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Para Ovarian Cyst in 16 Years Old Female; Borderline Ovarian Tumor: A Case Report

Mohamed El-Bakry Lashin[1], Mai Mohammed Abdelwahab[2], Walid Mohamed Elnagar[1,*]

[1]Obstetrics and Gynecology Department, Faculty of Medicine, Zagazig University, Egypt
[2]Pathology Department, Faculty of Medicine, Zagazig University, Egypt
*Corresponding author: Maiabdelwahab@ymail.com

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Abstract Paraovarian cysts are common, accounting for 10–20% of all adnexal lesions, and most of them are benign. On the other side, paraovarian tumors of borderline malignancy are very rare, only about forty cases were reported all over the world. Here, we present a case of 16 years female with left lower abdominal pain, pelvic sonography showed a cystic mass in Douglas pouch with solid mural nodule. She underwent laparoscopy, we detected and extirpated a paraovarian cyst of about 10x10 cm with solid component 3x3 cm and corpus luteum cyst 5x5 cm. After histopathological analysis, it was proved to be borderline ovarian serous tumor Stage Ic according to the International Federation of Gynecology and Obstetrics (FIGO) staging. As till now, there is no clear guideline regarding the management of this tumor; the treatment strategy was determined on the basis of ovarian tumor guidelines, with preservation of fertility. In conclusion, paraovarian cysts is a common disease and usually benign. It rarely causes clinical problems, but caution is necessary as there is a possibility of a malignant or borderline tumor. As in ovarian tumors, even if the size of the solid component of the tumor detected by diagnostic imaging is very tiny, malignant or borderline malignant tumors should be considered.

Keywords: *borderline, ovarian tumor, paraovarian cyst, laparoscopy, imaging*

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1. Introduction

Paraovarian cysts represent 10–20% of all adnexal masses [1]. They usually arise from the mesothelium that cover the peritoneum or from remnants of mesonephric and paramesonephric origin. So microscopically, they are covered by a single layer of flattened cells or ciliated columnar cells [2]. They mostly represent in third to fifth decades, though they can be seen in all age groups. The origin of these cysts can be non-neoplastic, simple cystic or neoplastic [3]. Usually, they are benign and hardly 2 cm in maximum diameter [4].

Para ovarian cysts are usually asymptomatic and incidentally discovered at laparotomy or laparoscopy. If symptomatic, symptoms are usually due to pressure on nearby organ e.g. Bowel and bladder or due to complications such as perforation, enlargement, infection, or haemorrhage [5]. The definitive management of simple paraovarian cyst is enucleation of cyst with preservation of both ovaries and fallopian tube. In case of complicated cyst, excision of ovary and/or fallopian tube may be required [6].

Paratubal borderline cysts show epithelial proliferation without stromal invasion; these tumors are reported only as case reports in the literature.

2. Case Report

A 16 year old virgin female, was referred to our gynecological clinic with abdominal pain in left iliac fossa. On examination, there was mild abdominal tenderness in left iliac fossa with mild rebound tenderness.

Pelvic sonographic evaluation revealed normal sized uterus with no polyps nor fibroids and normal endometrial thickness. It showed a cystic mass in Douglas pouch measuring 76x47 mm. with a solid mural nodule at its right side measuring 27x23 mm. The nodule had a well defined margins and showed internal vascularity.The mass was inseparable from both ovaries, and there was a mild fluid in the pelvis (Figure 1). The values of the tumor markers cancer antigen (CA)125 was 8.7 U/ml.

Then, the patient underwent laparoscopy, the uterus was normal and there was no finding of endometriosis. We detected and extirpated a paraovarian cyst of about 10x10 cm with solid component 3x3 cm and also corpus luteum cyst 5x5 cm. (Figures 2). The cysts were sent to pathology. Grossly, the paraovarian cyst measured 10X10 cm, contained serous fluid and coarse papillary projections. The corpus luteum cyst was 5x5 cm with thick fibrous wall and hemorrhagic contents.

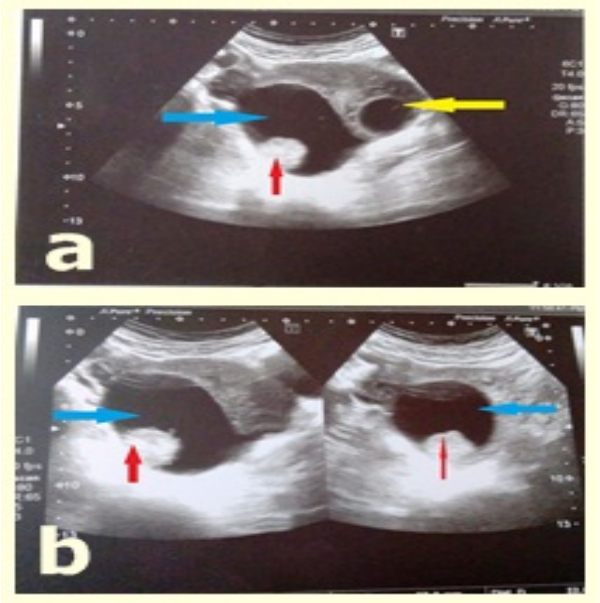

Figure 1(a,b). Ultasonographic picture of the case showing ovarian cyst (yellow arrow) and paraovarian cystic structure (blue arrow) with a solid component (red arrow). **Note**. The paraovarian cyst is close to but clearly separated from the ipsilateral ovary

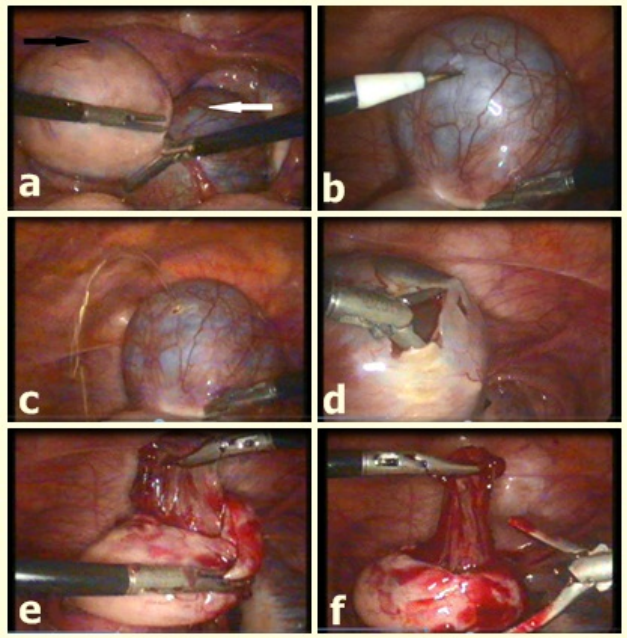

Figure 2. Laparoscopic removal of the paraovarian cyst(a,b,c) and the ovarian cyst (d,e,f)

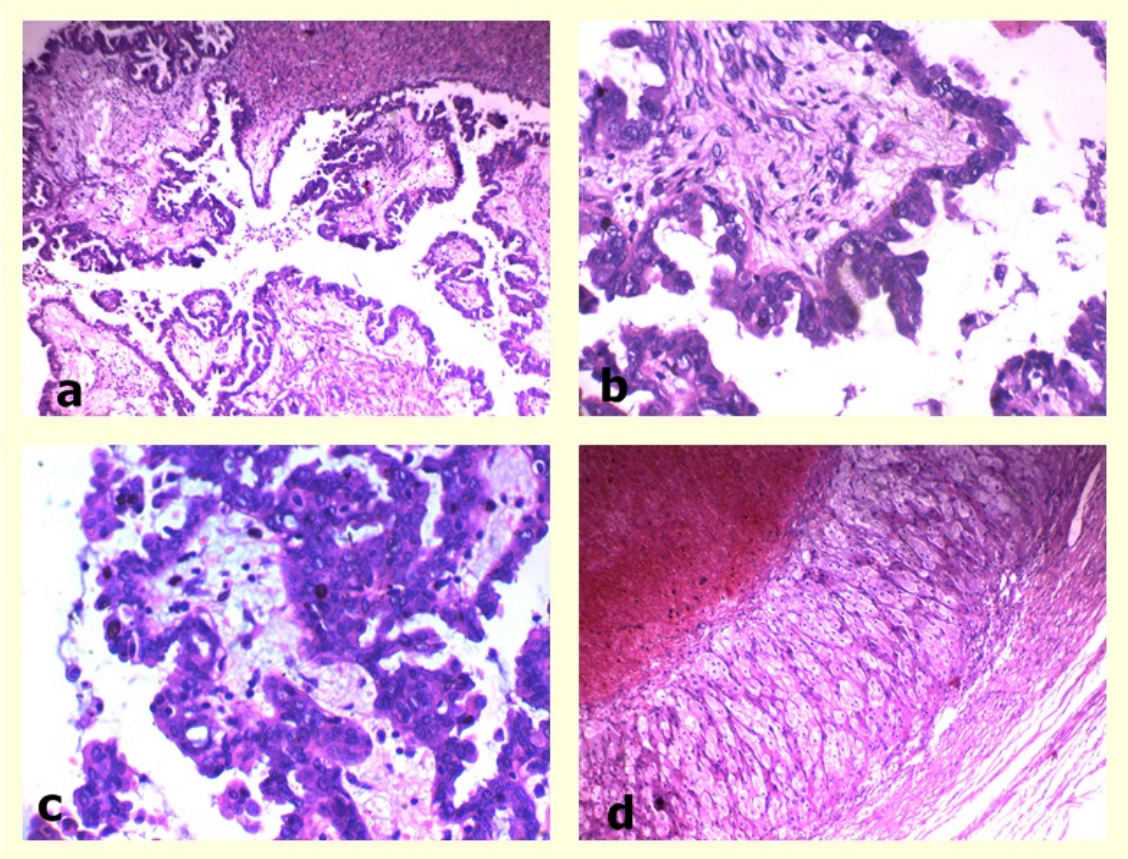

Figure 3. Microscopic picture of paraovrain cyst of borderline malignancy (a,b,c) showing multiple papillary projections lined by multilayring of cells (not exceeding 5 layers) with moderate nuclear basophilia and pleomorphism with no obvious stromal invasion. d) Corpus luteum cyst showing luteinized granulosa and theca cells with prominent outer fibrous layer

Microscopically; paraovarian cyst was unilocular, containing multiple papillary projections and lined by multilayered cells with few mitotic figures, light to moderate dyskaryosis, and without stromal invasion. While ovarian cyst showed convolutes lining by luteinized granulosa and theca cells with outer fibrous layer (Figure 3).

Based on the previous findings, the tumor was diagnosed as a serous borderline tumor, stage Ic in the International Federation of Gynecology and Obstetrics (FIGO) staging, as the cyst was ruptured during operation and the ovarian cyst was diagnosed as hemorrhagic corpus luteum cyst. As there is no obvious guideline for the

management of this tumor; the treatment plan was decided on the basis of the ovarian tumor guiding principle, with preservation of fertility.

Follow up by sonography and tumor markers, there has been no recurrence 1 year after surgery.

3. Discussion

"Paraovarian cysts" is a general term for cystic lesions that occurring between the hilum of ovary and fimbria within the mesosalpinx and broad ligament, and originate from the Wolffian duct, Müllerian duct, or mesothelium [7]. Paraovarian cysts are common, accounting for 10-20% of all adnexal lesions, and most of them are benign. On the other side, paraovarian tumors of borderline malignancy are very rare, only about forty cases have been reported worldwide [8]. These cysts are commonly asymptomatic, however, they can occasionally be presented by clinical symptoms resulting from excessive growth, rupture, hemorrhage or torsion [9]. Paraovarian cysts are usually anechoic unilocular cysts, with a uniform thin wall observed on ultrasound examination. However, preoperative diagnosis of these cysts is usually difficult, and they are commonly misdiagnosed as other pelvic cystic masses like peritoneal inclusion cysts and lymphoceles [10]. In one previous report , a preoperative diagnosis of paraovarian cysts by ultrasound was possible only in 6.7% of cases, and 73.3% were misdiagnosed as tumors of ovarian origin [11].

Another report showed that only 2% of patients with paraovarian cysts had malignant potential [12]. However, up to now, there is no reports that describe the detailed imaging characteristics of borderline/malignant paraovarian tumors.

Savelli et al., 2006 [13], compared the ultrasound findings of 50 cases of paraovarian tumors, they found that 15 cases showed multiple papillary projections arising from the cyst wall into the lumen. Eleven out of these 15 cases, showed no blood vessels within the papillary projections, as were visualized by color Doppler. However, 13 out of the 15 cases with multiple papillary projections were benign tumors, including eight cases of cystadenofibromas and five cases of cystadenomas. Only two cases were serous borderline tumors.

Generally, the presence of mural nodules and solid components in the lumen of cystic ovarian tumors indicates their malignant potential [14]. On the other hand, about 86% of paraovarian tumors with papillary projections were proved to be benign in Savelli study. So, detecting the nodular components of a paraovarian tumor does not always indicate that it is a malignant ones, making the differentiation between benign and malignant tumors very difficult.

In the literature, the first case of a serous borderline paratubal cyst (PTC) was reported by Seamon in 2009 [15]. Salamon et al., 2005 [16] 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 , 2010 [17]. Terek et al., 2011 [18] reported a twisted serous borderline paratubal tumor in a 19-year-old adolescent girl in 2011.

The differential diagnosis includes a simple ovarian cyst, peritoneal inclusion cyst and hydrosalpinx; many factors should be considered in differentiating theses lesions as proximity to the ovary, the presence of septations and small parietal papillae. *Peritoneal inclusion cysts* are multilocular cystic masses with a star like irregular morphology and no proper wall; septations are multiple and free to oscillate when moving the probe (flapping sail sign). *Hydrosalpinges* are tortuous and convoluted cystic adnexal masses having a distinct wall with small hyperechoic mural nodules on the cross-section of the salpinx, named (beads-on-a-string). Both hydrosalpinges and paraovarian cysts have the (split sign) identified by pushing the tip of the vaginal probe between this structures and ipsilateral ovary [19].

Although little is known about preoperative findings of imaging studies of patients with borderline paraovarian cysts, it is recommended that low-level echoes seen within the cyst and papillary projections on the cyst wall which should be searched carefully [20]. However, sometimes paraovarian cysts are removed, and considered benign; it is not until pathological assessment that the borderline tumor is recognized. This is what exactly happened in our case.

Borderline paraovarian cysts are usually early-stage at diagnosis. The microscopic appearance of these tumors is identical with that of borderline ovarian tumors; however, it is not known whether their biological behavior is also similar [17].

The management of these cases intraoperatively includes cystectomy, with or without partial or complete salpingectomy, adenextectomy, hysterectomy and bilateral oophorosalpingectomy, or salpingectomy along with pelvic-aortic lymphadenectomy or pelvic nodal sampling and omentectomy or biopsy with pelvic nodal sampling.

In review of literature, no positive nodal or distant metastatic disease were found in patients who underwent more comprehensive staging. Also, no recurrence was observed during the follow-up of such patients [21].

The optimal treatment procedure is unknown, however, patients desiring childbearing may be applied for fertility - sparing operation. If she has no desire for further fertility, more radical surgery is preferred (hysterectomy, bilateral salpingo-oophorectomy, pelvic-aortic lymphadenectomy, omentectomy, biopsies) [22]. Pelvic aortic lymphadenectomy is still a controversial subject [23].

In our case the treatment was only cystectomy. However, close follow-up is mandatory to detect recurrent disease after conservative, fertility-sparing surgery. Combining routine ultrasonography and markers (CA125) during follow-up examinations and prolongation of such follow-ups after 10 years were suggested.

4. Conclusion

Paraovarian cysts is a common disease and usually benign. It rarely causes clinical problems, but caution is necessary as there is a possibility of a malignant or borderline tumor. As in ovarian tumors, even if the size of the solid component of the tumor detected by diagnostic imaging is very tiny, malignant or borderline malignant tumors need to be considered.

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