



Dual Cavernous Hemangiomas in the Foot: A Case Study

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Purpose

Hemangiomas are a rare benign soft tissue tumor. Throughout the body they represent 7-10% of all soft-tissue tumors and intramuscular hemangiomas represent 0.8% of all hemangiomas. Forty-two to forty-five percent of intramuscular hemangiomas are found in the lower extremity [2] Malignant transformation is possible but rare. Presentation is usually seen before the age of 30 and is slightly more common in females.[4] Diagnosis is made using MRI and confirmed by pathologic examination of tissue specimen.[4] This poster presents a rare case where both an intramuscular and intraosseous hemangioma were present and discusses the surgical treatment necessary.

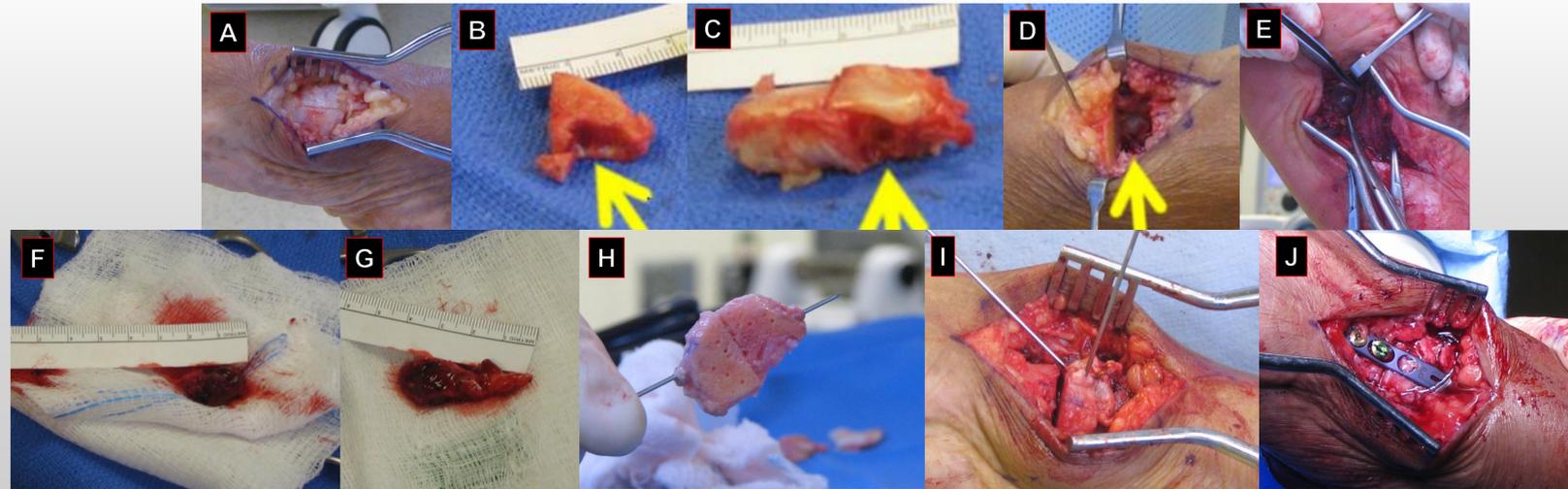
Case Study

The patient is a 76 year old female with a recent history of breast cancer, HTN, and HLD who presented to clinic with pain in the dorsal midfoot for several years. The pain was a sharp 6/10 pain, localized to the dorsal medial midfoot. She had failed numerous conservative therapies including cortisone injections, orthotics and heat therapy. The only relief she received was from NSAIDS, which she took regularly. On exam she was found to have pain along the entire midfoot, with pain being equal both medially and laterally. She also had pain in the plantar arch. There was edema across the midfoot, particularly over the first metatarsocuneiform joint laterally. A MRI was obtained detailing a 3.3 x 1.0 x 1.3cm mass in the flexor digitorum and another mass measuring 1.3 x 1.7 x 1.7cm in the 1st intermetatarsal space. To complete the workup, an additional CT was obtained to assess the articular changes to the joint, and a MRA was ordered to rule out AV malformation. A CT-guided needle biopsy was attempted and unsuccessful at diagnosing the masses. Patient was taken for surgical correction and to obtain biopsies of the masses.

Pre-Operative Imaging



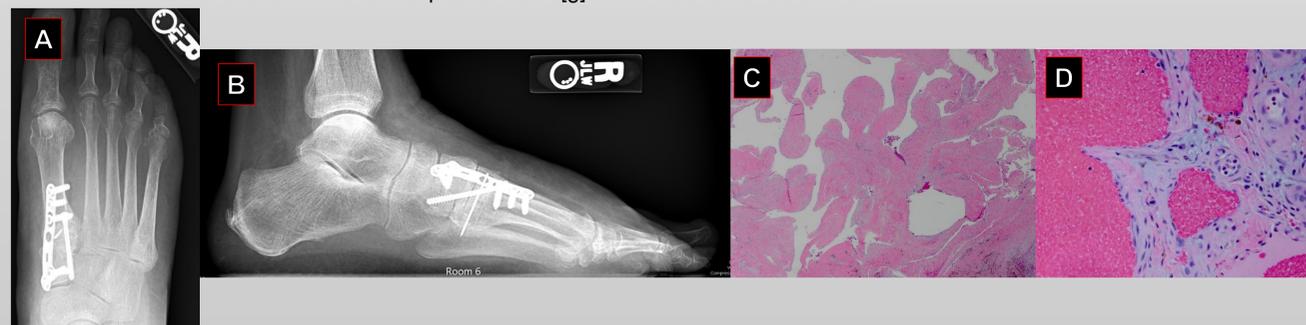
A: Initial right foot radiograph showing a cyst in the base of the first metatarsal. (Yellow Arrow)
 B: Pre operative CT showing lesion in the 1 MTCJ. (Yellow Arrow)
 C: Pre operative MRI demonstrating enhancing intramuscular mass in the plantar forefoot and 1st MTCJ (Yellow Arrow)



(a) Normal medial appearing joint (b) resected 1st metatarsal base to gain access (c) notch where the tumor eroded the plantar lateral 1st metatarsal base (d) erosion of the medial cuneiform (e) purple cluster of grapes appearance (f) plantar incision showing the intramuscular hemangioma (g) excised lateral 1st MTCJ hemangioma (h) excised plantar hemangioma (i) 1st metatarsal base reconstructed (j) temporary fixation (k) final fixation

Procedure

Incision was made through a medial approach revealing a normal appearing first metatarsocuneiform joint [a]. The metatarsal base was removed and the mass was revealed. The interosseous mass had eroded the lateral portion of the 1st metatarsal base [b] and medial cuneiform [c]. The mass appeared as a cluster of grapes. After mass removal, a bony void [d] was present. Cartilage of the arthrodesis site was removed. The bone void was reconstructed with the combination of healthy bone from the removed metatarsal base and a combined calcium sulfate and calcium phosphate [h]. Fixation and arthrodesis of the MTC joint was obtained with a dorsal locking plate and interfragmentary screw [j]. The mass in the plantar musculature was excised through a plantar incision [e] and was found to envelope an intramuscular branch of the lateral plantar nerve [g] which was removed as well.



A/B: Post-operative AP/lateral radiographs at 7 months showing consolidation of the previous tumor area
 C: 10x Histologic picture of the intramuscular mass showing cystic spaces lined by benign endothelial cells filled with RBCs
 D: 40x Histologic picture of the interosseous soft tissue mass showing thin walled vessels making up much of the vascular proliferation. Benign appearing endothelial cells
 *Photos and histologic description by Dr. Joseph Milo Hibbert, DO, Pathology Resident

Results

Pathology results confirmed that the mass in the flexor digitorum muscle belly and the 1st intermetatarsal space were both benign cavernous hemangiomas. The intramuscular mass on histology demonstrated large cystic spaces lined with endothelial cells. The interosseous mass presented with thin walled vessels making up much of the vascular proliferation and benign appearing endothelial cells without atypia. Following surgery the patient was treated with a 12 week course of NWB until fusion was obtained. Patient has returned to regular activity without pain. She has had no recurrence of the hemangioma at 8 months post operatively.

Disclaimer: The views expressed are those of the author and do not reflect the official policy or position of the Department of the Army, DOD, or the U.S. Government.

Author	Age	Sex	Location	Treatment	Recurrence
Zheng	71	F	Dorsum of the foot	Surgery with tumor free margins	6 months
Saste	29	M	3rd metatarsal mid-distal shaft/shaft intertrochanteric region of the femur	1) Wide surgical excision 2) Adjuvant radiotherapy 3) chemotherapy	None
Chang	11	F	FDB muscle belly	En-bloc excision	None at 6 year follow up
	15	M	FDB muscle belly	En-bloc excision	None
Mitsionis	12	M	FDB muscle belly	Excision with wide margins	Not reported
Kramer	10	F	Right ankle and foot	Sclerotherapy	Asymptomatic
Bisbinas	41	M	Left foot and ankle	1) Curettage 2) Excision	1) Yes with curettage 2) 5.5 yrs w/o recurrence after excision
Uslu	7	F	FDB muscle belly	Excision	None at 22 months
	9	M	Plantar foot	Percutaneous sclerotherapy	Asymptomatic after 16 months

Discussion

-Hemangiomas are benign soft tissue tumors comprising 7-10% of all soft tissue tumors body wide and account for 1% of all primary bone tumors.[4] Intramuscular hemangiomas comprise 0.8% of all hemangiomas. Half of intramuscular hemangiomas are found in the lower extremity, most commonly in the thigh.[4] It is rare to see soft tissue hemangioma at the 1st metatarsal cuneiform level as presented in this poster.

-Excision with wide margins has been advocated as standard treatment[4,7] but other options have been explored. Sclerotherapy has been used to treat soft tissue hemangiomas of the foot. It has advantages of being less invasive and used more than once. Sclerotherapy does have complications including skin necrosis, allergic reaction, hyperpigmentation, thrombosis of normal veins, intra-arterial injection and neurolysis.[7].

-Recurrence rates have proven to be high at 18-61%.[7] In the table above, a review of the literature shows recurrence in 2 out of 8 cases of hemangiomas found in the foot and ankle. Due to risk of recurrence, long-term surveillance will be necessary. Therefore, repeat MRI is planned for 1 year from surgery.

-Future research is needed to evaluate various treatment methods to reduce recurrence rates for this rare tumor type.

References

1. Bisbinas I, Karabouta Z, Georgiannos D, Lampridis V, Badekas A. Multifocal epithelioid hemangioendothelioma of the foot and ankle: a case report. J Orthop Surg (Hong Kong). 2014 Apr;22(1):122-5
2. Chang JJ, Lui TH. Intramuscular haemangioma of flexor digitorum brevis. Foot Ankle Surg. 2010 Jun;16(2):e8-11.
3. Kramer D, Downey C, Vargas P, Castro A. Multifocal spindle cell hemangioma: Report of two cases. Indian J Dermatol Venereol Leprol. 2016 Jan-Feb;82(1):93-5. doi: 10.4103/0378-6323.172907.
4. Mitsionis GI, Pakos EE, Kosta P, Batistatou A, Beris A. Intramuscular hemangioma of the foot: A case report and review of the literature. Foot Ankle Surg. 2010 Jun;16(2):e27-9.
5. Ozsahin M, Uslu M, Inanmaz E, Buyukkaya R, Erdem H. Intramuscular hemangioma of flexor digitorum brevis muscle. Am J Phys Med Rehabil. 2012 Oct;91(10):910.
6. Saste A, Cabrera Fernandez DF, Gulati R, Gamalski S. A trimodality approach in the management of metastatic low-grade epithelioid hemangioendothelioma of the bone. BMJ Case Rep. 2015 Jul 16:2015.
7. Uslu M, Besir H, Turan H, Bozkaya H, Erdem H. Two different treatment options for intramuscular plantar hemangioma: surgery versus percutaneous sclerotherapy. J Foot Ankle Surg. 2014 Nov-Dec;53(6):759-62.
8. Zheng LQ, Han XC, Huang Y, Fan JY. Cutaneous retiform hemangioendothelioma on the right foot with an unusual clinicopathological feature. Am J Dermatopathol. 2014 Sep;36(9):757-9.