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CASE REPORT

# Fibro-osseous pseudotumor of the hand: a case report of a 22-year-old young woman

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## Abstract

We present a case of a 22-year-old patient with an important expansive neoformation of the soft tissues near the proximal phalanx of the fourth finger of the right hand. There was no evidence of past direct traumas, wounds, or local infections. The Rx, CT scan and MRI exams showed a lesion of the soft tissues measured around 3 × 2 × 2.8 cm, without hinting at a connection with the bone structure with a hypointense signal in T1 and an isointense signal in T2. First diagnostic speculations pointed at a giant cell tumor of the tendon sheath (GCTTS). Since there seemed to be no indication of a malignant tumor, it was decided to execute an excisional biopsy on the patient, rather than an eco-guided core needle. The imaging studies of the histological characteristics showed spindle-shaped elements with low-grade atypia, fibromyxoid and osseous stroma in different maturity stages, and always delimited by osteoblast cells. Osteoclastic giant cells were also found, as well as foci of osseous metaplasia peripherally aligned around proliferating cell aggregates. Considering all the following data, the final diagnosis clearly indicated a fibro-osseous pseudotumor of digits (FOPD). At the last checkup after 6 months, the wound healed correctly and without any restriction in finger movement.
Level of evidence: Level V, diagnostic study.

**Keywords** Fibro-osseous pseudotumor of digits (FOPD) · Hand surgery · Benign tumor lesion of the hand

## Introduction

Among the known uncommon benign lesions, fibro-osseous pseudotumor of digits (FOPD) of the hand is characterized by fibroblastic proliferation and an osteoid formation [1]. This type of tumor usually occurs in young women as a subcutaneous, painful mass, and is typically located in the proximal phalanges. FOPD generally shows aggressive clinical features, including rapid growth, increasing pain and swelling, with aggressive histological and radiological patterns [2]. Because of the latter, FOPD may be falsely labeled as a malignant bone-forming tumor like extraskeletal chondrosarcoma, extraskeletal osteosarcoma, or parosteal osteosarcoma [3]. Furthermore, it sometimes can be wrongly diagnosed as Turret exostoses or Nora's disease [4]. For this reason, it should be mandatory to have an accurate and precocious differential diagnosis, in order to discriminate a reactive lesion from a malignant neoplasm, and to avoid unnecessary radical treatment. Although there is no case of malignant transformation of FOPD in literature, a good prognosis with a low recurrence rate is only obtained if complete surgical excision is accomplished [5]. We hereby introduce a retrospective case of FOPD of the hand treated in a tertiary referral hospital. We also will describe the clinical, diagnostic, and surgical charts.

## Case presentation

A 22-year-old patient treated in a tertiary referral hospital showed the following characteristics: an important expansive neoformation of the soft tissues near the proximal phalanx of the fourth finger of the right hand; the lesion extended predominantly to the dorsal and radial side of the interested region (Fig. 1). Clinically, the patient did not report any pain; still, the range of movement was widely limited. At palpation, the lesion presented itself as mildly tender, only

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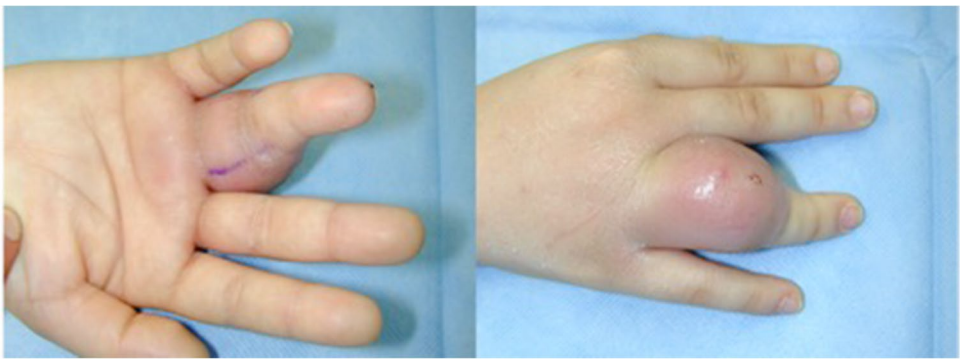

**Fig. 1** Neoformation of the soft tissues near the proximal phalanx of the fourth finger of the right hand extended to the dorsal and radial side of the finger

partially movable and without clear skin adherences. The only apparent visible skin connection was a small ulcer on the dorsal side of the mass. The patient reported that the mass had presented itself about 12 months before clinical presentation, but had grown out of proportion in the last 6–8 months. There was no evidence of past direct traumas, wounds, or local infections. First radiological exams, such as X-rays, confirmed a lesion of the soft tissues, without hinting at a connection with the bone structure (Fig. 2). Subsequently, the patient was subjected to a MRI scan: the lesion showed a hypointense signal in T1 and an isointense signal in T2, with an intense and uneven enhancement in post-contrast evaluation. It was furthermore characterized by a low signal intensity rim (Fig. 3). A following CT scan did not show any cortical bone erosion of the phalanx. In both radiological exams, an abundant perilesional edema was clearly noticeable, and the neoformation measured about $3 \times 2 \times 2.8$ cm. First diagnostic considerations, based on both radiological exams and clinical presentation, pointed at a giant cell tumor of the tendon sheath (GCTTS). Since there seemed to be no indication of a malignant tumor, the patient underwent an excisional biopsy, rather than an eco-guided core needle [6]. An informed consent was obtained before the treatment, with all possible risks associated with it explained. Before surgical treatment, prophylactic antibiotics were administered regularly and the anesthesiologist performed an ultrasound-guided axillary brachial plexus block.

After that, a longitudinal dorsal incision was made along the proximal interphalangeal area of the fourth finger, extending to the shaft of the fourth metacarpus. Exploration revealed an extensive subcutaneous fiber-adipose mass surrounding the radial digital neurovascular bundle. The excision was performed by dissecting the tumor, preserving the digital nerve bundle; the mass ultimately measured $4.5 \times 4 \times 1$ cm (Fig. 4). The imaging studies of the histological characteristics showed spindle-shaped elements with low-grade atypia and fibromyxoid and osseous stroma in different maturity stages; these elements were always delimited by osteoblast cells. Osteoclastic giant cells were also found, as well as foci of osseous metaplasia peripherally aligned around proliferating cell aggregates. Lastly, no evidence of necrosis or chondroid differentiation was found. Considering all the following data, the final diagnosis clearly indicated a fibro-osseous pseudotumor of digits. Both the surgery and the postoperative follow-up went smoothly and without complications. At the last follow-up, after 6 months,

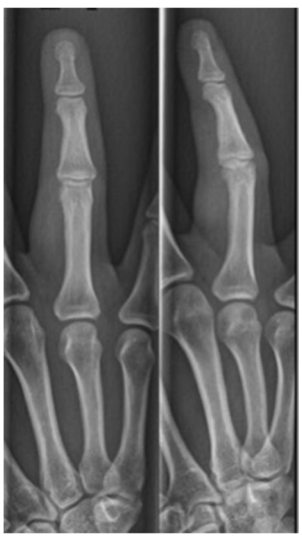

**Fig. 2** X-ray examination showed absence of bone structure involvement

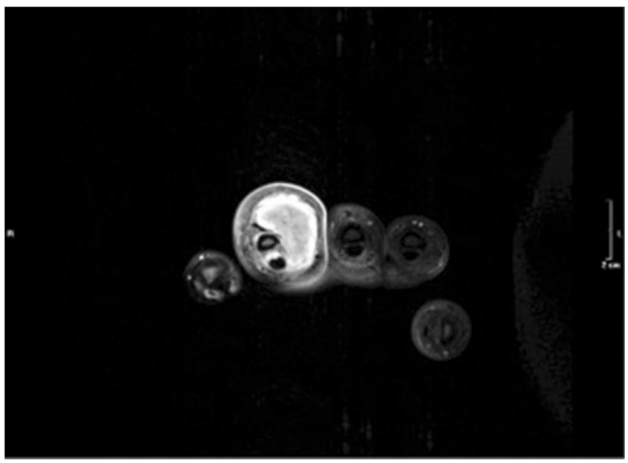

**Fig. 3** MRI showed a lesion with a hypointense signal in T1 and an isointense signal in T2, with an intense and irregular increase in post-contrast evaluation

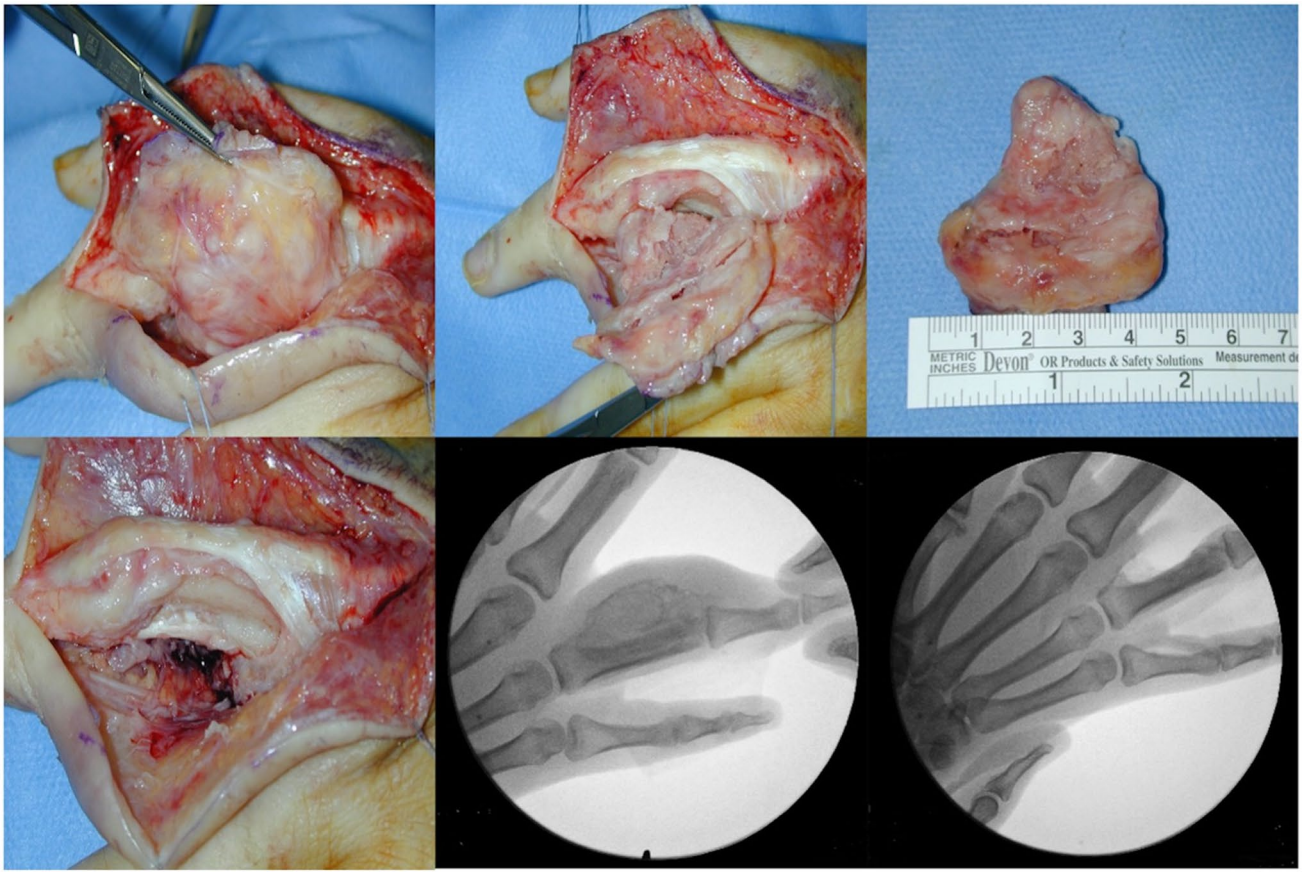


**Fig. 4** Surgical excision of the neoformation of 4.5 × 4 × 1 cm with intraoperative radiographic control

the wound had healed correctly and without any restriction in finger movement (Fig. 5).

## Discussion

Fibro-osseous pseudotumor of digits or FOPD is an entity closely connected to ossifying myositis [7, 8]; this type of lesion often develops in the soft subcutaneous tissue of digits, typically in the region of the proximal phalanx and less commonly in the middle finger of the hand. It is a difficult pathology to identify, and it often gets mixed up with other types of lesions, such as ossifying myositis and exostosis, or even worse, with malignant pathologies such as extraskeletal chondrosarcoma, extraskeletal osteosarcoma, and parosteal osteosarcoma . It is important to correctly and prematurely diagnose FOPD in order to avoid radical surgical treatment. FOPD typically afflicts young adults, age ranging between 10 and 64 years with a median age of 40, and it more specifically plagues women [9]. At clinical presentation, it manifests itself as a localized lesion, often painful and with erythematous swelling in the soft tissues encompassing the phalanges. In our study case, the patient did not report any direct traumas of the concerned hand; there have been however different cases noted in literature where neoformations developed after a local trauma. Such a history of trauma can be obtained in approximately 40% of patients. Furthermore, cases exist in which a chronic repetitive microtrauma has been identified. Radiographic imaging of FOPD can be difficult to read and identify: it commonly shows an ill-defined mass in the soft tissues overlapping the bone, and containing calcification. This type of neoplasm is commonly well separated from the phalanx, without any local periosteal reactions; however, it can be tricky to distinguish this kind of separation between the mass and the healthy bone [10].

MRI appears to be an optimal and adequate choice for a good diagnostic methodology, since it allows to distinguish the fibroproliferative lesion with signal intensities on T2-weighted imaging together with contrast enhancement, and to therefore determine how much of the soft perilesional tissues, together with tendons and neurovascular bundles, is involved. This type of analysis significantly reduces the risk of iatrogenic surgical damage.

On histologic imaging, a FOPD lesion appears grossly, gritty, gray-white, and zonal in approximately one-half of

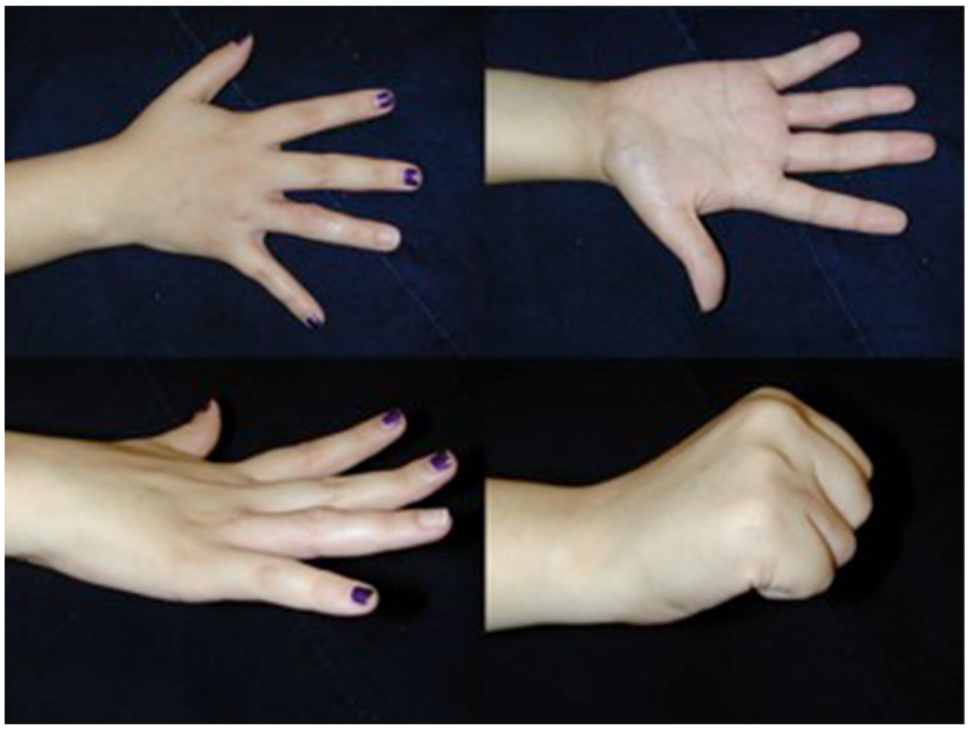

**Fig. 5** At the last checkup after 6 months, the wound healed correctly and without any restriction in finger movement

the known cases. The zonal organization includes mature woven bone in more peripheral zones, while immature woven bone in central areas: the whole bone structure demonstrates osteoblastic rimming [9]. FOPD is furthermore composed of a mixture of typical and atypical fibroblasts, osteoblasts, and trabeculae of bone showing varying degrees of maturation giving the appearance of a fibrous to myxoid matrix. Intramembranous ossification can be commonly noticed. Trabeculae of osteoid matrix and bone are rimmed by osteoblasts without evidence of peripheral zoning, with rare presence of osteoclasts (Fig. 6). On a first glance, a differential diagnosis would primarily include extraskeletal osteosarcoma that is extremely rare in the digits and most commonly arises in the lower extremities; nevertheless, the rim of osteoblast cells, the transitions from fibroblasts to osteoblasts and osteocytes, the absence of mitoses, and the transformation of collagen to osteoid are distinguishing features of FOPD. Turret and subungual exostosis may clinically simulate the FOPD, but radiologic connection to the underlying bone and histopathologic demonstration of fibrocartilage cap separate out the two entities; in fact, the fibro-osseous pseudotumor remains unattached to the bone with an intramembranous ossification and without evidence of cartilaginous cap. Another common entity that can be considered in a differential diagnosis is myositis ossificans [8]; however, it is usually diagnosed after a trauma, and its histopathological feature shows a typical zonation pattern.

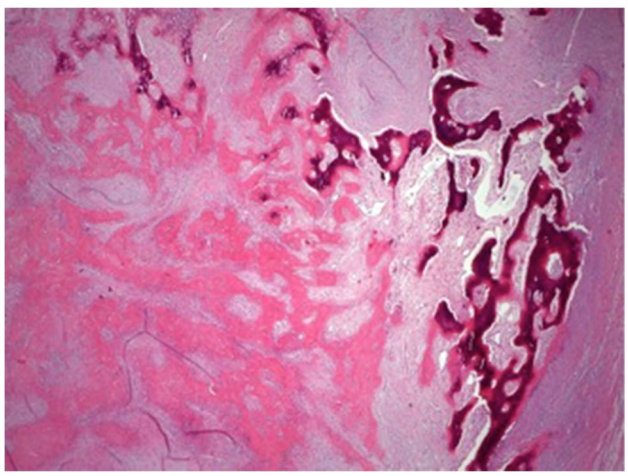

**Fig. 6** The histological examination shows that the lesion is composed of a mixture of typical and atypical fibroblasts, osteoblasts, and bone trabeculae which show variable degrees of maturation giving the appearance of a fibrous matrix with myxoid matrix

## Conclusions

Finally, this study, like many others, underlines how an accurate and premature diagnosis can be of the uttermost importance in the clinical journey of a patient. A correct diagnostic and therapeutic course can make the difference. FOPD is a fairly uncommon benign lesion that presents itself with such peculiar clinical and radiological characteristics, that it can easily be mistaken for other malignant pathologies. Considering that a core needle biopsy is not always easy to perform, especially in cases characterized by small lesions, and considering also that an excisional biopsy presents a risk

of intralesional or contaminated margins, it is paramount to understand the nature of the neoplasm, to avoid radical surgery. A correct employment of X-rays, CT scan, and MRI scans enables to identify the above-mentioned characteristics that can thus give an accurate understanding of the lesion and permit an adequate choice of treatment.

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**Author contribution** Dr. Tuzzato Gianmarco, Dr. Tosi Daniele, and Dr. Vita Fabio researched literature and conceived the study. Dr. Tuzzato Gianmarco wrote the first draft of the manuscript. Dr. Giuseppe Bianchi and Dr. Roberto Adani reviewed and edited the manuscript and approved the final version of the manuscript.

## Declarations

**Ethical approval** All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki. Declaration of 1975, as revised in 2008. Statement of human rights approval was obtained from the institutional review board regarding procedures to ensure protection of human rights and informed consent.

**Informed consent** Written informed consent was obtained from the patient included in this study.

**Conflict of interest** Tuzzato Gianmarco, Vita Fabio, Bianchi Giuseppe, Daniele Tosi, and Adani Roberto declare no competing interests.

**Patient consent** Patient signed informed consent regarding publishing her data and photographs.

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