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Case Report

Giant juvenile granulosa cell tumor torsion: a case report

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Abstract: This report describes a 17-year-old patient with ovarian granulosa cell tumor torsion, which is rare. The patient presented with lower abdominal pain for 1 day after defecation, accompanied by nausea and vomiting. Gynecological examination revealed a palpable mass of about 150 mm × 130 mm in the uterus. The mass had high surface tension, tight cysts, well-defined borders, and marked tenderness at its pedicle. Gynecological B-ultrasonography showed that the uterus was not compressed, and a mixed echo of about 152 mm × 112 mm was seen in the pelvic cavity. The initial diagnosis was ovarian tumor torsion. After explaining the condition to the patient's family, an emergent open resection of left ovarian tumor was performed. The postoperative pathology report indicated juvenile granulosa cell tumor. The patient's body temperature was normal, and there was no infection or thrombosis after operation.

Keywords: Acute abdomen, juvenile granulosa cell tumor, laparotomy, ovarian cyst torsion, ovarian granulosa cell tumor

Introduction

Statistically, about 10% of ovarian tumors are complicated by torsion [1]. Ovarian cyst torsion is one of the common gynecological causes of acute abdomen, which is an emergency [2-5]. Therefore, once an ovarian cyst torsion is suspected, immediate surgery is required. Recently, case reports have described the color of the ovary as purple and black intraoperatively, which may not necessarily represent ovarian necrosis, thus the ovary should be preserved as much as possible. Ovarian tumor torsion is mostly from benign tumors, with the most common being simple cysts, followed by dermoid cysts and serous cystadenoma. Granulosa cell tumor (GCT) is a rare tumor, accounting for about 2-5% of all ovarian tumors. Meanwhile, juvenile granulosa cell tumor (JGTC) is a rare GCT, mainly seen in pre-adolescent women and women under the age of 30 [6]. Here, we report a case of ovarian torsion of granular cell tumor in a pubertal-aged girl.

Case report

A 17-year-old Chinese girl presented to the emergency department complaining of acute lower abdominal pain. The patient complained of lower abdominal pain for one day after defecation, associated with nausea and vomiting. The patient had no diarrhea, no constipation, no hematochezia, no fever, no dizziness, and no history of syncope. She had menarche at 14 years old, with regular menstrual periods of 5 days, menstrual volume was normal and with no significant history of dysmenorrhea. The patient was sexually active, but no had birth history or abortion history. No ovarian cysts were identified in the previous examination, with no family history of tumor, no history of smoking, and no long history of drug use. As indicated by the physical examination, the abdomen was bulging, like a woman at 5 months of pregnancy, with abdominal tension and significant tenderness. During a combined vaginal abdominal examination, a huge mass was felt in the pelvic cavity above the uterus. The upper margin of the lump reacheed the level of the navel and the anterior axillary line, with great tension and obvious tenderness. There was significant tenderness in the pedicle of the left proximal pelvic tumor. Ultrasound examination indicated that uterine compression was unclear, with a mixed

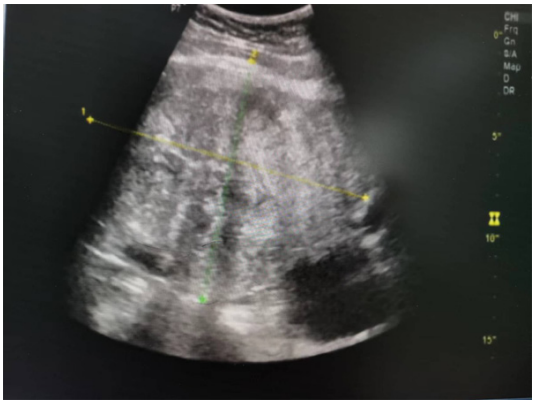

Figure 1. Ultrasound examination.

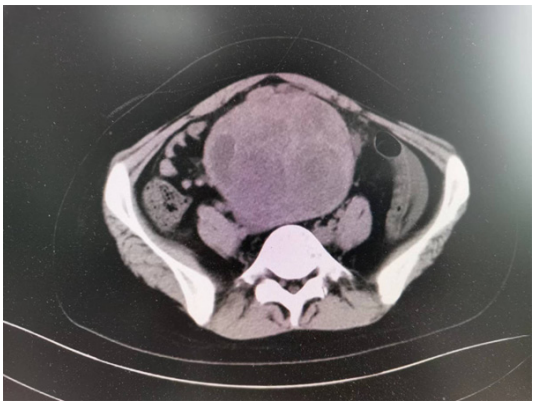

Figure 2. Computed Tomography examination.

echo of about 152 mm × 112 mm in the pelvic cavity. A liquid dark area was visible in the pelvic cavity, and the depth was about 33 mm (as shown in **Figure 1**). Subsequently, computed tomography (CT) examination showed a mass and other low mixed density lesion in the pelvic cavity with the maximum cross-sectional area about 122 mm × 106 mm; the boundary was clear, part of it was close to the uterus, and the pelvic space was occupied. It was considered that the mass may be attached at some point (as shown in **Figure 2**).

Given the patient's clinical symptoms, ovarian tumor torsion was considered. The patient and her guardians were informed about the condition, and then an emergent open resection of left ovarian tumor was performed after obtaining their consent. Emergency laparotomy revealed that the surface of the left ovary was purple-black, the longest and widest part was about 150 mm × 130 mm, the surface was smooth, and there was no adhesion to the surrounding tissues. The left fallopian tube and ovary were twisted 720° clockwise. Intraoperative examination of the uterus and the right fallopian tube and ovary revealed no abnormalities. A little bloody effusion was seen in the retrorectal depression of the uterus (as shown in **Figure 3**). Subsequently, a left-sided ovarian cystectomy was performed after detorsion of the left adnexa. During the operation, the surgical incision film was protected, and there was no tumor rupture or capsule rupture. The patient's body temperature was normal after operation, and there was no infection or thrombosis. The incision healed well, and the sutures were removed normally one week after the operation.

The postoperative pathology report suggested juvenile granulosa cell tumor with infarction (**Figure 4**). Immunopathological examination confirmed juvenile granulosa cell tumor with vimentin (+), CD99 (+), CD56 (+), p53 focus (+), S-100 (-), inhibin (-), CD34 (-), calretinin (-), WT1 (-), and Ki-67 20% (+), as shown in **Figure 5**.

Discussion

Torsion of ovarian neoplasms is a gynecological emergency that occurs in women of all ages, but is less common in prepubertal and postmenopausal women [2-5]. For patients with abdominal pain as the main clinical presentation, accompanied by nausea or vomiting, ovarian tumor torsion should be considered first after ultrasound or CT indicating an ovarian mass. According to relevant literature statistics, the correlation between these examination results and tumor torsion is up to 85% [7]. If patients miss the best time for treatment, it can lead to ovarian loss and infertility [8]. Therefore, once the ovarian tumor is evaluated to have torsion, immediate surgical exploration is required. However, there is no unified standard on whether an ovary with torsion should be removed. Multiple studies have shown that even black or purple ovaries may retain ovarian function after torsion is corrected [8-11]. It has been reported that with the extension of time, the retention rate of the twisted ovary decreases [9, 12], so once torsion is determined, surgical exploration should be performed as early as possible to reduce the risk of ovarian loss.

It was previously thought that a torsed ovary should be excised, mainly because of postoperative infection or a malignant tendency [13,

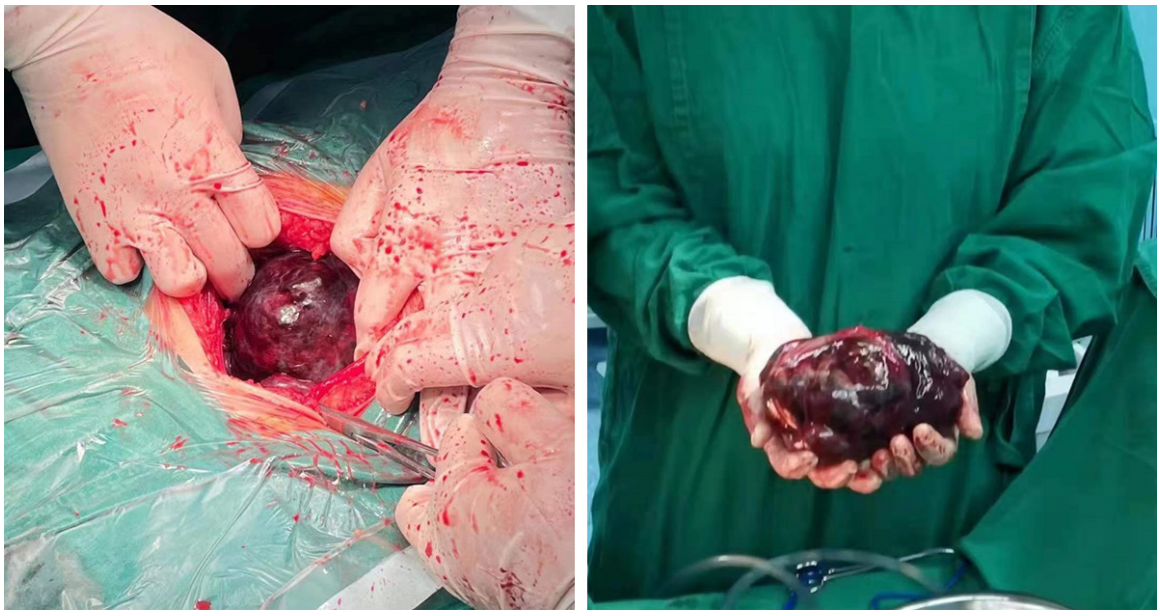

Figure 3. Laparotomy revealed a purplish black surface of the left ovary, about 150 mm × 130 mm in size, and smooth surface.

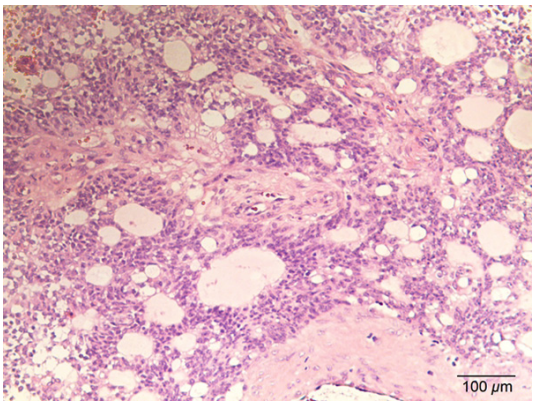

Figure 4. Hematoxylin-eosin staining demonstrated granulosa cell tumor with infarction (Magnification: 10 × 10).

14]. However, in a 5-year retrospective study of torsion of ovarian cysts, simple cysts were found to be the most common, followed by dermoid cysts and serous cystadenomas, and none of these patients developed a malignancy [6].

GCT is a rare tumor. Adult granulosa cell tumor (AGCT) is most common in women aged 50-55, while JGCT is most common in young women younger than 20 [15]. Due to the different ages of onset, the clinical manifestations are also different. AGCT mostly has typical postmenopausal bleeding symptoms, while JGCT has typical precocious puberty symptoms [15]. AGCT is a low-grade malignant ovarian tumor [16], but the age of onset and clinical symptoms cannot completely distinguish AGCT from JGCT. Due to the rarity of GCT, there are no unified imaging standards for the diagnosis of GCT. In this case, GCT was confirmed by pathology. Typical pathologic hallmarks of AGCT are Call-Exner bodies and nuclear grooves. JGCT is characterized by a well-demarcated tumor, formation of papillary structures, numerous follicle-like structures, thin eosinophilic secretions, and no Call-Exner bodies or nuclear grooves [17]. However, the follicular structure is not unique to JGCT. Similarly, some AGCTs may not have Call-Exner bodies [15]. In such cases, other pathological manifestations need to be identified. It is reported that small hair follicle structure can be seen in JGCT and can also be used to identify JGCT. The differential diagnosis of AGCT and JGCT is very important for prognosis.

There are no reported cases of torsion of granulosa cell tumors in the literature. The peculiarity of our case is the torsion of a giant granulosa cell tumor that occurred in an adolescent. The imaging in this case lacked specificity, the nature of the tumor before surgery was unknown, and a definitive diagnosis of the patient depended on pathology. Therefore, in

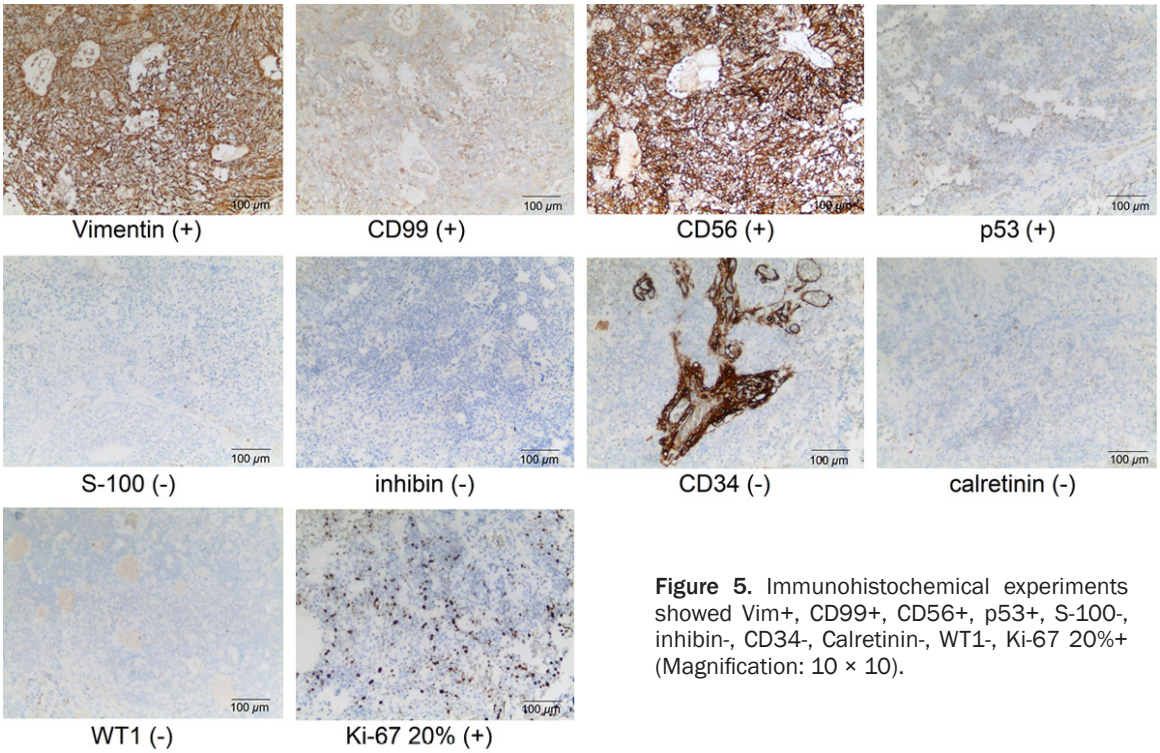

Figure 5. Immunohistochemical experiments showed Vim+, CD99+, CD56+, p53+, S-100-, inhibin-, CD34-, Calretinin-, WT1-, Ki-67 20%+ (Magnification: 10 × 10).

ovarian tumor torsion surgery, it is necessary to carefully protect the incision and surrounding tissues, avoid rupture of the tumor capsule during the operation, and carefully explore the surrounding tissues. For patients with ascites, the ascites should be collected for exfoliated cell examination. The surgical method that is most beneficial to the patient should be adopted to avoid the risk of reoperation. The GCT patient in this article has a good prognosis and the patient's fertility was preserved, so further efforts are needed to improve the diagnosis of GCT by imaging.

Conclusion

In this case, a combined vaginal-abdominal examination was performed first, and a large palpable mass was found in the pelvis. There was obvious tenderness at the left near the pelvic tumor pedicle. Ultrasound examination and CT examination results showed suspected torsion but could not determine whether it was a malignant tumor. Postoperative pathologic examination showed granulosa cell tumor. Therefore, it is necessary to improve the diagnostic ability of imaging for benign and malignant tumors.

Disclosure of conflict of interest

None.

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