Intravascular papillary endothelial hyperplasia occurring in the foot
A case report and review of literature
Yeeun Han, MD, Sangchul Yun, MD, PhD, In Ho Choi, MD.

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.[1] It occurs most frequently in the head, neck, trunk, or upper extremities, but its occurrence in the foot is rare.[2] Herein, we present a case of IPEH occurred on the dorsum of foot with a review of literature.

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 2cm 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 x 1.2 x 0.6cm-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...
atypia of endothelial cells or mitotic features were found. It was confirmed as IPEH. She has complained no complication within 2 years.

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.[3] However, Clearkin and Enzinger[2] 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.[4–9] A few cases of IPEH developed in foot were reported as summarized in Table 1.[10,11]

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...
Table 1

| Author, y | No. of case | Age/Sex | Location | Size | Duration | Trauma history | Treatment and prognosis (if stated) |
|-----------|-------------|---------|----------|------|----------|----------------|-------------------------------------|
| Fink et al[11] | 1 | 44/F | Left foot, planter | 2 cm | Insidiously, over 2 years | (-) | Excised, no recurrence in followed 2 years |
| Hashimoto et al[13] | 3 | N/D | Toe ×1, Foot ×2 | N/D | N/D | N/D | N/D |
| Clearkin and Enzinger[2] | 4 | N/D | Foot ×4 | N/D | N/D | N/D | N/D |
| Katsumata et al[14] | 2 | 40/F | Right heel | 1 cm | 2 decades | Suspected | Excised, N/D |
| Espinosa et al[10] | 1 | 23/F | Left foot, 1st toe, dorsum | 2 cm | Unknown | (+) | Excised, No recurrence in followed 4 years |
| Uchiyama et al[17] | 1 | 34/M | Right sole | 0.8 cm | 2 years | N/D | N/D |
| Shirai et al[20] | 1 | 46/F | Right sole | 0.9 cm | 40 years | N/D | N/D |
| Takahashi et al[19] | 1 | 8/F | Left sole | 2 cm | 3 months | N/D | N/D |
| Han et al[12] | 3 | N/D | Toe | | | | |

N/D = not described.

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.[11-20]

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[11] 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,[12] 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.
Supervention: In Ho Choi.
Writing – original draft: Yeenun Han, Sangchul Yun, In Ho Choi.
Writing – review & editing: Yeenun Han, In Ho Choi.

References

[1] Salyer WR, Salyer DC. Intravascular angiomatosis: development and distinction from angiosarcoma. Cancer 1973;36:993–1001.

[2] Clearkin KP, Enzinger FM. Intravascular papillary endothelial hyperplasia. Arch Pathol Lab Med 1976;100:441–4.

[3] Mason P. Intravascular vegetative hemangioendothelioma. Bull Soc Anat 1923;95:517–32.

[4] Akhtar M, Aslam M, Al-Mana H, et al. Intravascular papillary endothelial hyperplasia of renal vein: report of 2 cases. Arch Pathol Lab Med 2005;129:516–9.

[5] Kawashima A, Johnsen T, Murayama S, et al. Intravascular papillary endothelial hyperplasia of the adrenal gland. Br J Radiol 1986;59:610–3.

[6] Petty M, Brown MA, Hesselink JR, et al. Multilocular intravascular papillary endothelial hyperplasia in the retroperitoneum and spine: a case report and review of the literature. J Magn Reson Imaging 2009;29:947–51.

[7] Abad C, Campo E, Estruch R, et al. Cardiac hemangioma with papillary endothelial hyperplasia: report of a resected case and review of the literature. Ann Thorac Surg 1990;49:305–8.

[8] Hong SG, Cho HM, Shin HM, et al. Intravascular papillary endothelial hyperplasia (Masson’s hemangioma) of the liver: a new hepatic lesion. J Korean Med Sci 2004;19:305–8.

[9] Liu YJ, Tian YX, Ge DF, et al. Intravascular papillary endothelial hyperplasia of small intestine: report of a case. Zhonghua Bing Li Xue Za Zhi 2013;42:696–7.

[10] Espinosa A, Gonzalez J, Garcia-Navas F. Intravascular papillary endothelial hyperplasia at foot level: a case report and literature review. J Foot Ankle Surg 2017;56:724–8.

[11] Fink B, Temple HT, Miesel MS. Intravascular papillary endothelial hyperplasia: a pseudotumor presenting on the plantar foot. Foot Ankle Int 2003;24:871–4.

[12] Pins MR, Rosenthal DI, Springfield DS, et al. Florid extravascular papillary endothelial hyperplasia (Masson’s pseudosangosarcoma) presenting as a soft-tissue sarcoma. Arch Pathol Lab Med 1993;117:239–43.

[13] Hashimoto H, Daimaru Y, Enjoji M. Intravascular papillary endothelial hyperplasia. A clinicopathologic study of 91 cases. Am J Dermatopathol 1983;5:359–46.

[14] Katoh H. Two cases of intravascular papillary endothelial hyperplasia developing on the sole. J Dermatol 1996;23:655–7.

[15] Yamamoto T, Marui T, Mizuno K. Recurrent intravascular papillary endothelial hyperplasia of the toes. Dermatology 2000;200:72–4.

[16] Miyamoto H, Nagatani T, Mohri S, et al. Intravascular papillary endothelial hyperplasia. Clin Exp Dermatol 1988;13:411–5.

[17] Uchiyama N, Matsus MH, Kawauchi S. A probable case of intravascular papillary endothelial hyperplasia. Jpn J Dermatol 1984;94:756–7.

[18] Kitagawa NAJ. Three cases of intravascular papillary endothelial hyperplasia. Rinsho Hifuka 1986;40:765–8.

[19] Takahashi A, Higuchi WA, Takeuchi M. Three cases of intravascular papillary endothelial hyperplasia. Jpn J Dermatol 1986;96:865.

[20] Shirai TNM, Hirota S, Muramatsu T, et al. Two cases of intravascular papillary endothelial hyperplasia. Jpn J Dermatol 1987;97:183.

[21] Levere SM, Barsky SH, Meads RA. Intravascular papillary endothelial hyperplasia: a neoplastic “actor” representing an exaggerated attempt at recanalization mediated by basic fibroblast growth factor. J Hand Surg Am 1994;19:539–64.