CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 77 (2020) 682–685

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Intramuscular haemangioma of abductor hallucis muscle – A rare case report

Dimas Radithya Boedijono [a], Andi Praja Wira Yudha Luthfi [b,*], Erlina [c]

[a] Department of Orthopaedics and Traumatology, Fatmawati General Hospital, Jakarta, Indonesia
[b] Department of Orthopaedics and Traumatology, Bhayangkara Tk.I R. Said Sukanto Police Hospital, Jakarta, Indonesia
[c] Department of Pathological Anatomy, Fatmawati General Hospital, Jakarta, Indonesia

ARTICLE INFO

Article history:
Received 20 October 2020
Received in revised form 2 November 2020
Accepted 8 November 2020
Available online 24 November 2020

Keywords:
Haemangioma
Intramuscular Haemangioma
Abductor hallucis muscle

ABSTRACT

INTRODUCTION: Haemangioma is a slow growing benign soft tissue tumor and its presentation in the foot is rare. Intramuscular haemangioma (IH) are usually found before 30 years of age, with gender predominance is still inconclusive.
PRESENTATION OF CASE: An 18-year-old woman came with pain and mass in the left foot for the past 3 years. Magnetic Resonance Imaging (MRI) of the left foot shown a heterogenous multilobulated mass, with previously thought originated from flexor digitorum brevis (FDB) muscle. Wide excision was performed and intraoperative findings showed that the mass actually originated from abductor hallucis muscle. Post-operative histopathological findings confirmed the diagnosis of cavernous-type of intramuscular haemangioma.
DISCUSSION: The rare occurrence of intramuscular haemangioma of the foot can cause a delayed diagnosis and treatment to the patient. The differential diagnosis include lipoma, fibroma, enlargement of the lymph nodes, compartment syndrome, hematoma, hernia, and soft-tissue sarcoma. Anytime a soft tissue mass is identified in the skeletal muscle of a young adult, haemangioma should be considered.
CONCLUSION: Literature research identified very few cases of intramuscular haemangioma of the foot. Wide excision of the muscle is a feasible surgical treatment option.

1. Introduction

Haemangioma is a slow growing benign soft tissue tumor, which makes up for 7–10% of all soft tissue tumors. Despite possible acceleration in growth due to growth spurt or trauma, haemangiomas will regress spontaneously with only rare cases of malignant transformation [1,2]. Intramuscular haemangioma (IH) are usually found later in life, before 30 years of age, as opposed to cutaneous haemangiomas predominantly found in young children [3]. The prevalence between genders are still inconclusive, with some studies showing higher prevalence in female while others found no difference between male and female. Approximately 55% of IH present with pain and swelling, the symptoms of which usually occur for 1–5 years. In 98% of cases, a mass was found during physical examination [2,4]. Muscle contraction was found to have an effect on lesion size. The mass could move transversely, but not in line with the fibers. IHs are mostly found in the lower limb (42–45%), specifically the thigh. Latest literatures currently have very limited amount of case reports regarding intramuscular haemangiomas found in the foot [2]. This paper will discuss the case of an intramuscular cavernous haemangioma of the abductor hallucis muscle of the left foot of an 18-year-old female who received surgical excision. To the best of our knowledge, this is the first paper from Indonesia reported intramuscular haemangioma of abductor hallucis muscle.

2. Case report

An 18-year-old woman came to the outpatient clinic with a history of pain in the left foot for the past 3 years. The patient was an active student with no recent history of trauma. Aside from pain, there was swelling in the medial plantar side of her left foot. The mass started out small but slowly progressed to the size of a quail egg. The pain was said to be 7/10 in ratings. The patient had previously consulted an orthopaedic surgeon in another hospital. She had received surgical excision 3 years prior, but evidently, the mass reappeared. There was no history with same condition in her family, and she ignored to take regular medication previously. The patient thought that the first surgery had not finished yet.

The patient appeared to be of healthy condition without any signs of acute stress. She had non-antalgic gait with a slightly high-

* Corresponding author at: Department of Orthopaedic and Traumatology, Bhayangkara Tk.I R. Said Sukanto Police Hospital, Jl. RS Polri Sukanto, Kramat Jati, Jakarta Timur, Jakarta, Indonesia.
E-mail address: wirapraja.med@gmail.com (A.P.W.Y. Luthfi).

https://doi.org/10.1016/j.ijscr.2020.11.035

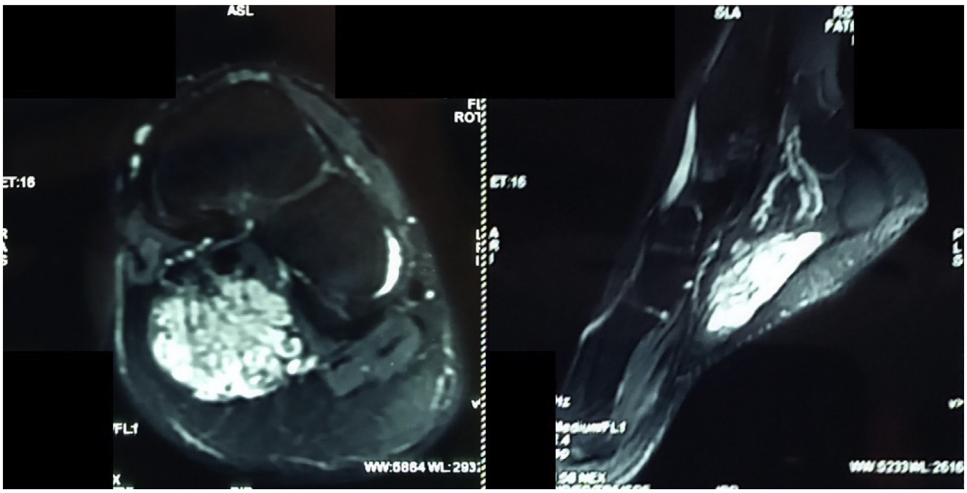

Fig. 1. Coronal and sagittal view of the patient's left foot MRI showed a heterogenous multilobulated mass.

arched feet. The surface of the left foot plantar was mildly swollen in the origin of the plantar fascia, which extended to the arch of the foot. The color, temperature, and capillary refill time were within normal range. There was tenderness at the distal of the plantar fascia with no evidence of tenderness elsewhere. The passive range of motion of the left foot at the ankle was found to be symmetric and the midtarsal joints also showed no abnormality during passive movement. The motoric strength of the lower extremity was optimal and both legs showed no signs of pain. There wasn't any pain elicited by passive dorsiflexion of the ankle and the foot thumb. Normal sensation and reflex were found in both legs and Tinel's test showed negative results at the tarsal tunnel. Initial radiographs showed no signs of abnormality.

Magnetic Resonance Imaging (MRI) of the left foot was conducted, the showed a heterogenous multilobulated mass (Fig. 1). The expertise of the MRI stated that the mass originating from the flexor digitorum brevis (FDB) muscle. Some part of the mass was located around the plantar medial nerve and artery. There was an increased signal demonstrated in T1 and T2 images. These findings were first considered suggestive of malignancy.

A surgical excision was conducted by the first author, who has more than 10 years of experience in foot and ankle surgery. A longitudinal approach across the medial border of the foot was made, which facilitated the exposure of the abductor hallucis muscle and FDB muscle. Intraoperatively, the mass found within the multilobulated lesion was in abductor hallucis muscle (Fig. 2).

The lesion was completely removed through excision with wide surgical margins. Histological examination showed vascular channels within the skeletal muscle, based on the size of the vessels, it is classified as cavernous-like spaces (Fig. 3). The patient was then mobilized using two crunches and partial weight bearing for around 3 weeks post-operatively, with weekly visit to the clinic to evaluate any improvements of her complaints. There was no restriction in movement of the toes with no signs of deformities. The foot arch was maintained, there was no complaint of sole pain.

This case report has been reported in line with the Updating Consensus Surgical Case Report (SCARE) 2018 criteria [5].

3. Discussion

Haemangioma of the skeletal muscle is the most common type of deep soft tissue tumor and also the most common type of benign tumor found in the muscle. It is widely known as a benign yet locally aggressive tumor comprising of ectatic blood vessels. They commonly can be distinguished from deep-seated sarcoma of the soft tissue through MRI. Nevertheless, it might be difficult to distinguish from other types of benign vascular proliferations. They are thought to be caused by trauma but are most likely congenital [6,7].

Plenty of case reports refer other types of benign vascular tumor as haemangioma, including arteriovenous malformations, intramuscular haemangioma, lymphangioma, and juvenile capillary haemangiomas, but it is important to note that each type differ in clinical presentation and management [6]. Pain is considered to be the main symptom, found in 60% of cases, while a mass was present in 98% of the case. Our patient complained of the two major symptoms for 3 years despite having had surgery. These symptoms could be difficult to distinguish from other conditions such as plantar fasciitis. A case of acute compartment syndrome due to haemangioma was reported in a paper, which came as a surprise for surgeons as no recent trauma was reported, and intramuscular haemangioma of the foot itself is quite rare [8].

The rare occurrence of IH of the foot is the reason for the patient's delay in getting proper treatment for 3 years, having had prior surgical excision. Plenty of studies reported delayed diagnosis for months and even years [3,7]. The differential diagnosis include lipoma, fibroma, enlargement of the lymph nodes, compartment syndrome, hematoma, hernia, and soft-tissue sarcoma. IH should be considered in patients with soft tissue mass and swelling or in cases of no improvement with traditional care [3].

In 25% cases, a calcified thrombus known as phlebolith are present. These phleboliths are better identified using radiograph and Computed Tomography (CT) rather than MRI. With haemangioma present, bones near the site of lesion could show periosteal reaction which needs to be differentiated from X-ray findings of osteomyelitis or bone tumors [7].

Anytime a soft tissue mass is identified in the skeletal muscle of a young adult, haemangioma should be considered. The probability of haemangioma rises if USG examination shows hyperechoic lesion. Through the use of technetium-99 m methylene diphosphonate (MDP) bone scan, intramuscular haemangioma shows a rise of absorbance during the early scintigraphy phase, which could possibly be the result of hyperemia of the surrounding structures with activation and parasitizing of blood vessels [9].

Each modality has their own advantage in the diagnosis of soft tissue tumors, but when it comes to tumors of the soft tissues and soft tissues of the bone with extraosseous extension, MRI is superior. With MRI, high signal intensity on T1- and T2-weighted images could be identified, which is different from that of other soft tissue

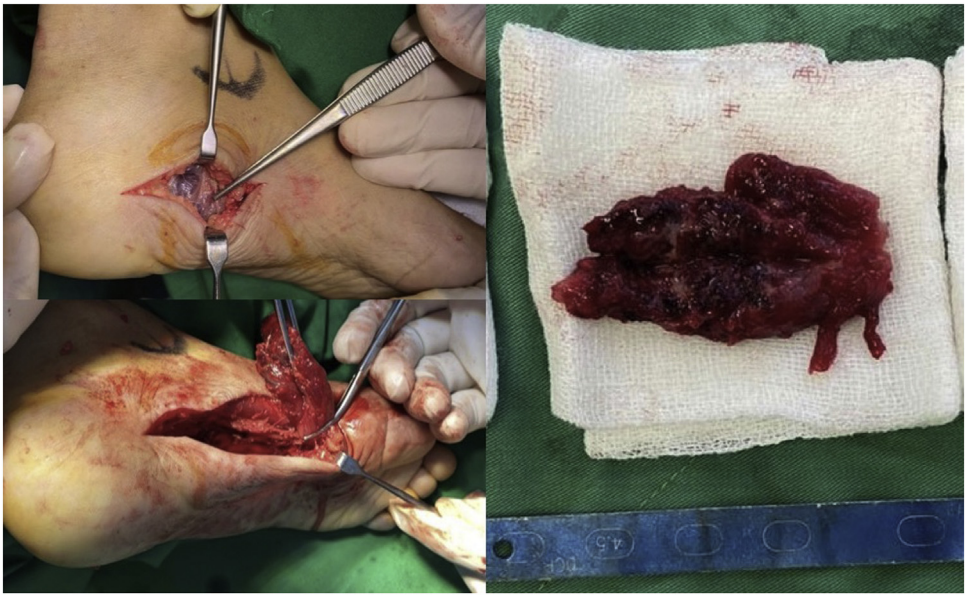

Fig. 2. Red-bluish colored mass was found intraoperatively. En-bloc excision of the abductor hallucis muscle was performed.

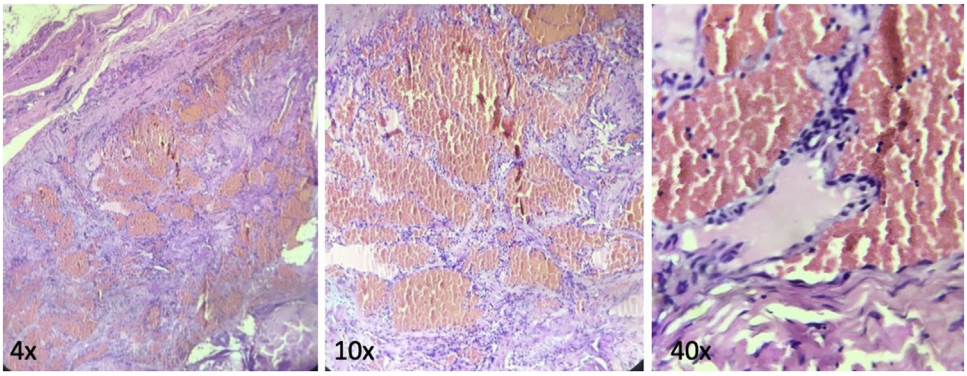

Fig. 3. Histopathological findings with low magnification showed some vascular channels within skeletal muscles with mature adipose tissue (haematoxylin and eosin).

tumors with intermediate signal intensity on T1-weighted images and a high intensity on T2-weighted images [9,10].

In the patient discussed in this case, no abnormalities were identified on plain X-ray. The expertise of the MRI examination in this case showed that the mass was originating from FDB muscle, which later we found the mass was originating from abductor hallucis muscle intraoperatively. This could be happened because the location of abductor hallucis muscle and FDB muscle was near and was sometimes overlap each other on MRI. The MRI could also differentiate between IH and soft tissue sarcoma with the appearance of lobulated and septate found in haemangiomas and not present in soft tissue sarcoma.

In cases of asymptomatic or mildly presenting symptoms of IH, observation should be considered as the management of choice. Indications for further treatment include persistent pain, progressive increase of the lesion size, functional impairment, and patient's concern [11]. The surgical options include partial excision, wide excision, and other choices such as sclerotherapy. It is important to note the recurrence rate of 18%, which was found on partial excision reported on a previous study, while total excision showed significantly better outcome, especially in cases of localized lesion. In cases of high risk in performing resection, such as site of lesion near important neurovascular structures, alternative treatments should be considered. Amputation poses as the last option [2,3,12]. In this case, wide excision was successfully conducted, and the diagnosis was confirmed through histopathological examination.

The challenges we met in this study includes a very rare case, which can lead to a misdiagnosed case and undertreatment. It is important to remember the medial plantar nerve innervates the abductor hallucis muscle. Damage to this structure poses as a possible complication. A long-term follow-up is highly recommended to check on mass recurrence and foot function.

4. Conclusion

Despite the rarity of intramuscular haemangioma of the foot, they should be considered as a differential diagnosis. A team approach is needed to established a correct diagnosis and treatment. The literature research conducted by the author identified very few cases with the same location. Wide excision of the muscle is a feasible surgical treatment option.

Declaration of Competing Interest

The authors declare no conflicts of interest.

Funding

The authors report no external source of funding during the writing of this article.

Ethical approval

Ethical approval was not required in the treatment of the patient in this report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Andi Praja Wira Yudha Luthfi contributes to the study concept or design, data collection and writing the paper.

Dimas Radithya Boedijono contributes to the study concept or design, data collection, analysis and interpretation, oversight and leadership responsibility for the research activity planning and execution, including mentorship external to the core team.

Erlina contributes to the data collection, analysis and interpretation, and oversight for the research activity planning and execution.

Registration of research studies

This is a case report which does not need registration of research study.

Guarantor

Dimas Radithya Boedijono is the sole guarantor of this submitted article.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgment

The corresponding author, on behalf of all authors, declares that there is no conflict of interest.

References

[1] R.A. Brown, K. Crichton, G.M. Malouf, Intramuscular haemangioma of the thigh in a basketball player, Br. J. Sports Med. 38 (June (3)) (2004) 346–348.
[2] J.J.H.T. Chang, T.H. Lui, Intramuscular haemangioma of flexor digitorum brevis, Foot Ankle Surg. 16 (June (2)) (2010) e8–11.
[3] S. Patnaik, P. Kumar, B. Nayak, N. Mohapatra, Intramuscular arteriovenous hemangioma of thigh: a case report and review of literature, J. Orthop. Case Rep. 6 (5) (2016) 20–23.
[4] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A.J. Fowler, D.P. Orgill, et al., The SCARE 2018 statement: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 60 (December) (2018) 132–136.
[5] G.P.B. Bella, J.C. Manivel, R.C. Thompson, D.R. Clohisy, E.Y. Cheng, Intramuscular hemangioma: recurrence risk related to surgical margins, Clin. Orthop. Relat. Res. 459 (459) (2007) 186–191.
[6] A.T. Wild, P. Raab, R. Krauspe, Hemangioma of skeletal muscle, Arch. Orthop. Trauma Surg. 120 (3–4) (2000) 139–143.
[7] F.J. Downey-Carmona, P. González-Herranz, C. De La Fuente-González, M. Castro, Acute compartment syndrome of the foot caused by a hemangioma, J. Foot Ankle Surg. 45 (1) (2006) 52–55.
[8] S.J. Wisniewski, K. Newcomer, A.W. Stanson, Intramuscular hemangioma of the foot: a diagnostic dilemma, Med. Sci. Sports Exerc. 37 (October (10)) (2005) 1655–1657.
[9] N. Griffin, N. Khan, J.M. Thomas, C. Fisher, E.C. Moskovic, The radiological manifestations of intramuscular haemangiomas in adults: magnetic resonance imaging, computed tomography and ultrasound appearances, Skeletal Radiol. 36 (November (11)) (2007) 1051–1059.
[10] M. Ozsahin, M. Uslu, E. Inanmaz, R. Buyukkaya, H. Erdem, Intramuscular hemangioma of flexor digitorum brevis muscle, Am. J. Phys. Med. Rehabil. 91 (October (10)) (2012) 910.
[11] G.I. Mitsionis, E.E. Pakos, P. Kosta, A. Batistatou, A. Beris, Intramuscular hemangioma of the foot: a case report and review of the literature, Foot Ankle Surg. 16 (2) (2010) e27–9.
[12] J.M. Wierzbicki, J.H. Henderson, M.T. Scarborough, C.H. Bush, J.D. Reith, J.R. Clugston, Intramuscular hemangiomas, Sports Health 5 (September (5)) (2013) 448–454.