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Clinical diagnosis and complications of paratubal cysts: review of the literature and report of uncommon presentations

Mine Kiseli · Gamze S. Caglar · Sevim Dincer Cengiz · Demet Karadag · Muserref B. Yılmaz

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Abstract

Introduction Paraovarian or paratubal cysts (PTCs) constitute about 10 % of adnexial masses. Although they are not uncommon; they rarely cause symptoms and are usually incidentally found. Actual incidence is not known. The symptoms occur when they grow excessively, or in case of hemorrhage, rupture or torsion.

Methods Here, literature review reporting the incidence, presentation and complications of PTCs is performed. Uncommon presentations of PTCs in three different cases, a giant PTC, torsion of PTC and borderline paratubal tumor, are also reported and discussed.

Results Ultrasonography, CT or MRI may be performed in preoperative evaluation; but none of these imaging techniques have specific criteria for diagnosis. So, in most cases misdiagnosis as an ovarian mass remains to be a problem. Conclusion Paratubal cysts can become extremely big before causing symptoms. Torsion is another urgent issue regarding PTCs, necessiating urgent surgery for preservation of the ovary and the tube. Although malignancy is rare,

borderline paratubal tumors have been reported in the

Keywords Paratubal cyst · Paraovarian cyst · Adnexial mass · Torsion · Borderline tumor

M. Kiseli · G. S. Caglar (⋈) · S. D. Cengiz · M. B. Yılmaz Department of Obstetrics and Gynecology, Faculty of Medicine, University of Ufuk, Mevlana Bulvarı No 86-88, Konya Yolu, Balgat 06520, Ankara, Turkey e-mail: gamzesinem@hotmail.com

D. Karadag

literature.

Department of Radiology, Faculty of Medicine, University of Ufuk, Mevlana Bulvarı No 86-88, Konya Yolu, Balgat 06520, Ankara, Turkey

Introduction

Paratubal cysts (PTCs) are located in the broad ligament between the ovary and the fallopian tube and constitute about 10 % of adnexial masses [1, 2]. They originate from mesothelium or are thought to be remnants of paramesonephric (Müllerian) and mesonephric (Wolffian) ducts. Paramesonephric duct remnants tend to occur more commonly within the broad ligament rather than at the fimbriated ends of the fallopian tube [3]. The size of the PTCs seen in the broad ligament might range from 1 to 8 cm in diameter [3]. Moreover, under the influence of hormonal factors they can reach huge sizes [4]. When a PTC is pedunculated and located near the fimbria of the fallopian tube, it is referred to as a hydatid cyst of Morgagni, which is usually smaller than 2 cm.

Paraovarian and PTCs are usually used synonymously. The incidence of PTCs is not clearly known, but in an Italian population an incidence of 29 per 1,000 (\sim 3 %) has been reported with a peak age of occurence in the third and fourth decades of life [5]. In pediatric and adolescent population, a much higher incidence of PTCs was reported (7.3 %) [6]. Paraovarian cysts have been reported in all age groups, beginning from premenarchial period up to menopause.

A certain diagnosis of PTCs is not usually possible preoperatively. Therefore, they are usually found incidentally during operative procedures for other indications. A preoperative misdiagnosis as true ovarian cysts is very common and creates a major problem. Barloon et al. [2] concluded that these cysts are difficult to diagnose before surgery with ultrasonography and preoperative diagnosis was only possible in 6.6 % of the cases.

Although they are not uncommon, they rarely cause symptoms and are usually incidentally found. The



symptoms occur when they grow excessively, or in case of hemorrhage, rupture, or torsion. These rare occasions of PTC complications are reported as case reports. Paraovarian tumors are mostly benign. Although ovarian borderline tumors are well characterized in the literature, their fallopian tube and paratubal counterparts are limited to case reports [5]. In this report, three cases of PTCs diagnosed and treated in our department with uncommon complications are presented, and a brief review of the literature considering the subject is performed.

Case 1

A 33-year-old multigravid woman, presented with cyclic pelvic pain during the last few months. She was found to have a 30 × 20 cm mass by ultrasonography which is filling up all the abdominal cavity. The ultrasonography revealed thin-walled mass of right adnexial origin with fine particles inside. The right ovary was not visible by abdominal or vaginal ultrasonography. The left ovary was normal. Tumor markers were normal except Ca 125 which 69.15 IU/ml. The abdominal MRI revealed a 30×25 cm mesenteric or paraovarian cyst (Fig. 1). We decided to perform laparotomy because of huge size and high levels of Ca 125. Intraoperatively, a giant 25×30 cm mass of right tubal origin, lying towards the infundibulopelvic ligament, was seen (Fig. 2). After the suction of 2,500 mL serous fluid from the cyst, cystectomy and right ovarian wedge resection was done. The cyst was diagnosed as simple serous cyst in the frozen sections, whereas the



Fig. 1 MRI view of a giant paratubal cyst: sagittal contrast-enhanced view show a large unilocular cystic mass (arrow)





Fig. 2 Intraoperative view of the giant paratubal cyst capsule with prominent vessels after aspiration of 2,500 mL of serous fluid

final histopatological diagnosis was serous papillary cystadenofibroma.

Case 2

A 22-year-old primigravid patient was admitted to emergency service for acute onset of right lower quadrant pain, nausea, and vomiting. Transvaginal ultrasonography revealed 4.9 \times 4.1 cm heterogeneous cyst of right ovarian origin (Fig. 3). White blood cell count was high (18,700/ μ L) and CRP was 4.5 mg/L. The patient underwent urgent laparoscopy because decreased vascularisation was found at Doppler ultrasound. Intraoperative finding was 5 cm hemorrhagic right paratubal cyst, twisted two times around its stalk. After aspiration of cyst fluid, capsule excision was



Fig. 3 Transvaginal ultrasound image of a heterogeneous right twisted paratubal cyst (*calipers*), misdiagnosed as ovarian torsion after Doppler ultrasound

done laparoscopically. The final pathologic diagnosis revealed simple serous cyst.

Case 3

A 17-year-old virgin girl was admitted to the gynecology clinic for menstrual irregularity. She was suffering from oligomenorrhea and transabdominal ultrasonography showed 7×4.9 cm right abdominal mass in cystic pattern with papillary projections inside (Fig. 4). Her history revealed the diagnosis of a cystic right ovarian mass of



Fig. 4 Transabdominal ultrasound image of a paratubal cyst with papillary projections and thick wall (calipers): misdiagnosed as ovarian complicated cyst

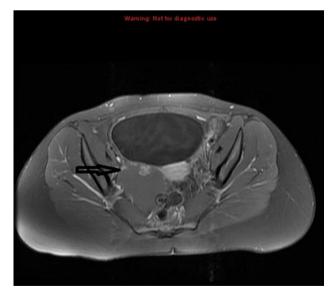


Fig. 5 MRI view of a paratubal cyst: coronal image with unilocular cystic mass with papillary projections (*arrow*); histopathological diagnosis was borderline paratubal cyst



Fig. 6 Photomicrograph of papillae corresponding to a serous borderline tumor with atypia, and stratification (H&E ×400)

5 cm in diameter a year ago. A slow progression of the mass was confirmed from previous ultrasonography reports. Tumor markers were normal (Ca 125 9 IU/ml). Abdominal MRI revealed 7 × 5 cm complicated right ovarian mass with papillary projections (Fig. 5). In the laparotomy, the cystic mass was about 8×7 cm with smooth surface, unbound to right ovary under the right tuba. The cyst was excised without rupture and right ovarian wedge resection was performed. Frozen specimens were benign, whereas final diagnosis was borderline serous cystadenoma (Fig. 6) and ovarian pathology revealed corpus luteum cyst, and she was referred to medical oncology department. Another surgical procedure has not been advised because there was no stromal invasion seen in the specimens. She has been disease-free after 1-year follow-up.

Discussion

The initial evaluation of an adnexial mass starts with ultrasonography either transvaginally or transabdominally. The differential diagnosis of paratubal and paraovarian cyst by ultrasonography requires greater awareness and accuracy [7]. The dissocation of the cyst from the ovary when pushing the probe is a useful sign, called as "Split sign", for discriminating paraovarian masses [8]. Darwish et al. [7], reported definite preoperative transvaginal sonography diagnosis in 44 % of the cases (52/118 patients). In another study, preoperative diagnosis of PTC was 26.3 % (29/110) based on ultrasound or computed tomography [6]. Gray scale and color Doppler ultrasound for diagnosing paraovarian or PTCs has 99 % specifity but low sensitivity (14 %) [9]. Others [10] reported 47 % sensitivity by transvaginal ultrasound. The explanation for discordant



results of the studies might be due to experience of the examiner and the quality of the ultrasound used. Moreover, serum Ca 125 measurement does not increase the diagnostic accuracy of transvaginal sonography [10]. Other imaging techniques like computerized tomography (CT) and magnetic resonance imaging (MRI) can also be used in differential diagnosis. MRI features of paraovarian cysts were described as homogeneous cystic masses near the ipsilateral round ligament and the uterus [11]. Especially demonstration of a normal ovary seperate from the cyst is an important MRI finding. There is no specific finding on CT other than unilocular cystic masses near the adjacent ovary. In two of the cases reported here, the preoperative diagnosis was ovarian mass, but the giant PTC was reported as paraovarian mass in MRI (Case 1).

Paratubal cysts are mostly asymptomatic, but they may occasionally give rise to clinical problems due to enlargement, hemorrhage, torsion, or malignancy. When enlargement of a paraovarian or PTC occurs, patients might admit with pelvic pain or feeling of a mass in the abdomen. They may also admit with other symptoms such as menstrual irregularity as in our third case. As the mass increases in size, risk of torsion also increases. In the literature, the biggest paraovarian cyst reported by Letourneur [12] is a 36×25 cm mass in a 19-year-old girl, which was managed by minimal suprapubic laparotomy following laparoscopy. In 2008, Kostov et al. [13] reported a $30 \times 26 \times 12$ cm large hypodense cystic mass with double torsion in a 14-year-old obese girl. Laparotomic cyst excision had been performed [13]. The case reported here (Case 1) is the third giant PTC in the literature (Table 1). Our patient was quite older than the two previously reported ones. Due to the large size of the cyst, we also performed laparotomy as the other authors. Luckily, all of these giant masses were benign; and no further surgical procedures were required.

Another uncommon presentation of PTC is torsion. The incidence of torsion among patients with paraovarian cysts ranges from 2.1 to 16 % [4] compared to 2.3 % in adnexal cysts [14]. Especially in children, torsion is a more common issue while the infundibulopelvic ligament

is longer. In young females, (age range 4–14 years old) operated for right lower quadrant abdominal pain, 13.6 % had PTCs (46/338) and two of them had isolated torsion of a large paratubal cyst [15]. Beginning from 2002, only six cases of benign twisted paraovarian masses had been reported up today (Table 2). The case reported here as twisted paraovarian cyst (Case 2) is the seventh in the literature.

Symptomatic PTCs complicated by acute tuboovarian torsion share a common clinical presentation with other diseases, such as acute appendicitis, ruptured ovarian cyst, acute ureteric colic, or pelvic inflammatory disease. Physical findings are abdominal and adnexal tenderness on pelvic examination. Peritoneal signs, sometimes with a palpable mass may or may not occur [3]. Laboratory values are usually non-specific; there may be an elevated white blood cell count [16] as in our case (Case 2). So, gynecologists would rather keep in mind torsion of PTCs while performing differential diagnosis of acute pelvic pain. In our case only torsion of the PTC was present but the previous cases in the literature also reported accompanying tuboovarian torsion or torsed fallopian tube. Therefore, surgery on time is important for ovarian preservation and tubal preservation, as well.

Ultrasound features of tubal torsion include visualization of the twisted vascular pedicle and dilated tube with thick echogenic walls and internal debris/hemorrhage [17]. Color Doppler is also helpful, which may show a high impedance waveform with reversal of diastolic flow in the affected tube [3]. In the second case reported here, preoperative diagnosis was ovarian mass with altered Doppler findings that necessiated laparoscopic evaluation. Low et al. [18] reported torsion of a large paratubal cyst in 12 years old obese girl. They defined the CT features as a thickened fallopian tube and smooth eccentric cyst wall thickening and suggested that CT is an alternative for overweight patients where USG may have been of limited value. Laparoscopy is the gold-standard for diagnosis and treatment of torsion. De-torsion is the treatment of choice today; in addition excision of the mass is necessary to avoid recurrence, as performed in our case.

Table 1 Reported giant paratubal cyts in the literature

Year	Author [ref.]	Age (years)	Imaging results	Surgical procedure	Intraoperative findings
2006	Letourneur [12]	19	USG: 36 × 25 cm mass	L/S Mini-LPT	Benign paraovarian cyst
2008	Kostov [13]	14	CT: $30 \times 26 \times 12$ cm large hypodens cystic mass	LPT	Giant paraovarian cyst with double adnexal torsion
2012	Case 1	33	USG: 30×20 cm thin walled mass with fine particles inside, MRI: 30×25 cm mesenteric or paraovarian cyst	LPT	Giant Paraovarian Cyst

CT computerized tomography, LPT laparotomy, L/S laparoscopy, USG ultrasonography



Table 2 Reported paratubal cysts complicated by acute tubooyarian torsion in the literature

Year	Author [ref.]	Age (years)	Presentation	Imaging results	Surgical procedure	Pathology
2002	Okada (2 cases)	14	POC torsion Torsed fallopian tube with a POC	CT: twisted right fallopian tube CT: homogenous cyst close to the uterus	LPT	Benign paraovarian cyst
2005	Low [18]	12	PTC with tuboovarian torsion	CT: $11 \times 8 \times 10$ cm thin-walled cyst with a few thin internal septa	L/S	Benign PTC
2008	Said MR	12	Twisted POC	USG: 9.7×9.1 cm multicystic pelvic mass	L/S	Mesothelial cyst
2008	Kostov [13]	14	Giant POC with double torsion	CT: $30 \times 26 \times 12$ cm large hypodens cystic mass	LPT	Serous cystadenoma
2009	Seshadri S	12	Bilateral PTC with coexisting fallopian tube torsion	USG: 7.7×6.5 cm and 8×4.6 cm bilateral ovarian cysts	L/S	Benign PTC
2012	Case 2	32	PTC twisted two times	USG: 50×41 cm right ovarian mass	L/S	Simple serous cyst

CT computerized tomography, LPT laparotomy, L/S laparoscopy, POC paraovarian cyst, PTC paratubal cyst, USG ultrasonography

Table 3 Reported borderline paratubal tumors in the literature

Year	Author	Age (years)	Intraoperative findings	Procedure	Pathology	Follow-up
2005	Salamon [22]	45	3 cm twisted PTC with a 2 cm right ovarian cyst	Right SO	Endometrioid BT in a PTC	1 year no recurrence
2009	Seamon [20]	26	12.5 cm simple PTC	Right SO, partial omentectomy, appendectomy, PPALND	Serous BT	1 year no recurrence
2010	Kumbak [23]	39	6 cm PTC during C/S	Cystectomy, omentectomy, appendectomy, PPALND	Serous BT	15 months no recurrence
2011	Terek [24]	19	10 cm twisted left PTC	Cystectomy and peritoneal washing	Serous BT	7 months no recurrence
2011	Im [25]	20	Complex right adnexal mass	Laparoscopic cyst excision	Mucinous BT	
2012	Case 3	17	$\begin{array}{c} \text{Complex 7} \times \text{5 cm right adnexal} \\ \text{mass} \end{array}$	Cystectomy and right ovarian wedge resection	Serous BT	1 year no recurrence

BT borderline tumor, PPLND pelvic paraaortic lymphadenectomy, PTC paratubal cyst, SO salpingooophorectomy

Histologically paraovarian or PTCs are usually simple cysts filled with serous fluid. Neoplastic features of these cysts have also been reported, but malignant PTC or paraovarian lesions are very rare. Although there are not large population based series, the histopathological analyses of cystic paraovarian lesions revealed 74.6 % simple cysts and 25.4 % neoplastic lesions (seven cystadenomas, eight cystadenofibromas) [19]. As far as we know, there are only 18 cases of malignant or borderline paraovarian epithelial tumors reported up today. Among the borderline tumors, the one reported here is the sixth borderline PTC in the literature (Table 3). In the literature, the first case of a serous borderline PTC was reported by Seamon in 2009 [20]. Salamon et al. [22] reported the first and the only borderline endometrioid tumor in a PTC. Another case of serous borderline paratubal tumor which was incidentally found during cesarean section, was reported by Kumbak [23]. Terek et al. [24] reported a twisted serous borderline paratubal tumor in a 19-year-old adolescent girl in 2011.

Paraovarian or PTCs usually represent as unilocular simple cysts with thin walls. If papillary projections are present, risk of malignancy increases [8]. The previously defined sonographic features of paraovarian and PTCs are as follows: unilocular cysts in 66 %, multilocular cysts in 4 %, and papillary projections in the cyst wall in 30 % [8]. The histopathological diagnosis of masses containing papillary projections (n = 15) were all neoplastic tumors in a series of 94 PTC/paraovarian cysts; eight cystadenofibromas (8/15), five cystadenomas (5/15), and two serous papillary borderline tumors (2/15). In the last case presented here, ultrasonography and MRI revealed a complicated cyst involving papillary projections. Although the tumor markers were normal, the progression in size of the mass and the presence of intramural papillary projections were suspicious for malignancy, and frozen section was



performed at laparotomy. As a result, if the size of the PTC is greater than 5 cm and papillary projections are present, the gynecologists should be aware of the increased risk of malignancy [8].

For an appropriate treatment, the nature of the mass is important. Even if there are no well-described criteria operating PTC, they are usually treated as ovarian masses. Therefore, small, unilocular, simple cysts are managed expectantly, while persistent thick-walled cysts involving papillary projections, or masses showing signs of torsion are managed surgically. Surgery may be performed laparoscopically or via open surgery. Size of the mass is the limiting factor for laparoscopy. In a total of 1,853 patients undergoing laparoscopy, Darwish et al. [7] detected 118 (15.7 %) paraovarian or PTCs. They defined endocystic-endoscopic visualization of these cysts as a simple and valuable step prior to cystectomy.

Fertility-sparing surgery seems to be a suitable option for young patients with borderline paraovarian/PTCs, but close follow-up is necessary. For stage IA borderline serous ovarian tumors, the indications for restaging surgery remain controversial. A recent French multicenter study found no difference in recurrence rate between women who underwent restaging and those who did not [21]. Rarity of paratubal borderline tumors makes the management particularly challenging [22]. In the previous five reports of paratubal borderline tumors, no recurrence was reported in mean follow-up of 1 year. In our case (Case 3), further restaging surgery was not performed due to lack of stromal invasion. Our case is 17-year-old and is the youngest patient in the literature with borderline paratubal tumor.

In conclusion, gynecologists should always be aware of paraovarian or paratubal masses in differential diagnosis of adnexal masses. Possible complications possess a risk for future fertility. A careful and detailed evaluation reminding paraovarian/paratubal pathologies before surgery is necessary. Presence of papillary projections indicates the possibility of malignancy. Therefore, frozen-section analysis is strongly suggested. More reports of such cases of malign paratubal/paraovarian pathologies are needed before a treatment modality is established.

Conflict of interest The authors declare that they have no conflict of interest.

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