

CASE REPORT

Osteosarcoma arising from acetabulum extended to femoral head through round ligament: a case report

Toru Sasaki^{1,†}, Tabu Gokita², Keisuke Ae¹, Taisuke Tanizawa¹, Keiko Hayakawa¹, Yuki Funauichi¹, Noriko Motoi³ and Seiichi Matsumoto^{1,*,‡}

¹Department of Orthopedic Oncology, Cancer Institute Hospital, Japanese Foundation for Cancer Research, Tokyo, Japan, ²Department of Orthopedic Surgery, Saitama Cancer Center, Saitama-ken, Japan, ³Division of Pathology, Cancer Institute Hospital, Japanese Foundation for Cancer Research, Tokyo, Japan

*Correspondence address. Department of Orthopedic Oncology, Cancer Institute Hospital, Japanese Foundation for Cancer Research, Ariake, Tokyo 135-8550, Japan. Tel: +81-3-3813-6111; E-mail: smatsumoto@jfc.or.jp

Abstract

A skip metastasis was defined as a solitary separate focus of osteosarcoma occurring synchronously with a primary osteosarcoma in the absence of anatomic extension. The progression of skip metastasis is considered less likely because the articular cartilage acts as a barrier, so there have been few reports on progression of the extremity bone tumor across a joint. In our case report, the acetabular osteosarcoma progressed to the femoral head through the ligament of the femoral head. From the findings of magnetic resonance imaging and resected specimen and tissue specimen, we considered that the tumor progressed between ligament and synovial tissue covering the ligament, and not passing through the inside of the ligament. This case suggested a possibility that the tumor might progress through the synovium around the ligament of femoral head in the cases of osteosarcoma arising from the proximal femur and acetabulum.

INTRODUCTION

In a malignant bone tumor occurring in the long bones of the extremities, a skip metastasis was defined as a solitary separate focus of osteosarcoma occurring synchronously with a primary osteosarcoma in the absence of anatomic extension. This skip metastasis occurs in the same bone as the primary lesion or in the bone adjacent to the primary lesion across a joint [1]. Joint involvement by osteosarcoma is uncommon because articular cartilage being a relative barrier to tumor invasion [2]. Looking at reports on the metastasis across a joint, there have been a few reports on a hip joint. Here, we report a case that acetabular osteosarcoma progressed to the femoral head through the

ligament of the femoral head (round ligament). We obtained written informed consent by the patient.

CASE REPORT

Case: a 63-year-old male was presented with pain in the left proximal thigh. A tumor was suspected by X-ray and magnetic resonance imaging (MRI).

Imaging findings: X-ray showed slight uptake in the left acetabulum (Fig. 1).

Computed tomography showed bone destruction images mainly in the left acetabulum extending to femoral head (Fig. 2).

[†]Toru Sasaki, <http://orcid.org/0000-0003-0939-3296>

[‡]Seiichi Matsumoto, <http://orcid.org/0000-0001-7834-2156>

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Figure 1: Frontal X-ray of the pelvis shows a radiolucent lesion in the left acetabulum.

MRI revealed a multilocular lesion from left acetabulum to pubis (Fig. 3). There was a slight accumulation of joint fluid. A lesion having similar abnormal signal was detected in the femoral head. The tumorous cords were well circumscribed from the surroundings, and there were findings suggestive of the existence of ligament component in the center.

Clinical course: an incision biopsy was performed and a diagnosis of chondrogenic sarcoma, suspect of chondrogenic osteosarcoma, was established. Tumor cells were fusiform or round shaped with atypical nuclei, often binucleated and scattered mitotic figures, suggesting high-grade sarcoma (Fig. 4). After preoperative chemotherapy was started, a deterioration of renal function was observed, and therefore, the chemotherapy (cisplatin + doxorubicin) was stopped after one course. The surgery consisted of wide excision and femoral head replacement for tumor (Fig. 5).

Pathology: gross cut surface of the resected specimen revealed that the tumor in the acetabulum progressed through the round ligament to the femoral head side. The articular cartilage was not destroyed by the tumor, neither on the acetabular side nor on the femoral head side. There was no tumor extending into the joint cavity (Fig. 6a and b). Final diagnosis was chondroblastic osteosarcoma with slight osteoid formation and little therapeutic effect (Fig. 7). Histological findings were similar to the preoperative biopsied specimen. Microscopic examination confirmed the unique extension of tumor cells on the gross findings. The tumor invaded along to the ligament from the acetabular site to the femoral head (Fig. 6c and d). The pathological assessment of preoperative chemotherapy was grade 1 (General Rules for Clinical and Pathological Studies on Malignant Bone Tumors, Japanese Orthopaedic Association), according to the response evaluation criteria [3].

Postoperative course: postoperative chemotherapy (carboplatin and doxorubicin) was administered. As of 3 years after surgery, neither local recurrence nor metastasis has been observed.

DISCUSSION

There have been few reports on progression of the extremity bone tumor across a joint. Simon has reported on the three mechanisms of progression into a joint [4]. First, the tumor may directly invade and cross the articular cartilage. Second, sarcomas may extend around the articular cartilage, beneath the joint capsule, and into the joint. Finally, intra-articular penetration can occur by direct extension of the tumor through or around an osseous-tendinous junction of an intra-articular ligament, usually the cruciate ligaments. In addition, Simon proved progression through cruciate ligament histologically [4, 5]. Since then, progression of osteosarcoma of the hip, shoulder, elbow, ankle [2] and sacroiliac joint [6, 7] have been reported occasionally.

Simon has reported that capillary vessels extending from the femur are distributed in cruciate ligaments and tumors progress through the ligament tissue directly. However, in the case of femoral head, it has been proved that the bloodstream distributes in round ligament disappeared at 6 months old [8],

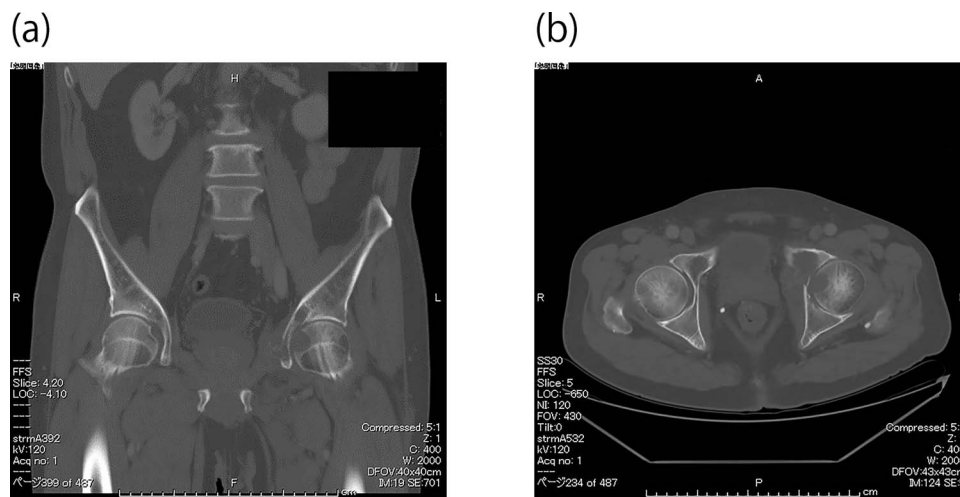


Figure 2: Computed tomography images at the time of visit show bone destruction mainly in the left acetabulum, extending to the femoral head; (a) coronal, (b) axial.

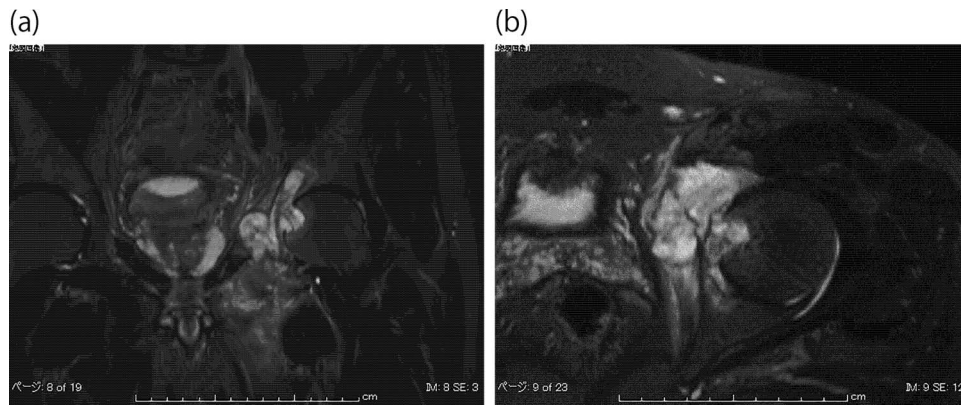


Figure 3: MRI (T2 STIR :Short TI inversion Recovery) shows a multilocular mass mainly in the anterior rim of left acetabulum. (a) Coronal, (b) axial.

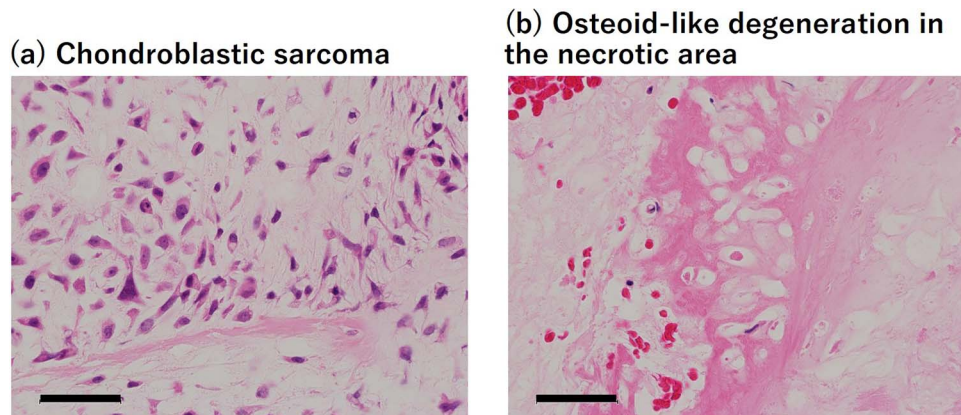


Figure 4: Preoperative biopsy specimen showed high-grade sarcoma, mainly with chondroid matrix. (Hematoxylin and eosin-stained slides, bar = 50 μ m).



Figure 5: Postoperative X-ray: extracapsular resection of the hip joint and femoral head replacement for tumor (KLS System).

and there has been no report on the progression of tumors in detail.

As for reports on progression of acetabular osteosarcoma extending to the femur, there have been only few reports [9, 10]. These reports were cases with no precise imaging findings and pathological findings. There were no reports showing whether or not the tumor had progressed through the round ligament. In our case, MRI revealed little accumulation of joint fluid and the structure of ligament components was maintained, which means that the tumor did not progress into the joint cavity, but progressed to the femoral head surrounding the ligament without destruction of the ligament (Fig. 3). Gross observation of the resected specimen revealed that the tumor did not disseminate in the joint cavity (Fig. 6a). On the other hand, looking at the tissue specimen, the tumor cell in contact with the normal ligament reached the femoral head side, and the cell was considered to progress through some kind of structure adjacent to the ligament (Fig. 6c and d). Considering the findings of MRI and resected specimen and tissue specimen, in this case, we considered that the tumor cell did not disseminate in the joint cavity, but progressed between ligament and synovial tissue covering the ligament (Fig. 6b).

Since synovial fold existed on the surface of normal round ligament, there was a high possibility that the tumor progressed between ligament tissue and synovial fold. In addition, the synovial fold was considered to prevent the tumor from expanding into the joint cavity to some extent. Unlike cruciate ligament, blood vessels in the round ligament in adults had disappeared,

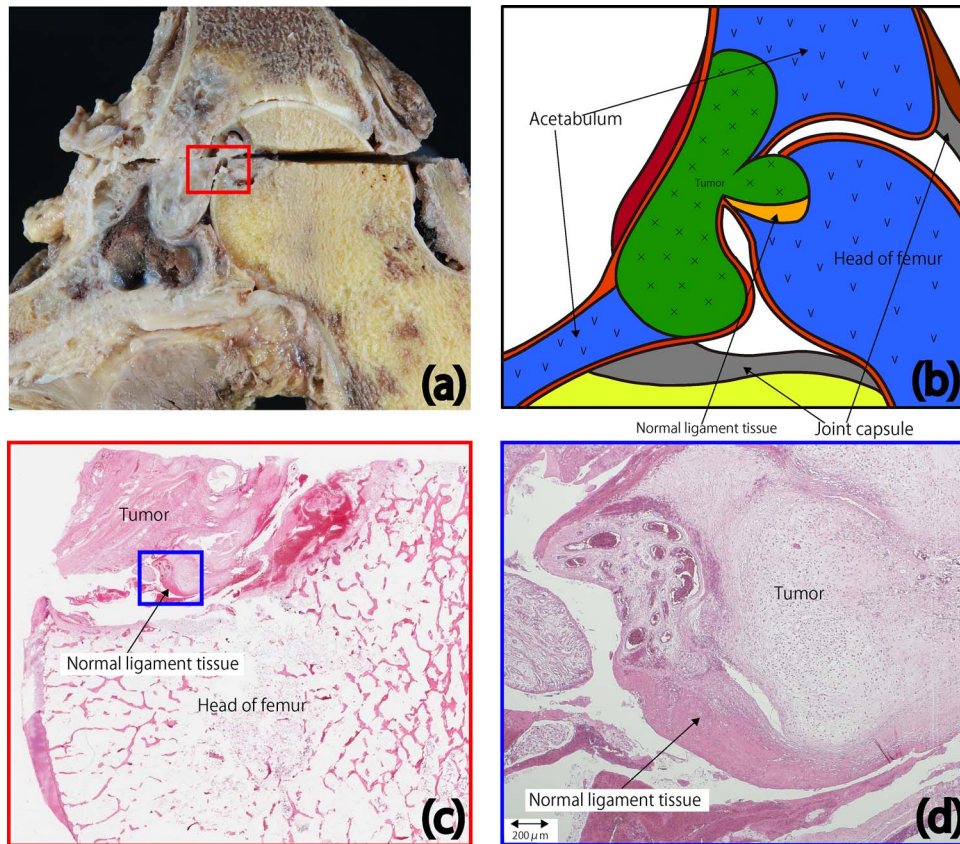
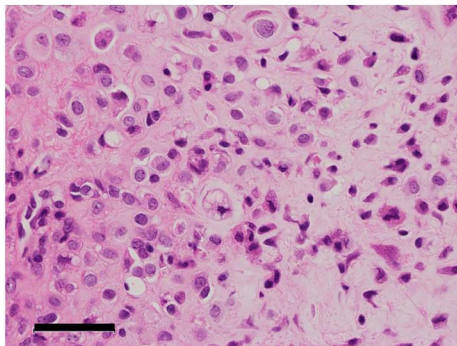


Figure 6: Pathological findings. (a) Macroscopic imaging shows a tumor extending from the acetabulum to the femoral head. (b) Illustration of the macro image shows progression through ligament, but no progression to articular capsule and cartilage. (c) Slightly magnified image of ligament attachment shows the existence of tumor cells on the surface of normal ligament issue. (d) Magnified image shows normal ligament components are in contact with tumor.

(a) Chondroblastic osteosarcoma



(b) Focal osteoid formation

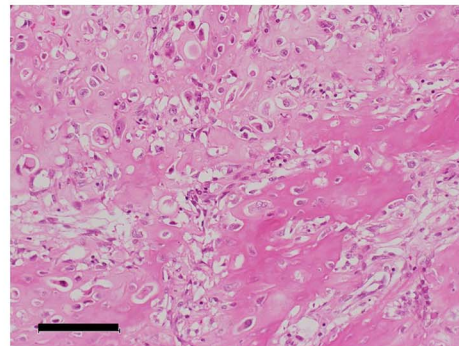


Figure 7: Histology of the resected specimen: Chondroblastic osteosarcoma with nuclear atypia (a) and focal osteoid formation (b) (Hematoxylin and eosin-stained slide, bar (a) = 50 μm , (b) = 100 μm).

and therefore, the tumor might have progressed through microvascular of the synovium between ligament and synovial fold covering the ligament, and not passing through the inside of the ligament.

Based on the findings stated above, this case suggested that the tumor might progress through the synovium around the ligament of femoral head in the cases of osteosarcoma arising from the proximal femur and acetabulum, even if accumulation of joint fluid or invasion to ligament is not obvious by imaging. Therefore, in acetabular osteosarcoma cases, we considered that

it would be important to take into account the existence of the tumor progression when planning surgical margins.

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None.

CONFLICT OF INTEREST

None declared.

FUNDING

None declared.

ETHICAL APPROVAL

No ethical approval required.

CONSENT

The patient has given written consent for publication of this case report.

GUARANTOR

Seiichi Matsumoto, MD, PhD.

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