Case Report

Fixed Flexion Deformity of the Middle and Ring Fingers in Adult Caused by Intramuscular Hemangioma of the Forearm: A Case Report

Vilai P, MD[1], Manop P, MD[1], Vanichanon C, MD[1]

[1] Department of Orthopaedics, Faculty of Medicine, Srinakharinwirot University, Nakhon Nayok, Thailand

Intramuscular hemangioma (IMH) of the upper extremity is extremely rare and mostly found in children. The authors presented a 39-year-old female with an unusual case of fixed flexion deformity of the middle finger and ring finger with deep forearm pain for three years. The patient was diagnosed with IMH of the flexor digitorum superficialis muscle. Excision of the IMH was performed and intra-operatively passive extension of PIP joints was achieved. After 12 months follow-up, the patient was satisfied with the outcomes with no recurrence of flexion contracture. IMH of the upper extremity is an uncommon disease, especially in adults. It should be considered in patient who presented with deep forearm pain and finger flexion deformity. Early investigation should be performed and surgical removal remains the treatment of choice. Furthermore, tendon transfer should be reserved in case of functional deficits.

Keywords: Intramuscular hemangioma, Flexion contracture, Forearm, Soft tissue tumor

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Hemangioma is a benign vascular tumor that occurs in 7% of all benign soft tissue tumors. This tumor usually occurs during infancy in superficial structures more than in deep structures. This type of tumor is mostly found in the bones of the head and neck with only 0.8% occurring in muscles[1].

Intramuscular hemangiomas (IMH) are mostly found in the first three decades of life[1-3], and extremely rare in the upper limbs, with cases reported in children[4-8].

The symptoms and signs of the disease remain unclear due to its rarity and pathogenesis. Most patients present with pain. Other symptoms could occur as deformity of the extremities.

Correspondence to:

Manop P.

Department of Orthopaedics, Faculty of Medicine, Srinakharinwirot University, 62 Moo 7 Rangsit-Nakornnayok Road, Ongkarak, Nakhon Nayok 26120, Thailand.

Phone: +66-37-395085

Email: pratchaya.mnop@gmail.com

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In the present case, the authors reported IMH of the forearm in a 39-year-old female who presented with flexion contracture of the middle and ring fingers caused by invasion of the tumor in the extrinsic flexor muscle, and the result after treatment.

Case Report

A 39-year-old female was diagnosed with flexion deformity of the middle and ring fingers of her left hand without history of trauma three years ago (Figure 1). There was gradual increased in the flexion deformity of her fingers. She felt deep pain in her proximal forearm. There was no swelling or palpable mass on her forearm.

Physical examination showed no abnormal discolorations, no mass, no bruits, and no thrills. There was mild tenderness of her proximal forearm. The patient could not passively extend the PIP joint of middle and ring fingers. No abnormal nerve symptoms were observed. Although, no abnormalities were detected on plain radiographs, magnetic resonance imaging (MRI) showed the infiltrative slow flow vascular malformation in deep anterior muscular compartment on medial side of proximal left forearm predominantly involving flexor digitorum superficialis (FDS) muscle, at about 2.4×2.6 cm in cross-section and 6.6 cm in length (Figure 2).

Figure 1. Flexion deformity involving middle and ring finger at PIP joint.

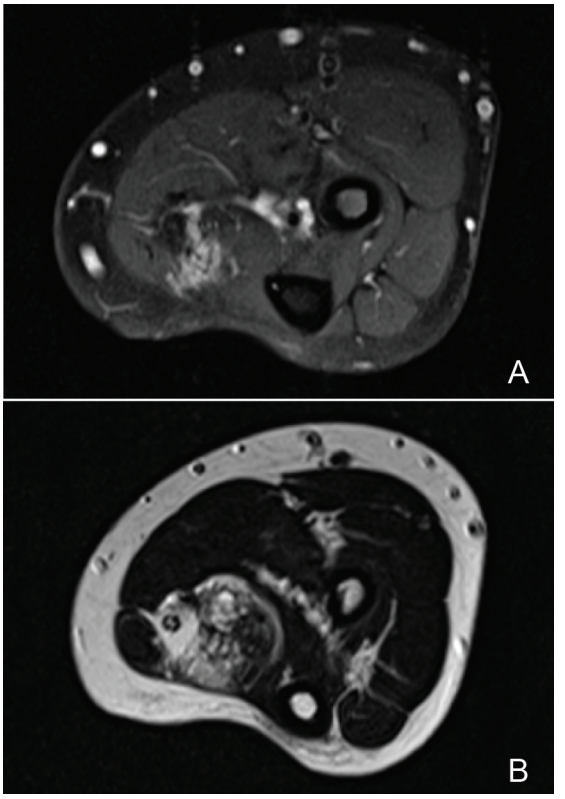

Figure 2. MRI showed intermediate intensely enhancing tortuous soft tissue lesion in deep anterior muscular compartment on medial side of proximal left forearm in T1 (A), high signal tubular structure associated with fatty proliferation and multiple enhancing dilated draining vessels in T2 (B).

In the present case, the author performed excision and removal of the IMH due to bothersome symptoms. Intraoperative finding showed that the flexor muscle was invaded by IMH especially FDS,

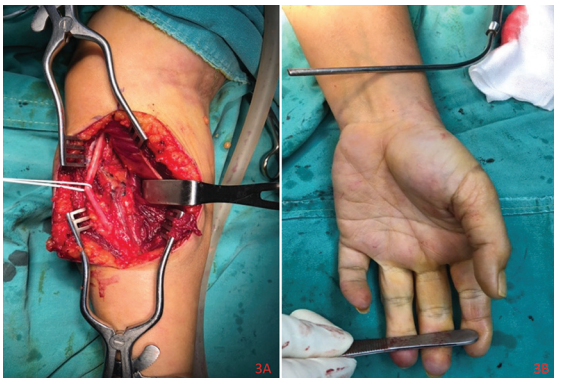

Figure 3. Intramuscular hemangioma was found under the forearm structure near the median nerve (white arrow) (A). Full finger extension were achieved after surgery (B).

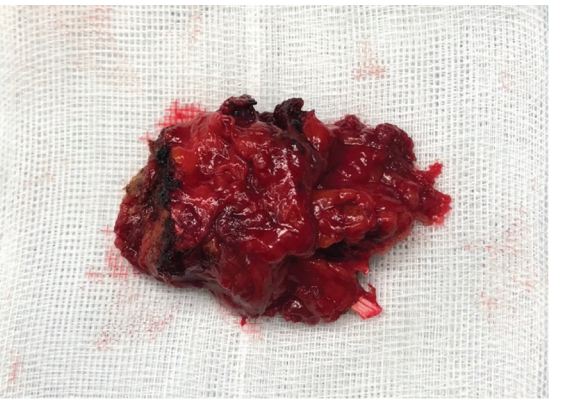

Figure 4. Excised tissue sent for pathological diagnosis.

with no neurovascular involvement (Figure 3A). After excision, both affected fingers could be passively extended (Figure 3B). The lesion was totally removed and sent for pathological diagnosis (Figure 4).

Histological examination review showed intramuscular lesion composed of mixture of thin and thick-walled vessels, a cluster of cavernous vessels, and capillary vessels associated with surrounding mature adipose tissue, compatible with IMH.

After 12 months follow-up, no recurrence was detected, and the patient regained full hand motion (Figure 5).

Discussion

Hemangiomas are benign vascular neoplasms, making up to 7% of all benign soft tissue tumors[1,3,7,9], usually occuring in infants and children[1,3,5]. About 55% of these tumors are presented at birth, with other cases developing in the first few weeks of life[5].

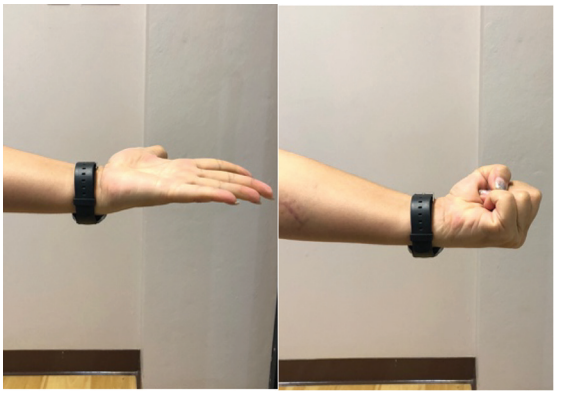

Figure 5. Normal active finger extension both of middle and ring finger after excision.

IMH is a rare type of hemangioma, constituting about 0.8% of all benign hemangiomas[3,8]. They were first described by Liston' in 1843[2]. IMH usually involves muscles of the lower extremity, most commonly the quadriceps muscles[9]. IMH of the forearm is extremely rare and it has been rarely reported in the literature. About 80% to 90% of IMH occurs in the first three decades of life, with no predominance in either gender[1].

IMH has a variety of clinical presentations where some patients may be asymptomatic while others present with pain at site, palpable mass or soft tissue swelling, subcutaneous discolorations, and less frequently, neurologic symptoms or deformity of extremities. Those symptoms are mimicking Dupuytren contracture or Volkmann ischemic contracture. Owing that muscle invasion by IMH is an extremely rare condition, there are only three case reports of this nature in the English literature[6-8]. Median nerve symptoms were also a result of tumor invasion into the nerve and the surrounding tissue that may present with a positive Tinel sign in the wrist[7]. Those reports were all in children[4-8].

In the present case, the deep pain of the forearm and the flexion contracture of the fingers gradually progressed and occurred in an older age, presenting in both the middle finger and ring finger. Although, pain is a common symptom present in 60% of the cases[6], IMH cannot be diagnosed without investigation.

Rounded soft tissue calcification may be seen in plain radiograph but not in all cases. Ultrasound does not reliably identify the pathognomonic features of hemangiomas, phleboliths when they are present because they are seen as an echogenic focus with acoustic shadowing[3]. Computed tomography may be necessary in case of associated bone involvement. The MRI is the modality of choice for IMH[1,3]. The MRI finding can show, in T1-weighted images, high signal intensity reflecting fat content of lesion of hemangiomas, indistinct lesion borders, and areas of signal void indicating muscle atrophy. In T2-weighted images, the MRI can show high signal intensity relative to muscle, multilobulated, "bag of worms" or tubular appearance. Furthermore, the central area of low intensity is highly specific, "dot sign". However, if the diagnosis following clinical examination and imaging were not conclusive, open or needle biopsy is recommended[3].

Conservative management is the first line of treatment in isolated IMH. Surgical excision maybe offered in cases of highly localized, well-circumscribed single-muscle, with minimal loculations or if the patients did not respond to conservative treatments.

Incomplete surgical excision is the greatest risk factor for recurrence, ranging from 18% to 61%, although hemorrhage remains the most common complication[10]. Functional impairment after excision should also be of concern.

Conclusion

IMH of the upper extremity is an uncommon disease, especially in adults. It should be considered for differential diagnosis in patient presenting with deep forearm pain and finger flexion deformity. Early investigation should be performed, and surgical removal remains the treatment of choice. Furthermore, tendon transfer should be reserved in case of functional deficits after operation.

What is already known on this topic?

IMH in the extremities is rare, mostly found in the first three decades of life, with most case reports in children.

The presentation is unclear due to its rarity and pathogenesis. Most patients present with pain. Other symptoms could occur such as deformity of the extremities.

What this study adds?

For patients who present with finger flexion like deformity or deep pain of the forearm, space occupying lesion such as IMH should be one of the differential diagnosis.

Acknowledgement

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Conflicts of interest

The authors declare no conflict of interest.

References

1. Ramon F. Tumors and tumorlike lesions of blood vessels. In: De Schepper AM, Vanhoenacker F, Gielen J, Parizel PM, editors. Imaging of soft tissue tumors. Berlin, Heidelberg: Springer; 2006. p. 263-82.
2. Rossiter JL, Hendrix RA, Tom LW, Potsic WP. Intramuscular hemangioma of the head and neck. Otolaryngol Head Neck Surg 1993;108:18-26.
3. Wierzbicki JM, Henderson JH, Scarborough MT, Bush CH, Reith JD, Clugston JR. Intramuscular hemangiomas. Sports Health 2013;5:448-54.
4. Shajrawi I, Dreyfuss UY, Stahl S, Boss JH. Intramuscular haemangioma of the forearm. J Hand Surg Br 1990;15:362-5.
5. Vakilha M, Farhan F, Samiei F, Shariat S. Intramuscular hemangioma of the forearm: report of a case. Iran J Radiat Res 2003;1:175-9.
6. Sunil TM. Intramuscular hemangioma complicated by a Volkmann's like contracture of the forearm muscles. Indian Pediatr 2004;41:270-3.
7. Eyesan SU, Itie JC, Adesina SA, Kehinde GT, Olasode BJ. Cavernous hemangioma of the left forearm. Rare Tumors 2015;7:5895.
8. Kamath JB, Jayasheelan N, Shenoy T, Bansal A, Shetty K, Sujir P. Surgery with timed wake-up anesthesia for hemangioma of ring finger flexor digitorum superficialis. Online J Health Allied Sci 2017;16:1-2.
9. Patnaik S, Kumar P, Nayak B, Mohapatra N. Intramuscular arteriovenous hemangioma of thigh: A case report and review of literature. J Orthop Case Rep 2016;6:20-3.
10. Bella GP, Manivel JC, Thompson RC Jr, Clohisy DR, Cheng EY. Intramuscular hemangioma: recurrence risk related to surgical margins. Clin Orthop Relat Res 2007;459:186-91.