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Radiation-Induced Osteosarcoma of the Mandible: A Case Report and Review of the Literature

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ABSTRACT

We report the case of a 69-year-old man with radiation-induced osteosarcoma of the mandible diagnosed four years after radiation treatment for squamous cell carcinoma of the tongue.Although radiation therapy is a proven treatment modality for malignancies of the head and neck, it can have adverse effects. Late complications in bone following radiotherapy include osteoradionecrosis, radiation-induced osteitis, and osteosarcoma, which is an uncommun tumor, diagnosed by meeting certain diagnostic criteria. The mandible is a location very sensitive to radiation effects.

Introduction

Radiation therapy is an adjuvant in the treatment of many tumors. Second primary malignancy following radiotherapy is one of the late sequelae of radiation therapy, which is of concern in long-term cancer survivors.

We report the case of a 69-year-old patient, operated in 2017 for squamous cell carcinoma of the tongue, admitted to our structure for a right submandibular swelling, study of which was in favor of a radiation-induced osteosarcoma.

Observation

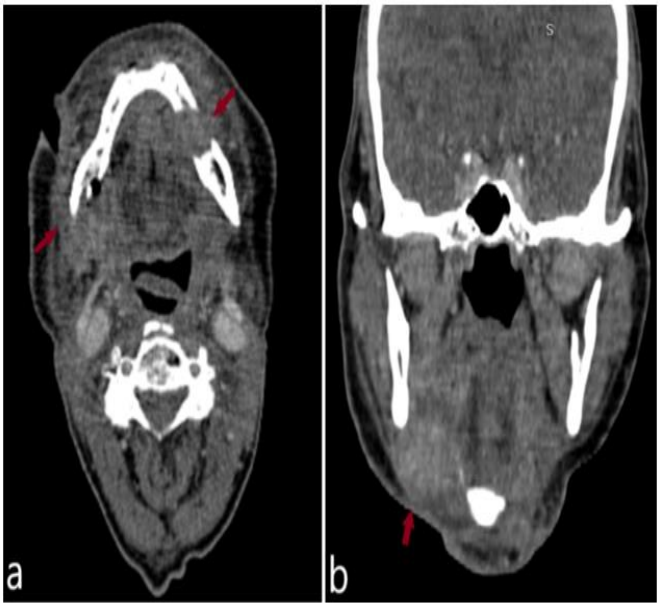

Figure 1. Parenchymal window CT scan of the facial mass in axial (a) and coronal (b) sections showing two bilateral mandibular heterogeneous lytic tissue processes infiltrating the submandibular gland on the left and fistulating to the skin on the right

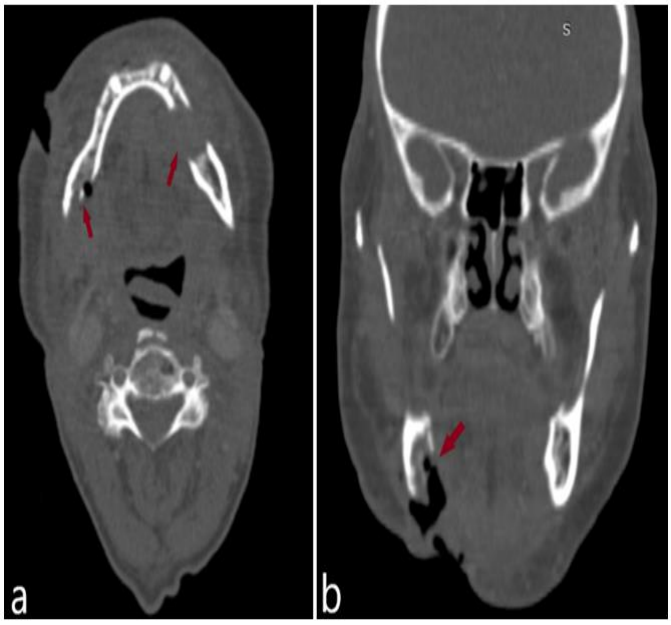

Figure 2. Bone window CT scan of the facial mass in axial (a) and coronal (b) sections showing two bilateral mandibular osteolytic lesions with cortical rupture

We report the case of a 69-year-old patient with no medical or toxic history, operated in 2017 for squamous cell carcinoma of the tongue. Our patient had an adjuvant chemotherapy with cisplatine et 5-fluorouracil and radiotherapy 65 Gy, Classic fractionation 2 Gy / fractions, 5 times/week for 7 weeks.

Two years after his last irradiation, he was admitted to our structure for a right submandibular swelling. A CT scan of the facial mass was performed (figure 1 and 2) showing, two bilateral mandibular heterogeneous lytic tissue lesional processes with cortical rupture infiltrating the submandibular gland on the left and fistulating to the skin on the right.

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The patient had a biopsy, and the anatomopathological study of which was in favor of a radiation-induced osteosarcoma.

Discussion

Radiation therapy is an adjuvant in the treatment of many tumors. Second primary malignancy following radiotherapy is one of the late sequelae of radiation therapy, which is of concern in long-term cancer survivors. [1]

Radiation induced sarcoma is a rare but recognized complication of radiotherapy and is associated with poor prognosis. Several cases have been reported in the literature, In a review article on radiation-induced sarcomas, Malcolm Feign had summarized 10 reports totalling 14,000 patients, the reported incidence being 0.16%. [2].

Wei et al. described an increase in incidence of radiation-induced sarcoma from 0.06% to 0.17%; with a median latency of 7.7 years and 3-year overall survival of 19.1% [3]. In a study examining outcomes in radiation-induced sarcoma in NPC patients, Xi et al. reported a median latency of 9.3 years and median overall survival 21.2 months [4].

Risk factors for developing RIS are young age at treatment and treatment-related factors, including high radiation dose and simultaneous chemotherapy with alkylating agents. Our patient had received a radiation of 50 Gy. He received chemotherapy as well. Radiation above 50 Gy cause cell death, while lower doses (<30 Gy) cause genomic instability and damage cell repair mechanisms.

RIS typically occur within or at the edges of the radiation field. At the edges of the radiation field, the dose of radiation is not homogeneous and may be less than the tumor killing dose [5].

The diagnostic criteria of post-irradiation osteosarcoma include a lesion centered in irradiated bone without a primary malignant osteoblastic lesion, arising after a latency period of at least 3 years after the completion of radiation therapy [6].

Both CT and MR imaging are excellent for revealing tumor extent and as aids in presurgical planning. CT is superior in localizing matrix mineralization, periosteal bone reaction, and cortical destruction, it shows an iso or hyperdense heterogeneous tissue mass enhancing after injection of the contrast medium with bone lysis and formation of new tumor bone. Whereas MRI is better in delineating soft-tissue infiltration [7], it shows a heterogeneous mass with an intermediate T1 signal and a marked T2 hypersignal, strongly enhancing after injection of gadolinium, with frequently associated bone erosion.

Osteosarcomas of the mandibleare are osteoblastic in 50% of patients often with a periosteal reaction [7]. Tumor matrix mineralization and aggressive bone destruction is strongly suggestive of osteosarcoma [8]. Presence of matrix mineralization and periosteal new bone formation favors a diagnosis of osteosarcoma over metastatic carcinoma, lymphoma, and myeloma; a destructive mass is generally not found in radiation osteitis. A less aggressive osteosarcoma may be radiologically indistinguishable from chondrosarcoma [8].

The histologic criteria for diagnosis include the presence of nuclear pleomorphism, bizarre cell forms, a pattern of interlacing bundles of smooth muscle cells, and a high rate of mitoses. The literature review shows that the best choice of treatment is complete surgical resection of the lesion with tumor-free margins [9]

As, in head and neck sarcomas, it is difficult to obtain wide margins during surgical treatment, because of anatomical constraints most patients with locally resectable tumors undergo postoperative irradiation. Adjuvant systemic chemotherapy seems to improve outcome, but its benefit must be weighed against associated toxicities [10].

Conclusion

Radiation induced sarcoma is a rare but recognized complication of radiotherapy and is associated with poor prognosis. Several cases have been reported in the literature. The diagnostic criteria of post-irradiation osteosarcoma include a lesion centered in irradiated bone without a primary malignant osteoblastic lesion, arising after a latency period of at least 3 years after the completion of radiation therapy. Both CT and MR imaging are excellent for revealing tumor extent and as aids in presurgical planning.

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