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# Case Report

# A rapidly recurrent synovial hemangioma involving tendon sheath: A rare case in the finger

Tien-Ching Lee a,\*, Song-Hsiung Chien , Ping-Cheng Liu , Yuh-Min Cheng

- <sup>a</sup> Department of Orthopaedics, Kaohsiung Medical University Hospital, Kaohsiung Medical University, Kaohsiung, Taiwan
- b Department of Orthopaedics, Kaohsiung Municipal Hsiao-Kang Hospital, Kaohsiung Medical University, Kaohsiung, Taiwan

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#### ABSTRACT

Synovial hemangiomas in the hands are very rare. We present a 14-year-old girl who had a rapidly recurrent tenosynovial hemangioma at the volar aspect of the proximal interphalangeal joint of her left ring finger with pain on motion and numbness over the fingertip. There are no earlier reports in the literature of synovial hemangioma found distal to A2 pulley. Its benign property and rarity may lead to inappropriate excision and recurrence. Complete excision with the involved tendon sheath is recommended.

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### 1. Introduction

Synovial hemangiomas are uncommon tumors, arising from surfaces lined by synovium, such as tendons and joint cavities. They are found most frequently in the knee (60%), followed by the elbow (30%), and are usually present in children or adolescents. They are rarely present in the fingers. To our knowledge, there are only five cases involving tenosynovium of the hand reported in the literature, <sup>2–6</sup> four of them found to have infiltrated within the underlying tendon. Among these cases, none was found distal to the A2 pulley.

We present a rare case of a rapidly recurrent tenosynovial hemangioma at the ventral aspect of the proximal interphalangeal joint of the left ring finger with encasing of the flexor digitorum sublimis tendon, causing distal numbness and local tenderness.

# 2. Case report

This 14-year-old girl had no underlying systemic disease. She had a history of a capillary hemangioma over the left ring finger, which was excised 1 year ago. Before surgical intervention,

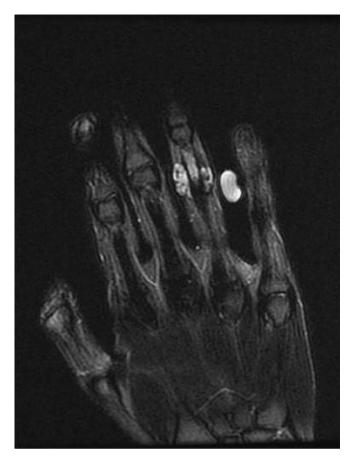
E-mail address: tn916943@gmail.com (T.-C. Lee).

a palpable mass was found and reported to have existed over the volar side of the proximal interphalangeal (PIP) joint of the left ring finger for about 3 years with mild pain on flexion, local tenderness, and distal numbness. Magnetic resonance imaging (MRI) revealed a well-defined soft tissue tumor, which was hyperintense on T1-and T2-weighted images and enhanced by contrast media, with encasing of the flexor tendon. Intra-operatively, a cystic dark red soft tissue tumor with adhesion to the flexor tendon sheath was noted. The postoperative pathological report was compatible with hemangioma.

However, a progressively enlarging mass almost at the same location was found about 1 month later, with similar symptoms as before. The patient denied any trauma history. MRI showed a well-defined soft tissue lesion (1.5 cm  $\times$  0.7 cm  $\times$  1.1 cm) at the volar aspect of the PIP joint of the left ring finger. This lesion was significantly hyperintense on T2-weighted images, and enhanced by contrast media (Fig. 1). The flexor tendon was encased by the enhanced lesion, but intact. The ventral surface of the adjacent bony structure was slightly compressed by the lesion. Recurrent hemangioma was impressed. Differential diagnosis included giant cell tumor of the tendon sheath.

Using a volar Brunner incision, we incised the C2, C3, A3 and the distal part of the A2 pulley and then the soft tissue mass was revealed above the flexor digitorum profundus tendon. We dissected the cystic dark red tumor bluntly and then retracted the deep flexor tendon. The tumor tissue was noted beneath the tendon

<sup>\*</sup> Corresponding author. Department of Orthopaedics, Kaohsiung Medical University Hospital, 100 Tzyou 1st Road, Kaohsiung 807, Taiwan. Tel.: +886 7 3121101x5751; +886 928795356 (cell); fax: +886 7 3119544.



**Fig. 1.** Magnetic resonance, T2-weighted coronal image revealing a significantly hyperintense, well-defined soft tissue lesion at the palmar aspect of the proximal interphalangeal joint of the left ring finger.





**Fig. 3.** (A) Cystic hemangioma encasing the flexor digitorum superficialis tendon with compression of the digital nerve. (B) After excision of the main tumor mass, the residual tumor tissue can be seen beneath the superficial flexor tendon without infiltration of the tendon.

with infiltration to the tendon sheath and compression of the digital nerve (Fig. 2). The whole tumor was excised grossly along with involved tendon sheath. No infiltration of the tendon was noted (Fig. 3).

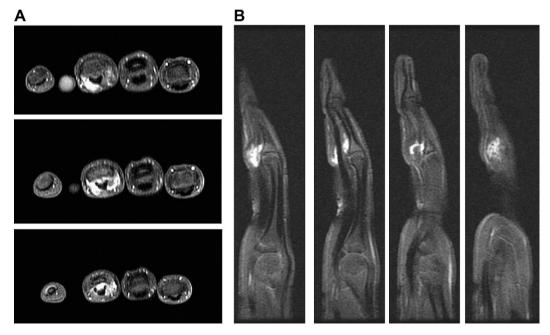
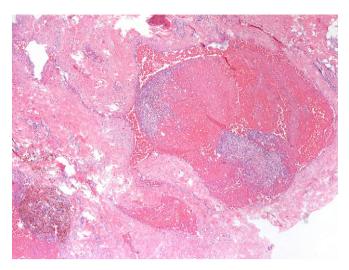


Fig. 2. (A) MR, STIR coronal image showing the flexor tendon of the ring finger encased by the enhanced lesion. (B) MR, STIR sagittal image showing the ventral surface of the adjacent bony structure slightly compressed by the soft tissue lesion. MR = magnetic resonance; STIR = short T1 inversion recovery.



**Fig. 4.** Microscopic appearance of the tumor (hematoxylin and eosin staining), showing dilated blood vessels with organizing thrombus mucosa surrounded by fibrous tissue.

After the operation, active motion was preserved without weakness, pain, tenderness or numbness. Histologically, the specimen showed dilated blood vessels with organizing thrombus mucosa, surrounded by fibro-adipose tissue, consistent with a capillary hemangioma (Fig. 4). There was no arterial component, such as elastic fiber within the vessel wall. No atypism was found. There was no evidence of recurrence noted after more than 1 year of follow-up.

# 3. Discussion

Hemangiomas occurring in the hand are considerably rare. They may arise from peripheral nerves, <sup>7,8</sup> muscles, <sup>9</sup> volar plate <sup>10</sup> or

tendon sheaths.  $^{2,4-6}$  Including the case presented here, there are six reported cases of hemangiomas in relation to tendon sheath in the hands (listed in Table 1) $^{2-6}$  with an average age of 22.5 years (14–33 years old) and a predominance of females (66.7%), similar to synovial hemangioma in relation to other sites. Involvement of the flexor side is more common than of the extensor side (2:1).

The clinical manifestation of hemangiomas of the tendon sheath in the hand is nonspecific. Most are palpable masses without discoloration. Pain and tenderness may be caused by local nerve compression or inflammatory response. Plain X-ray film plays no role in the diagnosis of hemangiomas. MRI usually provides more accurate information for diagnosis and surgical planning. The typical presentation of hemangiomas in MRI includes ill-defined margin, heterogeneity, content of fatty tissue (intermediate or high-signal intensity at T1-weighted imaging), high-signal intensity at T2-weighted imaging, strong enhancement with contrast media, vascular content (i.e., fluid-fluid levels), and presence of obvious flow void artifacts. 11 Differential diagnosis of hemangioma should include giant cell tumor of the tendon sheath, aneurysm, vascular malformation, and malignancy. Local invasive properties of giant cell tumor may be helpful in differential diagnosis. In the histological examination of this case, no arterial, venous or lymphatic component or atypism ruled out vascular malformation and malignancy respectively.<sup>12</sup>

The principal treatment for benign tumor in the hand is complete excision. As is known, surgical intervention in zone II of the hand may result in restrictive adhesion to limit gliding of the tendons. In our literature review, however, there was no obvious postoperative adhesion after excision of hemangioma of the tendon sheath. There were two cases with the presence of infiltration of the tendon. Postoperative deformity or some degree of limited range of motion was reported after tumor excision with the involved tendon. <sup>3,4</sup>

Our patient was the only case who had hemangioma of the tendon sheath occurring in the finger distal to the A2 pulley. Unlike

**Table 1**Six known cases of hemangioma in relation to tendon sheath in the hands.

Study	Location	Age (years)	Gender	Symptom/sign	Infiltration to tendon	Operation	Outcome
Webster and Geschickter 1945 <sup>2</sup>	Flexor tendon over left index finger base	22	Male	Nontender Full ROM No discoloration No N/V disturbance	Yes	Tumor excision with tendon sheath removed	Full ROM
Waddell 1967 <sup>4</sup>	Extensor tendon at MP joint of right <u>thumb</u>	27	Female	Tender (+) Bruit (+) No discoloration No N/V disturbance	Yes	Tumor excision with infiltrated EPL/ EPB tendons removed (+ extensor incidis tendon transfer)	Lack of active thumb extension
Spinner et al 1983 <sup>5</sup>	Extensor tendon proximal to MP joint of left middle and ring fingers	33	Female	Recurrence (+) Nontender Full ROM No discoloration No N/V disturbance	Yes	Tumor excision with infiltrated extensor tendons to the middle and ring fingers, and a portion of the third dorsal interosseous muscle + palmaris longus tendon transfer	Full ROM
Rico et al 1994 <sup>6</sup>	Flexor tendon proximal to A1 pulley of right index finger	23	Male	Acute pain (+) Fixed in 40° flexion at MP and PIP and 10° flexion at DIP	No	Tumor excision with tendon sheath removed	Uneventful
Talwalkar et al 2005 <sup>3</sup>	Flexor sublimis tendon at zone II of left <u>little</u> finger	16	Female	Nontender No discoloration No N/V disturbance	Yes	Tumor excision with infiltrated sublimis tendon and a part of A2 pulley removed	20° flexion deformity at PIP joint with full further flexion
Lee et al 2011	Flexor sublimis tendon at proximal interphalangeal joint of left ring finger	14 y/o female	Female	Recurrence(+) Tender(+) Distal numbness(+) No discoloration Full ROM	No	Tumor excision with tendon sheath removed	Full ROM without distal numbness

DIP = distal interphalangeal; EPB = extensor pollicis brevis; EPL = extensor pollicis longus; MP = metacarpo-phalangeal; N/V = neurovascular; PIP = proximal interphalangeal; ROM = range of motion.

previously reported cases proximally to the A1 pulley presenting as trigger fingers, the symptoms in our case were mainly caused by digital nerve compression. The risks in treatment are the possibility of neurovascular bundles and A2 pulley injury. Previous biomechanical studies showed less than 25% excision of the distal portion of the A2 pulley causes little loss of function.<sup>13</sup> There was no weakness noted after operation in our patient.

The reason for rapid recurrence may be incomplete removal and unsuccessful ligation of the feeding vessel at the first operation. We did not clearly determine the origin of the hemangioma, so there might be some residual tumor tissue or feeding vessels beneath the involved tendon sheath. During the second operation, we resected the hemangioma with the involved tendon sheath. We think that it is necessary to excise the involved tendon sheath to achieve complete removal and avoid recurrence. The presence of infiltration of the tendon is difficult to manage, because of high complication rates after excision of the involved tendon. <sup>3,4</sup> Preservation of the tendon may be reasonable as a result of the benign property of hemangioma.

In conclusion, synovial hemangioma involving the tendon sheath in the finger is very rare. Its rarity may lead to inadequate excision and recurrence. Correct diagnosis and complete excision of the tumor with involved tendon sheath is recommended.

#### Disclaimer

No benefits in any form have been received or will be received related directly or indirectly to the subject of this article.

#### Acknowledgments

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