



Case report

Fibro-osseous pseudotumor of the digit: Case report and surgical experience with extensive digital lesion abutting on neurovascular bundles

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ABSTRACT

Background: Fibro-osseous pseudotumor (FOPD) of the digit is a rare benign lesion of subcutaneous tissue characterized by fibroblastic proliferation and osteoid formation. Herein, we present a case of massive FOPD lesion in the base of ring finger with extensive involvement of the neurovascular bundles with challenging surgical approach.

Case description: A 27-year old female patient, presented with 7-months history of a progressively enlarging mass on her left hand. Upon assessment, the mass was located over the proximal phalanx of the left ring finger with extensive involvement of the 4th web space. Her neurovascular examination was normal. Radiological investigations showed partial involvement of the radial sided bundle together with complete involvement of the ulnar sided neurovascular bundle. The patient was bothered by the mass being painful with overlying skin ulceration. She was taken afterwards to the operating room where the mass was dissected freely from those bundles while preserving the radial and ulnar structures. The resected margins were however, positive for residual lesions due to the extensive nature of the mass. The patient was informed about the need for close follow-ups for both clinical and radiological signs of lesion recurrence pending early surgical intervention.

Conclusion: FOPD although benign, a soft tissue osteosarcoma is one of the differential diagnosis. Meticulous attention to the clinical, pathological and histological features of FOPD is required. Early diagnosis and treatment of FOPD is very crucial in optimizing the overall outcome. Pre-operative planning with various radiological modalities was of great help anticipating the surgical course.

1. Introduction

Fibro-osseous pseudotumor of the digit (FOPD), is a rare benign lesion of subcutaneous tissue characterized by fibroblastic proliferation and osteoid formation [1,2]. Fibro-osseous pseudotumor of the digit was previously prescribed in literature under various names like; florid reactive periostitis, parosteal fasciitis and fasciitis ossificans [1,3]. It is crucial to know the clinical, pathological and histological features of this lesion in order to avoid diagnostic ambiguity. The major differential diagnoses include; myositis ossificans, extraskeletal osteosarcoma, parosteal osteosarcoma and subungual exostosis [1,4,5]. Fibro-osseous pseudotumor of the digit usually presents as a localized proximal nodule that is slightly mobile and affecting more commonly women than men. This lesion is known for its good prognosis if complete surgical excision was attained, with a very low recurrence rate

and no reports of malignant transformation [1,4,5].

2. Methodology

This is a retrospective chart review and case presentation of a patient presented to our tertiary care hospital. Patient's related history and examination are summarized. This work has been reported in line with the SCARE criteria [6]. The patient was consented for the publication of this work.

3. Case presentation

Our patient is a 27-year old female that denied any past medical and surgical history. Her family and drug history were also unremarkable. The patient presented with a 7-month history of progressively enlarging

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Fig. 1. Clinical presentation of digit Fibro-osseous pseudotumor mass. It shows the pre-operative clinical presentation of the mass at the base of the left ring finger. Notice the overlying ulcerative skin. Pre-operative incision marking is also shown.

mass on her left ring finger. The patient mentioned that the mass appeared suddenly with no history of trauma and that she was concerned about the potential malignant nature of the mass. The patient also mentioned that she underwent incision and drainage of that mass 2 months after its appearance with no improvement and provided no detailed surgical or pathological reports, which was the reason for her delayed presentation. Upon her assessment, the mass was located over the ulnar side of the proximal phalanx of left ring finger with extensive involvement of the 4th web space. The overlying skin coverage was ulcerative with no active signs of infection. Range of motion of the involved digit was limited, however neurovascular examination was normal. (Fig. 1).

Radiological evaluation of the involved hand showed a soft tissue swelling with no evidence of bone involvement (Fig. 2). Further magnetic resonant (MRI) evaluation showed a mass on the volar aspect of the ring finger encasing about 50% of the flexor tendons of that digit with low signal intensity on T1 and high signal intensity on T2 evaluation with strong enhancement in post contrast evaluation. Assessment of neurovascular structures showed partial abutment of the radial sided bundle together with complete encirclement of the ulnar sided neurovascular bundle. The surrounding bone was free of any masses and associated mass effect.

The patient was taken to the OR for exploration and mass excision by the senior author. Possible risks associated with such intervention were explained. Intra-operatively, bruner type incision was designed together with island of skin involved in the mass. Exploration revealed extensive subcutaneous mass with fibro-fatty consistency with

extensive fascia like extension to the surrounding soft tissue. The mass was encircling the ulnar neurovascular bundle with mass abutment over the radial bundle as seen in pre-op assessment. The mass was dissected freely from its attachment to those bundles preserving both radial and ulnar structures. The mass was then excised en-bloc having a dimension of $3.5 \times 4 \times 2.5$ cm (Fig. 3). Histological assessment showed a lesion with fasciitis like features, myofibroblastic proliferation and scattered foci of osteoid formation that was positive for Alpha-Smooth Muscle Actin (ASMA 1A4) immune staining and no evidence of malignancy (Fig. 4). The resected margins were however, positive for residual lesion with difficulty in obtaining negative margins due to the extensive nature of the mass. Post-operatively, the patient had an uneventful course. She was informed about the need for close follow-ups for both clinical and/or radiological signs of lesion recurrence, pending early surgical intervention (see Fig. 5).

4. Discussion

Fibro-osseous pseudotumor (FOPD) of the digit is a rare benign lesion that usually presents clinically as a localized erythematous, painful, subcutaneous fusiform swelling or mass that appears suddenly or gradually in the proximal phalanx of the digits, most commonly in the index finger and very rarely in toes [1,4,5,7]. On gross assessment, it was observed as a well-circumscribed, firm or rubbery lesion with a gray-whitish cut surface and some calcification [1]. It predominantly affects young adults, unlike myositis ossificans, that is more common in women [7]. Although a history of trauma was not considered related to diagnose this lesion, such history can occasionally present in patients with FOPD [1,7,8].

On X-ray, the lesion presents as an ill-defined soft tissue mass with focal calcification, and lack geographic distribution or zoning patterns of myositis ossificans. Focal periosteal thickening adjacent to the mass can also be seen, and in some rare cases, distinct cortical erosions were observed [1,7,9,10]. Magnetic resonant imaging of such tumor was shown to exhibit low to intermediate signal intensity on T1-weighted imaging and variable signal intensities on T2-weighted imaging together with contrast enhancement that were consistent with fibro-proliferative lesions [11]. In our patient pre-operative radiologic imaging was of great help anticipating the nature of mass extensive extension and involvement of neurovascular bundles. Meticulous dissection and handling helped in optimizing the surgical outcome with no residual effect on any vital structures.

Histologically the lesion appears multi-nodular with irregular margins. It consists of a mixture of typical and atypical fibroblasts, osteoblasts and trabeculae of bone with variable degrees of maturations. The proliferation of fibroblasts was either loosely disseminated within a richly mucoid matrix or forming a netlike pattern associated with collagen and islands of loosely cellular tissue. Fibroblasts were seen with abundant amphophilic cytoplasm with indistinct borders and large, slightly pleomorphic nuclei, distinct nuclear membranes, and occasionally small nucleoli. Fibroblast transition to osteoblasts and osteocytes is also a common feature seen under the microscope, as well as the transformation of collagen to osteoid and bone. Osteoblasts however show no cellular atypia, and osteoclasts and bone marrow elements are rarely seen. Multinucleated giant cells are also present. Signs of inflammation were usually minimal and consist of small aggregates of lymphocytes and occasional plasma cells [1,7]. Immune staining of such lesions were found to have focal positivity for alpha smooth muscle (ASMA), S100 and CD 34 markers [8]. Histological assessment of the excised mass in this patient showed similar finding with scattered osteoid formation, myofibroblast proliferation and positivity for ASMA immune-staining.

A diagnostic algorithm was suggested when evaluating such lesions. The first step was to assess the stromal component of the lesion for any evidence of malignancy including; nuclear atypia, pleomorphism, mitotic activity and growth pattern. If malignant evidence was found, then

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