» Case Report «

Intravenous Lobular Capillary Hemangioma Occurring after Needle Insertion during Routine Health Checkup

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Surgery was performed on a 53-year-old male patient with a painful mass in front of the elbow. The mass originally occurred after needle insertion during a routine health checkup and grew in size during a 1-year period. Intravenous tumor with arterio-venous fistula was diagnosed, and it was resected. Histopathological diagnosis of intravenous lobular capillary hemangioma was made. Occurrence of this tumor after a routine health checkup is rare. The etiology of this tumor occurring simultaneously with arterio-venous fistula is discussed.

Keywords: intravenous lobular capillary hemangioma, needle insertion, routine health checkup

INTRODUCTION

Lobular capillary hemangioma (LCH) is commonly a dermatic tumor occurring anywhere on the skin or even on a mucosal surface. It is a benign tumor and is also called a pyogenic granuloma. The tumor is often related to a history of prior trauma. In rare cases, the tumor occurs intravenously and is called intravenous lobular capillary hemangioma (IVLCH). Although there are no known risk factors for IVLCH, some reports describe

an association with arterio-venous malformations,¹⁾ and hyperproliferative vascular response to trauma or infection.²⁾ We present a rare case of IVLCH occurring after needle insertion during a routine health checkup.

CASE REPORT

A 53-year-old man was referred to our hospital due to a painful mass in front of his left elbow. Although he had hypertension and hypercholesterolemia, he had been living a normal life. He had received a routine health checkup 1 year prior. During the checkup, a blood sample was taken. For sampling, needle insertion was performed on his left median cubital vein, which was normal with no prior trauma. He remembers insertion being attempted twice with difficulty, with the tip of the needle being moved several times to catch the vein. Although hemostasis occurred with no problems, he noticed a small hard mass several days after insertion. Because there was no pain, he left the mass untreated. The mass increased in size gradually, and he had been experiencing intermittent pain in the area of the mass for the 3 months prior to referral.

On consultation, a hard mass of about 2 × 1 cm was palpable, and darkened skin was observed around the

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Fig. 1 Preoperative images. (A) Color Doppler sonography. Intravenous mass (arrowhead) with internal anechoic tubular structures, showing internal blood flow. Connection (arrow) from a small artery behind the vein to the internal tubular structure. (B) Computed tomography. Contrast medium that flowed into the mass (arrow) during the early arterial phase. (C) Magnetic resonance imaging (T2*-weighted). Areas of spotted low-signal intensity scattered inside the mass (arrow).

area of the left median cubital vein in front of the elbow. Doppler stethoscope showed a continuous murmur around the area.

Color Doppler sonography revealed an intravenous mass with internal anechoic tubular structures, and internal blood flow in the structures (**Fig. 1A**, arrowhead). A small artery was observed behind the vein, which supplied blood flow to the internal tubular structure (**Fig. 1A**, arrow). Therefore, a diagnosis of intravenous tumor with arterio-venous fistula was made.

Computed tomography revealed a mass of $15 \times 9 \times 24$ mm inside the vein. Contrast medium flowed into the mass from behind in the early arterial phase (**Fig. 1B**), which led to the diagnosis of micro arterio-venous fistula.

Magnetic resonance imaging (MRI) revealed a mass; on T1-weighted images it was isointense to muscles; on T2*-weighted images, areas of spotted low-signal intensity

were scattered inside, reflecting internal flow or hemosiderin deposition (**Fig. 1C**).

Surgical treatment was performed. Strong adhesion due to inflammation was observed around the mass. The median cubital vein, which included the mass, was dissected. A small artery of less than 1 mm in diameter connected to the vein was identified (**Fig. 2A**, arrow). The vein including the mass was excised. The vein was incised, and a solid tumor attached to the inner wall of the vein appeared (**Fig. 2B**, arrow).

Microscopically, the mass was diagnosed as IVLCH with no evidence of malignancy. Proliferation of small diameter capillaries lined with flattened endothelial cells was observed (**Fig. 3A**). The lobules of capillaries were separated by fibromyxoid stroma (**Fig. 3B**). For immunohistochemistry, the endothelial cells were stained CD34 positive. Also, a large vessel existed inside the tumor (**Fig. 3C**).

Fig. 2 Surgical and macroscopic photographs. (A) Small artery (arrow) connected to an enlarged vein. (B) Incised vein and appearance of solid tumor (arrow).

Fig. 3 Microscopic photographs. (A) Proliferation of small diameter capillaries. (Hematoxylin-Eosin stain, magnification $\times 100$). (B) Lobules of capillaries separated by fibromyxoid stroma. (Hematoxylin-Eosin stain, magnification $\times 100$). (C) A large vessel existed inside (arrow). (Hematoxylin-Eosin stain, magnification $\times 40$).

COMMENT

IVLCH was first reported by Cooper, et al.³⁾ The appearance of lobules of capillaries separated by a fibromyxoid stroma was similar to dermatic cases. They thought that the tumor received its blood supply from a small artery in the region of the stalk to the wall of the vein. The terminology of pyogenic granuloma, which is in the

title of their report, is a classic term of LCH. When involved in the skin and mucosal surfaces, ulceration and suppuration may occur, which lead to the term pyogenic.⁴⁾

Although we were not able to detect a small artery that enters the lesion microscopically, we were able to detect a large vessel inside the tumor. This vessel is definitely one of the internal anechoic tubular structures observed by the sonography.

After Cooper, et al., several reports of IVLCH have been published.¹⁾ To the best of our knowledge, there is no report in the literature of IVLCH occurring after needle insertion during a routine health checkup.

Dermatic and mucous cases of LCH often relate to a prior trauma. It is described that IVLCH is associated with arterio-venous malformations,¹⁾ and hyperproliferative vascular response to trauma or infection.²⁾ Our case occurred in a previously normal median cubital vein. The basis of this case is a formation of an arterio-venous fistula. Formation of the fistula was verified with color Doppler sonography. The existence of a small artery entering the vein was observed as shown on the photograph.

The histogenesis of IVLCH, whether reactive or neoplastic, still remains obscure.⁵⁾ Microscopically, the tumor consists of multiple capillaries in a lobular fashion which are separated by fibrous stroma. Observing **Fig. 3**, two types of cells are obvious. One is the endothelial cells of the capillaries, and the other is a fusiform stromal cells with flattened nuclei. Seeing this bicellular, mixed endothelial and pericytic composition of LCH, Nichols, et al. supported reactive, rather than monocellular neoplastic process in these lesions.⁶⁾ But reading their report precisely, we must be careful that they admitted that bicellular composition has also been demonstrated in neoplastic lesions. Although our case can be estimated as a reactive procedure against the fistula, we were unable to obtain a concrete scientific evidence to describe whether reactive or neoplastic.

Rusin, et al. elegantly described the formation of pyogenic granuloma (LCH) in cases of acquired arterio-venous fistula.⁷⁾ They described the mechanism by relating it to the formation of collateral circulation and to the inflexibility of the rim surrounding the fistulas. Again, we were not able to prove this estimation from the results of our case.

Ghersin, et al. describes the color Doppler sonographic manifestation of IVLCH.⁸⁾ Although similar to their sonography, our case is unique in visualizing the existence of an arterio-venous fistula, and the fistula flowing directly into the tumor. The first examination we

performed was a color Doppler sonography, and this is just what Maher, et al. insist which is to perform ultrasound exam as a first-line diagnostic tool.²⁾

MRI of our case was similar to the IVLCH reported previously.^{1,4)} Our case had an opportunity to acquire a T2*-weighted image, which was not reported previously to the best of our knowledge.

In conclusion, this is the first report of an IVLCH occurring after needle insertion during a routine health checkup. The formation of an arterio-venous fistula was verified by sonography and proven macroscopically. The findings confirm the lesion to be associated with arterio-venous fistula, but the etiology whether reactive or neoplastic needs further investigations.

DISCLOSURE STATEMENT

The authors have no conflicts of interest to declare.

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