

Complete Response to Adjuvant Full Dose Photon Megavoltage Radiotherapy of Giant Cell Tumor of the Temporal Bone

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Abstract

Giant cell tumors (GCTs) of the skull bones are extremely rare and can be very aggressive. The tumor location and the frequent associated local infiltration did not allow adequate surgical treatment. However, with modern microsurgery techniques, a more complete removal of tumor may be possible. Nevertheless, postoperative radiation therapy appears to be the most effective tool in achieving local control of the tumor. We report a 30-year-old young male, treated initially for a GCT of the right temporal bone with surgery alone, which presented nine months later. A local recurrence necessitated surgery and adjuvant radiotherapy using high energy photons without mixing with protons. The patient achieved a good outcome and local control for more than 12 months of follow-up, with good cosmetic result and without significant toxicity. The aim of our work is to demonstrate that 3D conformal radiation therapy using only photons of high energy is a safe and effective adjuvant treatment of GCT of the temporal bone, and therefore equivalent to proton therapy.

Keywords: Giant cell tumor; Skull bone; Temporal bone; Surgery; Adjuvant radiotherapy; Photons; Protons; Complete response

Introduction

Giant cell tumors (GCTs) of bone are uncommon primary bone neoplasms that usually occur in the epiphysis of long

bones. When these lesions are encountered in the skull, which is an extremely rare site of development of these tumors, they are preferentially seen to involve the sphenoid, and temporal bones. GCTs are benign neoplasms but can be locally aggressive, especially with local recurrence after surgery. A tendency towards late malignant change with metastases especially to the lung has been reported. We report a clinical case of GCT of the right temporal bone in a 30-year-old young man. The patient received initial treatment based on surgery alone, with a good local control obtained during nine months. The tumor had local recurrence, with adjacent soft tissue infiltrating. The patient then received surgery and adjuvant radiotherapy. He is recurrence free for more than 15 months of follow-up.

Case Report

We report a clinical case of 30-year-old young man, with no history of disease, presenting three years ago with right otalgia associated to an ipsilateral temporal mass well visualized on the CT scan. Patient underwent surgery, and pathological examination demonstrated a tumor of myeloblastosis.



Figure 1. CT SCAN showing a process of the right temporal bone.

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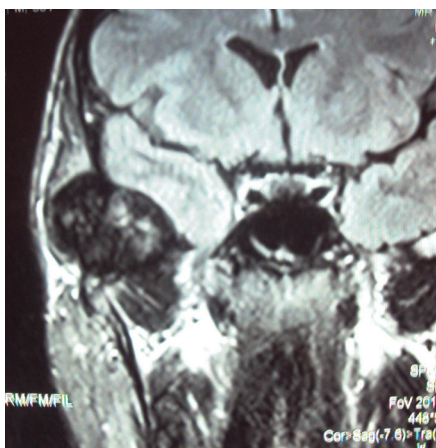


Figure 2. MRI showing a process of the right temporal bone.

After nine months of a well control, patient had local recurrence with hearing loss clinically, associated to a larger and a consistent mass in the temporal area. CT scan showed in the temporal bone, the presence of multiple lacunar images, associated to a tissue component that invades the subcutaneous soft tissues in outer part and in outside the dure-mere and cerebral parenchyma, reaching the rocher with an extent to the ossicular chain of the middle ear . The patient was lost from view for two years. A new CT scan showed an increase of the tumor size. Then, the patient had surgery; with macroscopic complete resection. Pathological examination showed a giant cell tumor, grade II of Jaffe Lichtenstein. The patient received adjuvant radiotherapy with photons 6 and 18MV, at a dose of 60 Gy (2 Gy per day, five days a week). After

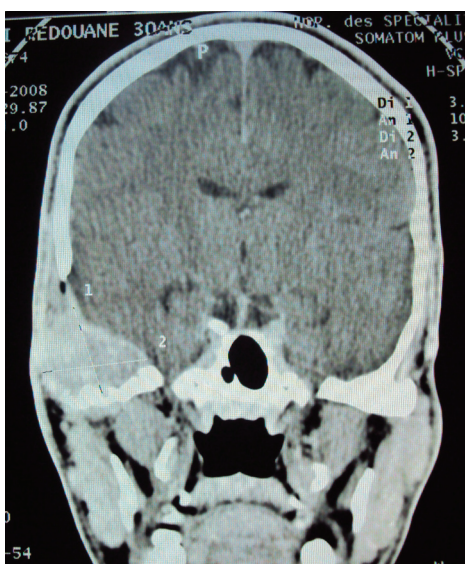


Figure 3. CT SCAN showing recurrence in the same area.



Figure 4. CT SCAN showing complete response after radiotherapy.

more than 15 months of follow-up, the patient achieved local control of complete response.

Discussion

GCT is a relatively common skeletal tumor, accounting for 4-9.5% of all primary osseous tumors and 18-23% of benign bone neoplasms [1]. Sir Astley Cooper was the first to describe a GCT in 1818. They were previously known as myeloid sarcoma, tumor of myeloplexus, osteoblastoclastoma, or osteoclastoma [2]. Approximately 2% of GCTs present in the head and neck, primarily in the sphenoid, ethmoid, or temporal bone as these sites develop from enchondral ossification [2]. Given the lack of pathognomic radiographic features, a biopsy of a solid (not cystic) component of the lesion is the only definitive way to confirm the diagnosis of GCTs [2]. When they occur in unusual sites, such as the skull, it may be difficult to make a correct preoperative diagnosis. Giant cell tumors are locally aggressive bone lesions characterized by vascularized tissue that contains numerous multinucleated giant cells dispersed through plump, spindly and/or ovoid cells [3]. Jaffe and Lichtenstein had described three levels of aggressiveness: (1) Grade I with absence of atypia and rare mitoses; (2) Grade II, where stromal cells show some atypical characteristics, but insufficient to be classified as malignant tumors, with a sub-grade II+ corresponding to potentially malignant lesions; (3) Grade III reserved for malignant tumors [4]

Otalgia, headache, hearing loss, and cranial nerve palsies are the most common presenting symptoms of temporal bone GCTs. As to ancillary diagnostic tests, skull X rays and angiography have represented the traditional

investigations for cranial osteoclastomas [5]. The advent of modern neuroimaging technology has changed the traditional scenario in this as well as generally in all cranial lesions. CT and magnetic resonance imaging (MRI) have maximally restricted the indications for diagnostic arteriography. With a well-defined, expansile bony lesion of significant hypointensity on T2-weighting, bony erosions and no abundant matrix calcifications on CT, one should consider giant cell tumor [6]. Prognosis of GCTs is mainly related to extent of surgical excision, with little contribution from radiographic and histologic grading systems [7]. Treatment options and prognosis are mainly derived from the literature on tumors in long bones [8]. Primary management was recommended by Prosser et al, using curettage for intraosseous giant-cell tumors without adjuvant treatment or filling agents, but tumors with soft tissue extension or with local recurrence require more aggressive treatment [9]. The treatment of choice is complete surgical excision which if achieved can be curative [10]. However, the skull base location of these tumors can make total surgical excision hazardous and not possible [10]. While adjuvant radiotherapy is recommended to eliminate residual tumor tissue, there is evidence that irradiation predisposes the tumor to subsequent sarcomatous degeneration [7]. Therefore, a single course of moderate dose super voltage radiation is recommended in achieving a high success rate and at the same time lowering the likelihood of malignant transformation [10]. A single modality extremity radiation therapy protocol using 40 to 60 Gy over 3 to 6 weeks yielded 85% local control rate, as determined by examination, resolution of pretreatment symptoms, and radiographic studies [11]. Doses less than 30 Gy with prolonged fractionation result in increased local recurrence and are not recommended [12]. With large doses delivered by protons (57.6 to 61.2 Gy), Hug et al reported the control of the disease in four children, all followed for more than three years [13]. The use of protons allow to best protect the healthy tissues adjacent to the tumor and deliver a higher dose which is confined to the target volume [13], but in our case we achieved the same objectives using 3D conformal radiotherapy with high energy photons, the homogeneity of the dose and the protection of healthy tissue were ensured by using wedge filters and a multileaf collimator. For unresectable tumors, radiotherapy remains the only option [14]. Chemotherapy, on the other hand, has not been largely evaluated in the skull base GCTs. Yamamoto et al reported two cases of cranial base GCT that responded well to chemotherapy on periodic CT scans with regression [15], where adriamycin was used after partial tumor excisions. Metastases occur in only 2% of cases and are usually to the lungs, but spread to rare areas like lymph nodes, mediastinum, skin, scalp and pelvis has been reported [16]. However, the rate of malignant transformation in previously irradiated lesions is between 5 and 10% [2].

Although rare, GCT can occur in the skull. Preferential

sites for GCT of the skull are the temporal and the sphenoid bone. The location and the infiltrative quality of these tumors can be extremely devastating, leaving the patient severely disabled. Radiotherapy alone or in combination with surgery is a safe and effective treatment for primary and recurrent giant cell tumors of the temporal bone. Radiotherapy should be used in their management if surgery would result in significant functional morbidity, and should be considered in this select site where the probability of recurrence is high and where there is potential for significant morbidity from relapse or subsequent surgery.

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