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Retroperitoneal Müllerian Cyst Found Incidentally During Total Laparoscopic Hysterectomy: Is There a Role for MRI in Pre-Operative Evaluation?

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ABSTRACT

Retroperitoneal cysts of Müllerian origin are rare and often discovered incidentally during surgical procedures or imaging studies. We present a case of a retroperitoneal cyst found during a total laparoscopic hysterectomy performed for myoma removal. A 40-year-old female presented with symptomatic myomas and underwent pre-operative evaluation, including pelvic ultrasound, which revealed no abnormalities apart from the myomas. A cyst was incidentally discovered in the retroperitoneal space during a total laparoscopic hysterectomy procedure. The cyst was excised and sent for histological examination, which confirmed its Müllerian origin. Retroperitoneal cysts of Müllerian origin are rare findings during TLH. This case report highlights the importance of thorough pre-operative evaluation and imaging studies and the significance of intraoperative awareness in identifying incidental findings. Further research is warranted to determine the optimal diagnostic imaging strategies for such cysts.

INTRODUCTION

Retroperitoneal cysts of Müllerian origin are rare findings, often discovered incidentally during surgical procedures or imaging studies. These cysts arise from remnants of Müllerian duct remnants and can occur in various locations within the retroperitoneal space (Santana Gonzalez *et al.*, 2021). Uterine fibroids, also known as leiomyomas, are benign tumors originating from the smooth muscle cells of the uterus. Although up to 70% of cases may be asymptomatic, they are sometimes associated with heavy menstrual bleeding, intermenstrual symptoms, pelvic pain, and pressure symptoms, necessitating treatment by medical therapies, interventional radiology, and surgical procedures (Giuliani *et al.*, 2020). Total laparoscopic hysterectomy (TLH) is a minimally invasive surgical procedure frequently performed to manage symptomatic fibroids.

While uterine fibroids are a common indication for surgical intervention, the discovery of an incidental retroperitoneal cyst during a TLH in a patient with uterine fibroids is a unique occurrence. Incidental findings during TLH can pose diagnostic challenges and require careful consideration. Although rare, retroperitoneal cysts of Müllerian origin have been reported in the literature (Yang *et al.*, 2004). These cysts can present as an incidental discovery during surgery, often lacking pre-operative imaging clues (Rivas *et al.*, 2022). Understanding such incidental findings' characteristics, management, and implications is crucial for appropriate patient care.

This case report presents the incidental discovery of a retroperitoneal cyst of Müllerian origin during TLH performed in a patient with uterine fibroids. By highlighting this unique case, we aim to contribute to

the existing literature and raise awareness about the importance of thorough pre-operative imaging and management of incidental retroperitoneal cyst findings during TLH.

LITERATURE REVIEW

Müllerian cysts are uncommon findings that often present as incidental discoveries during imaging studies or surgical procedures. These cysts arise from remnants of the Müllerian duct system (Giuliani *et al.*, 2020; Wilson & Bordoni, 2024). The Müllerian ducts, also known as paramesonephric ducts, are paired ducts that originate from the intermediate mesoderm in the embryo (Wilson & Bordoni, 2024). During early fetal development, these ducts emerge alongside the urogenital ridge and run laterally. They terminate at the Müllerian eminence within the primitive urogenital sinus. The Müllerian ducts play a crucial role in the development of the female reproductive system (Sugi *et al.*, 2021). These paired structures emerge during early fetal life and contribute to the formation of several female reproductive organs, including the fallopian tubes, uterus, cervix, and the upper two-thirds of the vagina. In males, the Müllerian ducts typically regress due to the influence of anti-Müllerian hormone (AMH) secreted by the developing testes (Wilson & Bordoni, 2024). However, remnants of these ducts such as Müllerian cysts may persist in both sexes, leading to various clinical conditions. Müllerian cysts result from focal incomplete regression of the Müllerian ducts (Sugi *et al.*, 2021). Areas where complete regression did not occur become localized cystic formations. These cysts can occur anywhere along the path of Müllerian duct regression. Retroperitoneal Müllerian cysts are rare,

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with limited prevalence data (Yang *et al.*, 2004). They are typically discovered incidentally during imaging studies or surgery. The exact incidence remains uncertain due to their asymptomatic nature. When symptomatic, patients may experience Abdominal pain or discomfort due to cyst enlargement or pressure symptoms related to adjacent structures such as the urinary tract (Renzulli and Candinas, 2009). Imaging Modalities that can be used in the detection of Mullerian cysts include Ultrasound, CT and MRI (Stefanopol *et al.*, 2022; Yacoub *et al.*, 2021; Yang *et al.*, 2004). Ultrasound can visualize Müllerian cysts as hypoechoic lesions. However, ultrasound may not always provide detailed anatomical information compared to MRI. Ultrasound-guided aspiration can be performed for diagnostic purposes (Yacoub *et al.*, 2021). On CT Scan Mullerian cysts may appear as well-defined hypoattenuating cystic masses. MRI however provides superior soft tissue characterization and multiplanar imaging capabilities for accurate pre-operative assessment. MRI characteristic features for Mullerian cysts include well-defined, unilocular cystic lesions, hypointense signal on T1-weighted images and hyperintense signal on T2-weighted images (Renzulli & Candinas, 2009). Pelvic MRI remains the imaging modality of choice for diagnosing Müllerian cysts, allowing accurate assessment and guiding clinical management (Yacoub *et al.*, 2021). Treatment options for Müllerian cysts include observation and monitoring for asymptomatic cysts with regular follow-up imaging to exclude any other conditions (Johan *et al.*, 2020). Surgical excision is indicated for symptomatic or large cysts. Pre-operative MRI guides surgical planning and should be carried out whenever possible (Johan *et al.*, 2020). Complete cyst excision is usually the goal intraoperatively (Yacoub *et al.*, 2021). Although rare, complications of Müllerian cysts may include infection, hemorrhage, mass effect and transition to malignancy (Zhu *et al.*, 2023).

METHODOLOGY

Case Presentation

A 40-year-old African female, Para 1+0, presented to PCEA Tumutumu Hospital in Nyeri, Kenya, with a ten-year history of intermenstrual bleeding and heavy

and prolonged periods associated with dysmenorrhea and intermittent lower abdominal pain radiating to the back. In 2013, she was diagnosed with uterine fibroids through a pelvic ultrasound scan and had been managed conservatively for her symptoms. However, she recently visited the gynaecology outpatient clinic with persistent lower abdominal pain and irregular periods. A repeat ultrasound scan revealed two intramural hypoechoic masses measuring 96.74x95.75x115.52 mm and 26.44x17.51x36.71 mm, respectively, along with a pedunculated fibroid in the left adnexal region. The decision was made to proceed with TLH.

Laboratory Investigation

The patient was admitted on 17th May 2023, and routine pre-operative tests were conducted, including full blood count, urea, electrolytes, creatinine, random blood sugar, and group crossmatch. The only abnormal findings were a microcytic anemia with Hb of 7.5 g/dl (Ref 11.5-16.5 g/dl) and MCV of 67 fl (Ref 75-100 fl), due to chronic blood loss. Serum tumor marker tests were not carried out.

RESULTS

The TLH procedure was performed under general anesthesia by consultant obstetrician/gynecologist and laparoscopic surgeon Dr Njagi. Intraoperatively, the leiomyomas were noted to have fatty degeneration. After the removal of the uterus, a retroperitoneal cyst of size 40mm by 5mm was incidentally noted (Figure 1A). Macroscopic examination revealed a white encapsulated lesion adherent to the left lower abdominal wall. The cystic lesion was filled with brownish fluid (Figure 1B). The lesion was surgically excised as a whole and sent for histopathological analysis. The whole surgical procedure took a total of 2 hours and 50 minutes with a total intraoperative blood loss of about 50cc.

The post-operative stay was uneventful, with the patient experiencing mild pain. Vitals remained stable, and a physical examination indicated normal bowel sounds. Post-operative medication consisted of Paracetamol 1g IV TDS for 3 days and Ceftriaxone 1g IV twice daily (BD) for 3 days. Upon discharge, the patient was

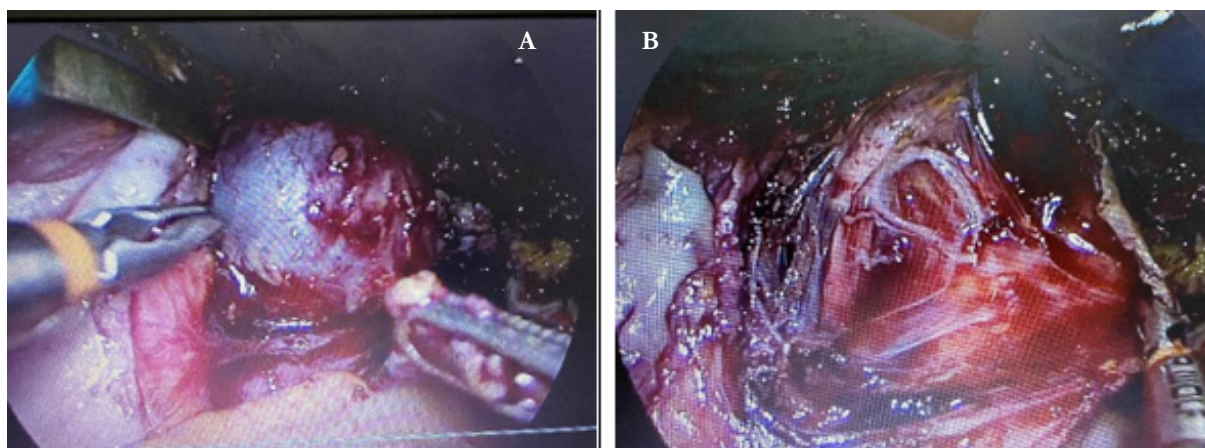


Figure 1: A retroperitoneal cyst of size 40mm by 5mm was incidentally noted (A). The cystic lesion was filled with brownish fluid

Patient: [REDACTED]	Age: 40 years
Doctors Ref: [REDACTED]	Sex: Female
IP/OP. Num.: [REDACTED]	Facility: PCEA Tumutumu Hospital

Specimen: RETROPERITONEAL CYST	Collection Date: 18/05/2023
Lab. No: UH/1540/2023	Received Date: 22/5/2023
	Final Report Date: 12/06/2023

Requested: HISTOPATHOLOGY

Diagnosis:
 Retroperitoneal Cyst, Excision:
 - Features Favor Mullerian Type Cyst, see note

NOTE
 Immunohistochemistry report to follow.

Clinical History:
 Para 1+0 with multiple Leiomyoma noted to have fatty degeneration. Retroperitoneal cyst noted incidentally and excised as whole.

Macroscopy:
 Received in formalin, labeled with "Patients name and retroperitoneal cyst," and is cyst biopsy measuring 40mm by 5mm. Cut section multilocular wall and clear fluid released. All are processed in six cassettes.

Microscopy:
 Sections show a thick walled multilocular cyst lined by benign ciliated epithelium. Adjacent reactive nodes noted attached to the external wall.


Reporting Doctor: Dr. Obiero Okoth Signature:  Date: 13/06/2023

Figure 2: Histopathological analysis of the excised retroperitoneal cyst showed a thick-walled multilocular cyst lined by benign ciliated epithelium

prescribed oral Metronidazole 400mg TDS, Cefuroxime 500mg BD, Aclosara SP (Aceclofenac/Paracetamol and Serratiopeptidase 100mg/325mg/15mg, Sara Remedies) PO TDS for 5 days, and IFAS (Iron and Folic Acid) 2 tabs PO BD for 1 month.

Histopathological analysis of the excised retroperitoneal cyst showed a thick-walled multilocular cyst lined by benign ciliated epithelium (Figure 2). Adjacent reactive nodes were noted attached to the external wall. The features were consistent with a cyst of Müllerian origin, further supporting the incidental finding during TLH. The cyst wall consisted of a lining epithelium resembling the Müllerian epithelium, surrounded by fibrous tissue. No atypical cells or malignancy was identified. During a follow-up visit in the gynecology clinic scheduled two weeks post-surgery the patient had no complaints and the physical exam was unremarkable. The surgical wound was healing well. We explained the histology results to the patient.

DISCUSSION

The incidental discovery of a retroperitoneal cyst of Müllerian origin during a TLH in a patient with uterine fibroids presents an intriguing case and raises several important considerations. Retroperitoneal cysts are often misdiagnosed pre-operatively (Johan *et al.*, 2020). In our case, the patient presented with a history of metrorrhagia, dysmenorrhea, and lower abdominal pain radiating to the back. These symptoms, along with the presence of uterine fibroids on ultrasound imaging, prompted further investigation and the decision to proceed with TLH. The intraoperative discovery of a retroperitoneal

cyst, unrelated to the uterine fibroids, highlights the importance of appropriate pre-operative imaging and evaluation to detect such lesions early. Additionally, thorough exploration during surgical interventions is important, even in cases where the primary pathology is well-defined.

Pre-operative ultrasound imaging may not reliably detect retroperitoneal cysts of Müllerian origin, as seen in our case where the cyst was not visualized on the pre-operative ultrasound scan (Yacoub *et al.*, 2021). Magnetic resonance imaging (MRI) provides a sophisticated method of distinguishing mullerian anomalies from one another, characterizing the degree of defect severity, and evaluating for concomitant urogenital anomalies non-invasively and without radiation exposure, with superior soft-tissue delineation and availability of advanced functional sequences (Udayakumar *et al.*, 2023). MRI is also a commonly used tool for evaluation of pelvic malignancies. Mullerian cysts may occasionally have malignant transformations that would only be picked by careful imaging using MRI, as has been shown by occasional case reports (Xiao *et al.*, 2022; Zhu *et al.*, 2023). Further studies may be done to determine the utility of pre-operative MRI imaging in TLH to facilitate the identification of pathology.

Histopathological analysis confirmed the cyst to be of Müllerian origin, characterized by a lining epithelium resembling the Müllerian epithelium, surrounded by fibrous tissue. The absence of atypical cells or malignancy supported the benign nature of the cyst. It is essential to differentiate retroperitoneal cysts of Müllerian origin from other retroperitoneal cystic lesions, such as

lymphatic cysts or cystic teratomas, to guide appropriate management and prevent unnecessary interventions. The retroperitoneal cysts of Müllerian origin are believed to be embryological remnants of the Müllerian duct system (Santana Gonzalez *et al.*, 2021). These remnants can give rise to various cystic structures in the retroperitoneal space, presenting a diagnostic challenge due to their rarity and diverse clinical presentations.

Managing retroperitoneal cysts of Müllerian origin largely depends on the clinical presentation, size, and symptoms associated with the cyst. Asymptomatic cysts can be managed conservatively with regular monitoring. However, in cases where the cyst is symptomatic, surgical excision is warranted (Renzulli and Candinas, 2009). The incidental discovery of retroperitoneal cysts during TLH presents a unique opportunity for their complete removal, minimizing the risk of future complications or recurrence (Stefanopol *et al.*, 2022). The importance of appropriate post-operative care, including pain management and prophylactic antibiotics, cannot be overstated to ensure optimal recovery.

This case report emphasizes the need for thorough pre-operative evaluation and intraoperative vigilance during TLH. The incidental discovery of retroperitoneal cysts of Müllerian origin during TLH highlights the importance of considering these rare entities in the differential diagnosis of retroperitoneal cystic lesions. Further research is warranted to explore the prevalence, clinical characteristics, and optimal management strategies for retroperitoneal cysts of Müllerian origin, particularly when encountered during TLH. Enhanced awareness among clinicians and gynaecological surgeons regarding possible incidental findings during TLH can aid in early detection, appropriate management, and improved patient outcomes.

CONCLUSION

The incidental discovery of a retroperitoneal cyst of Müllerian origin during TLH in a patient with uterine fibroids highlights the diagnostic challenge and importance of thorough intraoperative exploration. This case underscores the need for vigilance and consideration of rare entities in the differential diagnosis of retroperitoneal cystic lesions encountered during TLH. Further research is necessary to advance our understanding of retroperitoneal cysts of Müllerian origin and optimize pre-operative detection using MRI.

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