



Case report

Synchronous chondroblastomas in the knee joint: A case report

Takao Sakai^a, Hisaki Aiba^{a,*}, Hideo Hattori^b, Shinji Miwa^c, Hideki Murakami^a, Hiroaki Kimura^a

^a Department of Orthopedic Surgery, Nagoya City University, 1, Kawasumi, Mizuho-cho, Mizuho-ku Nagoya, Aichi 467-8601, Japan

^b Department of Pathology and Molecular Diagnostics, Nagoya City University, 1, Kawasumi, Mizuho-cho, Mizuho-ku Nagoya, Aichi 467-8601, Japan

^c Department of Orthopedic Surgery, Kanazawa University, 13-1, Takaramachi, Kanazawa, Ishikawa 920-8641, Japan

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ABSTRACT

Introduction: A chondroblastoma (CB) is a benign chondroid bone tumor that typically arises in growing children. It frequently occurs in the proximal tibia, femur, and humerus. However, the biological behavior of CBs remains unclear because of their rarity. Herein, we report a rare case of synchronous CBs arising on the bilateral sides of the knee joint, suggesting the etiology of chronological tumorigenesis.

Presentation of case: An 18-year-old Japanese man with a complaint of right knee pain was referred to our hospital. Radiography revealed an expanding osteolytic lesion in the right proximal tibia. A small lesion in the distal femur was detected on magnetic resonance imaging. A biopsy of the tibial lesion revealed a pathological diagnosis of CB. Two-stage curettages were performed in the tibia and femur, and the pathological diagnosis of the femoral lesion was CB. No recurrence had occurred for 5 years in the tibia and a year in the femur with stable ambulation and a full range of motion.

Discussion: In our case, the CBs in the proximal tibia and distal femur co-occurred, suggesting that the seed of the CB emerged before the separation of these two bones. The key point to the generation of CB is that its origin could be initiated during mesenchymal condensation before the separation and differentiation of bones as if continental drifts.

Conclusion: We experienced a rare and suggestive case in CB etiology. An experience of synchronous CBs in the consecutive bones was suggestive of CB etiology and careful management.

1. Introduction

Finding the origin of the tumorigenesis is important for developing new diagnostic and treatment strategies. A chondroblastoma (CB) is a benign chondroid bone tumor that typically arises in growing children [1]. CBs frequently occur in the proximal tibia, proximal femur, and humeral epiphysis [1]. CB can occur even in the distal phalanx of a finger [2]. CB rarely metastasizes to the lungs [3]. A histologically proven case of CB with multiple distant soft tissue metastases has been reported as a rare finding [4]. However, reports on CBs are usually case reports and retrospective studies, and the biological behavior of CBs remains unclear because of their rare occurrence in only 1% of primary bone tumors [5]. The recurrence rates of curettage have been reported to be 3.3% to 20% [6–8], particularly in cases with epiphyseal lesions [9]. Herein, we report a rare case of synchronous CBs arising in the bilateral sides of the knee joint, suggesting the etiology of chronological

tumorigenesis. Moreover, we discussed recent genetic findings of CB in relation to tumor generation.

2. Presentation of case

A Japanese 18-year-old male student without any drug history, family history or psychosocial history, complaining of right knee pain during exercise, visited a family clinic and received conservative treatment for a month. Owing to persistent pain, magnetic resonance imaging (MRI) was performed, in which bone tumors in the proximal tibia and distal femur were detected. Therefore, the patient was referred to the university hospital (Nagoya-city, Aichi, Japan).

Physical examination revealed swelling in the right knee, tenderness of the proximal tibia, and restricted range of motion of the knee with painful claudication. Blood tests revealed normal complete blood counts and serum chemistry of liver and kidney functions.

* Corresponding author.

E-mail address: h-aiba@med.nagoya-cu.ac.jp (H. Aiba).

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An X-radiography revealed a gradually expanding osteolytic lesion in the right proximal tibia (Fig. 1). The lesion in the proximal tibia demonstrated a low T1-weighted image (WI) (Fig. 2a) and high T2-WI (Fig. 2b), with fluid-fluid level suggesting aneurysmal bone cyst change with a maximum diameter of 50 mm on MRI. Moreover, another small solid lesion with maximum diameter of 6 mm in the distal femur on the same side with the same intensity was observed on MRI. During this period, the femoral lesion was considered too small to be a neoplasm. Chest radiography revealed no mass lesions in the lungs.

Fine needle biopsy of the tibial lesion revealed monomorphic neoplastic proliferation of mononuclear cells and scattered giant cells in the chondroid-like tissues possessing chicken-wire calcification with aneurysmal bone cysts change, which was compatible with CB (Fig. 3a).

This patient had no contraindications for surgery, and the anesthesiologists in charge considered tolerance for the subsequent interventions with echocardiogram, electrocardiogram and tidal volume of the mechanical ventilator. As a definitive treatment of the tibial lesion, a thorough sharp curettage, with the immersion of the tumoral cavity with an ethanol adjuvant as a local adjuvant, was performed by an experienced certified orthopedic surgeon (SM). Post-operation, no restrictions on weight-bearing were advised. Four years after the tibial surgery, an unexpected growth of the lesion in the right distal femur (Fig. 2c) was revealed without pain sensation or altered range of motion. An experienced certified orthopedic surgeon (HA) performed an additional sharp curettage with ethanol adjuvant. Pathological diagnosis of the femoral lesion was confirmed as CB, similar to that of the tibia. (Fig. 3b) Although the femoral lesion had no aneurysmal bone cyst changes, the accompanying tumor cells had the same findings in the femoral lesion, such as the sheets of proliferated mononuclear cells along with scattered giant cells. No recurrence occurred after five years in the tibia and a year in the femur with stable ambulation without any post-operative comorbidity. This case report is presented based on the Surgical Case Report (SCARE) Guideline [10].

3. Discussion

Here, we report a rare case of synchronous CB arising in successive bones. Based on the identical pathological images, it was assumed that tibial and femoral lesions co-occurred. Although distinguishing bone metastasis or ectopic generations in different bones is difficult, we inferred that the latter might be possible. This is because Korosh et al. have reported a case of pathologically proven multiple soft tissue metastases of CB, suggesting that the main sites of metastasis are the lungs and soft tissues and that metastasis of CB in the bones is an extremely



Fig. 1. The X-radiography image (lateral view) revealed a transparent osteogenic tumor in the anterior proximal epiphysis of the tibia. No femoral lesions were observed.



Fig. 2. Preoperative magnetic resonance imaging revealed a tumor (46 mm × 32 mm; lateral view) with (a) low intensity on T1-weighted image (WI) (b) high intensity on T2-WI, suggesting a change in the aneurysmal bone cyst of the tibia (arrowhead). A small round lesion with low intensity on both T1-WI and T2-WI was also included in the epiphysis of the femur (arrow). (c) The femoral lesion increased in size four years after the follow-up (arrow).

rare event [4]. As the origin of CB was determined to be the primitive cartilage cell [11], a CB may be generated as “seeds” before the generation of two continuous bones and division into different bones, which can be comparable to the famous theory “plate tectonic theory” in geology [12].

Similarly, Miwa et al. analyzed patients with enchondromas in the digital bones. They found that the lesions successively occurred in the

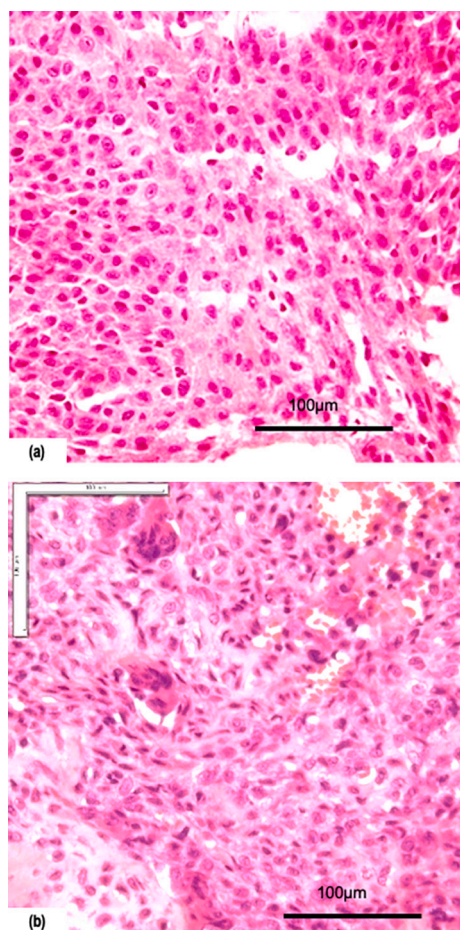


Fig. 3. For the open biopsy, specimens were collected from the proximal tibia. (a) These specimens consisted of monomorphous neoplastic proliferation of mononuclear cells and scattered giant cells in the chondroid-like tissues possessing chicken-wire calcification. Specimens collected from the distal femur (b) showed pathological similarity to the former resected lesions.

same digital ray and hypothesized that the occurrence of the lesions preceded the separation of the phalanges during the period of organogenesis [13]. Likewise, Fukunaga et al. have reported a case of CBs involving multiple tarsal bones in a middle-aged woman [14]. From the histological findings, no malignant features were observed, suggesting that metastasis of the multiple lesions may be unlikely and that the lesions rather arose during the bone formation. These cases provided astute ideas on the origin of the tumor and suggested a similar etiology in this case.

Expression of the sex-determining region Y-box 9 gene was shown in all CBs proven in a study by Dancer et al. [15] This gene was related to mesenchymal condensation of chondroblasts and osteoblasts. Mesenchymal condensation is a gathering of mesenchymal pluripotent cells, leading to differentiation into chondroblasts and osteoblasts. Also, the involvement of the sex-determining region Y-box 9 gene in the early phases of cartilage differentiation has been suggested [15]. These findings supported our hypothesis that CB was generated in mesenchymal condensation and could have been separated into the femur and tibia.

In the pediatric population, the CBs were predominantly located in the epiphysis of the long tubular bones [9]. Meanwhile, Andrea et al. have reported that CB in patients aged >30 years was predominantly located in the small bones, such as the talus, calcaneus, acromion, cuneiform, and metatarsals [16]. Fink et al. have reported cases of CB in the foot and found that the mean age of patients was 25.5 years, which was different from that population of patients with CBs in the long bones [17]. These epidemiological findings of CB locations suggest that the

epiphyses of long bones have a higher possibility of recruiting CB cells during mesenchymal condensation, which accelerates the process of tumor development faster than that seen in small bones.

Because CB is a rare bone tumor, identifying the risk of recurrence is difficult, although we have some clues. Suneja et al. have reported that young patients had a high risk of recurrence after curettage [7]. Saihan et al. have reported that epiphyseal CBs, especially in pediatric patients with an open epiphyseal line, had a higher risk of recurrence than CBs in other parts of the bones because the growth plate, being close to an epiphysis can make complete removal of CBs difficult [9]. We should carefully treat the CB around the epiphysis because, although cases are rare, the formation of multiple synchronous CB occurs.

4. Conclusion

Although CBs are presumed to originate from mesenchymal condensation in the fetal period, researching this topic may be difficult because of its rarity. The synchronous CBs in the adjacent joints might be a clue to elucidating the etiology of CBs. When treating CBs, we should pay attention to the consecutive bones.

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Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the ethical committee of the Nagoya City University Hospital and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Case reports, however, are exempt from the approval in our institution.

Consent

Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available on request for review by the Editor-in-Chief of this journal.

Research registration

Not applicable.

Guarantor

Hisaki Aiba.

CRediT authorship contribution statement

Conceptualization, TS, HA, HM; Data curation; HH, Formal analysis, HH; Funding acquisition, HA, HM; Investigation, HA, HH; Methodology, HA, SM; Project administration, TS, HA, HK; Resources, HM; Software, not applicable; Supervision, HM; Validation, HA, HK; Visualization, HH; Roles/Writing—original draft; TS, HA; Writing—review & editing, TS, HA, HK.

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Declaration of competing interest

There were no conflicts of interest.

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