

Intravenous Lobular Capillary Hemangioma A Case Report

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Abstract : Lobular capillary hemangioma, so-called pyogenic granuloma, is a relatively common mucocutaneous vascular neoplasm that commonly occurs in skin. Intravenous lobular capillary hemangioma is an unusual variant that usually occurs in the neck and upper extremities. We have experienced a case of intravenous lobular capillary hemangioma. The patient was a 19-year-old female who had a soft, nontender neck mass, 0.8 cm x 0.7 cm, in the medial side of left sternocleidomastoid muscle for a month. The clinical impression was lymphadenitis. On gross examination, the tumor was pale pink to gray and nodular. Microscopically, the tumor showed typical patterns of lobular capillary hemangioma, however, they arose from the vessel wall and protruded into the vessel lumen. By immunohistochemistry, the tumor cells and surrounding vascular walls were positive for CD31 and smooth muscle actin. The pathologic finding and the differential diagnosis are discussed.

Key Words : Intravenous lobular capillary hemangioma, pyogenic granuloma

Introduction

Lobular capillary hemangioma (LCH) is a relatively common vascular neoplasm. It was formerly called as pyogenic granuloma and considered as a reactive lesion secondary to trauma and pyogenic infection [1]. However, the term of LCH is widely accepted

nowadays and the underlying process of lobular vascular proliferation is regarded as neoplastic in nature [2].

Intravenous lobular capillary hemangioma (IVLCH) is a rare solitary form of lobular capillary hemangioma. This entity was first described in 1979. There are less than 30 IVLVH documented in the most recent review

of articles [3]. The most common sites are the head, neck and upper extremities.

We present a case of IVLCH and discuss pathological findings with differential diagnosis.

Case Report

A 19-year-old female presented with a palpable soft, nontender neck mass in the medial side of left sternocleidomastoid muscle for a month. Ultrasonography showed an oval and homogeneous nodule at subcutaneous fat layer of the left neck. The tumor was excised under the clinical impression of cervical lymphadenitis. On gross examination, the tumor was pale pink to gray, nodular and measures 0.8 cm x 0.7 cm. Microscopically, the tumor was composed of lobulated capillaries lined by endothelial cells containing mitotic figures and supported by fibrous stroma. Few prominent vascular structures were situated in the center of the lesion. It arose from the vessel wall and protruded into the vessel lumen (Fig. 1 A & B). There was no evidence of ulceration or inflammation. Immunohistochemically, the tumor cells and surrounding vascular walls were found to be strongly positive for CD31 and smooth muscle actin (Fig. 2 A & B). The histopathological finding was diagnostic for IVLCH. The patient was in good health at the time of discharge. The patient has been followed up, and there has been no recurrence since then.

Discussion

Lobular capillary hemangioma (LCH) is a

relatively common benign mucocutaneous lesion. This lesion was also called as pyogenic granuloma and considered as a reactive lesion secondary to trauma and pyogenic infection.

Intravenous lobular capillary hemangioma (IVLCH) is a rare benign lesion which seems reasonable to attribute the intravenous occurrence of LCH to a proliferation of the endothelium of the patient venous vasa vasorum with capillary formation [4].

IVLCH was first described in 1979. The most common sites are the veins of neck and upper extremities. Most patients present in their mid- to late 30s and there is a slight female predilection [1]. The clinical and radiological features are not characteristic enough to suggest the diagnosis. Most patients do not present any specific symptoms, but some patients with IVLCH present with extremity edema or a painless mass.

On gross examination, most lesions appear as a soft, whitish-gray, polypoid mass extending from the vein wall by a fibrovascular stalk. There is a characteristic perivenous artery, contained within the stalk, that serves as the feeding vessel. Usually, no thrombus, necrosis, or hemorrhage are found.

At light microscopical examination, the lesion shows an intraluminal polyp attached to the vein wall and composed of lobules of capillaries lined by flattened or rounded endothelial cells, separated by an edematous fibromyxoid stroma. There are elongated spindle cells with numerous mitotic figures. Vascular dilatation can be seen. There are no inflammatory cells within the mass [5].

The risk factors associated with IVLCH are not identified; however, some reports suggest these lesions are linked with

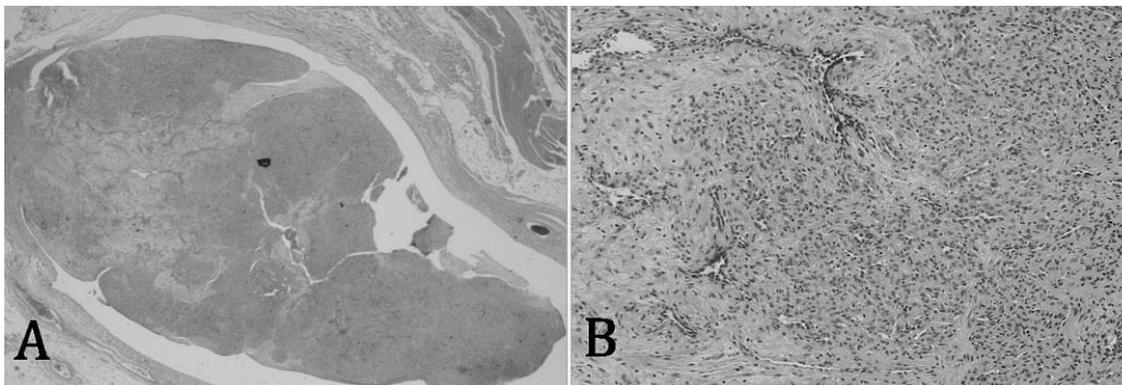


Fig. 1. (A) Low-power view of the lesion shows an intravascular lobulated soft tissue lesion, protruding from the vascular space (H and E, x 40) (B) High-power view shows a tumor composed of lobulated capillaries and lined by endothelial cells containing mitotic figures and supported by fibrous stroma. Some larger vascular structures are situated in the center of the lesion showing capillary proliferation in a fibromyxoid matrix (H and E, x 200).

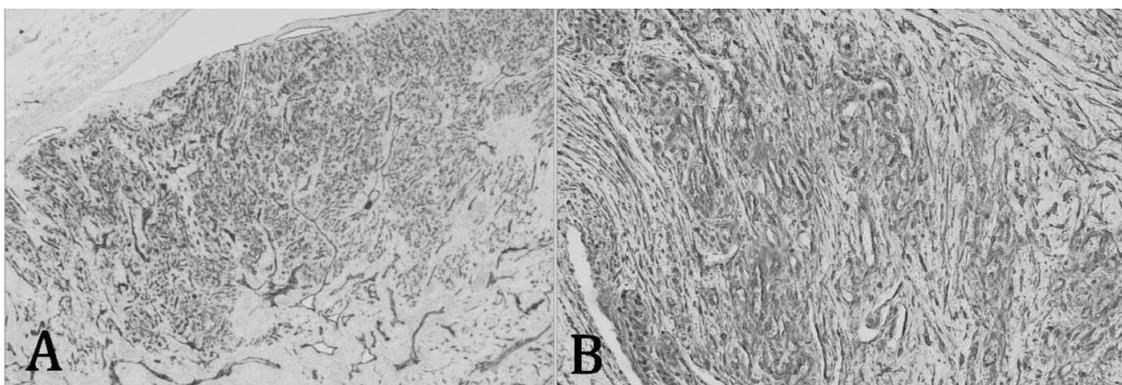


Fig. 2. The tumor cells stain positively for CD31(A) and smooth muscle actin(B).

arteriovenous malformations. The pathogenesis of IVLCH remains unclear, with a neoplastic process favored [2].

The correct pathological diagnosis is necessary to differentiate IVLCH from other intravascular lesions of angiomatous elements, such as angiosarcoma, intravascular papillary endothelial hyperplasia (IVPEP), intravenous atypical vascular proliferation (IVAVP), intravascular fasciitis and organized thrombus [4].

Angiosarcomas, which often appear as pale, gray-white solid, fleshy tumors, are

macroscopically indistinguishable from IVLCHs. On histologic examination, however, angiosarcomas does not show a lobular pattern, is usually seen in an older age group, and does not frequently involve the neck, arm or hand. It typically displays nuclear hyperchromasia, pleomorphism, and a high rate of mitoses. Staining for CD-31, CD-34, and von Willebrand factor is helpful in confirming the diagnosis [6].

Hemangioendotheliomas, another vascular tumor, demonstrate an intermediate histology. Microscopically, these tumors exhibit either

plump, cuboidal cells or more flattened, epithelioid cells [6].

Benign tumors such as IVPEP can be distinguished from IVLCH by its complex distinctive papillary architecture, presence of hemosiderin deposits, and association with organized thrombi [4].

A thrombus can be distinguished from IVLCH by the presence of the different fibrin deposits, the relatively paucity of capillaries, and the lack of lobular arrangement of the intraluminal growth.

IAVP has been described by Rosai and Ackerman [7]. This lesion, made up of proliferating endothelial cells occurs within the lumina of veins of the lower dermis and underlying soft tissues. IAVP can be distinguished by the greater cellularity and atypia of the proliferating endothelial cells [4]. Another differential feature of IAVP is the involvement of multiple veins, usually of medium size.

Furthermore, the association with thrombosis, whether a primary or secondary event, can be helpful by demonstrating fibrin within the lesion. In the past, recanalized DVT (deep vein thrombosis), which can be associated with infiltrating leukocytes, may have often been misdiagnosed as pyogenic granulomas. However, IVLCH clearly have a distinct, noninfectious, noninflammatory appearance that should not be mistaken for DVT [5].

The treatment of choice is complete local excision with a small portion of the vein. The prognosis is excellent. There is no tendency for recurrence or hematogenous spread.

Summary

IVLCH is a rare benign lesion, which is usually found in the veins of neck and the upper extremity. Controversy exists about the underlying pathogenesis of such lesions, but histologic examination demonstrates that these lesions are not of an inflammatory or infectious nature. With no characteristic clinical findings associated with these lesions, diagnosis can be difficult. Definitive diagnosis requires histologic examination of the excised lesion. Special stains, including immuno-histochemistry can be used for differential diagnosis. With complete excision, the prognosis is excellent and there is little risk of recurrence.

References

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