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Giant Cell Tumor of the Distal Phalanx of the Fourth Toe: A Case Report

Masahiro Yokouchi, MD, PhD [1], Yoshiya Arishima, MD, PhD [2], Satoshi Nagano, MD, PhD [3], Hirofumi Shimada, MD [2], Shunsuke Nakamura, MD, PhD [2], Takao Setoguchi, MD, PhD [4], Ichiro Kawamura, MD, PhD [2], Yasuhiro Ishidou, MD, PhD [5], Setsuro Komiya, MD, PhD [6]

[1] *Associate Professor, Department of Orthopaedic Surgery, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*
[2] *Orthopedist, Department of Orthopaedic Surgery, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*
[3] *Lecturer, Department of Orthopaedic Surgery, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*
[4] *Associate Professor, The Near-Future Locomotor Organ Medicine Creation Course, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*
[5] *Associate Professor, Department of Medical Joint Materials, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*
[6] *Professor, Department of Orthopaedic Surgery, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Japan*

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ABSTRACT

Giant cell tumor of the bone is a benign, but locally aggressive, primary bone tumor of unknown origin. It most commonly occurs in the long bones and is only rarely found in the phalangeal bones, such as the distal phalanx of the foot. In our review of English-language published studies, only 4 other cases of giant cell tumor involving the distal phalangeal bone of the foot had been reported to date. We report a case of giant cell tumor arising in the distal phalanx of the fourth toe in a 28-year-old female. Although bisphosphonate therapy was administered, the tumor showed highly aggressive behavior with ulceration of the overlying skin, and the patient underwent phalangeal amputation 1.5 months after diagnosis.

Giant cell tumor (GCT) is a relatively common benign bone tumor that accounts for 5% of all primary bone tumors and almost 20% of benign primary bone tumors (1). The tumor most frequently occurs in the metaphyseal region of the long bones, in particular, the distal femur, proximal tibia, distal radius, and proximal humerus. Its occurrence in the small bones is very rare (1,2). GCT of the small bones is characterized by extensive bony destruction and displays more aggressive behavior, with a greater recurrence rate, than that of GCT involving the ends of the long bones (2). Although the mainstay of treatment of GCT of the long bones is surgery, including en bloc or wide resection or curettage with adjuvant therapy, no consensus has been reached regarding the ideal treatment of GCT located in the small bones owing to the rarity of this condition (2). Among the cases of small bone GCT, GCT of the distal phalanx of the foot is particularly rare. Our review of published English-language studies to date yielded only 4 reported cases of GCTs involving the distal phalanx of the foot (3–6). In the present report, we describe a case of GCT located in the distal phalanx of the foot in a patient who was initially treated with conservative bisphosphonate therapy. To the best of our knowledge, no isolated case reports of phalangeal bone GCT treated with bisphosphonates have been previously reported. Although several reports have demonstrated the efficacy of bisphosphonate therapy in the treatment of GCT (7,8), no response to bisphosphonate therapy was observed in our patient. The tumor grew rapidly, completely displacing the distal phalanx, and causing ulceration of the overlying skin; therefore, the patient underwent phalangeal amputation. The present case report illustrates a pathologic rarity and incorporates a brief review of the published data relevant to surgeons.

Case Report

A previously healthy 28-year-old female visited a local hospital because of increasing pain and swelling in the distal part of her fourth toe that she had first experienced 1 month previously. She had no history of trauma to this area. The patient was diagnosed with a bone tumor of the distal phalanx of the fourth toe from the radiographic findings and was subsequently referred to our hospital. At her first visit to our hospital, an enlarged fourth toe and toenail were observed, with severe tenderness in the distal part of

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Address correspondence to: Masahiro Yokouchi, MD, PhD, Department of Orthopaedic Surgery, Graduate School of Medical and Dental Sciences, Kagoshima University, 8-35-1 Sakuragaoka, Kagoshima 890-8520, Japan.

E-mail address: masahiro@m3.kufm.kagoshima-u.ac.jp (M. Yokouchi).

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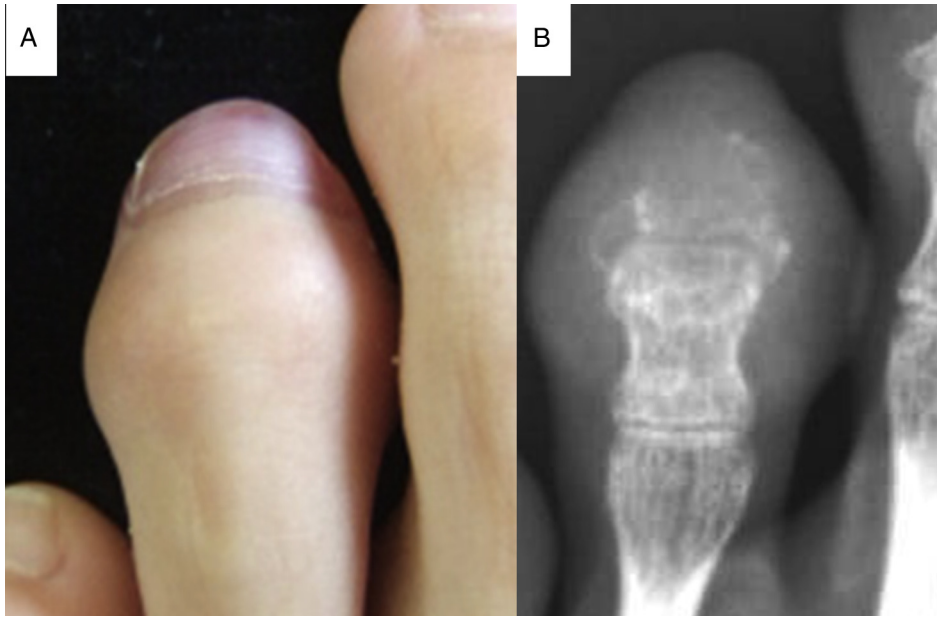

Fig. 1. (*A*) Clinical photograph showing enlargement of the toe and toenail in the distal part of the patient's fourth toe. (*B*) Anteroposterior radiograph of the distal phalanx of the fourth toe at initial presentation showing osteolysis with cortical destruction.

the fourth toe (Fig. 1*A*). The overlying skin was smooth and nonadherent, and no redness or warmth was present in the area. The laboratory test results were all within the normal range. Radiographs showed osteolysis with cortical destruction in the region of the distal phalanx of the fourth toe (Fig. 1*B*). No calcification or periosteal reaction was observed. A tissue biopsy was performed, revealing round to oval mononuclear cells with numerous osteoclast-like giant cells, leading to the diagnosis of GCT of the bone (1). The patient did not agree to radical surgical treatment such as en bloc resection; thus, conservative bisphosphonate therapy was initially administered (1 cycle of 4 mg of zoledronic acid). However, the tumor did not show any response to the chemotherapy and began to show highly aggressive behavior with skin ulcerations forming (Fig. 2*A*). Radiographs and magnetic resonance imaging revealed complete displacement of the distal phalanx by the tumor (Figs. 2*B* and 3*A*). The patient underwent phalangeal amputation only 1.5 months after the diagnosis (Fig. 3*B*). Histopathologic analysis of the resected specimen (Fig. 3*C*) confirmed the

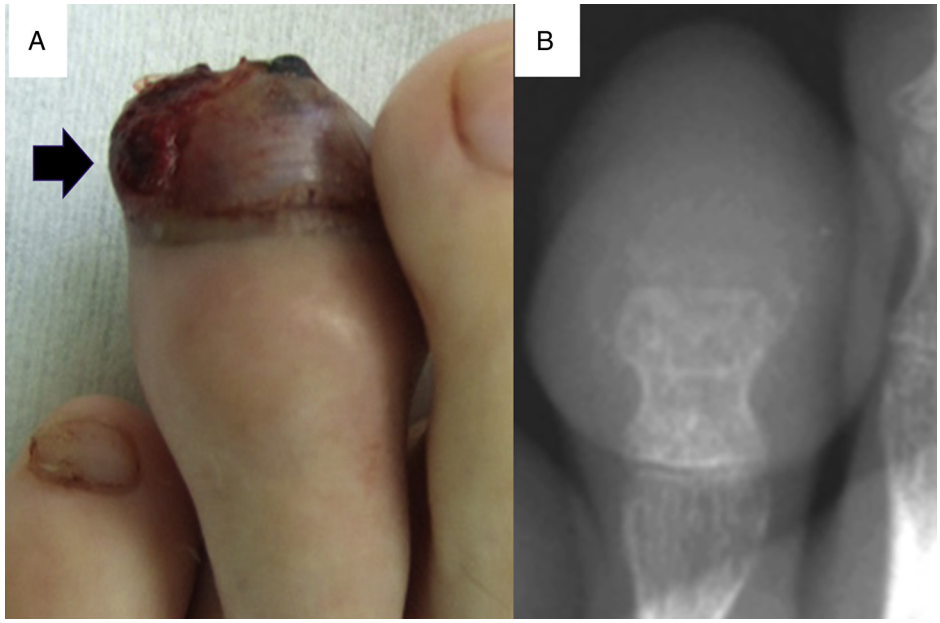

Fig. 2. (*A*) Clinical photograph showing skin ulceration associated with the tumor (*arrow*). (*B*) Anteroposterior radiographs of the distal phalanx of the fourth toe 1 month after bisphosphonate therapy showing rapid progression of the tumor with cortical destruction.