

Chemical peritonitis resulting from spontaneous rupture of a mature ovarian cystic teratoma: a case report

Diana Bužinskienė^{1,2*},

Matas Mongirdas³,

Saulius Mikėnas^{4,5},

Grażina Drąsutienė^{1,2},

Linas Andreika²,

Indrė Sakalauskaitė^{6,7}

¹ Clinic of Obstetrics and Gynaecology,
Institute of Clinical Medicine,
Faculty of Medicine,
Vilnius University,
Vilnius, Lithuania

² Centre of Obstetrics and Gynaecology,
Vilnius University Hospital
Santaros Klinikos,
Vilnius, Lithuania

³ Faculty of Medicine,
Vilnius University,
Vilnius, Lithuania

⁴ Clinic of Gastroenterology,
Nephro-Urology and Surgery,
Centre of Urology,
Institute of Clinical Medicine,
Faculty of Medicine,
Vilnius University,
Vilnius, Lithuania

⁵ Centre of Urology,
Vilnius University Hospital
Santaros Klinikos,
Vilnius, Lithuania

Background. Mature cystic teratomas (dermoid cysts) are the most common germ cell tumours with 10–25% incidence of adult and 50% of paediatric ovarian tumours. The aetiology of dermoid cysts is still unclear, although currently the parthenogenic theory is most widely accepted. The tumour is slow-growing and in the majority of cases it is an accidental finding. Presenting symptoms are vague and nonspecific. The main complication of a dermoid cyst is cyst torsion (15%); other reported complications include malignant transformation (1–2%), infection (1%), and rupture (0.3–2%). Prolonged pressure during pregnancy, torsion with infarction, or a direct trauma are the main risk factors for a spontaneous dermoid rupture that can lead to acute or chronic peritonitis. The diagnosis of mature cystic teratoma is often made in retrospect after surgical resection of an ovarian cyst, because such imaging modalities as ultrasound, computer tomography, or magnetic resonance imaging cannot yet accurately and reliably distinguish between benign and malignant pathology.

Materials and methods. We present a report of a clinical case of a 35-years-old female, who was referred to the hospital due to abdominal pain spreading to her feet for three successive days. She had a history of a normal vaginal delivery one month before. Abdominal examination revealed mild tenderness in the lower abdomen; no obvious muscle rigidity was noted. Transvaginal ultrasound showed a multiloculated cystic mass measuring 16 × 10 cm in the pelvis. In the absence of urgency, planned surgical treatment was recommended. The next day the patient was referred to the hospital again, with a complaint of stronger abdominal pain (7/10), nausea, and vomiting. This time abdominal examination revealed symptoms of acute peritonitis. The ultrasound scan differed from the previous one. This time, the transvaginal ultrasound scan revealed abnormally changed ovaries bilaterally. There was a large amount of free fluid in the abdominal cavity. The patient was operated on – left laparoscopic cystectomy and right adnexectomy were performed. Postoperative antibacterial treatment, infusion of fluids, painkillers, prophylaxis of the thromboembolism were administered. The patient was discharged from the hospital on the seventh postoperative day and was sent for outpatient observation.

⁶*Clinic of Rheumatology,
Orthopaedics Traumatology and
Reconstructive Surgery,
Institute of Clinical Medicine,
Faculty of Medicine,
Vilnius University,
Vilnius, Lithuania*

⁷*Centre of Plastic and
Reconstructive Surgery,
Vilnius University Hospital
Santaros Klinikos,
Vilnius, Lithuania*

Results and conclusions. Ultrasound is the imaging modality of choice for a dermoid cyst because it is safe, non-invasive, and quick to perform. Leakage or spillage of dermoid cyst contents can cause chemical peritonitis, which is an aseptic inflammatory peritoneal reaction. Once a rupture of an ovarian cystic teratoma is diagnosed, immediate surgical intervention with prompt removal of the spontaneously ruptured ovarian cyst and thorough peritoneal lavage are required.

Keywords: chemical peritonitis, dermoid cyst, mature cystic teratoma, peritoneal lavage, spontaneous rupture

INTRODUCTION

Mature cystic teratomas or dermoid cysts are the most common germ cell tumours with the incidence of 10–25% and 50% of paediatric ovarian tumours (1). Dermoid cysts comprise about 30% of all benign ovary tumours and can occur at any age, but they are more common during reproductive years, especially in patients under 30 years of age (2). These tumours can be bilateral in 8–15% of all cases (3). The tumour is slow-growing and usually asymptomatic, and because of that it is an accidental finding in the majority of cases. Clinical symptoms are associated with complications. The main complication of a dermoid cyst is cyst torsion (15%). Other reported complications include malignant transformation (1–2%), infection (1%), and rupture (0.3–2%) (4). A spontaneous rupture is an extremely rare complication of mature cystic teratoma because of its usually thick capsule. Prolonged pressure during pregnancy, cyst torsion with infarction, or a direct trauma are the main risk factors for a spontaneous dermoid rupture that can lead to acute or chronic peritonitis (5). The incidence of chemical peritonitis after a cyst rupture is less than 1%. This condition

requires urgent surgical treatment (6). We report a case of 35-year-old female, who developed chemical peritonitis resulting from a spontaneous intraperitoneal rupture of a dermoid cyst.

THE CLINICAL CASE

A 35-year-old female, gravida 3, para 3, presented to our emergency room with a complaint of abdominal pain spreading to her feet for three successive days. She had a medical history of a normal vaginal delivery one month before. She had no gynaecological operations or diseases.

On physical examination, the patient's vital signs were stable, temperature was normal. Abdominal examination revealed mild tenderness in the lower abdomen; no obvious muscle rigidity was noted. A big mass in the pelvis was felt during pelvic examination.

Transvaginal ultrasound showed a multiloculated cystic mass measuring 16 × 10 cm in the pelvis (Fig. 1). There was no fluid in the pelvis. In the absence of urgency, planned surgical treatment was prescribed.

The next day, the patient presented to our clinic again, with a complaint of stronger abdominal pain (7/10), nausea, and vomiting. This time abdominal examination revealed symptoms of peritonitis: tenderness in the lower abdomen and muscle rigidity. Total white blood cell count was

* Correspondence to: Diana Bužinskienė, Centre of Obstetrics and Gynaecology, Vilnius University Hospital Santaros Klinikos, 2 Santariškių St., Vilnius 08661, Lithuania.
Email: diana.buzinskiene@santa.lt

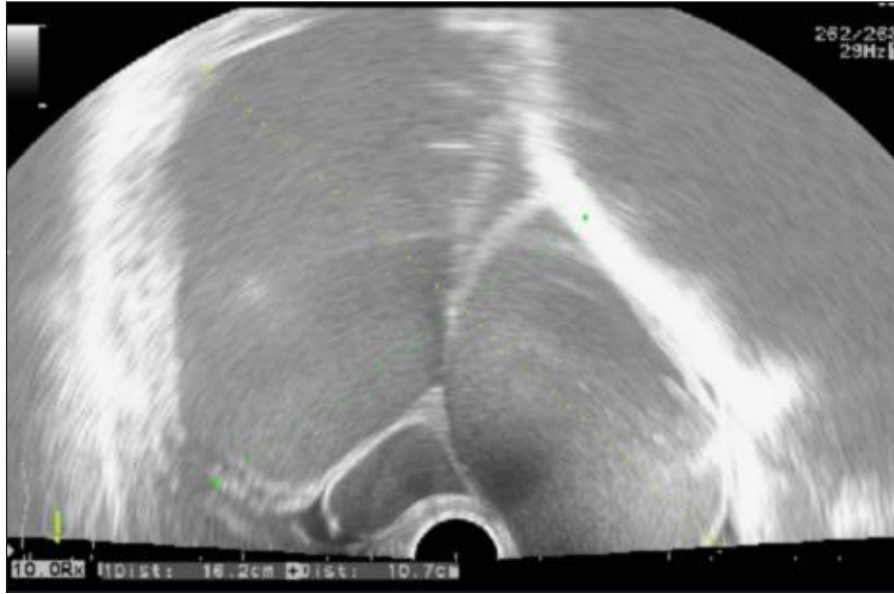


Fig. 1. Transvaginal ultrasound: a multiloculated cystic mass measuring 16×10 cm in the pelvis

$15.60 \times 10^9/L$ and C-reactive protein (CRP) was increased (96 mg/l). Ultrasound scan pictures were different from previous. This time transvaginal ultrasound scan revealed abnormally changed ovaries bilaterally. The multiloculated cystic mass in right ovary measured 8.8×5.2 cm (Fig. 2) and the multiloculated cystic mass in the left ovary was 4.5×5.5 cm in size (Fig. 3). Uterine size was 4.7×3.2 cm, with endometrial thickness of 6 mm. There was a moderate amount of free fluid

in the abdominal cavity. A CT scan was not performed. The patient was admitted to the gynaecological department for surgical treatment.

Laparoscopic surgery was undertaken. Operative findings revealed about 1500 ml of yellowish ascites containing adipose component. The peritoneum, the bowel, the stomach, the liver, the uterus, and both adnexa were covered by diffuse, thick, white to yellowish lesion. A cystic mass of 7 cm in size was seen in the left ovary

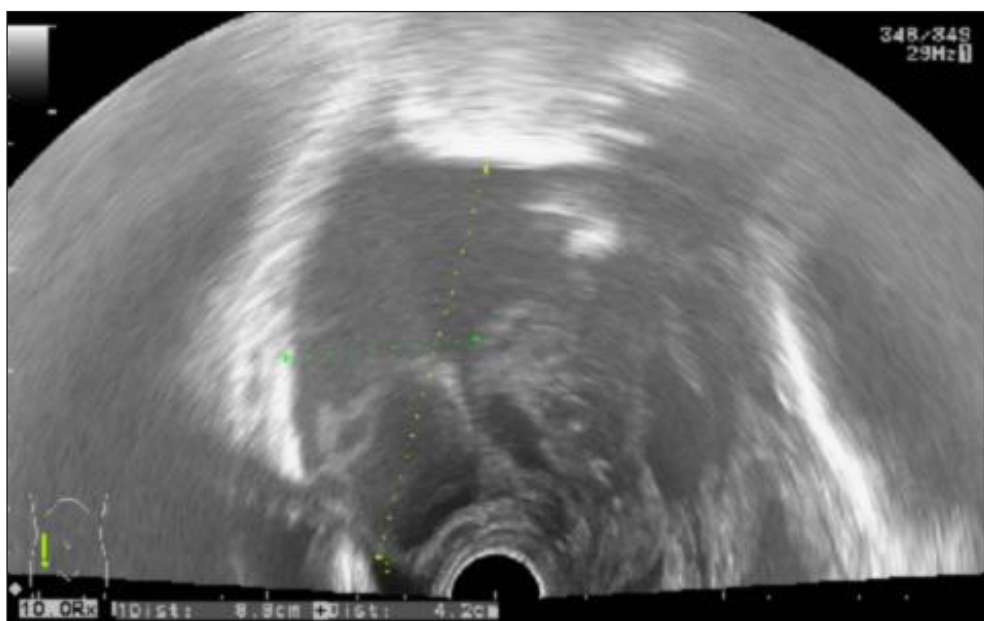


Fig. 2. Transvaginal ultrasound: multiloculated cystic mass in the right ovary

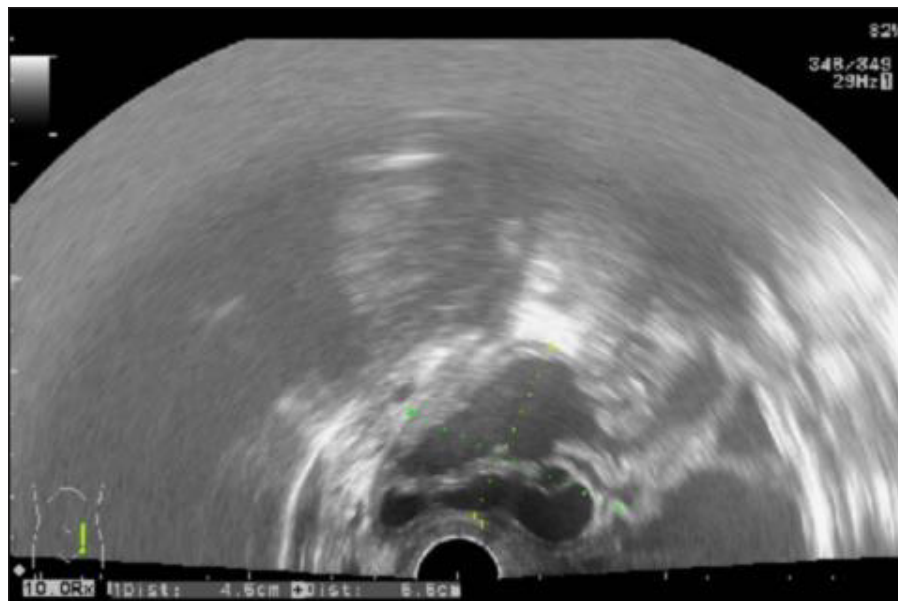


Fig. 3. Transvaginal ultrasound: a multiloculated cystic mass in the left ovary

and it was strongly adhered to the superior part of the rectum. The right ovary with a cystic mass approximately 20 cm in size was torsed, ruptured, and necrotized. The right fallopian tube was fixed to the unilateral ovary (Fig. 4).

Abdominal surgeons were called into the operating room to evaluate the possibility of surgical pathology. The pathology of the peritoneum, the bowels, the stomach, the liver, and the appendix was declined, and there were no signs of perforation or diverticulation. Adherence between

the left adnexa and the superior part of the rectum were dissected.

Left cystectomy and right oophorectomy were performed (a hair tuft was present inside the cyst). Peritoneal lavage with 4000 mL of warm saline, until the solution was clean, was administered. The abdominal cavity was drained with two drains.

The histological examination showed bilateral mature dermoid cysts. The section of the left cystic ovary revealed accumulation of various mature



Fig. 4. Intraoperative laparoscopic findings: a massive left ovarian cyst

tissues such as adipose, muscle tissue, mucous gland, cartilage, hair follicles with focal wall lymphoplasmocytic infiltration, and foreign body giant cells. The wall of the right cystic ovary was partially necrosed with digestive tract cells and diffuse neutrophil infiltration. The ascitic fluid cytology was negative for bacteria. Antibiotic therapy with Sol Amoxiclavi 1.2 g I/V every 6 h and Sol. Metronidazoli 500 mg I/V every 8 h for seven days, infusion therapy, and prophylaxis of thrombembolism were administered.

The postoperative course was uneventful. The patient was discharged on the seventh postoperative day and sent for further outpatient observation.

LITERATURE REVIEW AND DISCUSSION

Definitions and classification

Ovarian germ cell tumours (OGCTs) are heterogeneous tumours that are derived from primitive germ cells of the embryonic gonads. According to the 2014 classification by the World Health Organisation (WHO), the tumours are divided into two main categories: primitive malignant neoplasms of germinogenic cells (dysgerminomas) and teratomas, most of which are mature cystic teratomas (7).

Teratomas are the most common germ cell tumours, which might consist of multiple cell types derived from one or more of three germ layers. The word teratoma is derived from Greek word *teras*, which means monster. This term was formulated in 1863 in the first edition of Virchow's book about tumours (8). Teratomas can be divided into two groups:

- a) mature teratomas, which include dermoid cysts, and are generally benign
- b) immature teratomas, which can be cancerous (9).

Epidemiology

OGCTs constitute about 20–25% of all ovarian neoplasms. Ninety-five per cent of tumours are benign. Mature cystic teratomas (MCT) are the most common germ cell tumours that comprise about 95% of all ovarian germ cell tumours and 10–25% of all ovarian tumours (10). The incidence rate of dermoid cysts is 1.2–14.2/100 000 females (11).

This type of tumour can occur at any age, but is more common during reproductive years, espe-

cially in patients under 30 years of age (2). Early diagnosis and operative treatment (cystectomy) in childhood and adolescence are important for the development of normal puberty and future fertility (12). In their study, Zupi et al. compared tumour-affected ovaries before and after laparoscopic ovarian cystectomy. Of 632 patients, 56 had 3 cm³ or larger ovarian volume at 12 months post-operatively on transvaginal ultrasound. Moreover, 24 of these patients had no normal ovarian tissue pre-operatively during transvaginal ultrasound (13).

Aetiology

The exact mechanisms of the development of teratomas remain unclear. Currently, the parthenogenic theory is the most widely accepted. Most have a 46XX karyotype and by parthenogenesis develop from a single haploid germ cell. This theory supports the presence of Barr bodies (nuclear sex chromatin) in the tumour cells (8). There are no new data about the risk factors for the development of the benign dermoid cyst. Parazzini et al. revealed that there is no relation between the marital status, the age at menarche, the menopausal status, abortions, the age at the first pregnancy, oral contraceptive use and the risk of ovarian benign teratomas (14). In their retrospective study, Park et al. revealed that elevated CA-125 levels, older age, large tumour masses, and the postmenopausal status are the main risk factors for malignant transformation of MCT (15).

Location

Although mature cystic teratomas (MCT) commonly occur in ovaries, they can be found anywhere in the body. Dermoid cysts may occur at extragonadal sites, usually along the midline of the body: anterior mediastinum, the sacrococcygeal region, the retroperitoneal area, the head, the neck, and the central nervous system. Mature cystic teratomas have been reported throughout the gastrointestinal tract in caecum, pancreas, and omentum (16, 17). Dermoid cysts are bilateral in 8–15% of cases (18). Sometimes ipsilateral multiple ovarian teratomas can occur (with the incidence of about 9%) (9). Ruchi Rathore et al. reviewed different studies and revealed that pathology side in unilateral cases may vary depending on studies (19).

Symptoms

In at least a quarter of all cases, MCT are asymptomatic and discovered accidentally on a routine pelvic examination, imaging, or during surgery (20). If it causes symptoms, the most common symptom of ovarian MCT is lower abdomen pain, which occurs in nearly half of the patients (9). On the other hand, this type of tumour may also become symptomatic due to complications. The latter include dermoid cyst torsion (15%), rupture (0.3–2%), malignant transformation (1–2%), and infection (1%) (4). Nausea, vomiting, constipation, urinary retention and hydronephrosis are less common symptoms (21).

Moreover, due to the slow growth of the cystic mass, 1.8 mm/year, dermoid cysts are usually large at the time of diagnosis. Growth factors remain unclear, but increasing levels of hormones oestrogen and progesterone may explain the increase in the size of dermoid cysts after puberty and their arrested growth after menopause (22). Big size of the tumour may cause symptoms caused by pressure of the adjacent organs, bowel obstruction, and fistula formation into the small and large bowel (23). Garrido-Abad et al. described an unusual cause of a renal colic case caused by a giant dermoid cyst (24).

An extremely rare manifestation of mature cystic teratoma is anti-NMDA-receptor encephalitis. Neurological symptoms develop because of the antibodies generated in response to the neural elements within the teratoma (25).

Diagnostics

Preoperative diagnosis of mature cystic teratoma still remains a very challenging task for clinicians, because primary symptoms are often vague and nonspecific. It is advisable to start from vaginal examination (it is important for differential diagnosis). The clinician can evaluate the size, the contour, and mobility of the uterus with adnexes. Abdominal examination can reveal a large teratoma.

In imaging, MCTs have a wide spectrum of radiological presentation ranging from a purely cystic mass to a complex cystic mass with a considerable solid component. Ultrasound is the imaging modality of choice for a dermoid cyst because it is safe, non-invasive, and quick to perform, although it is not always easy to differentiate MCT from the haemorrhagic cyst (26). MCTs ultrasound findings include the presence of a Rokitansky nodule, dermoid mesh, and the “tip of iceberg sign”. The Rokitansky nodule

is a cystic lesion with a densely echogenic tubercle projecting into the cystic lumen. Dermoid mesh refers to hyperechogenic lines arising from hairs (27). Ultrasound is also useful in differentiating benign ovarian masses from malignant lesions. Malignancy characteristics include:

- 1) ovarian volume above 20 cm³;
- 2) septations;
- 3) papillary projections;
- 4) heterogeneous tissue echogenicity;
- 5) vascular support to a lesion (Color Doppler) (28).

Computed tomography (CT) has an excellent sensitivity (93–98%) due to detecting fat in the diagnosis of a dermoid cyst. The presence of fat (areas with low Hounsfield units) inside an ovarian tumour and calcifications appear to be specific to a mature cystic teratoma (29).

The exact diagnosis of a dermoid cyst is determined by histological examination. Macroscopically, dermoid cysts usually are multicystic and contain sebaceous fluid and tissue of two or more germ cell layers, e.g., ectoderm (hair, skin, brain), mesoderm (teeth, bone, cartilage, muscle, fat), or endoderm (mucous and ciliated epithelium) (30). Furthermore, it is important to emphasize, that attentive histopathological examination of postoperative specimen is mandatory. MCTs can frequently arise in combination with over ovarian neoplasms, that can be fatal without an additional treatment (31).

Dermoid cyst rupture and chemical peritonitis

MTC can rupture spontaneously or during surgery. The rarity of a spontaneous intraperitoneal dermoid cyst rupture (0.3–0.7%) is due to a thick capsule. It may rupture into the peritoneal cavity or into the adjacent hollow organs like bowels, the bladder, or the vagina (10). The exact causes of the rupture are mostly unknown, but factors such as prolonged pressure during pregnancy, torsion with infarction, direct trauma, malignant transformation, and internal pressure from rapid growth of the mass are the most likely explanations (5). It is important to mention that in our case the patient had a vaginal delivery a month before the symptoms occurred.

The traditional surgical treatment of dermoid cysts was cystectomy by laparotomy, although laparoscopic cystectomy is now widely accepted. The rates of spillage of dermoid cysts into the peritoneal

cavity after the removal of cysts by laparoscopy were 15% to 100% in several series, compared to much lower spillage rates (only 4% to 13%) when cysts were removed by laparotomy (32). Leakage or spillage of dermoid cyst contents can cause chemical peritonitis, which is an aseptic inflammatory peritoneal reaction. Chemical peritonitis after spillage of teratoma contents after laparoscopic surgeries is a rare complication with an incidence of less than 1% if the contents are fully removed (6). If chemical peritonitis develops, early repeat surgery may be advantageous, especially if remnants are suspected.

There are two different types of clinical presentation, which depend on the leakage time: acute and chronic. Acute peritonitis is caused by a sudden wide cyst rupture and abrupt leakage of sebaceous material, while chronic granulomatous peritonitis is caused by a slow leak from a tiny tear in the cyst wall (3).

The clinical presentation might be insignificant in the early period, however, the patient would complain of progressive abdominal pain, abdominal distention, fever, and gastrointestinal disturbances such as anorexia, nausea, vomiting, and diarrhoea (33). Chemical peritonitis can result in pelvic adhesive disease or bowel obstruction, abdominal wall abscesses, enterocutaneous fistula formation, and other complications, which require intensive medical management and repeat surgical treatments (32). Since the acute abdomen can be a result of wide range of diseases, differential diagnosis plays an important role.

An accurate diagnosis of a ruptured ovarian teratoma can be confirmed when the discontinuity of the cyst wall is noted at ultrasonography (US), CT, and magnetic resonance (MR) imaging (3). In the presented case, we had an opportunity to perform pelvic ultrasonography on two successive days and it was obvious that the appearance of the multiloculated cystic mass changed. Furthermore, during ultrasonography scan on the second day, there was free fluid in the abdominal cavity. The clinical diagnosis of the ruptured ovarian teratoma was made with a high positive predictive value. We did not perform any other visual diagnostic tests.

Only in a small number of cases, the diagnosis of a ruptured ovarian dermoid cyst can be made by ultrasonography alone. A computed tomography

scan can be helpful because this modality is very sensitive (93–98%) for the detection of an intraperitoneal fatty implant, most commonly around the liver surface (34).

Although there is a lack of literature on the management of chemical peritonitis due to an ovarian cyst rupture, the treatment of choice, once rupture of an ovarian cystic teratoma is diagnosed, should be immediate surgical intervention. The purpose of surgery is prompt removal of a spontaneously ruptured ovarian cyst with thorough peritoneal lavage. These steps are sufficient to prevent prolonged chemical peritonitis (5).

CONCLUSIONS

1) Mature cystic teratomas or dermoid cysts are the most common germ cell tumours that comprise about 30% of all benign ovarian tumours and are the most commonly diagnosed in females of the reproductive age.

2) The exact mechanisms of teratoma development remains unclear.

3) Preoperative diagnosis of mature cystic teratoma still remains a very challenging task for clinicians, because primary symptoms are often nonspecific. In at least 25% of all cases, MCTs are asymptomatic and discovered accidentally on a routine pelvic examination, imaging, or during surgery.

4) Ultrasound is the imaging modality of choice for a dermoid cyst because it is safe, non-invasive, and quick to perform.

5) The main complications of dermoid cysts are cystic torsion (15%), malignant transformation (1–2%), infection (1%), and rupture (0.3–2%).

6) Prolonged pressure during pregnancy, torsion with infarction, or a direct trauma are the main risk factors for spontaneous dermoid rupture, which can lead to acute or chronic peritonitis.

7) Leakage or spillage of the contents of a dermoid cyst can cause chemical peritonitis, which is an aseptic inflammatory peritoneal reaction.

8) Once a rupture of an ovarian cystic teratoma is diagnosed, immediate surgical intervention with prompt removal of a spontaneously ruptured ovarian cyst with thorough peritoneal lavage is required.

Received 12 January 2020

Accepted 15 January 2020

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Saulius Mikėnas, Gražina Drąsutienė,
Linus Andreika, Indrė Sakalauskaitė

BRANDŽIOS CISTINĖS TERATOMOS SPONTANINIO PLYŠIMO SUKELTAS CHEMINIS PERITONITAS: KLINIKINIS ATVEJIS IR LITERATŪROS APŽVALGA

Santrauka

Tikslas. Brandi cistinė teratoma (dermoidinė cista) – labiausiai paplitęs gemalo ląstelių navikas, sudarantis 10–25 % suaugusiųjų ir 50 % vaikų kiaušidžių navikų. Dermoidinių cistų etiologija lieka neaiški. Labiausiai tikėtina partenogenetinė teorija. Šiems navikams būdingas lėtas augimas ir asimptomė eiga, todėl jie nerečiau aptinkami kaip atsitiktiniai radiniai. Ligai nebūdingi specifiniai simptomai. Pagrindinė komplikacija – cistos apsisukimas (15 %), mokslinėje literatūroje aprašomos ir kitos komplikacijos: piktybiniai pokyčiai (1–2 %), infekcija (1 %) ar cistos plyšimas (0,3–2 %). Ilgalaikis spaudimas nėštumo metu, cistos apsisukimas su kraujotakos sutrikimu ar tiesioginė trauma yra pagrindiniai dermoidinės cistos spontaninio plyšimo rizikos veiksniai, galintys sukelti ūminį ar lėtinį peritonitą. Dažnai šių navikų diagnozė nustatoma tik po chirurginės naviko operacijos, nes radiologiniai tyrimo metodai (ultragarsinis tyrimas, kompiuterinė tomografija ir magnetinio rezonanso tomografija) negali tiksliai ir patikimai atskirti gerybinės ir piktybinės patologijos.

Metodai ir medžiaga. Pristatome 35 metų pacientės klinikinį atvejį. Ligonė atvyko į priėmimo skyrių dėl tris dienas besitęsiančio pilvo skausmo, plintančio į kojas. Iš anamnezės buvo žinoma, kad pacientė prieš mėnesį gimdė natūraliais takais. Moteris teigė iki tol neturėjusi ginekologinių operacijų ir nesirgusi ginekologinėmis ligo-

mis. Pilvo apčiuopos metu nustatytas jautrumas pilvo apatinėje dalyje be pilvo raumenų įtempimo ir pilvaplėvės dirginimo reiškinių. Transvaginalinio ultragarsinio tyrimo metu dubenyje buvo nustatytas daugiakamerinis cistinis 16 × 10 cm diametro darinys (kiaušidės cista), laisvo skysčio dubenyje nerasta. Nesant ūminės ginekologinės patologijos, pacientei buvo rekomenduotas planinis operacinis gydymas – laparoskopinė kiaušidės cistos šalinimo operacija. Dėl sustiprėjusio pilvo skausmo (VAS 7/10), pykinimo ir vėmimo kitą dieną pacientė pakartotinai kreipėsi į priėmimo skyrių. Pilvo apčiuopos metu buvo nustatyti peritonito simptomai. Ultragarsiniu gimdos ir kiaušidžių tyrimu nustatyti pokyčiai abiejose kiaušidėse (cistiniai dariniai), taip pat didelis kiekis laisvo skysčio dubenyje. Pacientei buvo skubiai atlikta laparoskopinė operacija: pašalinta kairės kiaušidės cista ir atlikta dešinės ooforektomija (dėl didelių matmenų dešinės kiaušidės naviko, nesant sveiko dešinės kiaušidės audinio), kruopščiai išplauta pilvo ertmė. Po operacijos skirtas antibakterinis gydymas, lašinės skysčių infuzijos, skausmą malšinantys vaistai, tromboembolijų profilaktika. Septintą parą po operacijos pacientė buvo išrašyta iš ligoninės ambulatoriniam gydymui.

Rezultatai ir išvados. Ultragarsinis tyrimas yra pirmoji priemonė pilvo ir dubens organų būklei įvertinti. Į pilvaplėvės ertmę išsiliejęs cistos turinys gali sukelti cheminį peritonitą – aseptinį pilvaplėvės uždegimą. Dermoidinės cistos plyšimo sąlygoto cheminio peritonito grėsmės atveju pirmas pasirenkamas gydymo metodas – skubi chirurginė operacija, kruopščiai išvalant pilvaplėvės ertmę.

Raktažodžiai: brandi cistinė teratoma, cheminis peritonitas, dermoidinė cista, pilvo ertmės lavažas, spontaninis plyšimas