Case Report / Olgu Sunumu

Hyoid osteoradionecrosis accompanied by candida infection

Kandida enfeksiyonuna eşlik eden hyoid osteoradyonekrozu

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ABSTRACT

Osteoradionecrosis of the hyoid bone is a rare complication of therapeutic irradiation performed for head and neck cancer. In this article, we present a 52-year-old male patient who admitted with severe odynophagia following chemo-radiotherapy administration for tonsil carcinoma. Fluorine-18-fluorodeoxy-glucose positron emission tomography-computed tomography revealed a metabolic activity in hyoid bone. The pathological findings were consistent with fungal infection and hyoid bone necrosis. Hyoid osteoradionecrosis should be kept in mind in patients with intractable dysphagia following irradiation for head and neck tumors.

Keywords: Head and neck cancer; hyoid; osteoradionecrosis; radiotherapy; squamous cell carcinoma.

ÖZ

Hyoid kemiğin osteoradyonekrozu baş ve boyun kanseri için uygulanan terapötik irradyasyonun nadir bir komplikasyonudur. Bu yazıda, tonsil karsinomu nedeni ile kemoradyoterapi uygulamasının ardından ciddi odinofaji ile başvuran 52 yaşında bir erkek hasta sunuldu. Florin-18-florodeoksi-glukoz pozitron emisyon tomografi-bilgisayarlı tomografide hyoid kemikte metabolik aktivite görüldü. Patolojik bulgular fungal enfeksiyon ve hyoid kemik nekrozu ile uyumlu idi. Baş ve boyun tümörlerine yönelik irradyasyon ardından inatçı disfajisi olan hastalarda hyoid osteoradyonekrozu akılda tutulmalıdır.

Anahtar Sözcükler: Baş ve boyun kanseri; hyoid; osteoradyonekroz; radyoterapi; yassı hücreli karsinom.

Osteoradionecrosis (ORN) is a well-known long-term complication of therapeutic head and neck irradiation. Osteoradionecrosis describes the process where irradiated bone undergoes necrosis and becomes exposed through soft-tissue. The most common site of ORN is the mandible and the incidence of mandibular ORN is reported to be between 2-22%.[1] On the other hand, ORN of the hyoid bone has been described in only a few patients.[2-8] Herein we report a case of hyoid bone ORN and discuss the reported cases in the literature. Our case was unique in that it was accompanied by candida infection.

CASE REPORT

A 52-year-old man with a history of dysphagia presented to our institution. Physical examination revealed a mass on the right tonsil accompanied

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with right cervical lymphadenopathies. The diagnosis was confirmed by excisional biopsy of the right tonsil, which revealed squamous cell carcinoma. The patient subsequently underwent fluorine-18-fluorodeoxy-glucose (FDG) positron emission tomography computed tomography (PET/CT). Metabolic imaging with dual modality PET/CT examination was obtained after intravenous administration of 370 megabecquerel FDG using an integrated scanner (Biograph mCT, Siemens Healthcare, Erlangen, Germany). Computed tomography was performed without intravenous contrast injection with 120 kV, 35 mA, pitch of 1.5, section thickness of 5 mm and field of view 70 cm. The PET scan was started immediately after unenhanced CT. Standardized uptake value (SUV) was calculated using "region of interest" (ROI) technique. Positron emission tomography CT examination revealed a mass lesion from base of tongue to the soft palate expanding through the lingual and palatine tonsil with a maximum standardized uptake value (SUV_{max}) of 14.50. There were also conglomerated lymph nodes on the right cervical region with high FDG uptake (SUV_{max}: 21.50). The patient was diagnosed with $T_3N_{2b}M_0$ tonsil squamous cell carcinoma and three cycles of induction taxotere, cisplatin and fluorouracil (TCF) was administered. We repeated the PET/CT examination in order to define the tumor response to induction chemotherapy. Extensive glucose uptake was observed in the right jugulodigastric lymph nodes (SUV_{max}: 3.60). We obtained a good response as both the primary tumor and metastatic right cervical lymph nodes were diminished in size and metabolic activity. Therefore we planned to continue with concomitant chemo-radiotherapy. We applied 7000 cGy in 35 fractions to the primary tumor, 6600 cGy to right neck and 5000 cGy to the supraclavicular and left neck regions with concomitant weekly cisplatin. We used three-dimensional conformal radiotherapy technique with linear accelerator. After 10 months of treatment, the patient developed hoarseness and severe odynophagia. He did not respond to symptomatic treatments. A detailed physical and endoscopic examination revealed an edematous appearance of the mucosal surfaces of the oral, oropharynx, hypopharynx and larynx. Magnetic resonance imaging showed a 3x1.5 cm contrast-enhancing solid mass extending from the hypopharynx to the right vocal cord (Figure 1). A biopsy revealed candida infection on histopathological examination. The patient received antifungal treatment; however his odynophagia was not totally improved. Repeat physical and endoscopic examination showed a suspicious lesion in the left great cornu of the hyoid bone. Repeat PET/CT imaging performed to differentiate the radiation-induced mucosal swelling and tumor recurrence revealed metabolic

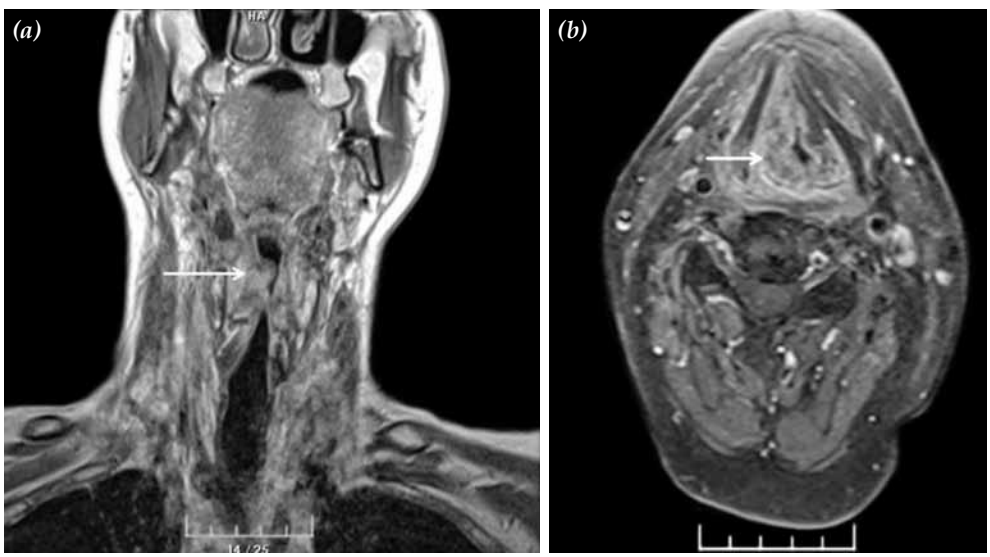

Figure 1. ***(a)*** *Coronal, and* ***(b)*** *axial magnetic resonance imaging of the patient. A 3x1.5 cm contrast-enhancing solid mass is seen extending from hypopharynx to the right vocal cord (arrow). The mass is narrowing the airway.*

activity in the hyoid bone (SUV_{max}: 12.89, Figure 2) and a lymph node in the right lower paratracheal region (SUV_{max}: 5.98). The direct laryngoscopy and biopsy was repeated and pathological findings revealed resistant fungal infection and hyoid bone necrosis. The left great cornu of the hyoid bone had become exposed through soft-tissue which explained the severe odynophagia (Figure 3). Antifungal treatment was resumed.

DISCUSSION

Osteoradionecrosis is a well-recognized complication of radiotherapy. It is defined by non-healing bone in an area of previous irradiation without evidence of malignancy.[9] With its high density and poor vascularity, the mandible is more prone to develop ORN. Although ORN of the mandible is commonly discussed in the context of head and neck radiation; its occurrence in the hyoid bone is rarely reported and likely underappreciated. A review of the literature reveals only 22 previously reported cases.[2-8]

Bhatia et al.[2] reported the first case of hyoid bone ORN in 1979. The case was a 55-year-old woman with the diagnosis of priform fossa tumor who was treated with cobalt therapy. Parallel to the developments in the field of radiation oncology the radiation exposures of normal regional tissues are significantly reduced. It is well-known that different radiation delivery methods present different levels of risk for developing ORN.[10] In the case reported by Bhatia et al.[2] the radiation technique was two-dimensional radiotherapy. On the other hand we used three-dimensional conformal radiotherapy in our case.

After this, Robertson et al.[3] reported the second case in 1995. In this case multimodal treatment was applied including surgery, chemotherapy and radiotherapy for laryngeal carcinoma. In our case, excisional biopsy was performed on the right tonsil in order to make the diagnosis and the patient received three cycles of TCF induction chemotherapy followed by concomitant chemoradiotherapy. Although using multimodal therapy in head and neck cancer patients has been shown to increase acute toxicity especially mucositis, there is no such data with respect to ORN.[10,11] However the variable outcome of the studies observing the risk of development of ORN indicates that the

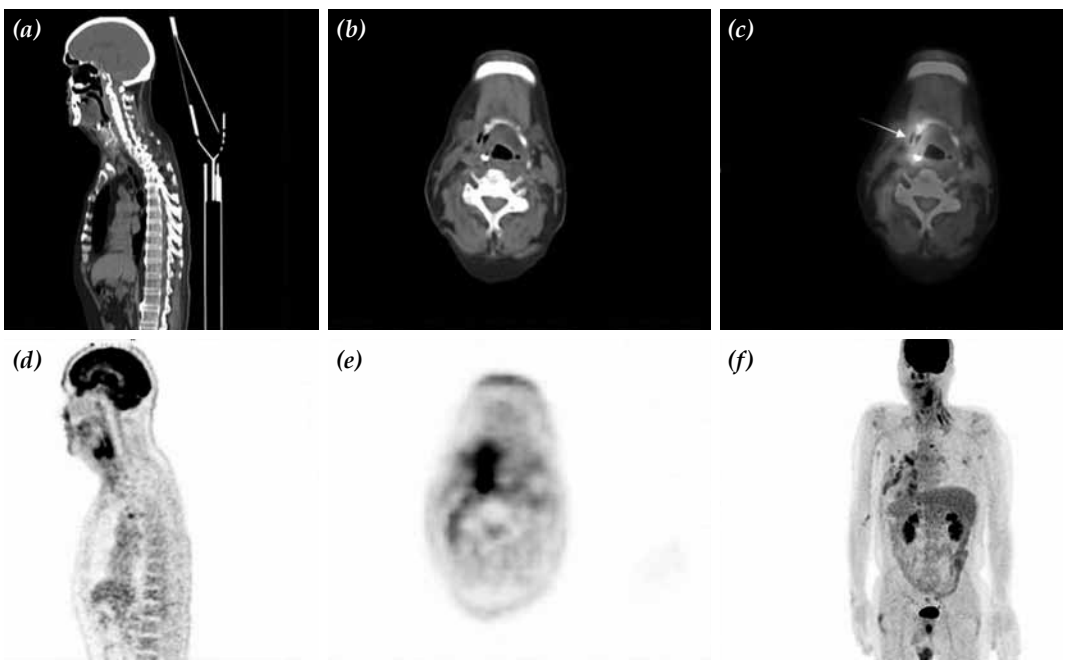

Figure 2. ***(a)*** *Sagittal, and* ***(b)*** *axial, computed tomography;* ***(c)*** *fused transaxial positron emission tomography/computed tomography,* ***(d)*** *sagittal,* ***(e)*** *axial and* ***(f)*** *maximum intensity projection positron emission tomography images of the patient. High fluorodeoxyglucose uptake with SUV_{max} value of 12.89 was observed in hyoid bone on transaxial fused positron emission tomography/computed tomography image (arrow).*

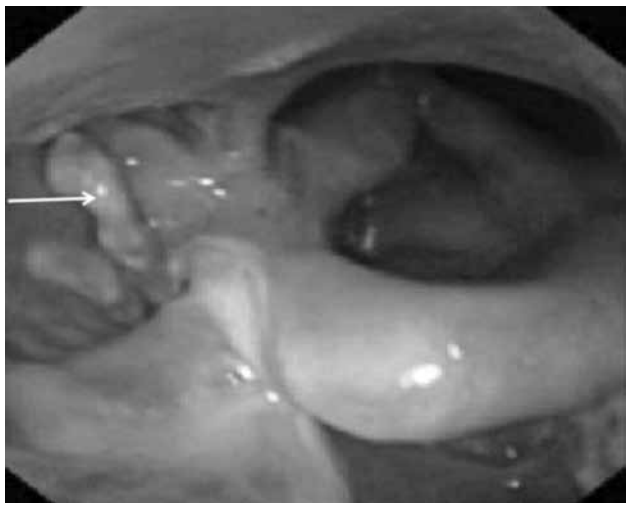

Figure 3. *Direct laryngoscopy revealed that the left great cornu of the hyoid bone had became exposed through soft-tissue (arrow).*

addition of chemotherapy to radiotherapy does not appear to increase the risk of developing ORN.[10]

It is also well-known that there is higher risk of developing ORN with higher radiation doses.[10] Yoo et al.[7] reported the largest series of hyoid ORN with imaging findings of 13 cases. The authors retrospectively contoured the hyoid bone from their treatment planning system and their findings showed that the mean dose of the hyoid bone ranged between 66.8-71.6 Gy. In our case the maximum dose measured in the hyoid bone was 69.5 Gy and the mean dose was 68.7 Gy. In the study by Yoo et al.[7] the elapsed time between completion of radiotherapy and radiologic observation of ORN ranged between 1-34 months. In our case the time from the end of radiotherapy to the diagnosis of hyoid ORN was 10 months.

In the reported cases, the most frequent associated symptom was odynophagia, which was also seen in our case. Patients with a history of malignancy closely approximating the hyoid, particularly with involvement of the overlying mucosa, seem to have a greatly increased risk of hyoid ORN following radiation therapy.[7] In the current case after observing intractable dysphasia, we initially wanted to rule out a recurrent tumor. Therefore we performed a detailed head and neck examination and neck MRI. The edematous appearance of the mucosal surfaces first suggested a radiation related side effect; however the MRI findings led us to suspect a recurrent tumor since there was a contrast-enhancing solid mass. However histopathological examination of the biopsy revealed candida infection. Although anti-fungal treatment was started, the odynophagia worsened a short time after treatment. Again we suspected recurrent disease and performed a PET/CT which revealed extensive glucose uptake in the hyoid bone.

Although hyoid bone ORN is a rare complication of head and neck irradiation; it should be kept in mind in patients with intractable dysphagia. Adjacent soft-tissue enhancement may suggest the possibility of tumor recurrence in these patients; however, the characteristic osseous changes of ORN make post-radiation sequelae more likely. The potential for false-positive findings in PET/CT in such cases must also be borne in mind; because the lesion may be accompanied by an infection.

Declaration of conflicting interests

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