颞骨巨细胞修复性肉芽肿影像表现与手术和病理对照

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摘　要：【目的】提高对颞骨巨细胞修复性肉芽肿(GCRG)的CT、MRI表现的认识，减少误诊。【方法】回顾性分析经病理证实的4例颞骨GCRG的CT、MRI表现，并与手术病理对照。【结果】4例均位于颞骨前、下份，侵犯范围较广。CT表现为膨胀性骨质破坏，骨性包壳不完整，病灶内及边缘见点状、条片状高密度钙化、骨化影，病灶边缘骨质均见部分增生硬化，与手术所见一致。MRI检查T_2WI信号混杂，T_1WI、T_2WI表现为大片低信号影，增强扫描不均匀强化。镜下为纤维组织增生，伴多量多核巨细胞及含铁血黄素沉积。【结论】CT和MR表现反映了颞骨GCRG骨内反复出血，肉芽组织包裹的形态和病理特点，具有一定的特征性，对该病的诊断具有重要意义。

关键词：颞骨；巨细胞修复性肉芽肿；体层摄影术，X线计算机；磁共振成像
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Imaging Analysis of Giant Cell Reparative Granuloma of Temporal Bone Compared with Operation and Pathology

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Abstract: 【Objective】To improve the understanding of CT and MRI features of giant cell reparative granuloma of temporal bone and reduce misdiagnosis. 【Methods】The CT and MR images of 4 cases of GCRG of temporal bone were analyzed, compared with their operation and pathology results. 【Results】All the lesions of the 4 cases were located in the anterior and lower parts of the temporal bone with widely destruction. The CT images showed expansive destruction of bone with disruption of osseous shell, strip and punctate calcification and ossification in and around the lesion, and osteosclerosis of the adjacent bone, which consistent with the scope of the operation. The MR images showed a large patchy of low signal intensity on both T1-weighted and T2-weighted images, which showed heterogeneous enhancement after injection of contrast. Fibrous proliferation with multiple multinuclear giant cells and hemosiderin deposition were showed under microscope. 【Conclusion】The morphological and pathological characteristics of recurrent intraosseous hemorrhage and parcels of granulation tissue in GCRG of the temporal bone could be reflected by CT and MR images, which has certain characteristics and is of important significance to the diagnosis of the tumor.

Key words: temporal bone; giant cell reparative granuloma; tomograpghy; X-ray computed; magnetic resonance imaging

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Giant cell reparative granuloma (GCRG), also known as central giant cell granuloma, repairable giant cell granuloma or central giant cell disease, is a rare, non-neoplastic, benign lesion with local invasiveness. The incidence rate was about 7%, occur mainly in the jaws, rarely in the temporal bone. In

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1974, Hirschl etc, reported the first case of GCRG in the sacrum. Most literature about GCRG were case reports [1-8]. Therefore, GCRG were often misdiagnosed as malignant tumors before surgery. This article retrospectively analyzed the CT and MRI features of 4 cases of GCRG of the temporal bone, compared with the findings of operation and pathology, and explored the pathological basis of CT signs and MR signals of the temporal bone and raised the awareness of GCRG.

1 Materials and methods

1.1 General materials

Four cases with GCRG diagnosed by surgery and pathology at the First Affiliated Hospital, Sun Yat-sen University from November 2007 to June 2017 were included in this study. Institutional review board approval was granted for this study and patients had signed informed consents. All four GCRG occurred in the right temporal squama, three cases involving the temporomandibular joint and Petrous bone. Among them, there were 3 males and 1 female, aged 23-56 years old. The main symptoms included middle ear pus, hearing loss in 1 case, swelling and pain in the temporal in 3 cases, merger of facial paralysis in 1 case, and open mouth limitation in 2 cases. Symptoms last from 1 week to more than 7 years, without local trauma or surgery history. One case was misdiagnosed as giant cell tumor, and 3 cases were misdiagnosed as malignant tumor before surgery.

1.2 Methods

In 4 cases, CT scans were performed in 3 cases before surgery, 3 cases underwent noncontrast-enhanced and contrast-enhanced MRI scans and 2 cases underwent both CT and MRI examinations.

3.0T MR imaging system (Siemens, Trio/Verio, Germany) was used for MR imaging. Scan sequence: T_1WI, T_2WI, FS-T_2WI (thickness/interval 2 ~ 6 mm/2 ~ 6 mm); Contrast medium (GD-DTPA, Beilu, China) was used according to body weight (0.2 mL/kg) for enhanced scan, sagittal 3D-T_1WI-VIBE and axial FS-T_1WI (thickness/ interval 2 ~ 6 mm) sequence (thickness and interval 0.7 mm) was obtained 20 seconds after contrast injection. Multiplanar reconstruction was done to 3D-T_1WI-VIBE sequence and axial, coronal, and sagittal images were obtained (slice thickness: 1 ~ 5 mm).

CT examination using Toshiba 64- or 320-slice spiral CT, tube voltage 120 kv, tube current 200 ~ 250 mAs, plain scan thickness/interval: 0.5 mm/0.5 mm, enhanced scan was obtained 60 ~ 80 s after contrast injection. Contrast agent (iopromide300, Bayer Schering) was used by weight of 1.5 mL/kg via cubital vein.

The lesion size, morphology, bone destruction and extent, lesion density and signal intensity, and the extent of lesions were observed, compared with surgery and pathology, and their signal-pathological characteristics were analyzed.

2 Results

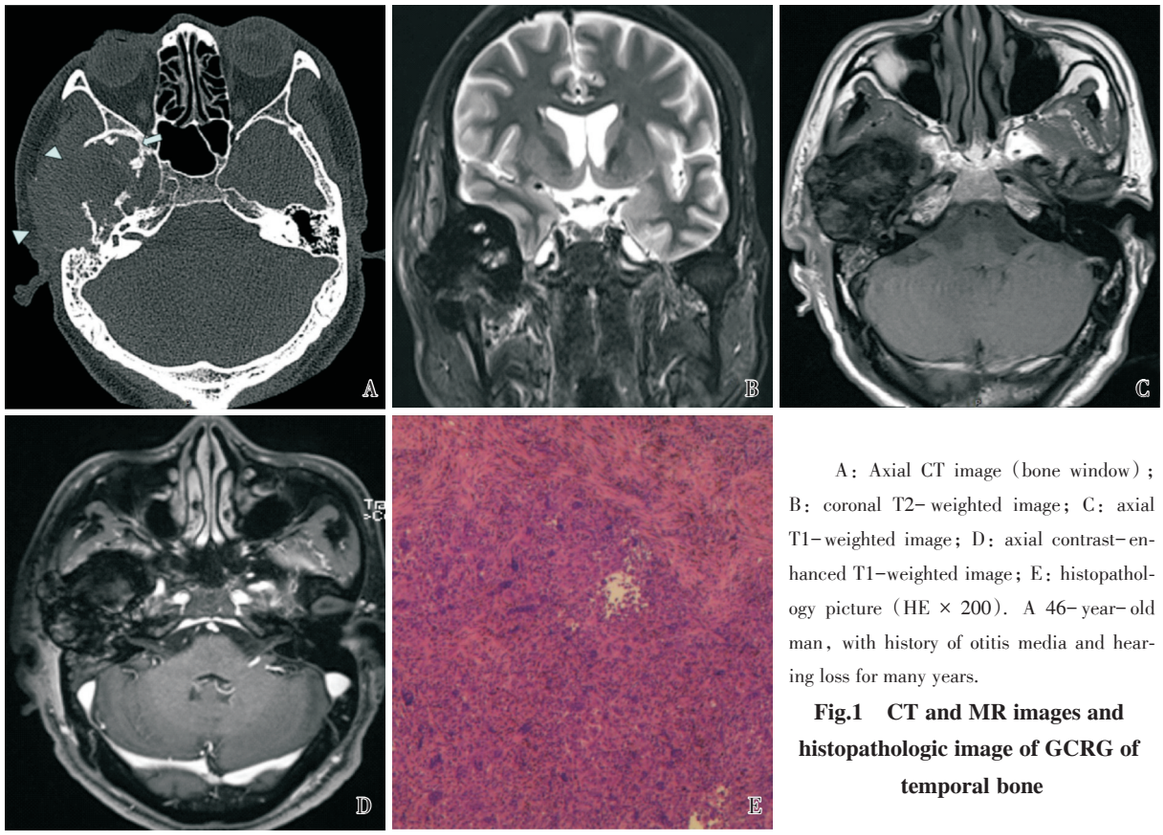
2.1 Lesion shape and size

The lesions were in the form of masses with mild lobulated, and the size were approximately 28 mm× 19 mm ×26 mm to 63 mm×41 mm×70 mm.

2.2 Bone destruction of the lesions

The range of bone destruction: the center of 4 lesions located on the anterior inferior part of the temporal squama, 3 invaded the petrous bone, 2 invaded the anterior portion of the mastoid, 1 invaded the anterior portion of the tympanum and sphenoid bone, 3 involved the temporomandibular joint and surrounded the mandibular condyle, but only 1 case invaded the mandibular condyle, and 1 case destroyed the head of the malleus.

The form of bone destruction: The bone window of CT images shows expansive destruction of bone with disruption of osseous shell, osteosclerosis of the adjacent bone, strip and punctate calcification and ossification in and around the lesion in 2 cases (Fig 1A). The lesions of 2 cases invaded and exceeded the bone cortex of the temporal squama and invaded the temporal muscle.



A: Axial CT image (bone window); B: coronal T2-weighted image; C: axial T1-weighted image; D: axial contrast-enhanced T1-weighted image; E: histopathology picture (HE × 200). A 46-year-old man, with history of otitis media and hearing loss for many years.

Fig.1　CT and MR images and histopathologic image of GCRG of temporal bone

2.3　The density and signal intensity of the lesions

Of the 4 cases, one lesion manifested as a solid mass, 3 lesions showed cystic and solid masses with central cystic degeneration.

Lesions showed mainly isodensity on CT plain scan images, with cystic or patchy low density foci in 3 cases, which were heterogeneous enhanced after contrast injection. The low density foci showed no enhancement.

On MR images, lesions were heterogeneous signal intensity on both T1-weighted and T2-weighted images. 3 cases showed low signal intensity mainly (compared to the gray matter) on T1-weighted and T2-weighted images (Fig. 1B, 1C), with patchy isointensity in the center of the lesion on T1-weighted images and large irregular extremely low signal intensity on T2-weighted images. In the center of the masses, scattered small cystic or patchy high signal intensity were seen on T2-weighted images (Fig. 1B), and fluid-fluid level was seen in one case (Fig.2). Lesions showed uneven patchy or nodular enhancement after contrast enhanced (Fig.1B), with enhancement of the adjacent dura. In the lesions, the regions that showed high signal intensity on T2-weighted images were not enhanced after contrast injection.

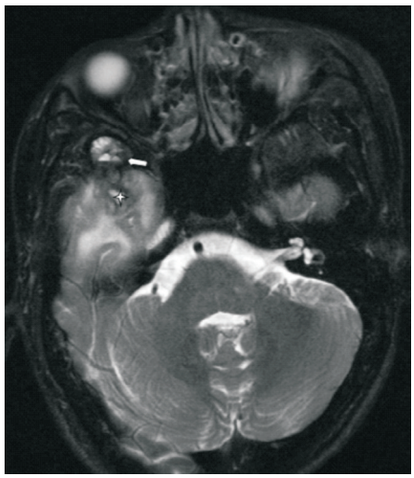
2.4　Operative findings

2 case underwent radical resection; another 2 cases underwent biopsy. The temporal squama, the middle ear tympanic cavity, and the temporomandibular joint fossa destruction with partial osteosclerosis was seen in surgery, which was consistent with the image findings. The gross appearance of the masses were dark-red. In 2 cases, brownish old blood flowed out when the surface was incised. The boundary of the masses were unclear and the deep part of the temporal muscles were invaded. 1 case involved the dura and the temporal lobe.

2.5　Pathology and immunohistochemistry

All 4 cases were diagnosed as giant cell reparative granuloma. The tumors were gray-red, gray-yel-

low and soft in macropathology. Polygonal cells and short spindle cells were the main components of the tumor, with many fibrous stroma nodular distribution under microscopic, which showed iso-intensity on T1-weighted images and T2-weighted images. Multinucleated giant cells and hemosiderin deposition (Fig.1E) (corresponding to the low signal portion on T_2WI and T_1WI) were seen, with inflammatory cell and scattered necrotic areas (corresponding to small cystic changes seen on CT and MR images). 2 cases showed focal calcification or reactive osteogenesis (corresponding to the spotty or striped high-density on CT images). There were no nuclear abnormalities and rare mitoses in the cells.



Axial T2-weighted image. A 46-year-old man, with history of temple pain for 20 days. T2-weighted image showed a mixed signal intensity mass with capsule, fluid-fluid level (white arrow) and temporal lobe invasion.

Fig.2 Fluid-fluid level and temporal lobe invasion of GCRG of temporal bone

Immunohistochemistry showed that Ki67 was 3% ~ 10% positive in 3 cases, Vimentin positive in 2 cases, P63, CD68, CD163 partial cells, actin, CD31, and CD34 were positive in 1 case.

3 Discussion

3.1 Overview

GCRG is a non-neoplastic, granulomatous lesion and was the body's repair response to bone bleeding or trauma and inflammation. The GCRG often occurs in the jawbone, rarely in the temporal bone. Its mechanism is unknown. It is considered to be a giant cell repair reaction caused by trauma, surgery, and infection[3-4.8-9], however, there were quite a few reported cases with no history of trauma, operation or infection[4-5, 9]. In this study group, 3 cases had no local trauma surgery, only 1 case had a history of otitis media. Some studies reported that young female GCRG of the jaw was more commom[10], GCRG of the temporal bone had no age and sex tendency[3, 5, 11], while young male of this group was more common (male / female = 3/1) and the age was 23 ~ 56 years old. The clinical manifestations were not special, and most of the cases were misdiagnosed before operation.

3.2 Imaging Analysis compared with operation and pathology

In this group, all of the 4 cases of temporal bone GCRG were located in the anterior lower portion of the temporal bone. Imaging findings and surgical findings showed a wide range of GCRG bone destruction in the temporal bone, extensive destruction of the temporal squama, the drums (including the temporomandibular fossa), the mastoid and petrous bone, and even the sphenoid bone. One of the cases invaded the temporal lobe, and there were few reports of in the literature[11].

The forms of bone destruction of the temporal bone GCRG consistented with the literature reports[2, 3, 5] were complex. Compared with the operation, the main manifestation of CT images was expansive destruction of bone with disruption of osseous shell, strip and punctate calcification and ossification in and around the lesion, which were similar to the invasive and malignant bone tumor. This type of GCRG complex bone destruction was also an important cause of misdiagnosis of malignant tumors before surgery. This was related to the dual nature of benign and invasive of the GCRG. Although GCRG is defined as a benign lesion, it is generally considered to be invasive[3]. Pathological lesions were rich in fi-

brous matrix, fibroblasts, giant cells, and scattered lymph nodes[2, 12-13]. The cells repaired the injuries caused by surgery, trauma, inflammation, and other unknown factors, but at the same time, macrophages´ osteoclast function and fibroblasts and fibrin matrix produced by continuous stimulation will also "erode" bone, which caused osteolytic destruction and even the bone cortex destruction. The presence of lesions stimulates osteoblasts to attempt to repair the osteolytic area through hyperplasia, forming the hardened edge of the lesion and strip and punctate high density seen by image and surgery, and new bone was seen by pathology[12, 13], but due to the speed of osteogenesis can not keep up with the speed of osteolysis, and the speed of osteolysis of the lesion is not as rapid as malignant, so spot-like high-density in and around the lesion were still visible, eventually leading to the formation of osteolysis and osteogenesis at the same time, but mainly osteolysis.

The MR signal intensity of the temporal GCRG had certain characteristics. 4 case were similar to those reported in the literature[3, 5, 11, 14-15], low signal intensity area on T1 and T2 weighted images of the mass were large, especially on T2 weighted image, large low signal was characteristics. These large areas of low signal were the old brown bleeding seen by surgery, according to image and surgical findings, we can confirm that under pathological examination, there were massive hemorrhage and hemosiderin deposition. The area of scattered patchy equal signal on T1W2 in the mass with enhancement was the solid tumor components by surgery, pathologically related to stroma hyperplasia and infiltration of giant cells and inflammatory cells. The soft tissue components of these fibrous stroma and inflammatory cells were often overshadowed by low signal caused by old blood and hemosiderin with shortened T2 effect, so the lesion was often expressed as a large number of very low signals on T2W2. The central area of the mass showed high signal on T2WI was cystic necrosis area without enhancement, suggesting scattered necrotic areas on pathology. There was a case of a liquid-liquid level found in this group, which was only a few case outside maxilla and mandible reported in previous studies[14], presumed to be caused by repeated old and new hemorrhagic and cyst changes in the same area.

In 4 cases, there were no nuclear abnormalities and rare mitoses in the cells, suggesting that the tumor cell morphology had no malignant signs.

Therefore, the CT signs of invasive bone destruction of the temporal bone and the signal characteristics of large low-signal area on T2WI reflected the morphological and pathological features of the temporal bone GCRG, supporting the multiple lesions in the temporal bone, which is surrounded by granulation tissue, and caused by a repair reaction, is a relatively characteristic feature of the temporal bone GCRG, which is of great significance in the diagnosis of the disease.

3.3 Differential diagnosis

The temporal bone GCRG needs to be differentiated from giant cell tumor of bone, aneurysmal bone cyst and malignant tumor of temporal bone. Giant cell tumor occurs predominantly in the age of 20～40 years, the extremities of the long bone. The CT showed expansive bone destruction with intact bony shell. The edge of the bone destruction was clear and sharp without hardening; the tumor was dominated by slightly high signal intensity on T2WI and significantly enhanced after contrast injection, which was different from the GCRG of the temporal. Aneurysmal bone cyst also showed expansive bone destruction. Sclerosing edge was visible on the dege of bone destruction. There was a large number of blood sinus in the tumor, which showed high and low signals of liquid-liquid level on T2WI. The cystic change was obvious. Malignant tumors of the temporal bone, with different pathological types, most show extensive invasive or wormy-like bone destruction, with mixed mixed signal intensity on T1WI and T2WI, unlike GCRG. The final diagnosis depends on the pathology.

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