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CASE REPORT

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Ultrasonographic features of an intravascular lobular capillary hemangioma originating in the basilic vein: Case report and literature review

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Abstract

Intravascular lobular capillary hemangioma (ILCH) is a rare variant of pyogenic granuloma, which develops within the lumen of a blood vessel. Here, we report the case of a 38-year-old male presenting with a mobile, incompressible, nonpulsatile nodule in the superficial fascia of the forearm. A first ultrasonographic examination diagnosed the mass as intravascular thrombosis with partial recanalization. Three months later, a second ultrasonographic examination concluded to the existence of an intravascular tumor, which finally turned out at pathological examination to be ILCH. Here, we report the ultrasonographic features of ILCH and the differential diagnosis between ILCH and other intravascular masses like thrombus.

KEYWORDS
intravascular lobular capillary hemangioma (ILCH), intravascular pyogenic granuloma, thrombus, ultrasonography

1 | INTRODUCTION

Lobular capillary hemangioma (LCH), also known as pyogenic granuloma (PG), is a common form of benign vascular tumor.[1,2] Intravascular lobular capillary hemangioma (ILCH) is a specific type of PG located within a blood vessel, which was first reported by Cooper et al.[3] Due to its clinical significance, studies concerning the diagnosis and treatment of ILCH have been abundantly carried out in recent years. Dermawan et al[4] reported 40 cases of ILCH to emphasize its clinicopathological features, which greatly promoted the understanding of disease stratification and severity acknowledgement. Although a number of individual cases have also been published, studies focused on imaging diagnosis, especially with ultrasonographic examination (US) are comparatively scarce. Here, we report a case that has been initially misdiagnosed as intravascular thrombus and was finally demonstrated by US to be ILCH.

2 | CASE REPORT

A 38-year-old male patient was referred to our hospital because of a small, palpable mass in his left forearm, found incidentally approximately 2 months ago. During the 2-month period, the mass showed progressive enlargement and tenderness. Physical examination in outpatient department revealed a mobile, incompressible, and nonpulsatile nodule in the medial aspect of the left forearm. The patient had no history of drug abuse, surgery, intravenous catheter insertion, or trauma of the left upper extremity.

Grayscale US using a iU22 ultrasound system (Philips Ultrasound, Bothell, Washington) with its 5 to 12 MHz linear probe revealed a well-defined nodular intraluminal mass in the basilic vein, with dimensions of approximately 8 × 2 × 6 mm (Figure 1A). The mass appeared as a heterogeneous, mainly hypoechoic structure, with distinct anechoic tunnel-like structures (Figure 1B). Color Doppler flow imaging (CDFI) showed abundant blood flow signals within the lesion, with vessels coming from the surrounding tissues (Figure 1C). Pulsed Doppler (PW) examination recorded typical arterial signals within the lesion (Figure 1D). The mass was diagnosed as thrombosis with partial recanalization, while intravascular tumor was not excluded. The patient was not given any specific therapy, but regular US follow-up was recommended.

Three months later, the patient was re-evaluated. The size of the mass within the basilic vein had increased to approximately 10 × 4

× 6 mm (Figure 2A). Its structure remained heterogeneous, mainly hypoechoic, but the anechoic tunnel-like structures had almost disappeared (Figure 2B). CDFI showed abundant angiogenesis within the lesion. As on the previous examination, the vessels feeding the lesion were coming from the periphery of the vein (Figure 2C). Pulsed Doppler showed low-resistance arterial signals within the nodule (Figure 2D). Therefore, the diagnosis was changed to intravascular tumor.

The mass was surgically resected, and no recurrence was found during three years of follow-up. The initial macroscopic examination found a well-defined polypoid mass in the lumen, in continuity with the venous wall. Further pathological examination demonstrated a mass composed of multiple capillaries lined with flattened endothelial cells arranged in a lobular fashion, in which the small capillaries were separated by loose fibrous mucinous stroma. Rich vascularity within the mass was confirmed by immunocytochemistry staining for CD34 and ERG. CD31, EMA, and podoplanin (D2-40) were negative (Figure 3).

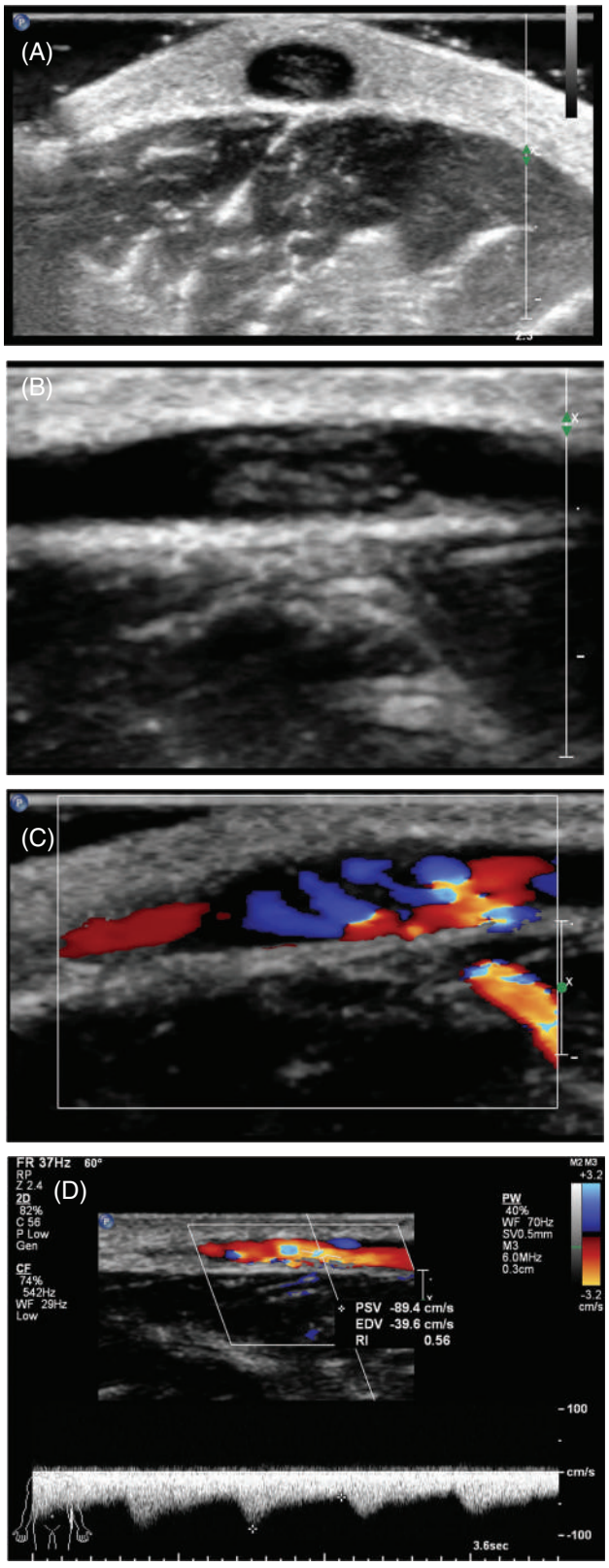

FIGURE 1 Legend on next page.

3 | DISCUSSION

ILCH is a rare variant of LCH, which develops within the lumen of a preexisting blood vessel. It occurs most frequently in the head, neck, and upper extremities.[3,4] In rare cases, ILCH might occur in the renal vein, iliac vein, adrenal gland, and corpus cavernosum of the penis.[5-8] Epidemiologically, ILCH usually occurs in middle-aged people, with a slight female predominance.[4] Most patients present for the first time with a well-defined, elevated skin mass which may remain stable or grow slowly over several months, as in the present case who presented with a self-palpable gradually enlarging mass. [4]

For superficial soft tissue masses such as ILCH, ultrasonography is one of the preferred imaging diagnostic techniques, especially when the masses are too small to be evaluated by computed tomography (CT) or magnetic resonance imaging (MRI). We searched PubMed from January 1979 to August 2020 for "intravascular lobular capillary hemangioma" and "intravascular pyogenic granuloma," which yielded 44 papers, including nine reports on ultrasonography.[9-17] Grayscale ultrasonographic features of ILCH were almost uniformly described as a well-defined mass within the lumen of a vein, except for one case where ultrasound failed to identify the host blood vessel.[12] Although some lesions were reported as iso-echoic, most were mainly hypoechoic. Typically, the lesion was echogenic with small hypoechogenic

FIGURE 1 At the first examination, B-mode grayscale ultrasonography on transverse section revealed a well-defined, heterogeneous hypoechoic nodule in the lumen of the left forearm basilic vein, A, with distinct anechoic tunnel-like structures (arrow) seen on the lesion longitudinal section. B, The size of the lesion was approximately 8 × 2 × 6 mm. C, Color-coded Doppler flow imaging showed abundant blood flow signals within the lesion, with vessels coming from the vein periphery. D, Pulsed Doppler examination showed arterial waveform within the lesion, with peak systolic velocity (PSV): 89.4 cm/s, end diastolic velocity (EDV): 39.6 cm/s, and resistance index (RI): 0.56

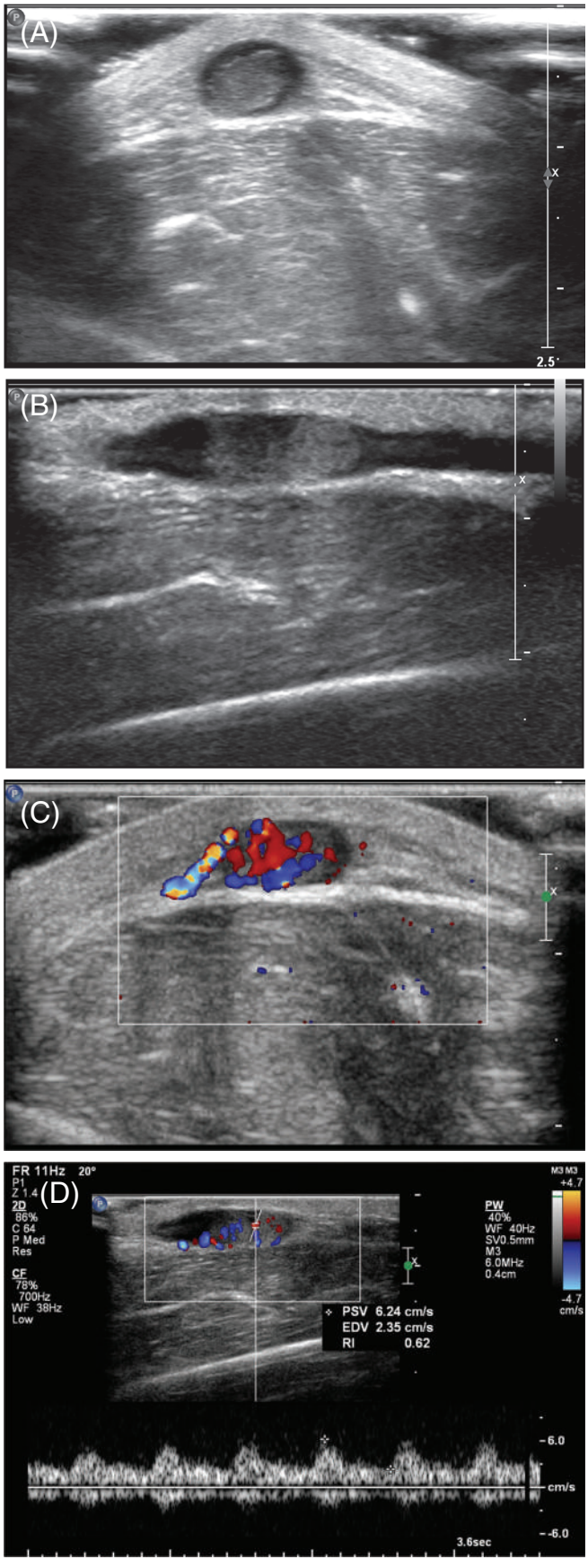

FIGURE 2 Legend on next page.

areas or internal tiny anechoic tubular or tunnel-like structures. The anechoic tunnel-like structures described in our case appeared thinner on the second examination, which has not been reported before. The lesion was adherent to the wall of the vein, and could not be collapsed. In most of the articles, CDFI showed intense hypervascularization within the lesion which adhered to the venous wall, with the blood vessels coming from surrounding tissues. Only four reports described the pulsed-Doppler signals, among which two studies showed arterial blood flow (as in our case),[13,16] and another two studies showed mixed arterial and venous blood flow.[12,15] One report described the use of ultrasound contrast agent which proved a high degree of vascularization.[9]

Under histological examination, ILCH was composed of lobules of capillaries separated by fibro-myxoid stroma, which was attached to the wall of the vein by a fibrovascular stalk.[3] Two different patterns of ILCH have been described in the literature: the classic, uncomplicated lobular pattern of capillaries mostly with small lumina, and the edematous pattern with stromal infiltration by inflammatory cells and dilated vessels, which has lost the lobular organization.[3] Therefore, we suggest that the sonographic features of the lesion may result from its histological pattern. The echogenic nodular intraluminal mass might correspond to the lobulated capillaries. The internal hypoechogenic areas and tiny anechoic tubular structures may correspond to edematous fibro-myxoid stroma. The vessels feeding the lesion apparently go through the fibrovascular stalk. In our case, the size of the mass increased between the first and the second US examinations, and the anechoic tunnel-like structures became thinner over time. We hypothesize that the pathological pattern of the lesion might change over time.

At US examination, ILCH should be differentiated from other lesions such as thrombus recanalization and intravascular papillary endothelial hyperplasia (IPEH) in particular. During recanalization, thrombi can show tunnel-like or tubular anechoic pattern on greyscale US, and abundant blood flow signals on CDFI. In addition, PW may detect arterial blood flow signals. This might be the reason why the mass, in our patient, was initially diagnosed as thrombus with partial recanalization, although tumor could not be completely ruled out. However, over time, ILCH might grow larger, while thrombi usually become smaller due to fibrinolysis. This is why we changed the diagnosis when we found that the mass had enlarged. Although ILCH maintains a relatively low morbidity, differential diagnosis with

FIGURE 2 At the second examination 3 months later, B-mode grayscale ultrasonography showed that the size of the lesion had increased to 10 × 4 × 6 mm in the cross-section, A, and longitudinal section, B. B, The anechoic tunnel-like structures within the lesion significantly had almost disappeared, while an anechoic fissure appeared. C, Color Doppler flow imaging showed abundant blood flow signals within the lesion and its feeding vessels. D, Pulsed Doppler examination showed low resistance arterial signals within the lesion, with peak systolic velocity (PSV): 6.24 cm/s, end-diastolic velocity (EDV): 2.35 cm/s, and resistance index (RI): 0.62

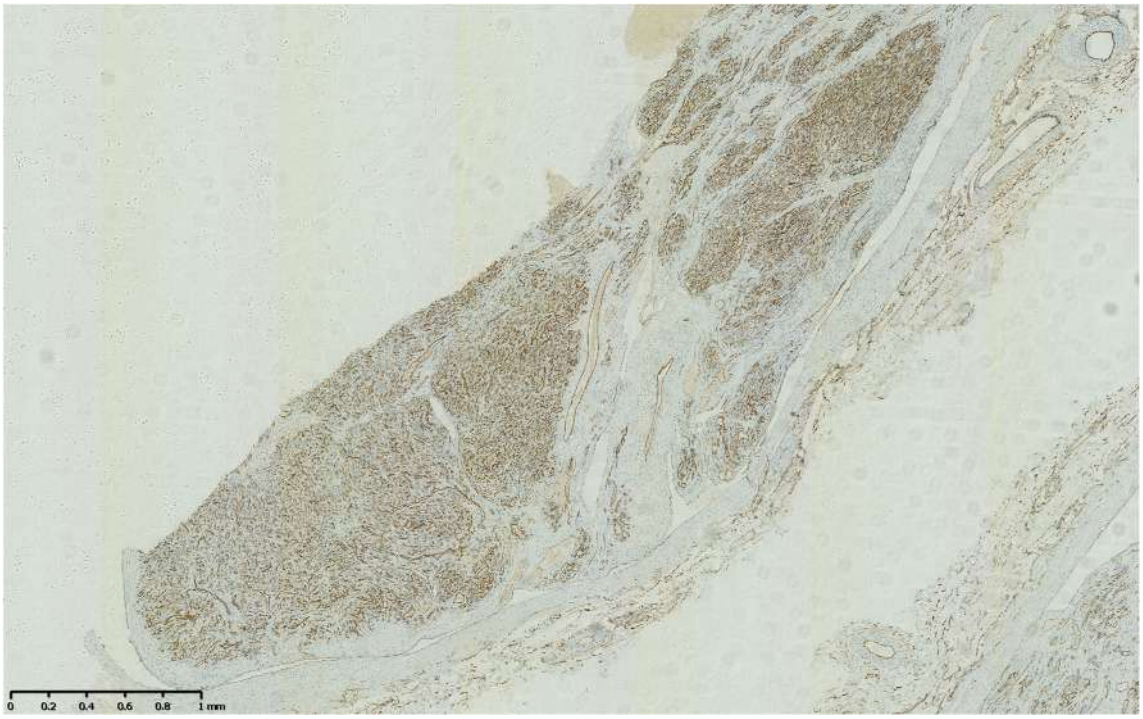

FIGURE 3 Immunohistochemical image. Rich vascularity in the lesion was confirmed with immunocytochemistry staining for CD34

thrombus is important. IPEH can be situated in subcutaneous tissues, or within muscles and blood vessels. [18,19] In contrast, almost all reported ILCH lesions occurred within the vascular lumen, with only one case where US could not identify the host vessel. There are two kinds of greyscale US manifestations of IPEH, most of which are heterogeneous, while some may show a relatively homogeneous structure. Heterogeneous lesions tend to show distinct internal septum-like structures.[20] However, ILCH lesions probably do not have the distinct internal septum-like structures, but are likely to maintain small hypoechogenic areas or internal tiny anechoic tubular or tunnel-like structures. Although there are some differences between the US images of both diseases, their differential diagnosis mainly relies on pathological examination. The characteristic histomorphological feature of IPEH is a papillary structure covered with hyperplastic endothelial cells within the vascular lumen.[21] However, the histologic feature of ILCH is a well-circumscribed, lobular proliferation of closely packed capillaries with central ectatic vessels.[4]

The first-line treatment for ILCH is radical local excision of the mass itself along with a small portion of the vein affected. The prognosis is excellent, without recurrence.[4] Therefore, accurate diagnosis of ILCH is essential to avoid unnecessary treatment and anxiety.

In conclusion, we report a rare case of vascular tumor presenting as a mall lobular heterogeneous hypoechoic mass with characteristic US features, including anechoic tubular or tunnel-like structures and peri-venous vessels connecting to the lesion, which should lead to consider ILCH.

CONFLICT OF INTEREST

The authors declare no potential conflict of interest.

AUTHORS' CONTRIBUTIONS

Wenli Jing and Xiaorong Wen contributed the central idea, analyzed most of the data, contributed to the writing and revisions. Mei Zhang, Chenyun Zhou, Jing Huang, Ying He provided the data, contributed significantly to manuscript preparation. Thanks to Dr Hong Wang and Dr Yujia Yang for their help and suggestions on the later revision of the manuscript.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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REFERENCES

1. Patrice SJ, Wiss K, Mulliken JB, et al. Pyogenic granuloma (lobular capillary hemangioma): a clinicopathologic study of 178 cases. *Pediatr Dermatol*. 1991;8:267-276.
2. Wassef M, Blei F, Adams D, et al. Vascular anomalies classification: recommendations from the International Society for the Study of vascular anomalies. *Pediatrics*. 2015;136:203.
3. Cooper PH, McAllister HA, Helwig EB. Intravenous pyogenic granuloma. A study of 18 cases. *Am J Surg Pathol*. 1979;3:221-228.

4. Dermawan JK, Ko JS, Billings SD, et al. Intravascular lobular capillary hemangioma (intravascular pyogenic granuloma): a clinicopathologic study of 40 cases. *Am J Surg Pathol*. 2020;44:1515-1521.
5. Philipp R, Jonas L, Georgi T, Cem A, Claudia G, Joanne Nyaboe ND. Intravenous pyogenic granuloma of the renal vein: a rare morphological differential diagnosis to renal cell carcinoma. *Urol Case Rep*. 2019;26:100951.
6. Pradhan S, Bazan H, Salem R, Gusberg RJ. Intravenous lobular capillary hemangioma originating in the iliac veins: a case report. *J Vasc Surg*. 2008;47:1346-1349.
7. Risio D, Selvaggi F, Viola P, et al. Intravenous pyogenic granuloma of the right adrenal gland: report of a case. *Surg Today*. 2013;43:569-573.
8. Gameiro A, Cardoso JC, Calonje E, et al. Intravascular lobular capillary hemangioma in the corpus spongiosum. *Am J Dermatopathol*. 2016;38:15.
9. Cera C, Calvagna C, Sgorlon G, et al. Internal jugular vein pyogenic capillary hemangioma: a case report. *Ann Vasc Surg*. 2015;29:361.
10. Taguchi S, Kakefuda T, Ono S, et al. Intravenous lobular capillary hemangioma occurring after needle insertion during routine health checkup. *Ann Vasc Dis*. 2013;6:102-105.
11. Ahn SE, Park JS, Ryu KN, Jin W, Park SY. Intravenous lobular capillary hemangioma occurring in the cephalic vein of a 39-year-old adult. *J Clin Ultrasound*. 2014;42:375-378.
12. Loftus WK, Spurrier AJ, Voyvodic F, Neuhaus SJ, James CL. Intravenous lobular capillary haemangioma (pyogenic granuloma): a case report and a review of imaging findings as reported in the literature. *J Med Imaging Radiat Oncol*. 2018;62:217-223.
13. Maddison A, Tew K, Orell S. Intravenous lobular capillary haemangioma: ultrasound and histology findings. *Australas Radiol*. 2006;50:186-188.
14. Jung JY, Goo B, Choi YJ, Chung HJ, Chung KY. A case of granuloma pyogenicum presenting as an intravascular mass: evaluation by ultrasonography. *J Eur Acad Dermatol Venereol*. 2008;22:515-517.
15. Ghersin E, Nitecki S, Brook OR, et al. Intraluminal pyogenic granuloma of the basilic vein. *J Ultrasound Med*. 2004;23:443-445.
16. Sarteschi LM, Bonanomi G, Mosca F, Ferrari M. External jugular vein hemangioma occurring as a lateral neck mass. *J Ultrasound Med*. 1999;18:719-721.
17. Ghekiere O, Galant C, Vande BB. Intravenous pyogenic granuloma or intravenous lobular capillary hemangioma. *Skeletal Radiol*. 2005;34:343-346.
18. Hashimoto H, Daimaru Y, Enjoji M. Intravascular papillary endothelial hyperplasia. A clinicopathologic study of 91 cases. *Am J Dermatopathol*. 1983;5:539-546.
19. Lysyy O, Schwartz I, Kolander Y, Strauss S. Sonographic features of intravascular papillary endothelial hyperplasia (Masson's tumor) in the forearm. *J Clin Ultrasound*. 2011;39:301-303.
20. Kim OH, Kim YM, Choo HJ, et al. Subcutaneous intravascular papillary endothelial hyperplasia: ultrasound features and pathological correlation. *Skeletal Radiol*. 2016;45:227-233.
21. Akdur NC, Donmez M, Gozel S, et al. Intravascular papillary endothelial hyperplasia: histomorphological and immunohistochemical features. *Diagn Pathol*. 2013;8:167.

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