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Case Reports and Series

Benign Fibrous Histiocytoma of the Talus: A Case Report

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ABSTRACT

Benign fibrous histiocytoma (BFH) is a rare benign primary bone lesion that occurs most frequently in the nonmetaphysis region of the long bones and the pelvic bones. The talus is a rare location for a BFH, which has not been reported previously in the literature. We report the case of a 19-year-old male patient with BFH of the talus, who was treated with curettage, followed by filling of the bone defect with calcium phosphate cement. The patient was free of pain and without local recurrence 5 years after the surgery. We describe the detailed radiographic findings of this rare lesion and discuss the differential diagnosis of such talar lesions.

Benign fibrous histiocytoma (BFH) of bone is a very rare benign bone tumor, and according to the World Health Organization's classification of tumors, it is categorized into the same disease entity as nonossifying fibroma (1). BFH and nonossifying fibroma have the same microscopic features. The tumors consist of bland, spindle-shaped fibroblasts, which are arranged in a storiform growth pattern and which include hemosiderin deposition within the stromal cells and the gathering of foamy histiocytes (1). However, the clinical presentation of the 2 entities is different (2). Nonossifying fibroma arises in the metaphysis of the long bones of the lower extremities, especially the distal femur and the proximal and distal tibia, typically in the second decade of life, whereas BFH arises in the nonmetaphyseal locations of the long bones and in the pelvic bones in subjects after the second decade of life.

We report the case of a patient with BFH of the talus. This site of involvement is extremely unusual. There has been only 1 previous report of BFH of the tarsal bone (3). We present the details of this rare condition, review the previous reports, and discuss the clinical and imaging features.

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Research Ethics Standard Compliance

The study was performed in accordance with the 1964 Declaration of Helsinki and its later amendments. The study was approved by the ethics committee of the University of Mie, and we received written informed consent from the patient for the publication of this case report.

Case Report

A 16-year-old male track and field athlete visited a private orthopedic clinic because he felt slight pain with mild swelling in the dorsal portion of his left foot during a workout. However, no abnormalities were indicated after a radiographic examination. Three years later, he visited the same clinic because he sometimes felt a sense of discomfort in his foot. He was referred to our hospital because the radiographic findings showed a lytic lesion of the talus.

On physical examination, there was a slight tenderness on the dorsal side of the talar head, without swelling, redness, or local heat. The range of motion of the left ankle joint was not restricted. The patient's full blood cell count and erythrocyte sedimentation rate were within normal limits. The radiographic findings showed a well-defined expanding lytic lesion with moderate marginal sclerosis at the dorsal portion of the talar neck and head (Fig. 1). Computed tomography indicated the partial disappearance of the dorsal cortical bone of the talar neck and head, with an irregular sclerotic margin. Neither calcification nor bone formation was observed (Fig. 2).

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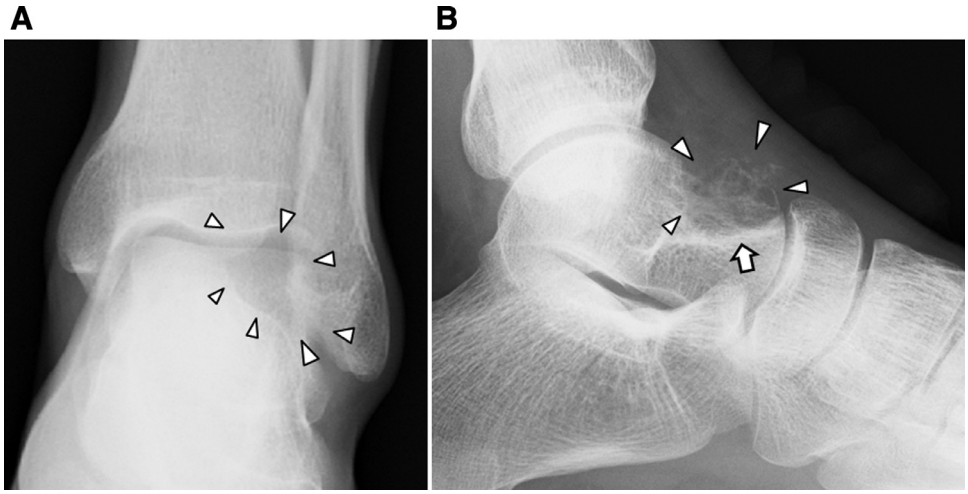

Fig. 1. The radiographic findings of the left foot show a well-defined expanding lytic lesion (*open triangle*) with moderate marginal sclerosis (*open arrow*) at the dorsal portion of the talar neck and head. (*A*) Anteroposterior view. (*B*) Lateral view.

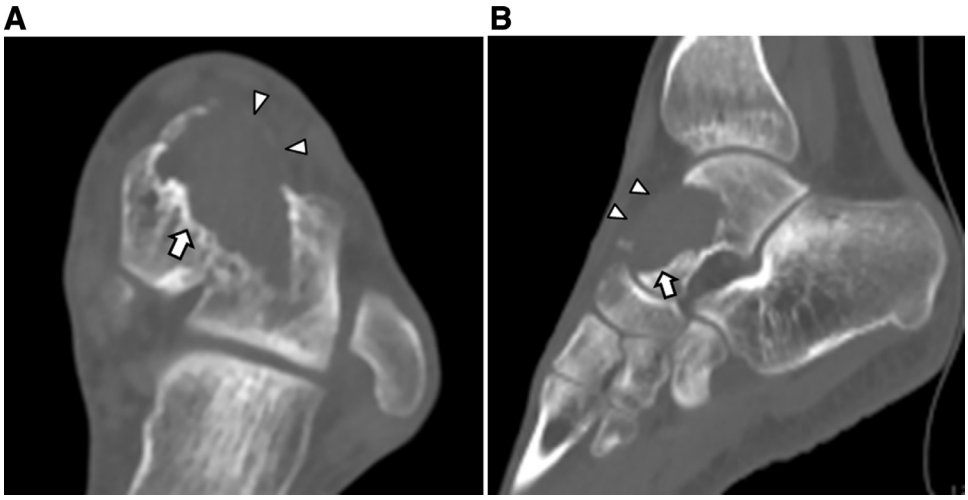

Fig. 2. A sagittal reformatted computed tomographic image shows the partial disappearance of the dorsal cortical bone of the talar neck (*open triangle*) and head, with irregular sclerotic margins (*open arrow*). Neither calcification nor bone formation was observed.

On magnetic resonance (MR) imaging, the T1-weighted MR images demonstrated that the tumor had intermediate signal intensity in comparison with the skeletal muscle and that it was homogeneously enhanced by the administration of gadolinium. The T2-weighted MR images revealed that the tumor had high signal intensity (Fig. 3).

An incisional biopsy was performed to obtain a pathologic diagnosis. The microscopic findings showed the proliferation of the

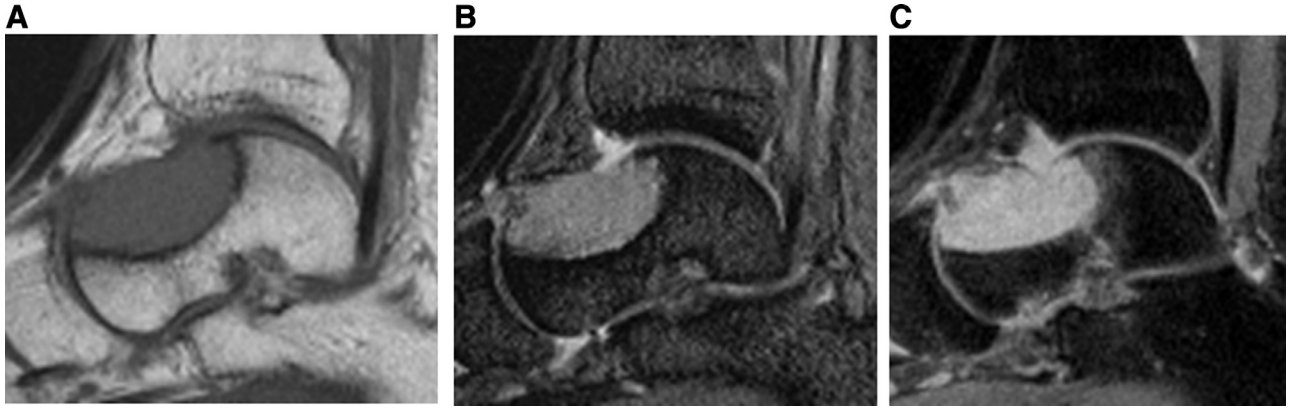

Fig. 3. The findings of the magnetic resonance (MR) imaging of the left foot (sagittal view). The T1-weighted MR images demonstrate intermediate signal intensity in comparison with the skeletal muscle (*A*) and homogeneous enhancement by gadolinium (*B*). (*C*) The T2-weighted MR images reveal high signal intensity.

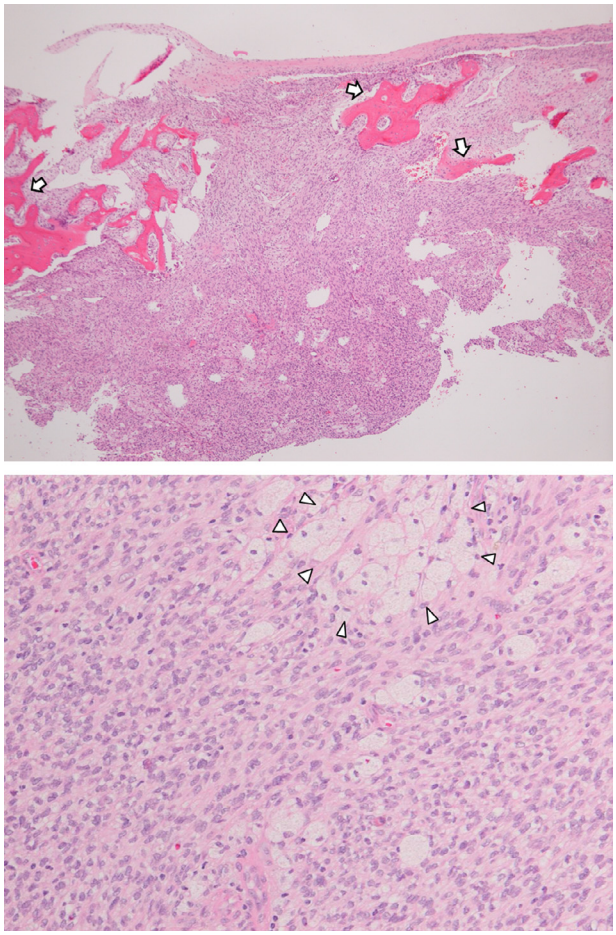

Fig. 4. (*A*) The low-power microscopic findings show the proliferation of spindle-shaped fibroblastic cells, which are arranged in a fascicular and storiform growth pattern with the reactive new bone (*open arrow*) (magnification × 40; hematoxylin and eosin stain). (*B*) Clusters of foamy histiocytes are also observed in a small part of the specimen (open triangle) (magnification × 200; hematoxylin and eosin stain).

spindle-shaped fibroblastic cells, which were arranged in a fascicular and storiform growth pattern, without any nuclear atypia. Clusters of foamy histiocytes were also observed in a small part of the specimen (Fig. 4). Immunohistochemical staining indicated negative findings for alpha-smooth muscle actin and S-100, weak staining for HHF35, and focal staining for CD34. The MIB-1 index was 3.6%. A diagnosis of BFH of the talus was provided, and curettage of the bone tumor, followed by the reconstruction of the bone defect with calcium phosphate cement, was performed. The patient has been free from symptoms and has not had any local recurrence in the 5 years after the operation.

Discussion

BFH arises in the nonmetaphyseal location of the long bones and in the pelvic bones of patients after the second decade of life (1). The Table shows a summary of the anatomic locations of the previous studies. Cases of BFH involving the talus could not be identified in the previous literature. Thus, to our knowledge, this presentation is the first report of BFH of the talus.

Bone tumors of the talus are uncommon. Dhillon et al (4) reviewed 12 cases of bony tumors involving the talus treated at a single institute and noted that giant cell tumors were the most common type of tumor.

Table
A summary of the locations of the previously reported cases of benign fibrous histiocytoma

Location	Cases (n)	References
Femur	7	1, 3, 4
Maxilla or mandible	6	3, 4, 8
Spine	6	2, 3, 4, 9
Pelvis	3	1, 3, 8
Radius	2	1, 3
Tibia	2	1, 3
Phalanx	1	3
Humerus	1	1, 3
Fibula	1	1, 3
Rib	1	1, 3
Calcaneus	1	3

Benign bone tumors, including aneurysmal bone cysts, chondroblastomas, intraosseous lipomas, and osteoblastomas, have also been reported as tarsal bone tumors (5–7). Osteosarcoma cannot be precluded from the differential diagnosis of these tumors because of their similar features.

In the present case, the radiographic findings showed well-defined and sclerotic margins. Computed tomographic images demonstrated the disappearance of the cortical bone of the dorsal side of the talus. Thus, it was first considered that the tumor had benign aggressive or low-grade malignant properties, similar to those of a giant cell tumor, chondroblastoma, or low-grade osteosarcoma. The MR images were helpful for excluding giant cell tumor and chondroblastoma from the differential diagnosis, because both of these demonstrate less likely homogeneous signal intensity on the T1- and T2-weighted images (1). The MR characteristics of BFH were of intermediate signal intensity, which was homogeneously enhanced by gadolinium, on the T1-weighted images and of high signal intensity on the T2-weighted images. These MR findings are consistent with the cases of BFH of bone reported previously (8). Thus, MR imaging is a useful diagnostic modality for the diagnosis of BFH of bone.

Curettage followed by the reconstruction of the bone defect by using a bone graft or an artificial bone is a common treatment for BFH of bone. Old case series indicate that BFH may have a poor prognosis in some cases (9,10). However, these old series included low-grade malignant bone tumors or more aggressive benign bone tumors, such as giant cell tumor of bone, and were not restricted to BFH of bone (3). The prognosis for BFH is excellent, with almost no recurrence after complete surgical resection.

In conclusion, we reported the case of a 19-year-old male patient with BFH of the talus. To our knowledge, this is the first report of BFH in the talus. The final diagnosis was made after a thorough evaluation of the histologic, radiologic, and clinical features. The tumor was treated with curettage, and the bone defect was filled with calcium phosphate cement. At the final follow-up visit 5 years after the operation, the patient was free of pain, without local recurrence.

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