# **CASE REPORT**



# Combined Pharmacological and surgical treatments for recurrent chemical peritonitis due to rupture of a bilateral mature cystic teratoma: a case report



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# Abstract

**Background** Mature cystic teratomas, a common type of benign ovarian tumors, are associated with complications such as twisting and tumor rupture; Rupture can cause severe chemical peritonitis, and no management policy has been established for the intraoperative and postoperative periods. Although peritoneal lavage and reoperation have been reported, the optimal treatment approach remains undetermined.

**Case presentation** A 30-year-old woman (gravida 0, para 0, and no sexual history) presented with abdominal pain and fever. Blood examination revealed high levels of WBC 9200/μL and CRP 23.7 mg/dL, although hemoglobin was normal. Serum tumor marker levels were also elevated (CA125 58.5 U/mL, CA19-9 36117 U/mL). Abdominal computed tomography revealed bilateral ovarian tumors (92 and 68 mm in the right and left ovaries, respectively). Each tumor cavity had calcification with increased fatty tissue density. We performed laparoscopic surgery for suspected diagnosis of torsion or rupture of a mature cystic teratoma. Intraoperative findings showed spontaneous rupture followed by chemical peritonitis. Therefore, we performed removal of the bilateral adnexal tumors and peritoneal lavage with 3000 mL warm saline to remove fatty components from the abdominal cavity. We also inserted an intra-abdominal drain to remove the residual fatty components. Amoxicillin was also administered for 10 days after surgery. The inflammatory response decreased, and the fever diminished 1 day postoperatively. The patient was discharged on the 10th postoperative day. However, on the 20th postoperative day, the fever and abdominal pain recurred. WBC 16,700/μL, CRP 26.46 mg/dL and tumor marker (CA125 172.3 U/mL, CA19-9 225.2 U/mL) levels were high. Intravenous administration of Prophylactic antibiotics was initiated. As no bacteria were detected in the blood cultures, we started oral prednisolone (5 mg/day) to treat the recurrent chemical peritonitis-induced inflammation. The blood test results and symptoms gradually improved. The patient was discharged on the 37th postoperative day.

**Conclusion** To date, no systematic review has focused on determining the treatment strategy for bilateral rupture of mature cystic teratomas and severe refractory chemical peritonitis. Treating the patient with laparoscopic surgery at the first occurrence and oral steroids for peritonitis recurrence can help avoid highly invasive treatments, such as reoperation or laparotomy.

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Page 2 of 7

**Keywords** Spontaneous rupture, Mature cystic teratoma, Chemical peritonitis, Laparoscopic surgery, Fertility preservation

# Background

Mature cystic teratomas are benign tumors that account for 10-20% of all ovarian tumors. The median age at onset is reportedly 30 years [1-3]. The complications of mature cystic teratomas include twisting and tumor rupture [4]. In the recent report, the incidence of torsion and rupture were 6.9% and 0.75%, of all mature cystic teratoma which underwent surgery [5]. In a recent study, torsion was found to occur more frequently than rupture [5]. The treatment for asymptomatic and torsional mature cystic teratomas in young patients includes tumor removal to preserve fertility [6]. However, cases of rupture are rare, and no management policy has been established for the intraoperative and postoperative periods.

Furthermore, mature cystic teratomas can cause chemical peritonitis due to tumor contents, with an incidence rate of 0.2% [7]. Capsule rupture is classified into rupture during surgery and spontaneous. Chemical peritonitis leads to adhesions and intestinal obstruction; it can also recur due to residual fatty matter, requiring reoperation or adnexal resection [8]. Moreover, chemical peritonitis can become severe after a spontaneous rupture. Thus, it is important to diagnose spontaneous rupture of mature cystic teratomas and chemical peritonitis preoperatively during intraoperative and postoperative management. Peritoneal lavage and reoperation have been reported as possible treatments; however, no established treatments exist [9, 10].

In this case report, we present the procedure of bilateral tumor removal for ovarian preservation and peritoneal lavage to treat extensive chemical peritonitis caused by the spontaneous rupture of bilateral mature cystic teratomas. The patient experienced a recurrence of chemical peritonitis 2 weeks after surgery and was treated conservatively with oral steroids. To the best of our knowledge, this is the first case wherein tumor treatment and fertility preservation were performed in a patient with recurrent chemical peritonitis due to the spontaneous rupture of a mature cystic teratoma.

# **Case presentation**

A 30-year-old woman (gravida 0, para 0, and no sexual history) presented to our hospital with abdominal pain and fever. The patient had previously undergone surgery for appendicitis when she was 4 years old. She had no comorbidities and did not take any regular medications. Her menstrual cycles were regular with a 28-day interval and lasts for 4 to 5 days. The last menstrual period was 2 weeks before her visit to the emergency department. She experienced severe abdominal pain, mainly in the

left abdomen, and a fever of 38 °C. She has never experienced similar symptoms. A blood test showed high white blood cell count (WBC; 9200/µL) and C-reactive protein (CRP; 23.7 mg/dL) level. The hemoglobin(Hb) level was 11.5 mg/dL, within the normal range. The tumor marker levels were also elevated. Serum cancer antigen 125 (CA125), carbohydrate antigen 19-9 (CA19-9), and squamous cell carcinoma antigen (SCC) levels were 58.5 U/mL, 36,117 U/mL, and 2.0 ng/mL, respectively. Carcinoembryonic antigen and alpha-fetoprotein levels were within normal limits at 3.6 and 0.8 ng/mL, respectively. Computed tomography (CT) revealed bilateral ovarian tumors (92 and 68 mm in the right and left ovaries, respectively). Calcification was observed in each tumor cavity, and the fatty tissue density increased (Figs. 1 and 2).

The tumor wall was smooth, and calcification was only observed in one area of each tumor. Imaging findings did not reveal any findings suggestive of malignant tumors, nor were there any findings suggestive of diseases of other organs, such as enteritis or pyelonephritis. Increased fatty tissue density and a small amount of ascites were observed around both tumors (Fig. 3).

Torsion or rupture of a mature cystic teratoma was suspected. Therefore, we decided to perform laparoscopic surgery. Intraoperative findings revealed bilateral ovarian tumors with hiatus in each tumor (Figs. 4 and 5).

The fat components in the tumors were widely dispersed from the pelvic cavity below the diaphragm (Fig. 6).

We diagnosed the patient with spontaneous rupture of bilateral mature cystic teratomas, followed by chemical peritonitis. We performed laparoscopic removal of the bilateral adnexal tumors and peritoneal lavage with 3000 mL of warm saline to remove fatty components from the abdominal cavity. We also inserted an intra-abdominal drain to remove the residual fatty components. The inflammatory response decreased, and the fever diminished from the day after surgery. After confirming the lack of drainage of fatty components, we removed the intra-abdominal drain on the 7th postoperative day. Amoxicillin was administered orally for 10 days starting the day after surgery. As both blood test data and symptoms improved, the patient was discharged on the 10th postoperative day. The removed tumor was macroscopically found to contain a large amount of hair and fat components. Histological examination of the resected specimen revealed keratinized stratified squamous epithelium, hair follicle, adipose tissue, and cartilage tissue. Histological findings were typical of a mature cystic



Fig. 1 Right ovarian tumor(arrow)



Fig. 2 Left ovarian tumor(arrow)



Fig. 4 Right ovarian tumor (location of capsula rupture) (arrow)



Fig. 5 Left ovarian tumor (location of capsula rupture) (arrow)



Fig. 3 Increased fatty tissue density(arrows)

teratoma (no malignant findings such as suggestive of immature teratoma were observed.) (Figs. 7 and 8).

After discharge, the progression was monitored on an outpatient basis. On the 20th postoperative day, the



Fig. 6 Fat components below the diaphragm(arrows)



Fig. 7 Gross appearance of the tumor



Fig. 8 Pathological findings of tumor (Hematoxylin Eosin staining × 4)



Fig. 9 Increased concentration and thickness of fatty tissue(arrows)

patient experienced a recurrence of fever of 38  $^{\circ}$ C and abdominal pain, so she was readmitted to the hospital. A blood test showed high WBC count (16700/µL) as well as elevated CRP(26.46 mg/dL) and tumor marker levels (CA125 172.3 U/mL, CA19-9 225.2 U/mL). Abdominal computed tomography showed increased concentration and thickness of fatty tissue in the abdominal cavity (Fig. 9).

We considered the possibility of recurrent chemical peritonitis, intraperitoneal infection and enteritis. We initiated intravenous administration of prophylactic antibiotics. As no bacteria were detected in the blood cultures and there were few gastrointestinal symptoms such as diarrhea, we diagnosed the patient with recurrent chemical peritonitis and we started oral prednisolone (5 mg/day) to treat the inflammation caused by recurrent chemical peritonitis. The patient's blood test results and symptoms gradually improved. The patient was discharged on the 17th day (37th postoperative day). Two months postoperatively, the patient's blood test results normalized and symptoms resolved completely. Three months postoperatively, no recurrence of peritonitis was observed, and serum CA125 and CA19-9 levels normalized to 30.5 and 11.3 U/mL, respectively.

# **Discussion and conclusions**

To the best of our knowledge, this is the first report of recurrent chemical peritonitis due to the spontaneous rupture of a mature cystic teratoma treated with laparoscopic surgery and postoperative management with fertility preservation.

Mature cystic teratomas are the most frequent type of benign ovarian tumors in women of reproductive age [1]. Acute abdominal manifestations of mature cystic teratomas include tumor pedicle torsion and rupture, with torsion occurring more frequently than rupture. The symptoms of both conditions are similar; however, in cases of torsion, blood tests show significant increases in tumor markers (CA125, CA19-9, and SCC) and CRP. In a previous review, the median values for CA125, CA19-9, and SCC were 90.3 U/mL, 160.9 U/mL, and 3.1 ng/ mL, respectively (CRP values not reported) [5]. In this case report, blood tests showed a significant increase in tumor marker levels, especially CA19-9, at 36,117 U/ mL. Thus, owing to the increased CA19-9 levels, we suspected rupture and chemical peritonitis. The correlation between the actual measured marker values and severity has not been reported previously. In this case, the values were higher than those previously reported, and the disease recurred after surgery; therefore, such a correlation may exist. In addition, CT scans show characteristic findings such as scattered low-attenuation areas (fat droplets) in the ascites, increased concentration of fatty tissue, and thickening of the omentum [11, 12]. In this

Tab	ble	1	Case reports on	chemical	peritonitis (	(spontaneous rupture
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Study	Age	Symptoms	Diameter (mm)	Localization	Tumor marker	Treatment	Hospitaliza- tion (days)
Buzinskiene	35	Abdominal pain	160	Unilateral	Not specified	Cystectomy(laparoscopy)	7
Tsapralis	17	Abdominal pain	50	Unilateral	Not specified	Antibiotics, cystectomy (laparoscopy→laparotomy)	5
Wong	35	Abdominal pain	Not specified	Bilateral	Not specified	Cystectomy(Not specified)	Not specified
Vulasala	33	Abdominal pain	Not specified	Unilateral	Not specified	Adnexal resection(laparoscopy)	Not specified
Li	66	Abdominal pain, fever	74	Unilateral	CA125: 29 U/mL	Adnexal resection (laparoscopy→laparotomy)	6
Tasaka	37	Abdominal pain	65	Unilateral	Not specified	Adnexal resection+PSL (laparoscopy→laparotomy)	22+a
Mazhoud	21	Abdominal pain	90	Unilateral	Not specified	Cystectomy(laparotomy)	Not specified
Chandar	32	Abdominal pain	Not specified	Unilateral	Not specified	Adnexal resection(laparoscopy)	Not specified
Our case	30	Abdominal pain, fever	92/68	Bilateral	CA125: 58.5 U/mL, CA19-9: 36,117 U/ mL	Cystectomy(laparoscopy)	11

CA125, cancer antigen 125; CA19-9, carbohydrate antigen 19-9; PSL, prednisolone

**Table 2** Case reports on chemical peritonitis (intraoperative rupture)

Study	Age	Symptoms	Diameter (mm)	Localization	Tumor marker	Onset	Treatment	Hospitalization (days)
Shamshirsaz	41	Abdominal pain, fever	28/25	Bilateral	Not specified	POD3	Reoperation (laparoscopic lavage)	4
Kim	39	Abdominal pain, fever	Not specified	Not specified	Not specified	POD90	Reoperation (laparoscopic lavage)	5
Yamawaki	40s	Abdominal pain	95/82	Bilateral	CA125: 9 U/mL CA19-9: 166 U/mL	POD30	Reoperation (laparoscopic lavage)	11
Yamawaki	20s	Abdominal pain	100/59	Bilateral	CA125: 13 U/mL] CA19-9: 107 U/mL	POD27	PSL	13
Our case	30	Abdominal pain, fever	92/68	Bilateral	CA125: 172.3 U/mL CA19-9: 225.2 U/mL	POD20	PSL	18

CA125, cancer antigen 125; CA19-9, carbohydrate antigen 19-9; POD, postoperative day; PSL, prednisolone

case, preoperative CT revealed increased density of fatty tissue surrounding the tumor. Moreover, CT performed at the time of recurrent chemical peritonitis showed fatty tissue around the intra-abdominal organs, including the liver. Predicting tumor rupture based on preoperative test findings may be useful for managing tumors during surgery.

Chemical peritonitis is a secondary form of peritonitis caused by tumor contents or drugs and has been reported in many cases after the rupture of mature cystic teratomas. In a review of cases of chemical peritonitis associated with mature cystic teratomas, nine cases (including our case) of spontaneous rupture (Table 1) and five of intraoperative rupture (including the in our case) (Tables 1 and 2) were reported.

In a previous report of spontaneous rupture (Table 1), the patients' ages ranged from 17 to 66 years. Eight patients experienced menstrual cycles, and one was postmenopausal. All patients presented with abdominal pain, and two had fever. The tumor size ranged from 50 to 160 mm, and the tumors were relatively large. Of all cases, seven were unilateral, and two were bilateral. Although a few studies have reported tumor marker values, our case was accompanied by a significant increase in tumor marker levels compared with those in previous reports. All patients with spontaneous rupture were simultaneously diagnosed with chemical peritonitis. In the case of spontaneous rupture, treatment consisted of laparoscopic cystectomy in three cases; cystectomy without mention of the surgical procedure in one; laparoscopic adnexal resection in two; abdominal cystectomy with antibiotics in one; abdominal adnexal resection in one, which was switched from laparoscopy; and abdominal adnexal resection combined with steroids in one, switched from laparoscopy. In contrast to the current report, most reports focused on open surgery and adnexal resection, which are highly invasive treatments. The length of hospitalization for spontaneous rupture cases ranged from 5 to 29 days, including that in our case.

In cases of intraoperative rupture and recurrence, the age of the patients ranged from 20 to 41 years, and all patients experienced menstruation. Furthermore, all patients exhibited abdominal pain, and three had fever. The onset of peritonitis occurred at 3, 20, 27, 30, and 90 days postoperatively. The tumor sizes ranged from 25 to 100 mm. All cases were bilateral, except for those that were not mentioned. In terms of tumor markers, CA19-9 was increased in three cases, including our case, whereas CA125 was increased only in our case. In intraoperative rupture and recurrence cases, treatment consisted of invasive laparoscopic reoperation in three cases, whereas conservative, noninvasive, and oral prednisolone treatment was administered in two, including our case. Considering the long hospital stay and increased tumor marker levels, our case might be more severe than others [8, 13–22].

To the best of our knowledge, this is the first case of spontaneous rupture of bilateral tumors, and also the first case of delayed recurrence after spontaneous rupture. In our case, the tumor markers were abnormally increased, which has not been previously reported. Considering that the disease recurred despite initial improvement, tumor markers were possibly correlated with disease severity.

There is currently no established preventive measure and treatment for chemical peritonitis associated with mature cystic teratoma. In previously reported cases, patients had undergone laparotomy or laparoscopic washing, or had received oral steroids. From the viewpoint of preventing capsule rupture, it has been reported that oophorectomy is superior to cystectomy in reducing the risk of intraoperative rupture [23]. On the other hand, it has also been reports that unilateral oophorectomy reduces fertility. Therefore, cystectomy is the first choice from the perspective of maintaining fertility regardless of whether rupture exists [24]. In ruptured mature cystic teratomas, intraoperative irrigation may reduce postoperative inflammation and adhesion in animal models [25]. In this case, despite adequate cleaning during surgery and postoperative drainage of fatty fluid, the disease recurred. However, after relapse, the symptoms improved with only oral steroids, suggesting that adequate washing during primary laparoscopic surgery may suppress the inflammatory reaction to a feasible extent.

Furthermore, when the condition recurred, oral steroids were initiated after the absence of bacterial infection was confirmed, and the condition progressed without recurrence. There have been reports of oral steroids being extremely effective against peritonitis caused by barium leakage after gastrointestinal contrast examinations, suggesting their effectiveness against aseptic peritonitis [26]. In this case, it is presumed that the antiinflammatory effect of steroids contributed to the chemical peritonitis. However, it is necessary to be careful about the susceptibility to infection due to immunosuppression when using steroids. Therefore, before administering steroids, a careful examination for the possibility of infection, such as a culture test, is necessary. In this case, blood cultures were performed to rule out infection. In the case of infection is suspected, antibiotics should be administered instead of steroids.

Our treatment options were based on these findings. In conclusion, laparoscopic tumor removal, peritoneal lavage, and intra-abdominal drainage are could be a relapse preventing and fertility sparing treatment for spontaneous rupture of mature cystic teratoma patients of reproductive age. Moreover, even if chemical peritonitis occurs, oral prednisolone could avoid reoperation.

These comprehensive treatments could be an optimal clinical approach for fertility preservation and avoidance of invasive procedure.

#### Abbreviations

CT	Computed Tomography
WBCs	White Blood Cells
CRP	C-Reactive Protein
CA125	Cancer Antigen 125
CA19-9	Carbohydrate Antigen 19–9
SCC	Squamous Cell Carcinoma Antigen

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#### Author contributions

SK, HT, and KT performed the surgeries. SK, HT, and YT were involved in the acquisition of data and preparing the figures. SK and HT wrote the manuscript. MO, and YY proofread and revised the manuscript. All authors read and approved the final manuscript.

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#### Data availability

No datasets were generated or analysed during the current study.

# Declarations

#### Ethics approval and consent to participate

This report was approved by the Hospital Ethics Committee of the University of Fukui

#### Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

#### **Competing interests**

The authors declare no competing interests.

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