Case Report

Ruptured Cystic Teratoma Associated with Mucinous Cystadenoma in a Pregnant Woman

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Abstract

Mature cystic teratoma is the most common ovarian neoplasm. However, mucinous cystadenoma with teratoma has been very rarely reported in literature. This case report, which is very rare, describes a clinical entity not previously reported in literature. A 34-year-old pregnant woman presented in the 23rd gestational week with severe right lower quadrant pain. She was diagnosed with acute abdomen and was then treated surgically. During the surgical intervention, a spontaneously ruptured mass was detected in the right ovary. This was reported histopathologically as a mature cystic teratoma in collision with mucinous cystadenoma. To the best of our knowledge, this case report is the first to have identified a ruptured mature cystic teratoma in collision with mucinous cystadenoma in a pregnant woman.

Keywords: *Acute abdomen, mucinous cystadenoma, ovarian neoplasm, pregnant woman, right lower quadrant pain, ruptured mature cystic teratoma*

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Introduction

Mature cystic teratoma, which is usually seen between the ages of 20 and 30 years, is the most common ovarian neoplasm, constituting 22%–40% of all ovarian tumors.[1] This also makes it the most common tumor observed during pregnancy.[2,3] Although mature cystic teratoma is generally asymptomatic, it may present as an acute abdomen due to torsion (16%), spontaneous rupture (1.3%), and infection (1.2%).[1,4] During pregnancy, there is a significantly increased risk of complications such as rupture, torsion, infection, and malignant degeneration, and when the cyst ruptures, the cystic component can incite a chemical or granulomatous peritonitis or pseudomyxoma peritonei with an estimated incidence of approximately 2 per 10,000 laparotomies or one per million population per year, with women mostly affected (two to three times more frequently than men).[2,3,5,6] Although mature cystic teratoma in collision with some other malignancies such as mucinous cystadenoma[1,7] has been reported in literature, there appears to be no case in literature, which describes a ruptured mature cystic teratoma coexisting with mucinous cystadenoma in a pregnant woman. In this report, the case described is of a pregnant woman who presented with a ruptured mature cystic teratoma; a collision tumor was diagnosed histopathologically

Case Report

A 34-year-old pregnant woman in the 23rd week of gestation presented at the Outpatients Clinic of the General Surgery Department with symptoms of right lower quadrant pain of inflammatory character and loss of appetite for 2 days. There was no history of previous surgery. On physical examination, there was rebound tenderness in the right lower quadrant. Abdominal ultrasonography revealed dilatation in the bilateral renal calyceal system which was more prominent on the right side and in the pelvis (grade 1 hydronephrosis). No free fluid was observed in the abdomen. Although the appendicular wall was not edematous, echogenicity was increased at the periappendicular fatty tissue of the appendix representing infiltration. Furthermore, the appendix could be compressed by the ultrasonography

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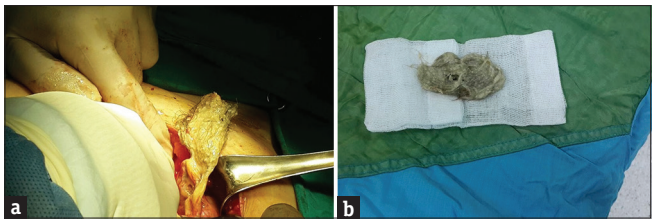

Figure 1: (a) The ruptured cystic lesion and hair within the cyst seen during surgical intervention. (b) The macroscopic image of the ruptured cystic component

probe, and the diameter of the appendix was measured as 6.4 mm at the distal end. Doppler ultrasonography revealed no increase in vascularity of the appendix wall. The laboratory test results were as follows: leukocyte count: 13,100/μL, hemoglobin: 10.9 g/dL, hematocrit: 32.9, aspartate amino transferase: 18 U/L, alanine amino transferase: 13 U/L, urea: 20.75 mg/dL, and creatinine: 0.45 mg/dL. The urine analysis was found to be in the normal range. The patient was hospitalized in the general surgery clinic with diagnosis of acute abdomen. Prior to surgical intervention, the gynecologist examined the patient and the gynecological ultrasonography images, but did not report any abnormality related to obstetrics or gynecology.

Due to the acute abdomen findings, the patient was admitted for emergency surgery. A McBurney incision was made under spinal anesthesia, and dense exudate-type fluid of approximately 50 mL in the right lower quadrant of the abdomen was seen. Upon exploration of the right ovarian region, a hairy glomus surrounded by exudates-type fluid and a ruptured ovarian cyst measuring 3 × 2 cm in the right ovary were discovered [Figure 1]. These findings suggested that this ovarian tumor may be a dermoid tumor. After the surgical area was cleaned, the mesenteric tissue of the appendix was observed to be edematous. The small intestines, colon, and stomach were seen to be normal. The ovarian cyst with its capsule was removed by the gynecologist, and then following appendectomy, the right adnexal region was observed for a few minutes, hemostasis secured, and a drainage tube was placed into the surgical field. The postoperative period was uneventful and the patient was discharged on the fourth day postoperatively.

The histopathological examination revealed that the cyst wall was paved with squamous epithelium within keratinous material and sebaceous glands [Figure 2a], and the cystic component was paved with a single row of benign mucinous epithelium in different areas of the specimen [Figure 2b]. These findings were reported histopathologically as "mature cystic teratoma in collision with mucinous cystadenoma"

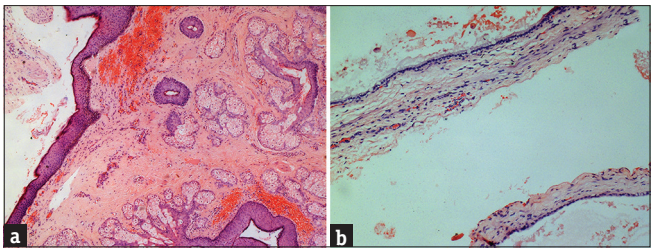

Figure 2: (a) The cyst wall paved with squamous epithelium within keratinous material and sebaceous glands in the histopathological specimen (H and E × 40). (b) The cystic component paved with benign mucinous epithelium in a single row was seen in the specimen (H and E × 100)

[Figure 2a and b]. Acute appendicitis was also revealed in the histopathological examination.

Discussion

The symptoms of right lower quadrant pain with tenderness and rebound without defense were thought to be related to acute abdomen in the present case. In clinical practice, it has been known that acute appendicitis, gastroenteritis, pelvic inflammatory disease, ruptured ectopic pregnancy, or ruptured corpus luteum can mimic these clinical signs and symptoms. Pelvic ultrasonography with color Doppler is the mainstay of diagnosis,[8] although in this case it did not reveal any intrabdominal abnormality in this pregnant woman except the finding of increased echogenicity in the periappendicular fatty tissue. Moreover, the gynecologist could not detect any obstetric or gynecological abnormality on the gynecological ultrasonography scan. The histopathological examination made with the consideration of diagnosis of adnexal cystic tumor revealed a mature cystic teratoma with mucinous cystadenoma. Moreover, acute appendicitis was also observed on pathological examination. During the surgery, it was not possible to determine which clinical entity, either teratoma or acute appendicitis, had caused the right lower quadrant pain.

In literature, it has been reported that mature cystic teratoma is thought to be derived from primordial stem cells.[2] The cyst is covered by an epidermis similar to the epithelial layer and consists of endo-, ecto-, or mesoderm derivatives such as hair, teeth, bone, or fat tissue.[1] Although it is generally asymptomatic, in pregnancy there is a significantly increased risk of complications such as chemical or granulomatous peritonitis associated with its rupture, torsion, infection, infarction, fetal intrauterine growth impairment, fetal malpresentation, preterm labor, and dystocia preventing normal vaginal delivery, and malignant degeneration.[2,3,6,9] Therefore, in literature, surgical excision of mature cystic teratoma is strongly recommended as soon as possible to prevent

complications especially in pregnant cases, and if the cyst is larger than 6 cm.[10] It is recommended that if a mature cystic teratoma is accidentally detected during the first trimester of pregnancy, surgical excision should be performed in the 14th–16th gestational weeks to prevent corpus luteum injury, and if it is diagnosed between the 16th and 22nd weeks, the surgery should be planned as soon as possible. However, if it is detected after the 22nd week, the surgical treatment should be delayed until after birth.[2,3] During the surgical procedure, it is recommended that the surgical area be washed with sufficient saline solution to minimize peritonitis and associated sequelae if there is cyst rupture.[2,11-13] The present case was in the 23rd gestational week of pregnancy, and the adnexal tumor or enlargement which could not be diagnosed before in routine follow-up examinations was complicated with the findings of acute abdomen. Therefore, to explore the etiological entity and to avoid the morbidity and/or mortality risks of acute abdomen, she had to be operated on urgently. After removing the tumor and its cystic component, to avoid the complications described above, the surgical field was washed out using a sufficient volume of saline solution, and a sump drainage (triple lumen 24F) was placed in that surgical area.

The origin and/or pathogenesis of mucinous cystadenoma shas not yet been fully identified. Various theories suggest that (i) most mucinous tumors associated with a teratoma are derived from the teratoma but that on occasion they may be collision tumors; (ii) mucinous cystadenomas can be a consequence of ectopic ovarian tissue or teratoma; or (iii) mucinous cystadenomas may originate from a peritoneal invagination, and therefore may form a cyst with further mesothelium mucinous metaplasia.[9,14] In the previous reported cases of mature teratoma ovary in collision with mucinous cystadenoma, it was believed that mucinous cyst adenoma originated from the epithelium of mature cystic teratoma.[14,15] However, in a case report by Naim *et al.*, an oval small size teratoma was described, which did not have mucinous epithelial constituents and the boundary could be differentiated anatomically from the mucinous lining of cystadenoma. It was concluded that this teratoma possibly originated from the ovarian germ cell remaining in the wall of the cyst adenomatous neoplasm or it resulted from the pathenogenesis of ovum evolved in a graafian follicle which failed to rupture, together with the mucinous adenoma cysts by "epithelial metaplasia" of the follicular lining.[16]

In conclusion, mature cystic teratoma coexisting with mucinous cystadenoma has been very rarely reported in literature,[7,16] and to the best of our knowledge this combination in a pregnant woman has not been previously reported in literature. Although it could not be substantiated in this patient whether the mucinous cyst adenoma originated from the epithelium of the mature cystic teratoma, this case report is the first report to have identified a ruptured mature cystic teratoma coexisting with mucinous cystadenoma in a pregnant woman.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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