Case Report

A case of retroperitoneal venous malformation resected by laparoscopic surgery

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Abbreviations & Acronyms
ADC = apparent diffusion coefficient
CT = computed tomography
DWI = diffusion weighted image
MRI = magnetic resonance imaging

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Introduction: Among vascular malformations, venous malformations are the most common type. Among these, retroperitoneal venous malformations are extremely rare.
Case presentation: A 60-year-old woman was diagnosed with a retroperitoneal tumor 4.5 cm in diameter by abdominal computed tomographic scan. We had difficulty judging whether the tumor was benign or malignant. We performed laparoscopic surgery in order to remove the tumor and make a precise diagnosis. The pathological diagnosis was a venous malformation.
Conclusion: Venous malformation located in the retroperitoneum is very rare, and there were few cases that could be removed by laparoscopic surgery. Laparoscopic surgery may be beneficial both for treatment and diagnosis of patients with a small retroperitoneal venous malformation.

Key words: laparoscopic surgery, retroperitoneal tumor, soft coagulation system, vascular malformation, venous malformation.

Keynote message

We report a case of retroperitoneal venous malformation that was removed by laparoscopic surgery. Laparoscopic surgery may be beneficial both for treatment and diagnosis of patients with a small retroperitoneal venous malformation.

Introduction

Among vascular malformations, venous malformations are the most common (54%), with a male–female ratio of 1:1–2, most of which is sporadic at about 90%.[1] The size and distribution of venous malformations vary and occur anywhere on the face, trunk, and limbs, most often in the head and neck. In contrast, retroperitoneal venous malformations are extremely rare. We report a case of venous malformation in the retroperitoneum that was discovered accidentally, with a review of the literature.

Case presentation

The patient was a 60-year-old female. A liver tumor was observed on abdominal ultrasonography during the disease screening. A CT was performed, a retroperitoneal tumor was found incidentally. The patient was referred to our department for a detailed examination and treatment for the retroperitoneal tumor. No prior medical history was collected. Family history was unremarkable. Laboratory analysis revealed that interleukin-2 receptor levels and adrenal hormone levels were all within the normal ranges. Contrast CT scan revealed an almost low-density heterogeneous tumor with slight enhancement. The tumor was 45 mm in diameter with calcifications and was located at the ventral portion of the right renal vein (Fig. 1). MRI showed a signal equal to skeletal muscle on T1WI, a high signal nodule on T2WI, a high signal on DWI, and no signal decrease on ADC (Fig. 2).

An adrenal tumor, schwannoma, myelolipoma, paraganglioma, lipoma, liposarcoma, teratoma, etc. were considered as differential diagnoses of the tumor. We suspected Castleman's

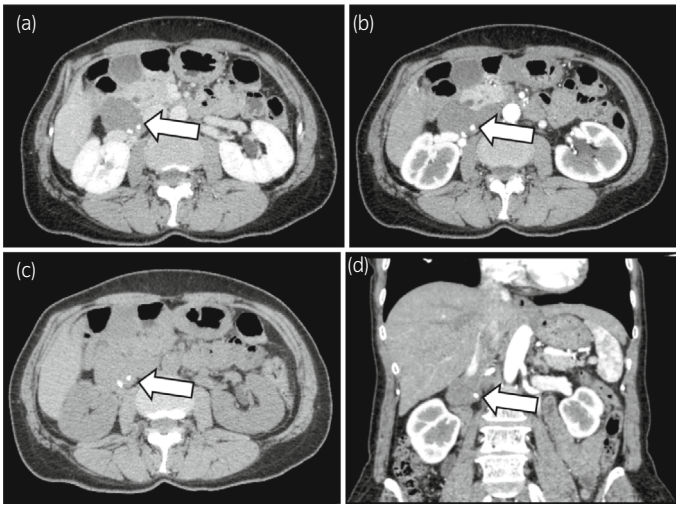


Fig. 1 Contrast-enhanced CT shows a well-defined round, 45 mm cystic mass with a rim of soft tissue in the retroperitoneal region and located at the ventral portion of the right renal vein. Slight enhancement of the peripheral rim of soft tissue is seen after intravenous administration of the contrast medium (a–c: axial image, d: coronal image).

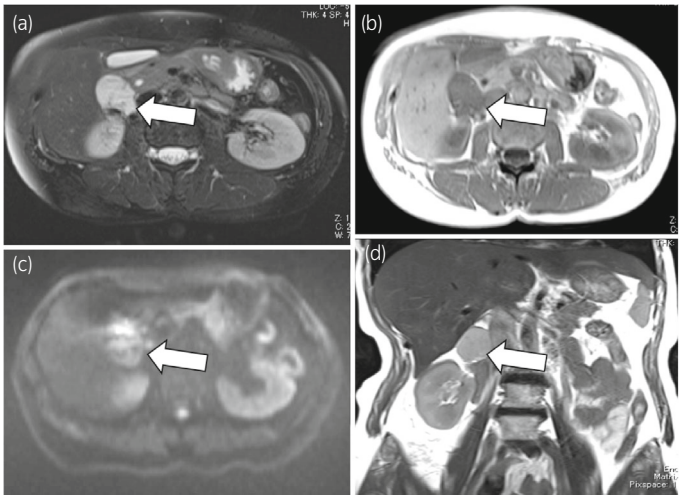


Fig. 2 MRI showed a signal equal to skeletal muscle on T1WI, a high signal nodule on T2WI, a high signal on DWI, and no signal decrease on ADC (a–c: axial image, d: coronal image).

disease or nerve-derived tumor because it was a retroperitoneal tumor with calcification. We had difficulty judging whether the tumor was benign or malignant. We performed laparoscopic surgery in order to remove the tumor and make a precise diagnosis. The surgery time was 240 min, and the blood loss was less than 5 mL. The tumor was passive but had mild adhesions to surrounding tissues (Fig. 3a). Bleeding was observed from the surface of the tumor detaching from the surrounding tissues (Fig. 3b), and a soft coagulation system with a monopolar electrode was effective for hemostasis (Fig. 3c). Feeding blood vessels from the right renal veins and right gonadal vein to the tumor were found, and each were treated with LigaSure™ (Fig. 3d). Macroscopic examination of the tumor revealed a multilocular mass, measuring 36 × 30 mm, consisting of cystic and solid components and containing two bean-sized calcifications (Fig. 4a). Hematoxylin–eosin staining showed that expanded blood vessels were growing against the background of fibrous connective tissues and mature adipocytes (Fig. 4b). Victoria blue staining-positive elastic fibers and Alpha-smooth muscle actin-positive cells were found on the vessel wall. No atypical cells with hyperproliferation or mitotic division were seen. The tumor was diagnosed as a venous malformation based on pathological results. The patient was discharged 6 days after the operation without any problems. The patient was alive without recurrence at 1 year after the operation.

Discussion

Venous malformations are thought to occur sporadically and account for about 94% of vascular malformations.[1] Retroperitoneal tumors account for 0.2% of all benign and malignant tumors, and retroperitoneal vascular malformations account for only 2% of them. Vascular malformations were previously classified as hemangiomas. In 1929, Harris *et al*. reported fibroangioma as the first case of retroperitoneal vascular malformation.[2] Although the mechanism of onset has not been clarified, many reports generally consider it congenital. In the process of development, it has been reported that the tumor is caused by a malformation generated from a specific blood vessel or from the residual tissue during the embryonic period.

The diagnosis of retroperitoneal venous malformation is difficult because they are located deep within the trunk. CT, MRI, Doppler sonography, and angiography are usually used for the diagnosis and delineation of anatomy and planning of treatment.[3] For vascular malformation, especially

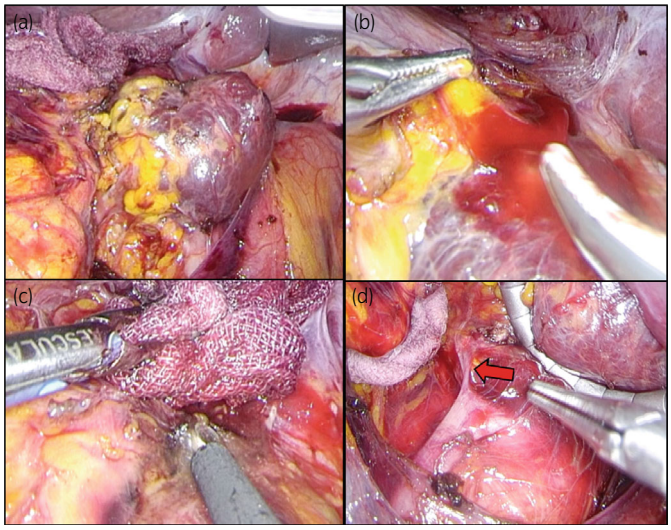
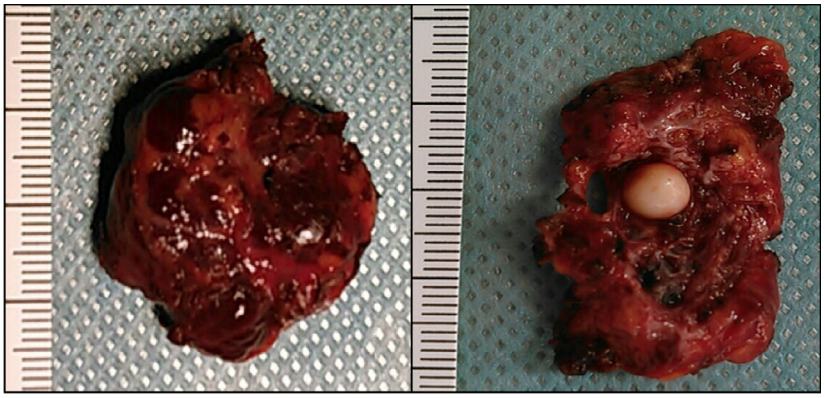


Fig. 3 Surgical images. (a) Retroperitoneal tumor. (b) Bleeding from the surface of the tumor. (c) Soft coagulation. (d) Feeding blood vessels from the right gonadal vein to the tumor.



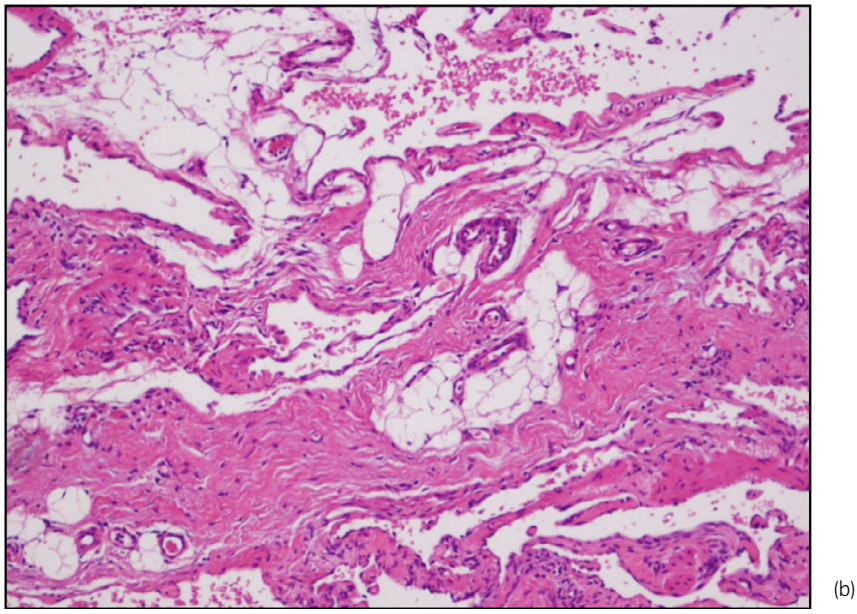


Fig. 4 (a) The resected specimen reveals a multilocular tumor, which measures 36 × 30 mm and consists of two Phleboliths, macroscopically. (b) Hematoxylin–eosin staining showed that expanded blood vessels are growing against the background of fibrous connective tissue and mature adipocytes.

arteriovenous malformation, angiography provides useful information to identify inflow arteries and outflow veins. If vascular malformation was assumed before surgery, it may have been possible to avoid surgery, and choose transvenous embolization by performing angiography.[3] We did not choose angiography as a diagnostic tool because we had not strongly suspected venous malformation prior to surgery. If we expected, angiography might be an option for less invasive treatment, such as intravenous treatment (e.g. embolization) or sclerotherapy.

Vascular malformations increase in proportion to growth if left untreated. Venous malformations can lead to collagen deposition and phlebolith formation when thrombi form, and x-rays and CT scans can show calcified lesions, which may aid in diagnosis.

Treatments for vascular malformation include follow-up and excision, vascular embolization, radiation and laser treatments, and sclerotherapy. In the case of proliferative or symptomatic vascular malformation, radical surgery is indicated.[3]

In this case, the laparoscopic surgery was used for tumor resection. A laparoscopic surgery under magnified view can be a reasonable and safe approach, and postoperative recovery is faster after laparoscopic surgery when compared with recovery after open surgery. Unexpected tumor adhesion to surrounding tissues can sometimes be appreciated during the laparoscopic procedure, and these findings often necessitate conversion to an open procedure. Recent reports suggest that laparoscopic resection is a safe and feasible operative approach for retroperitoneal hemangioma.[4,5] More accurate preoperative diagnosis of retroperitoneal tumors can help select more optimal treatment.

Vascular malformation is difficult to distinguish from malignant tumors due to invasive growth into surrounding tissues. Laparoscopic surgery for retroperitoneal venous malformation was performed in only 11 cases including our case in the world.[6–14] Since vascular malformation had a high rate of strong adhesions to the surroundings,[15] laparotomy was performed in most cases. Laparoscopic surgery might be indicated in those cases if it was detected earlier. Furthermore, a case of DaVinci robotic-assisted resection of cavernous hemangioma had been reported.[9]

In this case, the tumor showed slight adhesion to the surrounding tissues, and hemostasis by soft coagulation was effective for the bleeding from the tumor surface.

If the diagnosis is delayed, the tumor may grow invasively, which makes adhesion with surrounding tissues stronger and makes laparoscopic surgery difficult. According to a previous report, where the tumor was larger than 7 cm, adhesions to the surrounding tissue were strong, and the tumor was removed by laparotomy. For tumors of 5 cm or less, laparoscopic surgery was performed because of mild infiltration.[6] For large venous malformations larger than 7 cm, surgery may be performed on an inappropriate excision line for fear of major bleeding, resulting in positive surgical margins and recurrence.

Venous malformation consists of large and small atypical veins that are continuous with nearby veins. If the blood vessels that communicate with the tumor can be identified and treated first, the tumor may be completely resected while controlling bleeding. Retroperitoneal venous malformations are at risk of recurrence if not completely resected.[15,16] It has been reported that 70–80% of primary retroperitoneal tumors were malignant tumors;[17] therefore, in this case, surgery was decided to make a definitive diagnosis. Retroperitoneal venous malformation should be kept in mind as one of the differential diagnosis of retroperitoneal hypovascular tumors.

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Author contributions

Tomoki Kai: Data curation. Tadasuke Ando: Supervision. Toshitaka Shin: Supervision. Hiromitsu Mimata: Supervision.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an Institutional Reviewer Board

N/A.

Informed consent

Informed consent for publication was obtained from the patient.

Registry and the Registration No. of the study/trial

N/A.

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