

Postirradiation osteosarcoma of the occipital bone: A case report

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ABSTRACT

Postirradiation osteosarcoma is an uncommon complication of radiation treatment for brain tumors. It is associated with a poor prognosis and survival is usually <1 year. Herein, we report the case of a 58-year-old gentleman, who developed osteosarcoma of the occipital bone 5 years after the radiation treatment of oligodendroglioma in the right temporal lobe. He was treated for oligodendroglioma with surgery and radiation using two-dimensional radiation therapy technique to a total dose of 54 Gy through two lateral portals in two phases. Secondary osteosarcoma of the occipital bone occurred 5 years later on the ipsilateral side but within the previous field of irradiation. After metastatic workup, the patient was treated with surgery and adjuvant chemotherapy, but there was a progression of the disease after 6 cycles and the patient died 8 months after the diagnosis.

Key words: Irradiation, Oligodendroglioma, Osteosarcoma, Skull

Radiation therapy plays an important role as a curative modality in the management of most solid tumors, but its therapeutic use may be associated with the development of second malignancies [1,2]. Radiation-induced second malignancy is a stochastic effect. The criteria for the diagnosis of a radiation-induced secondary malignancy include the development of tumor in the previously irradiated field, histology of the second malignancy should be different from that of the original tumor and long latency period of at least 3 years after radiotherapy [3,4]. Radiation therapy in the management of central nervous system tumors may induce the second malignancies such as gliomas, meningiomas, and sarcomas [1,2,5,6]. Fibrosarcomas are the most common radiation-induced tumors after therapeutic radiation in the central nervous system.

Osteosarcoma is an uncommon complication of radiation treatment for brain tumors. Majority of the osteosarcomas arise *de novo* in bones; approximately 5.5% of all osteosarcomas are due to exposure to radiation. The risk of developing a radiation-induced osteosarcoma is estimated at 0.01–0.03% among all irradiated patients [6,7]. The most common site is the skull. Surgery and chemotherapy have been the mainstay of treatment modalities for osteosarcomas secondary to radiation exposure. However, postirradiation osteosarcoma is associated with a poor prognosis [8,9].

Herein, we report a case of a 58-year-old man who developed osteosarcoma of the occipital bone 5 years after radiation treatment for oligodendroglioma in the right temporal lobe and was treated with surgery and chemotherapy.

CASE REPORT

A 58-year-old man had presented to the hospital with a chief complaint of seizures in June 2009 and evaluation had revealed space-occupying lesion in the right temporal lobe. The right temporal craniotomy and excision of the lesion were done in November 2009. Post-operative histopathology was suggestive of the oligodendroglioma World Health Organization (WHO) Grade 2. He received adjuvant radiotherapy of 54 Gy in 28 fractions from January 2010 to February 2010 through two lateral portals in two phases (Fig. 1) (Phase 1: 50 Gy in 25 fractions at 2 Gy per fraction; Phase 2: 4 Gy in 2 fractions at 2 Gy per fraction). The patient also received concurrent chemotherapy with temozolomide during radiation therapy. He also received adjuvant chemotherapy with (procarbazine, lomustine, and vincristine) for 6 cycles from March to September 2010. Then, he was asymptomatic for 5 years. Contrast-enhanced computed tomography (CECT), done in September 2014, showed no evidence of tumor recurrence or any other lesion.

The patient presented again in September 2015 with complaints of painless, progressive swelling in the right occipital region for the past 6 months. On examination, there was a diffuse 6 cm × 3 cm × 3 cm swelling in the right occipital region with a small ulcer in the skin. The swelling was firm to hard in consistency. The patient was ambulant and was not having any neurological deficit. Ophthalmological examination revealed visual acuity of 6/9 in the right eye and 6/18 in the left eye with no evidence of papilledema.

CECT showed gliotic changes in the right parietotemporal region; lytic destruction of the right parietal and occipital

bone with diploic expansion and mildly enhancing intracranial (extradural plane) and extracranial (galeal plane) soft tissue (Fig. 2). Magnetic resonance imaging (MRI) study demonstrated multiloculated cystic lesion with septal enhancement and no increased cerebral blood volume (CBV), no restriction of diffusion, in the right superior temporal lobe, possibly radiation necrosis, and enhancing soft tissue lesion in the right occipital extracranial and extradural region, with destruction of the underlying occipital bone showing focal increased CBV (Fig. 3).

The patient underwent excision of the right occipital lesion and titanium mesh cranioplasty in December 2015. Immediately after raising the scalp flap, a large amount of necrotic material came out and the undersurface of scalp was found to be infiltrated. The infiltrated portion of the scalp along with the ulcerated skin margins was excised. The dura was found to be grossly intact and transverse sinus was identified. Titanium mesh cranioplasty was done and the wound was closed primarily in layers. Histological examination revealed neoplasm composed of spindled to polygonal cells in sheets. The cells showed moderate nuclear pleomorphism, increased nuclear-cytoplasmic ratio and active mitosis including atypical forms. There was the presence of malignant osteoid, focal myxoid change, and vast areas of necrosis. The tumor cells are glial fibrillary acidic protein negative with high MIB-1 labeling index (20–25%). The histological diagnosis was the right occipital osteosarcoma.

Post-operative CECT showed significant decompression of tumor with no intradural residual lesion with titanium mesh cranioplasty in situ. However, contrast MRI study showed a 4.8 × 1.9 cm sized T1 hypointense, T2/fluid-attenuation inversion recovery isointense extra-axial biconvex enhancing lesion in the right occipital region; it also showed multiseptated cystic lesion in the right temporal region with no significant choline elevation in magnetic resonance spectroscopy, features suggestive of radiation necrosis. CECT thorax and bone scan were done to rule out lung and bone metastases, respectively, and they were essentially normal. The patient underwent 3 cycles of chemotherapy with doxorubicin 60mg/m² D1 and ifosfamide 1.5g/m² D1–D3. MRI imaging revealed a mild reduction in the occipital lesion. Then, he received 3 more cycles of chemotherapy, but there was a progression of the local disease, and the patient died in August 2016, probably because of raised intracranial tension and significant mass effect.

DISCUSSION

Radiation therapy plays a significant role as adjuvant therapy in many brain tumors. However, its utility may be associated with a rare complication of secondary malignancies [1,2]. As mentioned above, the diagnostic criteria for radiation-induced second malignancy include three criteria, and in the present case, the patient fits all the criteria, thus confirming the diagnosis of radiation-induced second malignancy.

The latent period is defined as the interval between irradiation of an initial tumor and development of postirradiation

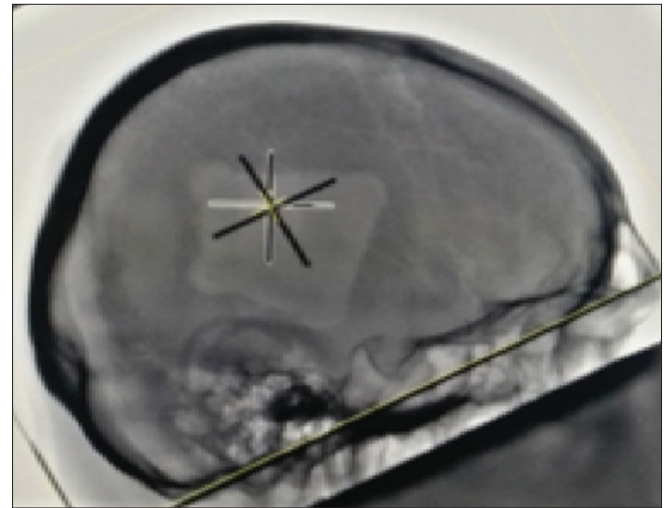


Figure 1: Radiation portals



Figure 2: Contrast-enhanced computed tomography showing the lesion

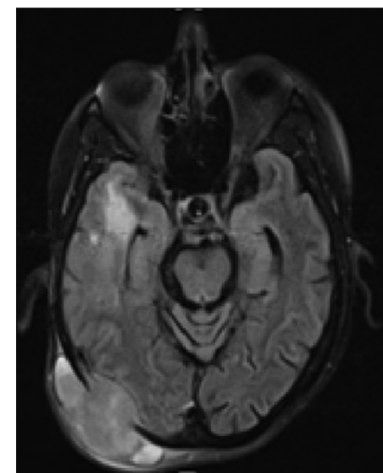


Figure 3: Magnetic resonance imaging showing the lesion

tumors. This latency is influenced by the dose and the size of the radiation field. High dose of radiation and large radiation volume may be associated with a short latent period. When we analyzed the literature, we found that the average latent period for the development of meningioma is 18.4 years when high-dose

radiation is given and 36.8 years when low-dose radiation is given. The average latent period for the development of osteosarcoma is about 9.1 years [5,6,10]. In our case, the patient developed osteosarcoma of the occipital bone adjacent to the primary lesion but within the irradiated field, approximately 5 years after radiation treatment for the brain tumor.

Development of osteosarcomas after radiation therapy for brain tumors such as craniopharyngioma, pituitary adenoma, medulloblastoma, and gliomas has been reported in the literature [11]. Watanabe *et al.* [12] reported radiation-induced osteosarcoma of the left parietal and occipital bone 16 years after surgery and radiation for glioma in the right occipital lobe, but the type of glioma was not known. Till now, the largest series had been reported by Patel *et al.* [13] in which they identified 16 patients with radiation-induced osteosarcomas of the calvarium and skull base, of which two patients had received initial radiation therapy for oligodendroglioma. In our case also, the patient developed osteosarcoma in the occipital bone after receiving radiation therapy for oligodendroglioma.

Surgery and chemotherapy have been the mainstay treatment modalities for osteosarcoma of the skull. However, postirradiation osteosarcoma is associated with a poor prognosis due to high rates of local recurrence [3,14]. In our case also, the patient underwent excision of the right occipital lesion and titanium mesh cranioplasty and received 6 cycles of adjuvant chemotherapy with doxorubicin and ifosfamide, but the patient developed progression of a local disease and died. The median survival for patients with postirradiation osteosarcomas ranges from 12 to 18 months, and it decreases to <12 months for those with intracranial extension [8,9].

CONCLUSION

Radiation-induced malignancy is one of the most deadly long-term complications of therapeutic radiation exposure. Since the role of radiation therapy in the radical treatment of most solid tumors continues to evolve and the survival of cancer patients continues to increase, it is mandatory for the treating radiation oncologist to be aware of the postirradiation secondary malignancies and

to counsel and to monitor the patients for the occurrence of this uncommon complication.

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