



Left Paratubal Cyst Torsion Mimicking Malignancy on Computed Tomography

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Taking into account the importance of fertility preservation, an accurate diagnosis of the nature of a pelvic mass in girls is crucial to the determination of the extent and procedure of surgery. Although a detailed preoperative imaging evaluation is mandatory for assessing the nature and size of a pelvic lesion as well as its relationships with the surrounding organs, it can only offer a preliminary diagnosis that needs to be correlated with the clinical picture. Here we reported a 14-year-old girl presenting with a huge pelvic mass for which preoperative abdominal and pelvic computed tomography suggested ovarian malignancy with extension outside the pelvic cavity, ascites, and mesenteric lymph node involvement. Nevertheless, laparoscopic exploration revealed a picture of left paratubal cyst with torsion. Based on the clinical diagnosis, laparoscopic detorsion and left tubal cystectomy were performed with preservation of the uterus and the ovaries without lymph node dissection. Pathological analysis of the excised left adnexa later demonstrated pictures of hemorrhagic necrosis compatible with paratubal cyst torsion without evidence of malignancy. The patient, who recovered uneventfully and was discharged three days after operation, remained symptom-free without surgical complication on following up for one month. The findings of the present case suggested that, a sound clinical judgment, rather than imaging findings, is of utmost importance in determining the nature of a pelvic lesion that guides the subsequent procedure and the extent of surgery to avoid unnecessary exploration as well as to preserve fertility for benign lesions, especially in children and adolescents.

Key words: adnexal torsion, ovarian malignancy, fertility preservation, computed tomography, laparoscopic surgery

Introduction

Although it appears that the fallopian tubes merely serve as conduits to convey the egg cells for fertilization, various paratubal

and tubal abnormalities have been reported. Some may cause significant morbidity and even mortality.¹ Previous studies have identified paratubal cysts as one of the most common causes of adnexal torsion, which is the fifth most common gynecologic emergencies,² in

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the pediatric and adolescent populations.^{3,4} Albeit rare, a previous large-scale study revealed a malignancy rate of 1.2% among children and adolescents presenting with adnexal torsion.⁴ Therefore, although prompt surgical detorsion with restoration of blood supply to the adnexa is of utmost importance in avoiding irreversible adnexal damage and in preserving ovarian function especially these young populations,⁵ preoperative evaluation to rule out the possibility of malignancy is critical for determining the appropriate procedure and the extent of operation. Here we presented a young girl who posed a dilemma in which the preoperative pelvic computed tomography (CT) suggested malignancy which did not fit the picture on laparoscopic exploration.

Case Report

A 14-year-old girl without prior medical conditions or trauma presented to the emergen-

cy department with sudden aggravation of left lower abdominal pain for one day accompanied by vomiting of food twice. There was no family history of breast, ovarian, or other malignancies among known family members. The severity of pain was seven out of a score of 10 and was not relieved by oral analgesics. There was no change in appetite before the acute pain attack and her bowel habit was normal. According to the patient, the pain first started one week ago and was alleviated when she assumed a sitting position. She had a regular but prolonged menstrual cycle that came every three months with menorrhagia. Her last menstrual period was six days ago. Otherwise, her gynecological history was remarkable; menarche at 11 years old, gravida 0/para 0 without known sexual activity. She was afebrile with stable hemodynamics on arriving at the emergency department. Physical examination showed tenderness of the lower abdomen on palpation without muscle guarding. Pelvic examination

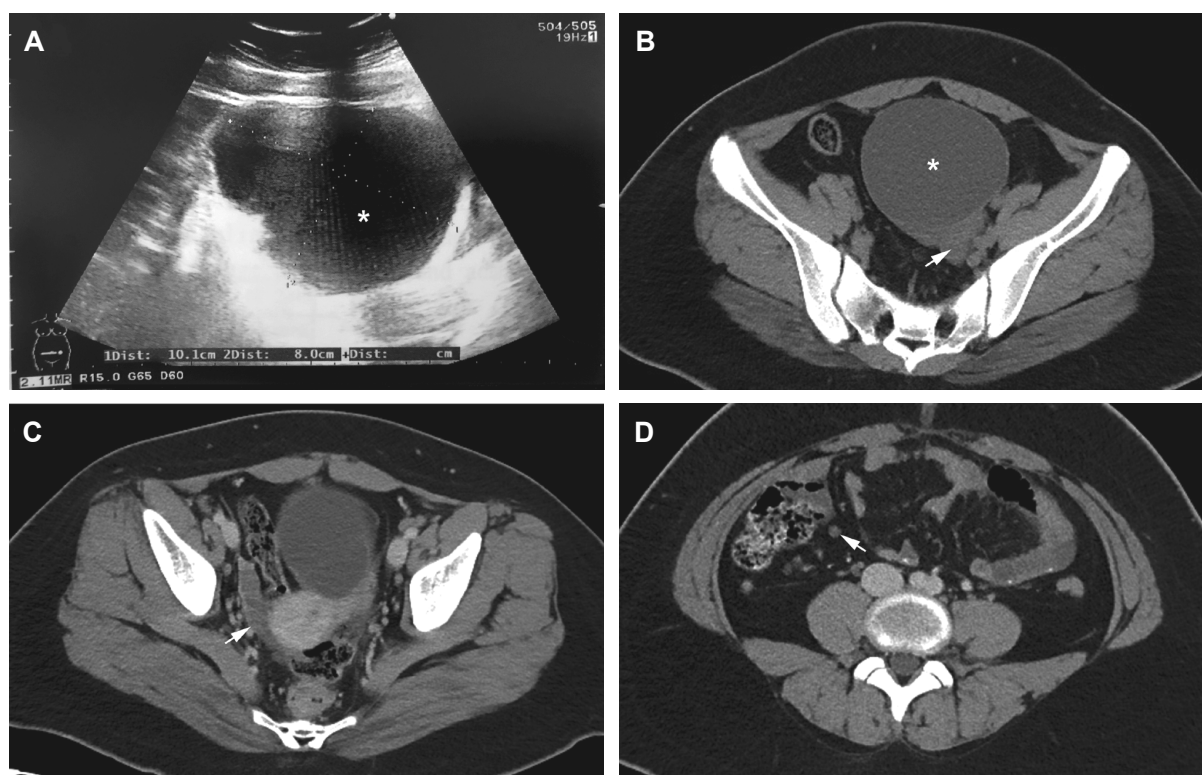


Fig. 1 (A) Abdominal ultrasound revealing a hypodense and homogenous left ovarian cystic lesion of size 10 cm × 9 cm without apparent septum. Abdominal and pelvic computed tomography showing (B) a cystic lesion of size 9.3 cm in the left parametrial region (asterisk) with (C) ascites (arrow) and (D) mesenteric lymph node enlargement (arrow), highly suggestive of pelvic extension of ovarian cancer.

was not performed because of her virginal status.

Hemogram showed mild leukocytosis with left shift (WBC: $11.35 \times 10^3/\mu\text{L}$, neutrophil: 87.7%, lymphocyte 10.3%). Serum biochemical studies demonstrated no abnormal findings (concentration of C-reactive protein: 8.54 mg/L, normally $< 10 \text{ mg/L}$) except for an elevated alanine aminotransferase concentration at 79 U/L (normal range, 7 – 56 U/L). Urinalysis gave normal findings. Pregnancy test was negative. Abdominal sonography revealed a hypodense and homogenous left ovarian cystic mass of size 10 cm \times 9 cm without apparent septum (Fig. 1A). The finding was supported by abdominal computed tomography (CT) that showed a 9.3 cm space-occupying adnexal lesion with mixed cystic and solid contents in left parametrial region (Fig. 1B). In addition to this finding, CT scan further revealed ascites (Fig. 1C) and suspicious pelvic extension of malignancy $> 2 \text{ cm}$ with mesenteric lymph node involvement (Fig. 1D) compatible with ovarian cancer with a preliminary image staging of T3cN1M0 [American Joint Committee on Cancer (AJCC), 8th Edition]. Differential diagnoses also included dysgerminoma and other germ cell tumors. However, the concentrations of serum tumor markers including alpha fetal protein, beta human chorionic gonadotropin (β -HCG), cancer antigen (CA) 125, CA 19-9, carcinoembryonic antigen (CEA), and lactate dehydrogenase (LDH) were all within normal limits. Although ovarian malignancy could not be ruled out, the preliminary diagnosis was left paratubal cyst with torsion for which emergent surgery was indicated.

Laparoscopic exploration of the pelvic cavity showed a large cystic structure attached to the fimbrium of the left fallopian tube compatible with the diagnosis of a paratubal cyst (Fig. 2A). Despite torsion of the left adnexa, there was no gross ischemic or necrotic change of the left ovary (Fig. 2B). Taking into consideration the young age of the patient, the rarity

of malignancy, and the absence of abnormal elevations in all tumor markers, laparoscopic detorsion and left tubal cystectomy were per-

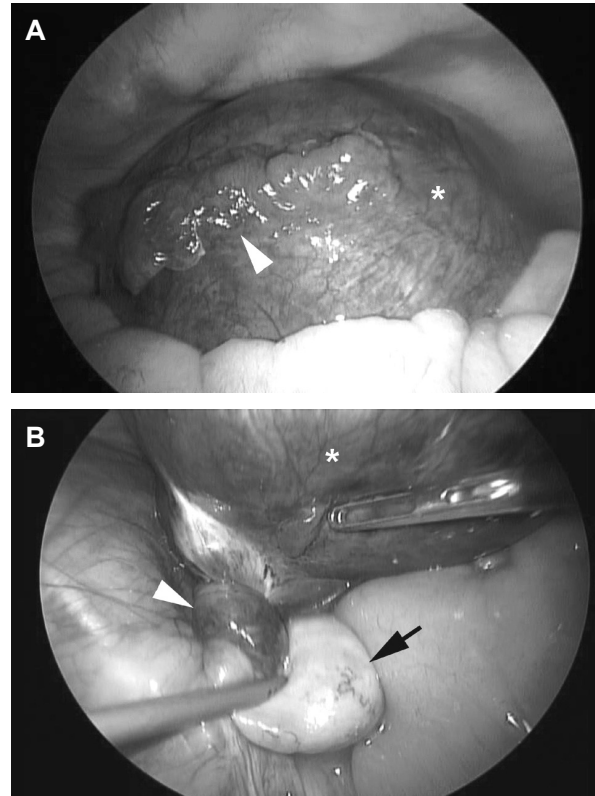


Fig. 2 Laparoscopic examination showing (A) a left paratubal cyst (asterisk) attached to the fimbrium (arrowhead) of the left fallopian tube, and (B) torsion of left fallopian tube (arrowhead). Note the left ovary (black arrow) and paratubal cyst (asterisk).

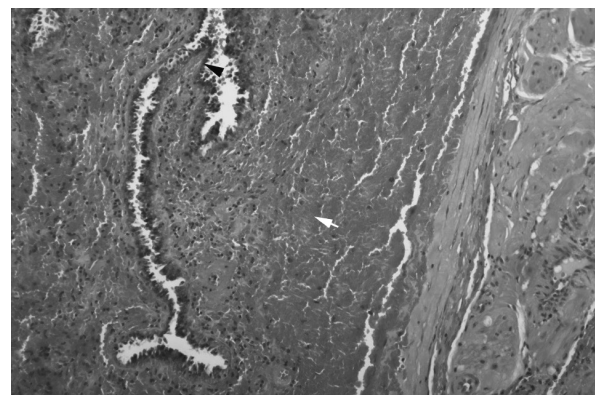


Fig. 3 Pathological analysis of left adnexal specimen from laparoscopic partial adenectomy demonstrating hemorrhagic necrosis (white arrow) compatible with paratubal cyst torsion without evidence of malignancy. Note the mucosal folds (plicae) typical of Fallopian tube (black arrowhead).

formed with preservation of the uterus and the ovaries without lymph node dissection. Pathological analysis of the excised left adnexa showed pictures of hemorrhagic necrosis compatible with paratubal cyst torsion without evidence of malignancy (Fig. 3). The patient's convalescence was uneventful and the patient was discharged three days after operation. She remained symptom-free without surgical complication on following up for one month.

Discussion

The blood supply of the ovaries comes from both the ovarian vessels that travel along the infundibulopelvic ligaments as well as the ovarian branches of the uterine arteries along the utero-ovarian ligaments that connect the ovaries to the uterus on both sides. Instead of being fixed to the pelvic wall, the adnexal tissue is suspended in the pelvic cavity through these ligaments. Therefore, a space-occupying lesion on the ovary or fallopian tube can act as a leading point that induces adnexal twisting, thereby compromising the ovarian blood supply.⁶ Adnexal torsion (AT) is a gynecologic surgical emergency because it can compromise blood supplies to the ovaries, leading to irreversible ovarian necrosis for which adnexectomy is the last resort.⁷ Although most ovarian torsion occurs in women of reproductive age and is less common (i.e., only 17.2%) in premenarchal girls as well as postmenopausal women,⁸ the potential fertility loss from surgery raises a lot of concerns among children and adolescents.⁵

Previous studies have reported age-related variations in the etiologies of adnexal torsion. For instance, compared with postmenarchal patients, their premenarchal counterparts were more likely to have a delayed diagnosis and develop ovarian necrosis as well as present without an associated adnexal mass.⁹ For adult patients, surgical treatment of adnexal masses,⁶ ovarian masses, and ovarian stimulation for in

vitro fertilization (IVF) are all risk factors for AT.⁷ A recent extensive review of up to 245 cases of AT in children and adolescents demonstrated that paratubal cysts and mature teratomas were the two most common causes of AT among children and adolescents.⁴ Consistently, another study also showed a close association between paratubal cysts and isolated tubal torsion in the pediatric population.³

A review of literature on AT in children and adolescent populations showed an average patient age of around 13.3³ and 12,⁴ which is compatible with that of our patient (i.e., 14). With respect to clinical manifestations, acute-onset lower abdominal pain is the most common symptom of ovarian torsion, followed by nausea and vomiting.⁶ The symptoms, therefore, are consistent with those of our patient. The fact that our patient sought medical help only after one week since the onset of pain was also in agreement with the common delay in patients' presenting for clinical evaluation of up to seven months as previously reported.⁶ Fever, which was absent in our patient, has been reported to be significantly associated with tissue necrosis.¹⁰ In terms of etiology, because ovarian tumors larger than 5 cm is known to be associated with the risk of ovarian torsion,⁶ the presence of a paratubal cyst of size more than 9 cm in our patient posed a definite risk to AT.

Regarding diagnosis-making, a recent meta-analysis on the accuracy of imaging modalities for AT identified ultrasound as a good first-line diagnostic test for the condition.¹¹ However, that study did not include CT in the analysis. Compared with ultrasound, CT has a higher sensitivity, specificity, and accuracy in the diagnosis of pelvic masses.¹² Nevertheless, in our case, it was the patient's pelvic CT findings that posed a dilemma to the gynecologist regarding the correct procedure as well as the extent of surgery to be performed.

The findings of our case suggested that, although CT may provide useful preoperative

information regarding the size of the lesion as well as the accompanying changes, a good clinical judgment is critical for determining the nature of the lesion. In our case, paratubal cyst torsion, rather than ovarian malignancy as suggested by pelvic CT, was the final diagnosis for which extensive pelvic dissection was avoided and the patient's fertility was successfully preserved based on clinical diagnosis.

References

1. Perlman S, Hertweck P, Fallat ME: Paratubal and tubal abnormalities. *Semin Pediatr Surg* 2005;14:124-34. doi: 10.1053/j.sempedsurg.2005.01.009.
2. Childress KJ, Dietrich JE: Pediatric ovarian torsion. *Surg Clin North Am* 2017;97:209-21. doi: 10.1016/j.suc.2016.08.008.
3. Webster KW, Scott SM, Huguelet PS: Clinical predictors of isolated tubal torsion: a case series. *J Pediatr Adolesc Gynecol* 2017;30:578-81. doi: 10.1016/j.jpag.2017.05.006.
4. Adeyemi-Fowode O, Lin EG, Syed F, et al: Adnexal torsion in children and adolescents: a retrospective review of 245 cases at a single institution. *J Pediatr Adolesc Gynecol* 2019;32:64-9. doi: 10.1016/j.jpag.2018.07.003.
5. Ssi-Yan-Kai G, Rivain AL, Trichot C, et al: What every radiologist should know about adnexal torsion. *Emerg Radiol* 2018;25:51-9. doi: 10.1007/s10140-017-1549-8.
6. Huang C, Hong MK, Ding DC: A review of ovary torsion. *Ci Ji Yi Xue Za Zhi* 2017;29:143-7.
7. Hasiakos D, Papakonstantinou K, Kontoravdis A, et al: Adnexal torsion during pregnancy: report of four cases and review of the literature. *J Obstet Gynaecol Res* 2008;34:683-7. doi: 10.1111/j.1447-0756.2008.00907.x.
8. Oelsner G, Shashar D: Adnexal torsion. *Clin Obstet Gynecol* 2006;49:459-63. doi: 10.1097/00003081-200609000-00006.
9. Prieto JM, Kling KM, Ignacio RC, et al: Premenarchal patients present differently: a twist on the typical patient presenting with ovarian torsion. *J Pediatr Surg* 2019;54:2614-6. doi: 10.1016/j.jpedsurg.2019.08.020.
10. Kruger E, Heller DS: Adnexal torsion. A clinicopathologic review of 31 cases. *J Reprod Med* 1999;44:71-5.
11. Wattar B, Rimmer M, Rogozinska E, et al: Accuracy of imaging modalities for adnexal torsion: a systematic review and meta-analysis. *BJOG* 2021;128:37-44. doi: 10.1111/1471-0528.16371.
12. Liu Y, Zhang H, Li X, et al: Combined application of ultrasound and CT increased diagnostic value in female patients with pelvic masses. *Comput Math Methods Med* 2016;2016:6146901. doi: 10.1155/2016/6146901.