International Journal of Women's Health

Dovepress
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CASE REPORT

An Adult Massive Retroperitoneal Mature Teratoma: A Case Report and Literature Review

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Background: Teratomas usually occur in the gonads. The retroperitoneum is the least common location, which accounts for approximately 4% of primary retroperitoneal tumors and 26% of retroperitoneal teratomas are malignant. Retroperitoneal teratomas occur mainly in childhood, and with a low incidence in adults. Due to its nonspecific clinical and imaging features, the disease is difficult to distinguish from cystadenoma and other diseases, making accurate diagnosis a persistent clinical challenge.

Case Presentation: Here, we present a 65-year-old female patient who was admitted to the hospital due to intermittent right upper abdominal pain for half a month. Preoperative abdominal computed tomography imaging revealed a large mass in the left abdominal cavity, which showed mild enhancement. However, the imaging findings were non-diagnostic, and the nature of the mass could not be determined. Postoperative pathological examination revealed a retroperitoneal mature cystic teratoma. Through close follow-up, the patient achieved a favorable prognosis, and had no local recurrence or distant metastasis 6 years after surgery.

Conclusion: This case highlights the importance of considering teratoma in the differential diagnosis of retroperitoneal tumor and emphasizes the importance of complete surgical resection for the patient's prognosis, which are critical to preventing misdiagnosis as malignancy and optimizing patient outcomes.

Keywords: retroperitoneal, mature teratoma, massive, adult, prognosis

Introduction

Primary retroperitoneal cystic mature teratoma is rare, with relatively few cases reported in medical literature, with incidence of 1%-11%.[1] Retroperitoneal teratomas most commonly occur in children aged 1 to 3 years and are even rarer in adults. Additionally, information regarding the surgical management and prognosis of massive retroperitoneal mature teratomas is relatively scarce. Retroperitoneal teratomas arise from germ cells that fail to migrate to normal locations.[2] They lack characteristic clinical manifestations in the early stage of the disease and are therefore not detected until they grow large enough to produce compressive symptoms or a mass effect.[3] The diagnosis of retroperitoneal teratomas mainly relies on imaging examinations and pathological diagnosis. Computed tomography (CT) and magnetic resonance imaging (MRI) are of great significance in evaluating the anatomical relationship between the tumor and surrounding organs, whether there is distant metastasis, and clinical staging;[4–6] however, they are still difficult to distinguish from other cystic tumors, and the final diagnosis often depends on postoperative pathological examination.[7] Here, we report a case of a huge primary retroperitoneal mature cystic teratoma in a 65-year-old woman. Following close postoperative follow-up, the patient demonstrated a favorable prognosis, with no evidence of local recurrence or distant metastasis observed at 6 years after surgery. Such a rare case with unusually large tumor size and long-term follow-up data is of great significance for the diagnosis and treatment of this disease.

Case Presentation

A 65-year-old female patient was admitted to the general surgery ward of our hospital in February 2019 due to "intermittent right upper quadrant abdominal pain of half a month". The patient had some concomitant symptoms,

Received: 26 September 2025
Accepted: 26 November 2025
Published: 2 December 2025

including abdominal distension, acid regurgitation and nausea. Physical examination: a mass about 15×10cm was palpable in the left upper - middle abdomen, with clear boundary, moderate activity, and tenderness. Laboratory examination: blood, urine and stool routine, tumor markers were normal. The abdominal contrast-enhanced CT showed 154.9 × 106.5 mm huge mass existed in left peritoneal cavity, with calcification and fat. The cystic wall and soft tissues showed slight enhancement. The surrounding tissues and organs were compressed (Figure 1A–C). Her previous and family histories were unremarkable, with no prior interventions.

Following completion of appropriate preoperative preparations, exploratory laparotomy with resection of the left retroperitoneal mass was elected, primarily based on the following considerations: First of all, large tumor (often >10cm) is hard to remove through small incisions without rupture, and limited working space increases risks of organ/vessel injury. Secondly, proximity to major vessels (aorta, vena cava) and adjacent organs (kidneys, pancreas) demands direct visualization for safe dissection. Thirdly, hard components (bone, calcifications) resist minimally invasive instruments, and intact resection (critical to avoid recurrence, especially for potential malignancy) is easier via open surgery. Ultimately, the mass was completely excised following meticulous dissection. The tumor was encapsulated intact without infiltration into adjacent tissues, with the resected specimen measuring approximately 130×110×60 mm and exhibiting a wall thickness of around 3 mm. (Figure 1D). The mass was mostly cystic, and the surface was smooth. Oily, greyish-yellow, and solid contents were observed after it was carefully incised, and calcification was observed. The histopathological diagnosis was retroperitoneal mature cystic teratoma with calcification. HE staining: the mass had one or more cystic cavities, and the walls were lined by stratified squamous epithelium, which was well stratified, with a pink keratinized layer on their surface. Mature tissue such as fat can be seen in solid areas (Figure 1E–H).

Postoperatively, closely monitor the patient's vital signs, observe the incision for oozing of blood and the condition of drainage fluid, and provide analgesic, nutritional support and infection prevention. The patient was discharged on the 7th postoperative day without complications such as infection, hemorrhage, or adjacent organ injury, and recovered well postoperatively. After discharge, the patient undergoes abdominal contrast-enhanced CT scan and tumor marker detection annually for follow-up. At 6-years follow-up, the patient was asymptomatic and showed no signs of recurrence or metastasis. She continues to lead a relatively active and independent life. Compared with most reported cases, this primary retroperitoneal teratoma case has a larger tumor volume and a longer follow-up duration, enriching the information on this disease entity (Table 1).

Discussion

Teratomas usually occur in the gonads. The retroperitoneum is the least common location, which accounts for approximately 4% of primary retroperitoneal tumors and 26% of retroperitoneal teratomas are malignant.[16] Retroperitoneal teratomas occur mainly in childhood, and with only 10% to 20% occurring in adults over 30 years of age, and it is twice as common in women as in men.[9] The mature teratoma grows slowly at an average rate of 1.8 mm per year,[17] with a malignant transformation rate of 1.5%-2.0%, which is higher in women aged 45 years and over. Here, we report a case of an aged woman with a massive retroperitoneal mature teratoma. The patient underwent surgery and had no local recurrence or distant metastasis 6 years after surgery.

Retroperitoneal teratoma, a subset of germ cell tumors, is postulated to arise from aberrant migration and differentiation of primordial germ cells during embryogenesis. The precise underlying mechanism remains elusive and may be closely related to genetics and the environment. Characterized by their occult nature and deep-seated location, retroperitoneal teratomas often present with no overt early-stage symptoms and lack specific clinical manifestations. About 50% of retroperitoneal tumors have similar symptoms, such as abdominal pain, distension, nausea, or compressive symptoms, making them difficult to distinguish.[18] A large proportion of such tumors are detected incidentally via abdominal imaging examinations. Commonly, retroperitoneal teratomas are commonly located near the upper pole of the kidney, with a predominance of left-sided involvement. In the case discussed herein, the patient was an aged female with a left-sided mass adjacent to the kidney and was admitted to hospital with abdominal pain as the main symptom.

Imaging has a vital role for diagnosis of retroperitoneal teratoma. Ultrasonography serves as the first-line imaging modality for the diagnosis of teratomas, characterized by its simplicity, safety, and cost-effectiveness. Most are retroperitoneal round mixed-echo masses with clear borders, showing uneven internal echoes, strong echo clusters, bright

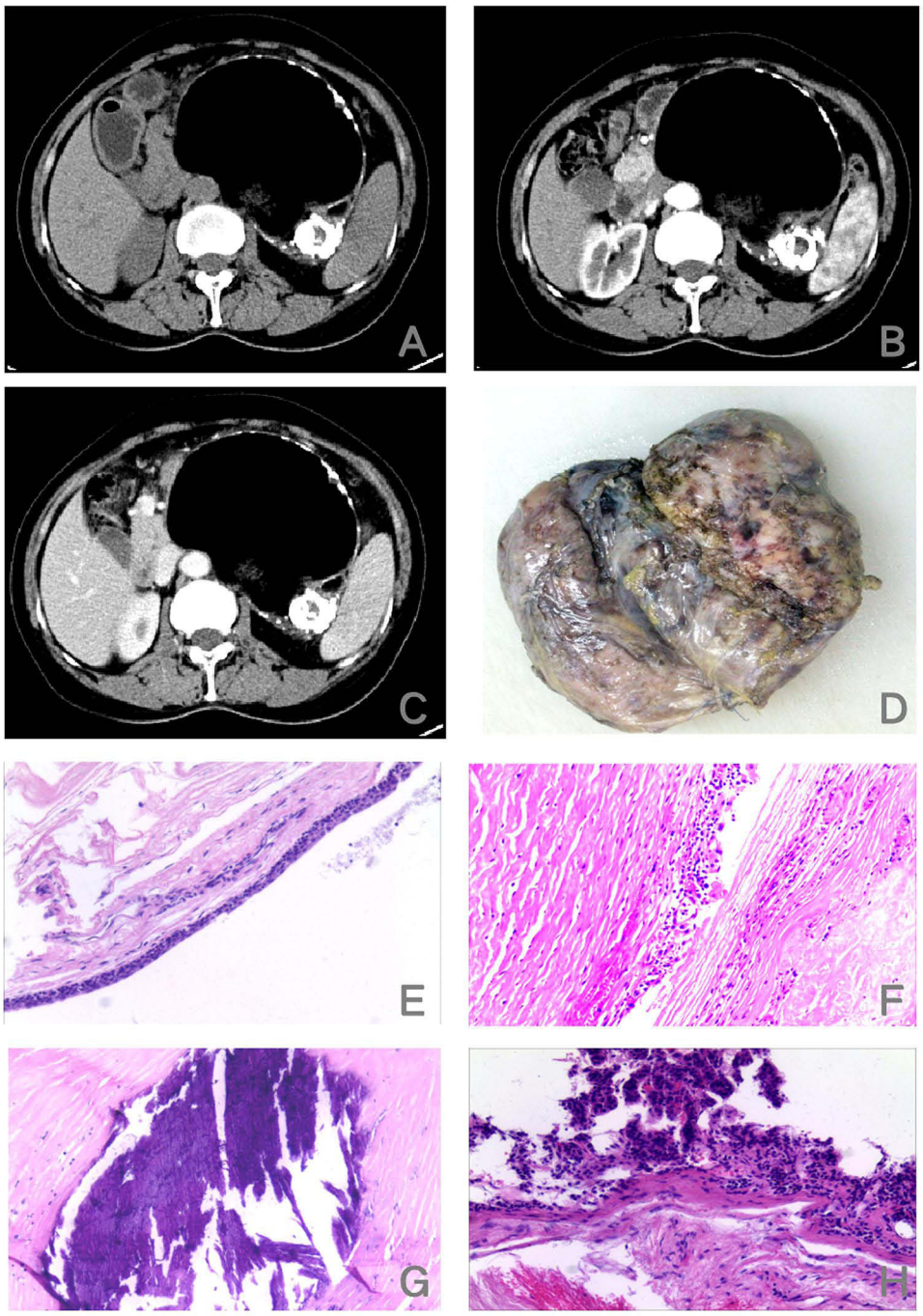

Figure 1 Calcification of the mass wall and soft tissue could be seen in unenhanced CT scan (**A**). Mild enhancement of the mass wall and soft tissue could be seen in arterial phase (**B**), and the degree of enhancement could be reduced in venous phase (**C**). A huge retroperitoneal mass with an intact tumor envelope, measuring approximately 130×110×60mm, and showing characteristic fat and calcification within the lesion (**D**). HE staining reveals mature squamous epithelium and underlying loose connective tissue, representing mature ectodermal skin components of teratoma, with mature differentiation and no atypia (**E**). Mature smooth muscle tissue noted, with regularly arranged myofibers and normal cellular morphology, representing mature mesodermal component of teratoma (**F**). Large area of deeply stained cartilage tissue; chondrocytes show mature distribution and morphology (**G**). Glandular epithelial structures identified; epithelial cells well-differentiated, representing mature endodermal-derived tissue (**H**).

spots, irregular hypoechoic areas, possible internal septa, and mostly no obvious blood flow signals. However, in the case of this patient, considering the possibility of acute abdomen, we first performed a CT scan on her. CT or MRI not only enables identification of tumor components such as osseous structures, soft-tissue density elements, adipose tissue, and sebaceous or serous fluids, but also plays a crucial role in preoperative planning by defining the tumor's precise location

https://doi.org/10.2147/IJWH.S569760

Table 1 Cases of Primary Mature Retroperitoneal Teratoma Occurring in Adult Patients

Sex	Age	Size	Site	Surgical Approach	Follow-Up
Female[8]	27	5.8×5.3cm	Left posterior	Laparoscopy	Regular follow-up without any recurrences
Male[9]	23	3.4×1.7cm	Left retroperitoneum	Laparoscopy	No report
Female[10]	40	15×13×16cm	Right retroperitoneum	Retroperitoneal lesion resection	No report
Male[1]	86	8×8.3×10cm	Above pancreas	Laparoscopic retroperitoneal tumor resection	No report
Female[2]	20	20×15.2×15cm	Left retroperitoneum	Surgical exploration	At one month postoperatively without any recurrences
Female[11]	29	11.6×11.4×14.5cm	Right suprarenal region	Modified Chevron incision	Lost
Male[12]	30	8 cm	Left retroperitoneum	Surgical exploration	At 3 months postoperatively without any recurrences
Female[13]	33	35×35 cm	Posterior to the pancreas and anterior to the left kidney	Laparotomy	At 2 weeks postoperatively without any recurrences
Female[14]	39	2×2cm	Near the abdominal aorta and behind the peritoneum of the lesser curvature of the stomach	Laparoscopy	Regular follow-up without any recurrences
Male[15]	58	10.1×8.8×7.0cm	Near the right kidney	Robot-Assisted laparoscopy	At 2 months postoperatively without any recurrences
Our case	65	13×11×6cm	Left retroperitoneum	Surgical exploration	At 6 years postoperatively without any recurrences

https://doi.org/10.2147/IJWH.S569760

and its relationship with adjacent structures. Besides, in retroperitoneal teratomas, serum tumor markers including CA19-9, CEA, and AFP may exhibit elevation. These biomarkers are also employed to monitor patients' response to therapy.

Based on their histopathological characteristics, teratomas can be categorized into mature, immature, and non-dermal subtypes. Teratomas are typically benign when they exhibit a cystic morphology with contents consisting of sebum and mature tissues. Mature teratoma often needs to be differentiated from the following diseases: Liposarcoma: fat density within the lesion, plus malignant signs (ill-defined borders, irregular shape, frequent surrounding tissue invasion, and obvious enhancement of solid components on contrast). Typically, there are atypical multinucleated adipocytes, pathological mitoses, and significant cellular pleomorphism. Cystic Lymphangioma: predominantly pure cystic lesion, with no obvious enhancement on contrast. Cyst wall lined by single-layer flat lymphatic endothelial cells, with a small amount of lymphoid tissue in the wall. Pancreatic Pseudocyst: pancreatic-derived inflammatory cystic lesion containing pancreatic juice, with mild enhancement on contrast, and no neoplastic cells.

Surgical resection holds paramount importance in both the diagnosis and management of the condition. A midline or transverse incision is employed for the majority of patients. When feasible, laparoscopic approaches represent an excellent alternative, albeit requiring proficient laparoscopic expertise. Prognosis is excellent after complete surgical excision with an overall 5-year survival rate of nearly 100%.[19] Additionally, teratomas are characterized by intrinsic resistance to both chemotherapy and radiotherapy; however, in patients with high-risk malignant features-such as positive surgical margins and unresectable disease-these modalities may be considered. In our case, as complete tumor resection was performed, the patient has a high probability of cure; moreover, no recurrence or metastasis was observed during the 6th year of follow-up after surgery.

The pathologically confirmed recurrence of ovarian mature teratoma was 10.5 per 100 person-months at 0.4 months.[20] In addition, overall prevalence of metachronous contralateral ovarian mature teratoma was 2.1%.[21] Besides, malignant transformation of mature teratoma is a rare complication (1–3% of cases).[22] Approximately 80% are squamous cell carcinoma; other malignancies include adenocarcinoma, sarcoma, etc.[23,24] A study has reported that enhancement of soft tissue components may be a risk factor for malignant transformation and poor prognosis.[25] Disease-free survival depends on complete tumor resection. In cases of incomplete tumor resection, up to 6% of mature teratomas undergo malignant transformation to sarcoma or cancer.[26] This study also has limitations. This study is a single case with potential selection bias and a lack of molecular-level research, so some conclusions may not be universally representative. Thus, further clinical and molecular mechanism studies with expanded sample sizes are still needed.

Conclusion

In conclusion, primary retroperitoneal teratoma constitutes a rare disease, particularly among adult populations. In the majority of cases, it remains asymptomatic. Preoperative imaging is crucial not only for diagnostic confirmation but also for formulating preoperative strategies. Surgical resection represents the mainstay of management for mature retroperitoneal teratomas. Despite their benign biological behavior, mature retroperitoneal teratomas necessitate close follow-up due to their inherent risk of malignant transformation, albeit low. It is generally recommended to review imaging and tumor markers annually after surgery. If the soft tissue components show obvious enhancement preoperatively, closer follow-up should be paid more attention to.

Trial Registration

Not applicable, because this article does not contain any clinical trials.

Data Sharing Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Ethics Approval and Consent to Participate

The present study was performed in accordance with the principles of the Declaration of Helsinki. Ethical exemption was received for the Medical Ethics Committee of the Fifth Clinical Medical College of Henan University of Traditional Chinese Medicine. Institutional approval was obtained for publication of the case details from the Medical Ethics Committee of the Fifth Clinical Medical College of Henan University of Traditional Chinese Medicine.

Patient Consent for Publication

The patient involved in the present study was subjected to standard clinical practice and provided written informed consent for the publication of medical data and images.

Acknowledgments

The authors thank the patient and families and caregivers; And we acknowledge the staff of the study for their important contributions.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Disclosure

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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