

Case Report

Volumetric-Modulated Arc Therapy for Giant-Cell Tumor of Temporal Bone: An Illustrative Case Report

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Keywords

Giant-cell tumor · Radiation therapy · Volumetric-modulated arc therapy · Magnetic resonance imaging

Abstract

Giant-cell tumor of the skull is extremely rare. Surgery is the main treatment for this disease, but not all cases are suitable for complete resection. In this report, we present the clinical features of a case of giant-cell tumor of temporal bone that demonstrated good outcome after radiation therapy (RT) using volumetric-modulated arc therapy (VMAT). The patient was a 55-year-old man with giant-cell tumor of temporal bone who received surgery as the first treatment. Three months after the initial surgery, the tumor regrew, and the patient received surgical resection again. Although second partial resection was undergone, it regrew. Therefore, 36 months after initial surgery, RT was conducted. The prescribed dose was 54 Gy in 1.8 Gy fractions using VMAT. The tumor began to shrink from 4 months after the initiation of RT and kept shrinking slowly and gradually. At the last follow-up, there was no evidence of local recurrence. There was no report about VMAT for giant-cell tumor of the skull, and no report revealed the radiographic details after recent radiation techniques. Therefore, this case report was meaningful in describing the details and response during and after VMAT for giant-cell tumor of temporal bone. The adjuvant RT using VMAT seemed to demonstrate a sufficient local control benefit without severe adverse effects in our case with giant-cell tumor of temporal bone.

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Introduction

Giant-cell tumor of bone is a rare primary bone neoplasm and accounts for 5% of all primary bone tumor [1]. Although histologically benign, clinically it is locally invasive and potentially metastatic. Giant-cell tumor of bone arises from undifferentiated mesenchymal cells of the bone marrow. It mainly develops in the epiphyses of long bones in the extremities and rarely occurs in the skull [2]. The therapy of giant-cell tumors of the skull represents a special challenge due to the anatomy, functionality, and cosmetics.

Surgery is the main treatment for giant-cell tumor of bone. Recent improvements in surgical techniques have resulted in sufficient local control [3, 4]. However, complete resection may cause deficits in cosmetic and function and be difficult in some areas, especially in such as the skull. In these cases, radiation therapy (RT) may be effective in the interest of the anatomy, functionality, and cosmetics. Some reports mentioned about RT for giant-cell tumor of the skull [2, 5]. However, the previous reports lacked recent radiation techniques, such as volumetric-modulated arc therapy (VMAT), which is a further technique that evolved from intensity-modulated radiotherapy.

In this report, we present the clinical features of a case with giant-cell tumor of temporal bone that demonstrated good outcome after RT using VMAT. Additionally, we discuss the details of this case in the context of the previously reported literature.

Case Presentation

The patient was a 55-year-old man with giant-cell tumor of temporal bone. He had no past medical history and complications. He presented with dizziness, tinnitus, and hearing loss on the right side. Imaging tests revealed the tumor of temporal bone. Figure 1 shows the computed tomography (CT) and magnetic resonance imaging (MRI) appearances before treatment. He received craniotomy for the tumor resection as the first treatment. While most of the tumor was removed, the tumor near right petrous bone was remained. However,

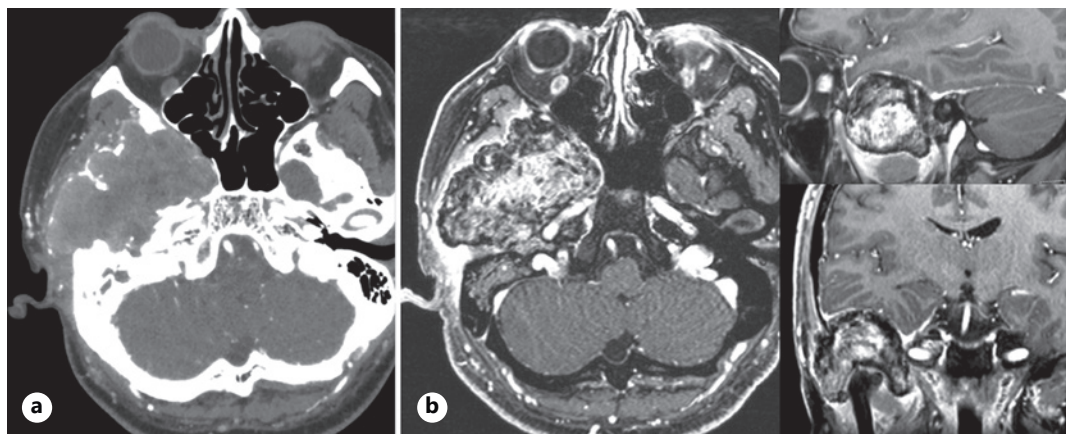


Fig. 1. Computed tomography (CT) and magnetic resonance imaging (MRI) appearances before craniotomy as the first treatment. **a** Axial: contrast-enhanced CT before treatment. The tumor originated from right temporal bone. The maximum tumor diameter was 5 cm. The tumor was largely composed of a soft tissue mass with heterogeneous contrast-enhanced component. **b** Axial, sagittal, coronal: T1-weighted MRI with gadolinium before treatment. The tumor consisted of low signal intensity in the peripheral area and heterogeneous enhancement in the central area.

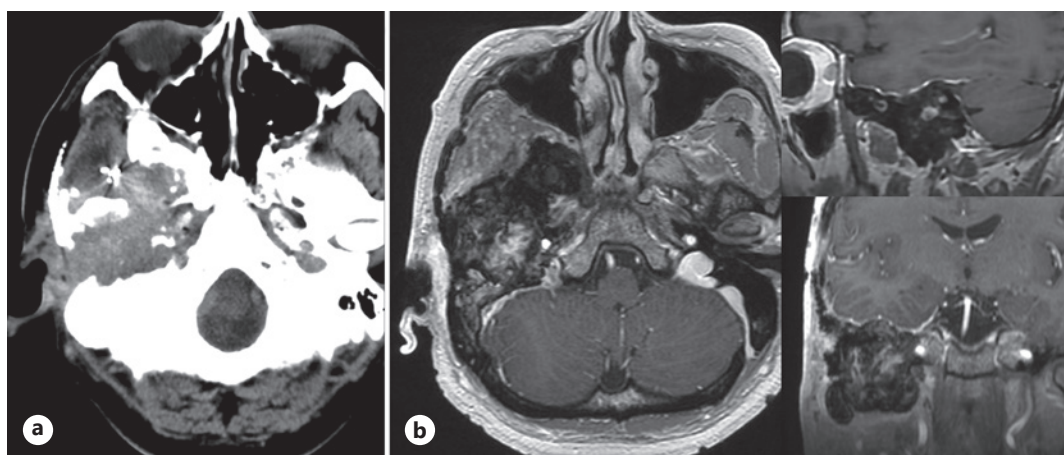


Fig. 2. Computed tomography (CT) and magnetic resonance imaging (MRI) appearances before RT using volumetric-modulated arc therapy (VMAT). **a** Axial: CT before VMAT. **b** Axial, sagittal, coronal: T1-weighted MRI with gadolinium before VMAT.

the careful follow-up without additional treatment was selected for the patient's request. Three months after the initial surgery, the follow-up MRI revealed that the tumor regrew, and the patient received surgical resection again. Again, the second resection was not able to remove the tumor completely. Fourteen months later, the 2nd recurrence was imaged. Third surgery was considered, but the patient refused it. Therefore, 36 months after initial surgery, RT was conducted. Figure 2 shows the CT and MRI appearances before RT. The tumor consisted of low signal and contrast-enhanced component on T1-weighted MRI with gadolinium.

Before RT, the head of the patient was noninvasively immobilized using a thermoplastic head mask and subjected to CT. CT images were acquired at a slice thickness of 1.25 mm and imported to the Eclipse treatment planning system (Varian Medical Systems, Palo Alto, CA, USA) for VMAT planning (Rapidarc; Varian Medical Systems, Palo Alto, CA, USA). Gross tumor volume (GTV) was based on radiological findings according to the abnormality on CT and MRI. Clinical target volume (CTV) was generated by adding a 10-mm margin from GTV up to 45 Gy, and, from 45 to 54 Gy, CTV boost was generated by adding a 3-mm margin from GTV to reduce the dose to normal brain. Planning target volume (PTV) was generated by adding a 5-mm margin from CTV. Two axial coplanar arcs were used for VMAT. The treatment plan was designed based on a TrueBeam STx linear accelerator equipped with 2.5 mm leaf-width multi-leaf collimators (Varian Medical Systems, Palo Alto, CA, USA). The plan was normalized so that PTV D50 (the dose that covers 50% of the PTV) was equal to the prescribed dose using 6-MV photon beams. PTV received a total dose of 54 Gy in 30 fractions. Organs at risk (OARs) included brain, brain stem, optic chiasm, eyes, optic nerves, and lens. Brain stem, optic chiasm, optic nerves, and lens were evaluated using planning OAR volume margin of 2 mm. Table 1 shows OAR dose constraints used for treatment plan. Figure 3 shows the treatment planning of VMAT up to 45 Gy. VMAT enabled irradiation to fit to the tumor with irregular shape at single isocenter. ExacTrac (version 6.2.1; Brainlab AG, Munich, Germany) was performed daily for patient set-up and positioning verification.

During RT, grade 2 dermatitis, grade 2 external ear pain, and grade 1 alopecia were observed (the Common Terminology Criteria for Adverse Events ver 4.0). One month after RT, grade 2 right facial nerve disorder that required steroid treatment was observed. All adverse effects improved during follow-up. There were no grade ≥ 3 toxicities. Figure 4 shows MRI appearance

Table 1. OAR dose constraints used for treatment plan

OAR	Dose constraints		This patient, Gy
Brain	D20%	<20 Gy	20.0
	D10%	<30 Gy	22.9
	D5%	<40 Gy	34.4
Brain stem	Dmax	<54 Gy	52.9
Optic chiasm	Dmax	<54 Gy	36.8
Left eye	D20%	<35 Gy	4.6
	D10%	<45 Gy	5.2
Left optic nerve	Dmax	<54 Gy	25.4
Left lens	Dmax	<10 Gy	4.3
Right eye	D20%	<35 Gy	19.6
	D10%	<45 Gy	14.5
Right optic nerve	Dmax	<54 Gy	53.0
Right lens	Dmax	<10 Gy	6.2

D20%, 10%, 5%, minimum dose to 20%, 10%, 5% of the organ; Dmax, maximum dose received by the organ.

of the giant-cell tumor of temporal bone after RT using VMAT. The tumor began to shrink from 4 months after the initiation of RT and kept shrinking slowly and gradually. Six months after the initiation of VMAT, there was a clear reduction in the size of the tumor, especially of the contrast-enhanced component. At the last follow-up (2 years after VMAT), there was no evidence of local recurrence. Denosumab, cytotoxic chemotherapy, or interferon was not conducted in this case.

Discussion

Giant-cell tumor of the skull is extremely rare. The mainstream of treatment is surgery, but complete resection is sometimes difficult. The rate of recurrence of giant-cell tumor is related to the complete resection and location [2], and no standard treatment exists other than surgery. Denosumab represented a treatment option for patients with giant-cell tumor of bone, and a previous report showed denosumab was associated with tumor responses and reduced the need for morbid surgery [6]. However, in our case, complete resection would have been impossible even if the tumor had shrunk. On the other hand, some reports described the use of RT alone or the combination with surgery. However, these treatment strategies seemed to be minority [2, 7, 8]. Fear of sarcomatous transformation and adverse effects after RT might be the reasons for the avoidance from RT [9]. Indeed, routine RT is unnecessary due to the high local control after surgery, but there might be a case in which RT is effective, such as a present case. RT may be effective in the interest of organ preservation while providing tumor control. In addition, VMAT is able to deliver highly conformal treatment, sparing nearby OARs, and ExacTrac allows more precise treatment. These advances in radiation techniques might contribute to the reduction of RT-related toxicities and improve organ preservation. However, there was no report about VMAT for giant-cell tumor of the skull, and no report revealed the radiographic details after recent radiation techniques. Therefore, this case report was meaningful in describing the details and response during and after VMAT for giant-cell tumor of temporal bone.

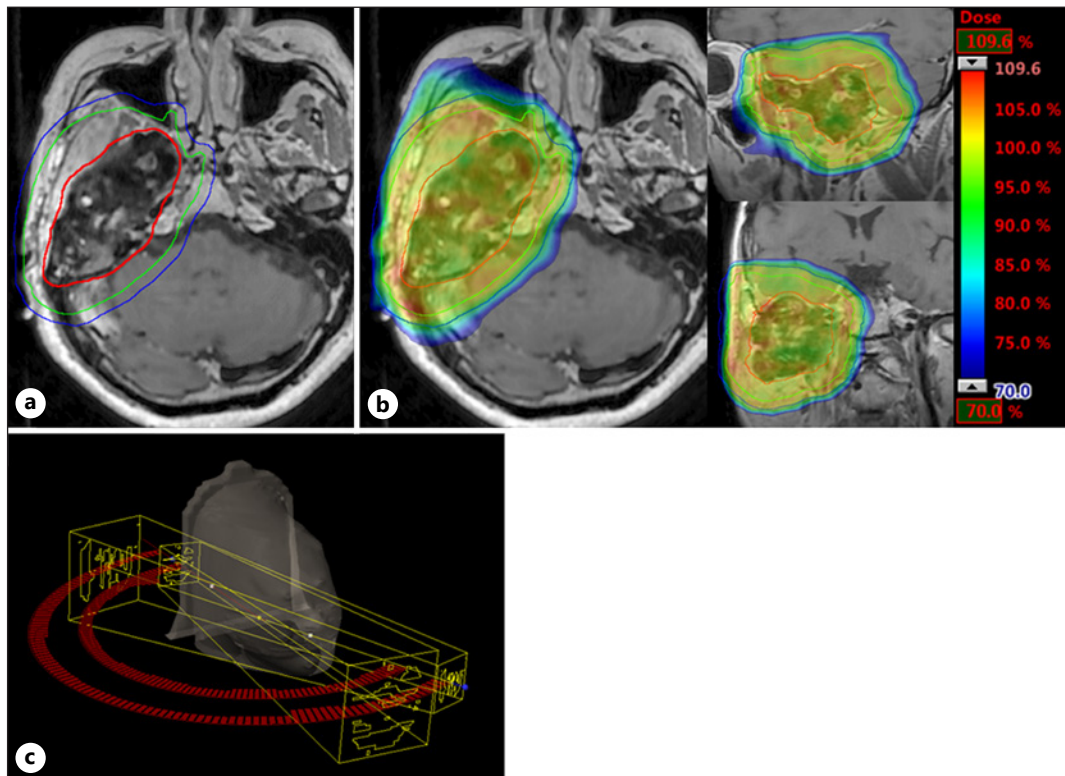


Fig. 3. Treatment plan of giant-cell tumor of temporal bone by RT using volumetric-modulated arc therapy (VMAT). **a** Axial: T1-weighted magnetic resonance imaging (MRI) with gadolinium before VMAT. GTV (red line) was defined as the abnormality on the T1-weighted MRI. **b** Axial, sagittal, coronal: dose distribution of VMAT. **c** Two axial coplanar arcs of VMAT.

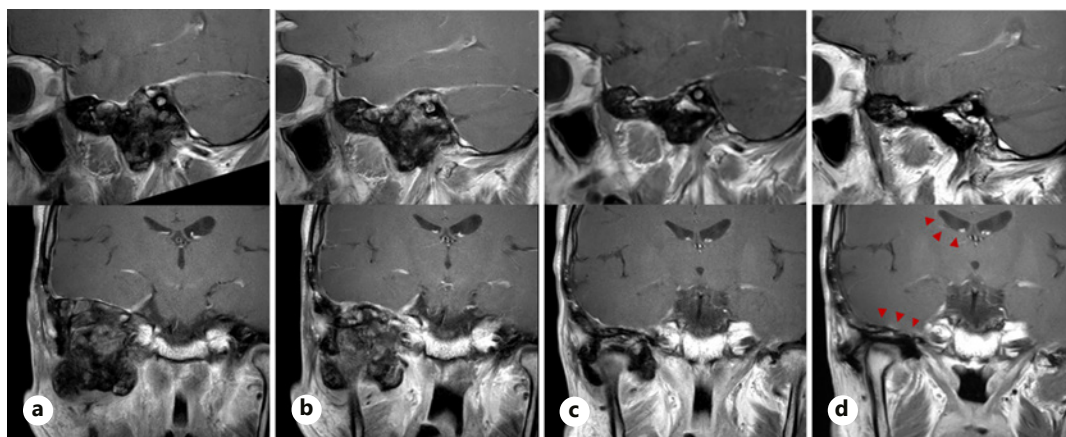


Fig. 4. Magnetic resonance imaging (MRI) appearance of the giant-cell tumor of temporal bone after RT using volumetric-modulated arc therapy (VMAT). **a** Sagittal, coronal: 2 months after the initiation of VMAT. No significant changes in the tumor were observed compared to the pre-treatment MRI. **b** Sagittal, coronal: 4 months after the initiation of VMAT. Slightly shrinkage of the tumor was observed. **c** Sagittal, coronal: 6 months after the initiation of VMAT. There was a clear reduction in the size of the tumor, especially of the contrast-enhanced lesions. **d** Sagittal, coronal: 20 months after the initiation of VMAT. An exclusion of the brain and third ventricle was reduced by the tumor shrinkage (arrow head).

The radiation dose recommendation for giant-cell tumor of the skull is unclear. According to previous reports, radiation dose ranged from 30 Gy to 76 Gy [5, 10]. An 85% local control rate was reported in patients with giant-cell tumors who were treated with more than 40 Gy [10]. In addition, Miszczyk et al. mentioned that the local tumor control was usually very high even for relatively low total doses for tumors smaller or equal to 2.5 cm in diameter, and the tendency suggested that giant-cell tumors of bone might be radiosensitive [11]. On the other hand, one series suggested the likelihood of local control seemed to be lower for patients who have undergone multiple prior therapies [5]. The higher dose may not be unreasonable for these patients with recurrent disease. However, it has been suggested that the risk of sarcomatous transformation might have been related to the higher radiation dose [5]. Therefore, we selected 54 Gy in 1.8 Gy fractions, and the irradiation volume was further reduced after 45 Gy to avoid adverse effects.

The reports of MRI appearance change after RT for giant-cell tumor of the skull are also limited [7, 8]. Tang et al. [7] reported CT and MRI appearance change before and after RT. However, in their study, follow-up images of CT and MRI were only available at 1 month after RT, and the radiographic details after RT were unclear. In our case, the tumor shrinkage was observed slowly and gradually. The appearance of the tumor did not change until 4 months after the initiation of RT and then shrank gradually (shown in Fig. 4). Giant-cell tumor of bone might have a very slow response to radiation, and contrast-enhanced component on T1-weighted MRI with gadolinium seemed to be reduced first as MRI appearance change after RT. In addition, Caudell et al. [5] mentioned the median time to development of local recurrence after RT was 11 months in their reports of giant-cell tumors. Our case has been followed up for more than 2 years, and the risk of local recurrence seems to be low. Of course, longer observation may be required to make a determination of effectiveness after RT for giant-cell tumor of the skull and to address the presence of sarcomatous transformation.

Conclusion

In conclusion, the adjuvant RT using VMAT seemed to demonstrate a sufficient local control benefit without severe adverse effects in our case with giant-cell tumor of temporal bone. However, long-term observation is needed for final conclusions.

Statement of Ethics

Ethical approval was not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Katsumaro Kubo and Masahiro Kenjo contributed equally to this work and were responsible for drafting the work. Katsumaro Kubo, Masahiro Kenjo, Hideharu Miura, Takuro Magaki, and Atsushi Tominaga were involved in the treatment of this patient. Hideo Kawabata contributed to the data collection. Masahiro Kenjo, Atsushi Tominaga, Takuro Magaki, Hideo Kawabata, Hideharu Miura, and Yasushi Nagata read and critically reviewed the report. All authors approved the final submitted version.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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