Vascular malformation of the flexor tendon presenting as tenosynovitis

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Congenital vascular malformations are rare and may involve any region of the body. Lesions are primarily venous, arterial, capillary, or a combination thereof and are commonly divided into two categories: dysplasia and hamartias. Dysplasia are normal formations of tissue with structural deviation; conversely hamartias are anomalous formations of normally present tissue [9].

The upper extremity is involved in 30% to 60% of patients with vascular malformations [6]. These often present in the early stages of life, with approximately 50% of lesions being clinically recognized before the age of 2 [9]. While these lesions are typically present since birth, they are often only noted following trauma, as this may cause hemorrhage or stimulate vascular growth. Swelling is the most common presentation, seen in 50% of patients, followed by pain (40%) and a noticeable mass (30%). Other presentations include asymmetric warmth, paresthesia, hyperhidrosis, and ischemic ulcers [4].

Initial investigation involves plain X-rays, demonstrating soft tissues masses in approximately 65% of cases. Ultrasound and computer tomography can often further define the extent of the bone or soft tissue involved. If surgical intervention is considered, an MRI or MRA may be used to evaluate bone and soft tissue involvement, while arteriography may be used to delineate the AVM [7].

Treatment represents a challenge, as recurrence is common and lesions are often much more extensive than

suggested by the clinical appearance. The decision to carry out surgical intervention often depends on the location of the malformation, symptoms, and the vessels involved.

Case Report

A 34-year-old woman presented for evaluation of severe pain in her left palm radiating along her ring and middle fingers that had been progressively worsening for 2 days. At the onset of symptoms, the patient noted a dull ache in her left palm, which she attributed to increased use of her left (non-dominant) hand. At this time, she also noted a 10×10-mm soft tissue mass at the volar base of her middle finger.

One day prior to hospital presentation, the patient attended her general practitioner where she was prescribed non-steroidal anti-inflammatory drugs and a splint. The pain escalated overnight and she presented in the morning to the hospital with severe pain. She had no preceding trauma and did not report any associated fevers or rigors. She was otherwise fit and well.

On physical examination, the left hand appeared swollen over all digits, which was significantly worse in her middle finger. On palpation, there was a 10×10-mm poorly defined mass at the volar base of her middle finger, which was tender to touch and normothermic. There was also pain along the length of the middle finger flexor tendon sheath. All fingers were in a flexion posture at the metacarpophalangeal, proximal interphalangeal, and distal interphalangeal joints, which was significantly worse in the left middle finger. There was moderate pain with passive and active extension of the middle finger, localized to the volar aspect of the third metacarpophalangeal joints and extending along the left middle finger.

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Laboratory tests were conducted and the results included a normal white cell and neutrophil count. A measure of her C-reactive protein was normal at 2 mg/L.

An ultrasound targeted at the swelling in the left palm demonstrated a hypoechoic heterogenous collection between the middle finger flexor digitorum superficialis and flexor digitorum profundus. The lesion was highly vascularized on Doppler assessment, measuring approximately 20 mm in length and 7 mm in width (Figs. 1 and 2). The radiology team suggested that this could represent infective tenosynovitis with phlegmon. X-ray examination of the hand showed no skeletal abnormalities.

Due to the clinical picture, pyogenic flexor tenosynovitis was deemed the most serious condition needing exclusion. However, without a clear source of infection, a broad differential was still considered. Due to the mild diffuse swelling and pain throughout the hand, the hospital's Rheumatology Department was consulted to investigate the possibility of arthritis including crystalopathies. By their guidance, further laboratory investigations were conducted, including rheumatoid factor, CCP antibody, and urate. The results of these investigations were within normal limits. They found no clear signs of arthritis and suggested an exploration and washout of the region if symptoms of tenosynovitis persisted on intravenous antibiotics.

Due to her early presentation, medical management was initially attempted. Her hand was splinted and she was started on intravenous antibiotics. As she suffered cephalosporin hypersensitivity, this consisted of lincomycin (600 mg, t.d.s.).

Following 48 h of intravenous antibiotics, the patient was still experiencing significant discomfort. The decision was made for surgical exploration. A zigzag incision was made in the palm over the proximal end of the sheath. A second

midline incision was made on the pulp of the distal phalanx. The middle finger tendon sheath was opened and a 10×10-mm non-pulsatile, dusky, vascular mass was identified inside the tendon sheath without any associated purulent material (Fig. 3). Swabs were taken of synovial fluid for microscopy and culture, and the tendon was washed out.

The intraoperative wound swab had no leukocytes or organisms on gram stain. There was no growth on culture after 2 days. Histopathology examination of the specimen showed a vascular malformation which consisted of large abnormal arteriovenous-like structures which had thickened muscular walls and lined by bland endothelial cells (Fig. 4).

The patient experienced an uneventful recovery with complete resolution of pain within 7 days of surgery. At 3 weeks, she had full range of movement with no functional deficits.

Discussion

Vascular malformations are a common condition and are usually diagnosed clinically. They are typically followed up periodically unless they are symptomatic or the diagnosis is in question. As in this case, a deep location or acute presentation may create a diagnostic challenge. The literature describes only three cases of a vascular malformation in a tendon sheath [3, 10, 13], two of which presented with signs of tenosynovitis [3, 13].

In the above case, our patient presented with all four of Kanavel's signs, with the addition of a mass at the base of her middle finger and mild pain and swelling to her whole hand. This mixed picture led to a significant diagnostic challenge. Due to the presence of mild diffuse symptoms throughout her hand, crystalopathies and rheumatoid arthritis were considered. Had this ultimately been the diagnosis, then

Fig. 1 Ultrasound of the left middle finger FDS/FDP tendons, longitudinal sections showing a hypoechoic heterogenous collection between the middle finger FDS and FDP with high vascularity on Doppler assessment

Fig. 2 Ultrasound of the left middle finger FDS/FDP tendons, transverse showing a hypoechoic heterogenous collection between the middle finger FDS and FDP with high vascularity on Doppler assessment

the presence of the vascular malformation may have been purely coincidental. However, as these tests were negative and the patient's signs and symptoms were significantly worse in the middle finger, the authors propose that the lesion itself led to the presentation.

We propose that the mass effect caused by the lesion created a situation similar to that of reactive tenosynovitis or stenosing tendovaginitis, where the narrowing of the tendon canals restricts tendon glide and causes further damage and irritation. That two of the other three cases of vascular malformations found in tendon sheaths also presented with signs of tenosynovitis supports this hypothesis [10, 13]. In addition, there have been two similar cases with nonvascular masses involving the tendon sheaths presenting as flexor tenosynovitis, consisting of a patient with metastatic small cell cancer in the phalanx, [1] and an angioliipoma [12].

The literature also describes a palmar arch vascular malformation which presented as subacute tenosynovitis despite ultimately not having any involvement with the tendon or sheath on surgical exploration. This suggests that there may also be a local reaction related to these lesions, in addition to their mass effect [5], and provides a potential explanation for the added mild diffuse swelling seen in our patient's hand.

Investigation of a soft tissue mass should initially include plain X-ray; however, diagnosis will be most significantly aided with a combination of ultrasound with or without MRI. Meanwhile arteriography may further be able to delineate the lesion. Ultrasound may be able to show an abnormal effusion,

Fig. 3 Intraoperative view of the dissection of the lesion from the flexor tendon

Fig. 4 Pathologic examination, a large abnormal arteriovenous structure with thickened muscular walls lined by bland endothelial cells, image is 7×4 mm

abscess, or vascular mass in the tendon sheath [2] but is often operator dependant and may not lead to the definitive diagnosis, as in the above case. MRI or MRA provide the best means to investigate a soft tissue mass in the hand, potentially aiding in both diagnosis and surgical planning but are often limited by availability and cost. MRI may speed up diagnosis in cases where vascular malformations are suspected, especially if an ultrasound is equivocal, and allow more prompt management. In two of the three cases of vascular malformation in tendon sheaths, MRI played a key role in diagnosis [3, 10]. In the above case, MRI could have potentially provided a preoperative diagnosis, allowing for a smaller procedure and decreasing recovery time.

The key additional challenge this case posed was that while the patient did not recall any trauma or recent infections, the speed at which symptoms developed and the clinical picture led to the concern of possible pyogenic flexor tenosynovitis. Hematogenous spread to the sheath (i.e., *Neisseria gonorrhoeae*) occurs rarely but should also be suspected if there is no puncture wound or history of trauma [8, 11]. Early diagnosis and treatment of pyogenic flexor tenosynovitis is necessary to prevent tendon necrosis, adhesion formation, and the spread of infection to the deep fascial spaces. Therefore, if its diagnosis cannot be excluded within an appropriate timeframe, as in our case, surgery is often warranted and should not be delayed.

Conflict of Interest The authors declare that they have no conflicts of interest, commercial associations, or intent of financial gain regarding this research.

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