A Case of Multiple Radiation-related Complications during Long-term Follow-up after Sacral Ulcer Surgery

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Summary: We report the clinical course of a patient who developed a sacral radiation ulcer 19 years after treatment for cervical cancer. The patient’s postoperative course after a free latissimus dorsi muscle flap transfer was favorable, but various late radiation complications, including rectal perforation, a rectal fistula, sacral necrosis, a rectointestinal fistula, and sacroiliac joint osteomyelitis, occurred within 11 years. Plastic surgeons who treat such ulcers need to know that patients may develop other serious radiation-related complications. Being aware of these complications will allow appropriate measures to be taken and aid decisions regarding future surgical strategies. More careful assessment of sacral necrosis and bone resection may have ameliorated some of the complications. When encountering similar patients, we believe that careful magnetic resonance imagery (MRI) and intraoperative evaluation are warranted, as sacral necrosis may be detectable in some patients. (Plast Reconstr Surg Glob Open 2022;10:e4686; doi: 10.1097/GOX.0000000000004686; Published online 22 November 2022.)

The incidence of radiation ulcers in the sacral region after radiotherapy for cervical cancer is unknown. However, a previous study indicated that 9.8% of 1558 patients developed severe complications requiring hospitalization after such treatment.1 Three of these patients developed subcutaneous fibrosis, and one developed bone problems. This indicates that skin ulcers account for 2.6% of the severe complications seen after such treatment. There have been reports about plastic surgery for sacral ulcers,2 but no reports of complications occurring a long time after such surgery.

We report the clinical course of a patient who developed a sacral radiation ulcer 19 years after cervical cancer treatment. The patient’s postoperative course after a free flap transfer was good, but various late radiation complications, such as sacral osteomyelitis and a rectal fistula, occurred within 11 years.

CASE REPORT

The patient had a history of diabetes. In 1988, at age 35, she underwent cobalt radiotherapy and balloon occlusive antineoplastic arterial infusion3 for cervical cancer (stage IIIb in the International Union of Gynecology and Obstetrics classification). The radiation dose and whether intracavitary brachytherapy was performed are unknown. In 2009, a small skin ulcer appeared on her coccyx, which was treated with a local flap. A partial gluteus maximus muscle and skin flap was used to treat the first recurrence of the ulcer. In 2011, a latissimus dorsi musculocutaneous free flap anastomosed to the right inferior gluteal blood vessels was used to treat the second recurrence of the ulcer. In 2011, a latissimus dorsi musculocutaneous free flap anastomosed to the right inferior gluteal blood vessels was used to treat the first recurrence of the ulcer. In 2012, the patient was diagnosed with IgA nephropathy. In 2016, because urinary incontinence appeared, lumbar cauda equina nerve compression was suspected, and L2-S laminectomy was performed. Based on her high fever, a perforated rectal ulcer and abscess formation were diagnosed, and colostomy was performed. In 2017, she was started on hemodialysis. In 2019, a subcutaneous abscess formed in the sacrum under the partial gluteus maximus muscle skin flap. During the debridement, a rectal fistula leading to the abscess was found (Fig. 3), and endoscopy confirmed that the residual rectum had contracted to a diameter of about 1 cm. We removed the necrotic bone and reused the gluteus maximus muscle and skin flap to close the fistula. As undigested food residue was being excreted from the rectal fistula, a small intestine-residual rectal-sacral cutaneous fistula was diagnosed. After 6 months, the rectal fistula closed (Fig. 4), but in 2020, the patient experienced a cardiopulmonary arrest from septic shock and was resuscitated.

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After that, she had recurrent episodes of unexplained sepsis involving methicillin-resistant *Staphylococcus aureus* or *Pseudomonas aeruginosa*. The sepsis was relieved each time by treatment with appropriate antibiotics. Finally, magnetic resonance imagery (MRI) confirmed the presence of sacroiliac joint osteomyelitis. The site of the osteomyelitis was close to the site of the rectal perforation seen in 2016, so strong bowel adhesions were expected, and a small intestine-residual rectum fistula had already formed. Therefore, it was determined that resection of the sacrum and ilium would require extensive resection of the nearby bowel and would likely result in anastomotic leakage. The patient was then transferred to a geriatric hospital.

**DISCUSSION**

Cervical cancer is treated with surgery, chemotherapy, radiotherapy, or a combination of these. Our patient received chemotherapy and radiotherapy without surgery. The 5-year survival rate for stage III cervical cancer is reported to be about 30%–50%. Fortunately, our patient survived a long time.

Various radiation-related complications of cervical cancer treatment have been reported within 5 years of follow-up. Gastrointestinal symptoms, such as enteritis, ulcers, stenosis, and perforation, are the most common. Genitourinary symptoms, such as incontinence and bladder atrophy, are the second most common. Pelvic fractures, osteonecrosis, myelitis, and gait disturbance have also been reported.

However, there are a few reports about the long-term observation of such cases for more than 20 years. The reported major complications that occurred after 5 years mainly involved the rectum, bladder, or small intestine. The reported incidence of Franco-Italian glossary grade 3 or higher complications was 9.3% at 5 years and 14.1% at 20 years. Most rectal adverse events occurred within 5 years, with a relatively low incidence of such complications after 5 years. In contrast, adverse events affecting the bladder or small intestine tended to occur after 5 years. However, the incidence rates of complications such as rectocutaneous fistulas and osteomyelitis, as were seen in our case, remain unknown. There has only been one other report of a patient who experienced complications.
about 20–30 years after radiotherapy for cervical cancer. In our patient, urinary incontinence occurred, and L2-S laminectomy was performed, but it was only partially effective, and her gait disturbance progressed gradually, which was also thought to have been due to radiation. Some patients who undergo radiotherapy for breast cancer experience complications, such as ulcers and osteonecrosis, more than 20 years after treatment. In the case of radiotherapy for cervical cancer, the skin and various other organs may be affected by complications. And those complications should be treated in collaboration with relevant departments. We consider that being aware of the various possible complications will help clinicians to take appropriate measures and decide on future surgical strategies.

For treatment of sacral radiation ulcer, in 2011, a gluteus maximus turnover flap could have been used, but she was already complaining of difficulty walking at that time and did not choose this method. The next option was to use a transverse rectus abdominus muscle flap through the retroperitoneum. However, a free flap was chosen because the patient was so obese that the muscle of a transverse rectus abdominus muscle flap would not have been able to cover the large ulcer, and its skin flap would have been too bulky above the sacrum.

The sacrum, which later became necrotic, was the area covered by the second surgery. If sacral necrosis was suspected before or during the third surgery, and if it could be treated with a minor excision, rectocutaneous fistula might have been ameliorated. Judgment is often based on the absence of bleeding from the bone during surgery or poor wound healing around the bone after surgery. However, no apparent sacral necrosis was seen in our patient. It was also impossible to predict later sacral necrosis. Nevertheless, when encountering similar patients, we believe that careful MRI and intraoperative evaluation are warranted as sacral necrosis may be detectable in some patients.

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