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Giant Cell-rich Osteosarcoma in Temporal Bone: Case Report and Literature Review

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Abstract

Objective: Describe the clinical, imageological, and pathological features of Giant cell-rich osteosarcoma (GCRO), as well as differential diagnosis.

Case Presentation: We describe a 43-year-old patient with temporal bone GCRO who accidentally noticed a painless hard bump about the size of a finger in the left tempus a year ago. Imageological examination showed obvious swelling of the temporal bone, dissolving bony destruction, invasive growth, wide range of involvement, and obvious enhancement of the substantial part of the tumor (found by enhancement scanning). Pathological studies also revealed a large number of osteoclast-like giant cells in the background of osteosarcoma.

Conclusion: GCRO can be easily misdiagnosed as GCT, and early diagnosis is very important for its prognosis. Clinically and imageologically, GCRO has certain characteristics but lacks specificity; thus, clinical, imageological, and pathological diagnoses should be integrated.

Keywords: Giant cell-rich osteosarcoma (GCRO); Osteosarcoma

Introduction

Giant cell-rich osteosarcoma (GCRO) is a subtype of common osteosarcoma, which is the most common primary malignant bone tumor [1]. GCRO can be easily misdiagnosed as giant-cell tumor (GCT) in pathology and imageology, and such misdiagnosis results in the loss of the optimal opportunity for treatment [2,3]. Therefore, early diagnosis is critical to GCRO prognosis. GCRO has low incidence [4], and the nine cases of GCRO reported by Bathurst et al. [5] for the first time in 1986 accounted for 3% of the osteosarcoma incidence during that period. GCRO occurs in the long bones of limbs and is extremely rare in skulls [2,6]. In this paper, the case of a 43-year-old patient with temporal bone GCRO was reported. In addition, the clinical, imageological, and pathological features of GCRO, as well as differential diagnosis, were discussed in combination with previous studies.

Case Presentation

Clinical history

The patient was a 43-year-old male who accidentally noticed a painless hard bump about the size of a finger in the left tempus a year ago and did not seek treatment. A month ago, the bump progressively grew and was accompanied by occasional dizziness. Physical examination revealed a local uplift bump in the left tempus felt by hand. The bump had a wide base and no obvious boundary, was hard and cannot be pushed, was about 3 cm × 4 cm in size, had no surface ulceration, and was swollen. Laboratory examination revealed that the hemoglobin level was 98.3 g/l. No other abnormality was found.

Imageological examination

German Siemens Multi-slice spiral CT (Siemens Sensation 16) was used to conduct head CT plain scan, enhancement, and three-dimensional multiplanar reconstruction (MPR). The CT scanning parameters were as follows: voltage, 120 kV; electric current, 100 mAs; reconstruction layer thickness, 2 mm; and layer spacing, 0.8 mm. About 370 mg/ml Iopamiro was used as the contrast agent for enhancement scanning at a dosage of 1.5 ml/kg and injection flow rate of 3 mL/s, with flushing by 30 ml of normal saline. Image post-processing was conducted in a Siemens random

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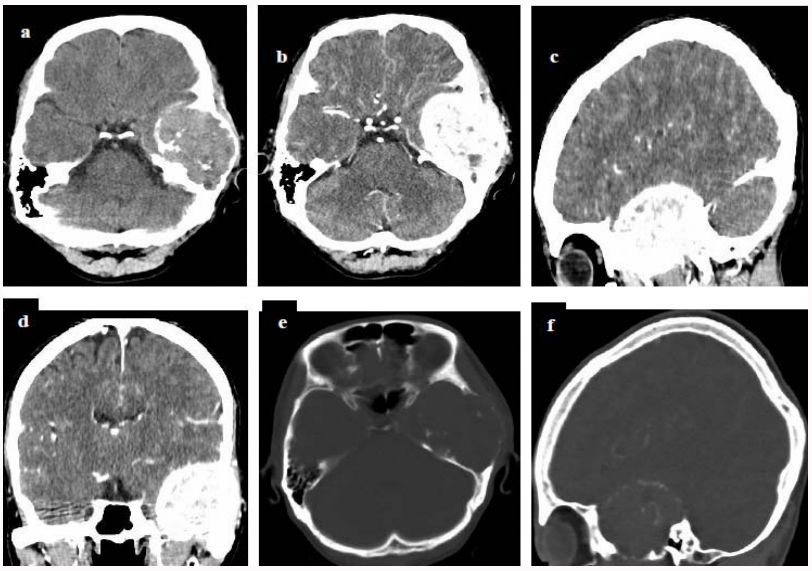

Figure 1: Plain CT and enhancement scans. Plain scan (a) shows quasi-circular bump in the left tempus, with uneven density, dotted and small patchy low-density lesions, and scattered high-density ossification. Enhancement scan (b–d) shows significant continuous and uneven enhancement of tumor. Bone window (e–f) shows swelling and osteolytic destruction of lesions, with irregular ossification and a thin, incomplete bony shell.

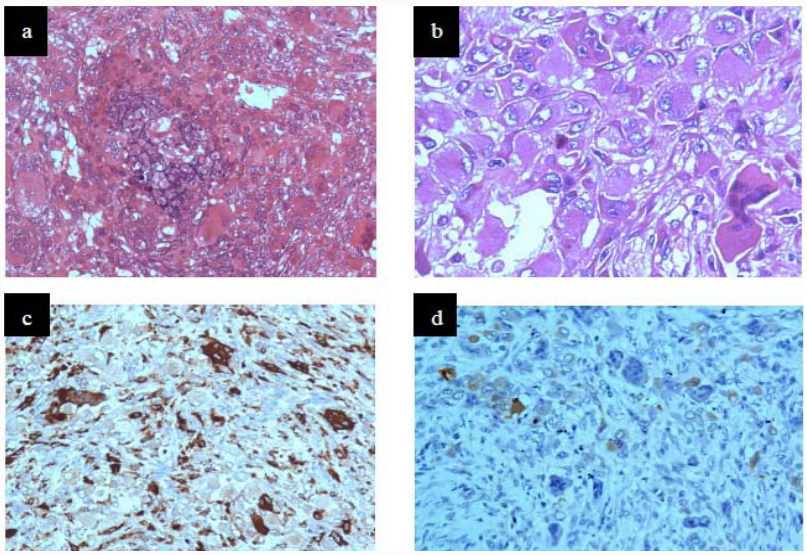

Figure 2: Pathological figure (HE staining and immunohistochemistry). (a) A large number of tumor cells, osteoclast-like giant cells, and a latticed osteoid matrix (HE staining, 200×). (b) Obvious atypia of tumor cells (HE staining, 400×). (c) Immunohistochemistry results showing multinucleated giant cells CD68 (+) (200×). (d) Immunohistochemistry results showing tumor cells S-100(+) (200×) scattered.

workstation. CT plain scan showed a quasi-round bump in the left tempus that was about 6.3 cm 4.7 cm 5.5 cm, had uneven density, was dotted, and had mottling low-density lesions. In addition, scattered dotted high-density calcification or ossification and osteolytic destruction were observed in the left temporal bone (involving the latter half of the left zygomatic arch, the left sphenoid wing, and the left mastoid and structures). The bump progressively grew and invasively crossed the inner and outer board of the skull. The bump edge showed visible thinning and an incomplete bony shell without periosteal reaction. The bump also moved inward into the middle cranial fossa, causing obvious displacement of the left temporal lobe, compression of the left lateral ventricle, and right shifting of the midline structure. Conversely, the bump moved outward causing local uplift of the left temporal soft tissue. The bump descended into the left temporal fossa along the left front part of the medial pterygoid. Enhancement scanning showed obvious continuity and uneven enhancement of the bump. The median and posterior cerebral arteries of the left brain were moved, and the anterior, median, and posterior cerebral arteries of both sides were not invaded (Figure 1). The preoperative diagnosis was malignant or low-grade malignant tumor in the left tempus, possibly a meningioma or temporal bone-sourced bone tumor.

Operation

After successful general anesthesia administration, the patient was asked to be in supine position with head to the right. The arc incision of the left temporal bone was marked, and the towel was regularly disinfected and paved. Craniotomy was conducted until the local bump of the left temporal bone was fully exposed. The left temporal bone was destroyed, becoming jelly-like and brittle. The jaw joints and the external auditory canal were eroded, but the

endocranium was not destroyed. The tumor boundary was stripped, and most of the tumor tissues were removed. The tumors around the temporomandibular joint were retained to reserve functions. After conducting thorough hemostasis and scalp suturing, an indwelling drainage tube was placed.

Pathological examination

Intraoperative inspection of grey red and grayish yellow brittle left temporal bone and tumor tissues with a size of 9 cm 8 cm 1 cm was conducted. HE staining showed diffuse infiltration of a large number of polygonal tumor cells, with large tumor cell nucleus, visible nucleoli, evident pleomorphism, osteoid matrix, and tumor osteogenesis. A large number of osteoclast-like giant cells were scattered among the tumor cells. Immunohistochemistry revealed CD68 (+) and S-100(-) of osteoclast-like giant cells, as well as S-100 (+) and CD68 (-) of mononuclear tumor cells. The pathological diagnosis was left temporal bone GCRO (Figure 2).

Follow-up visit

The patient received postoperative but not preoperative chemoradiotherapy. More than 2 years after the operation (May 2012), the patient experienced suppuration with no incentive in the left ear; accordingly, biopsy and surgery were conducted. The pathological diagnosis was recurrence of osteosarcoma, so adjuvant postoperative chemoradiotherapy was conducted. Three years later (March 20, 2013), head MRI detected tumor relapse (Figure 3), so radiotherapy and chemotherapy were conducted.

Discussion

Based on the WHO Classification of Bone Tumor Pathology and Genetics (2002), GCRO is an unusual osteosarcoma that is a subtype of normal bone osteosarcoma [1]. GCRO involves the presence of osteoclast-like giant cells with an abnormal increase in number, thereby almost disguising the osteosarcoma composition as the tumor core. GCRO can be easily misdiagnosed as GCT in pathology and imageology, thereby resulting in the loss of the optimal opportunity for treatment [2,3,7]. Therefore, early diagnosis is critical to GCRO prognosis.

According to previous reports, GCRO mostly occurs in adolescents, especially those under the age of 30. The long bones of limbs such as femur and tibia are the main pathogenic sites, and occurrences in the skull are extremely rare [2,6]. Clinically, GCRO is nonspecific and mainly characterized by local bumps and progressive pain. Laboratory tests showed increased alkaline phosphatase. Imageological examinations including X-ray, CT, and MRI showed that the main symptoms of GCRO were eccentricity (a few centricities), mild swelling, osteolytic destruction, visible tumor bone, formation of soft tissue bump in some cases, and obvious enhancement of the substantial part of tumor (found by enhancement scanning). Histologically, GCRO on HE dyeing showed a large number of osteoclast-like giant cells with a background of osseous osteosarcoma. Immunohistochemistry examination showed osteoclast-like giant cells, a small amount of histocyte-like mononuclear CD68(+) cells, abnormal mononuclear cells, and tumor CD68(-) cells [8]. We reported for the first time a case of GCRO that occurred in the temporal bone of a 43-year-old male. This age was higher than the average age reported in literature but was consistent with the age preference of GCT occurrence. Given that GCRO has a long course of disease and shows no progressive pain, no specificity was detected in the laboratory examination. This finding brought a certain challenge to the early diagnosis of this case. Imageological examination showed obvious swelling of the temporal bone, dissolving bony destruction, invasive growth, wide range of involvement, and obvious enhancement of the substantial part of the tumor (found by enhancement scanning). Given the dotted high-density lesions scattered in the tumor, we believed it was the pathological tumor bone. In this case, no obvious periosteal reaction was found. The above imageological findings were basically identical to previous reports. Pathological studies also revealed a large number of osteoclast-like giant cells in the background of osteosarcoma.

GCRO is treated the same as traditional osteosarcoma, i.e., by amputating the bone in the tumor segment and by implant surgery, supported with preoperative and postoperative radiotherapy and chemotherapy. GCRO prognosis is related to the pathological types of its background osteosarcoma. For example, if the background is centricity osteosarcoma, it is deemed to be highly malignant with poor prognosis. Conversely, if the background is periosteal osteosarcoma, it is deemed to be less malignant with better prognosis [6]. Differential diagnosis: GCRO should be mainly differentiated from conventional osteosarcoma and GCT. Key points to differentiation from conventional osteosarcoma: Imageologically, GCRO mainly manifests as osteolytic destruction, but the osteogenic tumor and periosteal reaction are often less obvious than those in conventional osteosarcoma. Pathologically, GCRO manifests as a large number of osteoclast-like giant cells in the background of osteosarcoma, whereas the latter shows no giant cells [6]. Key points to differentiation from GCT: 1) the onset age of GCRO is those of children and adolescents,

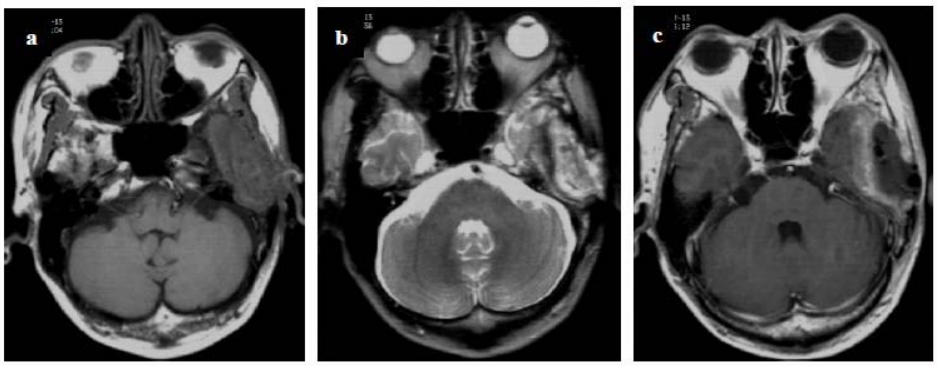

Figure 3: MRI plane and enhancement scans. TIWI scan (a) shows an oval bump shadow in the left tempus, with the center main body showing an uneven slightly low signal and an equisignal edge. T2WI (b) shows an uneven high signal in central lesions, and the signal of the edge is slightly low. T1WI enhancement (c) shows the central lesions mostly without enhancement, and the edge is irregular and obviously unevenly enhanced.

especially those under 30 years old, and is rare in those over 30 years old; GCT mostly occurs in adults over 30 years old. 2) The primary sites of GCRO are the metaphysis of long bones, whereas GCT mostly occurs in the capitulum of long bones (extremities after the epiphysis is closed, and the lesions often directly reaching under the bony articular surface). 3) Both GCRO and GCT show centrality, swelling, and osteolytic destruction, but GCRO has tumor osteogenesis and periosteal reaction, and GCT generally has neither periosteal reaction nor tumor osteogenesis. 4) Both GCRO and GCT have giant cells, but GCT has no tumor osteogenesis [8].

Conclusion

GCRO can be easily misdiagnosed as GCT, and early diagnosis is very important for its prognosis. Clinically, GCRO is mainly characterized by local bumps and progressive pain. Imageologically, GCRO is mainly characterized by osteolytic destruction, its tumor osteogenesis and periosteal reaction are often less obvious than those of conventional osteosarcoma, and it is obviously enhanced in enhancement scanning. Pathologically, GCRO is characterized by a large number of osteoclast-like giant cells in the background of osteosarcoma. Clinically and imageologically, GCRO has certain characteristics but lacks specificity; thus, clinical, imageological, and pathological diagnoses should be integrated.

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