Giant cell tumor of the patella with a secondary aneurysmal bone cyst: A case report

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Abstract. The substance of the patella is an uncommon location for tumor occurrence and development. The present study reports a case of giant cell tumor (GCT) of the patella, combined with an aneurysmal bone cyst (ABC). To the best of our knowledge, this is the second report of GCT with ABC published in English. GCT is the most common type of benign tumor. Secondary ABC is frequently associated with GCT, but this symbiotic tumor rarely occurs in the patella. A 27-year-old male patient was examined at the outpatient clinic, and clinico-pathological characteristics of the tumor were observed. X-ray and computed tomography (CT) scans revealed a lytic lesion located in the center of the right patella. Curettage, followed by autogenic and allograft bone grafting, was performed. Histopathologically, the lesion was diagnosed as a GCT with secondary ABC. No recurrence or metastasis was identified during the 1-year follow-up period. The present study reports a case of GCT with secondary ABC, and discusses the rare location and histopathological type of this tumor, in order to improve diagnosis and treatment of patellar tumors in general.

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Abbreviations: GCT, giant cell tumor; ABC, aneurysmal bone cyst; CT, computed tomography; MRI, magnetic resonance imaging

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Introduction

Tumors developing in the patella have a considerably rare occurrence, with the vast majority of them being giant cell tumors (GCTs) and chondroblastomas (1). Although the diagnosis and treatment of these histologies are typically straightforward, only a few patellar primary tumors have been reported to date (1). GCTs account for 33% of all patellar tumors, while aneurysmal bone cyst (ABC) accounts for 5% of all patellar tumors (1). Knee pain and/or swelling are the most common symptoms of these two patellar tumors (1). To the best of our knowledge, the occurrence of patellar symbiotic tumors is considerably more rare. GCT combined with ABC accounts for 14% of all GCTs (2), and has also been reported in other bone locations, such as the rib (3), calcaneus (4), talus (5), spine (6) and radius (7). Only Marudanayagam and Gnanadoss (8) have reported patellar symbiotic tumors thus far. Imaging data is helpful for the diagnosis of GCTs and ABCs and surgery is the main treatment used for the two tumors. More studies are required to raise awareness of this special type of tumor and to gain diagnostic and treatment experience. In the present study, a case of GCT of the patella with a secondary ABC is reported. Written informed consent was obtained from the patient for the publication of the present study.

Case report

On 10th March, 2014, a 27-year-old male patient visited the outpatient clinic of the Department of Orthopedics at The First Affiliated Hospital of Dalian Medical University (Dalian, China) complaining of right patellar pain, swelling and limited mobility for 10 days subsequent to an unexpected fall. The patient had no history of weight loss or exposure to tuberculosis.

Upon physical examination, swelling and localized tenderness was detected by palpation in the front aspect of the mid-patella. The right knee of the patient exhibited a decreased range of motion and severe pain upon reaching maximal knee flexion. The float and grinding tests of the whirlbone were positive. However, there was no evidence of a soft-tissue mass

and overlying skin lesion. Furthermore, no joint effusion or synovial thickening was noted.

Direct radiographs (Figs. 1 and 2) and computed tomography (CT) scans (SOMATOM Definition; Siemens Healthcare, Forchheim, Germany) of the knee were performed. Radiographic examination revealed a well-defined lytic lesion with a thin cortex occupying 2/3 of the patella, and no pathologic fracture or periosteal reaction was observed. Bone tracer scanning (Infinia Hawkeye 4; GE Healthcare Life Sciences, Pittsburgh, PA, USA) with ^{99m}Tc-methylene diphosphonate (Jiangsu Institute of Nuclear Medicine, Wuxi, China) revealed a moderate tracer uptake in the right patella. CT scan of the chest revealed no pulmonary metastasis.

Following laboratory tests, only the levels of C-reactive protein were observed to be slightly increased, at 13.5 mg/l (normal range, 0-10 mg/l). All other test results were within the normal ranges.

Intraoperatively, no abnormality was noticed in the soft tissue around the patella. The lesion was cystic and cavitary, and contained granulation tissue in addition to 2 ml light bloody fluids, which were discharged from the cavity. Subsequently, the patient underwent curettage of the lesion using a high-speed burr (Stryker, Mahwah, NJ, USA) through a 3x2-cm^2 window performed on the medial aspect of the patella. The material was sent for formal histopathological examination. Following massive saline irrigation, the cavities in the patella were filled with autogenic iliac bone and allograft bone graft. Histopathological findings revealed features of GCT with ABC. Macroscopically, the resected tumor tissue was grey, red and white; no necrosis was observed. An ABC component was found, with clotted blood filling the cystic cavities. Microscopic analysis (Leica DM-2500; Leica Microsystems, Wetzlar, Germany) (Fig. 3) revealed typical characteristics of benign GCT, including polygonal or cuboidal tumor cells, mitosis, thickened nuclear membranes and multinucleated giant cells. The resected margin was tumor-free. Immunohistochemistry results indicated that the tumor cells were partly positive for P63 (monoclonal mouse anti-human p63; #sc-8431; dilution, 1:1000), and negative for P53 (monoclonal mouse IgG_{2a} anti-human p53; #sc-126; dilution, 1:500) and cluster of differentiation 68 (monoclonal mouse IgG_1 anti-human CD68; #sc-20060; dilution, 1:100) (all antibodies from Santa Cruz Biotechnology, Inc., Santa Cruz, CA, USA).

Following surgery, the patient visited the outpatient clinic on a regular basis. Clinical and radiological examinations, including palpation and plain radiography, were performed (Fig. 4). No local recurrence or distant metastasis were identified 12 months following surgery. The patient had good functional outcome and obtained a full range of motion on the right knee.

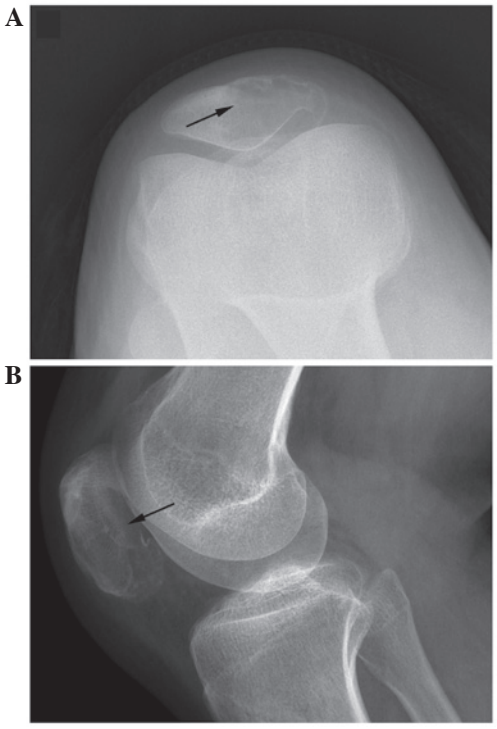

Figure 1. Preoperative plain radiograph. (A) Enlargement of the cystic lesion in the patella was visible on the axial view. (B) A bone translucency with peripheral rim change is observed at the lateral bottom part of the patella. Arrows indicate the lesion.

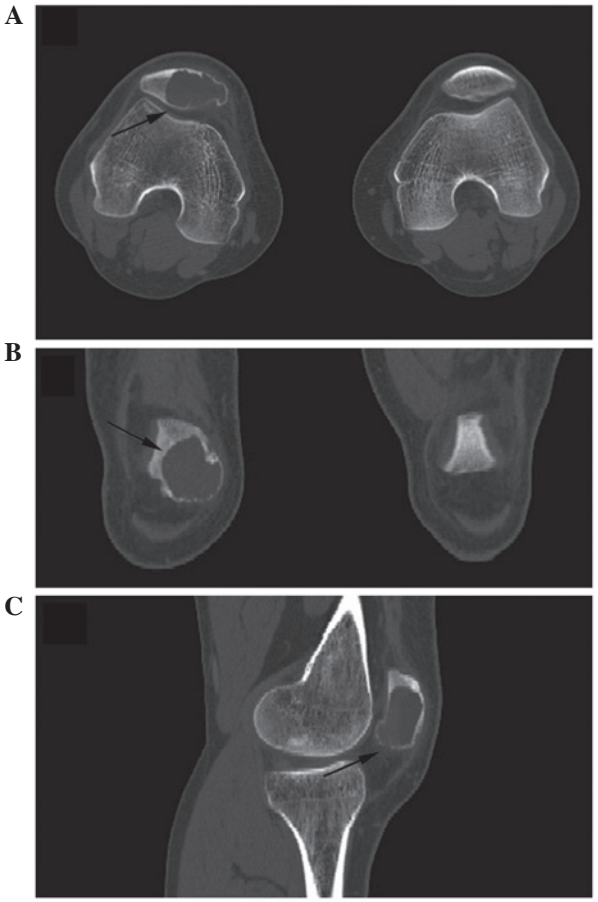

Figure 2. Preoperative computed tomography scan. (A) Axial, (B) coronal and (C) sagittal images showing a well-defined lytic lesion (arrows) with a thin cortex occupying 2/3 of the patella.

Discussion

Primary bone tumors originating from the patella are rare lesions. In a review by Mercuri and Casadei (1), it was reported that benign tumors of the patella are more frequent than malignant tumors. The most common diagnosis of patellar tumors is GCT, accounting for 33% of all patellar tumors, while patellar ABC accounts for only 5% of all patellar tumors (1). Despite the fact that GCT combined with ABC has also been reported

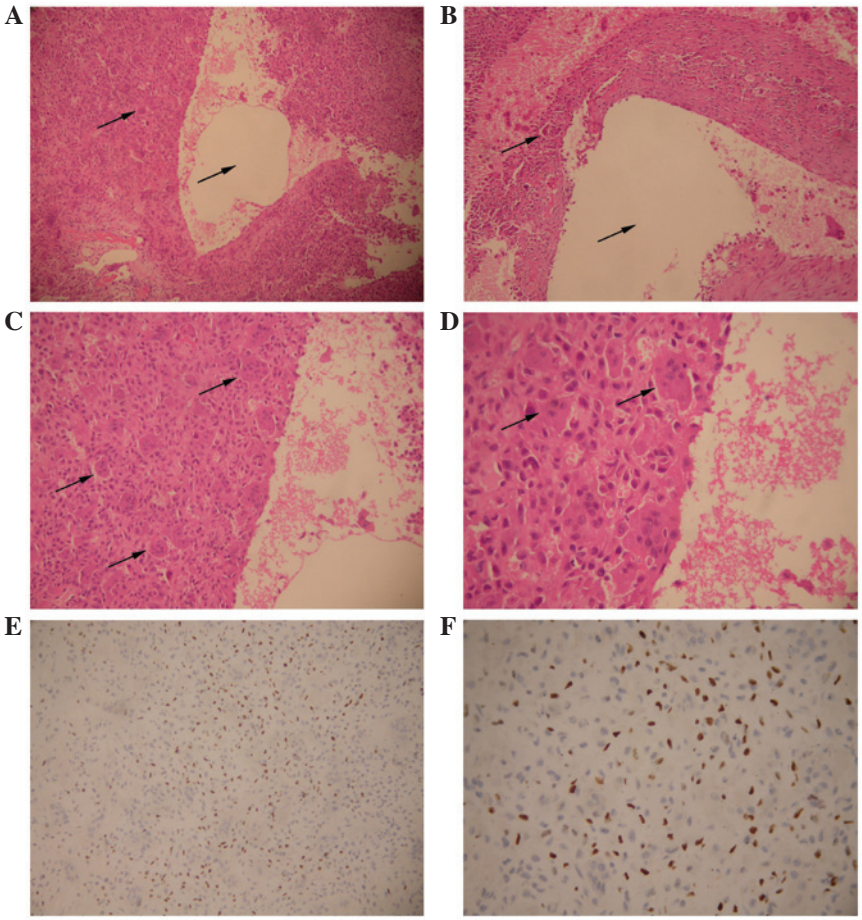

Figure 3. Photomicrograph of the resected tumor. (A and B) GCT lesion associated with aneurysmal bone cyst (arrows) (H&E staining; magnification, x40). Typical multinucleated giant cells (arrows) and uniform ovoid mononuclear cells were visible at (C) x100 and (D) x400 magnification (H&E staining). Immunohistochemistry revealed that the GCT cells were partly positive for P63: (E) x100 and (F) x400 magnification. GCT, giant cell tumor; H&E, hematoxylin and eosin.

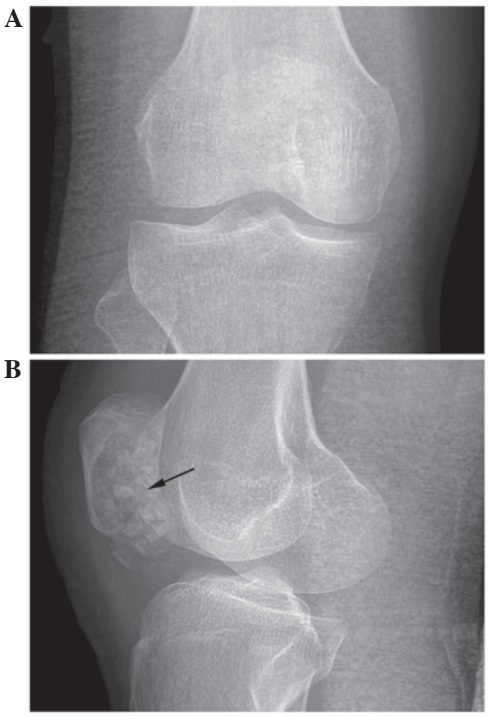

Figure 4. Postoperative plain radiograph. (A) No distinct change was observed on the frontal radiograph, while (B) the lateral radiograph showed that the cystic cavity had been filled with bone chips (arrow).

in other bone locations, including the rib (3), calcaneus (4), talus (5), spine (6) and radius (7), only 1 case of patellar symbiotic tumors has been reported to date (8).

GCT of the bone is a common benign, locally aggressive bone tumor that rarely metastasizes or causes mortality. Recurrent and malignant GCTs have been associated with higher rates of aneusomy than those exhibited by benign, diploidic GCT lesions. GCT has been reported to account for ~5% of all pathologically diagnosed primary bone tumors in Western populations (9-11) and 20% of all biopsy-analyzed primary bone tumors in the Chinese population (12). It can affect individuals of any age, but tends to occur in young adults aged 20-45 years. Women are considerably more susceptible to GCT than men. The majority of GCTs are located in the epiphyseal regions of long bones, with the sacrum or spine as secondary sites of involvement (13). Regional pain and tenderness upon palpation are the most common symptoms of GCT (14). Certain patients may present with a visible or palpable mass. In addition, effusion, decreased range of motion, activity-related pain or pathological fractures may also be observed (15).

Radiographically, GCT may involve the diseased patella, while ill-defined margins and pathological fractures are frequently observed (16). CT scan demonstrates cortical expansion and destruction, while magnetic resonance imaging

(MRI) reveals the presence of intra-articular fluid, as well as the involvement of any ligament, tendon and surrounding tissue and/or joint (17).

The typical histological appearance of GCT is that of a locally destructive neoplasm with tumors composed of mesenchymal fibroblast-like stromal cells (13). Other histological characteristics include the presence of monocytic, mononuclear cells of myeloid lineage and osteoclast-like, multinucleated giant cells (13). Treatment of GCT mainly includes curettage, followed by bone grafting, excision, irradiation, amputation (for certain patients who suffer from malignant GCT) and adjuvant therapy (such as polymethylmethacrylate, phenol and aqueous zinc chloride) following surgery (18).

ABC is a benign bone tumor with a low incidence (~5% of all patellar tumors) that may manifest as a primary or secondary lesion to another neoplasm, such as a GCT of the bone or chondroblastoma (1). The majority of ABC patients are females aged <20 years (19). Any bone may be affected by ABC; however, the most common location is the metaphysis of long bones, most frequently those of the lower extremities (20). Although the clinical manifestations of ABC highly vary, the most common ones are pain and swelling. Physical activity may aggravate the pain (15). The skin around the region of the cyst may display inflammation and tenderness upon palpation (15). ABC near a joint may be the reason for a decreased range of motion, while spinal ABC may cause nerve and cord impingement. Pathological fractures due to ABC occur in ≤20% of all ABC cases (21). Small lesions display minor or no symptoms, and may be discovered by radiographs following a pathological fracture, or incidentally. Plain film imaging reveals a radiolucent lytic lesion in the metaphysis of the bone, which is commonly eccentric and displays thinning of the cortex. ABCs are often described as 'soap bubbles' due to their expansile nature (20). CT scan may indicate the presence of fluid in the bone and soft tissue involvement, while MRI scan reveals multiple fluid-fluid levels within the lesion. In MRI, the outline of the cyst appears as an enhanced ring around the lesion, and is observed on T1- and T2-weighted images (20).

Macroscopically, the appearance of ABC is similar to that of a blood-filled sponge with a thin periosteal membrane (20). Microscopically, the lesion is composed of blood-filled spaces separated by connective tissue septa containing fibroblasts, osteoclast-type giant cells and reactive woven bone (22). Effective radical treatment of primary and recurring ABC includes complete resection of all tissues lining the cyst and any of its components from the surrounding soft tissues (23). Following resection, the cavity could be filled with bone chips, mesenchymal stem cells or polymethylmethacrylate bone cement (20). According to the position and size of the lesion, internal fixation may be used to maintain stabilization. Embolization has also been used for larger lesions (20).

The present study reported an unusual case of GCT combined with ABC in the patella. Despite numerous reports on GCT or ABC characteristics, the association between GCT and secondary ABC remains poorly understood. Due to the limited number of reports, the possibility of concurrent GCT with ABC may be easily overlooked. The aim of the present study was to supply clinical information in order to identify this rare type of patellar tumor.

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