—Case Reports—

Solitary Chondrosarcoma of the Right Ring Finger: A Case Report

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We present a case of solitary chondrosarcoma arising from the proximal phalanx of the ring finger in an elderly man. The chondrosarcoma developed over a period of 14 years, during which the phalanx became progressively more deformed. Several radiographic investigations were carried out, but the patient declined further suggested diagnostic examinations (computed tomography, magnetic resonance imaging, biopsy). Eventually, the lesion became significantly enlarged, and radiographs showed osteolytic lesions in the phalangeal bone. Ray amputation of the finger was required to establish a wide resection of the chondrosarcoma. Most osteochondral tumors arising from the phalanges are benign tumors such as enchondromas, but primary chondrogenic malignant bone tumors (chondrosarcomas) occasionally occur. Chondrosarcoma of the phalanx is difficult to distinguish from enchondroma of the phalanx, because histological investigations of the two neoplasms often produce similar findings. Even with a combination of clinical, biopsy, and imaging findings, differentiating these neoplasms is still challenging, because the characteristic clinical and radiological features of chondrosarcoma do not appear until it becomes aggressive and starts to cause destructive changes. Once that happens, radical expanded resection of the tumor is essential. Therefore, longstanding enchondroma-like lesions should be actively treated in elderly patients, even if a definite diagnosis of chondrosarcoma cannot be made.
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Key words: solitary chondrosarcoma, malignant tumor, finger, hand, sarcoma

Introduction

After osteosarcoma, chondrosarcoma is the most common malignant bone tumor; it most commonly arises in the pelvis, proximal femur, ribs, humerus, tibia, and scapula[1,2], and most frequently affects men in their fifties to seventies[1,2]. Chondrosarcoma of the finger is uncommon, however[1–4]. The first case was reported by Lichtenstein and Jaffe[5] in 1943, and only a limited number have been reported since[2,3,6–10]. We present a case of chondrosarcoma arising from the proximal phalanx of the ring finger in an elderly man. The tumor was initially suspected to be an enchondroma, but when it suddenly expanded eight years after the patient's first visit, a diagnosis of chondrosarcoma was made.

Case Report

An 88-year-old man was referred to our hospital with occasional light prickly pain in his right ring finger that had started after his right hand was struck by a cardboard box six years before his first visit. The patient had no clinical or family history of Ollier disease or Maffucci syndrome, and the symptoms of his presenting complaint had little effect on his daily life. Radiographic examination revealed a multilobate radiolucent region without calcification (**Fig. 1A～C**). The cortical bone was thin, and a ballooning deformity of the proximal phalanx of the ring finger was observed. Enchondroma of the phalanx was suspected, and further imaging examinations (magnetic resonance imaging and computed tomography) and a biopsy were scheduled for differential diag-

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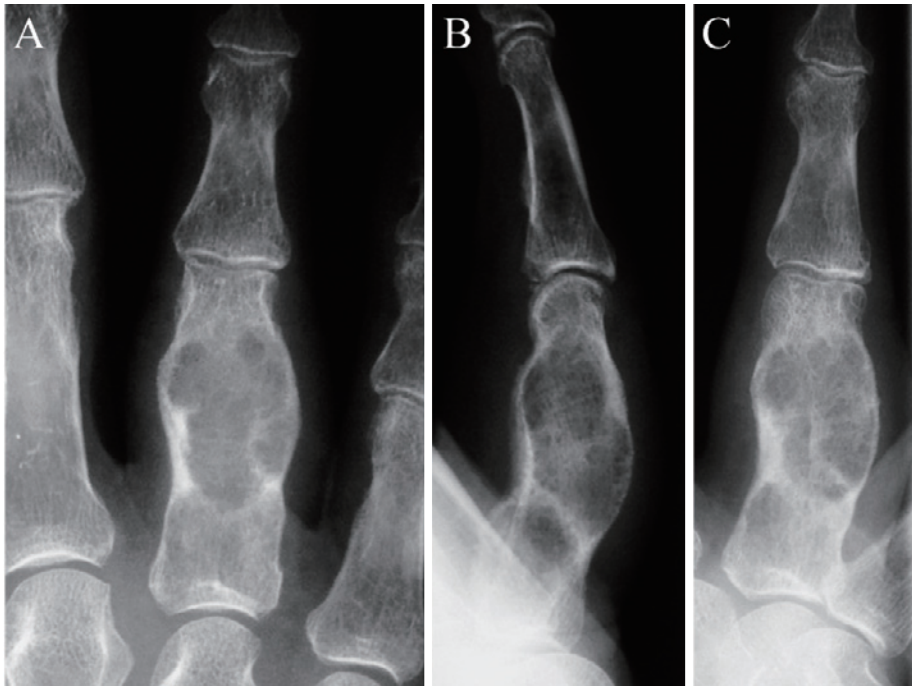

Fig. 1 Radiographs of the right ring finger of a right-handed 88-year-old man, showing multilobate radiolucent region without calcification when he initially visited our hospital.
A: PA view, B: lateral view, C: oblique view

nosis. Against our recommendation, however, the patient canceled these investigations, because he was almost asymptomatic. On a subsequent visit about three years later, radiographic imaging showed a growing radiopaque lesion protruding from the volar cortex of the proximal phalanx of his right ring finger (**Fig. 2A, B**), but again, the patient declined further investigations.

At the age of 96 years, eight years after his initial visit to our hospital, he presented with progressive swelling and pain in his right ring finger. He denied any history of trauma and said he occasionally took painkillers to relieve the pain. Physical examination revealed a firm mass measuring 8 x 7 x 7 cm, and radiography showed a dorsal cortical break of the proximal phalanx of the ring finger with a radiotransparent space-occupying lesion; a periosteal reaction was also observed (**Fig. 3A, B**). Chondrosarcoma of the ring finger was suspected this time, but the patient again declined further examinations and treatment. Over the succeeding year, the finger became inflamed, and the symptoms began to significantly affect the patient's activities of daily life: he could not bear any pressure on his hand because of the constant excruciating pain, which was not significantly relieved by painkillers. He finally requested surgery nine years after his first visit to our hospital.

Examination revealed a hard, bony, fusiform mass measuring 13 x 9 x 9 cm and involving the whole ring finger (**Fig. 4A～C**). Range of flexion was limited, and radiographs showed ballooning associated with the periosteal reaction in the proximal phalanx, along with unusual inflammation of the subcutaneous radiotransparent region (**Fig. 4D**). All routine preoperative laboratory findings were within normal limits, and neither whole-bone radiography nor computed tomography of the lung revealed any other metastatic lesions. No bone biopsy was done, but since malignancy was strongly suspected on the basis of the clinical and radiographic observations, the patient provided written informed consent for primary ray amputation of his ring finger; he also approved the publication of anonymized images in this case report.

Macroscopically, the resected specimen showed an infiltrative whitish tumor with necrosis, and a cavity in the proximal ring finger. The proximal and middle phalanges were unclear because of tumor infiltration. On gross examination, the proximal surgical margin was negative for malignancy, and the distal interphalangeal joint and distal phalanx were preserved (**Fig. 5**).

Histological examination showed that the cartilaginous tumor had a lobular configuration and was divided by fibrous septa (**Fig. 6A**). The tumor entrapped pre-existing bone trabeculae (**Fig. 6B**) and infiltrated the subcutaneous tissue (**Fig. 6C**). On higher magnification, increased

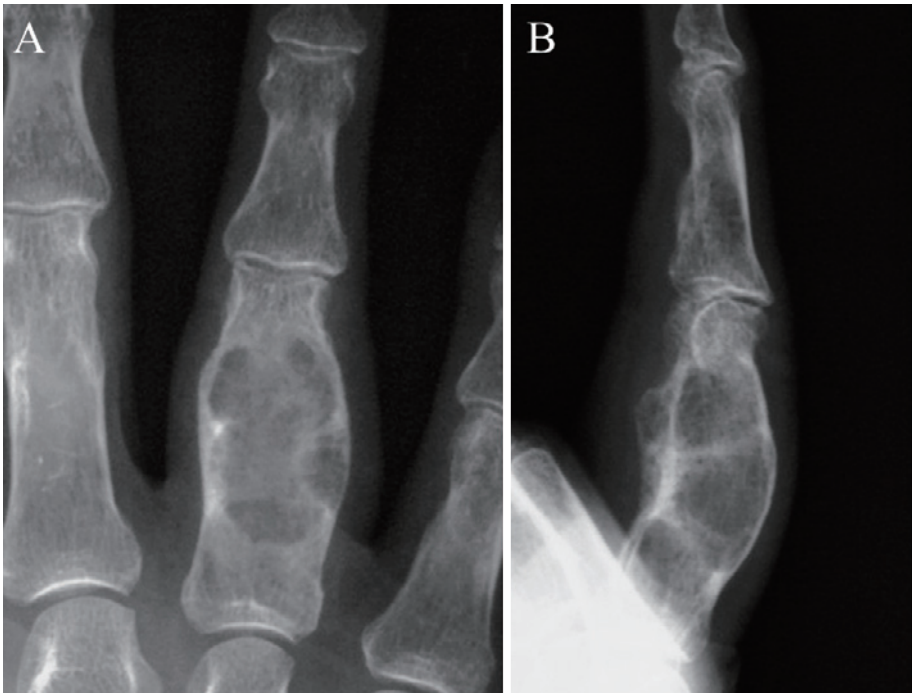

Fig. 2 Radiographs of the patient's right ring finger, showing a multilobate radiolucent region without calcification on his second visit to our hospital.
A: PA view, B: lateral view

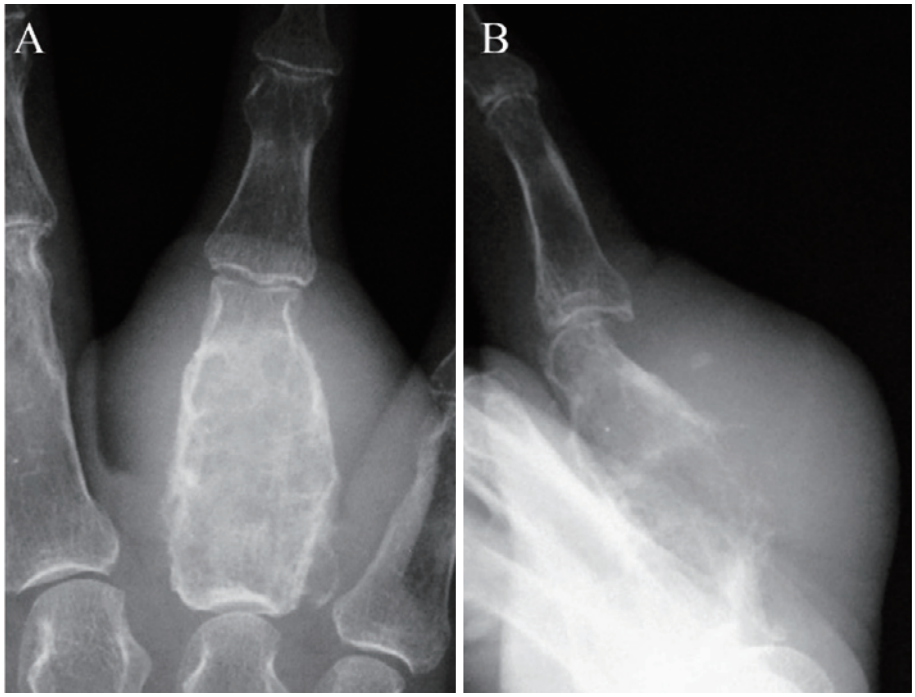

Fig. 3 Radiographs of the patient's right ring finger taken eight years after his first visit, showing a radiotransparent space-occupying lesion with periosteal reaction and a concomitant dorsal cortical break of the proximal phalange of the ring finger.
A: PA view, B: lateral view

numbers of atypical chondroid cells with myxoid matrix changes were observed (**Fig. 6D**). The tumor cells showed nuclear atypia and hyperchromasia, and the presence of mitoses (**Fig. 6D** inset). Although these histological features resembled those of enchondroma, we diagnosed central chondrosarcoma grade 2[11] because of

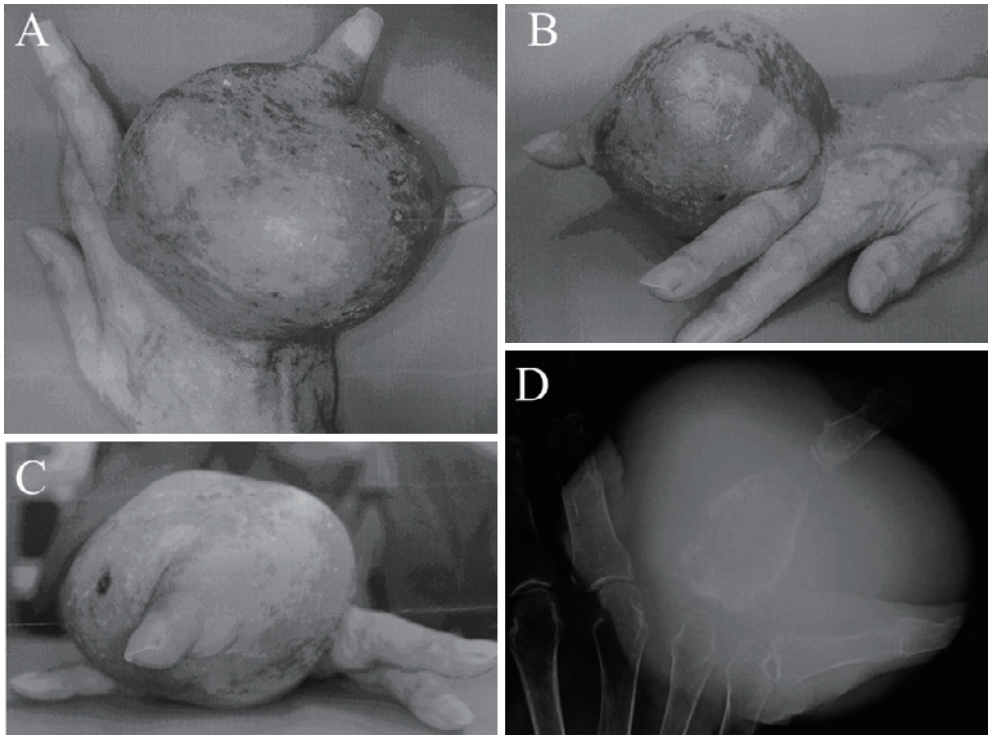

Fig. 4 A ～ C: Preoperative photographs of the patient's right ring finger, showing a hard, bony, fusiform mass measuring 13 × 9 × 9 cm and involving the whole ring finger. D: Radiographs of PA view showing ballooning associated with the periosteal reaction in the proximal phalanx of the right ring finger and unusual inflammation of the subcutaneous radiotransparent region

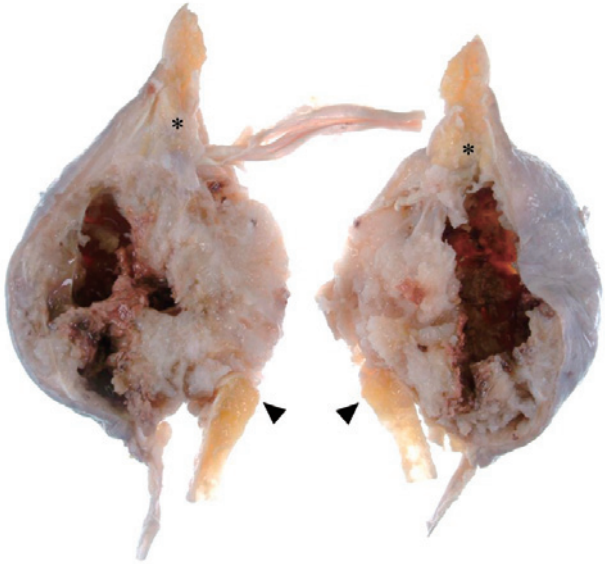

Fig. 5 Macroscopically, the resected specimen showed an infiltrative whitish tumor with necrosis, and a cavity in the proximal ring finger. The proximal and middle phalanges were unclear because of the tumor infiltration. On gross examination, the proximal surgical margin was negative for malignancy, and the distal interphalangeal joint and distal phalanx were preserved. The asterisk indicates the proximal part of the distal phalanx, and the black arrowhead indicates the head of the metacarpal bone.

the tumor's aggressive entrapment of lamellar bone and infiltration into the subcutaneous tissue.

Three months after the digital ray amputation, the patient returned to daily life, and he has since had no residual pain in his right hand.

Discussion

Chondrosarcomas arising in the fingers and hands are uncommon[1–4]: Posch reported only one chondrosarcoma out of 147 hand tumors[3], and Dahlin reported that chondrosarcomas arising from the small bones of the hand accounted for only two of 212 chondrosarcomas[1]. Moreover, Ogose et al. reviewed the files of 893 patients treated for chondrosarcomas at the Mayo Clinic for examples of chondrosarcoma of small bones of the hands and feet, and they found 54 cases of finger chondrosarcoma[4]. In a review of all 994 primary bone and soft tissue tumors treated at our hospital between 2002 and 2012, 170 were in the hands or fingers (fingers: 88, hands: 82). However, there was only one case of chondrosarcoma of the finger, a finding that is consistent with previous reports illustrating the rarity of chondrosarcoma of the finger.

Distinguishing between chondrosarcoma and enchondroma, the most commonly occurring neoplasm in the fingers, is essential to make a definite diagnosis of finger

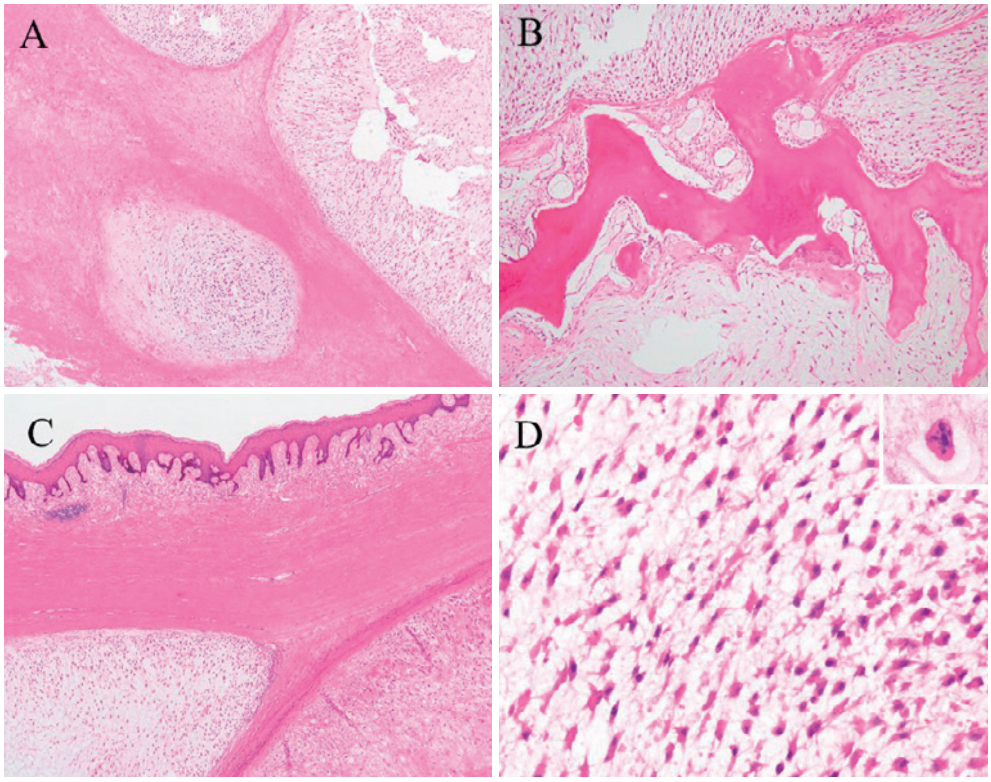

Fig. 6 Histological examination of the resected ring finger. The cartilaginous tumor was divided by fibrous septa with lobular configuration (A). The tumor entrapped pre-existing bone trabeculae (B) and infiltrated the subcutaneous tissue (C). Increased numbers of atypical chondroid cells with myxoid matrix changes were observed (D). The tumor cells showed nuclear atypia and hyperchromasia, and the presence of mitoses (D inset).
A ～ C: original magnification × 1.25, D: original magnification × 20

chondrosarcoma[12,13]. However, the distribution of enchondroma and chondrosarcoma in the hands and fingers is similar[9,12–14], and it is extremely difficult to distinguish histologically between chondrosarcoma and enchondroma in the phalangeal bones[9,12,15,16]. In long tubular bones, increased cellularity, characteristic cell distribution, and myxoid change of the matrix are the features of chondrosarcoma[1], but it is still difficult to distinguish between enchondroma and low-grade chondrosarcoma in phalangeal bones[11,12,17]. Therefore, in addition to histological findings, clinical and radiographic findings are also essential diagnostic indicators[5,11–13,18].

Almost all cases of chondrosarcoma of the hand occur in middle-aged or elderly patients[4,19], but although chondrosarcoma of the fingers often has a long clinical history, it is unclear whether it results from an enchondroma becoming malignant or develops independently over a long period[19]. A few cases of chondrosarcoma of the hand arising from solitary pre-existing enchondromas have been reported[20–24], and several authors have reported cases of chondrosarcoma of the hand with very long clinical histories[19]. Patil et al.[19] reported that in a clinical series of 23 patients with hand chondrosarcoma, the duration of symptoms before the patients sought medical care ranged from 6 weeks to 60 years, and that six of the patients complained of swelling of over ten years' duration (range: 10 to 60 years). It took our patient six years to seek medical care after he started experiencing pain in his finger, and another eight years passed before the tumor became aggressive and destructive.

Some clinical researchers have pointed out differences between finger chondrosarcoma and chondrosarcoma in other skeletal sites[9,10,17,19]. Bovée et al.[10] reported two cases of metastasis in a comprehensive review of 84 patients with chondrosarcomas arising from the phalanges and showed that chondrosarcomas at the phalangeal level of the hand and foot are locally aggressive and have minimal metastatic potential. Referring to previous studies of chondrosarcomas arising from the phalanges, Mankin[17] argued that chondrosarcomas of the fingers could be considered benign compared with chondrosarcomas in other skeletal sites. However, this cannot be said of our patient, in whom amputation of the ring finger was required after inflammation of the finger had been present for more

than ten years without treatment.

It has been reported that the lesion's clinical and radiological appearance (tumor size, cortical bone erosion and dilatation, fracture of the bone cortex, irregular calcification with rapid tumor growth, destruction, periosteal reaction, spicula of the phalanx, and the presence of a lesion extending to the extra-skeletal soft tissue) is useful in differentiating chondrosarcomas from enchondromas[4,8,9,11]. On magnetic resonance imaging, high T2-weighted images are characteristic of cartilaginous tumors, and both chondrosarcomas and enchondromas show low to equal signals on T1-weighted images and high signals on T2-weighted images[25]. However, such radiological findings are often not obvious until the aggressive character of the malignancy is tangible. Furthermore, it is very difficult to distinguish histologically between chondrosarcoma and enchondroma in the phalangeal bones[9,12,15,16]. And even with a combination of clinical, biopsy, and imaging findings[9,13,19], it is still difficult to distinguish chondrosarcomas in their early stages from enchondromas. Once a chondrosarcoma becomes aggressive, radical expanded resection is essential, so longstanding enchondroma-like neoplasms should be actively treated in elderly patients, even if a definite diagnosis of chondrosarcoma cannot be made.

Conclusion

Most osteochondral tumors of the phalanges are benign tumors such as enchondroma, but chondrosarcoma of the phalanges mimicking enchondroma can develop over an extended period. Chondrosarcoma of the phalanx is uncommon and difficult to distinguish from enchondroma of the phalanx, but even when it is impossible to reach a definite diagnosis on the basis of a combination of clinical, biopsy, and imaging findings, longstanding enchondroma-like neoplasms should be actively treated even in elderly patients.

Conflict of Interest: None.

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