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A case of ovarian goiter with ascites and significantly elevated CA125

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Abstract: *Struma ovarii (SO) refers to a type of monoblastic mature teratoma composed entirely or mainly of thyroid tissue, accounting for 1%-3% of all teratomas and 0. 3%-1. 0% of all ovarian tumors. At present, there is no complete diagnosis and treatment guide for the diagnosis and treatment of this disease, and the clinical misdiagnosis rate is very high, and the diagnosis is mainly based on clinicopathological diagnosis. The patient was a postmenopausal woman who underwent total hysterectomy combined with bilateral adannexectomy. The postoperative pathology was consistent with goiter, and CA125 decreased rapidly and ascites disappeared. Through case reports and literature review, it is hoped that clinicians can enhance their understanding of this disease and improve their diagnosis and treatment level.*

Keywords: *Mature cystic teratoma; Goiter ovarii; Ascites; CA125*

1. History and physical examination

The female patient, 56 years old, was admitted to hospital on 2022-04-13 due to "abdominal accumulation more than half a month, aggravated with abdominal distension for 1 day" as the chief complaint. The patient developed abdominal mass without obvious inducement half a month ago and was not paid attention to. 1 day ago, the patient felt aggravated abdominal accumulation accompanied by abdominal distension. Abdominal CT examination in the outpatient department of our hospital indicated that cirrhosis and ascites were considered. He has a history of varicose veins of lower limbs for more than 40 years. Specialty: Positive mobility dullness, mild edema of both lower limbs, no other abnormalities. Present symptoms: abdominal accumulation, dry mouth and bitter mouth, general fatigue, no fever, cough, no nausea and vomiting, poor diet, acid reflux heartburn, occasional shortness of breath, abdominal distension, no abdominal pain, poor night rest, normal bowel and urine.

2. Laboratory and auxiliary inspection

Epigastric CT: Cirrhosis and ascites were considered. The possibility of duodenal diverticulum was considered. Fecal routine: fecal occult blood (±);Urine analysis: urine white blood cells (+), urine protein (±), occult blood (±);There were no abnormalities in blood routine, liver A, liver E, infectious eight, liver function, kidney function, electrolyte, myocardial enzyme and glucose. Autoantibody series: anti-SSA /Ro60kD antibody positive (+), anti-SSA /Ro52kD antibody positive (+); Liver tumor: Golgi apparatus protein 73154. 10ng/ml;Liver fibrosis: hyaluronic acid 308. 76ng/mL, laminin 141. 21ng/mL, type III procollagen N-terminal peptide 15. 40ng/mL;Six coagulation items: plasma fibrinogen content determination (Fbg) 4. 20g/L, D-dimer 1. 78mg/L, fibrinogen degradation products 7. 7mg/L;Homocysteine 19. 8umol/L;Immunoglobulin quantity: immunoglobulin G18. 54g/L, immunoglobulin M0. 67g/L, immunoglobulin A3. 19g/L, complement C31. 34g/L, complement C40. 30g/L;Sedimentation rate (mm/H)26. 00mm/h;Anti-cardiolipin antibody and CRP were not significantly abnormal. Medium liquid dark areas were observed in the abdominal cavity, which were distributed as follows: 33mm perihepatic surface, 35mm perisplenic surface, 29mm left abdominal cavity, 39mm right abdominal cavity, 74mm lower abdominal cavity. There was a cyst solid mass about 107×65mm in the pelvic cavity. (Reexamination 7 days later) low-medium fluid dark areas were

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observed in the abdominal cavity, which were distributed as follows: perihepatic 56mm, right abdominal 29mm, right iliac fossa 60mm, and left iliac fossa 64mm. (Reexamination 14 days later) A small number of liquid dark areas were observed in the abdominal cavity, which were distributed as follows: 6mm around the liver, 31mm in the left abdomen, 40mm in the right abdomen, 69mm in the lower abdominal cavity. Gastroscopy: chronic superficial gastritis.

(Transferred to gynecology for further treatment) gynecological examination: vulva: married already delivered;Vagina: smooth, mucous membrane red;Cervix: atrophied, smooth;Uterine body: atrophy, palpation unclear;Accessories: The right attachment touched a cyst solid mass about 10×9×8cm in size, smooth surface, good motion, no tenderness, no mass and tenderness in the left attachment area. Gynecological B-ultrasound: the uterus was anterior, the size was about (40+25) ×46×29mm, the boundary was clear, the shape was regular, the parenchymal echo was uniform, the uterine line was centered, the endometrial thickness was 5mm, the echo was low, the intrauterine probe and the circular iUD was strong, and the position was normal. The size of the right ovary was 76×58mm, the volume increased, the internal echo was uneven, and the internal changed like a honeycomb. In addition, 44×39mm echoless sac dark area was detected, peripheral recording and blood flow signal. Hysterorectal depression and liquid dark area of 80×38mm. Diagnosis: Postmenopausal uterus, normal position of IUD, cystic solid mass of right ovary -nature undetermined. β-HCG:3. 42IU/L;Tumor markers:CA125:748. 00U/ml, CA19-9:7. 73U/mL, HE463. 20Pmol/L;AFP:2. 42ng/ml, CEA:0. 65ng/ml, SCC:0. 50ng/ml. CA125:519. 60U/ml.

3. Diagnosis and Treatment process

Hepatology department routinely provides symptomatic support treatment such as liver protection, diuresis, prevention of complications, etc. She was transferred to the department of gynecology and performed IUD removal + posterior fornix puncture. About 80ml of light yellow clear ascites were extracted. Postoperative ascites were examined: no malignant tumor cells were found. Ascites biochemistry: lactate dehydrogenase 115U/L, total protein 59. 2g/L. DNA quantitative cytology test, liquid based cytology test: no abnormal;Pelvic MRI: Pelvic malignant tumor, considered to be the source of ovarian serous cystadenous ca, pelvic effusion;Bilateral inguinal region and paravascular iliac lymph nodes slightly enlarged;Myoma of posterior wall of uterine body. Chest + mid-abdomen CT showed localized interstitial fibrosis in the lower lobe of the right lung, enlargement of the left axillary lymph nodes, thickening of the left adrenal base. Considering hyperplasia, dynamic follow-up review was recommended. Venous ultrasound of both lower limbs: the left great saphenous vein widened in diameter with venous valve insufficiency;Varicose great saphenous vein of the right lower limb with venous valve insufficiency.

Preoperative diagnosis: (1) Right ovarian tumor - serous cystadenocarcinoma?; (2)Abdominal fluid accumulation during decompensation of cirrhosis; (3) Lower limb varicose veins; (4) Connective tissue disease? (5) Sjogren's Syndrome? (6) Chronic superficial gastritis. Exploratory laparotomy (total ovarian CA staging operation) was planned. Intraoperative exploration showed a large amount of abdominal ascites with light yellow color, attracting about 1000ml of ascites. On the right side, the cystic enlargement was about 10×8×9cm, the uterine atrophy was smooth, and the appearance of the left accessory was not obvious abnormal. The surface of liver, stomach, omentum and intestine was smooth without nodules. Pelvic lymph nodes and abdominal aortic lymph nodes were not enlarged. Combined with the patient's condition, the whole uterus and bilateral salpingectomy were performed after talking with the family. Rapid freezing pathology: (right ovary) The tumor consists of follicular thyroid tissue with cystic change, interstitial degeneration, calcification, and considered proliferative goiter. Ascites exfoliation cytology: few alien cells. Routine pathology: (right) Struma ovarii with hemorrhage, cystic change, and local calcification;(right) tubal tissue;Adenomyopathy of uterus;Endometrial atrophy;Chronic cervicitis with retention cyst formation;(left attachment) shows ovarian and fallopian tissue, (right attachment) no obvious abnormality. Postoperative D1: Seven items of armour strength: no abnormalities were found, TSH0. 67uIU/ml, T31. 44nmol/L, T4121. 70nmol/L. Postoperative D5: CA125:207. 90 U/mL;Gynecological ultrasound: no abnormality;Thyroid ultrasound: diffuse thyroid lesion, please combine thyroid function;Slightly hyperechoic nodules in the left lobe of the thyroid gland were classified into 3 categories of TI-RADS. Postoperative D14: CA125:60. 60U /mL. CA125 was negative 1 month after surgery.

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4. Characteristics of cases

(a) With ascites and exudate, CA125 increased significantly;CA125 decreased with the decrease of abdominal water volume. After surgery, ascites disappeared and CA125 gradually decreased to normal.

(b) Gynecological physical examination: the right attachment touched about 10×9×8cm cystic solid mass, smooth surface, good range of motion, no tenderness;

(c) Imaging features: unilateral cystic consolidation, intracellular components showed high signal T2W1 and low signal T1W1, limited diffusion on DWI was not obvious, no enhanced consolidation on enhanced scan, separation components showed low signal and no significant enhancement on enhanced scan. (see Figure 1-5)

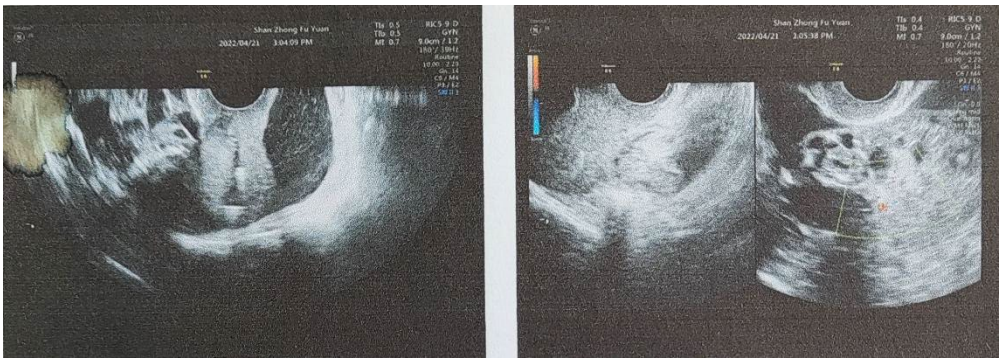

Figure 1: Transvaginal sonography

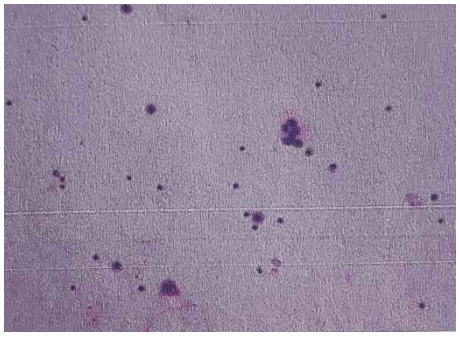

Figure 2: Peritoneal cytology

Figure 3: Postoperative thyroid ultrasound

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Figure 4: Tissue samples

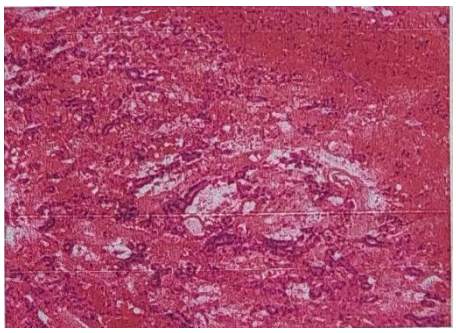

Figure 5: Pathological section

5. Discussion

Strumaovarii (SO) refers to a type of monoblastic mature teratoma composed entirely or mainly of thyroid tissue, accounting for 1%-3% of all teratomas and 0. 3%-1. 0% of all ovarian tumors [1]. Less than 5% are malignant [2]. The disease tends to occur at the age of 20-56, covering a wide range of ages, and very few patients are younger than 20 years old [3]. Perimenopausal patients are more common, and patients with thoracoabdominal hydrops are called pseudo-Meigs syndrome, which is rarely seen clinically, and abnormal elevation of serum CA125 is even rarest [4]. The clinical criteria of this disease are: thyroid tissue in pathological tissues is > 50%, or thyroid tissue is not more than 50%, but combined with hyperthyroidism symptoms [5]. Preoperative diagnosis of this disease is difficult, mainly due to its low clinical incidence and lack of atypia. Most patients are accidentally found to have abdominal distension, ascites, adjunct mass found in gynecological examination, or increased CA125 in admission [6], which cannot be distinguished from ovarian cancer through imaging and other clinical auxiliary examinations.

At present, there are no clear clinical guidelines for the treatment of this disease, and the postoperative diagnosis is the gold standard for this disease. The vast majority of patients are treated with ovarian cancer by staging operation, exploratory laparotomy, and intraoperative freezing is found to be SO. For young patients, simple tumor resection is often used in clinical treatment to preserve fertility function. In postmenopausal patients, the uterus combined with bilateral adnexectomy may be performed.

MRI findings of most struma ovarii showed clear boundary cystic mass, very low signal on T2W1 sequence, high signal on T1W1 [7], and low signal on T1WI and high signal on T2WI in cystic cavity [8]. The different concentrations of thyroxine, thyroglobulin and other substances in the cyst fluid lead to complex signals of the cystic components [9]. When the cyst fluid is highly concentrated and presents a high viscosity, gluey substance, low signals on T1WI and even lower signals on T2WI in MRI are most characteristic [10]. Preoperative imaging diagnosis of this patient was as follows: cystic components showed high signal of T2W1 and low signal of T1W1, combined with intraoperative freezing: thyroid follicular tissue composition accompanied by cystic change, interstitial glass change and calcification;As well as postoperative routine pathology: struma ovarii with hemorrhage, cystic change, local calcification. The reasons may be: (1) contain dilute serous fluid in the sac;② the composition of mucus is diverse;③ Hemorrhage in SO [7, 11].

CA125 is a glycoprotein derived from coelom epithelial cells and can be expressed in normal

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tissues. It has been recommended as a marker for early ovarian epithelial cancer, but it is characterized by high sensitivity and poor specificity. Compared with a single detection, the growth trend and speed of CA125 may have more clinical value [12]. There is no conclusive explanation for the apparent elevation of CA125 in benign tumors. Some scholars believe that the reasons are caused by biochemical factors, tumor mechanical stimulation, lymphatic obstruction and increased permeability, and peritoneal interstitial edema [13], which is more closely related to omentum and peritoneum [14]. Patients complicated with peritoneal effusion are easy to cause the increase of CA125 level, which may be due to the stimulation of ascites, but there is no obvious parallel relationship between abdominal water volume and the increase of CA125 level [15, 16], and most patients' CA125 decreased to normal after surgical treatment, which is also the case in this patient. It is worth noting that the patient in this case was indicated to have cirrhosis by imaging, but the liver function index was normal. 5 days after surgical treatment, ascites disappeared, which ruled out that ascites was caused by cirrhosis. After routine treatment such as liver protection and diuresis in the Department of Hepatology, abdominal water volume was less and CA125 levels were significantly reduced. The dynamic changes of CA125 before surgery were not monitored in previous reports. The reason for this phenomenon may be that the reduction of abdominal water volume leads to the reduction of the amount of stimulation, and the liver degradation of CA125 is enhanced after liver protection treatment. It may also indicate on the other hand that the decreasing trend and speed of CA125 or relatively large fluctuations have reference value for the diagnosis of SO. These possibilities await the reporting of a larger sample of cases.

At present, there is no conclusive explanation for the formation of ascites. Some scholars believe that it may be related to the leakage of tumor itself, peritoneal secretion, dysfunction of hypothalamic-pituitary goad axis, and release of inflammatory factors, as well as the stimulation of peritoneum by tumor and lymphatic reflux obstruction caused by tumor compression. Regarding the diagnosis of the patient's thyroid disease, it is a pity that thyroid function and thyroid ultrasound were not detected before surgery, and serology showed no abnormality in seven items of thyroid function 12h after surgery. Thyroid ultrasound examination on the fifth day after surgery showed:The patient's medical history was investigated. There was no history of thyroid diseases such as hyperthyroidism in the past. The reason may be the damage to the thyroid caused by long-term hormone secretion from the ovarian thyroid tissue.

In summary, when gynecological ultrasound indicates cystic consolidation in the adnexal region, accompanied by ascites and significantly elevated CA125, SO is easily misdiagnosed as ovarian malignant tumor, and only intraoperative freezing and postoperative pathology can suggest SO. Therefore, when the clinical findings of the above signs, we should refer to ultrasound, pelvic magnetic resonance, CT and other clinical examination means, including the most simple gynecological examination, clinical ovarian malignant tumors often show late signs, SO gynecological examination showed different signs with malignant tumors. At the same time, in this case, the change of abdominal water volume before surgery and the significant fluctuation of CA125 may also be a hint. Long-term follow-up should also be paid attention to after surgery.

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