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CASE REPORT
Reconstructive

Sacral Ulcer after Carbon Ion Radiotherapy Reconstructed with a Superior Gluteal Artery Perforator Flap

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Summary: As carbon ion radiotherapy (CIRT) was developed only recently, reports of CIRT-induced ulcers requiring plastic surgery are still rare, but the number of such cases is expected to increase. Here, we describe a case of a CIRT-induced ulcer to aid the treatment of such ulcers. An 82-year-old man had a sacral chordoma (12×7.5×7.5 cm), which extended from the fourth to fifth sacral vertebrae. He underwent CIRT (70.4 Gy). An ulcer developed 30 months after the treatment. The ulcer enlarged to 13 cm × 7 cm. Debridement, negative-pressure wound therapy, and antibiotics were used. We tried to avoid injuring the rectum and sciatic nerve, and covered the ulcer with a delayed superior gluteal artery perforator flap. Wound healing was difficult to achieve in the lower half of the flap. Further debridement was appropriate, but we considered that it was likely to cause complications. Once a post-CIRT ulcer develops, its progression and the required extent of debridement can be roughly predicted based on the radiotherapy treatment plan. In this case, the rectum and sciatic nerve were irradiated, but there were no related symptoms. Therefore, we performed surgery to preserve these structures. However, there was very thick scar tissue surrounding these structures, making debridement difficult, and the wound was slow to heal. It is desirable to use a flap with good blood flow, such as a myocutaneous flap, for covering post-CIRT ulcers.
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Surgery is the first-choice definitive treatment for chordoma, but carbon ion radiotherapy (CIRT) is often selected because the affected region is anatomically complex, and complete resection may be difficult.[1–3] CIRT results in a high-dose concentration, with the surrounding normal tissues being exposed to low radiation doses. However, it does have side effects. Of the 219 chordoma patients who received CIRT (mean radiation dose: 67.2 Gy), 6% exhibited grade 3 or higher delayed radiation injuries at a median follow-up period of 56 months.[4] Moreover, defecation problems,[1,2] peripheral neuropathy, and sacral insufficiency fractures[5] have been reported.

As CIRT, which delivers intense localized irradiation, is relatively new, there are few reports of CIRT-induced ulcers requiring plastic surgery.[1,2]

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When treating in the present case, we found it difficult to balance the various competing factors; that is, the patient's comorbidities (anemia and dementia), the possibility of tumor recurrence, the degree of debridement, and the choice of reconstruction method. Specifically, the rectum and sciatic nerve were irradiated, but there were no symptoms or necrosis, and a very thick scar was observed in this area. We report our experiences.

CASE

The patient was an 82-year-old man. Eight years ago, he had a sacral chordoma (12×7.5×7.5 cm), which extended from the fourth to fifth sacral vertebrae and had invaded ventrally until it was adjacent to the rectum and dorsally to the gluteus maximus muscle. He underwent CIRT (70.4 Gy/32 fractions; Fig. 1). Three years earlier, he had undergone surgery for left lung metastasis. Thirty months after the CIRT, a skin ulcer (diameter: 2 cm) developed on his sacral area. The ulcer enlarged,

Disclosure statements are at the end of this article, following the correspondence information.

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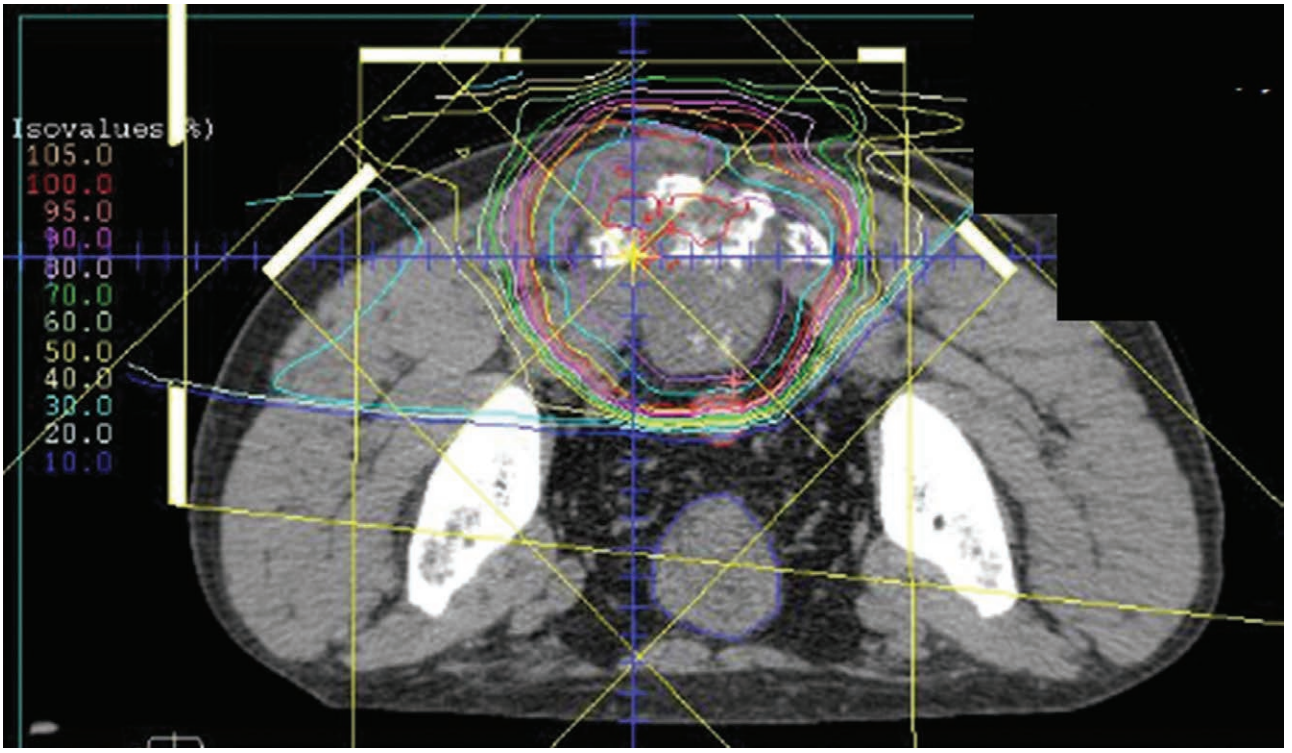

Fig. 1. Planning of the CIRT. The area within the red line indicates that the 100% isovalue included parts of the rectum and the left sacral plexus.

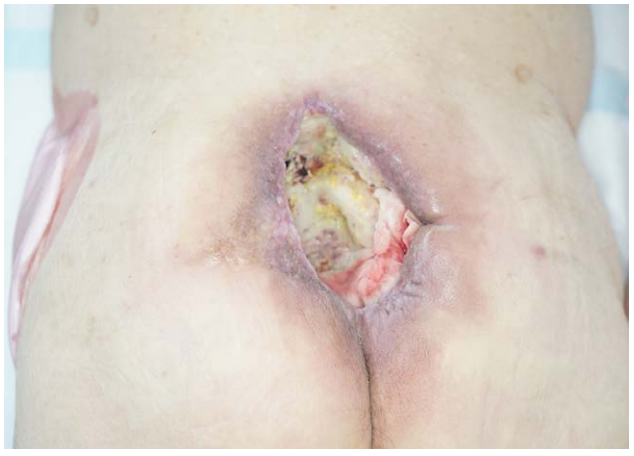

Fig. 2. First visit to our hospital. The ulcer measured 13 cm × 7 cm, and pockets of adherent necrotic tissue had formed on the left and right sides. The third, fourth, and fifth sacral vertebrae were absent.

and he visited another plastic surgeon, who determined that he was not a candidate for surgery and referred him to a local doctor. He underwent weekly debridement. However, the necrosis extended deeper, and bleeding occurred from the right inferior gluteal artery, and he was brought to our hospital.

The ulcer measured 13 cm × 7 cm; pockets had formed on both lateral sides; and necrotic tissue, which had produced copious purulent exudate, remained (Fig. 2). Osteomyelitis was seen near the left sacroiliac joint. A wound culture revealed multidrug-resistant *Pseudomonas aeruginosa* and extended-spectrum beta-lactamase-producing *Escherichia coli*. The patient had anemia, mild dementia, and paresthesia in both buttocks, but was ambulatory and had no obvious problems with urination or defecation. The facility where the CIRT was performed reported that the tumor had shrunk, but not disappeared. (**See figure, Supplemental Digital Content 1,** which shows a computed tomography scan obtained at the referring hospital before the removal of the fourth and fifth sacral vertebrae, **http://links.lww.com/PRSGO/D389**.)

Although the patient was at risk of recurrence, he and his family agreed that he should undergo debridement, negative-pressure wound therapy with instillation and dwell time, and antibiotic treatment (meropenem) to improve his quality of life. Because some granulation tissue was observed, and no tumor was found during a histopathological examination of the ulcer base, we decided to cover the ulcer with a delayed (2 weeks) superior gluteal artery perforator flap (Fig. 3). Wound healing was difficult to achieve in the lower half of the flap, but, as the patient's anemia had improved, we decided to treat him as an outpatient. However, after 1 year, the ulcer near the coccyx had not healed (it was still about 2 cm in diameter) and extended deeper toward the rectum. A delayed (2 weeks) skin flap was created using the previously transplanted skin flap (Fig. 4). To bring the flap into close contact with the deep defect, negative-pressure wound therapy was performed for 4 weeks while the secondary defect was left open, and the suprarectal and secondary

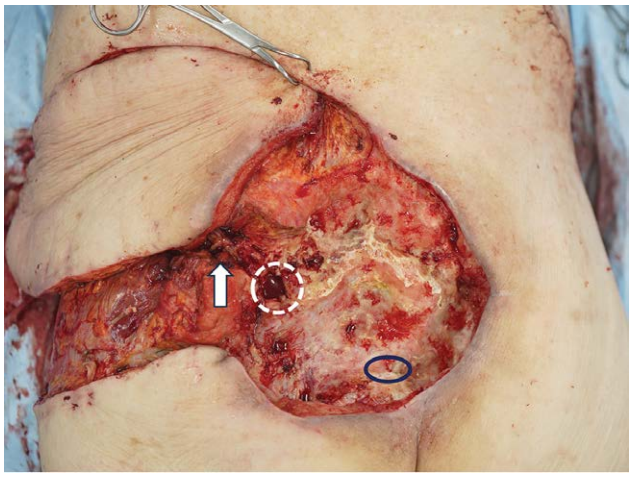

Fig. 3. Wound immediately before being covered with a superior gluteal artery perforator flap. White arrows: superior gluteal artery and vein. Dotted white circle: area of osteomyelitis near the left sacroiliac joint. Black circle: area where the wound was difficult to heal.

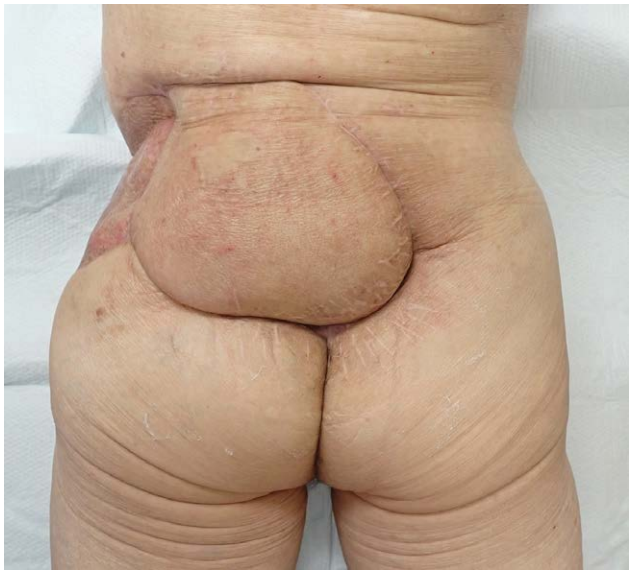

Fig. 4. It took a long time for the skin flap to fuse with the wound bed. No recurrence has been seen for 6 months from diagnostic imaging.

defect healed simultaneously. At 6 months after the last operation, the chordoma had not recurred. There were no problems with defecation, urination, or walking. (**See figure, Supplemental Digital Content 2,** which shows how a delayed skin flap was created using the skin flap that had already been transplanted to cover the rectum, **http://links.lww.com/PRSGO/D390**.)

DISCUSSION

In this case, the tumor had invaded a shallow area, including the gluteus maximus muscle, and shallow tumors are reported to be more likely to produce skin ulcers[6] (Fig. 1).

Post-CIRT rectal perforation has been reported previously.[1,2] To avoid irradiating the intestines, a spacer may be inserted between the intestinal tracts.[7]

We consider that once a post-CIRT ulcer develops, its progression and the area of debridement can be roughly predicted based on the radiotherapy treatment plan. In this case, there were no symptoms and no necrosis affecting the rectum or sciatic nerve, which were included in the 100% radiation dose area (Fig. 1).

Because CIRT delivers intense localized irradiation, it is thought to cause severe scarring at the irradiated area. Thick scarring was observed near the sciatic nerve and rectum, and the piriformis muscle was indistinguishable from the scar tissue, making the relationships between these important structures unclear. We only understood the depth of the ulcer when we injured the bilateral inferior gluteal arteries. Navigation may be required in such cases.[8] In this case, debridement to a depth equivalent to the point that received 30% of the maximum radiation dose managed to stop the progression of necrosis (Fig. 1).

Although the patient had dementia, he was able to walk, and a perforator flap was selected for reconstruction.[9] The selected perforator was the largest blood vessel seen on contrast-enhanced computed tomography; however, it was located inside the radiation field. There was no problem with blood flow of the flap, but postoperative wound healing was slow, and additional surgery was required. We should have performed the second surgery sooner. The tissue perfusion near the rectum and sciatic nerve was thought to be insufficient, and deeper debridement was considered appropriate, but it was also considered difficult to perform without complications. Therefore, it is desirable to use a flap with better blood flow, such as a myocutaneous flap, for such defects. We cannot determine whether our experience applies to all similar cases, and more cases should be studied.

In conclusion, reports of CIRT-induced ulcers requiring plastic surgery are still rare, but the number of such cases is expected to increase. We consider that after CIRT, ulcer progression and the required degree of debridement can be predicted based on the radiotherapy treatment plan. However, when attempting to preserve asymptomatic critical structures, debridement can be extremely difficult and leave poorly perfused tissue. It is preferable to cover such defects with the best-perfused tissue.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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