Case Report

Insufficiency fracture at an osteochondroma bridging the proximal fibula and the tibia: Case report

Akio Sakamoto*, Takeshi Okamoto, Shuichi Matsuda

Department of Orthopaedic Surgery, Graduate School of Medicine, Kyoto University, Shogoin, Kawahara-cho 54, Sakyo-ku, Kyoto 606-8507, Japan

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ABSTRACT

Osteochondromas are benign bone tumors that protrude from underlying bone. A fracture of an osteochondroma is rare, and no insufficiency fractures of an osteochondroma have been reported. A 57-year-old female complained of pain in her proximal leg that increased during walking. A computed tomographic image depicted bridging that had extended to the tibia. An incomplete fracture line was observed at the osteochondroma. The osteochondroma was resected to remove the bridge. The preoperative symptoms disappeared after surgery. It was challenging to diagnose an osteochondroma with an insufficiency fracture because the symptoms varied in accordance with daily activity.

1. Introduction

Osteochondromas are one of the most common benign tumors of bone and are characterized by an exophytic lesion comprising cortical and medullary bone with overlying hyaline cartilaginous tissue.¹ Two clinical presentations exist: one is a solitary osteochondroma characterized by a solitary lesion and the other is hereditary multiple exostoses denoted by multiple lesions.²

The appearance of symptoms depends on the site and the size of the osteochondroma. Mechanical effects of a neoplastic protuberance or bone deformities on adjacent tendons and ligaments can cause pain and restricted joint motion.¹ Painful symptoms usually occur in young patients; 75–80% of osteochondroma cases are detected based on reports of such symptoms before the age of 20 years.^{1,3,4} A fracture through the stalk of an osteochondroma is a rare complication.⁵

Stress fractures occur when the local load on bone exceeds its mechanical resistance.⁶ Stress fractures include fatigue fractures and insufficiency fractures. Fatigue fractures occur when an abnormal mechanical stress is applied to a normal bone, whereas insufficiency fractures occur when moderate or normal stress is applied to a bone that has inadequate resistance.⁶ One of the risk factors for an insufficiency fracture is postmenopausal osteoporosis.^{6,7} In the current report, a 57-year-old female with an insufficiency fracture at an osteochondroma is described.

2. Case presentation

A 57-year-old woman presented with pain on the medial aspect of

the proximal leg, which she had noticed for 6 months. The pain appeared during walking. The woman was 157 cm tall and weighed 46.3 kg. Her osteochondroma was diagnosed when she was 10 years old. There was no medical history related to bone health, steroid usage, diabetic mellitus, and/or endocrine disorders. There was also no history of trauma or injury, and she had not been examined for osteoporosis. On physical examination, the osteochondroma was not palpable on the bone surface. No tenderness over the tibia or fibula was reported. Range of motion at both knee joints was equivalent and normal.

Plain radiographs showed a lesion protruding from the fibula with continuity to the tibia (Fig. 1A). Three-dimensional reconstructed computed tomography (CT) images depicted the lesion bridging the proximal fibula and tibia (Fig. 1B) and showed an incomplete fracture line as well as an osteosclerotic line at the osteochondroma (Fig. 1C). The fracture line appeared along the outline of the lateral aspect of the tibia. Magnetic resonance imaging (MRI) depicted a protuberant bone lesion with continuity to normal bone. However, an overlying cartilaginous cap was not apparent. The MRI showed a band of low-signal intensity on T1- and T2-weighted images; corresponding to an area of osteosclerosis observed on CT images. Vague high-signal intensity was evident around the osteochondroma at the fracture site on the short tau inversion recovery (STIR; Fig. 1D).

After being diagnosed with an insufficiency fracture of the osteochondroma, the patient was followed-up within one month and reported consistent pain that did not decrease. Therefore, the osteochondroma was resected under general anesthesia. The lesion was reached by retracting the tibialis anterior muscle and the bridging site of the osteochondroma with the insufficiency fracture was resected. The

* Corresponding author at: Department of Orthopaedic Surgery, Graduate School of Medicine, Kyoto University, Shogoin, Kawahara-cho 54, Sakyo-ku, Kyoto 606-8507, Japan.
E-mail address: akiosaka@kuhp.kyoto-u.ac.jp (A. Sakamoto).

Fig. 1. A 57-year-old female with an osteochondroma. Plain radiographs show a lesion protruding from the proximal fibula extending toward the tibia (A, upper and lower panels). Three-dimensional CT shows that the lesion originating from the proximal fibula was forming a bridge with an osteochondroma (B, upper and lower panels). CTs (coronal view) depict an osteosclerotic line (yellow arrow) at the osteochondroma (C, upper panel). Note that the fracture line is not visible on the proximal axial section (B, middle panel), but can be observed at the distal section (yellow arrow; C, lower panel). MRIs showed low-signal intensity on T1-weighted images corresponding to calcification (D, upper panel). Vague high-signal intensity on the STIR image was observed (white arrow; D, lower panel).

Fig. 2. Plain radiographs taken 6 months after resection of the osteochondroma (A, upper and lower panels). Histopathology showed endochondral ossification of lamellar bone at the fracture site (B).

mobility of the fibulotibial joint was confirmed perioperatively. Because the tip of the osteochondroma was connected to bone, it was left in situ (Fig. 2A). Histologically, the specimen collected from the bridging site showed partially sclerotic lamellar bone and trabeculae undergoing endochondral ossification, compatible with the bone healing process of a fracture (Fig. 2B). Full weightbearing was allowed postoperatively. The preoperative pain disappeared after the operation. However, the patient reported vague pain in the proximal region of the lower leg. On physical examination, the pain reported at the proximal tibiofibular joint was not evident 3 months after the operation.

3. Discussion

An insufficiency fracture at the site of an osteochondroma has not been reported previously, although a fracture through the stalk of an osteochondroma has been reported as a rare complication.^{5,8–10} The cause of a stalk fracture at an osteochondroma occurs as a direct consequence of an impingement against the surrounding tissues and a direct or indirect injury as a result of strong muscle contraction.^{10–13} Reported cases have been localized to the knee joint.¹³ Spontaneous healing of an osteochondroma fracture at the stalk has been reported.⁹ However, surgery is recommended when symptoms continue after conservative management,^{10,13,14} and surgical resection reportedly provides a more rapid recovery and return to normal activity.¹³

The diagnosis of an insufficiency fracture is difficult and can easily be overlooked because plain radiographs are not accurate enough to detect the subtle changes in bone during the early phase.^{15,16} It can take 2 to 4 weeks after an incident to be able to detect changes, such as osteosclerosis, a periosteal reaction, a cortical radiolucent line, or a bone callus via radiography.¹⁷ CT is also used for the diagnosis of an insufficiency fracture. It is useful for identifying the fracture lines of insufficiency fractures in the late stages, although CT is not useful in the early stage.⁶ In our case, CT scans depicted an osteosclerotic line at the osteochondroma, suggesting a late stage insufficiency fracture.

MRI is also useful in the diagnosis of insufficiency fractures because it can detect abnormalities, even at an early stage. MRIs show diffuse bone marrow edema via low-signal intensity on T1-weighted images and high-signal intensity on T2-weighted images.^{15,18–20} In the current case, the fracture line was evident as a band of low-signal intensity on the MRI, both on T1- and T2-weighted images, corresponding to calcium deposition. Bone marrow edema at the osteochondroma was not clearly visible on T1- and T2-weighted images. However, a vague high-signal intensity band was observed on the STIR image, which is much more fluid sensitive. This signal abnormality supported the diagnosis of an insufficiency fracture.

Insufficiency fracture at an osteochondroma occurs in the absence of a traumatic trigger. Insufficiency fractures are most commonly sustained by elderly women with postmenopausal osteoporosis. In a previous study of 60 cases with 91 insufficiency fractures, females outnumbered males 11–1.⁶ The average age of the females among those case studies was 72.5 years.⁶ Our patient was a 57-year old female but a bone mineral density examination for the diagnosis of osteoporosis had not been performed. Postmenopausal status may have been a cause of the insufficiency fracture at the osteochondroma.

Rest or avoidance of weight bearing for 4–6 weeks is indicated for the treatment of insufficiency fractures.²¹ However, insufficiency fractures of the tibial shaft or the femoral neck could progress to a complete fracture. Surgical procedures can be a treatment option for a delayed union or nonunion.¹⁵ In the current case, the CT finding suggested a late stage fracture. Furthermore, the fracture line was located longitudinally at the osteochondroma, and may have been the result of shared stress. Spontaneous bone union was not expected, and resection seemed to be the appropriate treatment.

4. Conclusion

The diagnosis of an insufficiency fracture at an osteochondroma bridging two bones was supported by the disappearance of preoperative symptoms following resection and the emergence of pain at the tibiofibular joint postoperatively. An insufficiency fracture may be associated with postmenopausal osteoporosis. It was challenging to diagnose this osteochondroma with an insufficiency fracture because the degree of associated pain at the osteochondroma changed in relation to level of activity.

Informed consent

Informed consent was obtained from the individual in the case report.

Authors' contributions

Akio Sakamoto – Performed operation, data collection and analysis, manuscript writing.

Takeshi Okamoto – data collection and analysis, supervised development of work.

Shuichi Matsuda –Supervised development of work.

Conflicts of interest

None.

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