

Bipolar Head Perforation With Rhabdomyosarcoma of the Thigh: A Case Report With Literature Review

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Abstract. *Background/Aim:* High complication rates during the perioperative management of sarcomas around the pelvis have been reported; however, few include the detailed clinical course or complications in the late postoperative period. Radiotherapy is a multidisciplinary strategy for treating sarcomas. However, irradiated bone and soft tissues show a permanent loss of repair and immunocompetence. We present a case of pleomorphic rhabdomyosarcoma of the thigh that resulted in acetabular collapse induced by radiation and intestinal perforation during long-term follow-up. Additionally, we discuss the risk factors for late complications and pelvic reconstruction methods. *Case Report:* A 75-year-old man presented with a 1-month history of a recurring fever. Ten years prior, he was diagnosed with pleomorphic rhabdomyosarcoma of the right thigh and underwent a wide resection and bipolar hip arthroplasty, followed by chemotherapy and radiotherapy. Radiographs showed central dislocation of the bipolar head. Computed tomography revealed free air in the hip joint and thickening of the colon wall. Colonoscopy revealed displacement of the bipolar head into the colon wall. Colon resection and hip disarticulation were performed, as the bipolar head was contaminated with intestinal contents. Currently, he

is able to walk stably with a walker, and there is no evidence of recurrence or metastasis of the tumor. *Conclusion:* Irradiation of the periacetabular bone may induce resorptive destruction, resulting in future structural failure. Hardware should not be used for periacetabular bone reconstruction; the risk of pelvic organ damage should be considered when the acetabular collapse becomes deteriorated. Therefore, other reliable and permanent reconstruction options are required.

The complication rate for surgery of sarcomas around the pelvis is 1.5-3 times higher than those of the extremities (1). Postoperative organ symptoms, such as dysuria and neuropathy (2), intraoperative vascular and organ damage (3), postoperative infection and skin necrosis (4) have been reported; these occur mainly in the early postoperative period.

To treat sarcomas, radiotherapy is performed as an adjuvant, in addition to surgery and chemotherapy, to improve local control. Radiation causes cellular damage to both cancer cells and normal cells surrounding the tumor, resulting in acute and late adverse events (5). Osteonecrosis of the bone and cartilage is a late adverse event that occurs over a long period, ranging from months to years, with irreversible progression, complicating treatment and considerably reducing patients' quality of life (6).

We herein present a case of pleomorphic rhabdomyosarcoma of the thigh that resulted in acetabular collapse induced by radiation and intestinal perforation during long-term follow-up, and led to colon resection and hip disarticulation. We also aimed to discuss the risk factors for late complications and pelvic reconstruction methods. The patient was informed that data from the research would be submitted for publication and gave his consent.

Case Report

A 75-year-old man presented with a palpable mass in the right thigh and was diagnosed with pleomorphic rhabdomyosarcoma

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Key Words: Pelvic perforation, rhabdomyosarcoma, periacetabular bone reconstruction.

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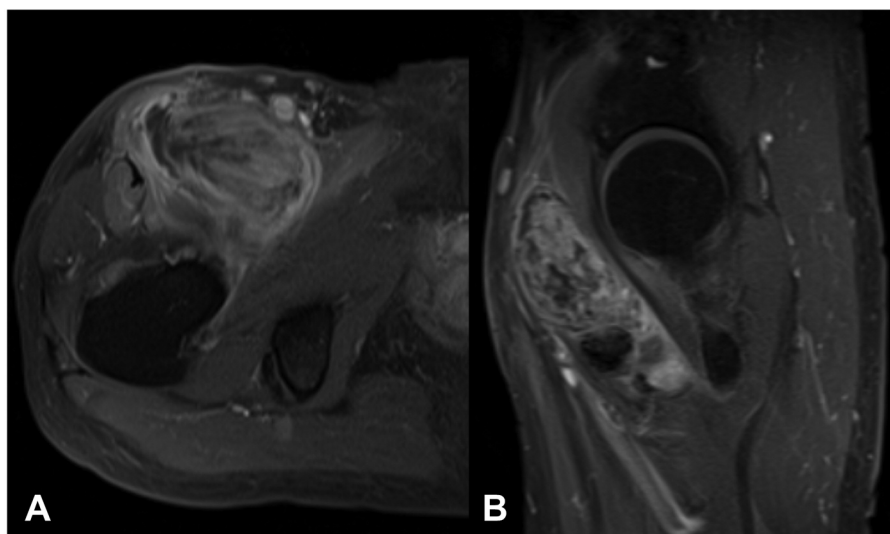


Figure 1. Preoperative gadolinium-enhanced magnetic resonance images (A) axial and (B) sagittal.

after an open biopsy. Neither distant nor lymph node metastases were detected during the primary diagnosis. After one course of doxorubicin ($20 \text{ mg/m}^2 \times 3$) and ifosfamide ($2.5 \text{ mg/m}^2 \times 5$) at a previous hospital, he was referred to our hospital for surgery. The sarcoma was localized within the iliopsoas muscle and measured at $10 \times 6.5 \times 3 \text{ cm}$. The femoral artery and vein connected with the anterior side of the sarcoma (Figure 1A and B). Thus, we planned wide resection for limb salvage.

Intraoperatively, the proximal femur, with the sarcoma located 3 cm distal to the lesser trochanter, was resected. After resection of the iliopsoas, sartorius, rectus, gluteus medius, adductor magnus, pectineus, and femoral veins (Figure 2A), the hip joint and soft tissue defects were replaced and reconstructed using a bipolar head (KLS system; Kyocera Medical, Kyoto, Japan), polypropylene mesh (Bard mesh; Medicon, Osaka, Japan), and gracilis muscle flap (Figure 2B). The surgical margin was wide (2 cm); however, the dissected external iliac lymph node was positive for sarcoma cells. Therefore, radiotherapy ($60 \text{ Gy}/30 \text{ Fr}$) was administered to the right pelvic lymph node 2 months postoperatively. Doxorubicin ($25 \text{ mg/m}^2 \times 3$) and ifosfamide ($1.8 \text{ mg/m}^2 \times 5$) was administered as adjuvant chemotherapy but was terminated midway owing to persistent wound infection and bacteremia. In subsequent periodic radiographic follow-up, no significant abnormality in the position of the bipolar head was observed.

Although radiographs showed that the femoral head had migrated upward 5 years postoperatively (Figure 3B), compared with that during the first postoperative week (Figure 3A), the patient was able to walk on his own. No recurrence or metastasis was observed during systemic follow-up.

Ten years after the surgery, the patient was urgently admitted to the hospital owing to a recurrent fever for a month, dull pain around the right hip joint, and difficulty in physical movement. Radiography showed central dislocation of the right hip joint (Figure 4A), and computed tomography (CT) revealed a perforation of the acetabulum by the bipolar head, with air contamination and thickening of the colon wall surrounding the bipolar head (Figure 4B), suggesting intestinal tract perforation. A colonoscopy and laparoscopy further revealed a migrated bipolar head within the ascending colon (Figure 5A and B). As the bipolar head was contaminated with intestinal contents, colon resection and right hip disarticulation were performed. The soft tissue specimens from the dissected right hip joint contained *Enterobacter cloacae*. One year after the last surgery, he is now waking stably with a walker. There has been no apparent recurrence or metastasis of the tumor.

Discussion

In the current case, the irradiated pelvic bone and wound infection and bacteremia, led to a massive bone resorption in the acetabulum which progressed over time. Ten years after the primary surgery, the acetabulum collapsed, resulting in the perforation of the abdominal cavity and ascending colon by the bipolar head. This is a rare complication and several hypotheses can be considered for its pathogenesis.

First, the acetabulum may have been destroyed due to radiation-induced osteonecrosis, followed by perforation of the colon by the bipolar head. Radiation osteonecrosis is a progressive necrosis of the bone and cartilage following

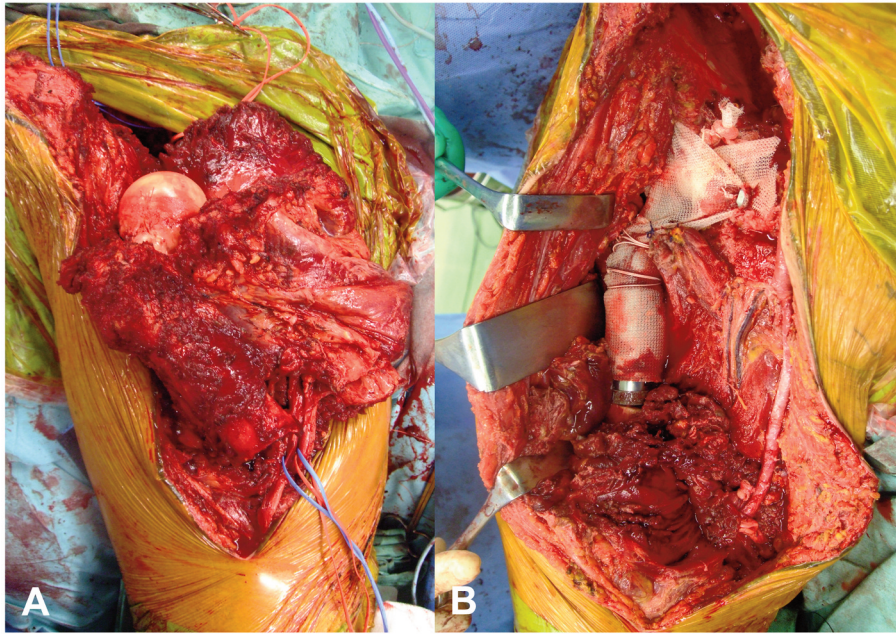


Figure 2. Intraoperative macroscopic images. (A) The femoral artery and vein were continuous after the femur and surrounding muscles were resected. (B) Bipolar head replacement and reconstruction of the hip joint capsule with mesh and gracilis muscle flap.

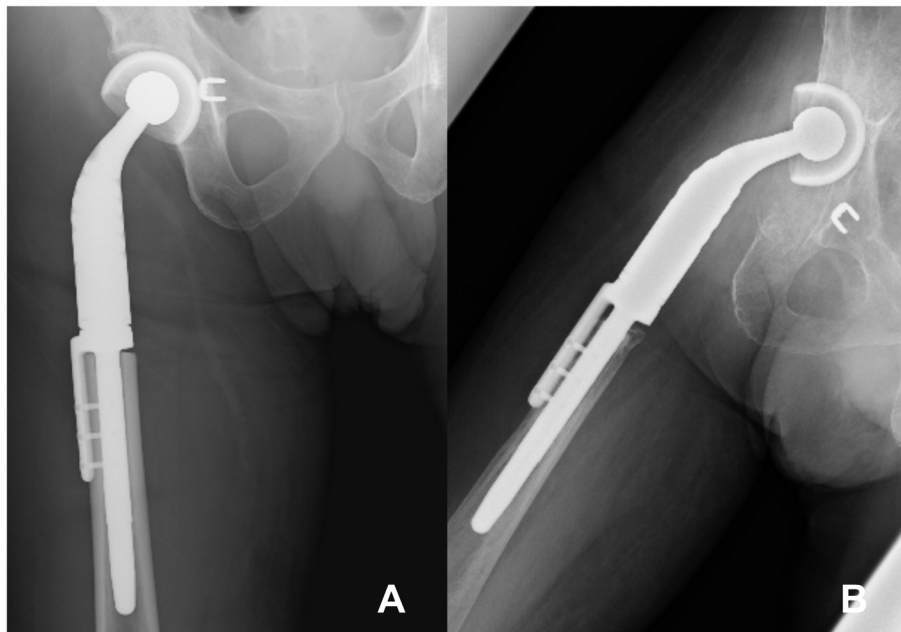


Figure 3. Postoperative X-ray images of the right hip joint (anteroposterior view). (A) One week after surgery. (B) Five years after surgery: the bipolar head migrated upward into the abdominal cavity.

radiotherapy. Marx *et al*. (5) described this as the irreversible progression of hypoxia, hypocellularity, and low blood flow in the tissue due to radiation-induced cell damage, resulting in a chronic loss of healing mechanisms. Delanian and

Lefaix (6) also proposed that radiation-induced fibroblast activation and dysregulation leads to hypocellularity, low blood flow, and fibrosis, resulting in fragile tissue. Radiation osteonecrosis can occur in 0.3%-0.4% of irradiated cases.

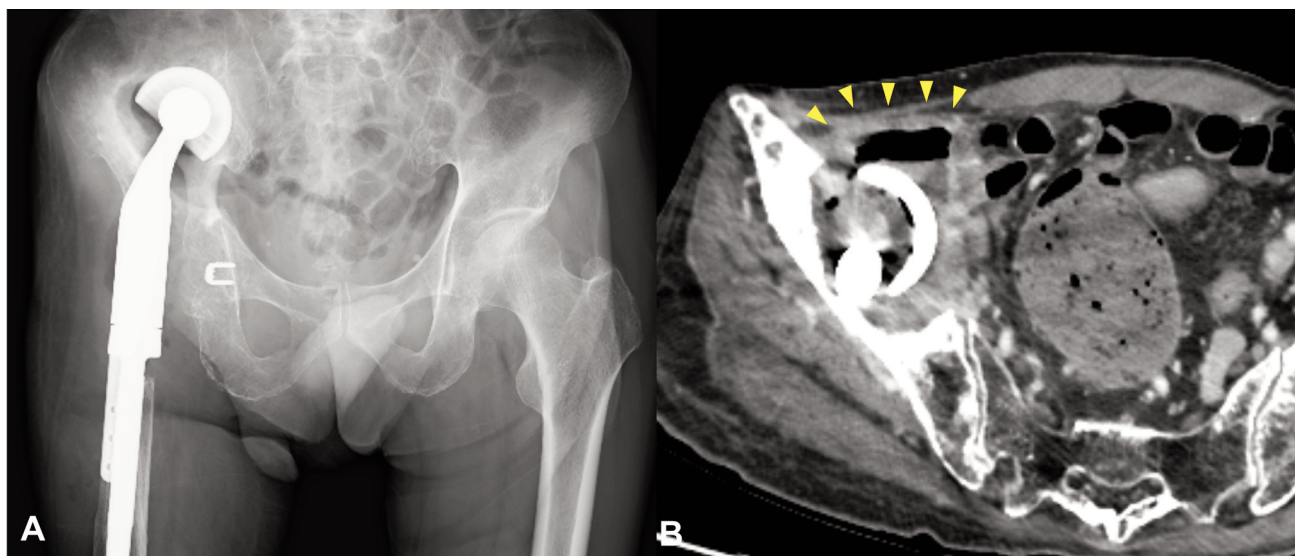


Figure 4. Postoperative images after 10 years. (A) X-ray image (anteroposterior view) showing central dislocation of the bipolar head. (B) Contrast-enhanced computed tomography image (axial) showing the bipolar head perforating the acetabulum with free air contamination and thickening of the surrounding colon wall (arrowheads).

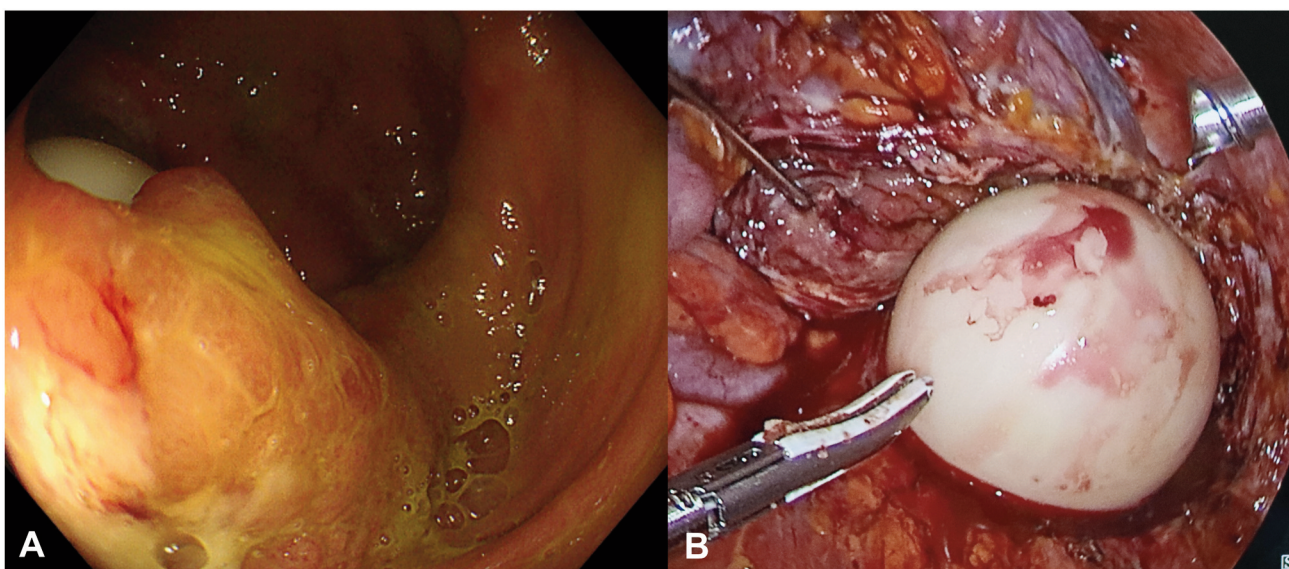


Figure 5. Postoperative scopic views: (A) colonoscopy and (B) laparoscopy images. The bipolar head migrated into the ascending colon.

Furthermore, a higher the risk of developing osteonecrosis has been reported with higher doses, especially with ≥ 30 Gy (7). Moreover, Kokubo *et al.* stated that acetabular necrosis typically occurs 7-16 years after radiotherapy, while femoral head necrosis typically occurs within 3 years (8). In the present case, 60 Gy of postoperative radiation therapy was performed to the acetabulum due to the metastasis in the

external iliac lymph node; its destruction occurred 10 years after the surgery. Radiation osteonecrosis could cause massive bone resorption in the acetabulum.

Second, the colon was perforated due to chronic radiation enteritis, followed by osteomyelitis of the acetabulum and central dislocation of the bipolar head. Radiation therapy may lead to intestinal wall injury with incapacitating

functional sequelae (9). Radiation enteritis is classified into reversible early damage, which is a local circulatory disturbance caused by edema in the intestinal epithelial cells, and irreversible late damage, a microcirculatory disturbance caused by vessel wall thickening due to arterial endarteritis, resulting in fibrosis and atherosclerotic changes (10). Late damages are often reported 9-24 months after irradiation (11), although some cases have been reported at 7 and 17 years after irradiation, as in the present case (12, 13).

Third, late onset infection of the bipolar head occurred followed by destruction of the acetabulum and perforation of the colon. *E. cloacae* detected in the dissected right periprosthetic soft tissue specimen possibly originated from the perforated intestinal tract. However, the radiation-induced immunocompromised acetabulum may have developed a chronic infection.

Although it is not certain which of the three hypotheses is correct, this case suggests that bone fragility increases from decreased local immunity and osteonecrosis following radiotherapy. Furthermore, the risk of serious complications due to pelvic organ damage increases, especially in cases that progress to central dislocation of the hip structure.

There have been reports of acetabular reconstruction using allografts, autologous irradiated bone, and allograft prosthetic composite materials (14-16). However, allografts are prone to nonunion and bone resorption (14-17), and once an infection occurs, treatment is difficult, and there are few options other than resection of the allograft (18). Moreover, there have been several reports of saddle prosthesis (19), total hip arthroplasty (THA) (20), and endoprosthesis with a pedestal cup to preserve the affected limb (21); however, all have complications, such as dislocation, infection, and implant loosening. To overcome these obstacles, acetabular reconstruction using vascularized bone grafts (22, 23) and cellular grafts, such as autologous fresh bone grafts (24) have been reported. However, postoperative failure of the reconstructed structure, arthropathic changes, and infections remain problems. Recently, Houdek *et al.* (25) compared the clinical outcomes of patients who underwent THA with those who did not undergo any acetabular reconstruction surgery for wide resection of pelvic chondrosarcomas. The Musculoskeletal Tumor Society score was higher in the reconstruction group with THA; however, the frequency of postoperative complications was also higher in the reconstruction group, and the final walking ability did not differ between the two groups.

Great clinical results have been reported for hip transposition, in which no initial attempt has been made to reconstruct the acetabulum (26, 27). Hip transposition was originally invented as a salvage procedure when other reconstruction techniques failed. However, owing to its low postoperative infection rate and risk of dislocation or other implant problems, some authors have recently recommended it as the first choice for acetabular reconstruction in wide

pelvic resection (28). The modified hip transposition reported by Xu *et al.* (29) addresses the problem of shortening of the affected limb, considered the biggest drawback of hip transposition, by rotating the femur.

In this case, if THA had been performed at the initial surgery, the wear on the sliding surface of the bipolar head would have been reduced and the central dislocation might have been prevented. However, THA for the single femoral resection was difficult as pelvic lymph node metastasis was detected postoperatively, leading to changes in our treatment plan to add radiotherapy to the pelvic area. Even with THA, irradiation causes fragility of the acetabulum and, ultimately, failure of the hip structure.

Conclusion

Hip joints with a history of irradiation may develop progressive resorptive destruction and structural failure. When considering surgery for sarcoma around the pelvis that involves the resection of hip structures, and if adjuvant radiotherapy is anticipated, reconstructive methods that consider late post-irradiation osteoarticular complications should be devised, and careful long-term follow-up is important.

Conflicts of Interest

The Authors declare that they have no conflicts of interest.

Authors' Contributions

Akane Ariga: Conceptualization; formal analysis; Investigation; Data curation; Writing-original draft; Writing-review and editing. Taisuke Tanizawa: Investigation. Shingo Sato: Investigation. Yuki Funauchi: Investigation. Seiichi Matsumoto: Investigation. Keisuke AE: Investigation; writing-review and editing. Project administration; supervision.

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