

Case Report

A Giant Cell Tumor of the Glenoid Fossa Extending to the Basal Skull: A 32-Year Follow-Up

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Abstract

Giant Cell Tumors (GCTs) are rare benign neoplasms that typically occur at the epiphyses of long bones in the extremities. Very rare is the occurrence of those lesions in the skull, where they show a tendency for significant bone destruction and local recurrence. We present a case of a 36-year-old woman that presented with right pre-auricular swelling. Imaging revealed destruction of the glenoid fossa with penetration of temporal bone, skull base and central cranial fossa. Histopathological analysis revealed a giant cell tumor of the bone. An incomplete resection of this lesion has been performed due to the extensiveness and location of the tumor into the skull. In contrary to literature findings which advocate additional radiotherapy after subtotal resection to prevent recurrence, a 32-year follow-up using MRI showed no volume increase nor recurrence of the lesion compared to the initial scans.

Keywords: Giant cell tumor; Glenoid fossa; Recurrence; Skull base

Introduction

A 36-year-old female was referred to the department of Oral & Maxillofacial Surgery of the Elkerliek hospital in Helmond, Netherlands in 1986. She presented with a slight swelling of the right pre-auricular area (Figure 1). Clinically a slight limitation of mouth opening was perceived, without further arguments for joint dysfunction. Neurological symptoms of recurrent headache with occasional vertigo and tinnitus were mentioned, without clinical arguments for dysfunction of the facial branches and other cranial nerves. Radiographic examination revealed destruction of the glenoid fossa and part of the zygomatic arch. Based on these findings a fine needle biopsy was carried out, which showed blood cells with histiocytes and scattered polynuclear giant cells. An additional angiogram showed a normal configuration of the external carotid artery and its branches. Computed tomography

showed, however, expansion of the tumor with penetration into the temporal bone, skull base and central cranial fossa (Figure 2).



Figure 1: Frontal and lateral view of the patient pre-operatively.

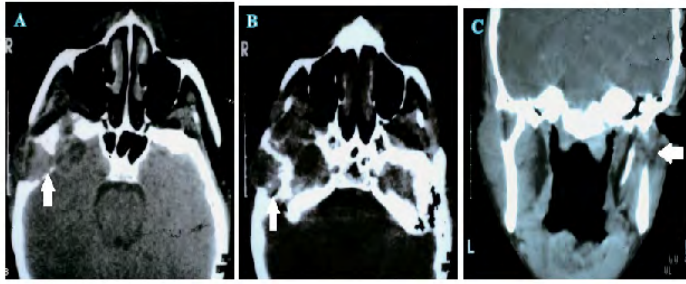


Figure 2: Axial and coronal CT view of osteolytic lesion destructing the glenoid fossa and expansion into the temporal bone, skull base and central cranial fossa.

A biopsy was performed which revealed the diagnosis giant cell tumor. Hyperparathyroidism was ruled out by the department of internal medicine. A chest X-ray was performed preoperatively without arguments of lung metastasis. Because of the location and cranial extension of the tumor, surgical intervention was decided in cooperation with the department of neurosurgery. Access was achieved by a pre-auricular approach with extension to the temporal area. The stem of the facial nerve was exposed along with the three main branches that were further dissected as deemed necessary to get sufficient access to the tumor. The tumor had also penetrated the temporalis muscle and was, thus, freed from this muscle. Further dissection towards the cranial base was continued by freeing the tumor from the dura, until firm attachment to the dura was perceived. A small perforation of the dura occurred, which was sutured. The tumor extended towards the midline of the cranial base, where it seemed to have reached the optical chiasm. At this point it was decided to stop further dissection. It was unclear if the tumor had been removed completely (Figures 3).

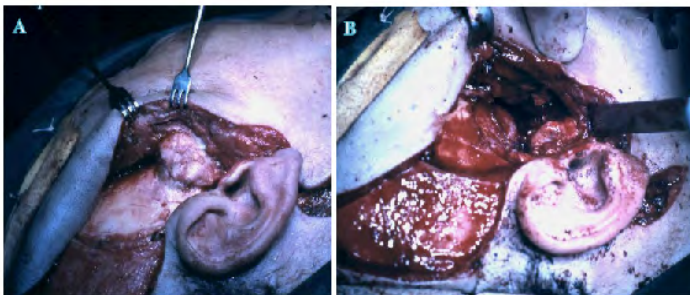


Figure 3: Perioperative images from tumor excision extending towards the cranial base.

The cranial bony defect and the glenoid fossa were subsequently reconstructed with a large cortical-cancellous, free graft from the anterior iliac crest, secured with metal wires (Figure 4). The wound was closed in layers and the patient received 2 million units of penicillin i.v, four times per day for a period of five days (Figure 5). The histo-pathological diagnosis was compatible with

giant cell tumor with only a very few mitoses. The diagnose was confirmed by the Dutch Committee of bone tumors. Immediately postoperative, a temporary partial facial nerve dysfunction of the buccal and zygomatic branch was perceived (House-Brackmann score eye 2, mouth 3) which was already recuperating during patients hospital stay for 8 days and cleared in a matter of six weeks. Further healing was uneventful.

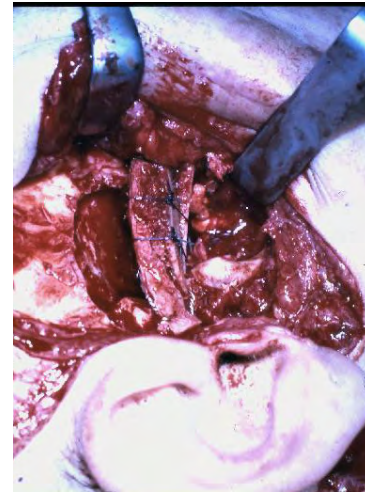


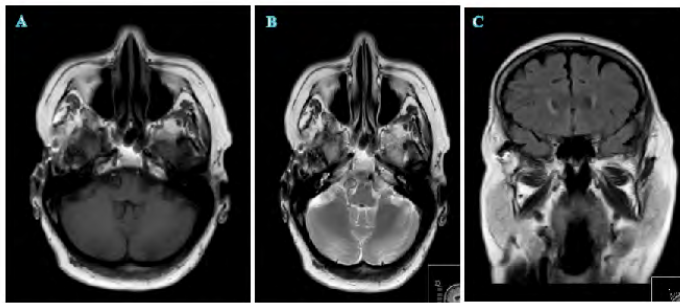
Figure 4: Reconstruction with a large cortical-cancellous, free graft from the anterior iliac crest, secured with metal wires.



Figure 5: Postoperative result after closure.

The patient presented for regular follow-up, initially every month, but after six months every three months, till 1991. Yearly follow-up chest X-ray showed no signs of pulmonary metastasis. No CT scan of the lungs was performed due to the benign nature of the tumour and the reduced accessibility of this examination at that time. Afterwards, patient had a follow-up appointment every year. During the first three years a cranial CT-scan was made twice, which did not show any further destruction. Adjuvant radiotherapy

was therefore not considered given the lack of radiological arguments for tumour remnants or recurrence as well as the risks of irradiation near the optic chiasm. As of 1992 several MRI's have been made showing for the first time tumour remnants at the skull base. Due to the absence of symptoms for the past six years, it was decided in consultation with the patient only to start additional treatment if neurological symptoms would occur. Last MRI was performed in 2018, which did not show any change as compared to the initial postoperative scans (Figure 6). The patient was last seen in 2018 and did not show any signs or symptoms of recurring disease (Figure 7). She has, thus, been free of tumor progression for 32years.



Figures 6: MRI T1 axial, T2 axial en T2 FLAIR coronal images 32 years after surgical procedure, without sign for tumor recurrence (November 2018).



Figure 7: Clinical photographs 32 years after surgery (November 2018).

Discussion

Giant Cell Tumors (GCTs) of the bone were first described by Cooper and Travers in 1818 and consist 4%-9,5% of all benign bone neoplasms [1-3]. They originate from different mesenchymal bone marrow cells and occur mostly in the epiphyses of the proximal humerus, distal radius, sacrum and distal femur, in contrary to most bony tumors that occur in the metaphysis. In less than 2% of the cases, they can be encountered in the skull, mostly in the central cranial fossa [4]. An invasion of the external auditory canal can occur on rare occasions [5].

Although benign, GCTs show a tendency for significant bone destruction and local recurrence. Occasionally metastases occur, mostly to the lungs [2,6]. It has proven to be difficult to predict the tumor's behavior due to lack of consistent correlation between the histological features and grade of malignancy of the GCT [7]. Although malignant metastasis of the GCT is possible following sarcomatous degeneration, it has not yet been described in the skull. Histology remains the gold standard for diagnosis of these lesions. However, several authors have studied new diagnostic tools that could be used to detect GCT earlier, such as biomarkers (serum phosphatase and creatinine kinase isoenzyme BB as a tumor marker) [8,9], and in the metastatic group, increased amplification of interleukin-6 and urokinase type plasminogen activator genes [10]. Although correlation of these laboratory tests with the presence of GCT is suggested, they are not pathognomonic and are not yet studied in GCTs of the cranial bones.

Due to the lack of literature regarding cranial GCTs, Freeman and colleagues (2016) conducted a literature search followed by a meta-analysis between 1982 and 2015 to describe the most adequate management of GCTs of the skull [11]. Of the 110 cases found, 67 cases remained after exclusion of the other cases due to lack of advanced CT- or MR-imaging or inadequate follow-up of at least 6 months. Average patient age was 33.7 years. The male: female ratio was roughly 1:1. The longest reported follow-up was 10 years, with a mean of 36,4 months [4,5]. In 20 patients GCT was found in the sphenoid bone, in 37 patients in the temporal bone and in 6 patients in the occipital bone. In two patients only, the tumor was found in the temporomandibular joint. Complete resection of the tumor with adjuvant radiotherapy was performed in 4 cases and complete resection without radiotherapy in 30 cases. Subtotal tumor resection with adjuvant radiotherapy treatment was found in 21 cases, and subtotal resection without radiotherapy in 10 cases [11]. Complete resection was associated with the lowest recurrence risk (3 of 34 patients - 8.8%) in comparison to subtotal resection and should be the goal of the treatment (odds ratio 0.203; 95% CI 0.033-0.937; p= 0.018). When complete resection cannot be achieved, subtotal resection in combination with adjuvant radiotherapy should be considered as an alternative. The risk of recurrence in a subtotal resection without radiation, is increased compared with subtotal resection with radiation (odds ratio 14.01,

95% CI 1,73-126,58; $p=0.0038$) [11]. However, a 32-year follow-up of our patient using MRI showed no volume increase nor recurrence of the lesion compared to the initial scans in case of subtotal resection without additional radiotherapy.

In some studies, alternative treatments like intralesional glucocorticoids, calcitonin and interferon alpha have been described [12]. Calcitonin injections and calcitonin nasal sprays have shown a fairly high success to prevent tumor recurrence. This is probably due to a higher expression of calcitonin receptors in giant cells, which when blocked, cause an influx of calcium into the bones and antagonize the function of parathormone [13-15]. However, this is based on a retrospective analysis and small sample size. A retrospective study by Nouri and colleagues (2011) rejects the benefit of using calcitonin as adjuvant therapy to prevent tumor recurrence [16]. In contrast, a double-blind clinical trial by Tabrizi and colleagues (2016) showed a significant benefit of calcitonin nasal spray as adjuvant post-surgical therapy to prevent recurrence in the jaw ($p=0,033$). In that study, however, no clear differential diagnosis was made between central giant cell granuloma and giant cell tumors [17,18].

In conclusion, in contrary to literature findings which advocate additional radiotherapy after subtotal resection to prevent recurrence, a 32-year follow-up using MRI showed no volume increase nor recurrence of the lesion compared to the initial scans. Due to lack of consensus about the management of this type of lesions in the maxillofacial region, it is important to continue reporting this entity and further investigation of alternative treatments in case of subtotal resections should be made.

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