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A Rare 40-cm Retroperitoneal Isolated Enteric Duplication Cyst Mimicking an Ovarian Neoplasm: A Case Report

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Abstract

Background: Isolated enteric duplication cysts (IEDCs) occurring in the retroperitoneum are extremely uncommon, and reports of those exceeding 40 cm in size are exceptionally rare. Diagnosing such lesions preoperatively remains difficult, especially when they mimic ovarian tumors. Case presentation: We present a 37-year-old woman (G3P3) with acute abdominal distension and pain. Radiologic imaging identified a 40-cm multiloculated cystic lesion containing calcifications and fat, initially suggestive of a giant mature teratoma with hydrosalpinx. Tumor markers (CA125, CA19-9, CEA) were elevated. Exploratory laparotomy revealed a smooth, large cystic mass that occupied the entire peritoneal cavity, and a small incision allowed for cyst drainage. The aspirated cystic fluid exhibited features consistent with intestinal contents, and the inner wall of the cyst demonstrated a mucosa-like appearance reminiscent of intestinal epithelium. Upon decompression, the uterus and adnexa appeared normal, and the mass was found to originate from the retroperitoneum adjacent to the descending colon. Histological evaluation established the diagnosis of a retroperitoneal IEDC. The lesion was excised completely one month later. One-year follow-up showed no recurrence. Conclusion: This case underscores the diagnostic complexity of massive retroperitoneal cystic tumors, the value of intraoperative assessment, and expands the clinical spectrum of giant IEDCs.

Keywords

Isolated Enteric Duplication Cyst, Ovarian Tumor Mimic, Giant Abdominal

Cyst, Tumor Marker Elevation, Surgical Diagnosis

1. Introduction

Retroperitoneal isolated enteric duplication cysts (IEDCs) are rare developmental anomalies typically reported in pediatric populations, with adult cases being exceedingly infrequent. These lesions are characterized by their lack of communication with the gastrointestinal tract and are thought to derive from aberrant embryological processes [1]. Retroperitoneal IEDCs often mimic ovarian or other intra-abdominal cystic masses, complicating preoperative diagnosis for gynecologists.

Although tumor markers like CA125, CA19-9, and CEA are frequently elevated in malignancies, they may also be nonspecifically increased in benign cystic entities such as IEDCs [2] [3]. Imaging modalities including computed tomography (CT) and magnetic resonance imaging (MRI) may demonstrate multiloculated cysts with calcified or fatty elements, yet distinguishing IEDCs from mature ovarian teratomas or other neoplasms based solely on imaging remains challenging [4] [5].

Massive retroperitoneal IEDCs exceeding 40 cm are particularly rare. This report details an unusual case of such a lesion, initially misdiagnosed as an ovarian tumor, and includes a literature review to contextualize its clinical and pathological characteristics.

2. Case

A 37-year-old woman (G3P3) with no notable medical or familial history presented with sudden-onset abdominal discomfort and bloating of two weeks' duration. Physical examination revealed a significantly distended and tender abdomen, precluding supine positioning.

Laboratory findings demonstrated elevated inflammatory markers (CRP: 15.46 mg/dL; WBC: 13,900/μL) and mildly impaired renal function (creatinine: 1.01 mg/dL). Tumor markers were also elevated: CA125 was 83.9 U/mL (normal range: 0 - 35 U/mL); CA19-9 was 691.5 U/mL (normal range: 0 - 37 U/mL); and CEA was 18.8 ng/mL (normal range: 0 - 5.0 ng/mL). Abdominal CT revealed a massive 40-cm multilocular cystic mass with tubular structures, internal calcifications, and adipose tissue. Left-sided hydronephrosis was also noted. Given the presence of multilocular cystic architecture with partial tubular structures, as well as internal calcifications and fatty components, the imaging findings suggested a mature cystic teratoma with possible hydrosalpinx (**Figure 1**).

Although malignancy could not be definitively excluded, the acute clinical presentation with suspected torsion or infection necessitated an emergency exploratory laparotomy. Intraoperatively, a smooth, large cystic mass occupied the entire peritoneal cavity without ascites or visible peritoneal metastasis. As the up-

per boundary of the mass and the reproductive organs were indiscernible, a small incision allowed for cyst drainage. The aspirated cystic fluid exhibited features consistent with intestinal contents, and the inner wall of the cyst demonstrated a mucosa-like appearance reminiscent of intestinal epithelium. Upon decompression, the uterus and adnexa appeared normal, and the mass was found to originate from the retroperitoneum adjacent to the descending colon (Figure 2). Biopsies were obtained, and histological analysis confirmed the diagnosis of an enteric duplication cyst. Because the tumor was exceedingly large and complete excision was considered technically difficult in a single-stage operation, complete surgical excision was achieved a month later via retroperitoneal dissection. The resected specimen measured 40 cm and showed features typical of enteric duplication: a mucosal lining akin to intestinal epithelium, organized smooth muscle, and a serosa-like outer wall. Microscopy revealed mucin-secreting epithelium, edematous submucosa, and interlacing smooth muscle bundles, consistent with IEDC (Figure 3). Foci of calcification and adipose tissue were observed. No malignancy or atypia was detected. Lack of anatomical continuity with the bowel confirmed the lesion as a retroperitoneal IEDC.

Postoperative recovery was uneventful. Tumor markers normalized, inflammation subsided, and hydronephrosis resolved. No recurrence was noted at the one-year follow-up.

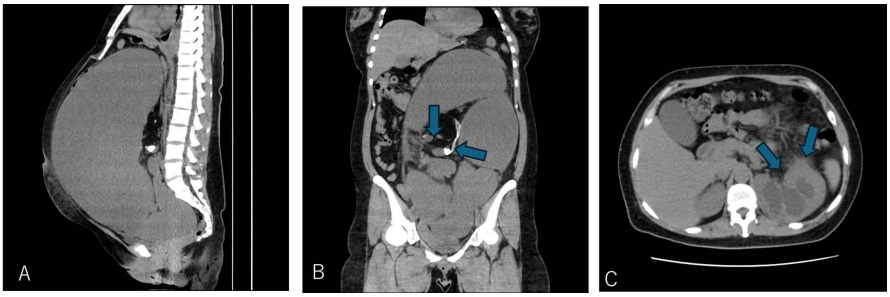

Figure 1. Abdominal CT revealed a massive 40-cm multilocular cystic mass (A) with tubular structures, internal calcifications, and adipose tissue (arrows), displacing the intestines to the right (B). Hydronephrosis of the left kidney was also noted (arrows) (C).

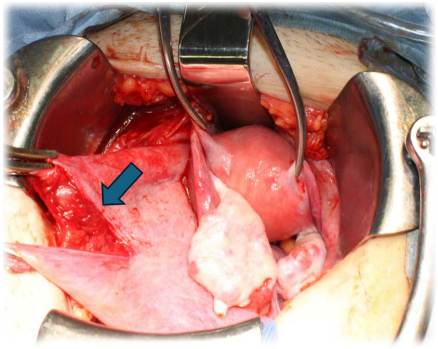

Figure 2. A diagnostic laparotomy revealed that the uterus and adnexa were normal. The mass originated from the retroperitoneum, adjacent to the lateral aspect of the descending colon (arrow).

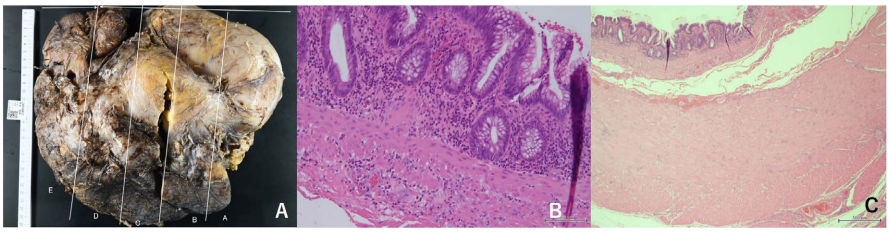

Figure 3. Macroscopic and microscopic findings of the tumor. Macroscopically, the cyst measures 40 cm in diameter (A). Microscopically, the cyst is lined by mucin-producing epithelium (B) with underlying submucosal edematous stroma and interspersed smooth muscle bundles (C).

3. Discussion

Enteric duplication cysts are histologically defined as spherical or tubular structures lined by gastrointestinal-type mucosa and enveloped by well-organized smooth muscle and serosal layers. In the present case, the duplication cyst was located in the retroperitoneum and exhibited no communication with the alimentary tract. These non-communicating, isolated cysts share histological features with conventional enteric duplication cysts—namely, gastrointestinal epithelial linings and smooth muscle layers—despite lacking anatomical continuity with the digestive system. Similar lesions have been reported [4] at various extraluminal sites, including the tongue, pleural cavity, liver, pancreas, biliary tract, and retroperitoneum, as exemplified in this case. IEDCs in adults remain exceedingly rare, and diagnostic standards, particularly regarding their potential for malignancy, are not yet well established.

This case is unique in several respects. First, the extraordinary size of the lesion—measuring 40 cm in diameter—is exceptional. A review of the literature reveals that most documented retroperitoneal IEDCs are significantly smaller, typically between 5 and 15 cm [4] [5]. To our knowledge, a lesion of this size ranks among the largest reported. Although imaging alone often fails to provide a definitive distinction between benign and malignant lesions, previous reports have indicated that large tumor size, the presence of calcification, and mural nodularity may suggest malignancy [5]. Second, the patient presented with acute symptoms resulting from rapid cystic enlargement, producing considerable mass effect, including hydronephrosis and displacement of intra-abdominal organs. Such rapid progression raised concern for malignant transformation—a phenomenon that, while rare in enteric duplication cysts, has been described in the literature [6] [7]. The markedly elevated tumor markers CA125, CA19-9, and CEA further reinforced this suspicion. However, elevated levels of CA19-9 and CEA have also been observed in duplication cysts lined by mucin-secreting epithelium in the absence of malignancy [2] [3]. Chronic irritation, cystic degeneration, or infection may contribute to these elevations. In the present case, the presence of inflammation within the cyst wall may have increased epithelial permeability, promoting the release of tumor markers. In addition, the rapid enlargement of the tumor may have elevated intracystic pressure, leading to diffusion or leakage of these markers

into the bloodstream. Third, establishing a preoperative diagnosis proved particularly challenging. The finding of hydronephrosis suggested a retroperitoneal lesion, but the identification of calcifications and fatty components on imaging was more consistent with a mature cystic teratoma [8] [9]. Inomata *et al.* [5] have noted that temporal changes in lesion size and increasing internal heterogeneity may aid in distinguishing IEDCs from other cystic masses. Nevertheless, definitive diagnostic criteria for IEDCs remain unclear. In contrast, multilocular cystic lesions such as lymphangioma or peritoneal mesothelioma rarely present with calcifications, which may help narrow the differential diagnosis.

In retrospect, the detection of a large multiloculated cyst containing both calcifications and fat should prompt the inclusion of IEDC in the differential diagnosis, along with teratoma, cystic lymphangioma, and cystic mesothelioma. However, no single imaging modality can definitively differentiate among these entities prior to surgery. Intraoperative assessment played a pivotal role in this case. Identification of intestinal-type mucosa within the cyst and confirmation of retroperitoneal origin upon decompression redirected the surgical team's suspicion from an ovarian neoplasm to a retroperitoneal enteric duplication cyst. This underscores the importance of meticulous intraoperative exploration, including evaluation of the cyst contents and anatomical origin, especially when preoperative imaging is inconclusive. Retroperitoneal cystic masses are uncommon and require consideration of a broad differential diagnosis, including cystic lymphangiomas, mesenteric cysts, tailgut cysts, and cystic teratomas [9]. Among these, the presence of calcification is often suggestive of a teratoma; however, calcifications may also occur in enteric duplication cysts, particularly those containing mucinous material or demonstrating chronic inflammation. Calcifications are rarely observed in lymphangiomas or peritoneal mesotheliomas. Histologically, unlike lymphangiomas derived from endothelial cells or peritoneal mesotheliomas arising from mesothelial cells, our case exhibited the classic features of an enteric duplication cyst: a mucosa-lined cyst wall composed of smooth muscle layers mimicking the intestinal wall and the absence of epithelial atypia. No communication with adjacent intestinal structures was observed, confirming its classification as an isolated lesion [1]. In terms of management, complete surgical excision remains the treatment of choice for retroperitoneal duplication cysts due to the associated risks of infection, hemorrhage, rupture, and rare malignant transformation [6] [7]. Incomplete resection may predispose to recurrence and should be avoided whenever possible. This case offers valuable clinical insights for gynecologists encountering large abdominal cystic lesions. Although rare in adults, retroperitoneal IEDCs should be considered within the differential diagnosis of giant abdominal cystic tumors. Importantly, elevated tumor markers are not definitive indicators of malignancy in these cases and should be interpreted cautiously. Additionally, intraoperative findings often play a crucial role in establishing the final diagnosis and guiding appropriate surgical management.

4. Conclusion

Retroperitoneal IEDCs in adults are exceedingly rare and may mimic large ovarian tumors, complicating preoperative diagnosis. Definitive identification relies on the integration of imaging, intraoperative evaluation, and histopathological confirmation. Increased clinical awareness of this unusual entity is essential for gynecologists managing atypical abdominal cystic lesions, to ensure precise diagnosis, tailored surgical planning, and optimal patient outcomes. When the ovarian origin cannot be clearly determined intraoperatively, assessment of the anatomical location after cyst decompression and evaluation of the cystic fluid characteristics can aid in differential diagnosis.

Availability of Data and Materials

The data are contained within this article.

Author Contributions

KT—Data curation; Formal analysis; Original draft writing. RA, AY—Data curation; Formal analysis. YY, MS, TI, HH, YK—Review & Editing.

Ethics Approval and Consent to Participate

All clinical information and images used in this paper were approved by the Ethical Committee of Kobe Medical Center. The patient gave her written informed consent to publish her case.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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