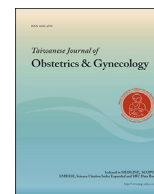




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Research Letter

Laparoscopic treatment of Castleman's disease in a patient with a history of malignant Brenner tumor

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Dear Editor,

Castleman's disease (CD) is a rare, atypical, lymphoproliferative disease of unknown etiology. The most common location for CD is the mediastinum (63%) [1]. A retroperitoneal location has been reported in 7% of patients, with only 2% of cases involving the pararenal region [2]. For this reason, CD located in the retroperitoneum is difficult to differentiate from other benign or malignant lesions. Here, we report a patient with CD located in the anterior lower pole of the right kidney who had been diagnosed with a malignant Brenner tumor after hysterectomy.

A 56-year-old woman visited our hospital with a chief complaint of a growing abnormal retroperitoneal mass. She had undergone a total abdominal hysterectomy (TAH) and bilateral salpingo-oophorectomy (BSO) due to a malignant Brenner tumor at another university hospital 3 months previously. Baseline serum levels of CA-125 and CA 19-9 were normal (5.59 U/mL and 8.11 U/mL, respectively). An abdominal computed tomography (ACT) scan revealed a 3.0-cm nodule in the anterior lower pole of the right kidney that had been visualized by ACT prior to the TAH and BSO. The mass had increased in size over the course of 3 months (Figure 1). No distant metastases or other abnormal findings were noted on ACT. We suspected that the mass was a metastatic lymph node and performed laparoscopic retroperitoneal removal.

The patient was placed in the dorsolithotomy position, and four ports were used for the procedure (Figure 2). The first assistant gradually rotated the telescope 180° clockwise from the pelvic

cavity toward the upper abdominal cavity during the procedure to allow observation of the same direction on the monitor as that viewed by the surgeon during the laparotomy. The retroperitoneal space was entered after washing cytology. Initially, we pushed aside the small intestine and mesentery to the left and inferior direction, followed by lifting of the transverse colon using atraumatic forceps. After performing a transverse mesocolon incision along the right side of the inferior vena cava and entering the lower pole of the right kidney, we discovered an oval-shaped mass (Figure 3). The mass was carefully dissected to avoid the ileocolic vessels, and the feeding vessels arising from the aorta were clipped using Hem-o-Lok clips (Teleflex Medical, Wayne, PA, USA). The tumor was delivered through a 10-mm port with an Endopouch (Ethicon, Somerville, NJ, USA). Histopathologic evaluation revealed unicentric, hyaline vascular-type CD (U-HVCD; Figure 4). The washing cytology specimens revealed no malignant cells. Post-operative recovery was uneventful, and the patient was discharged on the 3rd postoperative day. After 55 months, there continues to be no disease recurrence.

Many investigators proposed possible CD etiologies, but none of the suggestions have been proven. The low incidence of CD is responsible for the incomplete understanding of the disease, with current knowledge based largely upon case reports and histopathologic reviews. CD exists in two histologic forms: a HVCD and a plasma-cell-type CD (PCCD). Clinically, the two types were described as unicentric (localized) and multicentric types. Most cases of CD (70%) are U-HVCD, with younger patients usually presenting with isolated benign asymptomatic lymphadenopathies [3]. Unlike U-HVCD, most patients with PCCD have symptoms, such as fever, diaphoresis, weight loss, night sweats, and fatigue [4].

When CD occurs in the retroperitoneal space, it is important to distinguish it from other retroperitoneal masses. Differential diagnoses include benign neoplasms, lymphomas, sarcomas, neural tumors, and metastatic lymph nodes. Although the radiographic findings of CD are nonspecific, contrast-enhanced CT generally reveals a homogeneous, well-defined, localized abdominal mass, with enhancement probably due to the extensive vascularization [5]. On magnetic resonance imaging (MRI), the mass exhibits low signal intensity on T1-weighted images and high signal intensity on

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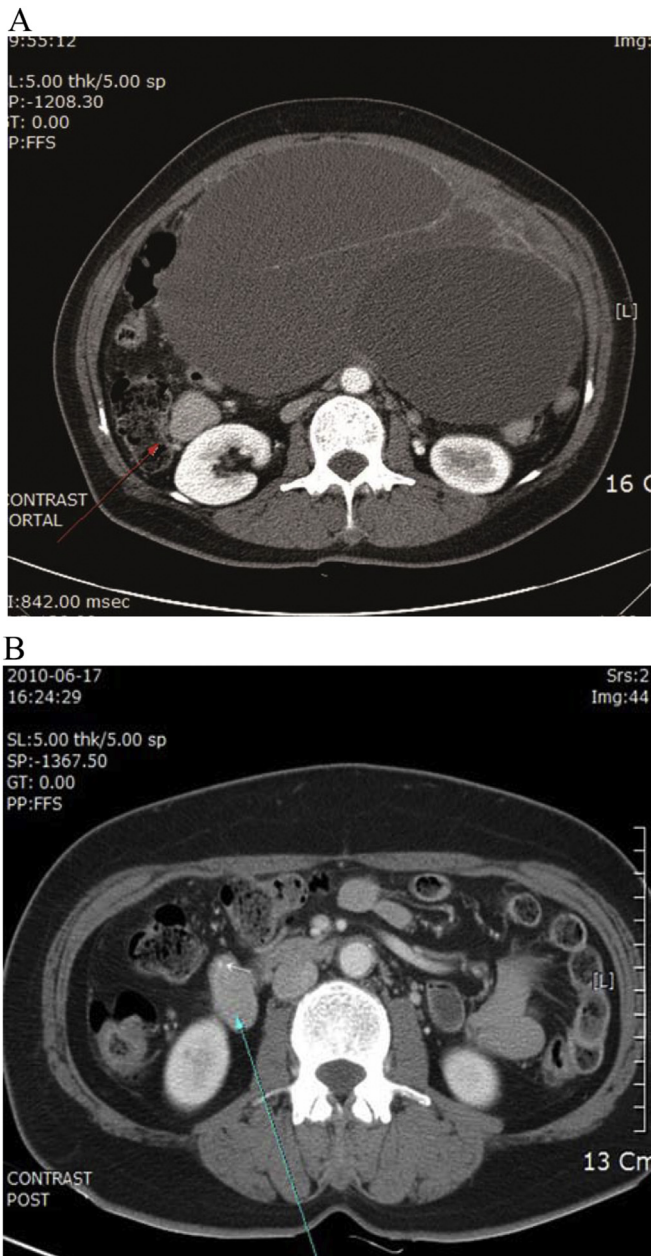


Figure 1. CT scans reveals a well-enhanced 3-cm mass in the anterior lower pole of the right kidney. This nodule was a homogenous solid mass with eccentric calcifications. (A) Pre-operative CT before total abdominal hysterectomy and bilateral salpingo-oophorectomy at another university hospital. (B) Axial contrast-enhanced CT shows a homogeneous retroperitoneal soft-tissue mass (blue arrow) displacing the pancreaticoduodenal vessel (white arrow) anteriorly. CT = computed tomography.

T2-weighted images [6]. A low apparent diffusion coefficient calculated from diffusion-weighted MRI can provide useful information in the differential diagnosis of retroperitoneal masses [5]. Because F-18 fluorodeoxyglucose positron-emission tomography/computed tomography (FDG-PET/CT) findings are seldom reported [7], the utility of FDG-PET/CT is uncertain. In this case, we used only ACT, and it was very difficult to predict CD, given the existence of no specific findings of CD in ACT. The fact that the patient had had a malignant Brenner tumor and a growing retroperitoneal mass confused us regarding the correct diagnosis.

High tissue concentrations of interleukin (IL)-6 (normal range, < 4.0 pg/mL), positive immunostaining for cluster of differentiation

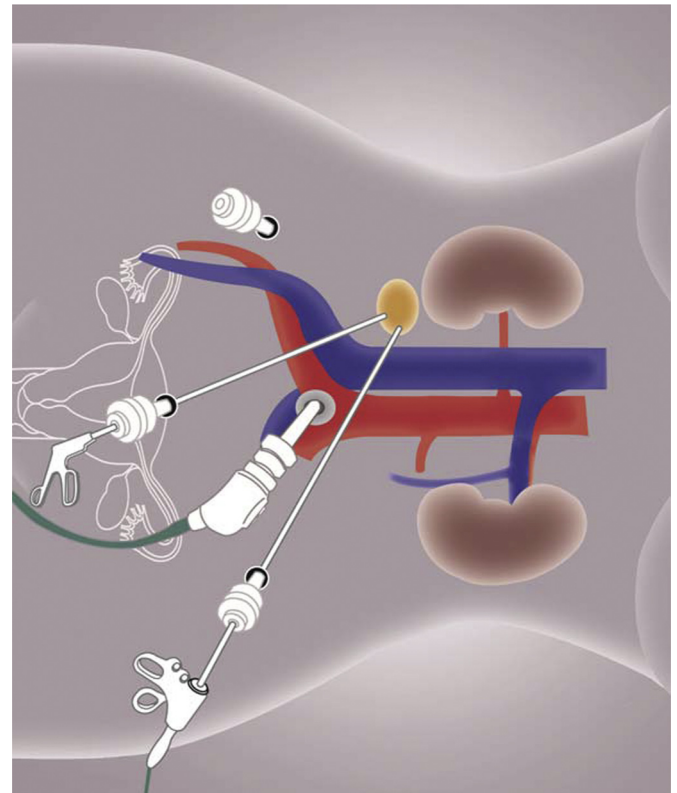


Figure 2. Intra-operative position and arrangement of the trocar and instruments.

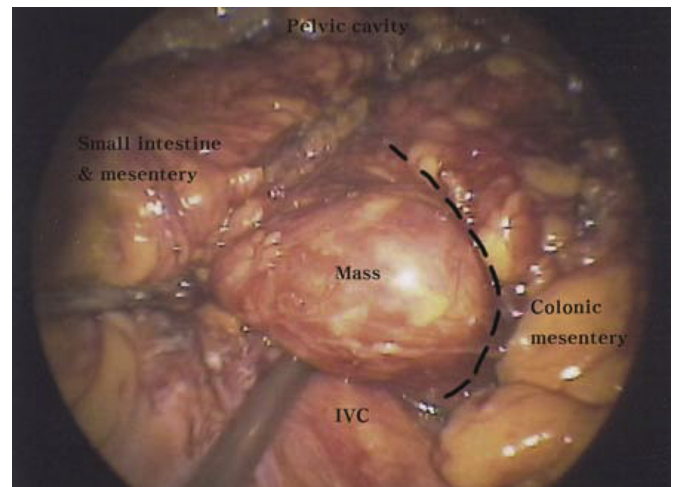


Figure 3. Intra-operative view of the tumor and surrounding structure after a transverse mesocolon incision (broken line represents the incision site). IVC = Inferior vena cava.

20 through fine-needle aspiration biopsy (FNAB), and measurement of serum IL-6 levels may provide useful information for a diagnosis of CD [8,9]. Therefore, to differentiate between CD and other retroperitoneal masses, appropriate clinical findings, typical radiologic features, and serum IL-6 levels should be considered. When physicians are confident of the clinical diagnosis of CD, it can be confirmed by FNAB; however, when cases are suspected of being malignant masses, such as lymphomas, FNAB is not recommended.

Although laparoscopic treatment of CD has only occasionally been reported, laparoscopic treatment of CD by a gynecologist has

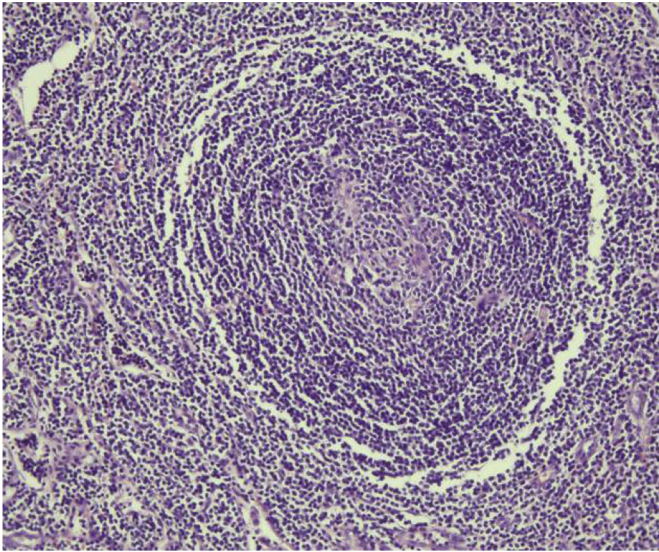


Figure 4. Hyaline vascular-type of Castleman's disease. Note the histopathologic "onion skin" appearance. The follicle is surrounded by a broad mantle zone composed of concentric rings of small lymphocytes. (H&E stain; 200 \times). H&E = hematoxylin and eosin.

not been previously described. Traditionally, many surgeons, especially urologists, have used a lateral transabdominal approach (LTA) or a posterior retroperitoneal approach (PRA). However, these approaches are not practical for gynecologists. Another method is the transumbilical approach, which suits gynecologists better than LTA and PRA. In the dorsolithotomy or supine position, the medial reflection of the colon causes a major problem for the lateral peritoneal approach. By contrast, for the transmesocolic approach, the blood vessels, such as the ileocolic and other feeding vessels from the aorta, are a major concern, given the risk of massive hemorrhage during surgery. However, the advantages of the transmesocolic approach include minimal handling of the bowel and restoration of the retroperitoneal anatomy after closing the mesocolic window [3].

Unicentric CD is generally cured after resection of the affected lymph nodes, and the prognosis is favorable. However, multicentric CD may be chronic, with remissions and exacerbations requiring continuous therapy and a reported median survival of 5–19 months [10]. Patient with unicentric disease should undergo an additional radiologic evaluation 6–12 months after treatment to confirm no recurrence, and for patients with multicentric disease, close follow-up and periodic surveillance are necessary to detect concurrent or ensuing malignant lesions [3].

In conclusion, although a correct pre-operative diagnosis was not made in our case, unicentric CD should be considered in the differential diagnosis of retroperitoneal masses when they present in an appropriate clinical setting and present typical radiographic features. If a correct diagnosis of retroperitoneal unicentric CD, including pararenal lesions, is established, the patient can be managed safely by laparoscopy using a transmesocolic approach.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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