Case Report

Fibro-osseous pseudotumor of the digit: a case report

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Abstract: Fibro-osseous pseudotumor is a poorly reported benign ossifying tumor. Due to its aggressiveness and lack of specificity, the lesion has previously been mistaken for a malignant lesion, leading to unnecessary radical treatment. Our case warns our readers of the aggressiveness of the tumor and rational surgical planning. In our case, a 25-year-old male patient presented with a painless, enlarging mass in the left index finger that had developed over the course of 5 months. The lesion was first partially surgically removed for biopsy, which confirmed the lesion to be fibro-osseous pseudotumor. Considering the possibility of skin necrosis from complete excision, complete removal was postponed until the second surgery. However, the residual lesion rapidly progressed, reaching its original size within 4 months. Another lesionectomy was performed to thoroughly remove the recurrent lesion surrounding the joint capsules. The intraoperative frozen section again supported the initial diagnosis and recurrence. During the 2-year follow-up, there were no signs of recurrence, and the function of the finger was fully recovered. Fibro-osseous pseudotumor should be considered in the differential diagnosis of rapidly progressive lesions affecting the digits. Complete surgical excision is the treatment of choice. However, the surgical strategy should be cautiously planned because of the aggressiveness of fibro-osseous pseudotumor and the possibility of saving the involved digit.

Keywords: Soft tissue neoplasms; differential diagnosis; fibro-osseous pseudotumor of the digits; tumor recurrence; case report

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Introduction

Fibro-osseous pseudotumor is a poorly reported benign ossifying tumor of the soft tissue that generally affects the dermis and subcutis of the extremities (1). Due to its aggressive nature and lack of specific signs or symptoms, the lesion can be misdiagnosed as a malignant lesion, leading to unnecessary radical treatment (2). It is commonly seen in young adults and is thought to be associated with a history of trauma (3). Thus, good understanding of its clinical and histopathological characteristics spares inappropriate aggressive surgical treatment due to misdiagnosis.

Here, we have reported a case of fibro-osseous pseudotumor involving the left index finger that aggressively recurred after the first surgical treatment. We believe that this case is of great reference value to fellow surgeons with regard to planning surgical treatment strategies. We present the following case in accordance with the CARE reporting checklist (available at https://dx.doi.org/10.21037/tcr-21-333).

Case presentation

A 25-year-old male patient presented with a painless, enlarging mass in the left index finger that had developed over the course of 5 months. The lesion gradually developed without any concurrent symptoms, except clumsiness owing to the presence of the lump. The patient did not have any history of related trauma, was previously fit and healthy, and had not received intervention prior to the first visit. The patient was mentally healthy, and no family history could

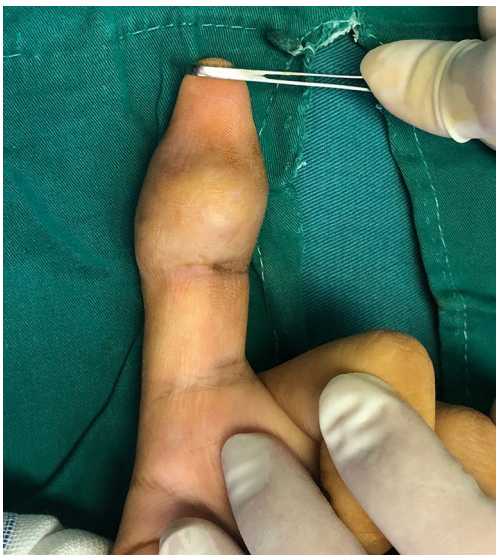

Figure 1 Photograph of the lesion before surgery.

be traced. At presentation, the lesion, involving the palmar and lateral aspect of the left index finger, was firm and fixed without any discharge of pus or mucus (*Figure 1*). X-ray revealed abnormal bone density surrounding the swollen soft tissue in the middle phalanx of the left hand. Enhanced magnetic resonance imaging indicated abnormal signals around the flexor tendon of the left index phalanx; hence, inflammatory lesions were initially considered (*Figure 2*). The results of other tests, including routine blood and urine analysis and tests for tumor markers, were normal.

Based on aggressive features of the tumor, we needed to obtain histopathological diagnosis. We decided to perform partial excisional biopsy to limit wound complications and decrease tumor burden. In the first stage, we performed a Z-shaped incision on the ventral side and discovered the subcutaneous diffuse tumor tissue that was wrapped around the flexor tendon sheath and neurovascular bundle. We carefully dissected around the neurovascular bundle, removed most of the mass and part of the tendon sheath, and excised the skin that infiltrated the dermis layer. The sample was sent for histopathological examination, which revealed spindle fibroblast-like cells and osteoid formation; spindle cells were densely arranged, mild in histology, and uniform in size. Further, nuclei divisions were rare, interstitial blood vessels were abundant, and osseous trabeculae demonstrated a complete zoning phenomenon (*Figure 3*). The final diagnosis was fibro-osseous pseudotumor (1).

Although we asked the patient to visit the clinic regularly, he did not present until 4 months later with a lump of almost the same size as the previous lesion on the radial side of the left index finger, which resembled the manifestation of the previous lesion. A sigmoid incision was carefully designed on the lateral side (*Figure 4*) and the mass was completely removed along with the proximal and distal joint capsules, ensuring the blood supply of the related flap. Since the adhesion occurred around the affected tendon, releasing the adhesion in the second excision surgery could not only improve the related joint function, but also make instant postoperative rehabilitation exercises possible. Once again, histopathological examination confirmed that the recurrent lesion was a fibro-osseous pseudotumor with no malignant transformation. Intraoperative frozen sections indicated the resection margins were negative.

The patient was encouraged to do flexion and extension for rehabilitation postoperatively. After the second surgery, he recovered favorably and was discharged within 2 days. No adverse events were observed. At the 2-year follow-up after the last surgery, there were no signs of tumor recurrence. The affected finger functionally recovered, and the patient did not experience any difficulty while flexing the finger. No numbness or weakness was observed in the follow-up. The patient was satisfied with the result (*Figure 5*). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

Discussion

Fibro-osseous pseudotumor is a rare entity that was first described by Dupree and Enzinger in 1986 (4). Although locally aggressive, it is a benign tumor that involves the dermis and subcutis of extremities (1,4). Due to its rarity, aggressiveness, non-specific clinical manifestation, and equivocal histopathological presentation, it has previously been misdiagnosed as a malignant lesion, such as osteosarcoma, resulting in unnecessary radical treatment (4). Extra attention should be given to suspicious nodule, and the decision to perform radical treatment should be carefully made after confirmation of histopathological examination. Although several similar cases have been reported in recent years (5,6), we believe that our case is of great importance to fellow surgeons because of its specificity and the customized surgical plan devised by us.

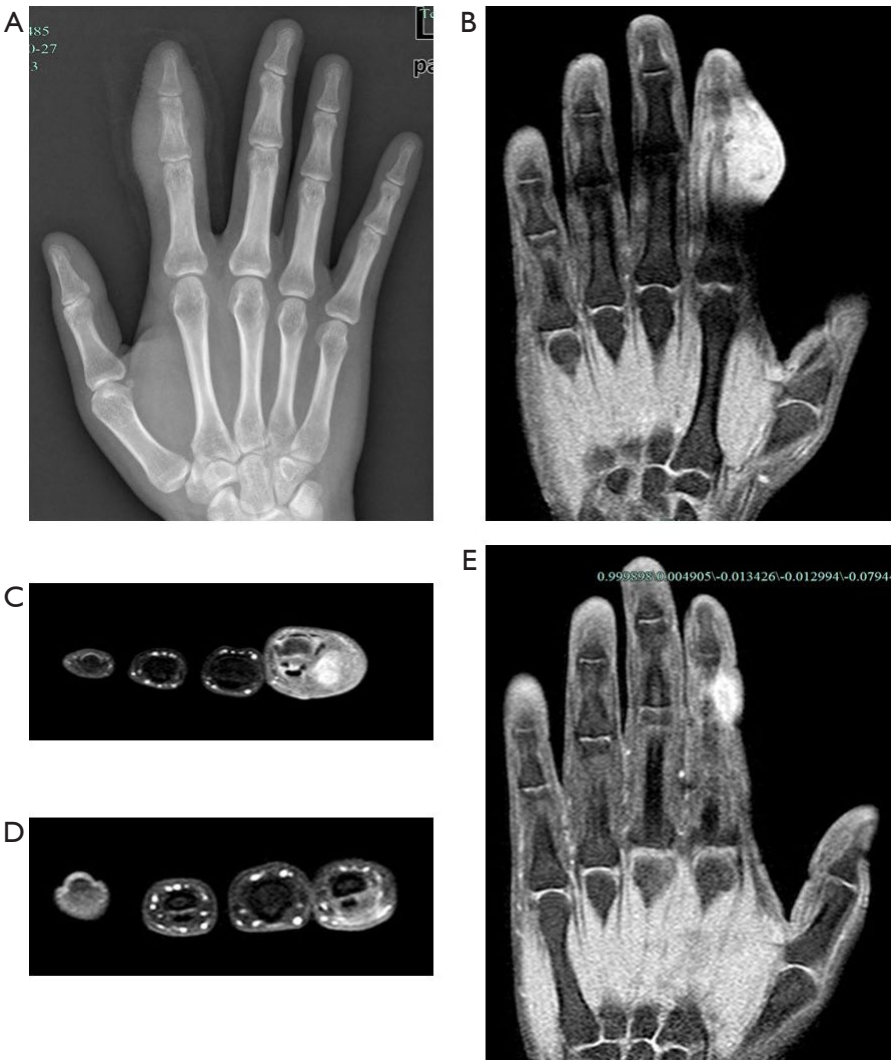

Figure 2 Imaging findings. (A) X-ray imaging prior to surgery; (B,C) coronal and transverse section of magnetic resonance imaging prior to the first surgery; (D,E) transverse and coronal section prior to the second surgery.

In this specific case, before surgical intervention, myositis ossificans (7), giant cell tumor of the tendon sheath (8), and extraskeletal and parosteal osteosarcomas (1,4) were considered in the differential diagnosis. Our surgical treatment strategy was planned based on the following. In preoperative communication with the patient, he expressed an intense will in retaining his finger and its function. Since malignant lesions could not be excluded and the affected finger's function was compromised, surgical intervention was considered inevitable. Considering the volume and specific location circling the second middle phalanx, complete removal of the lesion could not be performed through a single incision. If a thorough excision is performed by making two incisions and subdermal decollement, it is highly possible that the blood supply to the related tissue would be compromised, which would cause skin necrosis. Besides the potential trauma to nerves, vessels and tendons, it was possible to get necrotic tissues from needle biopsy. It would be safer and more accurate to biopsy in a direct and open way. Therefore, partial excision of the lesion was planned regarded as the preoperative biopsy we regularly do, saving the remaining for the second surgery to lower the possibility of skin necrosis. Even if the histopathological examination confirmed the lesion to be malignant, we could still arrange an extended resection

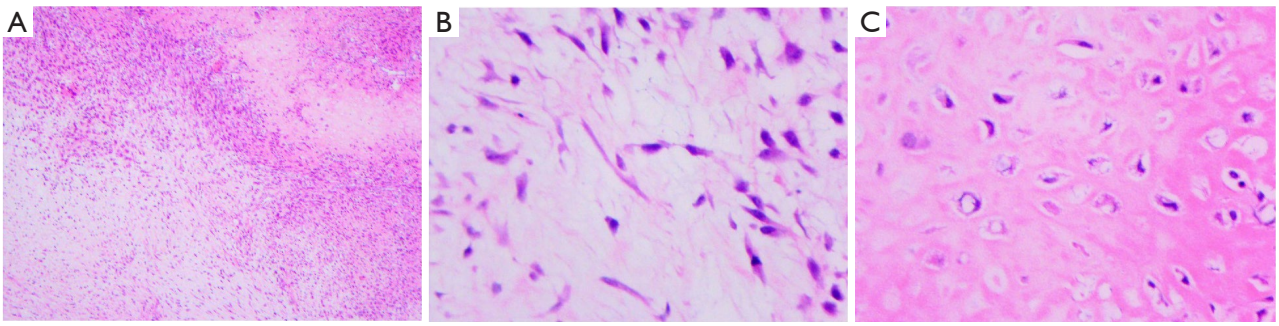

Figure 3 Fibro-osseous pseudotumor was considered in histopathological examination (hematoxylin and eosin staining). (A) Relatively uniform spindle cells with abundant interstitial vessels (×200); (B) spindle fibroblastic cell proliferation with varying degrees of atypia (×400); (C) osteoid formation with zoning phenomenon (×400).

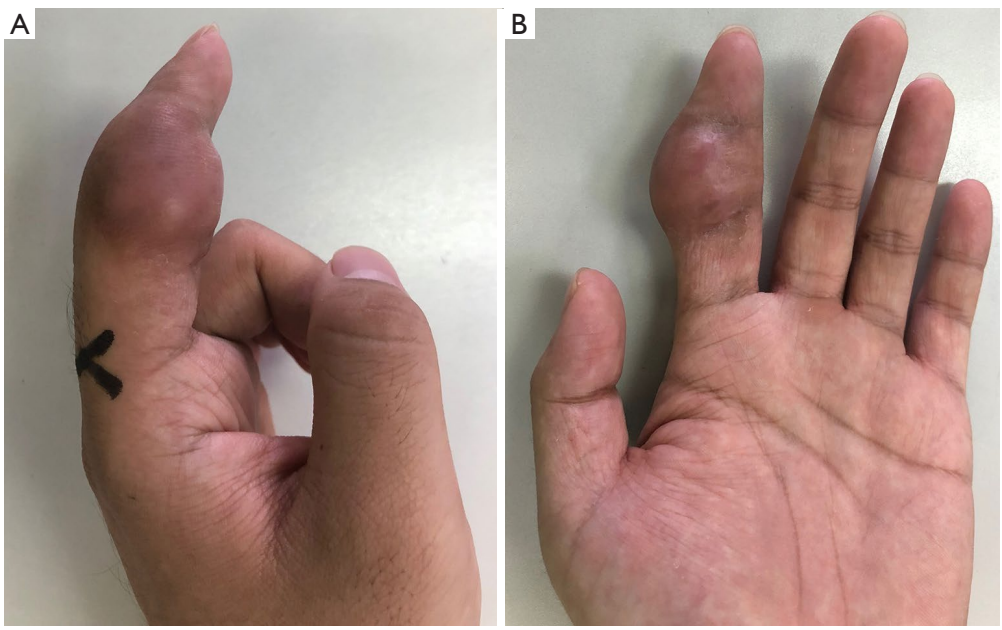

Figure 4 Photograph of tumor recurrence.

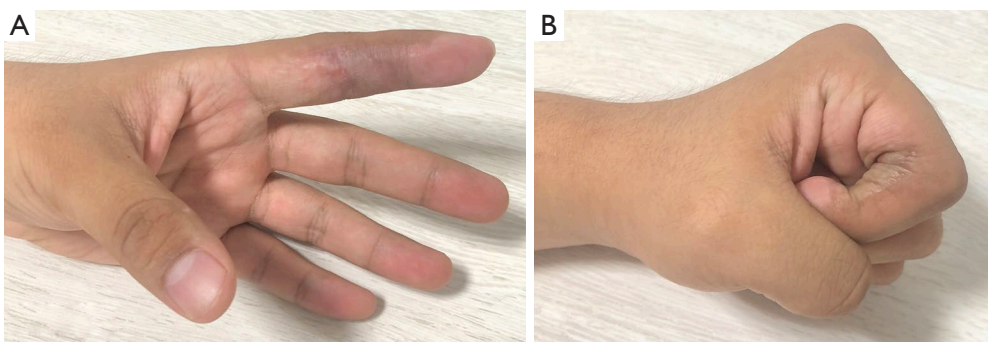

Figure 5 Follow-up after 2 years. (A) Lateral view. (B) Flexing position.

as soon as the results were obtained, and the long-term survival of the patient would not be affected.

Although regrowth of the residual tumor was predicted, the speed of the proceeding caught us off guard. During the intervening 4 months, the lesion recurred on the lateral side and progressed to almost the same size as that of the original mass, which further proved the aggressiveness of the tumor. In the second surgery, we thoroughly removed the mass along with the surrounding joint capsules, retaining essential nerves and muscle tendons. Intraoperative frozen sections and the 2-year follow-up showed that the lesion had been completely excised.

After reviewing the literature and summarizing our experience in treating this patient, we identified the following key points in diagnosing fibro-osseous pseudotumor (1,4,9): (I) dermis and subcutis involvement without muscle; (II) locally aggressive, with no malignant potential; (III) spindle fibroblastic cell proliferation with varying degrees of atypia; (IV) absence of cartilage; (V) osteoid formation with incomplete or complete zoning phenomenon; and (VI) possible periosteal reaction.

Although cases of recurrence have been reported previously (4,10) (location undefined, recurred one year later, completely re-excised; toe, recurred 4 months later, completely re-excised), the extent of the aggressiveness of fibro-osseous pseudotumor, as in this case, has not been reported thus far. It is quite reasonable to plan for a second-stage surgery if removal of the tumor is technically difficult or risky. However, when the treatment decision is made, the aggressiveness of the tumor should be considered. In addition, patients should be adequately informed of the risks preoperatively.

Fibro-osseous pseudotumor should be included in the differential diagnosis of rapidly progressive lesions of the digits. Complete surgical excision is the treatment of choice. However, the surgical strategy should be planned cautiously with respect to the aggressiveness of fibro-osseous pseudotumor and the possibility of saving the involved digit.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://dx.doi.org/10.21037/tcr-21-333

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://dx.doi.org/10.21037/tcr-21-333). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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