

Original Article

Intravenous lobular capillary hemangioma: report of a case and review of literature

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Abstract: Operation was done to excise a painful mass in the wrist of a 26-year-old woman. Its preoperative diagnosis was subcutaneous cysts in wrist, while B ultrasound showed that it came from excessive venous vessels. The mass was excised through operation. The pathological diagnosis was intravenous lobular capillary hemangioma. HE staining, microscopic observation and Immunohistochemistry pathologically confirmed the diagnosis of it being intravenous lobular capillary hemangioma (IVLCH). In addition, literature review was carried out to discuss its clinical manifestation, pathogenesis, histological features and differential diagnosis.

Keywords: Pathological diagnosis, intravenous lobular capillary hemangioma, literature review

Introduction

Lobular capillary hemangioma (LCH) is a common skin tumor, a polypoid capillary hemangioma which may appear in any surface of skin and membrane. Earlier pathologists considered it to be inflammatory infectious disease for the reason that there are often ulcers in the surface and inflammatory cell infiltrations inside the tumor, and its lesions resemble inflammatory granulation tissue. They called it pyogenic granuloma (PG). Nonetheless, some cases also have no inflammatory cell infiltrations or ulcers in surface. Instead, they proved to be a kind of benign hemangioma. In some rare cases, tumor was located inside the vein, which was then called intravenous lobular capillary hemangioma (IVLCH). It is hard to make definite diagnosis unless pathological examination was carried out. Since first report in 1979 [1, 2], IVLCH has been reported in no more than 30 reports abroad, while domestic reports show a much less figure. Wang Yang and others referred to 2 cases when reviewing and analyzing lobular capillary hemangioma. Yet they didn't report specifically. This report involves a case of IVLCH in wrist, in which the patient had resection of tumor tissue and some part of venous tissue with good outcome.

Materials and methods

Clinical data

The patient is a young woman aged 26, 162 centimeter tall and 85 kilogram in weight. Her left wrist was found having a tumor half a year ago. At that time, the tumor was only of bean size without any uncomfortable symptoms. So the patient did not pay attention. Later, she felt it increasing and tenderness became obvious in some parts. Some parts of the tumor were soft and had low active degree.

Generally speaking, the patient was in relatively mild condition, with blood glucose being 10.57 mmol/L. All the hepatic and renal functions were normal, urine ketone negative. It was diagnosed as Type 2 Diabetes Mellitus.

The patient had a local mass of 2*2 cm in size, oval in shape, soft in quality and low in active degree. No tenderness was felt, and it had no adhesion with surrounding tissue. Her wrist could move freely without obvious swelling.

By scanning the mass on the ulnar side of left wrist, stripy low echo area was detected with its range being 4.4*0.4 cm, borders unclear, inner echo uneven.

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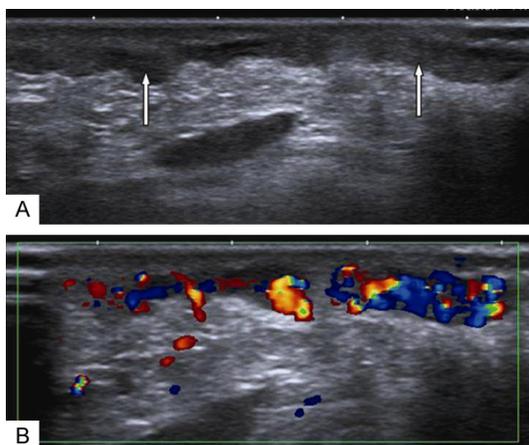


Figure 1. The B-ultrasound image of the patient with IVLCH in (A) detected stripy low echo area and rich blood signal, while (B) showed arterial spectrum in tumor.

Rich blood flow signal was detected in the stripy low echo area. Its inner measurement and arterial spectrum are showed in **Figure 1**.

The patient was diagnosed with subcutaneous cysts in left wrist.

Operation process

The longitudinal incision was cut on the surface of the mass, from skin to subcutaneous tissue. Blunt separation revealed mass inside the subcutaneous tissue, which was stripy, with transparent matrix and complete capsule. It was easy to separate and then resected completely. The patient recovered well, so left the hospital two days later, having dressing change every three days. She also had diabetic treatment.

Method

The sample was fixed by 4% neutral formalin, embedded in paraffin, prepared as usual, HE stained and observed under light microscope. Immunohistochemical staining (S-P method) adopted CD34, SMA, FVIII and S-100, DAB color. There was positive control in every step, which took the known positive slice of the corresponding antibody to make contrast, while negative control replaced the first antibody with PBS. Both the antibodies and kits were purchased from the Immunohistochemical staining of ZSGB-Bio.

Results

Grossly, a patch of greyish white tissue was there of 3*0.5*0.3 cm in size.

Microscopically, lesions consisted of hypertrophically capillaries, clustered or lobular. They were lined with lobular structures, no capsule but with clear border. Fibrous connective tissue was formed in interlobar area, while nutrient vessels were found inside the lobular structure. Under the high power lens, capillaries were seen to be covered with flattened epithelial cells, with stretched nucleus in some parts. This tumor was quite like LCH, but without ulceration or inflammatory cell infiltrations, which can be seen in **Figure 2**.

Immunohistochemical stains with automatic immunostainer (VENTANA) were performed with CD34, SMA, FVIII and S-100 antibodies. As a result, CD34, a kind of endothelial cell, stains strongly, while SMA and FVIII express markedly, S-100 negative. See **Figure 3**. To sum up, the final pathological diagnosis was IVLCH in wrist.

The patient recovered smoothly after operation, without complication. Three months of follow-up has seen no sign of recurrence.

Discussion

IVLCH is a rare LCH with benign lesions. Current reports have revealed that it mainly appears in heads, necks and the end of upper limbs. However, domestic reports about it are rare, and its pathogenesis has not been figured out. Unlike PG, which was previously referred to, it has no inflammatory reaction. For the deficiency of expression with characteristics, it is hard to do clinical diagnosis. Its first symptoms resemble the formation of deep venous thrombosis. However, imaging examination shows it is caused by excessive vessels. When Cooper and others reported IVLCH for the first time, they pointed out two of its pathological characteristics. The first one is the classic simple structure of lobular capillaries, while the other refers to interstitial of edema and swelling vessels [1].

IVLCH is a rare LCH with benign lesions. Current reports have revealed that it mainly appears in heads, necks and the end of upper limbs [1, 3-12], with few exceptions which involved locations including womb, adrenal, iliac vein and others [13-15]. Patients are 38 years old on average, ranging from 15 to 66, with more female than male [2, 5, 16-18]. Its first symptoms resemble the formation of deep venous thrombosis. Lesions can be located by B ultra-

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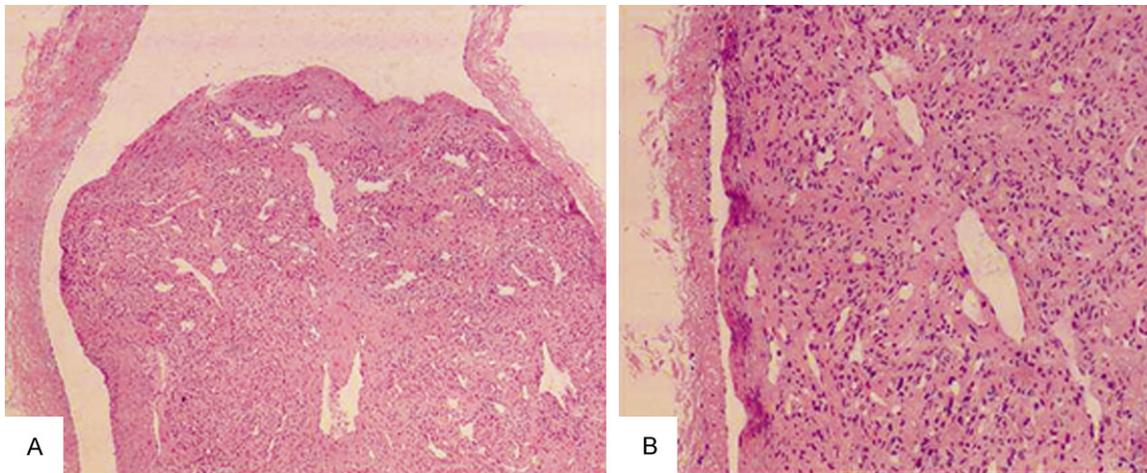


Figure 2. HE staining of tissue section of IVLCH was observed under microscope. Through low power lens ($\times 100$) in (A), lesions consisted of hypertrophic capillaries, clustered or lobular. They were lined with lobular structures, and fibrous connective tissue was formed in interlobar area. (B) Under the high power lens, capillaries were seen to be covered with flattened epithelial cells, without ulceration or inflammatory cell infiltrations.

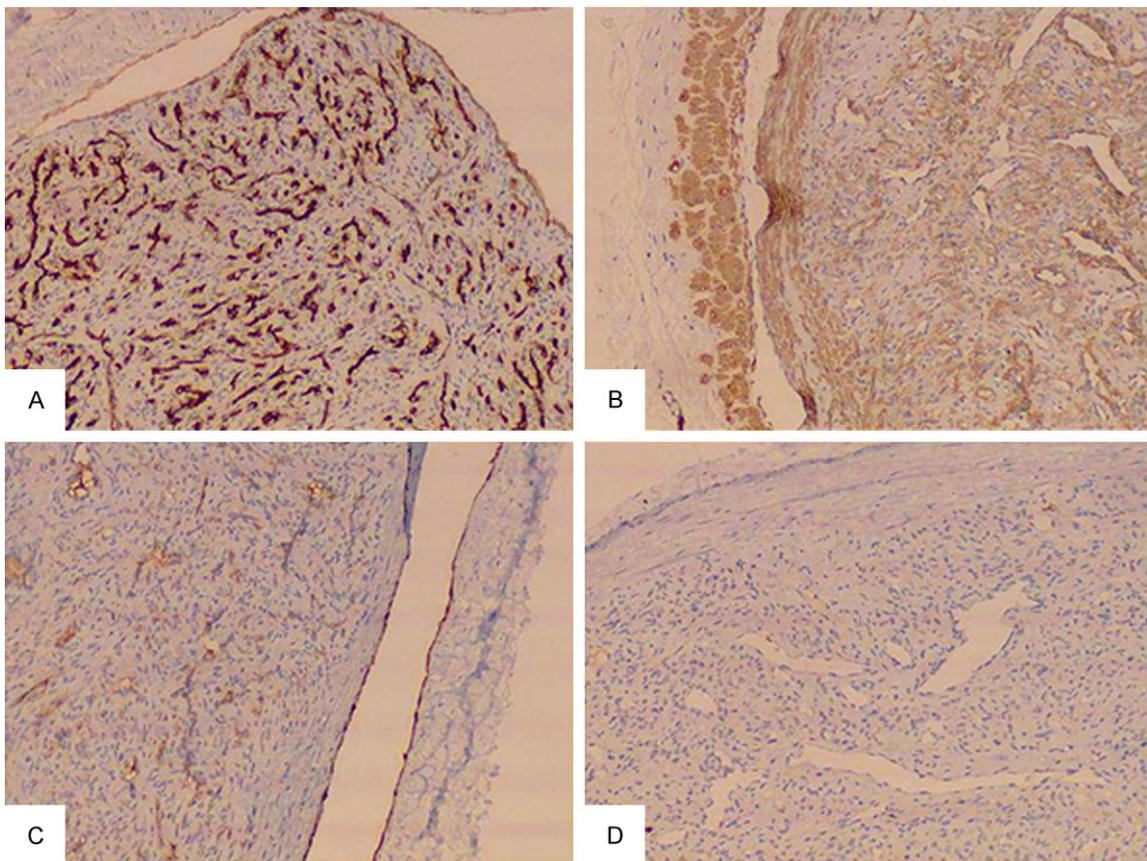


Figure 3. Immunohistochemistry staining ($\times 100$) of tumor tissue section of the patient showed in (A) that CD34, endothelial cell, stained strongly, while SMA and FVIII expressed markedly in (B). Peripheral venous wall also had smooth muscle tissue, which expressed the antibody. In (C and D), FVIII antibody expressed clearly, S-100 negative.

sound examination, which cannot yet exclude the possibility of neuropathy or cyst. Lesions of

finely rounded terminus and pulses of inner blood flow can eliminate the chance of throm-

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bus. For the deficiency of expression with characteristics, it is hard to do clinical diagnosis merely by clinical characteristics and physical examination. In this case, there were no other clinical characteristics than obvious swelling and tenderness.

Despite unknown risk factors, there have also been reports which claimed its connection with reactive arteriovenous malformation or hyperplasia of blood vessel which relate to trauma or infection [17]. The occurrence mechanism in histology, no matter it is of reaction or tumor, is not clear yet. Under microscope, this tumor consisted large amount of lobular capillaries, separated by fibrous matrix. The most obvious cell ingredients are the endothelial cells in capillaries and the spindle shaped stromal cells of flat nucleus. Based on these two cell ingredients, Nichols [19] and others tend to support the idea that this lesion is reactive, rather than tumor reproduction process. However, it still lacks hard evidence to confirm this lesion to be reactive or a kind of tumor.

Under low power lens, the tumor tissue was intravenous, connecting venous wall by fibrovascular axis. The tumor consisted of clustered or lobular hypertrophic capillaries, with disnatured, edema matrix as interstitial, which was like mucus. Collagen fiber was visible, while no obvious inflammatory cells were there as its previous name PG suggested. Under the high power lens, capillaries were seen to be covered with flattened epithelial cells, with stretched nucleus in some parts. However, there was no remarkable nuclear differentiation or division.

Except for CD31, CD34 and FVIII20 [20] as expression of endothelial markers, it also expressed SMA, inducible nitric oxide synthase (iNOS) and endothelial receptor tyrosine kinase [21, 22].

Benign as it is, the disease has to be differentiated from angiosarcoma, nipple-shaped endothelial proliferation in vessels, vascular fasciitis and organized thrombus.

Firstly, for organized thrombus, it also has obvious fibrin deposit in various periods, with relatively few capillaries and limited lobular structure. It is not difficult to do differentiation considering the clinical history of major operations and bed rest.

Secondly, vascular nipple-shaped endothelial proliferation is often caused by thrombus, colored mauve, characterized by sludge blood or thrombus. There lined numerous tiny nipples towards inside in vessel walls, the surface of which embodies monolayer of fat or swelling endothelial cells. The axes of the cells are fibrous tissue of collagen, like placental structure to some extent, often with hemosiderosis and organized thrombus.

Thirdly, vascular fasciitis constitute fibroblast-like spindle cells and myofibroblast of uniform obesity, whose nucleus have no anachromasis or polymorphism. There may be many mitotic figures, but no non-typical nuclear division. The cells are rich, but have loose areas, like mucus, with steoclast-like multinuclear giant cells, extravasation of red blood cells and the infiltration of lymphocyte. The capillaries are few and not lobular.

Fourthly, angiosarcoma was greyish white with solid mass on visual inspection, like fish flesh, which was difficult to distinguish from LCH. Under microscope, angiosarcoma was not lobular, with anachromasis and polymorphism. There are also lots of mitotic counts. Histological manifestations of the tumor vary greatly, with different zone of differentiation in different cases.

The clinical treatment of the tumor was to resect the tumor and some normal tissue 2-3 cm to the tumor by operation. The final diagnosis was based on the pathological diagnosis of the tumor tissue, the result of which was IVLCH with good prognosis. Although the growing rate of the tumor is not clear, the tumor is bound to grow in case of incomplete excision. Only when completely excised, can recurrence be prevented.

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Disclosure of conflict of interest

None.

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