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Case Report

Cervical Spine Osteoradionecrosis

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Osteoradionecrosis (ORN), a well-known complication of radiotherapy in the mandibular bone, is very rare in the cervical spine. The authors report the result of a 3-year follow-up of a 63-year-old female patient with ORN of the cervical spine. The patient had a history of laryngeal carcinoma and was treated with chemotherapy and radiation therapy with a total of 120 Gy. Eight years later, she developed acute, severe neck pain due to cervical spine necrosis. The authors performed vascularized fibular bone graft and posterior pedicle screw fixation to reconstruct her cervical spine. The patient was successfully treated with surgery, and cervical alignment was preserved. She had neither neurological deficits nor severe neck pain at her final follow-up 3 years later. Delaying treatment of ORN may be life threatening, so the early diagnosis of this condition is important for patients who receive radiotherapy. Otolaryngologists and spine surgeons should understand this potential complication to speed diagnosis and treatment as early as possible.

Key words: osteoradionecrosis, laryngeal carcinoma, cervical spine, radiotherapy

Radiation therapy is both a primary and supplementary treatment for head and neck cancers. There are advantages to using radiotherapy in cancer treatment, but the irradiated patient also may experience complications, including skin necrosis, acute visceral inflammation secondary to cancer and osteoradionecrosis (ORN) [1]. ORN of the mandible has been well reported [2]; however, spinal ORN is very rare [3,4]. Although severe neck pain is the most common symptom of cervical ORN [5], patients treated with neck radiation often complain of neck pain, and metastases to the spine are common. Therefore, early diagnosis of ORN can be very difficult, and a delay in diagnosis has been reported [6]. As a result, this complication of radiation therapy may become a life-threatening condition.

The authors report on a rare case of ORN of the cervical spine after laryngeal carcinoma. We also discuss the diagnosis and treatment of cervical ORN.

Case Report

Patient history. A 63-year-old woman was referred to our orthopedic department in February 2013 for severe neck pain. She had received radiotherapy and chemotherapy for laryngeal carcinoma twice at another hospital in 2004 and in 2005. A total dose of 120 Gy was given by 3 ports methods (Fig. 1). She also had mandibular reconstruction surgery through the Department of Plastic Surgery at our institute in January 2013. One month later, she complained of mild neck pain that gradually increased.

Physical examination. On examination, she

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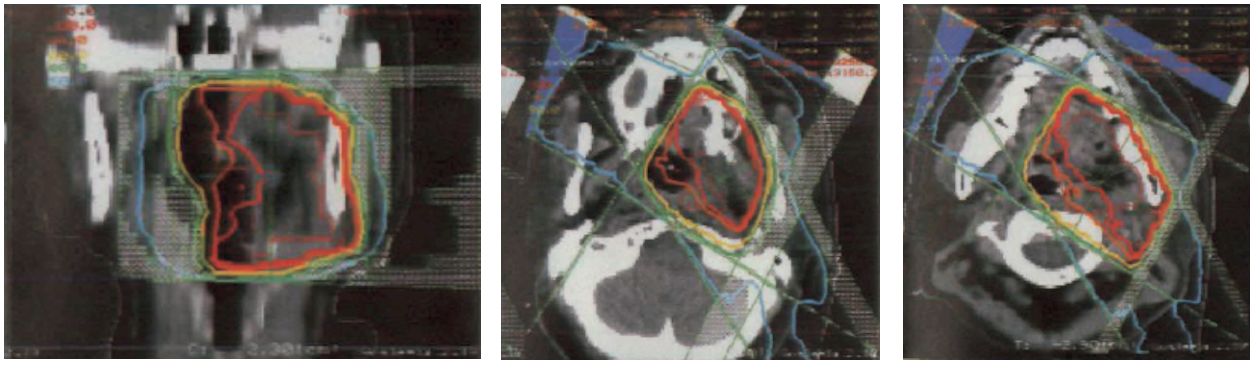

Fig. 1 The dose distribution. Total 60 Gy was irradiated by 3 ports method at one time.

could walk without support and had no neurological deficits. There was no hyperreflexia of her arms or legs and no abnormal abdominal reflex, but there was severe pain in her neck, and the range of neck motion was limited.

Preoperative imaging. CT and MRI at the patient's initial visit demonstrated subtle destruction of C2-4 vertebrae and loss of normal cervical lordosis (Fig. 2A, B). Follow-up plain radiographs and CT revealed progressive destruction of the cervical spine with bony sclerosing, which indicates ORN (Fig. 2C, D). The kyphotic angle of C2-4 was 23 degrees. MRI revealed bony collapse and necrosis of the C2-4 vertebrae (Fig. 2E). ORN appeared as low signal intensity on T1WI, mixed signal intensity on T2WI and enhanced on post contrast T1-weighted imaging (Fig. 2E).

Surgical intervention. We performed cervical reconstruction with a vascularized fibular bone graft (Fig. 3A) and pedicle screw fixation (Fig. 3B, C). The segmental pedicle screw fixation from skull to T2 produced an excellent curve correction and maintained good cervical alignment. Insertion of the pedicle screws using the Stealth Station navigation system Spine 7^{R} (Medtronic, Memphis, TN, USA) was accurate to less than 0.1 mm. For vascular reconstruction, the fibular artery and vein of the musculocutaneous flap were anastomosed with the internal thoracic artery and vein respectively. Operative time was 8 h and 22 min and estimated blood loss was 1,000 ml. A biopsy of C2 revealed necrotic bone without evidence of osteomyelitis. The tissue culture was also negative. There have been no postoperative complications and no neurological compromise.

Follow-up imaging. By 6 months, she was back to nearly full activity. The postoperative CTs demonstrated good correction of curve with an appropriate sagittal alignment (Fig. 3B). She has good cervical balance and no neurological deficit over 3 years later. She had no evidence of malignancy and good cervical alignment at the final follow-up (Fig. 3C).

Discussion

ORN subsequent to radiotherapy for neck cancer is rare. Regaud reported the first case of ORN in 1922 [7]. This condition is best described as a slow-healing radiation-induced ischemic necrosis of bone with associated soft tissue necrosis of variable extent occurring in the absence of local primary tumor necrosis, recurrence or metastatic disease [8]. ORN can occur in the upper and lower jaw 1-3 years following radiation therapy with an incidence of 8.2% in the mandible [9]. However, the incidence of cervical ORN is approximately 0.9% [10], and there are few cases in the literature [5]. This is mainly because the poor blood supply of the jaw [11] and the superinfection of the irradiated bone [12].

Demographic features of ORN. Among 22 reported cases [3-6, 13-15], we excluded 11 cases that were not fully documented; Table 1 summarizes the

Table 1 Summary of cervical ORN

Symptom	Number of patients	%
Neck pain	7/11	64%
Cervical kyphosis	6/11	55%
Dysphagia	4/11	36%
Myelopathy	3/11	27%
Radiculopathy	3/11	27%
Air way problem	3/11	27%
Headache	2/11	18%

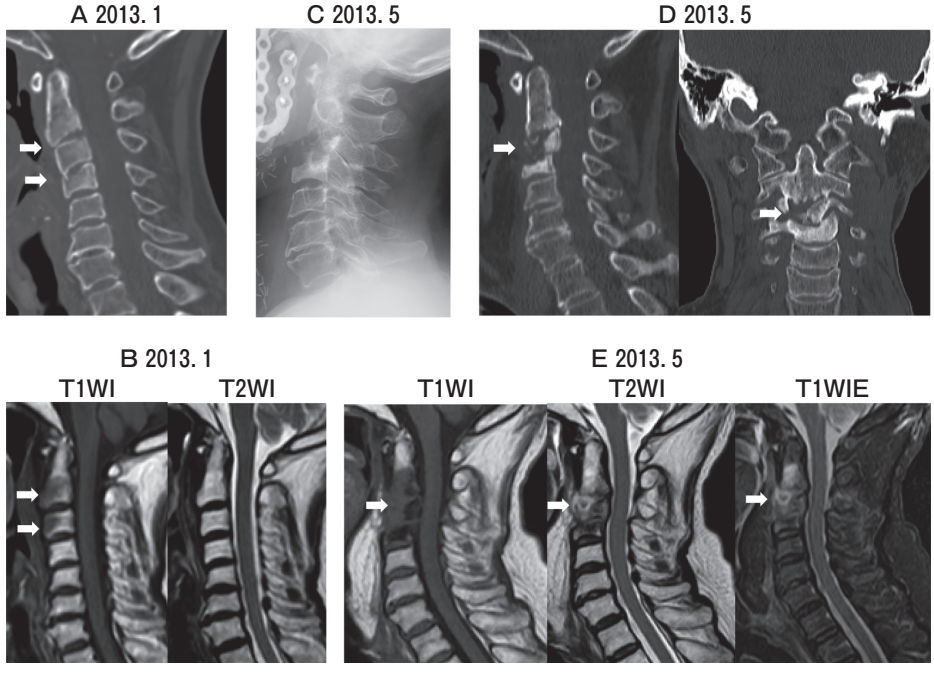

Fig. 2 Preoperative cervical CT and MRI. **A**, CT at 4 months before surgery. There is subtle bony destruction of C2-4 (white arrows); **B**, MRI at 7 months before surgery. There is only subtle low signal intensity at C2 and C3 vertebral bodies (white arrows); **C**, Radiogram at 2 weeks before surgery. There is local kyphosis at C3; **D**, CT at 2 weeks before surgery. C2-4 vertebrae are severely collapsed (white arrows); **E**, MRI at 2 weeks before surgery. ORN appears as low signal intensity on TWI and mixed signal intensity on T2WI. ORN appears as contrast enhancement on post contrast T1-weighted imaging (white arrows).

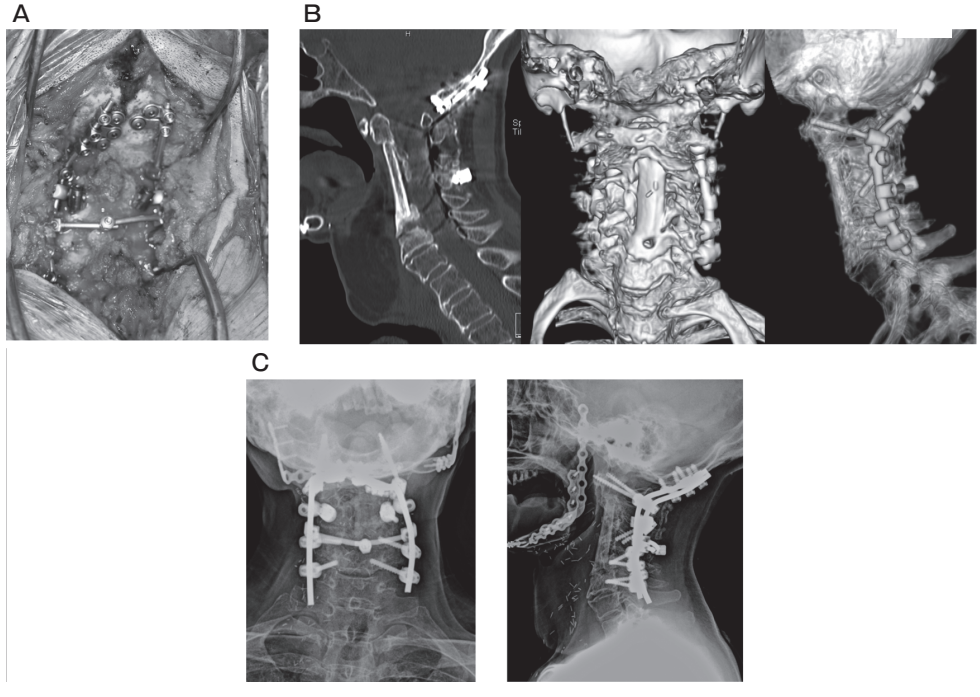

Fig. 3 Intraoperative view and postoperative imaging. **A**, Intraoperative view. A posterior cervical reconstruction was performed using pedicle screw fixation; **B**, CT at 16 months after surgery. Fibular graft was correctly inserted and fusion is apparent; **C**, Radiogram at 3 years after surgery. Solid bony fusion can be seen with good cervical alignment.

remaining 11 cases, which includes our case. The average radiation dose was 74 Gy, ranging from 57 to 120 Gy. Importantly, the onset of initial symptoms from radiation therapy varied widely from 15 months to 48 years. Considering that half the patients had a silent period of greater than 8 years, thorough history taking becomes extremely important for detecting certain patients with ORN of the cervical spine. Approximately 2/3 of the patients had neck pain (7/11, 64%). Dysphagia (4/11, 36%) was a relatively common symptom. Neurological deficits were also common: 3 patients demonstrated myelopathy and 3 patients complained of radiculopathy. Cervical kyphosis was the specific feature in these cases; 6 out of 11 patients had this condition.

Diagnosis of ORN. MRI is the most important tool for identifying a number of potential etiologies of cervical pain including tumor, infection, osteomyelitis, disc herniation and canal stenosis. King *et al.* [16] reported that cervical ORN appeared as low signal intensity on T1-weighted imaging and contrast enhancement on post contrast T1-weighted imaging in all cases. On the other hand, T2-weighted imaging was more variable with ORN appearing as a high signal or mixed intensity. These findings (bony high T2 signal intensity and contrast enhancement) are also often found in infection.

CT is also used to diagnose cervical ORN, especially to evaluate the bony destruction it causes. More importantly, clinical features of osteomyelitis may resemble symptoms of ORN. Prasad *et al.* [10] reported that ORN is the second most common cause of osteomyelitis in the head and neck (21%) after contiguous infection (33%). Many authors reported concomitant conditions of both ORN and osteomyelitis [13]. Osteomyelitis plays an important role in the rapid progression of ORN, which makes it important to exclude simple osteomyelitis or disease recurrence. A biopsy is necessary for a definitive diagnosis of cervical ORN versus malignancy or infection.

Treatment of ORN. The treatment options of ORN are immobilization with a collar or halo jacket, antibiotics, hyperbaric oxygen (HBO) and surgical intervention. Conservative treatment such as HBO may be effective only in the early stage of the disease [15]. However, in the advanced stage, with the presence of myelopathy or severe bony destruction, surgical intervention is necessary [5, 13, 14].

Of the 11 cases [3-6, 13-15], including ours, we summarized earlier, 6 cases received surgery. Four cases had anterior and posterior combined procedures, 1 had an anterior procedure and 1 a laminectomy. Three cases received vascularized fibular grafts for anterior reconstruction to secure solid bony fusion. This anterior cervical reconstruction using vascularized fibular grafts and posterior fusion is the most reasonable surgery for this condition [14]. Because nonvascularized bone grafts rely on fusion by a gradual process in which bone substitution occurs, and this may take up to 2 years and the grafts are at higher risk for failure [13]. On the other hand, a vascularized bone graft fuses faster, usually the union needs only 3-5 months [18, 19]. The posterior spinal fusion without anterior surgery is another option for this condition, if the bone necrosis is only limited area or the patient has a high risk for an invasive surgery. However, if there is a large ORN like this case, it is necessary for the patient to undergo the debridement of necrotic and infected tissue, decompression of neural structures, reestablishment of spinal alignment and stability by using a structural graft. Most importantly, the prognosis for cervical ORN was poor if the treatment was delayed. The mortality rate of these 11 cases was 27%, and there were good results in only 45%, so early diagnosis and treatment is the key to treating this complication.

In conclusion, ORN of the cervical spine is a rare but significant complication of head and neck radiation therapy. Delayed diagnosis can result in marked disability or life-threatening complications. It is very important for spine surgeons and otolaryngologists who treat head and neck malignancies to be aware of this potential complication. Early treatment may lead to more favorable outcomes; however, for patients in the advanced stage with myelopathy or cervical kyphosis, aggressive surgical intervention such as vascularized fibular grafts and posterior instrumentation are necessary.

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