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Giant Cell Tumor in the Cuboid

A case report

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Abstract

Giant cell tumors are uncommon in the hand or foot. Although several case reports with giant cell tumor in these locations are described, there have only been a few reported cases of this tumor type located in the cuboid of the foot. In this report, we present a case with giant cell tumor in the cuboid. A 19-year-old Japanese man was admitted with a three-month history of aching pain in his right foot. Radiographs demonstrated an osteolytic lesion in the cuboid. Magnetic resonance (MR) images showed that the lesion had homogeneously iso-signal intensity in T1-weighted images, and relatively high signal intensity in T2-weighted images. A biopsy specimen exhibited a proliferation of mononuclear round or oval cells and multinucleated giant cells, indicating a diagnosis of giant cell tumor. Curettage with cautery using 50°C high-temperature saline for 15 minutes followed by an artificial bone graft were performed. The patient has remained asymptomatic with no signs of local recurrence or pulmonary dissemination seven years after surgery.

Giant cell tumors typically occur in the epiphyseal/metaphyseal region of the long tubular bone but are uncommon in the hand or foot [1,2]. In English literature, several case reports with giant cell tumor in the hand or foot are certainly described, but there have only been a few reported cases of this tumor type located in the cuboid of the foot [2,3,4,5,6]. To our knowledge, no case of this type has been described in the Japanese literature. In this report, we present a 19-year-old Japanese man with giant cell tumor in the cuboid treated with curettage and cautery using high-temperature saline followed by an artificial bone graft. Radiological, histological differential diagnoses, and biological behavior are also discussed.

Case report

A 19-year-old Japanese man was admitted to our institution in July 1999 with a three-month history of aching pain in his right foot. Physical examination revealed tenderness at the cuboid of the right dorsal foot, but no local heat, swelling, or overlying venous distention were observed around the foot. Radiographs showed an osteolytic lesion in the cuboid (Figure 1). The cortical bone of the cuboid was thin and expanded. However, there was no apparent cortical destruction. Computed tomography demonstrated thinning of the cortex and an intramedullary lesion with an iso-density to the muscle and with no mineralization or trabeculation (Figure 2). Magnetic resonance (MR) images demonstrated that the lesion had homogeneously iso-signal intensity in T1-weighted MR images (Figure 3-A), and relatively high

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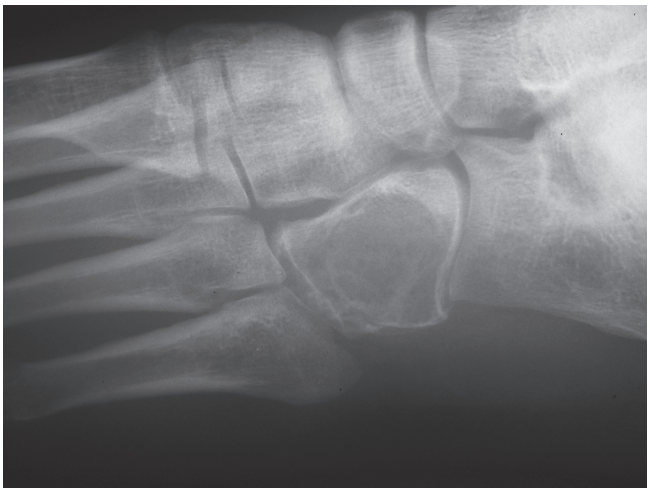

Figure 1. An oblique view of the left foot showing an osteolytic lesion in the cuboid. The margin between the lesion and preserved medullary portion of the cuboid is clear, and the cortical bone is thinned and expanded.

signal intensity in T2-weighted images (Figure 3-B). The margin between the lesion and the preserved medullary portion of the cuboid was clear. Gadolinium-enhanced MR images revealed diffuse and moderate enhancement of the lesion (Figure 3-C). The results of laboratory analyses were all within normal limits. A chest radiograph was normal, and bone scintigraphy showed a single high uptake portion of the cuboid, denying a multifocality of the disease.

A biopsy specimen was, grossly, friable and red-brown in color, and exhibited histologically a proliferation of mononuclear round or oval cells and multinucleated giant cells. Nuclear figures of the mononuclear cells were similar to those of the multinucleated giant cells. Giant cell tumor was considered as the diagnosis. Curettage and cautery using 50°C high-temperature saline for 15 minutes followed by an artificial bone (APACERAM®) graft were performed in August 1999. In the surgical specimen, proliferation of round-to oval or even spindle-shaped mononu-

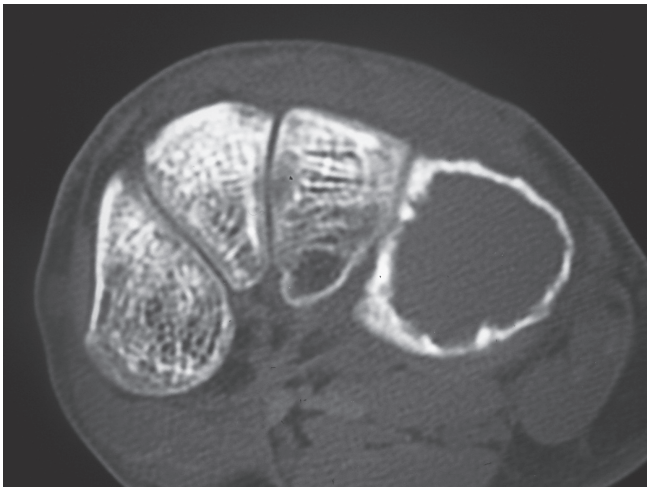

Figure 2. CT of the foot showing an intramedullary lesion with an iso-density to the muscle. Cortical bone is thin, but its continuity is preserved. There is no apparent mineralization or trabeculation.

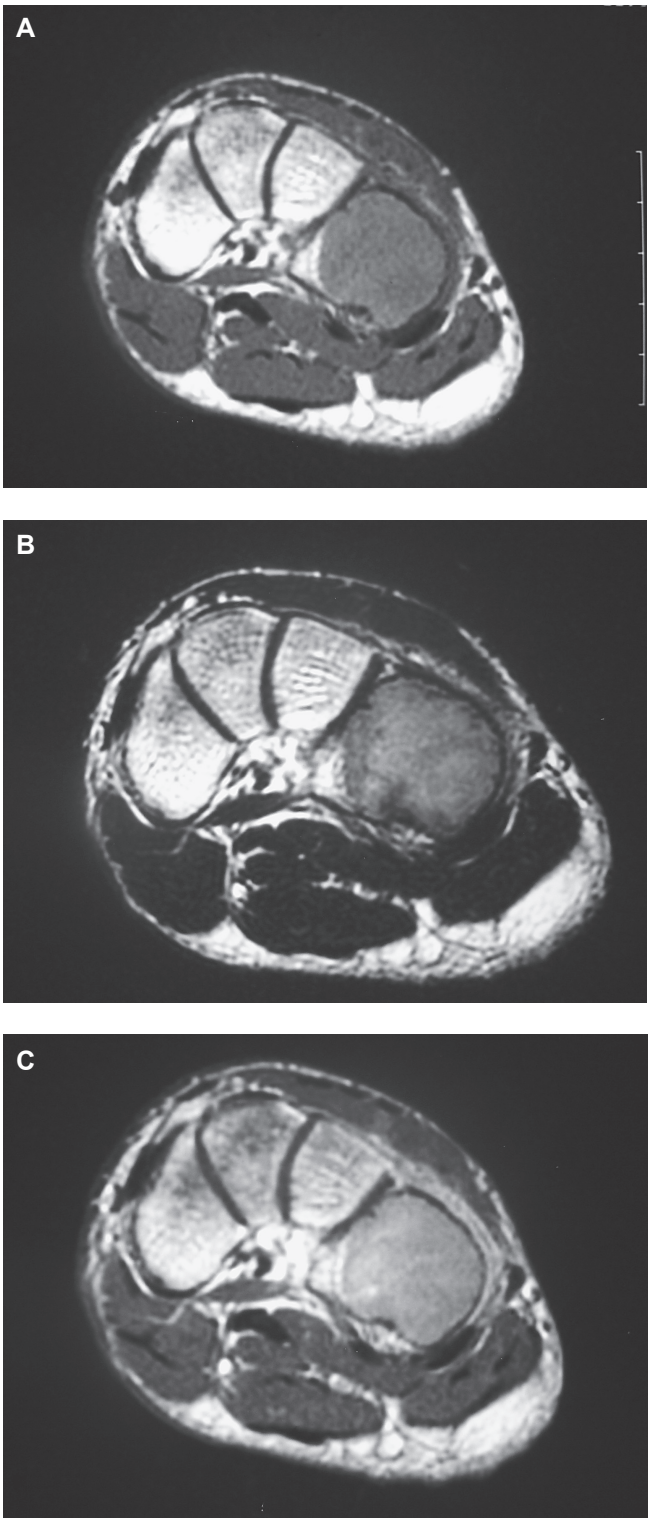

Figure 3. (A) The lesion in the cuboid showing a homogeneously iso-signal intensity in a T1-weighted image (TR:480, TE:9), and (B) the lesion demonstrating an irregular and relatively high signal in a T2-weighted image (TR:3500, TE:97.6). (C) Gadolinium-enhanced MR images showing diffuse enhancement of the lesion.

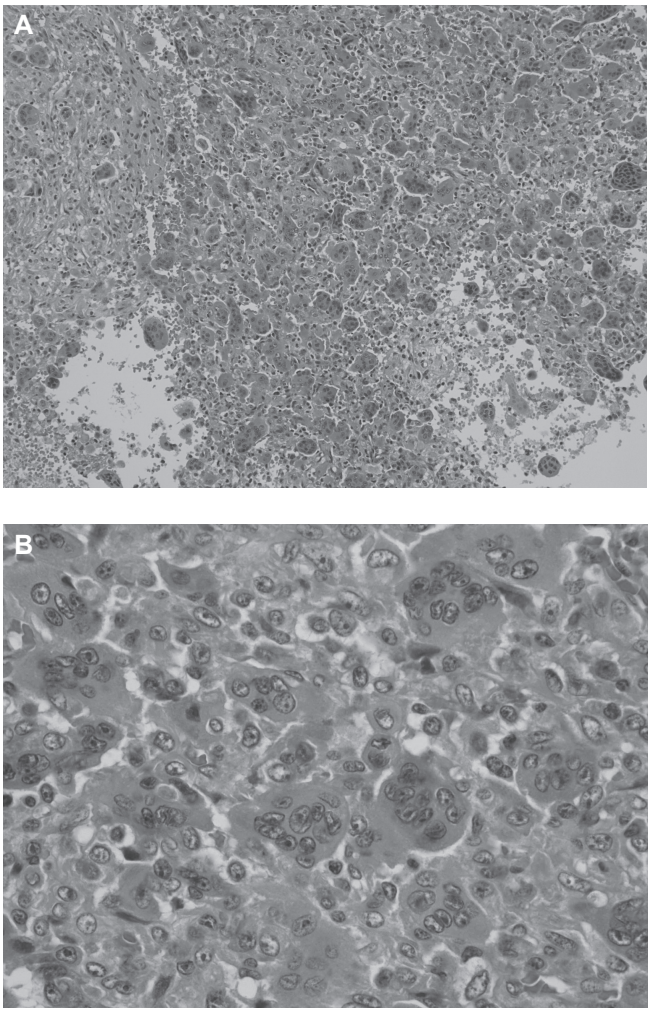

Figure 4. (A) A low-power view of histological specimen showing a proliferation of mononuclear round or short spindle cells and multinucleated giant cells. Microcystic changes were observed in the lower part. There was no chondroid island, cartilage, or reactive bone formation (H&E; original magnification x 100). (B) Nuclei of mononuclear cells were surrounded by an ill-defined cytoplasmic zone, and discernible intercellular substance was absent. The nuclei of the mononuclear cells were similar to the nuclei of multinucleated giant cells. Mitotic figures were occasionally observed, but there was no atypical mitosis (H&E; original magnification x 400).

clear cells and multinucleated giant cells were prominent. Nuclei of mononuclear cells were surrounded by an ill-defined cytoplasmic zone, and discernible intercellular substance was absent. The nuclei of the mononuclear cells were similar to those of multinucleated giant cells. Mitotic figures were observed in the number of 3 to 10 per 10 high power fields. However, there was no atypical mitosis, and nuclear atypia was generally mild. Occasionally, small collection of foam cells and cystic changes with proliferation of fibroblastic cells were observed. There was no chondroid island, cartilage or reactive bone formation. Diagnosis of giant cell tumor in the cuboid was made (Figure 4-A, B). The patient has remained asymptomatic with no signs of local recurrence and pulmonary dissemination seven years after the surgery.

Discussion

It is well known that nearly 70% of giant cell tumors occur in the region around the knee and wrist, while they are relatively rare in the foot, accounting for only 1.4% (8/574) of cases seen at the Mayo Clinic [1] and 1.8% (33/1849) of cases reported to the Japanese Bone Tumor Register [7]. Among cases of giant cell tumors occurring in the foot, cuboid is quite a rare site. According to a review of the literature from 1940 to 1984 by Mechlin et al., only one lesion in the cuboid was included among 38 giant cell tumors of tarsal bones [3]. In 1984, Wold and Swee reported 43 patients with giant cell tumors of small bones, and 3 of them were in the cuboid [2]. Recently, Biscaglia et al. reported the updated data of Rizzoli Institution, and they found that, in their series of 18 cases of giant cell tumors in the foot, 3 were located in the cuboid [6]. Other authors have reported only sporadic cases of cuboid lesions in their series of giant cell tumors [4,5]. However, in other large series, no giant cell tumor in the cuboid was found [8,9]. In Japanese literature, there has been no report of a case with giant cell tumor occurring in the cuboid, although Nishida et al. in 1993 reported a case with a giant cell tumor involving several tarsal bones including the cuboid [10]. To our knowledge, our case is therefore the first reported one with giant cell tumor in the cuboid in Japan.

The differential diagnosis of the current case includes slow growing and/or expansile osteolytic lesions, such as chondroblastoma and other cartilage tumors, aneurysmal bone cyst (ABC), and solid variant of ABC. However, mineralization, which is a common finding in cases of chondroblastoma and cartilage tumors, was not observed in the radiographs and CT. In addition, in the histological specimens, there were no chondroid islands or cartilage tissues. Chondroblastoma and other cartilage tumors were not likely as a diagnosis of the current case. In ABC, cystic structures are prominent and a fluid-fluid level is commonly seen in MR images or CT. In giant cell tumors such changes have been seen in 8.5% [11]. In addition, if the lesion consists of only solid components, solid variant of ABC (giant cell reparative granuloma), which is commonly seen in foot or hand [1], should be considered. Therefore, preoperative biopsy is mandatory to obtain a differential diagnosis between giant cell tumor and ABC or solid variant of ABC [1]. In both ABC and solid variant of ABC, large number of multinucleated giant cells is seen, and the basic and entire fibrogenic quality is the main features that differentiate ABC or solid variant ABC from true giant cell tumor. Since, in the current case, most of the lesion showed typical features of true giant cell tumor, the diagnosis of giant cell tumor in the cuboid was confirmed. The young age of the current case compared with those who had giant cell tumors in the long bones may assume a possibility of other diagnosis. However, the fact that patients with giant cell tumors in hand and foot location seem to be younger compared with those who have tumors in long bones has been stressed in the literature [1,2,6,12]. This features was also underlined by the present case.

In giant cell tumor, more aggressive biological behavior of a tumor arising in the hand than those arising in more central locations has been reported. Averill et

al. reported that there was a combined recurrence rate of 83% for giant cell tumors of the hand treated with curettage, and it has been recommended that these lesions be managed with resection or amputation [12]. In contrast to cases of the hand, O'Keefe et al. reported that giant cell tumor in the foot behaves not more aggressively than giant cell tumors found in more common locations, and they suggested that giant cell tumor of the foot can be treated effectively using intralesional procedures [9]. Biscaglia et al. reported a somewhat greater biological aggressiveness of giant cell tumors of the hand and foot, since the 7 of 11 cases developed local recurrences after curettage or curettage with bone graft. On the other hand, they stressed that none of 8 cases of foot treated by curettage followed by treatment with phenol showed local recurrence [6]. Therefore, we treated the current case with curettage and cautery using 50°C high-temperature saline followed by an artificial bone graft, and 7-year follow-up period was uneventful. A cautery using high-temperature saline might be useful to prevent a local recurrence of giant cell tumor of the foot provided that it is combined with a thorough curettage.

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