Case Report

# Case of caval lobular capillary hemangioma mimicking tumor thrombus

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**Abbreviations & Acronyms**
CA125 = carbohydrate antigen 125
CA19-9 = carbohydrate antigen 19-9
CD31 = cluster of differentiation 31
CD34 = cluster of differentiation 34
CT = computed tomography
IgG4 = immunoglobulin G4
IVC = inferior vena cava
IVLCH = intravenous lobular capillary hemangioma
MRI = magnetic resonance imaging
sIL-2R = soluble interleukin-2 receptor

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Abstract of this work has been partly presented and published at the International Symposium of Taiwan Urological Association (ISTUA) in 2016.

**Introduction:** We presented a rare case of caval lobular capillary hemangioma.
**Case presentation:** A 66-year-old female visited our department complaint with shadow defect in vena cava of right renal hilum appeared on computed tomography for periodically checking 3 years after radical hysterectomy with bilateral ovariectomy. Abdominal computed tomography identified a shadow defect of 35 mm in diameter in the inferior vena cava continuing posteriorly to a 35 mm mass of retroperitoneum. During the total removal of this lesion, we identified the lesion was connected to right ovarian vein. The specimen consisted of microcapillaries which formed reticular structure. Immunostaining of specimens identified positive CD31, CD34, and Factor 8 in all cells. Ki67 antibody was positive at 2–3% of all cells. These findings suggested the tumor was intravenous lobular papillary hemangioma.
**Conclusion:** This is the first report of intravenous lobular papillary hemangioma originated from right ovarian vein and extended to inferior vena cava.

**Key words:** inferior vena cava, intravenous lobular capillary hemangioma, ovarian vein.

## Keynote message

This is the first report of intravenous lobular papillary hemangioma originated from right ovarian vein and extended to inferior vena cava. Its etiology and clinical characteristics are reviewed based on literature search.

## Introduction

Lobular capillary hemangioma, also known as pyogenic granuloma, is a benign vascular tumor of the skin and mucosal membranes. Intravenous pyogenic granuloma also known as IVLCH is the intravascular counterpart of pyogenic granuloma, first reported by Cooper *et al.*[1] The pathogenesis of IVLCH remains unclear, with a neoplastic process favored.[2] Herein, we report a case with IVLCH originated from right ovarian vein, extending to IVC and mimicking a tumor thrombus.

## Case presentation

A 66-year-old female visited our department on account of shadow defect in vena cava of right renal hilum appeared on CT for periodically checking 3 years after radical hysterectomy with bilateral ovariectomy. Abdominal CT identified a shadow defect of 35 mm in diameter in the IVC continuing posteriorly to a 35 mm mass of retroperitoneum (Fig. 1). Physical examination revealed no mass in the abdomen. The results of complete blood count, chemistry, and urinalysis were all within the normal including carcinoembryonic antigen, CA19-9, CA125, neuron-specific enolase, sIL-2R, and IgG4. The mass showed an early phase strong enhancement followed by a delayed enhancement. By the abdominal MRI, the tumor showed a high signal intensity at T2-weighted images and slightly high signal at the diffusion-weighted images. The laboratory and image diagnosis indicated a tumor of vascular origin. However, we could not exclude angiosarcoma or tumor thrombus originated from prior

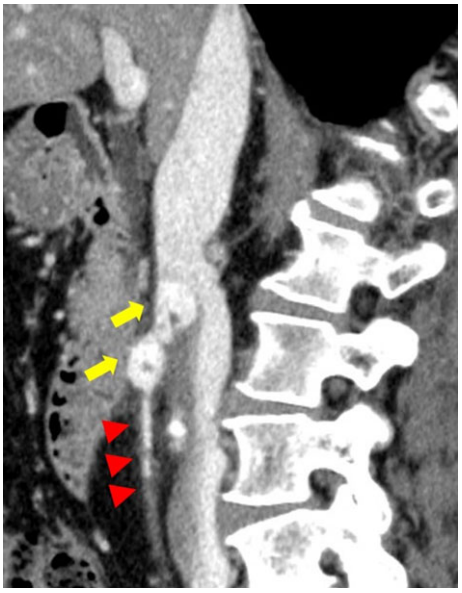

**Fig. 1** Contrast-enhanced computed tomographic image. Abdominal CT identified a shadow defect of 35 mm in diameter in the IVC continuing posteriorly to a 35 mm mass of retroperitoneum (yellow arrows). Red arrowheads indicate right ovarian vein.

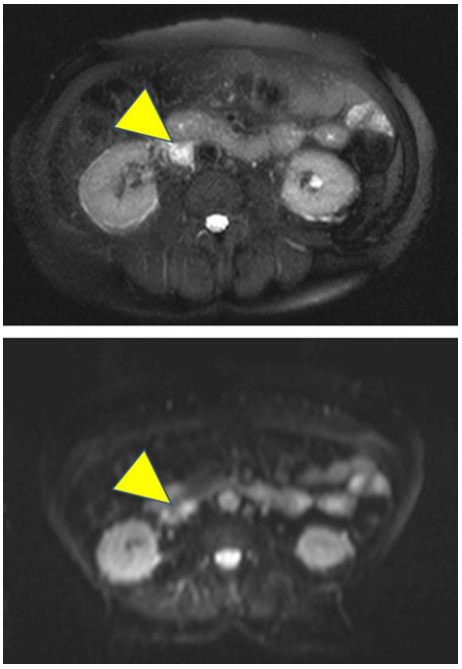

**Fig. 2** Plain MRI. MRI identifies an intracaval mass with a high signal intensity at T2-weighted image and a slightly high signal at the diffusion-weighted image (yellow arrowheads).

cancer; therefore, we performed a tumor removal. During the surgery, we identified an intravascular tumor of dumbbell like shape presenting in right ovarian vein extending to IVC. With the aid of vascular surgeon, we removed the mass together with the wall of ovarian vein and IVC. The defect of IVC was closed by using a graft of saphenous vein. Though we prepared extracorporeal circulation before surgery, the temporary blocking of IVC beneath the renal veins derived no reduction of blood pressure and we did not use the equipment.

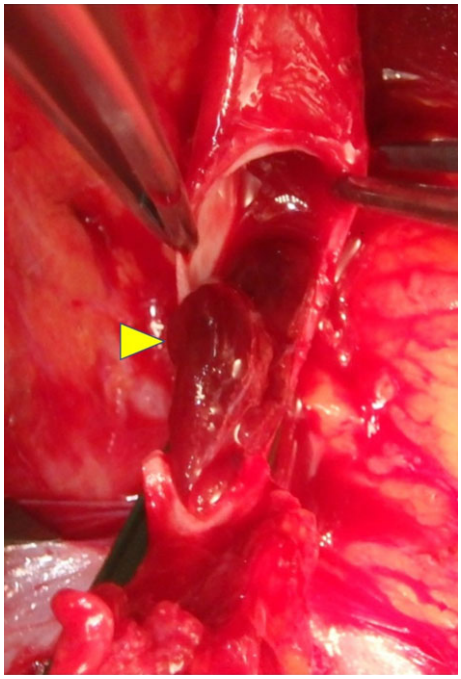

**Fig. 3** Macroscopic appearance of mass during surgery. A red and soft mass of 35 mm in diameter is identified in the IVC continuing posteriorly to a 35 mm mass of retroperitoneum (yellow arrowhead).

Microscopically, the tumor consisted of microcapillaries which formed reticular structure. Though the endothelial cells of them were relatively dense, there was no atypical cell or mitotic cell (Fig. 2). Immunostaining of specimens identified positive CD31, CD34, and Factor 8 in all cells. Ki67 antibody was positive at 2–3% of all cells (Fig. 2). These findings suggested the tumor was intravenous lobular papillary hemangioma. One year after surgery, she experiences no recurrence or new lesion of the tumor (Figs 3,4).

## Discussion

A PubMed search by using (*intravascular* or *intravenous) pyogenic granuloma* or *hemangioma,* and *intravascular lobular capillary hemangioma* following the selection, we obtained 74 cases among 38 reports (Table S1). According to these reports, the median age of this entity is 50 years old (range 3–75) with the male to female ratio of 2:3. The median size of lesion is 15 mm (range 4–85). Most of these tumors originated from the wall of veins of head and neck (33%), upper and lower extremity (28%), unknown (24%) and various miscellaneous veins including thoracic ($n$ = 2), renal ($n$ = 2), ovarian ($n$ = 2), mesenteric ($n$ = 1), azygos ($n$ = 1), iliac ($n$ = 1), and corpus carvenosum ($n$ = 1). One case with renal vein and present case with ovarian vein extended to IVC.

As precedent disease, 87% of cases accompanied no description, 9% had trauma or surgery, 3% inflammation, and 1% deep vein thrombosis. The patient of present case had a history of right ovariectomy and this might have induced IVLCH.

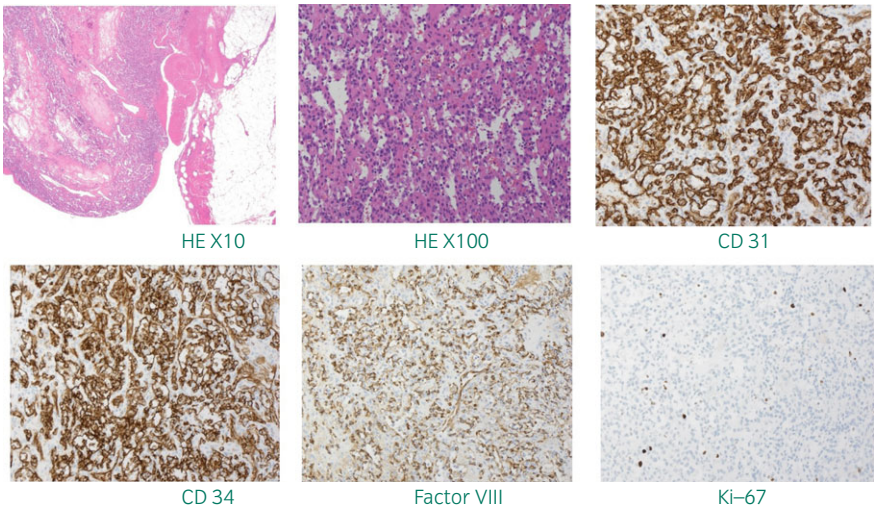


**Fig. 4** Pathological findings. Microscopically, the tumor consisted of microcapillaries which formed reticular structure. Though the endothelial cells of them were relatively dense, there was no atypical cell or mitotic cell. Immunostaining of specimens identified positive CD31, CD34, and Factor 8 in all cells. Ki67 antibody was positive at 2–3% of all cells.

Nine of 38 reports described its prognosis. Based on 10 cases with a median observation period of 1 year (0.5–7 years) only one case suffered from recurrence. These data indicate that it is a benign disease which shows favorable response for the surgical treatment.

In conclusion, we report a case of lobular capillary hemangioma in the right ovary vein. This is the first report of intravenous lobular papillary hemangioma originated from right ovarian vein extended to IVC.

## Conflict of interest

The authors declare no conflict of interest.

## References

1 Cooper PH, McAllister HA, Helwig EB. Intravenous pyogenic granuloma. A study of 18 cases. *Am. J. Surg. Pathol.* 1979; **3**: 221–8.

2 Maddison A, Tew K, Orell S. Intravenous lobular capillary haemangioma: ultrasound and histology findings. *Australas. Radiol.* 2006; **50**: 186–8.

## Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

**Table S1.** Literatures related to the current case.