

# Case Report: Unusual Presentation of Spontaneous Rupture of Ovarian Teratoma

Zhong Ning Neoh<sup>1</sup>, Ma Saung Oo<sup>2</sup>, Alik R Zakaria<sup>3</sup>, Anna L Roslani<sup>4</sup>

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## ABSTRACT

**Background:** Mature cystic teratoma (MCT) comprises 95% of ovarian germ cell tumors with 10–25% incidence in adults. The sequelae include torsion (15%), malignant transformation (1–2%), infection (1%), and rupture (0.3–2%). Spontaneous ruptures of MCT with spillage of sebaceous material into the abdominal cavity are rare due to its thick capsules. Ruptured ovarian teratoma can lead to chemical peritonitis, which is uncommon with diverse presentations. It presents with contralateral abdominal pain due to spillage of its contents into the opposite side of the abdominal cavity following tumor rupture.

**Case description:** This is an unusual presentation of twisted and ruptured ovarian teratoma. A 26-year-old para 1 presented with persistent left lower abdominal pain that radiated to right and sought treatment from various clinics for a week duration. She was referred to a tertiary hospital for ruling out acute appendicitis for right-sided lower abdominal pain where the appendix is located. At first, she presented with sudden onset of moderate-to-severe left iliac fossa (LIF) pain during a bowel opening 5 days ago. Subsequently, the pain radiated to the right iliac fossa (RIF) and worsened on the day of admission. She also had multiple episodes of vomiting and vague suprapubic mass for 1 week. Abdominal examination revealed a palpable cystic suprapubic mass which is about 10 × 8 cm with persistent RIF pain. Ultrasonography showed a solid cystic mass at the LIF measured about 11 × 6 cm and the right ovary was visualized, with normal size uterus and thin endometrial lining. There was significant amount of free fluid, with normal kidneys seen. Ultrasound examination provided an inconclusive diagnosis and an emergency laparotomy was performed for suspicion of a ruptured ovarian cyst with a differential diagnosis of a perforated appendix. There was a twisted and ruptured left ovarian teratoma with a gangrenous base found intraoperatively which contains sebum, hair, and slough at the anterior part of the uterus and thicken omentum. Left-sided salpingo-oophorectomy and omentectomy were done. The peritoneal cavity was washed with copious amounts of warm saline.

**Conclusion:** A ruptured dermoid cyst is one of the gynecological emergencies but is often an overlooked diagnosis. Mature ovarian teratoma consists of well-differentiated germ cell layers derivatives developing as hair, muscle, teeth, or bone. The size of dermoid cysts and torsion with infarction can contribute to their rupture. Correct diagnosis and timely surgical intervention are essential for preventing chemical peritonitis, long-term sequelae, and conserving the reproductive function.

**Keywords:** Case report, Chemical peritonitis, Mature cystic teratoma, Spontaneous rupture.

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## BACKGROUND

Mature cystic teratomas (MCT) of the ovary or dermoid cysts are benign ovarian lesions that are frequently seen in gynecological practice. These lesions frequently cause diagnostic misery and therapeutic challenges. Approximately, 10% of women have various approaches of surgical intervention for an ovarian pathology in their lifetime. Most are benign in premenopausal women and the risk of malignant increases from 1:1,000 to 3:1000 at 50 years old.<sup>1</sup> Twisted or ruptured ovarian cysts can be presented with various clinical presentations and misdiagnosed as perforated appendicitis or other inflammatory conditions of the gastrointestinal tract and urinary like in this case. Despite ovarian tumors having specific tumor markers, there are dilemmas in deciding on the best surgical intervention due to unreliability in the preoperative diagnosis and coinciding clinical presentation, a lack of consensus, and variations in surgical practices.

Mature cystic teratoma may be asymptomatic or present as gynecological emergencies with acute abdomen due to complications, such as chemical peritonitis in 20% of cases, as in our case. The primary sequelae include cyst torsion (15%), malignant transformation (1–2%), infection (1%), and rupture (0.3–2%).<sup>1</sup> Because of the cyst's thick capsule, spontaneous MCT rupture

<sup>1</sup>Department of Obstetrics and Gynaecology, Hospital Pakar Sultanah Fatimah Muar, Muar, Johor Darul Takzim, Malaysia

<sup>2</sup>Department of Obstetrics and Gynaecology, Universiti Sultan Zainal Abidin, Temangan, Kelantan Darul Naim, Malaysia

<sup>3</sup>Department of Obstetrics and Gynaecology, Universiti Sultan Zainal Abidin, Kuala Terengganu, Terengganu, Malaysia

<sup>4</sup>Department of Obstetrics and Gynaecology, KMI Kuantan Medical Centre, Kuantan, Pahang, Malaysia

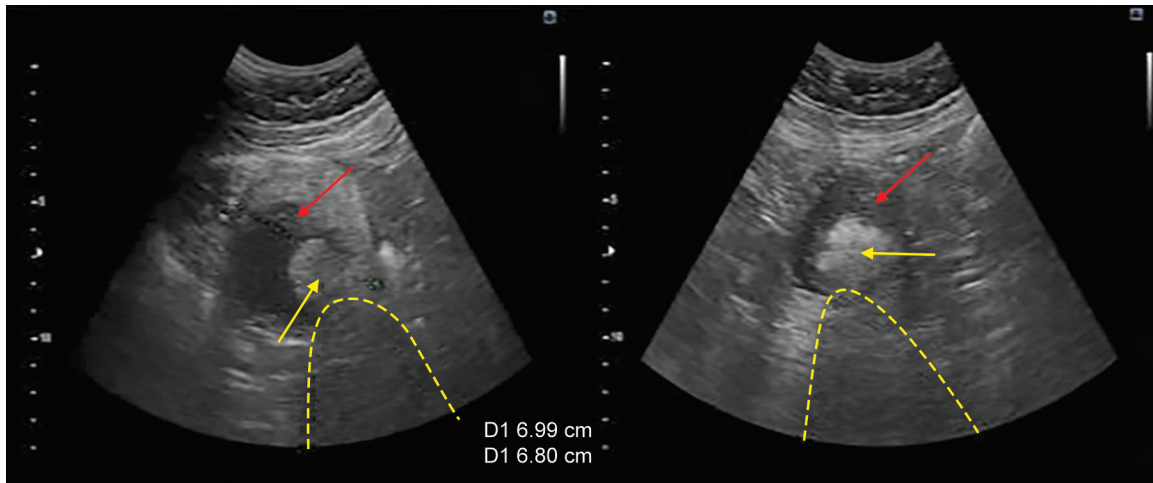
**Corresponding Author:** Zhong Ning Neoh, Department of Obstetrics and Gynaecology, Hospital Pakar Sultanah Fatimah Muar, Muar, Johor Darul Takzim, Malaysia, Phone: +0143382462, e-mail: jonineoh@gmail.com

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is highly uncommon and only occurs in 0.3–0.7% of instances, although they might cause chemical peritonitis, as in our case<sup>1</sup> (unusual presentation).



**Fig. 1:** Horizontal and transverse view of dermoid cyst showing Rokitansky nodule (yellow arrows) with acoustic shadowing—“tip of the iceberg” sign (outlined). This nodule is seen as an echogenic mass due to the presence of sebaceous material, hair follicles, and/or other solid elements with distal acoustic shadowing

Chemical peritonitis has varied presentation, with less than 1% occurrence. Here, we report a case of ruptured left ovarian MCT with contralateral severe abdominal pain due to chemical peritonitis.

## CASE DESCRIPTION

A 26-year-old para 1 lady presented with right lower abdominal pain refractory to medical treatment and was referred to tertiary hospital to rule out acute appendicitis. She presented with sudden continuous sharp moderate-to-severe left iliac fossa (LIF) pain during bowel opening 5 days ago which improved with intravenous analgesia. Subsequently, the sharp pain radiated to the right iliac fossa (RIF) which is associated with nausea and vomiting. On the day of admission, there was a sudden worsening of the RIF with a pain score of 10/10. She looked ill and presented multiple episodes of nausea, retching and minimal amount of vomiting with worsening RIF pain. She had constipation which was resolved with laxatives and enema. Otherwise, the patient was afebrile with no urinary frequency, early satiety, or leg edema. Her menses were regular with no abnormal vaginal discharge. Abdominal examination revealed a tender, smooth-surfaced cystic suprapubic mass more toward the left about 8 × 10 cm was palpable with rebound tenderness but no guarding. Normal bowel sound heard. Other systemic examinations showed no abnormalities.

Transabdominal and transvaginal ultrasound showed a solid cystic LIF mass up to left lumbar 11 × 6 cm, right ovary visualized, left ovary not seen, uterus 6 × 3.8 cm with thin endometrial thickness, a significant amount of free fluid present, with normal kidneys. Ultrasound findings are shown in [Figure 1](#).

There was mild leukocytosis ( $13.8 \times 10^9/L$ ) with neutrophilia ( $12.07 \times 10^9/L$ ) and marginal low microcytic anemia (Hb: 115 g/L). Other tumor markers were normal except for high CA-125 (42 U/mL). Serum amylase and blood urea serum electrolyte creatinine (BUSEC) showed normal results. UFEME was negative for leukocyte and nitrate, urine pregnancy test (UPT) was negative, COVID-19 Ag rapid test was negative and group screen and hold (GSH) was done.

With the diagnosis of a possible ruptured or twisted left ovarian tumor, the patient underwent an emergency laparotomy for “left cystectomy kept in view unilateral salpingo-oophorectomy” on the following day. Intraoperatively, there was a twisted and ruptured

10 × 7 cm left ovarian teratoma with a gangrenous base found, revealing sebum and hair; slough was present at the anterior part of the uterus and omentum and thickened omentum was observed. Left-sided salpingo-oophorectomy and omentectomy were done. The peritoneal cavity was washed with copious amounts of warm saline.

She was discharged from the hospital 2 days later with no complications. After 2 months, she was reviewed at the outpatient gynecology clinic and informed of the histopathology result.

The histopathology sections of the left ovary were consistent with MCT, revealing an infarcted hemorrhagic cyst wall with mature adipocytes, focally lined by keratinizing stratified squamous epithelium with underneath skin appendages. The fallopian tube showed extensive hemorrhage and infarction. The peritoneal fluid was grossly mucoid and slightly blood-stained; smears showed degenerated and inflammatory cells, no atypia. No malignant, lymph node involvement or immature elements were identified.

## DISCUSSION

Mature cystic teratoma consists of well-differentiated germ cell layer derivatives, ectoderm, mesoderm, and endoderm, developing as hair, muscle, teeth, or bone.<sup>1</sup> Mature cystic teratoma, the commonest ovarian tumor, comprises 5–25% of all cases.<sup>1</sup>

Typically, it is discovered during the reproductive years, and most patients are asymptomatic. The teratoma is usually found on a regular ultrasonographic test or with calcifications in standard abdominal X-ray imaging.

It is the commonest tumor that causes torsion, which occurs when the vascular pedicle supplying the ovary rotates, compressing and terminating the ovary's blood supply. Patients may present classically with acute or subacute abdominal pain, nausea, vomiting and mild shock, or there may be recurrent episodes of pain over some times as the pedicle twists and untwists. Finally, the pain persists as the ovarian blood supply compromises and gangrene of the ovary takes place.

In this case, the clinical presentation was rather peculiar with the patient's experience of severe radiation of the contralateral RIF pain that overrides the pathological site LIF and was misdiagnosed as a perforated appendix initially. The possible explanation could be

the spillage of the ruptured teratoma's extremely irritant sebaceous material into the RIF and the abdominal cavity, generating a pronounced granulomatous reaction, also known as chemical peritonitis, an aseptic inflammatory peritoneal reaction. In line with this, the patient was ill with lower third abdomen tenderness, while the peritoneal fluid was mucoid and blood-stained, with smears revealing inflammatory cells.

Clinically, the patient's symptom corresponded to the symptom of torsion and rupture of ovarian tumor in which she presented with sudden sharp LIF pain during Valsalva maneuver when having bowel opening associated with nausea and vomiting and rebound abdominal tenderness in RIF; while the fast-progressing nature of the suprapubic mass was more suggestive of an adnexa mass rather than perforated appendicitis.

Various clinical signs and symptoms of acute appendicitis, including fever, right lower quadrant (RLQ) pain, anorexia, and leukocytosis, which is similar to a ruptured ovarian cyst can cause a delay in diagnosis and management of acute abdomen. Acute appendicitis is a surgical emergency to be excluded as the patient later experienced sudden radiation of the sharp pain to RIF that worsened on the day of admission. Nevertheless, there was no fever, and on examination, the tenderness is more on the LIF.

Meanwhile, spontaneous ruptures of MCT can be asymptomatic or cause sudden unilateral, sharp lower abdomen pain, frequently during strenuous physical activities. Although rare, the size helps predict it as the bigger the size with a higher risk of rupture. Most cases fall into the 6–10 cm range, with torsion and infarction contributing to it, as in our case.

Other factors include prolonged pressure during pregnancy or direct trauma. The right ovary is most commonly affected, possibly due to the rectosigmoid colon protection of the left ovary from trauma and torsion, although the left ovary is involved in our instance.

Chemical peritonitis is categorized based on the leakage duration: acute and chronic. Acute peritonitis is caused by an abrupt large cyst rupture and leakage of sebaceous material, whereas chronic granulomatous peritonitis is characterized by a continuous leak from a tiny breach in the cyst wall.

The manifestation can be trivial at first. Progressively, patients may present with abdominal pain, distension, ileus, lymphadenopathy, constitutive symptoms or gastrointestinal disturbances.

Ultrasound is the noninvasive first-line investigation for women with a suspected pelvic mass or pelvic pain regardless of location. A transvaginal ultrasound provides better resolution for pelvic masses compared with the abdominal ultrasound and reveals the presence of a Rokitsky nodule (a densely echogenic tubercle projecting into the cystic lumen) (Fig. 1), dermoid mesh, and the "tip of iceberg sign." Doppler blood flow measurement may be helpful to diagnose torsion. When there is discontinuity of the cyst wall is noted at an ultrasound, CT or MRI, rupture can be conclusively proven.

Ultrasound also aids in detecting the malignant potential of pelvic masses. Risk of malignancy index (RMI), which includes the product of ultrasound score, menopausal status and serum CA-125 level has a sensitivity 78% and specificity 87%. Meanwhile, the international ovarian tumor analysis (IOTA) Group morphologically based ultrasound rule, which classifies masses as benign (B-rules) or malignant (M-rules), also reported of having high sensitivity 95%, specificity 91%.<sup>2</sup>

The ultrasound finding of this case indicated that the mass most likely arose from the left ovary, with a normal right ovary. Significant positive free fluid may indicate the tumor or cyst had ruptured, peritonitis or ascites. The RMI or IOTA score was not properly calculated as the patient was scheduled for an emergency laparotomy.

CA-125 is one of the tumor markers for an epithelial ovarian tumor which was mildly raised (42.0 U/mL) in our case. Serial CA-125 monitoring may be valuable in significantly high levels with rapidly growing tumor which is more prone to be malignant rather than benign conditions.<sup>2</sup> Otherwise, the beta-hCG (raised in gestational trophoblastic disease or choriocarcinoma), AFP (raised in yolk sac tumor), and CEA (raised in mucinous tumors) were within the normal range in this case.

The diagnosis of a twisted and ruptured MCT with a gangrenous base was only made intraoperatively and confirmed with histopathological examination to exclude malignancy.

Unfortunately, due to the vague symptoms and incorrect diagnosis despite multiple clinic visits, the surgical intervention was delayed, resulting in its rupture with chemical peritonitis. If prolonged, it can cause complications, such as pelvic adhesive disease, abdominal wall abscesses, enterocutaneous fistula, and chronic pelvic pain.

El Moussaoui M et al. demonstrated a case of ruptured teratoma leading to chemical peritonitis with a 1-month history of relapsing diffuse abdominal pain and bloating.<sup>3</sup> The clinical presentation, duration, and location of abdominal pain may vary with the nature and characteristic of affected adnexa with associated underlying pathology. Both patients were afebrile and had stable vital signs. The abdominal examination in both cases revealed distension and diffuse tenderness. Nevertheless, in our patient, the tenderness was started in the LIF, then radiation to the RIF with a tender cystic suprapubic mass.

Mature cystic teratoma is generally benign with rare recurrence (4.2%). Nevertheless, a malignant transformation may occur in 1–2% of cases, primarily after menopause.<sup>1</sup>

A review article by Sinha and Ewies indicated that MCT does not diminish ovarian reserves.<sup>4</sup> Most are unilateral, but even when bilaterally presented (10%), effective surgical intervention has no deleterious effect on fertility. Nevertheless, complex large cysts (>8 cm) can pose complications, and an improper surgical technique or fluid leak can cause inflammation, irritation, and adhesions. Subfertility may result if functional ovarian tissue is insufficient. Asymptomatic small cysts (less than 5 cm), have a low risk of torsion or malignant transformation. Thus, patients may be monitored until the fertility is completed.

As for the investigation, ultrasound was used as the first-line investigation in our patient instead of CT abdomen in Ek Moussaoui et al.'s case due to its wide availability and cost-effectiveness. However, the CT radiological signs of teratoma, rupture, and peritonitis with enlargement of lymph node and enhancing peritoneal layers (which suggestive of rupture and peritonitis) were unable to be appreciated in our case. The laparoscopic surgeries were done in both cases and confirmed the findings with HPE results.

The influencing factors for MCT surgery in reproductive period are tumor size, bilaterality, the risk of malignancy, possible complications, such as torsion and rupture, a complex solid cystic structure, increased tumor markers, and fertility status.

There is no role of conservative management when torsion or rupture is suspected as in our case and emergency surgical intervention is indicated. However, there is growing evidence of conservative surgery as a possible option for ovarian torsions, such as detorsion and oophorectomy especially in younger women to conserve ovarian function for further reproductive potential. Ovariopexy is not usually advised following untwisting.<sup>4</sup>

Unless the cyst is extremely large or there is an uncertain diagnosis, the laparoscopic approach is considered the gold standard.<sup>4</sup> In our case, an exploratory laparotomy was employed instead of laparoscopic, for the uncertainty of the diagnosis with significant sizeable ovarian mass with solid components. A unilateral salpingo-oophorectomy was done in our patient as infarction and gangrene of the ovary had occurred.

A sudden rupture of an MCT caused by trauma, torsion, or delivery can result in acute peritonitis with pronounced symptoms of shock or massive bleeding. Intraoperatively, an inflamed ruptured ovarian cyst along with peritoneal adhesions and spillage of peritoneal fluid may be seen. Despite its rarity, when assessing patients with discovered or known history of ovarian cyst, a diagnosis of spontaneous or traumatic rupture of ovarian cyst should be considered.

The risk of chemical peritonitis after contents spillage is extremely uncommon with a lack of literature on the treatment of choice. Immediate surgical removal of the cyst with thorough peritoneal lavage is recommended once the diagnosis is made, as in our case. These measures are sufficient to prevent prolonged chemical peritonitis.

## CONCLUSION

Mature cystic teratoma can be complicated with torsion and rupture, leading to chemical peritonitis with a highly heterogeneous presentation. A high index of clinical suspicion,

prompt assessment and appropriate investigation aid in making an early diagnosis. Timely effective surgical intervention with peritoneal irrigation is pivotal for a favorable outcome in preserving fertility.

## Clinical Significance

This article intended to highlight the possibility of overlapping clinical signs and symptoms between surgical emergency and gynecological emergency cases as clinical symptoms are identical. Those findings can be misunderstood as perforated appendicitis though a ruptured ovarian cyst is a definitive diagnosis.

Hence, detail assessment, appropriate investigations, correct diagnosis, and timely intervention are crucial for preventing peritonitis and conserving reproductive function, particularly in the young patients.

## ORCID

Zhong Ning Neoh  <https://orcid.org/0000-0001-9666-8635>

Ma Saung Oo  <https://orcid.org/0000-0003-3414-0258>

Alik R Zakaria  <https://orcid.org/0000-0002-4947-1060>

Anna L Roslani  <https://orcid.org/0000-0003-0518-4577>

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