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Case Report

Osteosarcoma of the temporal bone occurring 40 years after radiotherapy: A technical case report presenting *en bloc* resection of intra- and extracranial lesions followed by a one-stage reconstruction

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ABSTRACT

Background: Osteosarcoma (OS) is a malignant tumor of the bone, which rarely occurs in the head-and-neck regions as a primary or a secondary malignancy. Adequate surgical resection is currently the mainstay of treatment for head-and-neck OS; however, *en bloc* resection and reconstruction can be difficult because the anatomies of these regions are complex. We present a case of an OS arising from the temporal bone 40 years after radiation therapy, which was successfully treated with *en bloc* resection and a one-stage reconstruction using intraoperative tissue expansion technique.

Case Description: A 62-year-old woman who underwent surgery and radiotherapy for a left temporal lesion 40 years before presentation was hospitalized for aphasia and a right hemiparesis. She had a 4 × 3 cm subcutaneous mass in the left temporal area of the head. Computed tomography imaging showed destruction of the left temporal bone and a partially calcified mass. Magnetic resonance imaging showed an enhancing mass with intracranial and extracranial cystic components (5 cm and 3 cm in diameter, respectively). Due to rapid growth of the lesion, a semi-urgent surgery was performed. In this operation, a continuous narrow craniectomy was performed around the tumor using a ruler. Then, *en bloc* resection of the tumor, with adjacent skin, temporal muscle, skull, dura mater, and cerebral cortex, was achieved. Subsequently, a one-stage reconstruction of the dura mater, skull, and skin of the head was performed using fascia lata, artificial bone, and a local skin flap combined with intraoperative tissue expansion using a 20-French Foley catheter. Postoperative histological examinations revealed the tumor to be an OS.

Conclusion: We have presented a rare case of an OS occurring from the temporal bone 40 years after radiation therapy. We describe our experience and the surgical methods in this case to provide options for surgical strategies in patients with head-and-neck OS.

Keywords: *En bloc* resection, Osteosarcoma, Radiation, Reconstruction, Temporal bone

INTRODUCTION

Osteosarcoma (OS) is a malignant bone tumor that can occur anywhere in the body, though most commonly affects the long bones in young adults and children. The incidence of OS is estimated to be 1:100,000 per year,[9] and approximately 6%–10% of lesions occur in the

head-and-neck regions. OS rarely occurs as a secondary tumor after radio- or chemotherapy for other lesions.[5,17,19] Although adjuvant chemotherapy has been shown to have a good response rate in OS of the long bones in young patients, its role in head-and-neck OS remains unclear.[12] Therefore, adequate surgical resection is currently the mainstay of treatment for head-and-neck OS.[2,10,12] Unfortunately, it can be difficult to obtain negative surgical margins in these cases because the anatomy of the head and neck is more complex than the extremities.[7] We present a very rare case of a patient who presented with an OS arising from the temporal bone 40 years after radiation therapy. In this case, *en bloc* resection of the tumor with adjacent skin, temporal muscle, skull, dura mater, and cerebral cortex was executed. Subsequently, a one-stage reconstruction of dura mater, skull, and skin of the head was performed using fascia lata, artificial bone, and a local skin flap combined with an intraoperative tissue expansion technique.[4,11] We describe our experience and the surgical methods related to this case to provide options for surgical strategies in patients with head-and-neck OS.

CASE DESCRIPTION

Initial presentation

A 62-year-old woman who underwent surgery and radiation therapy for a left temporal lesion at another hospital 40 years before presentation was admitted to our hospital due to progressive aphasia and right hemiparesis. She was found to have a 4 × 3 cm subcutaneous mass in the left temporal area of the head [Figure 1a]. Computed tomography (CT) imaging showed destruction of the left temporal bone and a partially calcified mass lesion [Figure 1b]. T1-weighted contrast-enhanced magnetic resonance imaging (MRI) showed an enhancing mass with intracranial and extracranial cystic components (5 cm and 3 cm in diameter, respectively) [Figures 1c and d]. Although detailed information related to the previous surgery and radiotherapy was not available, a semi-urgent surgery was planned because two of the cystic lesions were growing rapidly, and the patient's symptoms continuously worsened after admission.

Operative procedure

The surgical procedure was performed in the right semilateral position under general anesthesia. The scar of the previous operation [Figure 1a] was reincised, preserving a frontal branch of the superficial temporal artery. An additional skin incision was made around the subcutaneous mass lesion, and an island-shaped portion of skin and temporal muscle covering the subcutaneous mass was left intact on the tumor side. The surrounding skin flap was then flipped, and artificial bone constructed during the previous surgery was observed on the vertex side of the tumor [Figure 2a]. A small bone window was made just above the artificial bone using a high-speed drill. Subsequently, a continuous narrow

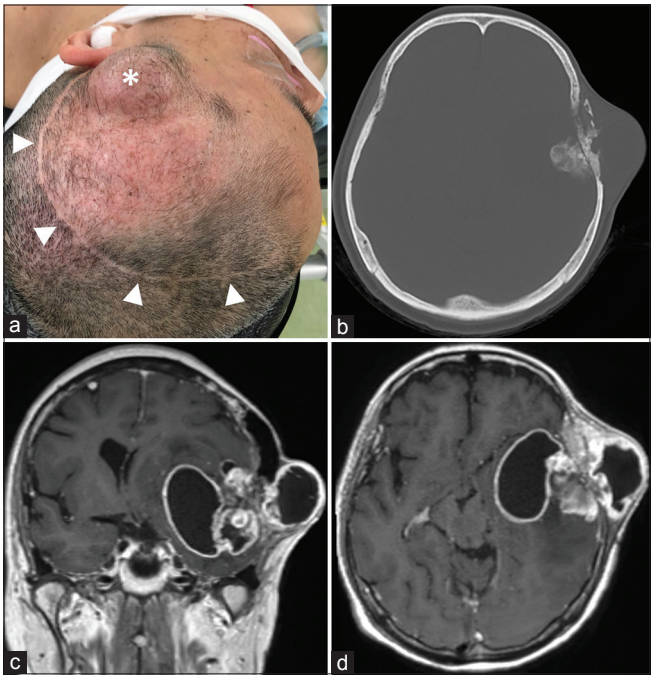

Figure 1: Clinical/radiologic findings at patient presentation. (a) Photograph showing a 4 × 3 cm subcutaneous mass in the left temporal area of the head (white asterisk) and a previous skin incision (white arrowheads). (b) Computed tomography imaging showing destruction of the left temporal bone and a partially calcified mass lesion. (c and d) T1-weighted contrast-enhanced magnetic resonance imaging showing an enhancing mass with intracranial and extracranial cystic components.

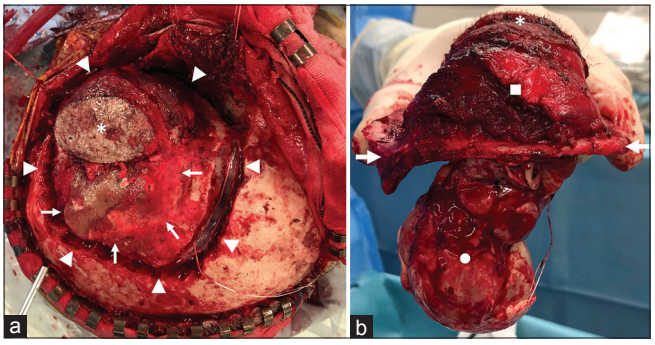

Figure 2: Operative photographs. (a) Photograph showing that the skin and the temporal muscle covering the subcutaneous mass were left on the tumor side (white asterisk), and the skin flap was flipped. An artificial bone constructed in the previous surgery existed on the vertex side of the tumor (white arrow). A continuous narrow craniectomy was performed surrounding the tumor and the artificial bone (white arrowheads), and a dural incision was made using the space created by this narrow craniectomy. (b) Photograph showing the specimen obtained by en bloc resection of the tumor with adjacent skin (white asterisk), temporal muscle and extracranial lesion (white square), temporal bone (white arrows), dura mater, and intracranial lesion (white circle).

craniectomy approximately 1 cm in width was performed surrounding the tumor and the artificial bone [Figure 2a]. This procedure was carefully performed using a ruler to maintain the safety margin of the tumor and to avoid tumor cell dissemination. A dural incision surrounding the tumor and the previous artificial bone were made using the space created by the narrow craniectomy [Figure 2a]. The middle meningeal artery, one of the main feeders of the tumor, was coagulated and cut during this procedure. The temporal bone, dura mater, and intracranial tumor were lifted gradually, and the branches of the middle cerebral artery and the cortical veins were carefully detached from the mass lesion. Subsequently, *en bloc* resection of the tumor with adjacent skin, temporal muscle, skull, dura mater, and a thin slice of the temporal lobe cerebral cortex was achieved [Figure 2b]. After completion of hemostasis, the dura mater defect was reconstructed with fascia lata, and the temporal bone defect was reconstructed with artificial bone (Cemex® Bone Cement, Tecres S.p.A., Italy). After repeated intraoperative tissue expansion of the adjacent scalp using a 30 ml balloon in a 20-French Foley catheter [Figures 3a and b], a local rotation skin flap was created on the dorsal side of the skin defect [Figures 3c and d]. The skin defect was covered with this rotation skin flap and the expanded surrounding scalp, and a primary closure of the skin of the head was achieved [Figure 4a].

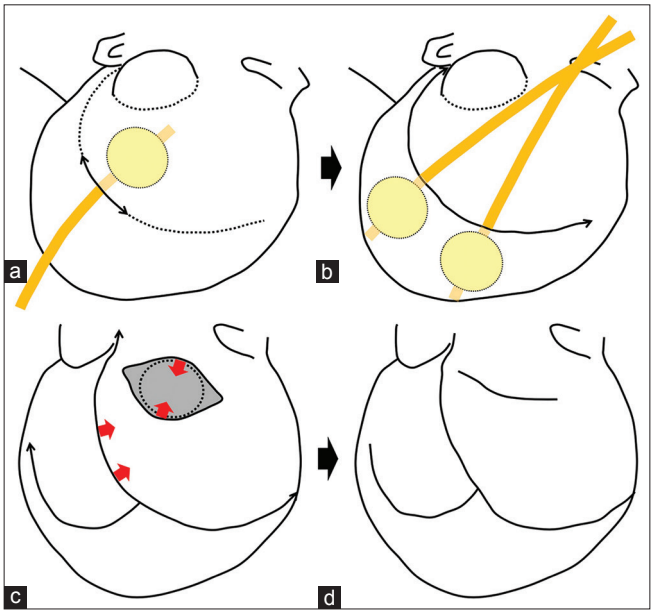

Figure 3: Illustration of the operative procedure. (a) Illustration showing intraoperative tissue expansion of the adjacent scalp using a 30 ml balloon in a 20-French Foley catheter after a small skin incision. The Foley catheter was inserted into the subcutaneous pocket, and the balloon was inflated with 30 ml of saline for 5 min and then deflated for 3 min. (b) Illustration showing intraoperative tissue expansion of the scalp on the parietal side with two Foley catheters. Repeated expansions were performed until sufficient expansion for a primary closure was achieved. (c) Illustration showing that the local rotation skin flap was created on the dorsal side of the skin defect. (d) Illustration showing that the skin defect was covered with this rotation skin flap and the expanded surrounding scalp, and a primary closure of the skin of the head was achieved. Thin black arrows show the skin incisions. Thick black arrows show the order of the procedures. Red arrows show the direction of the skin rotation.

Postoperative course

The temporal bone defect was shown to be adequately reconstructed with artificial bone on postoperative CT imaging [Figure 4c]. No residual tumor was identified on postoperative T1-weighted contrast-enhanced MRI [Figures 5a and b]. Histological analysis revealed that the tumor was comprised atypical spindle cell components with focal cartilaginous differentiation [Figures 6a and b]. The tumor was diagnosed as an OS, though the nature of the sarcoma remains unknown. Her symptoms, including aphasia and right hemiparesis, gradually improved within 2 months, and she was discharged from our hospital with no neurological deficits. Because a CT scan of the body revealed no evidence of tumor metastasis, and her blood test showed a mild leukocytopenia of unknown origin (leukocyte, 2860/μl; neutrophil, 1400/μl; and lymphocyte, 1200/μl), she was carefully observed in the outpatient clinic without postoperative adjuvant chemotherapy. The condition of the skin of the temporal region remained excellent without any signs of infection [Figure 4b], and no recurrence was found on follow-up MRI at 1 year after surgery [Figure 5c and d].

DISCUSSION

In this report, we have described a case of a patient who presented with OS arising from the temporal bone 40 years after an initial surgery and radiation therapy. The nature of the OS in this patient is still unknown, though there are two possibilities. First, the tumor could have represented a recurrence of her original malignancy from residual OS cells in the temporal bone. Second, it could have been a newly developed tumor induced by the patient's former radiation exposure. Unfortunately, information about this patient's previous clinical course was limited. While she remembered undergoing a prior surgery in the temporal region, as well as radiation therapy, she could not remember the diagnosis of her former malignancy. Although we contacted the previous hospital to confirm this patient's original diagnosis and her prior treatment course, her medical record was not available, and her primary surgeon was deceased. At present, we believe that the possibility of a radiation-induced secondary tumor is more likely because the 40-year latency period is likely too long for regrowth of residual tumor cells in the temporal bone.

Potential risk factors for the development of OS have been identified, including exposure to ionizing

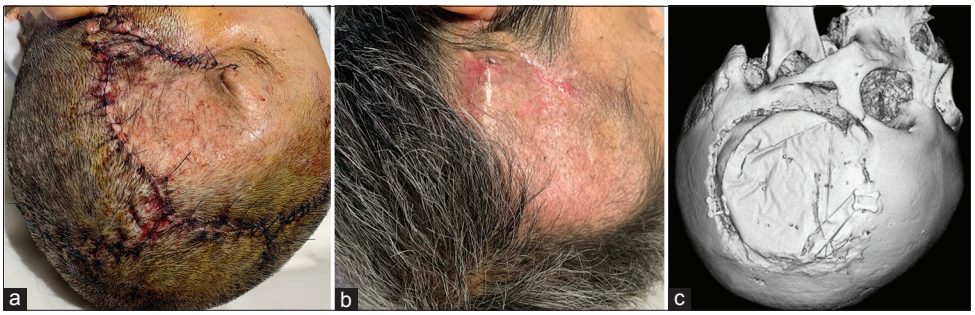

Figure 4: Postoperative photographs and computed tomography imaging. (a) Photograph showing the surgical head wound on postoperative day 1. (b) Photograph showing the condition of the surgical head wound on postoperative day 90. (c) Postoperative computed tomography imaging showing the temporal bone defect adequately reconstructed with artificial bone.

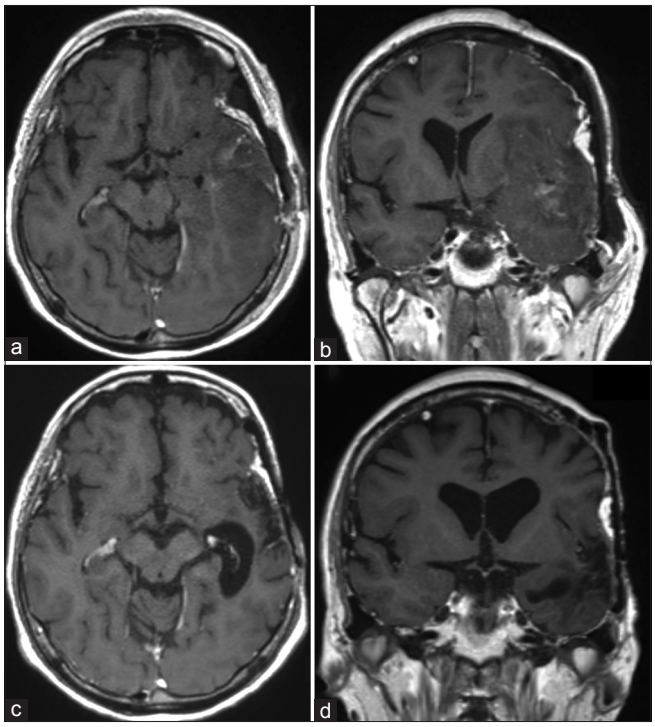

Figure 5: Postoperative magnetic resonance imaging. (a and b) Postoperative T1-weighted contrast-enhanced magnetic resonance imaging showing no residual tumor. (c and d) T1-weighted contrast-enhanced magnetic resonance imaging at 1 year after surgery showing no recurrence of the tumor.

radiation.[13,19] The incidence of radiation-induced malignant tumors is estimated to be 0.9% in patients who have undergone radiotherapy in any form for the head and neck.[15] Radiation-induced sarcomas represent 12% of these radiation-induced malignant tumors, with an overall incidence of 0.03–0.3% in patients exposed to radiotherapy.[8] The mean duration from initial radiotherapy to occurrence of radiation-induced sarcoma is reported to be 12.9–17.0 years.[13,19] Therefore, the present case is extremely rare, as the radiation-induced sarcoma developed 40 years after initial radiotherapy, an extraordinarily long latency period.

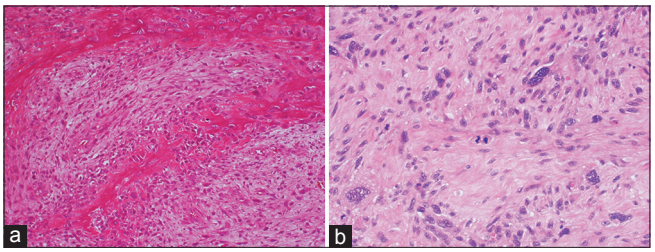

Figure 6: Micro-photographs of the specimens after hematoxylin and eosin staining. (a) The tumor was comprised atypical spindle cell components with focal cartilaginous differentiation. (b) Nuclear atypia and mitosis were occasionally seen.

A comprehensive treatment strategy, including surgery, chemotherapy, and radiotherapy, is generally preferred for patients with OS. In the literature, a high mortality in head-and-neck OS is associated with difficulty in local disease control, though it demonstrates less metastasis to the lung or other sites.[3] Complete tumor removal and negative resection margins have been reported to be significant prognostic factors in patients with OS.[7] Although *en bloc* resection with negative margins for skull base OS is difficult, Raza *et al.* reported that achieving negative margins also have a positive impact on prognosis of patients with these diseases.[14] Because radiation-induced OS is a rare condition, there are no disease-specific treatment guidelines; nevertheless, Wood *et al.* reported that complete tumor removal is also a significant prognostic factor for radiation-induced OS.[19] In the present case, we executed *en bloc* resection of the tumor with adjacent skin, temporal muscle, skull, dura mater, and a thin section of the temporal lobe cerebral cortex. On histological examination, there were no apparent tumor cells in the surrounding tissue, including the adjacent brain tissue. We performed a continuous narrow craniectomy surrounding the tumor using a ruler to obtain the safety margin more easily because the surgeon could feel bone abnormalities by the texture and solidity of the skull. Another advantage of this procedure was eliminating the risk of tumor cell dissemination through the use of a high-speed drill.

Intraoperative tissue expansion was first reported as "intraoperative sustained limited expansion" by Sasaki in 1987.[16] His report emphasized that extra tissue surrounding the skin defect was effectively obtained by cyclic loading of a temporary expander. We have recently adopted an intraoperative tissue expansion method using a 20-French Foley catheter with a 30 ml balloon as the standard expander apparatus.[4] In this case, we applied this technique for reconstruction of the large skin defect of the temporal area. The effect of skin expansion on the ventral side of the previous operation scar, which was likely irradiated because alopecia remained, was relatively limited compared with the dorsal side. Fibrotic skin from the previous radiation therapy appeared to decrease the elasticity of the skin in this area. Therefore, we created a local rotation skin flap combined with intraoperative tissue expansion on the dorsal side of the skin defect.

With current multimodal treatment strategies, approximately three-quarters of all patients with OS are cured, and 90%–95% of patients with OS can be successfully treated.[1] Radiotherapy was reported to improve overall survival and local control of tumor for patients with head-and-neck OS who have positive or uncertain resection margins.[6] Nevertheless, the role of adjuvant radiotherapy and chemotherapy for patients with radiation-induced OS is unclear.[18,19] In general, radiation-induced OS is resistant to radiation, and previous radiotherapy limits the possible irradiation dose and area.[8] In the present case, the patient was carefully observed in the outpatient clinic without postoperative adjuvant chemotherapy because there was no evidence of tumor metastasis on the whole-body CT scan and she had a mild leukocytopenia. At present, we are carefully following her condition by performing periodic CT scans of the body and MRIs of the brain. We plan on treating her with stereotactic radiotherapy, such as γ-knife or CyberKnife, if local recurrence or distant metastasis is identified.

CONCLUSION

We have presented a rare case of an OS occurring from the temporal bone 40 years after radiation therapy. Our experience and the surgical methods in this case will provide options for surgical strategies in patients with head and neck OS.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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