were summarized as followed: (1) previous history of giant cell tumor leading to left total hip arthroplasty, (2) recurrent hematoma around joint prosthesis with normal coagulation function, and (3) infection of the left thigh skin around hematoma area and risk of septicemia and prosthesis infection. The pathology report of the first specimen did not help in diagnosis.

Considering the possibility that the hematoma affected the blood supply of lower limb and chronic infection of artificial hip joint prosthesis, a filter was inserted in the vena cava and debridement was performed around the hematoma. In the operation, it was noticed that the mass was attached to the femur and prosthesis and invaded the acetabulum. Biopsy of the mass was conducted. Pathology revealed angiosarcoma, with heterotypic cell infiltration and CD31(+)/CD34(+), Ki-67 (about 20%). Antibiotics were applied continuously and chemotherapy was suggested by the department of oncology. However, the patient and his family members declined chemotherapy and other treatments. The patient died of respiratory failure 9 weeks after the operation.

DISCUSSION

Angiosarcoma, a rare and malignant tumor derived from mesenchymal cells, accounts for only 2–4% of all soft tissue sarcomas. However, the etiology of this neoplasm has not been very clear, despite several known risk factors such as traumas and radiation therapy.

The formation of angiosarcoma has been found in relation to Dacron graft as reported in several cases. [2] These cases all involve endovascular lesion with graft implantation made of Dacron. The tumor was described as developing

in contact with the prosthetic material and correlated with traumas existed in vascular in previous articles, which may lead to carcinogenic effect. Fenton *et al.*^[3] also reported an angiosarcoma case derived from embedding AneuRx graft, composed of nitinol exoskeleton sutured to a polyester fabric graft, because of abdominal aortic aneurysm. These cases suggest that angiosarcoma may be one of the complications of artificial grafts implantation because of its material characteristics.

However, there have been few reports on angiosarcoma secondary to prosthesis after joint arthroplasty. A case reported by Drexler et al.[1] also presented late recurrent hematoma as the clinic manifestation after his knee arthroplasty. The tumor grew in the bone near prosthesis, and the patient experienced a lateral tibial plateau fracture with open reduction and plating 25 years before sarcoma occurring. Our case is very similar as that the patient went through left proximal femur resection because of osteoclastoma 14 years ago. Addition to that in our case, the angiosarcoma was found just above trauma area and hip joint prosthesis in the proximal femur. However, most part of the tumor located in soft tissue of left thigh. Moreover, the angiosarcoma was adhered to hip joint prosthesis both in radiography and by eyes during surgery. However, the patient was diagnosed with two types of tumors around proximal femur. The giant cell tumor arose before joint prosthesis implantation and angiosarcoma occurred after arthroplasty. It indicated that the patient might be genetically susceptible to carcinogenesis. Although some studies debated that joints prosthesis could increase the risk of malignancy in normal population, the scenario in cancer susceptible patient may be different.

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Conflicts of interest

There are no conflicts of interest.

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