Nasopharyngeal carcinoma is the 9th common malignant tumor seen in male Chinese and the 12th common in female Chinese in Taiwan, full-course radiotherapy being the mainstay of treatment. With the improvement of radiotherapy techniques and consequent patient survival, the chance for long-term complications, such as radiation-induced tumors, would appear to increase.

CASE REPORT

In October 2000, a 57-year-old Chinese woman presented 5 years after radiotherapy for nasopharyngeal carcinoma with a 6-month history of a gradually enlarging left postauricular mass. The original nasopharyngeal carcinoma (Fig. 1) was staged T2N0M0 with a total of 7,000 cGy of radiation rendered for the malignancy. The patient appeared to be disease-free in the nasopharynx subsequent to the radiation therapy. A physical examination revealed a bony hard, immobile mass, 6.0 × 6.0 cm, overlying the mastoid tip and firmly attached to the skull. Computed tomographic scans of the mastoid bone demonstrated a densely calcified mass with radiating bony spicules, applied to left mastoid tip. The lesion was excised en-bloc through a postauricular incision. The histologic diagnosis was a parosteal osteosarcoma. Because of inadequate safe margins and the patient refusal of another surgery, 6,600 cGy of radiation was subsequently administered to the temporal bone. Post-operative follow-up in 3 years was negative for any evidence of tumor recurrence and post-irradiation complications.

Radiation-induced tumors subsequent to nasopharyngeal carcinoma are very rare. To date, no post-irradiation parosteal osteosarcoma of the craniofacial bone has been reported in the English literature. In October 2000, a 57-year-old Chinese woman presented 5 years after radiotherapy for nasopharyngeal carcinoma with a 6-month history of a gradually enlarging left postauricular mass. CT scans revealed a densely calcified mass with radiating bony spicules, applied to left mastoid tip. The lesion was excised en-bloc through a postauricular incision. The histologic diagnosis was a parosteal osteosarcoma. Because of inadequate safe margins and the patient refusal of another surgery, 6,600 cGy of radiation was subsequently administered to the temporal bone. Post-operative follow-up in 3 years was negative for any evidence of tumor recurrence and post-irradiation complications.

**Fig. 1** Histopathologic findings of the nasopharyngeal tumor showing plexiform masses of non-keratinizing tumor cells with pale eosinophilic cytoplasm and oval-shaped vesicular nucleus containing a prominent nucleolus (H&E, × 150).
scribed a densely calcified mass with radiating bony spicules, applied to the outer cortex of an otherwise normal mastoid (Fig. 2). The lesion was excised through a postauricular incision through which the ear canal, facial nerve and upper neck structures were well visualized. The bony attachments were divided with the aid of an osteotome and a drill. Histological examination of the specimen revealed an infiltration of lobules of pleomorphic spindle tumor cells exhibiting frequent mitoses and osteoid formation in fibrotic connective tissue and muscle (Fig. 3). The diagnosis of parosteal osteosarcoma was thus confirmed. The patient exhibited an excellent postoperative recovery without facial palsy. Because of inadequate safe margins and the patient refusal of another surgery, 6,600 cGy/33 fractions of radiation was subsequently administered to the temporal bone using 3-D conformal techniques. Post-operative follow-up in 3 years was negative for any evidence of tumor recurrence and post-irradiation complications.

**DISCUSSION**

The criteria for diagnosis of radiation-induced tumors, proposed by Cahan et al. and later modified by Arlen et al., include the following: 1. a history of previous radiotherapy; (2) the second neoplasm found within the previous field of radiation; (3) the histology of the second neoplasm distinctly different from that of the primary tumor; and (4) the existence of a fairly long latency period, eg. at least 5 years. The case we describe here fulfills all the criteria for a post-irradiation parosteal osteosarcoma of the mastoid bone. The location of this mass away from the margins of nasopharyngeal carcinoma and the presence of calcification in the mass, as revealed in the computed tomographic scans, would suggest the diagnosis of radiation-induced sarcoma rather than a recurrent nasopharyngeal carcinoma.

Parosteal osteosarcoma is an uncommon surface tumor of bone that is believed to arise in the periostium or in the immediate parosteal connective tissues. It is a rare variant of conventional osteosarcoma, comprising less than 4 percents of all osteosarcomas. Most often it occurs in the long bone with female predominance. From our extensive review, only 6 cases of parosteal osteosarcoma of the mastoid bone have been reported in the English-written literature, all of which have demonstrated the absence of any history of previous radiotherapy. The case we report here is an additional example, which, to the best of our knowledge, is the first case of radiation-induced parosteal osteosarcoma of the mastoid bone.

Parosteal osteosarcoma is a low-grade form of osteosarcoma, growing slowly with a significantly better prognosis than conventional osteosarcoma. Due to its bland microscopic appearance, the differential diagnosis may include giant osteoma, conventional osteosarcoma,
chondrosarcoma, sessile osteochondroma, organizing hematoma, heterotopic ossification and myositis ossificans. Furthermore, the differential diagnoses of osteoradionecrosis and metastasis from the primary tumor, and even a distant second tumor, may be considered as viable options for an irradiated patient with a mass in the mastoid bone. Calcification with radiating bony spicules, revealed in the computed tomographic scans, has been demonstrated to be characteristics of parosteal osteosarcoma as is the case for our cited example.

The mainstay of treatment for parosteal osteosarcoma of the mastoid bone is adequate surgical resection, which appears to be curative for most cases. Adjuvant chemoradiotherapy is only used for cases where wide surgical margins cannot be obtained. Because of the relative radio-resistance of osteosarcomas, a radiation dose exceeding 6,000cGy for it to be effective is needed. The possibility of subsequent osteoradionecrosis, radiation myelitis and brain abscess should be kept in mind and prospective preventive treatment, such as delicate 3-D conformal techniques plus a tailored mask, should be considered for a case of post-irradiation tumor.

REFERENCES