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Case report

Intra-articular nodular fasciitis of the proximal interphalangeal joint of a finger: A case report



Fasciite nodulaire de l'articulation interphalangienne proximale d'un doigt : à propos d'un cas

H. Choughri^{a,*}, J.-M. Coindre^b, F.M. Leclère^{a,c}

^a Service de chirurgie plastique, reconstructrice et esthétique-brûlés-chirurgie de la main, CHU de Bordeaux, groupe hospitalier Pellegrin, centre François-Xavier-Michelet, place Amélie-Raba-Léon, 33076 Bordeaux cedex, France

^b Institut Bergonié, département de pathologie-cancérologie, université de Bordeaux, 229, cours de l'Argonne, 33000 Bordeaux, France

^c Service de chirurgie plastique et chirurgie de la main, CHU de Poitiers, université de Poitiers, 2, rue de la Milétrie, 86000 Poitiers, France

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ABSTRACT

Nodular fasciitis is a benign reactive lesion, often mistaken for a soft-tissue sarcoma in clinical practice. Involvement of the finger is rare, and a finger joint even rarer. We report on the clinical, radiological and histological features of intra-articular nodular fasciitis in a 52-year-old man, originating from the proximal interphalangeal joint of the right ring finger, with cortical erosion of adjacent bone. The discussion is focused on the tumor diagnosis and therapeutic approach, the differential diagnosis and the importance of immunohistochemical staining to establish the final diagnosis.

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R É S U M É

La fasciite nodulaire est une lésion réactive bénigne, souvent confondue avec un sarcome des tissus mous dans notre pratique clinique. La localisation au doigt est rare, et en regard d'une articulation de doigt encore plus rare. Nous rapportons les caractéristiques cliniques, radiologiques et histologiques d'une fasciite nodulaire intra-articulaire chez un homme de 52 ans, localisée à l'articulation interphalangienne proximale de l'annulaire droit, et associée à une érosion corticale de l'os sous-jacent. La discussion s'est portée sur l'approche diagnostique et thérapeutique de la tumeur, le diagnostic différentiel et l'importance des marquages immunohistochimiques afin d'établir le diagnostic final.

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1. Introduction

Nodular fasciitis (NF) is a well-known benign, reactive myofibroblastic proliferation, with aggressive histological appearance, which is characterized clinically by a history of a rapidly growing single subcutaneous mass (over a few weeks) that may be mistaken for a sarcoma [1–3]. Here we present a rare case of NF, located under the volar plate of the proximal interphalangeal (PIP) joint of a finger, which caused erosion of the cortical surface of the base of the middle phalanx and the neck of the proximal phalanx. The initial biopsy did

not lead to a precise diagnosis but helped to rule out a neoplastic process. Histological differential diagnosis included a desmoid tumor (DT), NF and fibroma of the tendon sheath (FTS). Finally, complete excision of the tumor confirmed the diagnosis of NF after several immunohistochemical stains were done. Our discussion is focused on the treatment approach, the differential diagnosis and the importance of immunohistochemical staining to establish the final diagnosis. The patient was informed that data concerning this case would be submitted for publication and provided his consent.

2. Case report

A 54-year-old right-handed man who worked as a general practitioner, was referred to our outpatient clinic with a 3-month

* Corresponding author.

E-mail address: choughrih@yahoo.fr (H. Choughri).

history of a swollen painful ring finger of his dominant (right) hand, with limited flexion of his proximal interphalangeal (PIP) joint. The story began when he experienced spontaneous pain and swelling, without any notion of trauma. As a general practitioner, the patient decided to treat himself with non-steroidal anti-inflammatory drugs for a few weeks, thinking it was simply a finger sprain. He had temporary improvement, with partial regression of edema and slight improvement in PIP mobility. Faced with persistent symptoms, the patient requested a finger X-ray (Fig.1A), which was normal. One month later, he consulted a rheumatologist who did a blood test, another set of plain X-rays and an ultrasound of the finger. The blood test was normal. Ultrasound examination revealed PIP joint effusion with osteoarthritis. In comparison with the first set of normal X-rays, the second set of X-rays (Fig.1B) showed a well-defined cortical erosion at the base of the middle phalanx and the neck

of the proximal phalanx. Two joint aspirations were performed under ultrasound guidance and found no liquid. A magnetic resonance imaging (MRI) scan was performed which showed a non-specific, well-circumscribed mass, $2.2 \times 1.5 \times 0.8$ cm in size, that was isointense on T1-weighted images and had heterogeneous high signal intensity on T2-weighted images (Fig.1C and D).

The patient was referred to our department 3 months after the initial onset. He had a history of a traumatic spiral fracture of the middle phalanx of his finger 10 years ago, treated with a cast. Physical examination showed a painful right ring finger, with local swelling and deficit of the PIP joint range of motion (ROM) ($-10^{\circ}+60^{\circ}$ versus $0-80^{\circ}$ on the contralateral side). There was no triggering, sensory disturbance or joint instability. No palpable mass was identified, and the joint was not warm to touch. With the presumptive diagnosis of a non-specific soft-tissue tumor on MRI

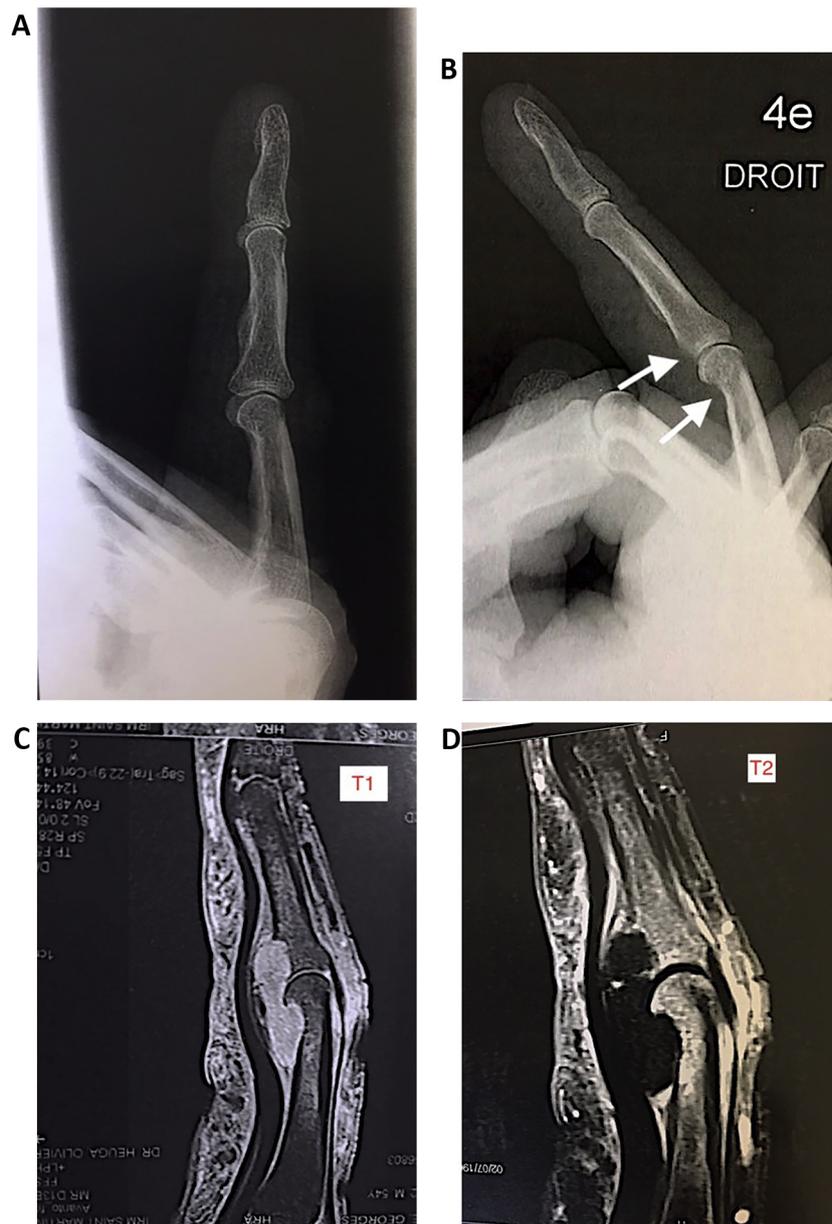


Fig. 1. Normal radiograph of the fourth finger (A). X-rays of the same finger, a few weeks later, showing well-defined cortical erosion at the base of the middle phalanx and the neck of the proximal phalanx of the PIP joint (white arrow) (B). Magnetic resonance imaging of the right ring finger showing a non-specific intra-articular well-circumscribed mass, with cortical erosion at the PIP joint, elevating the volar plate and the flexor tendon mechanism; T1-weighted images have a low signal intensity (C) and T2-weighted images have a homogeneously high signal intensity (D).

including the possibility of a sarcoma, an ultrasound-guided biopsy was performed to obtain the definitive diagnosis needed before surgery.

Histopathological examination ruled out any malignant tumoral proliferation, evoking a benign myofibroblastic fibrous lesion with spindle-shaped cells in a loosely textured myxoid matrix and wide bands of collagen, suggestive of a fibrous tumor. The differential diagnosis included NF, FTS and DT.

Given the patient's pain, continued rapid growth of the lesion radiologically with cortical bone erosion, size of the tumor, PIP joint location under the volar plate, and a benign histological diagnosis of a fibrous tumor, surgical excision was decided to ensure complete removal of the mass after discussion in a multidisciplinary cancer meeting. This was performed through a volar surgical approach using a Bruner incision with release of the A2, A3 and A4 pulleys, tendon dislocation, and complete detachment of the volar plate (Fig. 2A, B).

Histological examination of the surgical specimen (Fig. 3A, B) revealed a non-encapsulated tumor composed of cytologically bland plump spindle cells arranged in short, intersecting bundles within a variable loose myxoid to collagenous stroma, containing extravasated red blood cells and scattered lymphocytes. The lesion also showed prominent stromal development. Immunohistochemistry (IHC) staining revealed that the spindle cells were positive for alpha-smooth muscle actin (SMA, 1A4, 1:100; DAKO) and Ki67 (MIB-1, 1:150; DAKO), with an MIB-1 proliferation index of around 15%. It was negative for beta-catenin (Beta-catenin-1, 1:100; DAKO), S-100 protein (Polyclonal, 1:3000, DAKO), CD34 (Qbend10, 1:100, novocastra), HMGA2 (High Mobility Group AT-Hook 2), cytokeratin (AE1-AE3, 1:100, DAKO), EMA (E29, 1:200, DAKO), caldesmon (h-CD; 1:100, DAKO) and desmin (D33, 1:100; DAKO). From these findings, the diagnosis of DT was ruled out and a diagnosis of FTS proposed. This was finally changed to NF after considering all the IHC results, more specifically, the USP6 Break-Apart FISH probe results which confirmed rearrangement in 28% of the cells analyzed.

3. Discussion

Nodular fasciitis was first described by Konwaler et al. [4] in 1995 and was called pseudosarcomatous fibromatosis because of its rapid growth and histological presentation. It is often misdiagnosed clinically as a malignant neoplasm (fibrosarcoma or malignant fibrous histiocytoma). The etiology of NF is still unclear. NF is known to be a benign version of sarcoma, both clinically and histologically. It typically develops in the upper limbs (46%), especially the volar aspect of the forearm followed by the head and neck (20%), trunk (18%), and lower extremities (16%) [5]. NF can be divided into 3 major subtypes: subcutaneous, intramuscular, and fascial; it is often located in the subcutaneous tissue. Al-Qattan and Arafah reported four cases of NF of the hand treated surgically over a 20-year period. They preserved vital structures such as the digital nerve, both digital arteries of a finger, the volar plate, and the flexor profundus tendon; none of these tumors had joint involvement [6]. NF has also been reported in other unusual anatomical locations such as the parotid gland urinary bladder and or at the cubital fossa [7]. NF is extremely rare in the hand, and less than 30 cases have been reported in the literature. Here we describe a new location in the joint. Cortical erosion associated with NF in the hand has rarely been described [8]. To date, there are 19 cases of intraarticular NF reported. Of these, 13 were in the knee, three in the shoulder, two in the hand, one in the ankle [9]. All were reported in the English literature except for cases reported in the Japanese literature. NF in the hand is often mistaken for a soft-tissue sarcoma, such as malignant

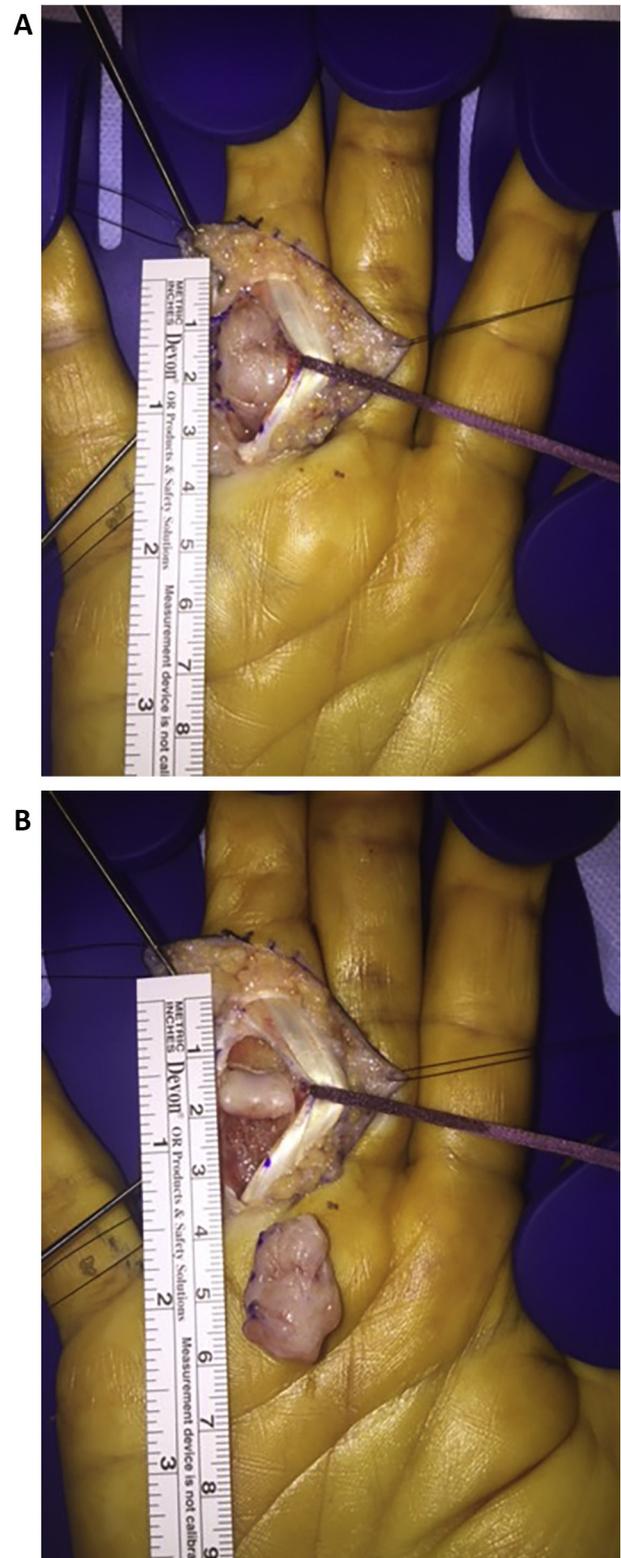


Fig. 2. Intraoperative view of the non-encapsulated mass after a Bruner volar approach, pulleys removed, tendons dislocated, and complete detachment of the volar plate (A). View of the PIP joint and tumor after total resection, measuring $2.2 \times 1.5 \times 0.8$ cm in size (B).

fibrous histiocytoma or fibrosarcoma, resulting in ray amputation or unnecessary irradiation [10,11].

Several factors make the diagnosis of intra-articular NF difficult, which is why several distinctive features deserve further comment, to avoid misdiagnosis which can result in incorrect

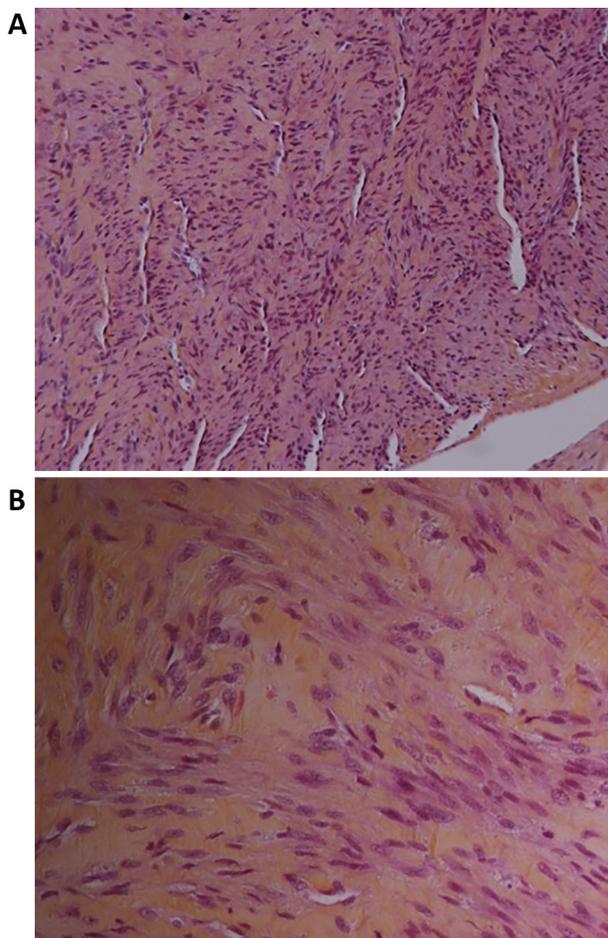


Fig. 3. Microscopic images of the intra-articular nodular fasciitis specimen from the marginal excision. Medium-power view of NF showing proliferation of non-atypical fusiform cells with background of numerous extravasated erythrocytes, hemosiderin deposition, and lymphocytic inflammation (A). High-power view of NF showing myofibroblastic-like spindle cells without nuclear atypia or mitotic activity (B).

treatment. First, when compared to NF lesions arising in subcutaneous or intramuscular locations, the intra-articular location seems to have a notably longer clinical history. This may in part be secondary to the fact that the primary symptom was non-specific joint pain with no palpable mass. Second, the initial X-rays is normal, the MRI findings is non-specific, and ultrasound is not very informative orienting the diagnosis towards osteoarthritis of the PIP joint. Indeed, MRI features of NF are often non-specific and somewhat variable. In the largest MRI study of musculoskeletal NF, it typically appeared isointense to slightly hyperintense to muscle on T1-weighted images, hyperintense on T2-weighted images and homogeneously enhanced after gadolinium administration [12]. Third, the tumor biopsy was suggestive of a fibrous tumor and was also non-specific, which could include the possibility of sarcoma, DT or FTS. The final diagnosis of intra-articular NF was made after complete excision of the tumor and its analysis with several IHC stains.

FTS is an uncommon benign tumor that almost always arises in the hands, typically within fibrotendinous tissue rather than in a joint. Pulitzer et al. [13] reported 32 cases of FTS. Most cases presented as a painless, firm and nodular mass in the distal portion of an extremity. They concluded that at least some cases of FTS described as fasciitis-like actually represent examples of nodular fasciitis.

Extra-abdominal DTs are extremely rare in the hand. These tumors do not metastasize; however, they are potentially locally invasive and have extremely high local recurrence rates after surgical excision with reports of up to 78% recurrence, specifically in the hand. IHC characteristics of DT are helpful, as they do not have immunoreactivity for desmin, similar to NF [14], but have proliferating spindle cells on a collagenous background with positive nuclear beta-catenin expression and CD34, which is not found with NF.

In a recent study [15], the diagnostic utility of USP6 fluorescence in situ-hybridization (FISH) analysis in a subset of spindle-cell proliferations for the differential diagnosis of NF was evaluated. In morphologically definitive cases, FISH analysis for USP6 had a sensitivity of 86% and specificity of 100% for a diagnosis of NF. The positive predictive value was 100% and the negative predictive value 90%. They concluded that USP6 FISH is a useful auxiliary test in cases where NF is a differential diagnosis. In our clinical case, the diagnosis of intra-articular nodular fasciitis was confirmed step by step and after considering the IHC results together and specifically with the USP6 Break-Apart FISH probe which confirmed rearrangement in 28% of the cells analyzed.

The patient has had 6 follow-up visits over 12 months so far and is free of recurrence. This has been confirmed with follow-up MRI. He has almost a full range of finger movements after physical therapy and dynamic splinting, with the exception of a 15° residual flexion deformity of the PIP joint in active extension.

4. Conclusion

Although NF of a finger is quite rare, it is important to keep in mind rarer pathologies such as intraarticular NF, especially with a history of trauma 10 years prior. NF should be included in the differential diagnosis of intraarticular masses discovered fortuitously even though it occurs in the finger joint. A biopsy is necessary before complete tumor excision. An experienced histologist must be part of the team to avoid possible misdiagnosis of malignant fibrous histiocytoma or sarcoma, which can lead to unnecessary irradiation or finger amputation. We want to emphasize the importance of careful clinical and histological examination to avoid unnecessary aggressive surgery, since marginal excision will provide excellent results even in a case with a difficult location such the finger PIP joint.

Disclosure of interest

The authors declare that they have no competing interest.

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