

Acetabular Osteosarcoma Treated by Irradiation-vascularized Hybrid Bone Graft

TADAHIKO KUBO¹, TAKASHI SUGITA², SHOJI SHIMOSE¹, HARUYUKI TANAKA¹,
HIROO NOBUTO¹, KUMI TANAKA¹ and MITSUO OCHI¹

¹Department of Orthopaedic Surgery, Graduate School of Biomedical Sciences, Hiroshima University;

²Department of Orthopaedic Surgery, Hiroshima Prefecture Hospital, Hiroshima, Japan

Abstract. *Background: Periacetabular reconstruction after malignant bone tumor resection to preserve limb function is extremely challenging. The optimal reconstruction method has not yet received consensus and the functional outcomes still remain unsatisfactory. Case Report: A thirteen-year-old girl who was suffering from acetabular osteosarcoma was treated with wide excision of the tumor, followed by periacetabular reconstruction with an autogenous extracorporeal irradiated osteoarticular graft combined with a vascularized fibula graft. Incorporation of the irradiated pelvic bone was achieved despite infection and limb function turned out to be acceptable. Conclusion: This is the first report on an irradiation-vascularized hybrid autograft to reconstruct massive bone loss in primary limb-sparing surgery.*

Periacetabular reconstruction after malignant bone tumor resection to preserve limb function is extremely challenging. The disruption and mechanical instability of the acetabulum may cause discomfort during walking or movement of the hip. A variety of reconstruction procedures for massive periacetabular bone loss at limb-saving surgery are currently available, including custom-made prosthesis, saddle prosthesis, arthrodesis, pseudoarthrosis, cement and pin or screw fixation, allograft and processed autograft (1-6). The optimal reconstruction method has not yet received consensus and the functional outcomes still remain unsatisfactory. The postoperative course is typically prolonged and morbid and there are generally high frequencies of major complications including infection,

loosening and hip dislocation. In an attempt to overcome those obstacles, we used a hybrid autograft consisting of an irradiated osteoarticular and a vascularized fibula to restore the hemipelvis after periacetabular resection.

Case Report

A thirteen-year-old girl had a six-month history of activity-related left buttock pain. She presented with a sharp pain in the buttock region when jumping up and down. Physical examinations did not reveal anything remarkable. The serum alkaline phosphatase value was 432 units/L, which is higher than the reference range (90-340 units/L). Plain radiographs showed an ill-defined osteolytic lesion involving the left acetabulum. The expanding destruction of the medial pelvic wall was apparent (Figure 1). Magnetic resonance (MR) images showed that an extensive Gadolinium-enhanced lesion was occupying the left whole iliac bone except the iliac crest. It also extended across the left iliosacral joint and into surrounding soft tissues (Figure 2A, B). There was no detectable distant metastasis evaluated on chest computed tomography (CT) scans and bone scintigraphs of the entire body. Histological examination of the biopsy specimens diagnosed small cell osteosarcoma, which is characterized by a proliferation of small round cells yielding osteoid (Figure 3). A negative periodic acid-Schiff stain and non-detection of the EWS-FLI1 fusion gene were consistent with the features of small cell osteosarcoma. The surgical stage was defined as IIB according to the Musculoskeletal Tumor Society classification (7). The patient received neoadjuvant chemotherapy comprising doxorubicin, cisplatin and ifosfamide, which resulted in densely sclerotic alteration of the cortical lesion and the depletion of a surrounding soft mass.

With the estimation of a good response to chemotherapy, limb-sparing surgery with wide excision and implantation of a hybrid autograft consisting of an irradiated osteoarticular and a vascularized fibula were performed (Figure 4A). The left whole pelvic bone containing the tumor was taken away along with the left iliosacral joint and the intact cuff of

Correspondence to: Dr. Tadahiko Kubo, Department of Orthopaedic Surgery, Graduate School of Biomedical Sciences, Hiroshima University, 1-2-3 Kasumi, Minami-ku, Hiroshima, 734-8551, Japan. Tel: (+81) 82-257-5232, Fax: (+81) 82-257-5234, e-mail: kubot@hiroshima-u.ac.jp

Key Words: Acetabular osteosarcoma, pelvic reconstruction, vascularized fibula graft, irradiated osteoarticular graft.



Figure 1. A plain radiograph of the pelvis at presentation showed an ill-defined osteolytic lesion predominantly involving the left acetabulum.

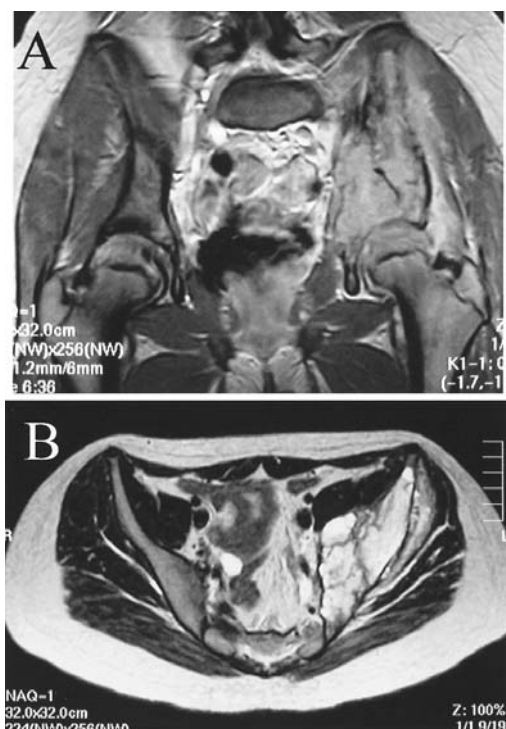


Figure 2. (A) A frontal view of enhanced T2-weighted MR image showed that an extensive lesion was occupying the whole iliac bone except the iliac crest. (B) An axial MR image showed the invasion across the iliosacral joint and the extension into surrounding soft tissues.

normal soft tissue surrounding the lesion. The capsule of the hip joint was circumferentially incised to ensure no intraarticular invasion by the tumor. The pelvic bone was stripped of the attached soft tissue except the capsule and muscle origins of hip flexors, abductors and extensors for

reparation at the time of reimplantation. The bulk of the intramedullary tumorous tissue was curettaged, then the remaining pelvis packed in the container filled with physiologic saline was extracorporeally irradiated at one fraction of 50 Gy dose, using a linear accelerator. The irradiated pelvis was returned to the original site of the host with a reconstruction plate immobilizing the pubic symphysis and three large cancellous screws firmly fixing the osteotomy site in the sacrum. A free vascularized fibula obtained from the ipsilateral limb was grafted along the medial wall of the irradiated pelvis to enhance the stability of the pelvic ring. Cancellous bone chips from the contralateral iliac crest were stuffed into the cavity right above the subchondral bone of the acetabulum in order to sustain weight-bearing force. The capsule and major muscles of the hip were repaired to the corresponding sites of preserved attachments or through drill holes in the iliac crest. An artificial peritoneum was used to cover the obturator foramen. The pathological evaluation of the curettaged tumor revealed total necrosis with no viable tumor cell remaining.

The patient had a high fever ($>38^{\circ}\text{C}$) immediately after the operation and a wound penetrating to the left pubic bone 2 weeks later. The exudate culture identified *Staphylococcus epidermidis* and a 10-week administration of combined anti-bacterial agents sedated the infection. Adjuvant chemotherapy comprising doxorubicin and cisplatin was performed subsequently.

Radiographs taken two months after surgery detected diffuse bone atrophy within the left femoral head. Serial radiographs showed that the collapse and shrinkage of the femoral head as well as the iliac crest had progressed. Six months postoperatively, however, the sclerotic and thickening changes had occurred predominantly around the acetabulum and grafted fibula, at which point partial weight-bearing was allowed. At the most recent follow-up visit four years postoperatively, the patient was pain-free and ambulatory, using crutches. Hip movement was slightly restricted (flexion, 80° ; abduction, 20° ; extension, 0°) and the functional score was 50% according to the Musculoskeletal Tumor Society rating system (8). There was no further bone resorption and the structure of the pelvic ring was maintained (Figure 4B).

Discussion

Osteosarcoma of the pelvis is rare, comprising less than 10% of all osteosarcomas. The therapeutic outcome for pelvic osteosarcoma has been reported as unfavorable. Several large series of studies has shown the 5-year overall survival to be between 18% and 34%, which is much worse than that of extremity osteosarcoma (9-11). Since intralesional excision was noted to be a significantly poor prognostic factor in stage IIB osteosarcoma of the pelvis, limb-sparing surgery should

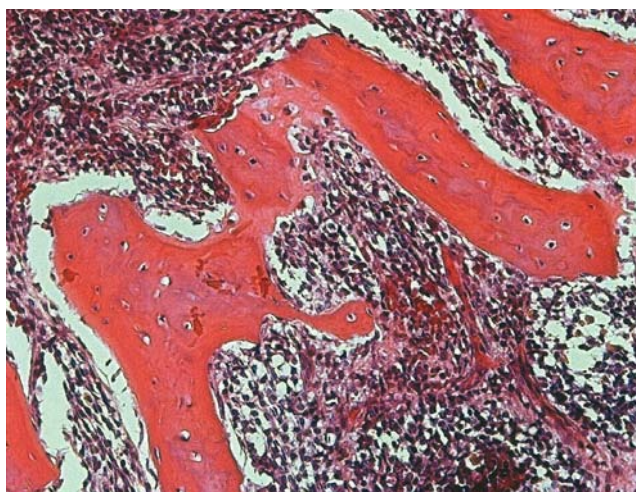


Figure 3. A photomicrograph of biopsy specimens revealed the features of small cell osteosarcoma with a proliferation of small round cells producing osteoid (Stain, hematoxylin and eosin; magnification, X100).

not jeopardize an adequate resection margin of the tumor. In this regard, reconstruction with an irradiated autogenous bone graft is oncologically safer than a prosthetic implantation. Custom-made endoprotheses are made based upon preoperative imaging data, which may not reflect the situation at surgery when more extensive resection than planned may be required, whereas irradiated autogenous bones can be modulated according to the actual involvement of bone so that the resection margin of the tumor is not restricted.

An intraoperative extracorporeal irradiated bone graft, one of processed autografts, was first reported by Uyttendaele *et al.* (12) in 1988 to reconstruct large amounts of bone defects after the excision of primary bone tumors. It not only has the theoretical advantages of being a biological material like an allograft, but also anatomically it adapts ideally to host bones with no risk of either an immunological response or disease transmission. Biological materials are supposed to improve the functional outcome by soft tissue adhesion and to enhance long-term survival by the biological capacity of the healing process to host bones.

The complication rate of irradiated bone grafts is relatively high. Araki *et al.* (13) reported that nonunion (20%) and infection (15%) were the two major complications in 20 patients treated with irradiated bone or osteoarticular grafts. Chen *et al.* (14) documented a similar incidence of failures, which included fractures at the rate of 26.3%, loss of articular cartilage at 21.1%, nonunion at 10.5% and wound infection at 10.5% in a total of 19 osteoarticular grafts. In an attempt to reduce these complications under the harshest circumstances of pelvic reconstruction, the complication rate of which is well known

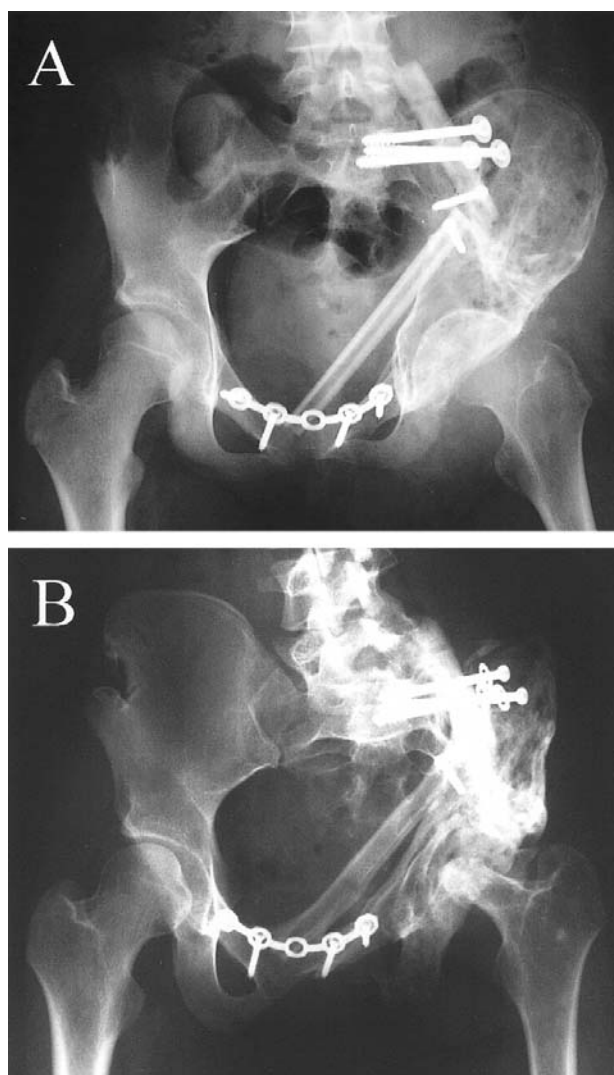


Figure 4. (A) An anteroposterior radiograph right after surgery. Limb-sparing surgery with wide excision and implantation of a hybrid autograft consisting of an irradiated osteoarticular and a vascularized fibula were performed. (B) Four years postoperatively, the structure of the pelvic ring was maintained. The collapse and shrinkage of the femoral head had progressed.

to be extremely high, we used a free vascularized fibula graft in addition to an irradiated osteoarticular graft to restore the hemipelvis after periacetabular resection. Vascularized bone can be expected to prevent large amounts of dead space or haematomas and provide the vascularity of the avascular graft bed and the mechanical stability of the host-graft junction. Despite all cautionary measures, the patient had deep infection, which was cured by the administration of combined anti-bacterial agents. The vascularized fibula may contribute to the healing process without requiring retrieval of the grafted pelvis.

In this case, the union of the host-graft junction and the incorporation of the reimplant was achieved radiologically 6 months after the surgery. Meanwhile, postoperative avascular necrosis of the femoral head had occurred, possibly due to the infection or damage of the medial femoral circumflex artery during the procedure. Clinical data on the state of the articular cartilage after irradiation are scant. Takahashi *et al.* (15) reported the high tolerance of articular cartilage to irradiation of 50 Gy in a rabbit model, but degenerative changes have been addressed in dog studies by other researchers (16). The current hip function of our patient is tolerable even as a physically active high school student, but the deformity of the femoral head and the predictable progression of degenerative changes of the articular cartilage will require salvage surgery.

This is the first report on a hybrid autograft consisting of an irradiated osteoarticular and a vascularized fibula to reconstruct massive bone loss in primary limb-sparing surgery. The incorporation of the irradiated pelvic bone was achieved despite infection and limb function turned out to be acceptable. This procedure is a good alternative to consider when treating patients with some perturbing tumors like acetabular osteosarcoma.

References

- 1 Abudu A, Grimer RJ, Cannon SR, Carter SR and Sneath RS: Reconstruction of the hemipelvis after the excision of malignant tumours. Complications and functional outcome of prostheses. *J Bone Joint Surg* 79B: 773-779, 1997.
- 2 Langlais F, Lambotte JC and Thomazeau H: Long-term results of hemipelvis reconstruction with allografts. *Clin Orthop* 388: 178-186, 2001.
- 3 Satcher RL Jr, O'Donnell RJ and Johnston JO: Reconstruction of the pelvis after resection of tumors about the acetabulum. *Clin Orthop* 409: 209-217, 2003.
- 4 Schwameis E, Dominkus M, Krepler P, Dorotka R, Lang S, Windhager R and Kotz R: Reconstruction of the pelvis after tumor resection in children and adolescents. *Clin Orthop* 402: 220-235, 2002.
- 5 Sys G, Uyttendaele D, Poffyn B, Verdonk R and Verstraete L: Extracorporeally irradiated autografts in pelvic reconstruction after malignant tumour resection. *Int Orthop* 26: 174-178, 2002.
- 6 Windhager R, Karner J, Kutschera HP, Polterauer P, Salzer-Kuntschik M and Kotz R: Limb salvage in periacetabular sarcomas: review of 21 consecutive cases. *Clin Orthop* 331: 265-276, 1996.
- 7 Enneking WF: A system of staging musculoskeletal neoplasms. *Clin Orthop* 204: 9-24, 1986.
- 8 Enneking WF, Dunham W, Gebhardt MC, Malawar M and Pritchard DJ: A system for the functional evaluation of reconstructive procedures after surgical treatment of tumors of the musculoskeletal system. *Clin Orthop* 286: 241-246, 1993.
- 9 Ozaki T, Flege S, Kevric M, Lindner N, Maas R, Delling G, Schwarz R, von Hochstetter AR, Salzer-Kuntschik M, Berdel WE, Jurgens H, Exner GU, Reichardt P, Mayer-Steinacker R, Ewerbeck V, Kotz R, Winkelmann W and Bielack SS: Osteosarcoma of the pelvis: experience of the Cooperative Osteosarcoma Study Group. *J Clin Oncol* 21: 334-341, 2003.
- 10 Grimer RJ, Carter SR, Tillman RM, Spooner D, Mangham DC and Kabukcuoglu Y: Osteosarcoma of the pelvis. *J Bone Joint Surg* 81B: 796-802, 1999.
- 11 Kawai A, Huvos AG, Meyers PA and Healey JH: Osteosarcoma of the pelvis. Oncologic results of 40 patients. *Clin Orthop* 348: 196-207, 1998.
- 12 Uyttendaele D, De Schryver A, Claessens H, Roels H, Berkvens P and Mondelaers W: Limb conservation in primary bone tumours by resection, extracorporeal irradiation and re-implantation. *J Bone Joint Surg* 70B: 384-353, 1988.
- 13 Araki N, Myoui A, Kuratsu S, Hashimoto N, Inoue T, Kudawara I, Ueda T, Yoshikawa H, Masaki N and Uchida A: Intraoperative extracorporeal autogenous irradiated bone grafts in tumor surgery. *Clin Orthop* 368: 196-206, 1999.
- 14 Chen WM, Chen TH, Huang CK, Chiang CC and Lo WH: Treatment of malignant bone tumours by extracorporeally irradiated autograft-prosthetic composite arthroplasty. *J Bone Joint Surg* 84B: 1156-1161, 2002.
- 15 Takahashi S, Sugimoto M, Kotoura Y, Oka M, Sasai K, Abe M and Yamamuro T: Long-lasting tolerance of articular cartilage after experimental intraoperative radiation in rabbits. *Clin Orthop* 275: 300-305, 1992.
- 16 Sabo D, Brocai DR, Eble M, Wannemacher M and Ewerbeck V: Influence of extracorporeal irradiation on the reintegration of autologous grafts of bone and joint. Study in a canine model. *J Bone Joint Surg* 82B: 276-282, 2000.

Received January 5, 2004

Accepted March 8, 2004