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CASE REPORT
Reconstructive

Radiation-induced Osteomyelitis/Osteonecrosis of the Rib: SPECT/CT Imaging for Successful Surgical Management

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Summary: Chronic radiation-induced osteomyelitis/necrosis of the rib was rarely encountered in breast cancer patients even before the era of breast-conserving therapy. Few studies have focused on how to evaluate the extent of rib osteomyelitis for surgical management. A 78-year-old woman who had received radiation therapy after a radical mastectomy due to breast cancer 30 years ago manifested a rib pain and chest skin ulcers. Because chest magnetic resonance imaging failed to visualize osteomyelitis of the rib, a wide-ranged rib resection was initially planned considering her radiation field. An additional imaging, 3-dimensional (3D) single-photon emission computed tomography (SPECT)/computed tomography (CT), was performed to obtain 3D virtual chest images highlighting the areas of inflammatory or necrotic bone tissues; her osteomyelitis was localized in the left anterior chest. A chest wall reconstruction was performed using a vascularized pedicled latissimus dorsi myocutaneous flap on the left side after the resection of the ribs, part of the sternum, and costal cartilage with radical debridement of all necrotic tissues. The 3D SPECT/CT contributed to a safe chest wall reconstruction with a 40% reduction in resected bone and soft tissues when compared to the magnetic resonance imaging-based surgical plan. Pathology results showed no evidence of inflammation or necrosis in the surgical margin. No complication related to the reconstruction or no recurrence was observed during a postoperative 12-month follow-up. The present case suggested that 3D SPECT/CT can be applied to preoperative surgical planning related to bone diseases including osteomyelitis, traumas, bone tumors, and diabetic foot. The appropriate application of 3D SPECT/CT requires full validation through significant clinical experience. *(Plast Reconstr Surg Glob Open 2019;7:e2536; doi: 10.1097/GOX.0000000000002536; Published online 30 December 2019.)*

In breast cancer, radiation therapy is generally performed to reduce the risk of local recurrence after surgery. However, it can cause complications such as upper extremity edema, skin ulcers, and radiation pneumonitis, which probably depend on the delivered dose.[1,2] Rib osteomyelitis/osteonecrosis (RO) is a rare complication, even in patients receiving radiotherapy after radical mastectomy. For members of a surgical team performing unfamiliar surgeries including RO, preoperative imaging often helps them accomplish successful operations. Especially, 3-dimensional (3D) virtual imaging is preferable to conventional tomographic imaging not only for determining surgical areas but for performing more realistic surgical simulation. Recently, our group has developed a novel technology to visualize the area of abnormal bone metabolism, which is based on tomographic imaging of bone scintigraphy (single-photon emission computed tomography, SPECT), onto 3D volume-rendered computed tomography (CT) images.[3] This 3D imaging technology (we call it 3D SPECT/CT) is thought to be useful to map the distribution of bone diseases such as mandibular osteomyelitis/osteonecrosis on 3D bone structures.[4] Here, we experienced a patient in which her radiation-induced osteomyelitis of the rib was successfully delineated and resected by using the patient's 3D SPECT/CT images. In the present case, 3D SPECT/CT contributed to a safe chest wall reconstruction with a 40% reduction

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Related Digital Media are available in the full-text version of the article on www.PRSGlobalOpen.com in resected bone and soft tissues when compared to magnetic resonance imaging (MRI)-based surgical plan. This was because metabolic information provided by SPECT pinpointed chronic inflammatory foci, which were difficult to exhibit with MRI.

CASE REPORT

The patient was a 78-year-old female patient with left breast cancer who had received radiation therapy after a radical mastectomy on the left side at a local hospital 30 years ago. She was admitted to our hospital, complaining of a rib pain and the progression of redness, pigmentation, and multiple ulcers of the skin on the left anterior chest wall. Her clinical records showed that the radiation therapy consisted of 5,400 cGy to her whole breast and 1,600 cGy to infra-supraclavicular fossa. Her previous surgical history included cholecystectomy and appendectomy. No recurrent tumor was found according to her chest and abdominal CT. The patient was diagnosed with RO on the basis of her chest wall condition (Fig. 1) and history of radiotherapy.

Contrast-enhanced MRI was initially performed to evaluate skin inflammation and RO; chest wall fistulae were clearly depicted as enhancing skin areas (**See figure, Supplemental Digital Content 1**, which displays MRI images at the levels of the second and fourth ribs. Enhancing subcutaneous soft tissue with small recesses was observed, **http://links.lww.com/PRSGO/B251**). However, it was difficult to delineate bone involvement, mainly due to a susceptibility artifact which is derived from air component surrounding the chest wall (ie, air in the lung and outside of the body). In addition, a respiratory motion artifact was also considered to make RO unclear. On the other hand, using an image browser (AW server 2; GE Healthcare, Milwaukee, WI), 3D SPECT/CT facilitates not only simultaneous evaluation of bone inflammation and anatomy from any angle but also focused display of the left ribs without displaying bone metabolic findings unrelated to RO. (**See Video [online],** which displays 3D SPECT/CT focusing on the affected area of the left ribs.) The patient's 3D SPECT/CT showed increased accumulation of injected radioisotope in the left lower side of the sternum body, anterior side of the left ribs, allowing us to reduce the surgical extent compared to her previously irradiated area (Fig. 2). The patient consented to undergo the surgery of her RO after an explanation of its surgical procedure using the video (**See Video [online],** which displays 3D SPECT/CT focusing on the affected area of the left ribs).

The resection of the ribs, part of the sternum, and costal cartilage was performed with radical debridement of all necrotic tissues. The parietal pleura was intact. The size of the resected skin and soft tissues was 13.0 cm × 6.5 cm (**See figure, Supplemental Digital Content 2**, which displays an intraoperative image (A), an intraoperative image fused with a 3D SPECT/CT image (B), and a 3D SPECT/CT image (C) in left anterior view, **http://links.lww.com/PRSGO/B252**). A 14-cm left pedicled latissimus dorsi myocutaneous flap was used for chest wall reconstruction after the vascularity of the vessel was confirmed by ultrasound (Fig. 3). Other possible flaps for the reconstruction were transverse rectus abdominis musculocutaneous, pectoralis major myocutaneous, and deltopectoral flaps, although these were not used because of the patient's surgical history and the flap volume. Intraoperative bone appearances in the areas of increased radiotracer accumulation were indicative of osteomyelitis/osteonecrosis, although it was difficult for our surgeons to determine the precise extent of osteomyelitis/osteonecrosis with high confidence without the reference of 3D SPECT/CT images. After all, 3D SPECT/CT contributed to a safe chest wall reconstruction with a 40% reduction in resected bone and soft tissues when compared to MRI-based surgical plan. The wound

Fig. 1. Preoperative chest appearance with previously irradiated area (red line).

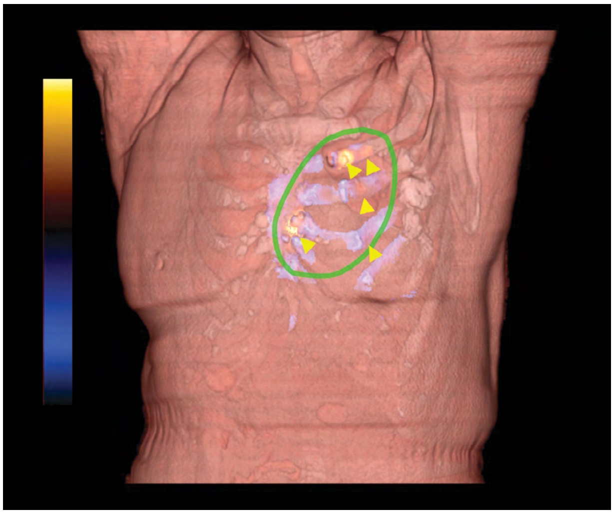

Fig. 2. Preoperative 3D SPECT/CT image. Increased accumulation of injected radioisotope suggesting osteomyelitis (yellow arrowheads) with the surrounding normal bone tissues (blue) is displayed in the patient's radiation field. Green circle indicates the actual surgical extent.

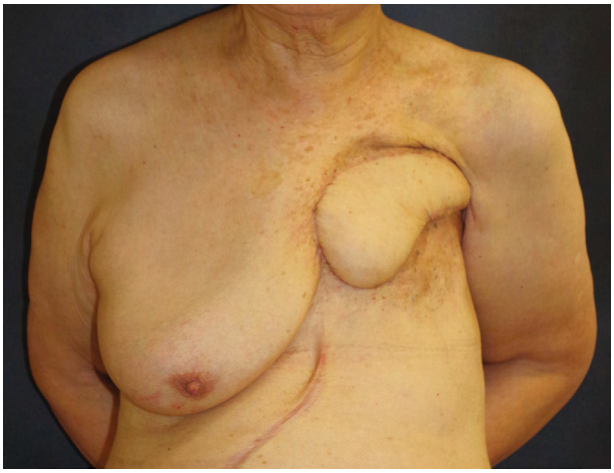

Fig. 3. Chest appearance after the reconstruction with the left pedicled latissimus dorsi flap vascularized by the left thoracodorsal artery.

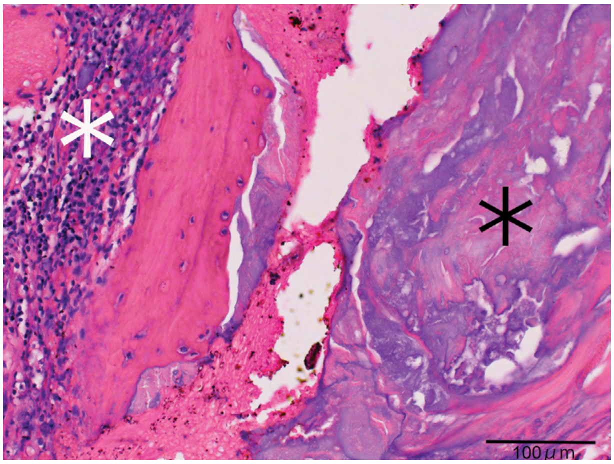

Fig. 4. Histopathological findings of the resected second ribs (hematoxylin and eosin stain, ×40) showing acute inflammatory cell infiltration (white asterisk) and necrosis (black asterisk).

of the donor area was closed with a deep and superficial layer. No complication related to the reconstruction or no recurrence was observed during a postoperative 12-month follow-up.

Pathology results revealed that the area of inflammatory cell infiltration in the resected part of the ribs and sternum was consistent with that of increased radiotracer in 3D SPECT/CT (Fig. 4). In addition, a substantial amount of osteonecrosis was observed mainly in the costal cartilages near the skin ulcers. There was no evidence of inflammation or necrosis in the surgical margin.

DISCUSSION

Although chronic radiation-induced osteomyelitis of the ribs is uncommon, it should be considered when chest wall fistulae are seen in patients with chest pain and a history of radical mastectomy followed by radiation therapy.[5] In such patients, radiation-induced toxicity would cause refractory tissue inflammation and subsequent necrosis in the chest wall as a result of compromising the tissue microenvironment by microvascular injury, inflammation, and infection.[6] Conservative therapy is ineffective for this chronic complication, and a surgical management would therefore be a key to improve the patient's quality of life.

There have been some case reports regarding the selection of the flap for chest wall reconstruction[1,7,8] or the disease management in patients with radiation-induced osteomyelitis.[9,10] However, to our knowledge, no study has focused on how to evaluate the extent of osteomyelitis. It seems that bone tissues within previously irradiated areas, where reduced tissue vascularity and loss of the ability to heal injuries could be expected,[6] should entirely be resected in combination with the debridement of the surrounding tissues. However, in fact, the boundary between the compromised part and the vital bone is not easy to predict based on skin examination,[8] and imaging technology to visualize the osteomyelitis is therefore practically helpful for surgical planning. Generally, the extent of osteoradionecrosis has been determined during surgery based on both the clinical appearance of the bone and the quality of blood supply. However, in cases with breast cancer receiving radiotherapy, it seems that even skilled or well-experienced surgeons have some difficulty in determining the extent of rib osteoradionecrosis during surgery without surveying the entire irradiated ribs with their own eyes. In the present case, chest MRI was not useful to evaluate RO, only making our plastic surgery team intend to perform a wide-ranged rib resection covering the entire irradiated area. 3D SPECT/CT revealed the limited area of RO compared to the radiation field, and its visual effect contributed to an explanation to the patient as well as a minute surgical simulation without exposing the entire irradiated ribs.

Regarding the benefit of imaging, without 3D SPECT/CT imaging, the extensive surgery covering her entire irradiated area would have required a much larger latissimus dorsi myocutaneous flap, entailing an additional skin grafting from other sites such as the thigh to cover the wide range of skin deficiency at the donor site. In such situation, the movement of a part related to the skin grafting would be restricted for about 1 week. (In fact, we explained the patient before operation that surgical procedure might change if 3D SPECT/CT findings should not visualize osteomyelitis/osteonecrosis correctly.) Some thoracic surgeons who participated in the surgery judged that an additional chest wall reconstruction using a surgical mesh was unnecessary.

Regarding the other applications of 3D SPECT/CT in the field of plastic surgery, we consider that bone and soft tissue reconstructions after bone traumas and malignant tumor resection as well as in osteomyelitis and diabetic foot are good indications for 3D SPECT/CT imaging. Its application to these diseases will lead to improved understanding of the value of this imaging. In addition, as shown in Supplemental Digital Content 2 (which displays an intraoperative image (A), an intraoperative image fused with a 3D SPECT/CT image (B), and a 3D SPECT/CT image (C) in left anterior view, http://links.lww.com/PRSGO/B252), 3D images can be fused into intraoperative images. We envisage that this technique will be applied to "Augmented Reality" using Google Glass during a live surgery or at training before surgery.

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