CASE REPORT

Intravenous Lobular Capillary Haemangioma of the Jugular Vein: A Case Report and Literature Review

Aiqin Peng, MS [a], Zhengdong Fei, MS [b,], Lu Zhang, MS [b], Dan Xu, MS [c]*

[a] Department of Radiology, Shuyang People's Hospital, the Affiliated Shuyang Hospital of Xuzhou Medical University, Shuyang, Jiangsu, China; [b] Department of Ultrasound, Shuyang People's Hospital, the Affiliated Shuyang Hospital of Xuzhou Medical University, Shuyang, Jiangsu, China; [c] Department of Pathology, Shuyang People's Hospital, the Affiliated Shuyang Hospital of Xuzhou Medical University, Shuyang, Jiangsu, China

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Abstract: Intravenous lobular capillary haemangioma (IVLCH) is extremely rare and occurs mostly in the head, neck, and upper extremity veins. This was a case of IVLCH that occurred in the right external jugular vein and was diagnosed by conventional ultrasound and ultrasound elastography. Herein, we described the conventional ultrasonographic and ultrasound elastographic features of one case of IVLCH and discussed the clinical manifestations, pathological and ultrasonographic features, and differential diagnosis of IVLCH.

Keywords: Intravenous lobular capillary haemangioma; Ultrasound; Elasticity imaging techniques

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Intravenous lobular capillary haemangioma (IVLCH), often called intravenous pyogenic granuloma, is a rare benign haemangioma. To our knowledge, extremely few reports have described the ultrasonographic features of IVLCH, and the tumor's elastographic features have not been reported. Here, we introduced the first reported case of the ultrasound elastography of IVLCH confirmed histologically in the right external jugular vein.

Case Report

A 55-year-old female patient was admitted to the hospital with a subcutaneous nodule located in the right cervical region. This subcutaneous nodule, which had no obvious cause, had been observed for four months and had gradually grown during the last two months. During her disease course, the patient experienced no other symptoms. A dermatological examination indicated that the neck was symmetrical and lacked any jugular venous distension or redness or swelling of the skin. A 1.5 cm × 1.0 cm subcutaneous nodule was detected in the right cervical region. The nodule's texture was hard, its surface was smooth, there was no tenderness, and it could be pushed laterally. The patient did not have a history of cervical trauma or venous catheterization.

Ultrasonography showed a well-defined, hyperechoic mural nodule approximately 1.4 cm × 0.7 cm in size located on the right external jugular venous wall; there was a hypoechoic signal within the nodule. The venous wall was intact, and no invasion of the venous wall or surrounding soft tissues by the nodule was observed. Color and power Doppler ultrasonography showed abundant blood flow signals, and pulsed Doppler ultrasonography revealed the spectrum of arterial blood flow in the nodule. The resistive index (RI) was 0.63. We performed ultrasound elastography by elasticity imaging (EI) on the patient by using the Siemens Acuson S3000 ultrasound system with the linear transducer 9L4. Ultrasound EI showed that most of the observed lesions were black, but a few lesions were white. Upon replaying the images, we recorded the elastic score according to the standard proposed by Itoh [1]. The EI score of the

[*] *Corresponding author: Department of Ultrasound, Shuyang People's Hospital, the Affiliated Shuyang Hospital of Xuzhou Medical University, No. 9, Yingbin Road, Shuyang, Jiangsu 223600, China.*
e-mail: zhengdongfei1982@163.com

nodule was 3 (Fig. 1). Based on these manifestations, the nodule was diagnosed as a benign vascular tumor, and the patient underwent surgical resection. The diagnosis of IVLCH was ultimately confirmed via a histopathological examination (Fig. 2).

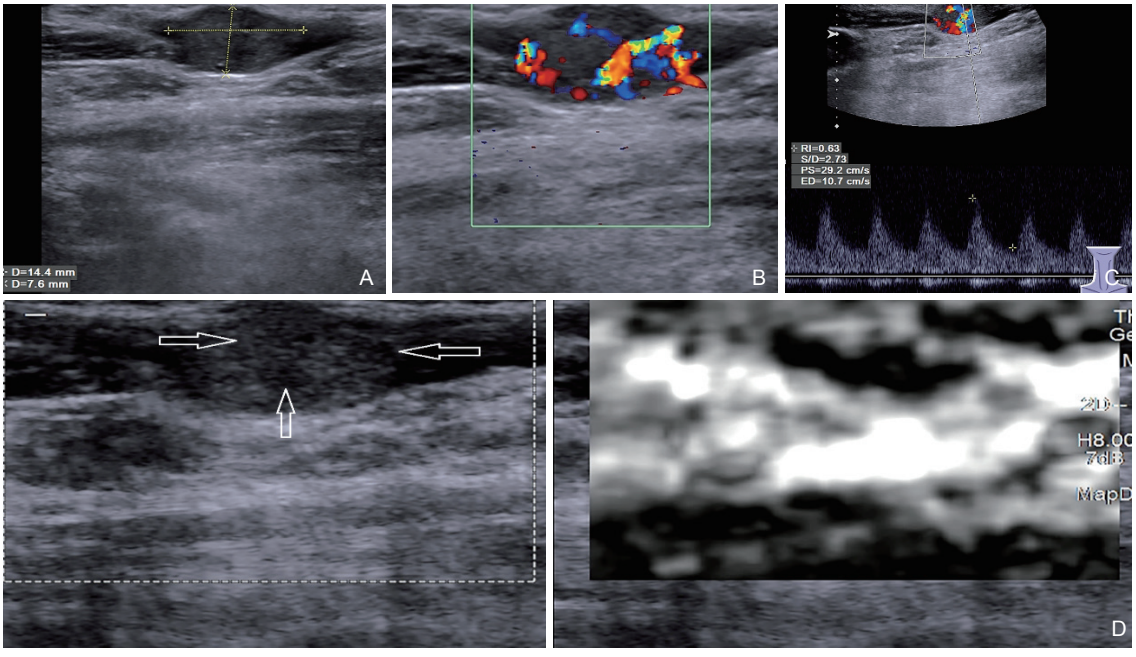

Figure 1 Intravenous lobular capillary haemangioma. Ultrasonographic findings. (A) Ultrasonographic findings for the hyperechoic mural nodule within the external jugular vein. (B) Color Doppler ultrasonography image of the nodule showing thick, strip-shaped and spot-like blood flow. (C) Pulsed Doppler ultrasonography revealing waveforms of arterial blood supply in the nodule. (D) EI showing that the nodule was mostly black, with a small white portion corresponding to the hypoechoic area on the two-dimensional image (hollow arrow).

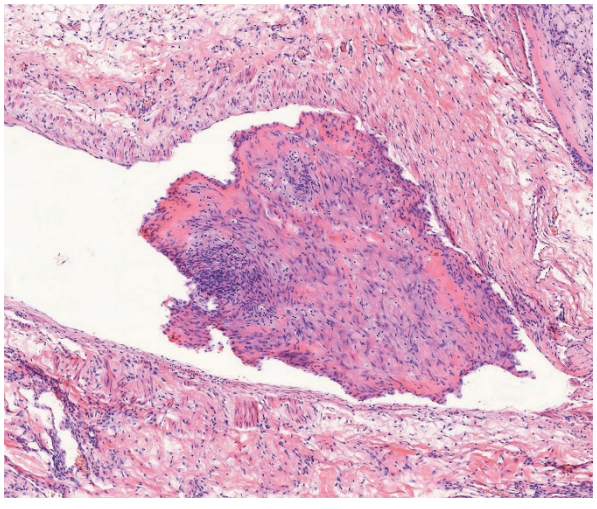

Figure 2 Numerous hyperplastic capillaries were observed in the dilated lumen of the vein; these capillaries were separated by fibrous tissue, forming a lobular structure. (HE stain ×40).

Discussion

IVLCH is an extremely rare, isolated lobular capillary haemangioma that is completely restricted to a venous lumen and lacks inflammatory cell infiltration. IVLCH was first reported by Cooper et al. [2] in 1979; to date, few cases have been reported. The frequency of this disease is slightly higher in women than in men, and this tumor is found mainly in the neck, upper arm, wrist, or fingers [3]. IVLCH has no characteristic clinical manifestations. In this case, the only manifestation was a palpable subcutaneous nodule located on the right cervical region that had gradually increased in size over the last two months.

As shown in our case, the ultrasonographic features of IVLCH included a well-defined hyperechoic mural nodule in the venous lumen that was accompanied by hypoechoic signals. Color Doppler ultrasonography showed abundant blood flow signals in the nodule. Pulsed Doppler ultrasonography showed that the spectrum of arterial blood flow could be measured in the nodule [4]. Loftus et al. [5] reported ultrasonographic features but not ultrasound elastography findings for one case involving clinically palpable IVLCH with a hard texture. In this case, EI showed that most of the observed lesions were black, but a few lesions were white. The EI score was 3. EI suggested that certain lesions had a harder texture, a possibility consistent with the findings from clinical palpation. In a comparison of conventional ultrasound, ultrasound elastography, and histopathological examination findings, the hyperechoic area in the nodule was regarded as lobulated capillaries

and had a hard texture; therefore, EI showed this area as being black. In contrast, the hypoechoic area was thought to be the edematous fibrotic myxoid stroma, and its texture was soft; thus, EI showed this area as being white [6]. Although EI can potentially be an important screening tool for the diagnosis of IVLCH, several limitations should be mentioned. First, lesions that are too small and very superficial may affect the result. Secondly, there are few soft tissues around the tumor in the venous cavity, so the repeatability of ultrasound EI may be poor.

IVLCH also needs to be distinguished from venous thrombosis, a tumor thrombus and intravascular papillary endothelial hyperplasia (Masson's tumour). An abundant blood supply was found in the intravenous mural nodule, and arterial blood flow was detectable in the nodule. These main ultrasonographic features helped us exclude venous thrombosis. Patients with intravenous tumor thrombus typically have a history of malignant tumours. In addition, an intravenous tumor thrombus rarely manifests as a solitary, well-defined hyperechoic mural nodule. Masson's tumour is a benign vascular disease caused by the papillary hyperplasia of endothelial cells. This disease is often found in the head, neck, fingers, and trunk and can coexist with other vascular diseases such as hemangioma and vascular malformation. Ultrasound and clinical examination cannot distinguish between IVLCH and Masson's tumour, and their differentiation relies mainly on the pathological examination. However, ultrasound elastography findings for IVLCH, we reported here for the first time, may help distinguish IVLCH from Masson's tumour. In particular, EI showed that the hyperechoic and hypoechoic areas of the nodule were black and white, respectively, which corresponded to the pathology of IVLCH. To our knowledge, prior studies have not reported ultrasound elastography findings for IVLCH and Masson's tumour; in the future, additional studies and reports addressing this issue are required.

Conclusion

IVLCH has unique ultrasonographic features. When the ultrasound examination reveals a well-defined, hyperechoic mural nodule within the veins that has an abundant blood supply, particularly if pulsed Doppler ultrasonography can measure arterial blood flow and ultrasound elastography shows that the nodule's texture is hard, IVLCH should be considered.

Conflict of Interest

Authors declare no conflicts of interest.

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