

Fibrous Dysplasia Arising from the Calcaneus

SHUJI ISEFUKU, MASAHITO HATORI, SHIGERU EHARA,¹
MASAMI HOSAKA, KATSU ITO² and SHOICHI KOKUBUN

Department of Orthopaedic Surgery, Tohoku University School of Medicine, Sendai 980-8574, ¹Department of Radiology, Iwate Medical University School of Medicine, Morioka 020-8505, and ²Department of Orthopaedic Surgery, Hukaya Hospital, Miyagi 987-1222

ISEFUKU, S., HATORI, M., EHARA, S., HOSAKA, M., ITO, K. and KOKUBUN, S. *Fibrous Dysplasia Arising from the Calcaneus*. Tohoku J. Exp. Med. 1999, 189 (3), 227-232 — A case of an 18-year-old woman with fibrous dysplasia arising in the calcaneus, which is extremely rare, is reported, with the emphasis placed on differential diagnosis from low-grade central osteosarcoma. She had a severe pain in her left ankle after sprain. Plain radiographs showed a radiolucent lesion measuring 6.3×2.5 cm with a sclerotic margin in the left calcaneus. CT scans showed a well-defined lytic lesion with disruption of the lateral cortex and an ossification or calcification in its center. On MR imaging, the lesion had iso-intensities and high intensities on T1 and T2 weighted images, respectively, but its central portions showed lower intensities both on T1 and T2 weighted images. The lesion was enhanced with gadolinium except for the central portions. The specimen obtained by open biopsy consisted of fibrous tissue and foci of irregular woven bone. None of the nuclear atypia, mitoses, longitudinal stream of bone or invasive nature of growth was detected. The diagnosis of fibrous dysplasia was histologically made. The lesion was curetted and packed with autogenous bone chips. No evidence of recurrence was noted postoperatively. ——— fibrous dysplasia; calcaneus; differential diagnosis © 1999 Tohoku University Medical Press

Fibrous dysplasia (FD) is a hamartomatous disorder characterized by fibro-osseous metaplasia (Mirra and Gold 1989). It commonly occurs in long bones, ribs, and skull (Mirra and Gold 1989), whereas it is rare in short bones, especially in the calcaneus (Schajowicz 1980; Mirra and Gold 1989; Unni 1996b). Low-grade central osteosarcoma sometimes mimics FD on radiographs. In this communication, we describe a case of FD arising in the calcaneus with emphasis placed on its differential diagnostic process.

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Address for reprints: Masahito Hatori, M.D., Department of Orthopaedic Surgery, Tohoku University School of Medicine, 1-1 Seiryomachi, Aoba-ku, Sendai 980-8574, Japan.
e-mail: mhato@gonryo.med.tohoku.ac.jp



Fig. 1. Lateral plain radiograph of the left hindfoot showing a well defined radiolucent lesion with a sclerotic margin in the anterior half of the calcaneus.

CASE REPORT

An 18-year-old woman had a severe pain in her left ankle after twisting. On physical examination one week after the accident, the medial aspect of the left calcaneus was tender to firm pressure. No mass was palpated. White blood cell count and C-reactive protein were in normal range. Plain radiographs showed a radiolucent lesion measuring 6.3×2.5 cm with a sclerotic margin in the anterior half of the left calcaneus (Fig. 1). CT scans (Toshiba Medical Systems Co., Ltd., Tokyo) showed disruption and partial disappearance of the lateral cortex of the calcaneus. Mineralization was noted in the center of the lesion (Figs. 2A and B). The lesion was well-defined on MR images (GE Yokogawa Medical Systems, Ltd., Tokyo). Its signal intensity was the same on T1 and higher on T2 weighted images, and the central portion showed lower intensity both on T1 and T2 images, as compared with the intensity of the muscle. Except for the central portions, the intensity of the lesion was enhanced by intravenous injection of gadolinium diethylene triamine pentaacetic acid (Figs. 3A, B and C). Based on those examinations, the most probable diagnosis was FD. But low-grade central osteosarcoma was not completely ruled out because of the cortical disruption. Biopsy revealed fibrous tissue and foci of irregular woven bones. None of the nuclear atypia, mitosis, longitudinal stream of bone and invasive nature of growth were



Fig. 2. CT scans of the left calcaneus (Toshiba Medical Systems Co., Ltd., Tokyo). A, Axial CT image (Window Level 60, Window Width 400) showing a radiolucent lesion with a sclerotic margin and the expansion of the lateral cortex of the calcaneus; B, Coronal CT image (Window Level 250, Window Width 1200) showing mineralization in the center of the lesion.

detected (Fig. 4). Consequently, the tumor was diagnosed as FD. The lesion was curetted and cancellous bones from the ilium were packed into the cavity. No evidence of recurrence was noted a year after surgery.

DISCUSSION

The incidence of the monostotic FD arising in the foot is only less than 2% (Schajowicz 1980; Mirra and Gold 1989; Unni 1996b). There was none arising in the calcaneus in Henry's (1969) and Gibson and Middlemiss's (Gibson and Middlemiss 1971) series consisting of 50 and 46 monostotic FD, respectively. To the best of our knowledge, only six cases of monostotic FD arising in the calcaneus have been reported: One by Dave et al. (1968), two by Schajowicz (1980) and three by Pandey (1971).

Differential diagnosis between FD and low-grade central osteosarcoma may sometimes be difficult because of their radiological similarities. We describe the differential diagnostic process to arrive at the correct diagnosis. The conditions are radiolucent with central calcifications described as "ground glass" appearances on plain radiographs (Mirra 1989; Mirra and Gold 1989). A thinned cortex with no disruption is a common finding in FD (Gibson and Middlemiss 1971). In contrast, low-grade central osteosarcoma is in general diagnosed depending on the

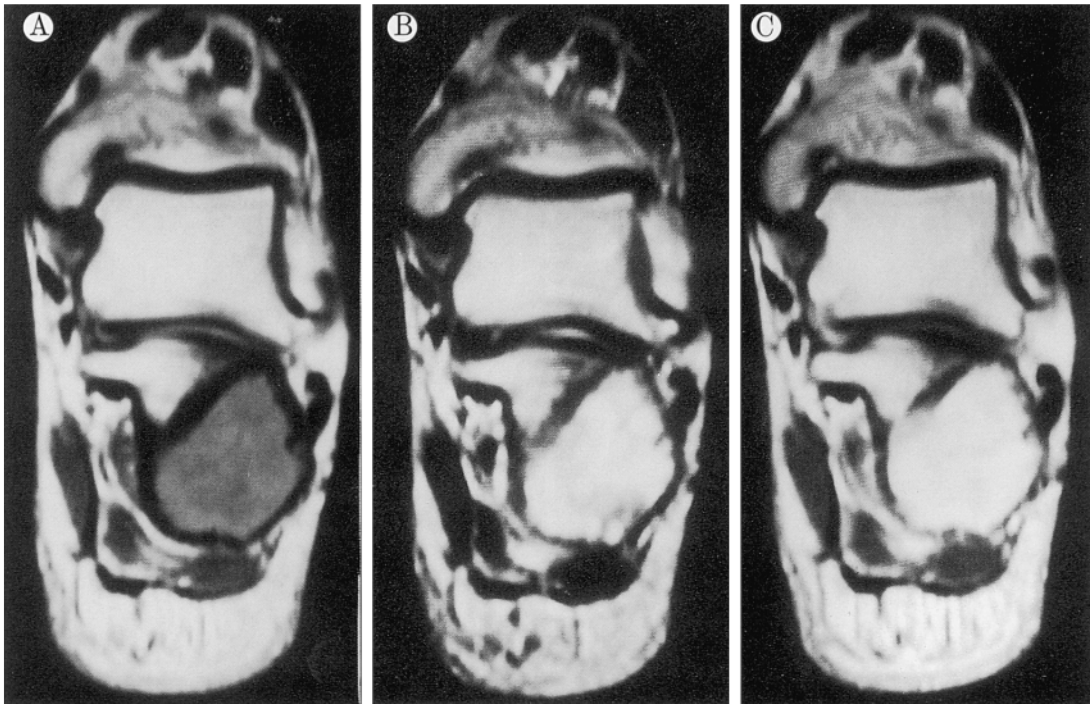


Fig. 3. MR images of the left calcaneus (GE Yokogawa Medical Systems, Ltd., Tokyo). A: Coronal T1 weighted image (spin echo, TR/TE = 360/25 milliseconds) showing a well defined lesion with the iso-signal intensity in the calcaneus as compared with the muscles. Central portion showed lower intensity. B, Coronal T2 weighted image (spin echo, TR/TE = 3000/100 milliseconds) showing the lesion with the high signal intensity. But central portion showed lower intensity. C, Coronal T1 weighted image after intravenous injection of gadolinium diethylene triamine pentaacetic acid (spin echo, TR/TE = 360/25 milliseconds) showing an enhancement of intensities of the lesion except for the central portions.

cortical destruction (Unni et al. 1977; Unni 1996a). But FD is occasionally found after a pathologic fracture (Henry 1969; Gibson and Middlemiss 1971), which may radiologically mimic the true cortical destruction by tumor invasion. In the present case, the disruption and partial disappearance of the lateral cortex of the calcaneus were clearly detected by CT scanning. Accordingly, the cortical disruption alone is not a finding sufficient to differentiate these two conditions.

Signal intensity of FD on MR images seem to vary case by case. According to Richardson and Gillespy (1993), the involved marrow has decreased signal intensity on T1 weighted images. On T2 weighted images, calcified or ossified portions have decreased or absent intensity and cystic portions show bright signal intensity. In other reports, signal intensity of the lesion were noted to be low to intermediate on both T1 and T2 weighted images (Casselman et al. 1993; Tajima et al. 1993). As to low-grade central osteosarcoma, only Kurt et al. (1990) mentioned its MR imaging. Without description of the signal intensities, he concluded that MR imaging is helpful to diagnose low-grade central osteosarcoma by detecting evidences of malignant nature: Intramedullary extension, soft tissue

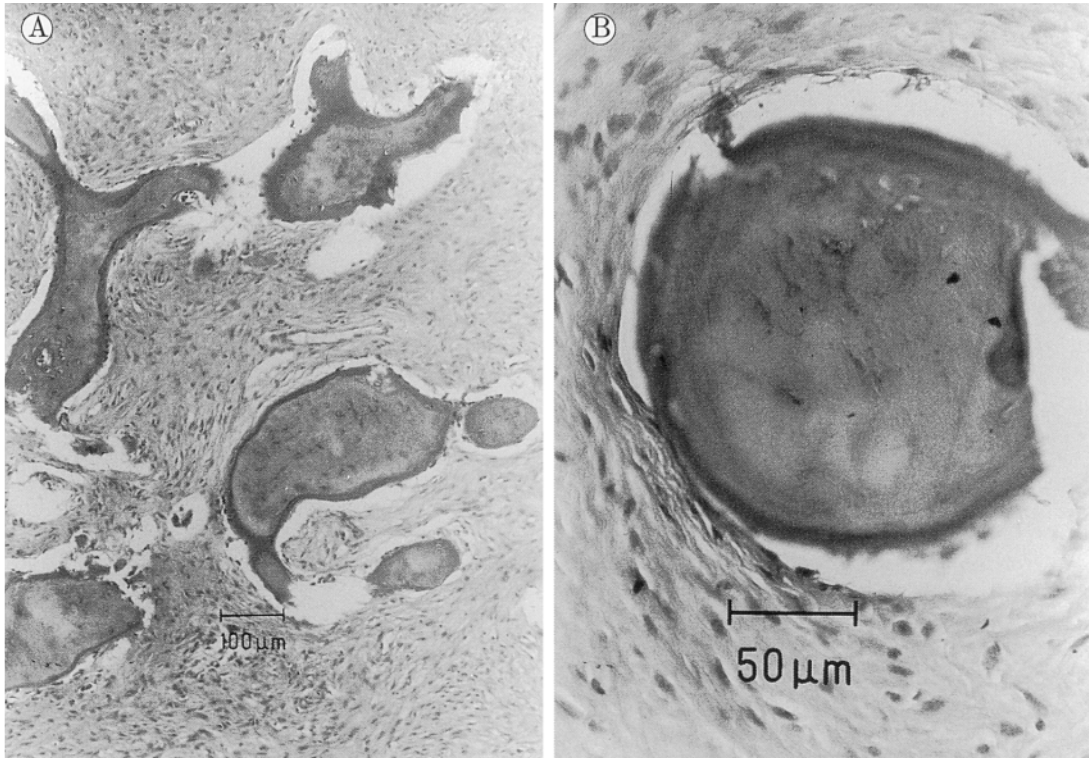


Fig. 4. Photomicrograph of a biopsy specimen showing fibrous tissue with woven bones in an irregular alignment. A, low-power view; B, high-power view.

mass and cortical destruction. In conventional osteosarcoma, tumor osteoid and soft tissue components are darker than normal bone marrow on T1 weighted images. On T2 weighted images, tumor osteoid remains dark, whereas the soft tissue component is brighter (Richardson and Gillespy 1993). The lesion of the present case had iso-intensities on T1 and high intensities on T2 weighted images and its central portions showed lower intensities on both T1 and T2 weighted images. Accordingly, these findings were similar to both those of FD described by Richardson (Richardson and Gillespy 1993) and of conventional osteosarcoma.

Even histologically, differential diagnosis between FD and low-grade central osteosarcoma may sometimes be difficult. Their tissue are composed of spindle cells with irregular woven bone (Unni et al. 1977; Mirra 1989; Kurt et al. 1990; Unni 1996a, b). Though low-grade osteosarcomas have metastatic potential, their spindle cells seldom show nuclear atypia and mitosis. Of Unni's (1997) 27 cases of low-grade central osteosarcoma, four were initially misdiagnosed as FD based on histological findings. The following two features of low-grade central osteosarcoma have been suggested as reliable findings for its differentiation from FD: "the longitudinal stream of woven- (partial) lamellar bone" (Mirra 1989), and the invasive nature of growth (Kurt et al. 1990). Based on the absence of these two features in addition to the absence of nuclear atypia and mitosis, the tumor in the present case was diagnosed as FD.

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