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CASE REPORT

Chondroblastoma of the Temporal Bone Removed by Utilizing a Replicated Model as a Navigator

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Purpose: To present a clinical features and surgical approach of a temporal bone chondroblastoma.

Materials and methods: A 42-year-old woman was diagnosed with temporal bone chondroblastoma. Her chief complaints were earache and hearing loss. A three dimensional model was replicated using rapid-prototyping method for the surgical navigation.

Result: At surgery, the tumor was found to extend from the temporomandibular fossa to the zygomatic base, squamous part and middle cranial fossa. The model allowed easy identification of the involved ossicles. The tumor was safely removed and the patient has been free from recurrence for 23 months.

Conclusions: The rapid-prototyped model was useful for safe and secure surgery for a lesion involving anatomically challenging region.

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Chondroblastoma is a rare tumor which constitutes 1 % of the primary bone tumors [1]. It most frequently arises from the epiphysis of the long bone. Involvement of the temporal bone is rare. Complete removal is a challenge due to anatomical complexity of the involved area including the middle ear and skull base. We encountered a case of temporal bone chondroblastoma that was safely removed by using a 3 dimensional replicated model as a navigator.

Case Report

42 year- old female, house wife This woman had left earache in the beginning of May 2006 after catching the common cold. She visited a local otolaryngologist in May 10. Medical treatment reduced the earache, but she started to notice left hearing loss. She was referred to our clinic on 12 May 2006. Her past and family history was negative.

Local and test findings

The left ear drum was thick and brownish in color. The upper wall of the ear canal was swollen. The right ear was normal. Other local findings were negative. The audiogram revealed mixed type deafness with averaged threshold of 55 dB. The tympanogram was normal. The Schueller x-ray showed a round translucent shadow extending superiorly and anteriorly from the ear canal and the temporomandibular area (Figure 1). The CT showed a round tumor shadow eroding the squamous part of the temporal bone, temporomandibular fossa, zygomatic base and extending to the middle cranial fossa (Figure 2). In the MRI, the tumor with T1 low and T2 high intensity compressing the dura mater was recognized. It was gadolinium-enhanced heterogenously (Figure 3).

The open biopsy was performed in June 23, 2006. The diagnosis was a tumor arising from the bony tissue, most suggestive of a giant cell tumor. Surgery was planned. Because of the wide extension, a three dimensional (3-D) model was replicated using the rapid-prototyping method. This is to create a 3-D model based on the CT scan DICOM data which were converted into the STL file. The powder material, a

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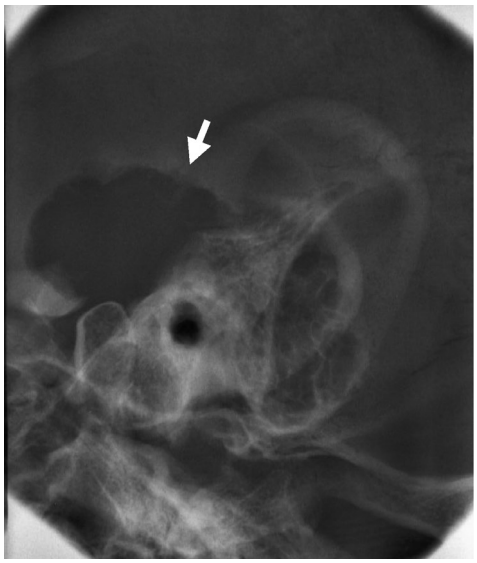

Figure 1. Schueller x-ray. A translucent round shadow in the squamous part is seen (arrow).

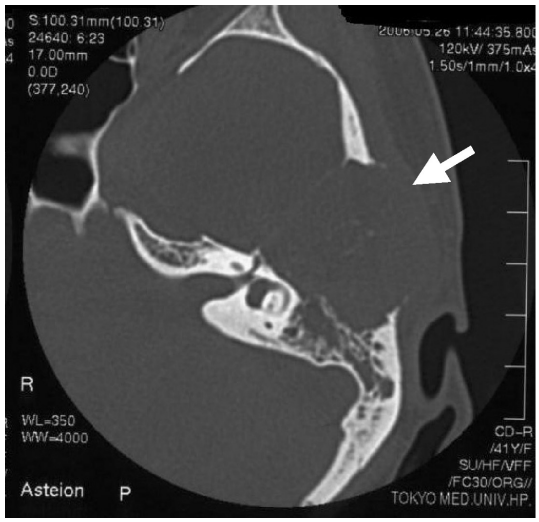

Figure 2. CT findings. A round tumor shadow eroding the squamous part of the temporal bone, temporomandibular fossa, zygomatic base and extending to the middle cranial fossa is shown (arrow).

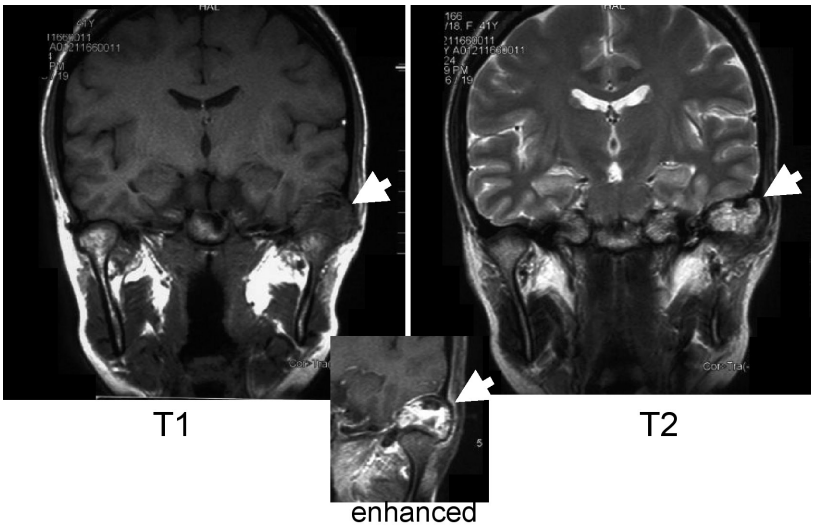

Figure 3. MRI findings. The tumor with T1 low and T2 high intensity compressing the dura mater is recognized. Gadolinium-enhance image is presented in the center.

polyamide nylon plus glass bead was laser sintered according to the STL protocol to create the model. The details of model replication had been reported previously [2, 3].

The model clearly showed a large bony defect involving the squamous part, temporomandibular fossa, zygomatic base and infratemporal fossa (Figure 4). The malleus head was missing, but the incus was identified. The horizontal section of the facial canal appeared intact.

Surgical findings

Surgery was performed under general anesthesia on August 21, 2006 with collaboration of a neurosurgeon. The inverted U shaped incision was placed on the supraauricular region. The temporal fascia and muscle flaps were preserved. A part of the tumor was exposed in the anterior region of the ear canal. Because of difficulty in orienting the ossicles from temporal route, the disinfected 3-D model was used as a navigator. The eroded part of the epitympanic tegmen was easily

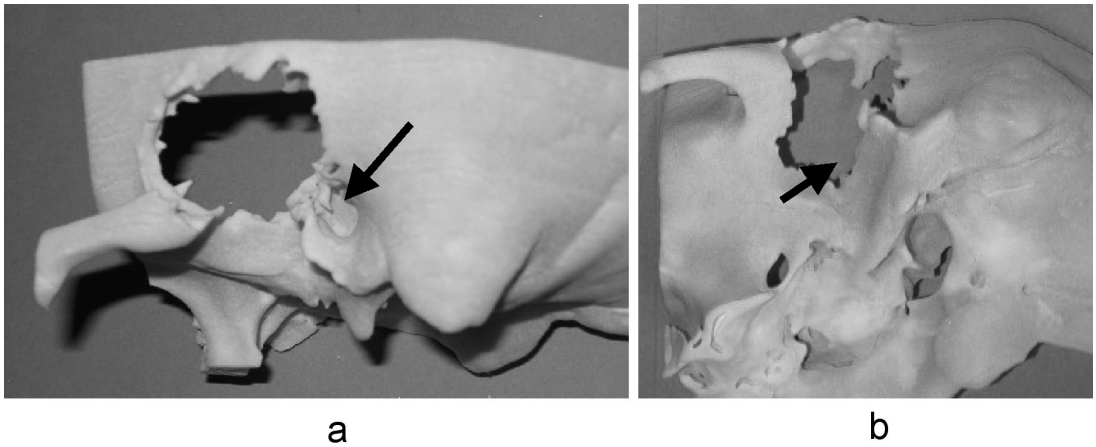

Figure 4. Three-D prototyped model. Lateral view (a) shows a large bone defect. The arrow is external ear canal. Downside view (b) shows a defect of the infratemporal fossa (arrow).

identified by scrutinizing the corresponding part of the model taken in surgeon's hand. The malleus and incus could safely be found by following down the eroded bony edge. The malleus head was missing as was shown in the model (Figure 5). The incus and the stapes were normal. The tumor did not involve the facial nerve. The chorda tympani nerve was sacrificed. A part of the tumor extended to the temporomandibular fossa and zygomatic base was removed en bloc. The mandibular joint capsule was preserved. The tumor adhering to the dura mater had to be removed in a piece-meal fashion. There was no defect in the dura mater. The surface of the dura mater to which the tumor attached was cauterized. The removed incus was used as a columella. The surgical defect was filled with pedicled temporalis fascia. The cranial bone defect was bridged by titanium mesh which was covered by the temporalis muscle. Total bleeding volume was 271 ml.

Pathological findings

The specimen showed a characteristic chicken wire calcification, multinuclear giant cells and cartilaginous cells (Figure 6). S-100 protein was positive. The diagnosis of chondroblastoma was established.

Postoperative course

The postoperative course was uneventful. The patient has been free from facial palsy and mastication disorder. However, a conductive deafness of 45 dB in average remained. No recurrence has been noticed for 23 months.

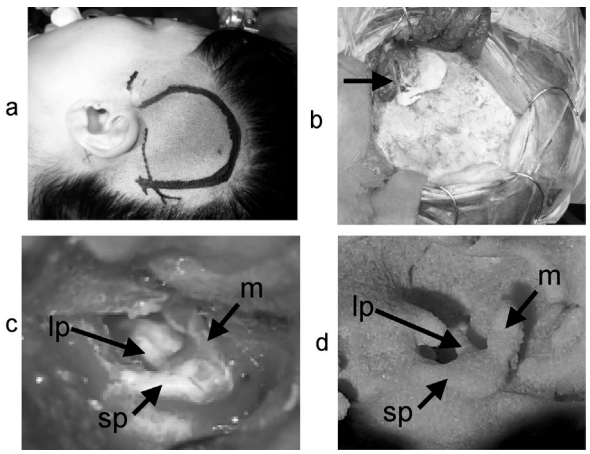

Figure 5. Surgical findings. Supraauricular incision (a). The tumor is recognized beneath the incision (b, arrow). Findings of the ossicles (c) are shown along with those of the model (d) for comparison. Malleus head is missing. Incus is intact.
m: malleus head, lp: incus long process, sp: incus short process

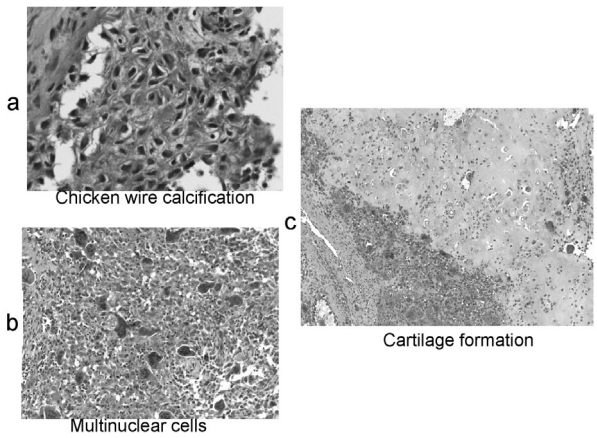

Figure 6. Pathological findings. Characteristic chicken wire calcification (a), multinuclear cells (b) and cartilage formation are shown (H-E staining).

Discussion

Chondroblastoma is a rare tumor that constitutes about 1 % of primary bone tumors [1] and usually occurs in the epiphysis of long bone. The temporal bone involvement is even rarer, comprising only 1-7 % of all chondroblastoma [4]. Approximately 50 cases had been reported in the English literature. The tumor is believed to arise from a cartilaginous remnant along the bone suture, such as petrosquamous suture [5]. The frequent sites of lesions are infratemporal fossa, temporomandibular fossa, squamous part and superior aspect of the ear canal [6, 7]. The present case involved all these areas, indicating a typical extension as a temporal bone chondroblastoma.

Chondroblastoma of the temporal bone prevails in the age of 30 to 40 years [7, 8] and male to female ratio is 2.3 to 1 [9]. The initial symptoms include local swelling, ear pain, hearing loss and tinnitus. The present case had all these symptoms.

Imagings show findings characteristic of chondroblastoma. Chondroblastoma typically shows osteolytic round lesion in CT with patchy calcification [10]. MRI shows hypo to isointense on T1 and iso or hyperintense on T2 [5, 10]. It is also heterogeneously enhanced. Differential diagnosis includes chondrosarcoma, giant cell tumor, aneurysmal bone cyst, chondromixoid fibroma and others. Chondrosarcoma may show lower signal in both T1 and T2 [6]. Aneurysmal bone cyst shows higher signal on T1 and high T2 depending on the degree of bleeding in the mass [6]. Final diagnosis is established by pathological findings. Positivity of S-100 protein gives high suspicion of chondroblastoma [6, 10].

For the treatment, total removal is the best option. Curettage leaves a high chance of recurrence. In the present case, a 3-D model was replicated for surgical planning, because of the large size and possible involvement of the ossicles. We had already created models for difficult cases such as congenital aural atresia and advanced cholesteatoma, and reported its usefulness for surgical simulation [2, 3]. Particularly, when the disinfected model is used as a navigator at surgical field, the surgery is safe and easy [3]. In the present case, the ossicles have been found safely by scrutinizing the model in surgeon's hand. However, a part of the tumor that attached to the dura had to be removed piece by piece. Possibility of recurrence increases when en bloc removal is not performed. For the present case, long term follow-up is mandatory.

Conclusion

We encountered a case of temporal bone chondroblastoma that arose in 42 year-old female. A three dimensional model was replicated using rapid-prototyping method for the surgical navigation. At surgery, the tumor was found to extend from the temporomandibular fossa to the zygomatic base, squamous part and middle cranial fossa. The model allowed easy identification of the involved ossicles. The rapid-prototyped model was useful for safe and secure surgery for a lesion involving anatomically challenging region.

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