International Journal of Research and Review
[Indexed, Peer-Reviewed Journal]
Volume 13; Issue: 7; July 2026
Website: www.ijrrjournal.com
Case Report *E-ISSN: 2349-9788; P-ISSN: 2454-2237*

Retroperitoneal Mucinous Cystadenoma of Ovarian origin: A Case Report

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DOI: https://doi.org/10.52403/ijrr.20260748

ABSTRACT

Retroperitoneal mucinous cystadenoma of ovarian origin is an extremely rare tumour, with fewer than 100 cases reported since 1965. Its histogenesis remains unclear, as no epithelial tissue is normally present in the retroperitoneum. We report the case of a 65-year-old woman who presented with a seven-day history of insidious, progressive dull aching pain in the right upper abdomen. Examination revealed right flank fullness with a palpable, soft, non-ballotable, retroperitoneal mass measuring approximately 8x10 cm in the right lumbar region. Ultrasonography and contrast-enhanced CT of the abdomen and pelvis showed a unilocular cystic lesion of 300 cc with internal septations, adjacent to the ascending colon and extending up to the upper pole of the right kidney, with foci of wall calcification. She underwent exploratory laparotomy with cyst excision. Histopathological examination showed a cyst wall lined by mucinous tall columnar epithelium arranged in papillary and micropapillary architecture, with a high nucleocytoplasmic ratio and hyperchromatic, pleomorphic nuclei, consistent with a borderline mucinous cystadenoma of ovarian origin. The patient had an uneventful recovery. This case highlights that although retroperitoneal mucinous cystadenoma is a diagnostic challenge due to its non-specific presentation, early diagnosis and complete surgical excision followed by regular imaging follow-up yield a favourable outcome.

Keywords: Retroperitoneal mucinous cystadenoma, ovarian origin, borderline tumour, exploratory laparotomy, case report.

INTRODUCTION

Retroperitoneal mucinous cystadenoma is a rare tumour occurring in the retroperitoneum, with fewer than 100 cases reported in the literature since 1965. Its histogenesis remains unclear, since no epithelial tissue is normally present in the retroperitoneum to give rise to such a lesion. Proposed origins include ectopic ovarian tissue seeded in the retroperitoneum, invagination of multipotential mesothelium with subsequent mucinous metaplasia, or, rarely, derivation from the appendix or pancreas. The primary presenting symptom is usually non-specific abdominal pain related to the mass effect of the lesion, although the tumour is often asymptomatic

in its early stages, making early diagnosis challenging. We report a case of a large retroperitoneal mucinous cystadenoma of ovarian origin in a 65-year-old woman, along with a review of the relevant literature, to highlight the clinical presentation, diagnostic work-up, and management of this rare entity.

CASE REPORT

A 65-year-old woman presented with pain in the upper abdomen of seven days duration, insidious in onset and progressive dull aching in nature, which had increased in intensity over the preceding two days. On examination, the abdomen was obese with an inverted umbilicus, and right flank fullness was noted. A palpable mass measuring 8x10 cm was noted in the right lumbar region, retroperitoneal in location, soft in consistency, non-ballotable, and dull to percussion, with normal bowel sounds. Ultrasonography of the abdomen and pelvis and contrast-enhanced CT of the abdomen and pelvis were performed, which revealed a unilocular cystic lesion of 300 cc with internal septations, adjacent to the ascending colon and extending up to the upper pole of the right kidney, with foci of wall calcification. The patient underwent exploratory laparotomy with cyst excision. On histopathological examination, the cyst wall was lined by mucinous-type tall columnar epithelium arranged in papillary and micropapillary architecture, with a high nucleocytoplasmic ratio and hyperchromatic, pleomorphic nuclei; the lumen contained proteinaceous fluid. These features were suggestive of a borderline mucinous cystadenoma of ovarian origin.

Representative investigation, intraoperative, and histopathological images

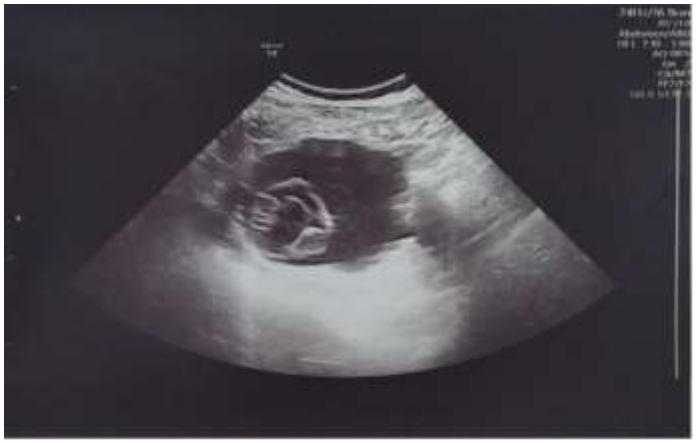

Figure 1: Ultrasonography of the abdomen and pelvis showing the retroperitoneal cystic lesion.

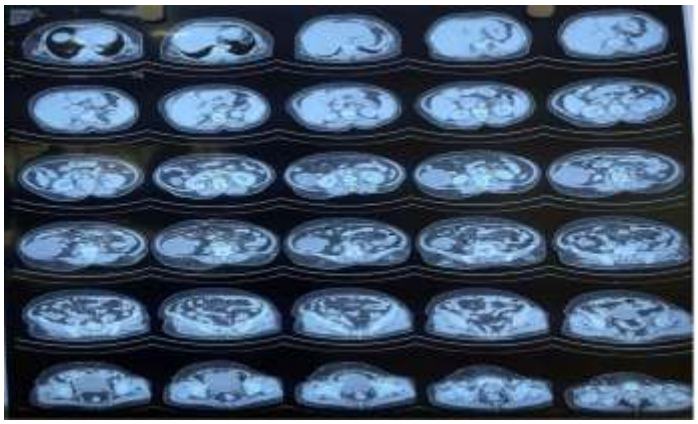

Figure 2: Contrast-enhanced CT of the abdomen and pelvis showing a unilocular retroperitoneal cystic lesion adjacent to the ascending colon.

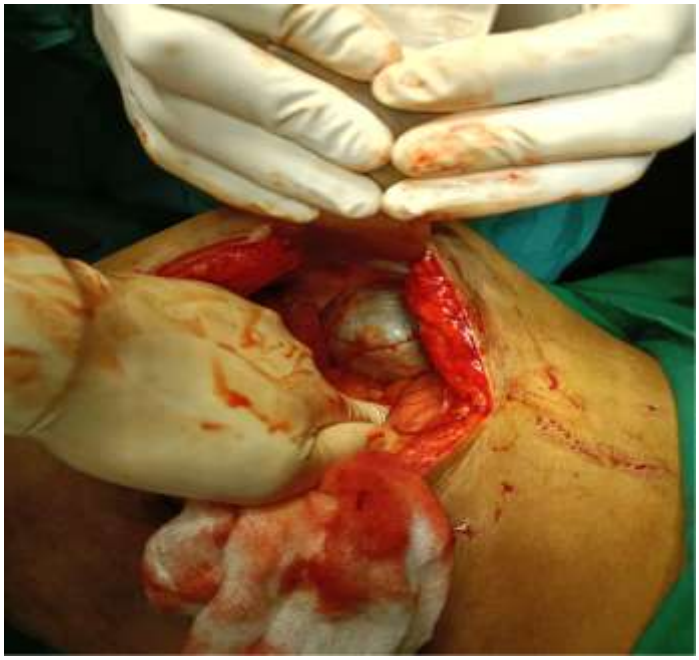

Figure 3: Intraoperative photograph showing the retroperitoneal cystic mass at exploratory laparotomy.

Figure 4: Gross photograph of the excised unilocular cyst with internal septations.

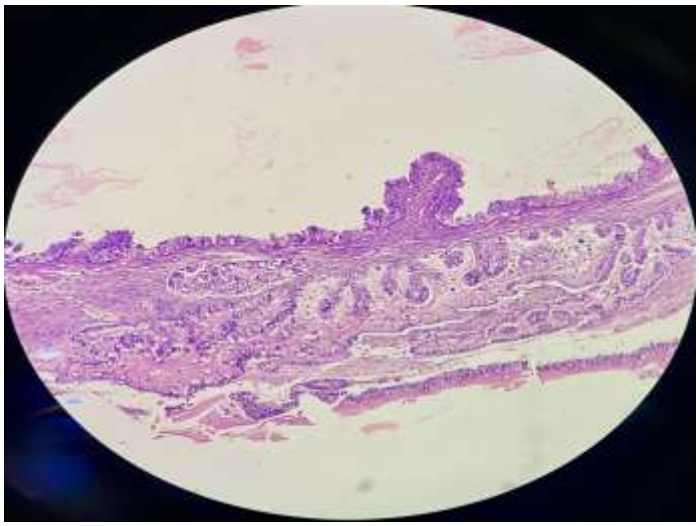

Figure 5: Photomicrograph (H&E) showing the cyst wall lined by mucinous tall columnar epithelium in papillary and micropapillary architecture, with hyperchromatic, pleomorphic nuclei, consistent with a borderline mucinous cystadenoma.

DISCUSSION

Retroperitoneal mucinous cystadenoma accounts for approximately 0.2% of all neoplasms and is a rare occurrence, as epithelial tissue is not normally present in the retroperitoneum. It affects women overwhelmingly more often than men, typically presenting in the fourth to seventh decades of life, and only a handful of cases have been described in male patients; those rare male cases are of particular interest because they argue against ectopic ovarian tissue being the sole explanation for the tumour's origin.

Several theories have been proposed to explain the histogenesis of this rare tumour. The first, and historically the most widely cited, suggests seeding of ectopic or heterotopic ovarian tissue within the retroperitoneum during embryogenesis, an explanation supported by the ovarian-type stroma and hormone-receptor positivity occasionally seen on immunohistochemistry. A second, increasingly favoured hypothesis proposes that these tumours arise from invagination of the coelomic or multipotential mesothelium lining the peritoneal and retroperitoneal cavities, which retains the capacity for Mullerian-type differentiation and subsequently undergoes mucinous metaplasia to form a cyst; this theory better accounts for the rare cases reported in men and in women with normal, unremarkable ovaries. A third, much less common pathway is derivation from misplaced or duplicated primitive gut endoderm, explaining the occasional tumour with an appendiceal or pancreatic-type lining.

Histologically, retroperitoneal mucinous cystic tumours mirror the spectrum seen in their ovarian counterparts and are classified into three categories of broadly increasing severity: benign mucinous cystadenoma, in which the cyst is lined by a single layer of bland, mucin-secreting columnar epithelium without atypia; borderline (low malignant potential) mucinous cystic tumour, characterised by epithelial stratification, papillary or micropapillary architecture, and nuclear atypia without stromal invasion, as seen in the present case; and mucinous cystadenocarcinoma, in which frank stromal invasion is present. This distinction has direct prognostic and follow-up implications, since borderline and malignant tumours carry a recognised risk of local recurrence even after apparently complete excision.

Clinically, these tumours are often silent for a long period and are discovered incidentally or only once they have grown large enough to produce mass effect, as reflected in the non-specific abdominal pain, fullness, or a palpable mass that most patients, including ours, eventually present with. This indolent course, combined with the rarity of the tumour, means it is seldom considered in the initial differential diagnosis of a retroperitoneal or abdominal mass, which more commonly includes conditions such as retroperitoneal lymphangioma, mesothelial or peritoneal inclusion cysts, cystic teratoma, a duplex or cystic renal anomaly, pancreatic pseudocyst, and, in women, an adnexal or paraovarian cyst. Cross-sectional imaging with ultrasonography, contrast-enhanced CT, or MRI reliably delineates the size, location, and septate or calcified nature of the lesion, as in this patient, but cannot distinguish a mucinous cystadenoma from these other cystic entities or reliably predict its histological grade. Serum tumour markers such as CA-125, CA19-9, and CEA are occasionally elevated but lack specificity and are of limited diagnostic value. Consequently, a definitive diagnosis rests on histopathological examination of the surgically excised specimen, as occurred in our patient.

Complete surgical excision, whether by open laparotomy or, increasingly, a laparoscopic or robot-assisted approach in selected cases, remains the mainstay of treatment and serves the dual purpose of confirming the diagnosis and preventing recurrence. Where imaging or intraoperative findings raise suspicion of a borderline or malignant lesion, care should be taken to

excise the cyst intact to avoid spillage, and some authors advocate intraoperative frozen-section assessment to guide the extent of resection. The prognosis for benign and borderline tumours is generally favourable after complete excision, whereas frank mucinous cystadenocarcinoma carries a more guarded prognosis and may warrant multidisciplinary discussion regarding adjuvant therapy. Given the recognised, if uncommon, risk of recurrence, particularly for borderline tumours, long-term surveillance with periodic imaging is recommended, as was planned for the patient in this report.

CONCLUSION

Retroperitoneal mucinous cystadenoma of ovarian origin is a rare entity that should be considered in the differential diagnosis of a retroperitoneal mass, even though it typically presents with non-specific symptoms. Early diagnosis and appropriate management, despite the non-specific presenting symptoms, yielded a favourable outcome in this case, where the mainstay of treatment was exploratory laparotomy with cyst excision, followed by histopathological examination and regular follow-up with imaging in view of the risk of recurrence.

Declaration by Authors

Acknowledgement: None
Source of Funding: None
Conflict of Interest: No conflicts of interest declared.

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How to cite this article: Nikhilesh, Lakshminarayan G, Ramesh M Tambat, Mahesh BN. Retroperitoneal mucinous cystadenoma of ovarian origin: a case report. *International Journal of Research and Review*. 2026; 13(7): 471-475. DOI: *10.52403/ijrr.20260748*

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