

# Intravascular papillary endothelial hyperplasia occurring in the foot

## A case report and review of literature

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### Abstract

**Rationale:** Intravascular papillary endothelial hyperplasia (IPEH; Masson tumor) is a type of vascular lesions composed of reactive proliferation of endothelial cells that occur in organizing thrombus. It commonly occurs on the head, neck, trunk, and upper extremities, but rarely in the foot.

**Patient concerns:** A 38-year-old woman visited the hospital with a mass on the dorsum of right foot, which gradually increased in size 3 months ago.

**Diagnoses:** Ultrasonographic examination suggested angiomyolipoma or hemangioma.

**Interventions:** The patient underwent excision under local anesthesia.

**Outcomes:** The lesion was confirmed to be IPEH by histological examination. There were no complications or recurrences after successful surgical excision.

**Lessons:** IPEH presenting on the dorsum of the foot is vary uncommon. Radiologic diagnosis may be limited for diagnosis, and histologic confirmation should be made after surgical excision. There are some reports suggesting an association between trauma and IPEH occurrence, but this is not yet conclusive.

**Abbreviation:** IPEH = intravascular papillary endothelial hyperplasia.

**Keywords:** foot, intravascular papillary endothelial hyperplasia, masson tumor, trauma

## 1. Introduction

Intravascular papillary endothelial hyperplasia (IPEH; Masson tumor) is a type of vascular lesion consisting of reactive proliferation of endothelial cells occurring in organizing thrombus.<sup>[1]</sup> It occurs most frequently in the head, neck, trunk, or upper extremities, but its occurrence in the foot is rare.<sup>[2]</sup> Herein, we present a case of IPEH occurred on the dorsum of foot with a review of literature.

Editor: N/A.

Consent for publication: Informed written consent was obtained from the patient for publication of this case report and accompanying images.

This work was supported by the Soonchunhyang University Research Fund.

The authors have no conflicts of interest to disclose.

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Medicine (2019) 98:28(e16391)

Received: 25 December 2018 / Received in final form: 9 May 2019 / Accepted: 13 June 2019

<http://dx.doi.org/10.1097/MD.00000000000016391>

## 2. Case report

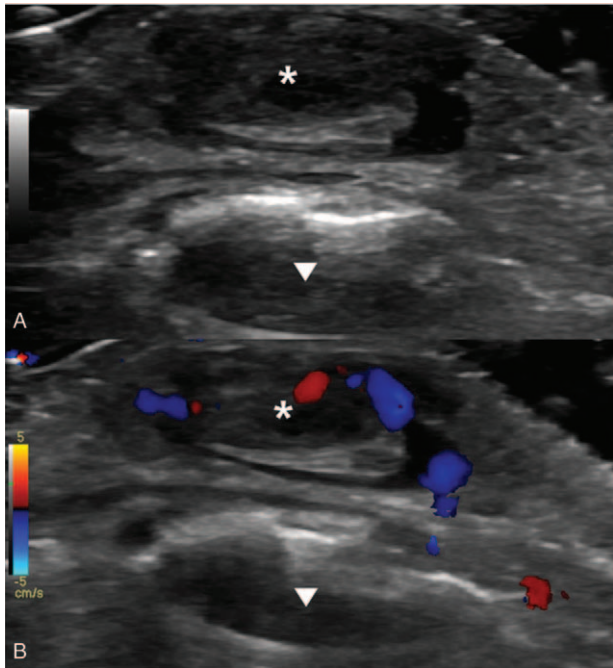
A 38-year-old female patient was referred to the hospital due to an asymptomatic mass on the right foot. It appeared 3 months ago and gradually increased in size. No trauma history to the lesion was mentioned.

A physical examination revealed that a 2 cm sized, soft movable mass was located on the dorsal aspect of the right forefoot above the second metatarsal bone. There were no significant changes on the overlying skin, such as erythema or erosion. There was a mild tenderness induced by the direct palpation, but digital pressure could not decrease the size of the mass. No lymphadenopathy was noted at the foot or right lower extremity.

Ultrasonographic examination revealed a 1.8 × 1.6 × 0.6 cm-sized, fairly well-defined, ovoid mass in the soft tissue of dorsum of the right foot. It was a heterogeneously hypoechoic mass with rich vascularity, suggesting angiomyolipoma or hemangioma (Fig. 1).

An excision was done under local anesthesia with aseptic conditions, and the mass was located in the dermis and subcutaneous tissue with no connection to the underlying fascia or tendons. Its feeding vessels were ligated before excision. After an excision, hemostasis was achieved and the surgical site was primarily closed using 4–0 prolene sutures (Fig. 2).

The largest diameter of the excised mass was measured as 2 cm, and its cut section seemed to have enlarged vessels with thrombus. Microscopically, an enlarged blood vessel with an intravascular thrombus was observed. The center of the thrombus showed anastomosing channels lined by benign endothelial cells with focal papillary growth (Fig. 3). No cellular



**Figure 1.** (A) Ultrasonographically, there was a 1.8 × 1.6 × 0.6 cm-sized, fairly well-defined, ovoid mass in the dorsum of right foot above second metatarsal bone. It is a heterogeneously hypoechoic mass (\*: mass, ▼: second metatarsal bone of right foot). (B) Doppler ultrasound reveals rich vascularity of the mass.



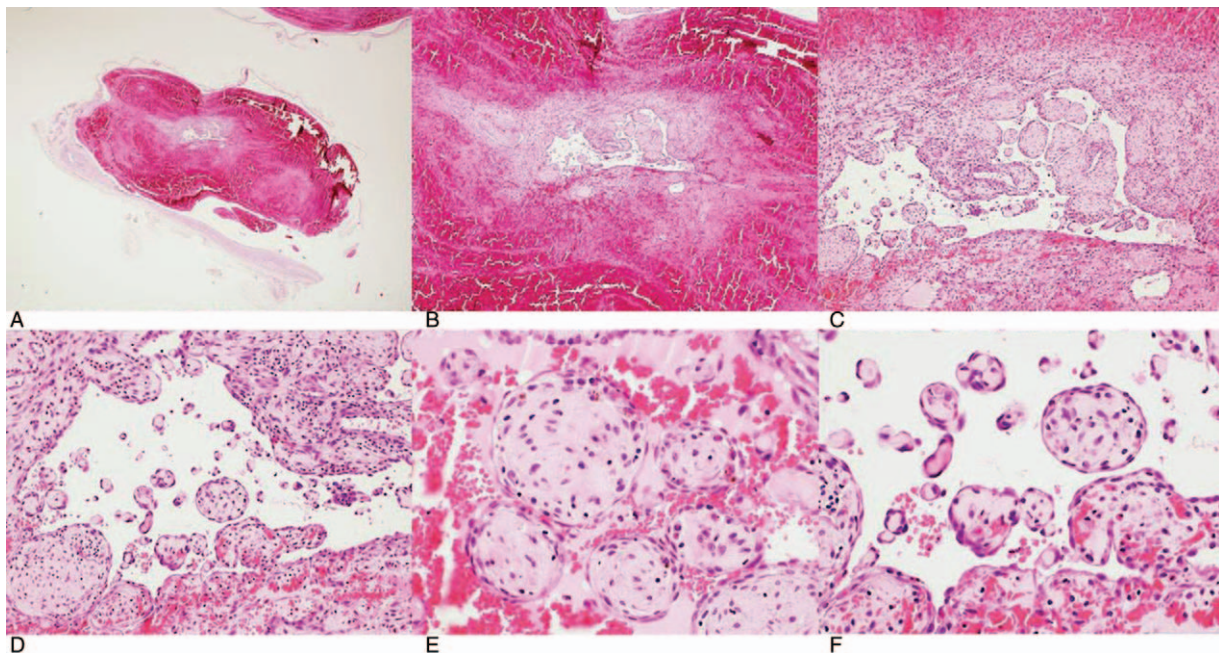
**Figure 2.** On operative field, mass measures 2 cm in length and seems to be having dilated blood vessel with intraluminal thrombus.

### 3. Discussion

In 1923, Pierre Masson first described a peculiar angiosarcoma-like appearance within thrombus in hemorrhoid plexus and termed it as “hemangioendotheliome vegetant intravasculaire,” regarding it as a true neoplasm.<sup>[3]</sup> However, Clearkin and Enzinger<sup>[2]</sup> recognized it as a reactive lesion rather than a neoplastic process. Although IPEH occurs commonly on the skin and subcutaneous tissue of head, neck, trunk, and upper extremities, it also occurs in the adrenal gland, heart, intestine, kidney, liver, and retroperitoneum.<sup>[4–9]</sup> A few cases of IPEH developed in foot were reported as summarized in Table 1.<sup>[10,11]</sup>

The main pathological problem might be its similarity with malignant vascular tumors like angiosarcoma or Kaposi sarcoma. However, intravascular location, papillary structures supported by the core of thrombus, lack of cellular atypia, and

atypia of endothelial cells or mitotic features were found. It was confirmed as IPEH. She has complained no complication within 2 years.



**Figure 3.** (A) A dilated vein with intraluminal organizing thrombus is seen. (H&E, ×12.5). (B–D) A detailed view of organizing thrombus shows papillary processes and anastomosing channels at the central portion. (H&E, (B): ×40, (C): ×100, (D): ×200). (E and F) Papillary processes are lined with benign endothelial cells without cellular atypia or mitotic figures. (H&E, (E) and (F): ×400).

**Table 1****Review of the literature on the intravascular papillary endothelial hyperplasia cases occurred in foot.**

Author, y	No. of case	Age/Sex	Location	Size	Duration	Trauma history	Treatment and prognosis (if stated)
Fink et al <sup>[11]</sup>	1	44/F	Left foot, plantar	2 cm	Insidiously, over 2 years	(-)	Excised, no recurrence in followed 2 years
Hashimoto et al <sup>[13]</sup>	3	N/D	Toe ×1, Foot ×2	N/D	N/D	N/D	N/D
Clarkin and Enzinger <sup>[2]</sup>	4	N/D	Foot ×4	N/D	N/D	N/D	N/D
Kato <sup>[14]</sup>	2	49/F	Right heel	1 cm	2 decades	Suspected	Excised, N/D
Espinosa et al <sup>[10]</sup>	1	23/F	Left foot, 1 <sup>st</sup> toe, dorsum	0.8 cm	2 week	(-)	Excised, No recurrence in followed 4 years
Yamamoto et al <sup>[15]</sup>	1	50/M	Between 4 <sup>th</sup> and 5 <sup>th</sup> toe	1.5 cm	Unknown	(+)	Excised, recurred, twice
Miyamoto et al <sup>[16]</sup>	1	45/M	Left sole	2.3 cm	2–3 years	N/D	N/D
Uchiyama et al <sup>[17]</sup>	1	34/M	Right sole	0.8 cm	2 years	N/D	N/D
Kitagawa et al <sup>[18]</sup>	1	8/F	Left sole	2 cm	3 months	N/D	N/D
Takahashi et al <sup>[19]</sup>	1	46/M	Left sole	0.9 cm	40 years	N/D	N/D
Shirai et al <sup>[20]</sup>	1	80/F	Right sole	N/D	6 months	(+)	N/D
	1	46/F	Right sole	N/D	8 months	N/D	N/D

N/D = not described.

rare but normal mitotic figures might be the histologically essential characteristics for the differential diagnosis.<sup>[11]</sup>

The pathogenesis of IPEH is unclear. Several investigators have described that this reactive process could be related to the antecedent history of trauma of endothelium and consequently formed thrombus.<sup>[1,2]</sup> However, only 4% of the 314 cases in the reviewed literature was related to trauma.<sup>[12]</sup>

In this case, the patient did not mention any trauma history on the right foot, but we could not completely rule out the possibility of repetitive minor irritation by high heels or tight shoes. There were relatively many reports of IPEH occurring on foot in Japanese.<sup>[13–20]</sup> We carefully suppose the possible relationship between mechanical irritation induced by their habits of wearing geta (wooden sandals) and increased occurrence of IPEH in the foot. Levere et al<sup>[21]</sup> suggested that IPEH involves an autocrine loop of endothelial b-FGF secretion stimulating endothelial cell proliferation, and also regarded minor trauma or irritation as one of the causes for releasing b-FGF from macrophages or vascular endothelial cells. Although only 4% of the cases were related to the trauma according to the study by Pins et al,<sup>[12]</sup> further research on the relationship between mechanical injury and occurrence of IPEH in foot is needed.

#### 4. Conclusion

Intravascular papillary endothelial hyperplasia is a rare reactive lesion occurring more frequently in the head and neck region, but rarely in the foot. Radiologic diagnosis may be limited in predicting IPEH, especially due to infrequent occurrence in the foot. Therefore, histological confirmation after surgical excision is necessary. There are some reports suggesting an association between trauma and the occurrence of IPEH, but this is not yet conclusive.

#### Author contributions

**Conceptualization:** Sangchul Yun, In Ho Choi.

**Methodology:** Sangchul Yun.

**Supervision:** In Ho Choi.

**Writing – original draft:** Yeeun Han, Sangchul Yun, In Ho Choi.

**Writing – review & editing:** Yeeun Han, In Ho Choi.

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