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Curettage and external fixation for displaced pathological fracture through non-ossifying fibroma in the radius: A case report☆

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ABSTRACT

Non-ossifying fibromas are extremely rare in the upper extremity, such as those in the radius and humerus. The treatment of completely displaced fractures following non-ossifying fibromas in the radius has not been reported. We present the case of a pathological fracture caused by a non-ossifying fibroma in the radius treated using external fixation. The patient was a 10-year-old girl who presented to our hospital with right forearm pain after an accidental fall. She had no remarkable medical history. Radiographs and computed tomography showed a pathological fracture through osteolytic lesions with sclerotic rims in the diaphysis of the radius, suggesting a pathological fracture through a non-ossifying fibroma in the radius. We performed tumor curettage and external fixation due to marked fracture displacement. Histological findings were compatible with those of non-ossifying fibroma. Six months post-surgery, there were no limitations in the range of motion of supination and pronation of the forearm, and radiographs confirmed a bone union. Although non-ossifying fibromas in the radius are rare, cases with large lesions can cause pathological fractures after minor trauma. This case suggests that curettage and external fixation are beneficial, especially if early surgery is required due to unacceptable displacement and when there is no time for a biopsy to rule out malignancy.

Introduction

Non-ossifying fibromas (NOFs) are benign lesions frequently located in the metaphysis of the lower extremities of children and adolescents [1–3]. Most NOF lesions spontaneously resolve with growth [4]; however, large lesions can cause fractures. Most cases of pathological fractures through NOFs have been reported in the lower extremities, with fewer cases in the upper extremities, such as those in the radius or humerus [1–3].

The treatment of NOF-induced pathological fractures includes immobilization with casts and curettage with or without bone graft after the bone union is achieved when the displacement is acceptable [3]. However, when the fracture is unstable and cannot be acceptably aligned by closed means, early open reduction and fracture fixation are considered necessary in addition to curettage [3]. To our knowledge, no reports exist on treating completely displaced fractures following NOFs in the radius.

Here, we present a case of a pathological fracture caused by a NOF in the radius, suggesting that curettage and external fixation are beneficial for early surgery for displaced pathological fractures while minimizing the extent of contamination by the tumor.

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Fig. 1. Radiographs showing a pathological fracture through osteolytic lesions with sclerotic rims in the diaphysis of the right radius.

Fig. 2. Computed tomography of the right forearm shows polycystic translucent lesions with thinning of the cortex and a pathological fracture through the middle of the tumor lesion.

Case presentation

A 10-year-old girl presented to our hospital with a complaint of right forearm pain after being caught between the floor and a friend who fell upon her. She had no remarkable medical history or previous symptoms at the fracture site.

On physical examination, deformation of the right forearm was noted, but there were no signs of neurovascular injury. Radiographs revealed a pathological fracture through osteolytic lesions with sclerotic rims in the diaphysis of the right radius (Fig. 1). Computed tomography (CT) showed polycystic translucent lesions of 33.5 mm in a longitudinal diameter with thinning of the cortex in the diaphysis of her right radius and a transverse fracture in the middle of the tumor lesion (Fig. 2). The differential diseases, in this case, include NOF, fibrous dysplasia, simple bone cyst, aneurysmal bone cyst, chondromyxoid fibroma, osteomyelitis, and tuberculosis. The characteristic radiological findings of NOF, such as the eccentric position of the lesions, the osteolytic lesions with marked sclerotic margin, and the orientation in the long axis of bone, led us to strongly suspect NOF rather than fibrous dysplasia, solitary bone cyst, aneurysmal bone cyst, chondromyxoid fibroma [1]. Osteomyelitis and tuberculosis were ruled out in this case based on clinical and laboratory findings. We scheduled curettage of the tumor and osteosynthesis due to the lack of evidence suggesting malignancy.

The operation was performed under general anesthesia. First, a 4-cm skin incision was made longitudinally above the fracture site.

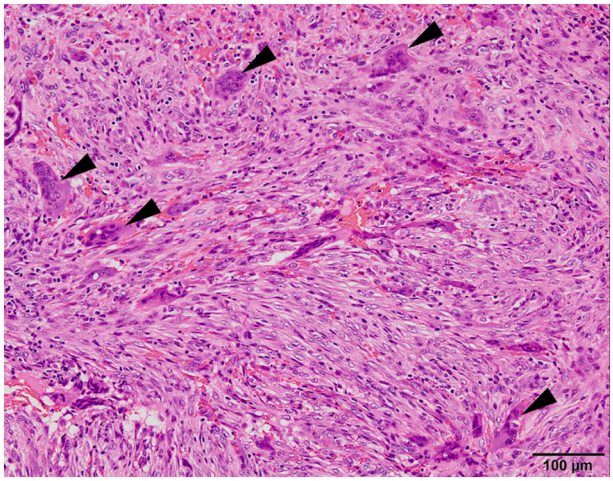

Fig. 3. Histopathological study
A photomicrograph of the surgical specimen in hematoxylin and eosin staining showed proliferative spindle-shaped cells in a convoluted manner. Multinucleated osteoclast-type giant cells (arrow heads) are also observed.

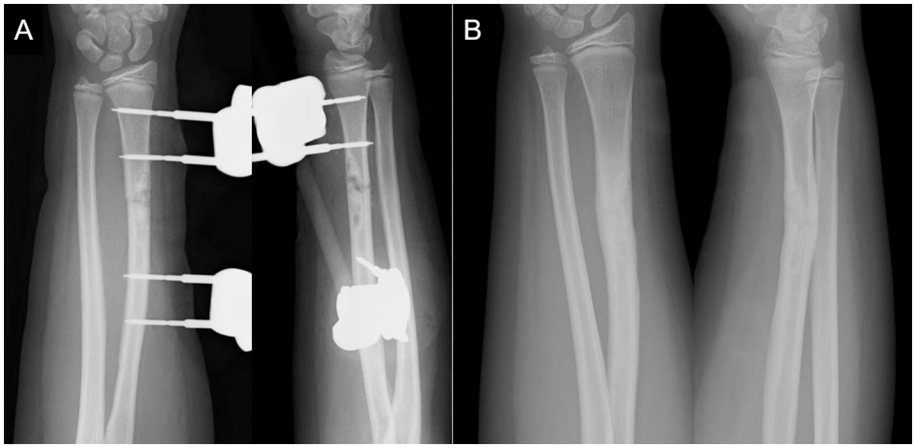

Fig. 4. Radiographs immediately after surgery (a). Radiographs at 6 months after surgery showing bone union (b).

Using Henry's anterior approach, the fracture site was identified by retracting the flexor pollicis longus muscle belly to the ulnar side. Tumor tissue was scraped piece by piece using a sharp curette under fluoroscopy. After tumor curettage, cavities were filled with β-tricalcium phosphate granules. Subsequently, two 2.5-mm half-pins were introduced into each distal and proximal bone fragment at a 45° angle dorsal to the frontal plane, and the pins were fastened with the fracture reduced.

Histological findings were compatible with NOF, showing proliferated spindle-shaped cells with oval nuclei and presenting a storiform pattern. Multinucleated osteoclast-type giant cells were also observed. There was no evidence of malignancy (Fig. 3).

Radiographs obtained immediately after the surgery are shown in Fig. 4a. The postoperative course was uneventful. We removed the external fixation after 5 weeks and applied a long-arm cast for 2 weeks. Six months postoperatively, there were no limitations in the range of motion of forearm supination and pronation, and radiographs confirmed bone union (Fig. 4b).

Discussion

This case presents two clinical issues. First, a NOF occurring in the radius is rare; however, it can cause pathological fractures following minor trauma, as in this case. Since a NOF is frequently located in the metaphysis of the distal femur and proximal and distal tibia [1], most pathological fractures caused by NOF have been reported to occur in the lower extremities, with only four cases of pathological fractures reported in the radius (Supplementary Table 1). This case is also atypical in that the lesion was located in the diaphysis, in addition to its origin in the upper extremity. NOF usually occurs in the metaphysis of long bones. However, as they grow, the lesions are thought to migrate proximally toward the diaphysis, away from the origin [5]. The location of the tumor in the diaphysis

suggests that the tumor may have migrated over the course of remodeling in this case. In addition, two of the four previously reported cases of NOF in the distal radius were located in the diaphysis and moved more proximally over the course of treatment, suggesting that this may be a characteristic imaging finding of NOF occurring in the distal radius.

To date, risk factors for pathological fractures through NOFs in the lower extremities have been identified as inhibiting >50 % cortical involvement and height measurement of >33 mm^3, and Ritschl's classification [6] of stage B lesions [7] (variable distance from the physis with thin sclerotic borders). However, NOFs in the upper extremities, such as those in the radius and humerus, have more aggressive biological features regarding thinning and expansion at the adjacent cortex, rapid increase in size, and slow ossification than those in the lower extremities [8]. In this case, although there were no radiographic images before the injury, CT taken at the time of injury showed a large lesion occupying approximately 90 % of the radius in the transverse plane with thinning at the cortex, similar to the radiographic findings in the previous literature [8]. Also, pathological fractures in the radius caused by NOFs have a high rate of large osteolytic lesions remaining at the fracture site even after fracture healing, resulting in subsequent refractures [1,8,9]. Therefore, careful follow-up is needed to prevent pathological fractures, especially when NOF in the radius is identified incidentally.

Second, this case demonstrates that curettage and external fixation can be treatment options for pathological fractures of the radius that require early surgery due to fracture displacement. Treatment of pathological fractures through NOFs in the radius includes a long arm cast [2] or curettage with a bone graft or artificial bone substitute [1,6]. Unlike previous reports [1,2,8,9], this case required open reduction and fixation in addition to curettage as the fracture was unstable and could not be acceptably aligned by closed means. Curettage and external fixation for benign bone tumors, including NOFs, have the advantages of treating deformity, limb-length discrepancy, and bone defects produced by benign bone tumors, and satisfactory outcomes have been reported [10]. In this case, other fracture fixation procedures include plate fixation and intramedullary fixation using Kirschner wires. However, external fixation has the advantage over these surgical procedures in that, if the tumor is malignant, the contamination extent of the tumor is minimized. In addition, in this case, the tumor had spread in the longitudinal direction of the radius; therefore, plate fixation would have required a long plate, which would have resulted in a long skin incision and greater invasiveness. Intramedullary wire fixation may not have provided firm fixation of the fracture against rotation due to the large bone defect of the tumor after curettage. Nonetheless, external fixation also has some disadvantages, such as pin-track infection and the need for repeated frame adjustment. Therefore, we removed the external fixation relatively early in the postoperative period (at 5 weeks postoperatively when bone bridging appeared) and changed the long arm cast for 2 weeks in this case.

Conclusions

This case provides new information on treating pathological fractures of the radius through NOFs. Although NOFs occurring in the radius are rare, cases with large lesions, such as in this case, can cause pathological fractures after minor trauma. This case suggests that curettage and external fixation are beneficial, especially in cases that require early surgery due to unacceptable displacement and when there is no time to perform a biopsy to rule out malignancy.

Supplementary data to this article can be found online at https://doi.org/10.1016/j.tcr.2023.100895.

Ethics approval and consent to participate

Consent to participate is not applicable in this type of study. A statement of the ethics committee was not required from this anonymized case report in accordance with the legislation of the Institutional Review Committee of our institution.

Consent for publication

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

Availability of data and materials

Data that support the findings of this study are available from the corresponding author on reasonable request.

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CRediT authorship contribution statement

Sakura Yamaguchi: Data curation, Writing-Original draft preparation, Investigation. **Ryogo Furuhata:** Writing-Original draft preparation, Conceptualization, Methodology. **Atsushi Tanji:** Supervision, Validation. **Tadahisa Urabe:** Writing- Reviewing and Editing.

Declaration of competing interest

The authors (Sakura Yamaguchi, Ryogo Furuhata, Atsushi Tanji, Tadahisa Urabe) have no interests to declare.

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