Benign growing mass of the digit presenting as an ulcerated mass – case report and review of the literature

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ABSTRACT
A 68 year-old female presents with an ulcerated mass of the 5th digit, with rapid growth during the previous month to surgery. The mass was excised and covered with a 4th dorsal metacarpal artery perforator flap. The histologic analysis was compatible with the diagnosis of fibro-osseous pseudotumor of the digit.

ARTICLE HISTORY
Received 17 March 2021
Accepted 26 July 2021

KEYWORDS
Fibro-osseous pseudotumor
digit; hand tumor;
hand surgery

Introduction
The hand surgeon acts both as an oncological surgeon, ablating all the tumor mass, potentially compromising aesthetics and function, and as a reconstructive surgeon, trying to optimize hand function. Balancing these goals may be a demanding task [1].

Even though the majority of hand tumors are benign, especially if they do not involve the skin, some malignant tumors arise in the hand. In the latter, a more aggressive approach would be justified [2].

Fibro-osseous Pseudotumor of the Digits (FOPD) is a rare benign tumor with clinical characteristics that can mimic a malignant tumor. We present a case of FOPD and a review of the literature.

Case description
A 68 year-old female, smoker, presented to the ER with an ulcerated mass of the dorsum of the 5th finger involving the proximal phalanx to the DIP joint (Figure 1). It had been progressively enlarging for the past year with substantial growth in the previous month. It was presented as a painless mass that limited PIP flexion. The neurovascular examination was normal. The ultrasound showed a 4.4 x 2.6 cm hypoechoic mass with a significant doppler sign.

Excision with the overlying skin was performed down to a tumor less surgical plane, preserving the extensor apparatus (Figure 2). The resulting defect was covered with a 4th dorsal metacarpal artery perforator flap and the donor area primarily closed (Figure 3). Histopathological study revealed a lobulated tumor, self-limited and centered in the dermis, causing epidermal ulceration. Histologically, it is composed of fascicles of uniform spindle cells, admixed woven bone without zonation. There’s a mixture of fibroblasts and myofibroblasts, arranged in hyper and hypocellular hyalinized areas and deposits of osteoid rimmed by uniform osteoblasts. Cells have bland cytology and no necrosis or mitotic figures are seen, compatible with the diagnosis of FOPD (Figure 4 and 5). The patient was followed for 22 months with no evidence of recurrence (Figure 6).

Discussion
FOPD is a rare entity. There are 173 cases documented in the literature [3–54], totaling 174 with the present case report (Table 1). The first case dates back to 1931. There have been several terms to classify this entity including: ‘pseudo-malignant osseous tumor of soft tissue’ [36], ‘parosteal fasciitis’ [49], and ‘florid reactive periostitis’ [3,4,7,13]. This disease was unified under the term ‘fibro-osseous pseudotumor of the...
digits’ in 1986 by Dupree and Enzinger [50]. The tumor is characterized by reactive fibroblastic proliferation and focal bone formation limited to the skin and subcutaneous tissue. The tumor is not of bone origin, though [52]. It presents as an enlarging mass, which can be tender or painless. Even though a history of trauma has been described in as many as 40% of patients, it is hard to assess the relevance and prevalence of injury since minor trauma of the extremities is common. Skin ulceration may occur; the tumor mimics pyogenic granuloma, especially if it affects the toes [7,31,38,48]. Due to its growth and local aggressiveness, it can simulate malignancies. When mentioned, we found a high rate of suspicion of sarcoma [14,19,50,51], namely osteogenic sarcoma. Some case series did not report the individual patient data regarding age [19,46,50]. The age ranged from 5 to 50 years.

Figure 1. Preoperative photographs of the tumor, dorsal view (A), palmar view (B).

Figure 2. Post-excisional photographs of tumor: inferior (A) and lateral (B) views.

Figure 3. Intra-operative views after surgical excision of the tumor with preservation of the extensor apparatus (A) and after raising the flap (B).
81, with a mean age of 36 and a median of 30 years old. The majority of patients were female (56.9%, 99 cases). It is more frequent in the upper limb (82.6%) than in the lower limb (16.9%). There is a sole case of FOPD outside the extremities [28]. In the upper limb, the most common segment affected is the finger (76.8%), more precisely the 2nd finger (32%) and the proximal phalanx (62.3%), followed by the hand (21.1%) and wrist (2.1%).

Radiologically, FOPD usually presents as a soft tissue mass with ill-defined margins and might reveal extraosseous ossification and bone formation. The periosteal reaction has been described [42]. Cortical involvement or bony destruction seldom occurs. MRI findings are commonly non-specific and frequently demonstrate a non-invasive soft tissue mass.

Histologically, FOPD presents as a multi-nodular lesion with irregular margins and it is localized in the dermis and subcutaneous tissues. Muscular and cartilage involvement is absent.

Typically, FOPD shows fibroblastic/myofibroblastic proliferation without atypia, in a myxoid stroma, and immature trabeculae with osteoid rimmed by osteoblasts, without zoning phenomenon [10, 23, 51]. Osteoclasts and bone marrow elements are rarely seen [42].

Immunohistochemically, the majority of studies has demonstrated positive staining for vimentin 14 [11, 21, 25, 26, 34], and focal reactivity to actin [18, 21, 26, 27, 40, 51, 53], which is in favor of myofibroblastic differentiation [18]. There is, however, controversial data, since Chan et al. found no reactivity to actin, myosin, myoglobin, desmin, and no dense bodies on electron micrograph, which is against the involvement

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**Figure 4.** Fibro-osseous pseudotumor of digits. The lesion presents as a well-circumscribed dermal mass, with epidermal ulceration. Hematoxylin-eosin × 25.

**Figure 5.** Fibro-osseous pseudotumor of digits. At medium power, the mixture of arranged fibroblasts and myofibroblasts and the deposits of osteoid is rimmed by uniform osteoblasts. Hematoxylin-eosin × 100.

**Figure 6.** Post-operative views at 22 months showing complete wound healing without functional limitation: dorsal (A), and medial (B,C) views.
| References           | Reported cases | Age (years) | Gender | Patient history | Clinical presentation | Anatomical location | Ulceration | Treatment          | Recurrence | Follow up |
|----------------------|----------------|-------------|--------|-----------------|------------------------|---------------------|-------------|-------------------|------------|-----------|
| Mallory [3]          | 1              | 17          | F      | –               | Wart-like lesion for 3 years | 3rd intermetacarpal space right hand | –           | Local excision      | –          | –         |
| Gonçalves [4]        | 1              | 23          | F      | –               | Painful swelling        | P1D2 left hand       | –           | Transmetacarpal amputation | –          | 60 months |
| McCarthy et al. [5]  | 1              | 40          | M      | –               | Growing mass, 6 months | P1D4 right hand      | –           | Ray amputation       | –          | 12 months |
| Spjut and Dorfman [6]| 12             | 5–46 (mean 22, median 19) | 5 M, 7 F | –               | Swelling mass (12), pain (5) and redness (3) | Hand: D3 (4); D4 (3); D5 (2); Foot: D1 (2); Hand: P1 (6); P2 (3), MC (1); Foot: P3 (1); MT (1) | –           | Local excision (1), ray amputation (1) | 1 Recurrence 10 months after local excision. | 2 months-11 years |
| De Smet and Vercauteren [7] | 1             | 58          | F      | –               | Painful swelling mass with, impaired flexion of joint | P1D3 left hand       | –           | Transmetacarpal amputation | –          | 12 months |
| Bulstrode et al. [8] | 1              | 30          | F      | –               | Stiffness and weakness, 2 years + tender nodule, 2 weeks | Left hand            | –           | Local excision followed by transmetacarpal amputation of 3 digits | Recurrence 2 months after associated with refractory pain, no recurrence after amputation | 12 months |
| Patel and Desai [9]  | 1              | 35          | F      | –               | Painful growing mass, 1 year | P1D5 left hand       | –           | Local excision followed by ray amputation | Recurrence after local excision, no recurrence after amputation | 8 years |
| Dupree and Enzinger [10] | 21            | 8–70 (median 33) | 13 F 8 M | –               | History of trauma injury site | Sudden (9)/slowly (3) growing mass; Painful (12), erythematous (11); hematoma (1), cellulitis (5) | DD1 (3); D2 (6); D3 (5); D4 (3); D5 (4); P1 (6); P2 (1); P3 (3); not stated (1) | –           | Amputation(2), radical excision (1), Local Excision (18) | Follow up in 7 cases, up to 15 years |
| Chan et al. [11]     | 1              | 17          | M      | History of trauma injury site | Enlarging polyloid nodule 2 months | P3D2 left foot       | Present Local excision > 2, followed by amputation by DIP joint | Recurrence after local excision, no recurrence after amputation | 12 months |
| Franchi et al. [12]  | 1              | 55          | M      | –               | Painless mass for 3 years | P1D3 right hand      | –           | Local excision       | –          | 24 months |
| Aboujaoude et al. [13]| 1              | 13          | F      | –               | Painful polyloid mass, 1 month, pyogenic granuloma-like | P2D1 right foot, subungual | Present Local excision       | –          | –         |
| Horie and Morimura [14] | 1            | 59          | F      | –               | –                        | –                    | –           | –                 | –          | 4 months |
| Prevel and Hanell [15]| 1              | 25          | F      | –               | Growing mass, 6 months | P3D2 right hand, ungual | –           | Local excision ×2 | Recurrence 10 months after excision | 24 months |
| Riaz et al. [16]     | 1              | 48          | M      | –               | Swelling mass associated with pain and stiffness | P1D1 left hand       | –           | Local excision       | –          | 6 months |
| Tang et al. [17]     | 1              | 53          | F      | –               | Painful swelling        | M4 left hand         | –           | Local excision       | –          | –         |
| Sleater et al. [18]  | 3              | 47–81 (mean 61.3333) | 2 M 1 F | –               | Growing mass 4 months with hyperkeratotic cap | Subungual P3 hallux, right foot | 1 out of 3 P3D3 (1) | –          | –         |
| Moon et al. [19]     | 1              | 10          | M      | –               | Growing mass 4 months | Subungual P3 hallux, right foot | –           | Local excision       | –          | 9 months |
| Takahashi et al. [20] | 1             | 60          | F      | –               | Painless swelling for 3 months | P3D2 right hand, subungual | –           | –                 | –          | –         |
| Nishio et al. [21]   | 1              | 30          | F      | –               | Painful swelling with rapid increase over 4 weeks and restriction of PIP joint | P2D4 left hand | Local excision       | –          | 7 months |
| Solana et al. [22]   | 1              | 56          | M      | –               | Tender growing mass, 3 months | P1D1 left hand       | –           | Local excision       | –          | 24 months |

(continued)
| References                     | Reported cases | Age (years) | Gender | Patient history | Clinical presentation | Anatomical location | Ulceration | Treatment | Recurrence | Follow up   |
|-------------------------------|----------------|-------------|--------|-----------------|------------------------|---------------------|-------------|-----------|------------|-------------|
| de Silva and Reid [23]        | 14             | 17–75 (mean 38.1, median 35.5) | 7 M 7 F |                  | Painful growing mass 75%, mean duration 9.7 weeks | D1 (2), D2 (4), D3 (5), D5 (3) | – | (4) Biopsy, (9) Excision, (1) finger amputation *Initial misdiagnosed as osteosarcoma | – | Local recurrence (1) 24 months after excision (1) |
| Hiso et al. [24]              | 1              | 29          | F      |                  | Swelling and painful mass, pyogenic granuloma-like | 1st Intermetatarsal space, left foot | – | – | Local excision | – | 17 months |
| Coleman [25]                  | 1              | 70          | F      |                  | Growing nodule for the past 2 years with rapid growth over 2 months | P3D4 left hand | – | – | Ray amputation | – | 10 months |
| Usta et al. [26]              | 1              | 57          | M      | History of trauma (6) + occupation with continuous hand-work | Painful swelling 2 years | P1D2 left hand | 6 out of 7 | Local excision | – | – |
| Moosavi et al. [27]           | 4              | 3           | 1      | 0                | Pain and edema | Hand (6), Finger (1), D1 hand (3), D2 hand (11), D3 (10), D4 hand (4), D5 hand (2), hypothermic eminence (1), web space (2), wrist (1), foot (1), D1 foot (1) | – | – |
| Nalbantoglu et al. [28]       | 1              | 30          | M      |                  | Pain and swelling on the radial side of the right wrist, 4 months | Wrist: 1 cm distal to the radial styloid over the first dorsal-extensor compartment | – | Local excision | – | 24 months |
| Kaddoura and Zaatari [29]     | 1              | 42          | M      |                  | Erythematous nodule with rapid growth over 2 months and progressively painful, limiting flexion and abduction of thumb | Thenar eminence and 1st interdigital space, left hand | – | Local excision | – | 18 months |
| Bettex et al. [30]            | 1              | 15          | M      | History of trauma on injury site (2) | Slow-growing mass Nodule (12), swelling (3), verrucous lesion (1), purulent lesion (1), duration from 2 weeks to 2 years | D1 Ungual D1 foot (1), Ungual D2 foot (1), Ungual D4 foot (1), D4 foot (1), D5 foot (1) D1 foot (2), D2 hand (3), D3 hand (2), D5 hand (1), foot (1), forehead (1) | 7 out of 17 | Local excision | Local excision (16), Finger amputation (1) * recurrence with later diagnosis of sarcoma | – | 15 months |
| Chaudhry et al. [31]          | 17             | 5–64 (median 34) | 8 M 9 F | History of trauma on injury site (2) | Swelling, 3 months M4 left hand | – | 14 cases: 12–168 months. Median 45 months, average 62 months |
| Song et al. [32]              | 1              | 20          | F      |                  | Painful mass | P1D1 left foot | – | – | Local excision | – | – |
| Tan et al. [33]               | 1              | 54          | F      |                  | Nodule mimicking pyogenic granuloma: tender, mobile and erythematous nodule, growing for 2 months | P2D1 left foot | Present | Local excision | – | – |
| Sayar et al. [34]             | 1              | 30          | F      | History of trauma injury site | Painful swelling mass | P2D5 left foot | – | Local excision | – | 24 months |
| Anand et al. [35]             | 1              | 21          | F      |                  | Swelling, 3 months | M4 left hand | – | – | Local excision | – | 6 months |
| Lee et al. [36]               | 1              | 22          | F      |                  | – | D2 left hand | – | Local excision | – | 7 months |
| Seeger et al. [37]            | 1              | 5           | M      |                  | Swelling mass | M3 right hand | – | Local excision | – | 12 months |
| Javidan and Tahristian [38]   | 1              | 30          | M      |                  | – | P2D2 left hand | – | Local excision | – | – |

(continued)
| References          | Reported cases | Age (years) | Gender | Patient history | Clinical presentation | Anatomical location | Ulceration | Treatment                                                                 | Recurrence | Follow up |
|---------------------|----------------|-------------|--------|-----------------|-----------------------|---------------------|------------|---------------------------------------------------------------------------|------------|-----------|
| Comer et al. [39]   | 1              | 61          | F      | Rheumatoid Arthritis treatment with methotrexate Rural laborer by occupation | Painful growing mass for 12 months, finger motion slightly limited | P2D2 right hand | –          | Amputation                                                                | –          | –         |
| Hashmi et al. [40]  | 1              | 32          | M      |                  | Painless tumefaction with increase over 1 month | D3                  | Present    | Local excision                                                            | –          | 12 months |
| Kwak et al. [41]    | 1              | 19          | F      |                  | Painful growing mass, 12 months | P1D3 right hand | –          | Local excision                                                            | –          | 12 months |
| Zhou et al. [42]    | 1              | 48          | M      |                  | Painful erythematous mass worsening 2 months | P2D4                | –          | Narrow-margin excision with recurrence followed by complete excision      | –          | 6 months  |
| Meani et al. [43]   | 1              | 16          | M      | Crush injury 2 years before | Painful subungal hematoma and swelling of toe, 2 years after trauma | P2D1 left foot | –          | Curettage and nail plate removal + local excision                         | Recurrence 6 weeks after curettage, no recurrence after local excision | 12 months |
| Kontogeorgakos et al. [44] | 1        | 28          | F      | Hairdresser working with scissors with the hand | Erythematous swelling with progressive pain over 3 months, Tinel's sign positive | Dorsum of 1st intermetacarpal space, left hand | –          | Local excision                                                            | –          | 12 months |
| Choi et al. [45]    | 1              | 68          | M      | Type 2 DM       | Growing tender subcutaneous mass for 3 months | P1D1                | –          | Local excision                                                            | –          | 6 months  |
| Singal et al. [46]  | 1              | 36          | F      |                  | Painless lesion growing for the past 6 months | Subungual P3D2 right hand | –          | Local excision                                                            | –          | 8 months  |
| Gómez-Zubaur et al. [47] | 1        | 13          | F      | Cuticle manipulation | Mass mimicking pyogenic granuloma: Painful, erythematous mass with granulomatous bleeding surface growing for the previous 2 months | P3D3 left hand periungual | Present | Shaving followed by local excision                                        | –          | –         |
| Cho et al. [48]     | 1              | 27          | M      | History of trauma injury site | Painful erythematous mass for 2 months, viral wart like | P3D3 foot          | Present    | Local excision                                                            | –          | 6 months  |
| Takahashi et al. [49] | 1          | 36          | M      |                  | Subungual painful keratotic nodule, 2 months | Subungual P3D5 left foot | –          | Local excision                                                            | –          | –         |
| Flucke et al. [50]  | 5              | 33–72 (mean 48) | 2 M 3 F |                  |                         | Palm (2), D3 (1), thenar eminence (1), D1 foot (1) | –          | Local excision (5)                                                        | –          | –         |
| Jawad et al. [51]   | 1              | 27          | F      |                  | Growing mass for 7 months, Mass (9), granulation tissue (1), painful mass (1), osteochondroma (1) | P1D4                | Present    | –                                                                          | –          | –         |
| Švajdler et al. [52] | 12             | 5–64 (median 32) | 6 M 6 F |                  |                         | Palm (2), D4 foot (1), D4 foot (2), 1st metacarpal, thenar eminence (1), digit of the hand (1), D5 foot (1) | –          | –                                                                          | –          | –         |

(continued)
of myofibroblasts [11], and Sayar et al. found no evidence of immune-reactivity to actin and desmin [34]. The majority of cases demonstrated negative staining to S100, CD34, cytokeratin (MAK-6 and CAM 5.2), desmin, and epithelial membrane antigen (EMA) [3,11,18,25–27]. Diagnosis is challenging – FOPD is a rare disease with only 174 cases documented, and it shares many histological and clinical features with malignant disease. This might lead to incorrect diagnoses, such as extraskeletal osteosarcoma, resulting in unnecessary procedures, including amputation. It should, however, be considered as a potential diagnosis of fast-growing tumors of the hand or feet with no previous history of trauma [42]. Osteosarcoma is usually diagnosed in older people (scarcely under 40) and is rarely found in the fingers, being more common in the large bones of the upper and lower extremities [42]. Myositis ossificans has been associated with FOPD, being the latter considered by some authors as a superficial variant of the first. However, in myositis ossificans there is commonly a history of trauma [38,42,51]. Other differential diagnosis include bizarre parosteal osteochondromatous proliferation (Nora’s lesion), ossifying plexiform tumor, acral osteoma cutis, and subungual exostosis [45,50].

FOPD might be included in a group of USP-6-rearranged myofibroblastic neoplasms that share a genetic rearrangement of the USP-6 gene and are known for their rapid yet self-limited growth and low recurrence rate [52,55]. Recent studies have identified COL1A1-USP6 fusions in the majority of their FOPD cases [50,52,56]. Molecular and genetic studies can therefore help diagnosis and avoid over-treatment, particularly in cases where biopsy is difficult to obtain [52,55], or clinical history is insufficient [52] since these lesions frequently mimic soft tissue sarcomas [52], but are self-limited and can be cured with a more conservative surgical excision [55].

The treatment for FOPD is surgical excision. Prognosis is excellent as recurrence has been related to incomplete excision [10,11,42]. We found 13 cases in which amputation was the treatment of choice [4–11,23,25,27,39]. Some of these cases were initially suspected or diagnosed as malignant – de Silva and Reid described a 13% of malignancy assumption by radiologists and a 9% incorrect malignant diagnosis by pathologists [23], while in others the extension of the disease and bone destruction led the surgeons to decide for amputation.

Nine cases of recurrence were described in the literature [6,8–11,15,23,31,43]. Most of them did not provide information about the reason for the recurrence.

| References | Reported cases | Anatomical location | Patient history | Clinical presentation | Treatment | Recurrence | Follow up |
|------------|----------------|---------------------|----------------|----------------------|------------|-------------|----------|
| Sakuda et al. [53] | 1 | 30 | M | Suspected occupational injury due to excessive load | Local excision | - | 6 months |
| Rela and Banik [54] | 1 | 60 | F | Hypothenar hyperkeratosis due to a metal rod insertion | Washout and debridement | - | 6 months |

Table 1. Continued.

M: male; F: female; D: digit; P: phalanx; M: metacarpal.
However, in three cases incomplete excision seems to be the reason for it thus reinforcing the need for complete excision.

After complete removal of the tumor, no recurrences were found with a median follow up of 15 (4–168) months [4,5,7–16,19,21–25,28–31,34–37,40–46,48,53]. Even though no malignant transformation has been described in the literature, close follow-ups are recommended.

Conclusion

FOPD should be considered in the differential diagnosis of fast-growing lesions of the extremities. An excisional biopsy allows for complete tumor removal and avoids overzealous treatment. FOPD should be treated without radical excisions provided if totally removed.

Disclosure statement

The authors report no conflict of interest.

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