

Radiation-Induced Osteosarcoma in the Cervical Spine after Definitive Radiotherapy for Esophageal Cancer: A Case Report

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We experienced a case of an extremely rare radiation-induced osteosarcoma (RIOS) that occurred in the cervical spine after definitive radiotherapy for esophageal cancer. Be-

cause of its rarity, obtaining an appropriate diagnosis may be occasionally delayed. The therapeutic choice is very limited because total resection of the tumor is impossible in the

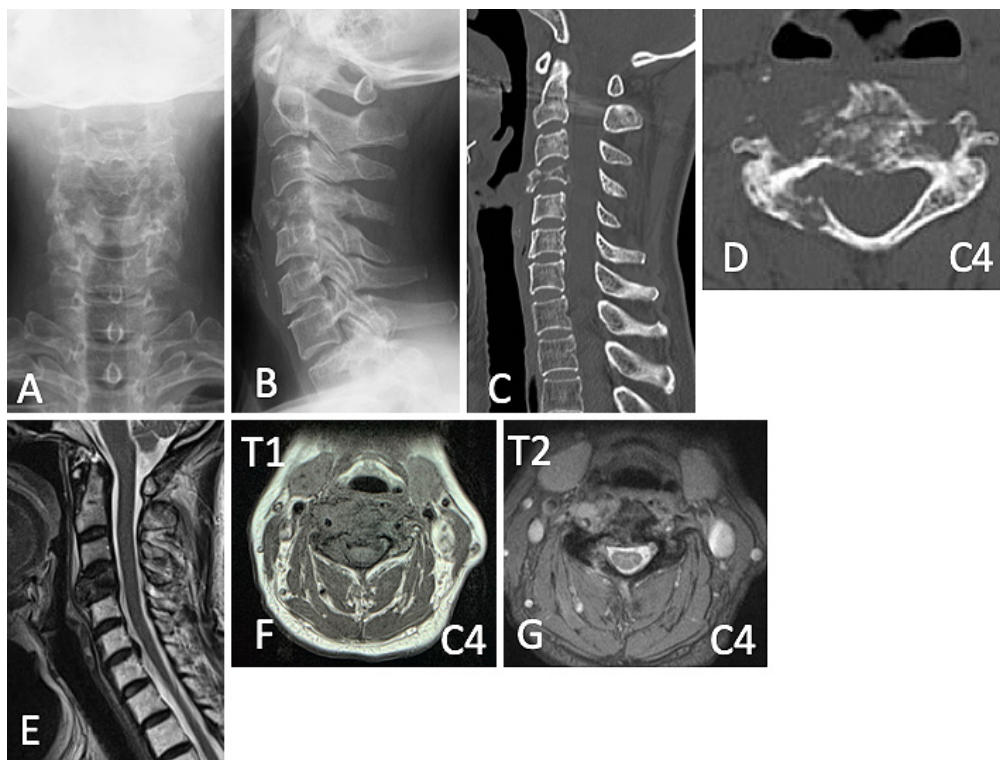


Figure 1. A-B. Plain lateral radiograph showing a vertebral fracture at C4. C-D. Sagittal CT showing a vertebral fracture at C4 (A). Coronal CT of C4 showing destructive changes of the vertebra body, the right-side facet, and the right-side lamina (B). E-G. Magnetic resonance images showing heterogenic low-intensity lesions at the C4 vertebra in T1- and T2-wighted images.

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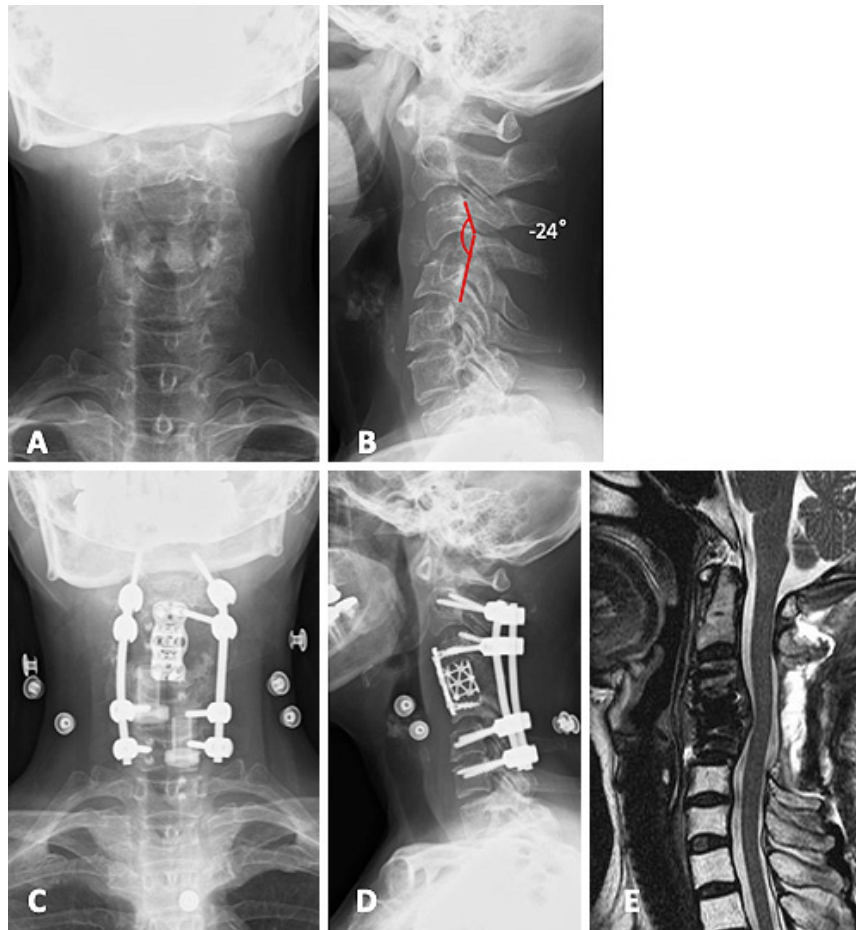


Figure 2. A-B. Lateral radiographs showing the progress of collapse at the C4 vertebral body and local kyphosis of 24 degrees between C3 and C5. C-E. Postoperative radiographs (C) anteroposterior view (D) lateral view (E) postoperative T2-weighted sagittal MRI.

cervical region owing to the presence of the spinal cord and vertebral artery.

A 58-year old woman suffered nape pain and visited our institute. She had a medical history of undergoing chemotherapy with FP (fluorouracil + cisplatin) and radiotherapy (60 Gy) for esophageal cancer (cT2N1M0) at the age of 50 years, and subsequent DCF (docetaxel, cisplatin, and 5-fluorouracil) chemotherapy in two courses was added. Her prognosis was good, and she received follow-up.

Her plain radiographs showed a vertebral fracture at C4 (Fig. 1A, B). Her computed tomography (CT) showed destructive changes of the vertebral body, the right-side facet, and the right-side lamina at C4 (Fig. 1C, D). Her magnetic resonance images showed heterogenic low-intensity lesions at the C4 vertebra in T1- and T2-weighted images (Fig. 1E-G). No neurological deficit was found at her initial visit.

Because of her medical history, metastatic bone tumor or osteonecrosis after radiation therapy was suspected. The tumor markers were all negative. Her positron emission tomography-CT showed high accumulation of SUV MAX 36.93 at the C4 vertebra without any hot spots elsewhere.

Incisional biopsy harvesting of substantial amounts of the tissue from the right-side facet and the lamina at C4 was

performed for pathological examination. However, the finding was “fibrous tissue with no malignancy.”

We carefully performed follow-up, but she showed subsequent augmentation of neck pain 1 month after biopsy. The radiographs showed the progress of collapse at the C4 vertebral body and local kyphosis of 24 degrees between C3 and C5 (Fig. 2A, B). Therefore, we performed reconstruction surgery, i.e., subtotal corpectomy of C4; cage insertion (Phyramesh, Medtronic, Memphis, TN, USA) at C4; anterior plating (Zepher, Medtronic, Memphis, TN, USA) between C3 and C5; laminectomy of C3, C4, C5, and C6; and posterior fixation using cervical pedicle screw between C2 and C7 (Vertex MAX, Medtronic, Memphis, TN, USA) (Fig. 2C-E). During surgery, we recognized thickened soft tissue in front of the fourth cervical vertebra and the body was filled with fibrous tissues. These tissues were submitted for pathological examination. With intraoperative histological examination, no malignant findings were reported. However, 1 month later, we finally obtained the diagnosis of post-radiation spindle cell sarcoma (Fig. 3A, B).

As radiation-induced sarcoma (RIS), malignant fibrous histiocytoma is the most common subtype. Other frequent subtypes include angiosarcoma, leiomyosarcoma, and fibro-

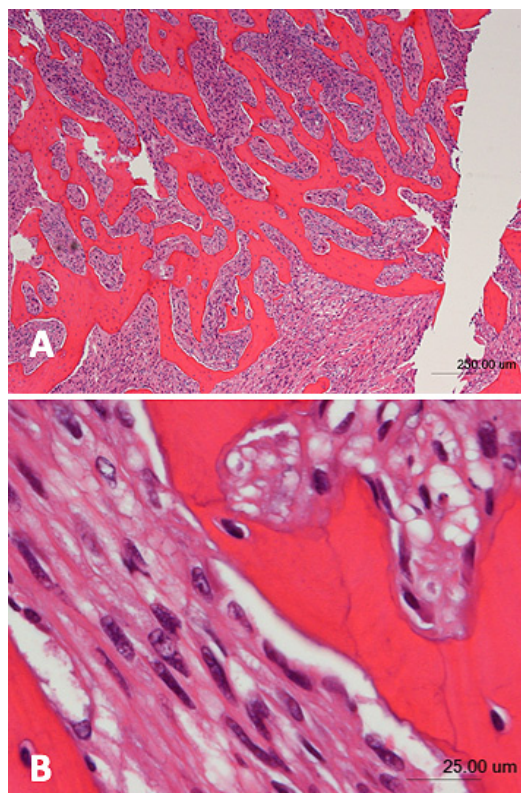


Figure 3. A-B. Scale bar in Figure 3A is equal to 250.00 um and in Figure 3B is equal to 25.00 um. Microscopic findings revealed that spindle cells were relatively fascicular in growth. Nuclear atypia was seen. A somewhat irregular cement line was seen in the trabecular bone, but it was relatively mature trabecula from trabecular arrays and substrate state. Additionally, invasion of malignant lesions into cancellous bone was observed.

sarcoma^{1,2}). Extra-skeletal osteosarcoma is a rare spontaneous tumor constituting 2%-4% of all osteosarcomas, but it seems to be more common among RIS²). RIOS, which presumably arises in the soft tissues from pluripotential fibroblastic cells, is classified into osteoblastic, chondroblastic, or fibroblastic subtypes depending on the morphologic variability³). The most common site of PROS is pelvis⁴). To our knowledge, only one patient with RIOS in whom the site was intradural of the cervical spine has been reported⁵).

We started treatment with the anti-cancer drug adriamycin after the surgery. However, the tumor was continuously extended (Fig. 4A, B). Hu et al reported that the effect of chemotherapy on RIOS was poor⁵). Rubino et al reported that the grade of malignancy of RIOS was often high⁶). The radical resection of the tumor is impossible owing to the existence of the spinal cord and vertebral artery. Therefore, the prognosis of RIOS is expected to be poorer in cases in

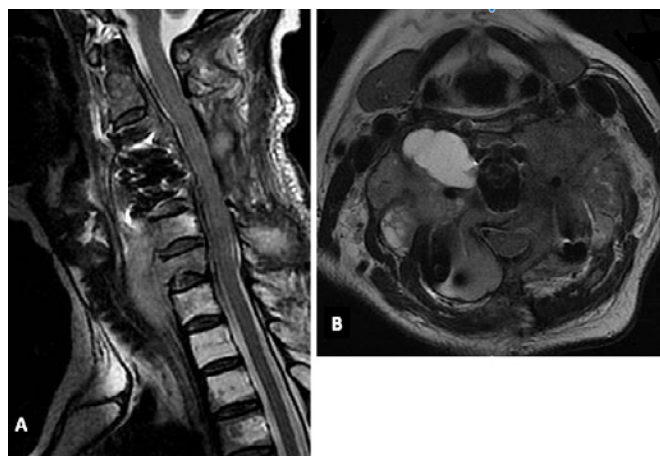


Figure 4. A-B. T2-weighted magnetic resonance imaging performed 3 months after surgery. The tumor was seen to extend cephalad, caudad, front, and back of the cervical spine.

which it occurs in the cervical spine than in an extremity. We advised our patient to follow palliative care.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

Ethical Approval: None

Informed Consent: Cervical vertebral body destruction is progressing and needs to be fixed. A biopsy is also performed by surgery to investigate the cause. Informed consent was obtained by the participant in this study.

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