An adolescent with an asymptomatic adnexal cyst: To worry or not to worry? Medical versus surgical management options

Vincenzo De Sanctis1, Ashraf T Soliman2, Heba Elsedfy1, Nada A Soliman4, Rania Elalaily3, Salvatore Di Maio6, Alaa Y Ahmed3, Giuseppe Millimaggi7

1Pediatric and Adolescent Outpatient Clinic, Quisisana Hospital, Ferrara, Italy; 2Department of Pediatrics, Division of Endocrinology, Alexandria University Children's Hospital, Alexandria, Egypt; 3Department of Pediatrics, Ain Shams University, Cairo, Egypt; 4Ministry of Health, Alexandria, Egypt; 5Department of Primary Health Care, Abu Nakha Hospital, Doha, Qatar; 6Emeritus Director in Pediatrics, “Santobono-Pausilipon” Hospital, Naples, Italy; 7Radiology Clinic, Quisisana Hospital, Ferrara, Italy

Summary. Paraovarian cysts or paratubal cysts (PTCs) arise from either the mesothelium or from paramesonephric remnants. These present as either adnexal mass or as an incidental finding. Diagnosis is usually established on ultrasound and it is important to differentiate these from ovarian cysts. Typically PCTs appear as simple cysts by ultrasound and are indistinguishable from ovarian cysts if one does not recognize the extraovarian location. Occasionally, PTCs have internal echoes due to hemorrhage. PTCs are usually asymptomatic and benign. The differential diagnosis includes a simple ovarian cyst, peritoneal inclusion cyst and hydrosalpinx. Malignant changes have been reported in about 2% to 3%, and it should be suspected if papillary projections are present. PTCs management depend upon the presence and severity of the symptoms, the cyst size and US characteristics, CA 125 results, age of the patient and the risk of malignancy. Simple PTCs can be expected to regress and may be managed expectantly. When surgery is indicated, a joint multidisciplinary management by the paediatric surgeons and trained paediatric gynaecologists should be the gold standard. (www.actabiomedica.it)

Key words: paraovarian cysts, paratubal cysts, diagnosis, treatment, complications

Introduction

In gynecology, the adnexa refer to the region adjoining the uterus that contains the ovary and fallopian tube, as well as associated vessels, ligaments, and connective tissue. Pathology in this area may arise from the uterus, bowel, retroperitoneum, or metastatic disease from another site, such as the breast or stomach (1, 2).

The adnexal cysts may be classified as paratubal or paraovarian depending on their proximity to either the tube or the ovary (1-4). Both are usually used synonymously and have been reported in all age groups, beginning from premenarchial period up to menopause (1). The incidence of PTCs is not clearly known. In an Italian population an incidence of 29 per 1,000 (~3 %) has been reported with a peak age of occurrence in the third and fourth decades of life (3). In pediatric and adolescent population, a much higher incidence of PTCs was reported (7.3 %) (5).

PTCs usually range in size from 2 to 8 cm. The smaller cysts are most commonly found in middle-aged women (in the 30 to 40 years age group), and are often indistinguishable from simple ovarian cysts.
Larger PTCs (up to 20 cm) tend to develop in younger women, quite often during pregnancy, at which time they have a tendency to grow rapidly (1-4).

The aim of this report is to present an adolescent referred to our adolescent outpatient clinic for a persistent “large unilocular ovarian cyst”. The most common types of extraovarian masses are reviewed with emphasis on diagnosis and management.

**Case presentation**

A 15-year-old virgin adolescent was referred, in March 2013, for persistent “large unilocular ovarian cyst” (4.3 x 4 cm) contiguous with, but not clearly separate from, the ovary. The cyst was found incidentally on pelvic sonogram 6 months before, because of menstrual irregularity and was treated for 3 months by her general practitioner with a contraceptive pill without success.

Her family and medical histories were unremarkable. Menstrual cycles were irregular with no dysmenorrhea. Her breast and pubic hair were at Tanner stage 5. The patient standing height was 158 cm (26th centile), weight 56 kg (63th centile) and body mass index (BMI) 22.4 Kg/m². Her pulse rate was 88 beats/minute and blood pressure 105/60 mmHg. Abdominal examination was negative. No signs of peritoneal irritation were noted. The patient did not have hirsutism, acne or galactorrhea.

Her lab investigations at the time of presentation were as follows: estradiol: 84 pmol/L; follicle-stimulating hormone (FSH): 4.0 IU/L; luteinizing hormone (LH): 5.4 IU/L; prolactin: 15.1 ng/ml; testosterone: 1.1 nmol/L and thyroid-stimulating hormone (TSH): 3.5 mIU/L. The routine urine, haematological and biochemical profiles, and CA-125 level were within the normal range.

Ultrasonographic (US) scan of the pelvic region showed a normal sized uterus, with a thin and regular endometrial lining. The left ovary was normal. In the right adnexal region a anechoic thin-walled cystic mass (4 x 3.8 cm) with regular contours was visible (Figure 1). The right ovary was not visible. No free abdominal fluid was noticed. She had her last menstruation 24 days back.

Abdominal magnetic resonance imaging (MRI) showed a cystic lesion (4 x 3.5 cm in size) in the abdomen that was separate from the right ovary and thought to be a paraovarian cyst.

The patient was discharged with a diagnosis of paraovarian cystic mass and managed conservatively. She was followed up clinically and by US every 6 months. The patient responded well to conservative management, and a significant regression in the size of the cystic lesion was observed at the end of the 12-month follow-up. A complete resolution was observed after 24 months without any need for surgical intervention. At last clinic visit her menstrual cycle intervals were between 24 to 35 days.

**Discussion**

PTCs are not uncommon. They rarely cause symptoms and therefore are usually incidentally found. The symptoms occur when they grow excessively, or in case of hemorrhage, rupture, or torsion. They arise from either the mesothelium or from paramesonephric remnants or from the invagination of the tube’s serosa (creating a mesothelial cyst). The mesonephric ducts begin to develop at 20-30 days of gestation and contribute to the development of the male reproductive excretory system (vas deferens, epididymis and seminal vesicles) (Figure 2). In females, these ducts remain as
vestigial structures and are often located in the broad ligaments. Parts of the epithelial lining may unusually remain active and continue to proliferate resulting in cystic masses (6).

In one large study, laparoscopic evaluation showed that these cysts were paratubal in 40% of patients and paraovarian in 60%. They were unilateral in 67.7% and bilateral in 15.3%. More than one small cyst occurred on one side in the remaining 17% of patients (7).

Small PTCs are usually asymptomatic and may disappear spontaneously. However, patients with larger PTCs frequently complain of pelvic pain, usually on one side, irregular periods, abnormal uterine bleeding, and dyspareunia. These symptomatic PTCs may need a laparoscopic procedure for histological diagnosis and treatment (8, 9).

Diagnosis is usually established by US imaging. It is important to differentiate these PTCs from true ovarian cysts. US can accurately diagnose PTCs in 87.5% of cases. When the mass is large or cannot be visually separate from the ovary an additional MRI may be necessary (10-12).

The majority of these cysts are benign. However, in rare cases borderline tumours and carcinomas have been reported arising from paratubal and paraovarian masses (1-2% of cases) (1, 2, 5, 13). In a retrospective study of symptomatic cysts by Savelli et al. (14) an incidence of 3-5% of malignancy was reported.

Certain sonographic findings (mural nodules, internal blood flow, papillary excrescences and ascites) are more pertaining to malignancy than others (15-21).

MRI may be useful when the ipsilateral ovary is not visualized separately, in the presence of complex paraovarian masses or suggestive US features of neoplasia. MRI offers better delineation of the mass and accurate evaluation of its vascularity (22).

The differential diagnosis of paraovarian and paratubal cysts includes peritoneal inclusion cysts and hydrosalpinx (23, 24) (Table 1).

Table 1. Ultrasound (US) differential diagnosis of simple paraovarian cysts, paraovarian cystadenomas, hydrosalpinx and peritoneal inclusion cysts

| Paraovarian cyst mass        | US characteristics                                                                 |
|------------------------------|-------------------------------------------------------------------------------------|
| Simple paraovarian cyst      | Paraovarian cysts can show a wide range of sonographic features. Sonographically they are usually thin-walled, smoothly margined, unilocular cysts. Occasionally, paraovarian cysts have internal echoes due to hemorrhage. |
| Paraovarian cystadenoma      | Paraovarian cystadenomas or cystadenofibromas are uncommon but should be considered when an extraovarian cyst contains a mural nodule or septation. |
| Hydrosalpinx                 | A hydrosalpinx should be considered when one encounters an elongated cystic mass with a tubular shape, with a partial septation and multiple small nodular areas along the wall because of thickened endosalpingeal folds. |
| Peritoneal inclusion cyst    | Peritoneal inclusion cysts are multilocular cystic masses with an irregular, star like morphology and no proper wall; septations are multiple and free to oscillate when moving the probe (flapping sail sign). On sonography, the presence of the ovary inside a large, ovoid or irregular, anechoic cyst is characteristic of a peritoneal inclusion cyst. |
Conclusions

Paraovarian or paratubal cysts (PTCs) constitute about 10% of adnexal masses. 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. Although malignancy is rare, borderline para- tubal tumors have been reported in the literature.

PTCs management depends upon the presence and severity of the symptoms, the cyst size and US characteristics, CA 125 results, age of the patient and the risk of malignancy. US imaging is currently considered as the first-line imaging technique for discriminating between benign and malignant adnexal masses. However, this technique is highly dependent on the expertise of the examiner (25-27).

Simple PTC can be expected to regress and may be managed expectantly. US morphological and functional properties must be periodically monitored as an alternative to surgery since malignant transformation is rare. However, “a cautious decision is necessary because larger and prospective patient series are needed to more definitely answer the question of which patients can be managed expectantly, and which patients need surgical management” (28).

When surgery is indicated, enucleation of the cyst from the mesosalpinx, with careful avoidance of cyst rupture or damage to ovary and fallopian tubes is recommended (29, 30). Joint multidisciplinary management of girls by the paediatric surgeons and trained paediatric gynaecologists should be the gold standard.

References

1. Muto MG. Approach to the patient with an adnexal mass. www.uptodate.com/contents/approach-to-the-patient-with-an-adnexal. May 4, 2016.
2. Samaha M, Woodruff JD. Paratubal cysts: Frequency, histogenesis, and associated clinical features. Obstet Gynecol 1985; 65: 691-694.
3. Pepe F, Panella M, Pepe G, Panella P. Paraovarian tumors. Eur J Gynaecol Oncol 1986; 7: 159-161.
4. Vlahakis-Miliaras E, Miliaras D, Koutsounis G, Miliaras S, Spyridakis I, Papadopoulos MS. Paratubal cysts in young females as an incidental finding in laparotomies performed for right lower quadrant abdominal pain. Pediatr Surg Int 1998;13:141-142
5. Muolokwu E, Sanchez J, Bercaw JL, Sangi-Haghpeykar H, Banszek T, Brandt ML, Dietrich JE. The incidence and surgical management of paratubal cysts in a pediatric and adolescent population. J Ped Surg 2011; 46: 2161-2163.
6. Akkawi R, Valente AL, Badawy SZA. Large mesonephric cyst with acute adnexal torsion in a teenage girl. Journal Pediatri Adolesc Gynecol 2012; 25: 143-145.
7. Darwish AM, Amin AF, Mohammad SA. Laparoscopic management of paratubal and paraovarian cysts. JSLS 2003; 7: 101-106.
8. Dotters-Katz SK, James AH, Jaffe TA. Paratubal/Paraovarian Masses: A Study of Surgical and Non-Surgical Outcomes. Med J Obstet Gynecol 2014; 2: 1019-1023.
9. Liu JH, Zanotti KM. Management of the adnexal mass. Obstet Gynecol 2011; 117: 1413-1428.
10. Timmerman D, Schwarzer P, Collins WP, Claerbout F, Coenen M, Amant F, Vergote I, Bourne TH. Subjective assessment of adnexal masses with the use of ultrasonography: an analysis of interobserver variability and experience. Ultrasound Obstet Gynecol 1999; 13: 11-16.
11. Guerrieri S, Alcazar JL, Pascual MA, Ajossa S, Gerada M, Bargellini R, Virgilio B, Melis GB. Diagnosis of the most frequent benign ovarian cysts: is ultrasonography accurate and reproducible? J Womens Health 2009; 18: 519-527.
12. Van Holsbeke C, Daemen A, Yazbek J, Holland TK, Bourne T, Mesens T, Lannoo L, Boes AS, Joos A, Van De Vijver A, Roggen N, de Moor B, de Jonge E, Testa AC, Valentijn L, Jurkovic D, Timmerman D. Ultrasound experience substantially impacts on diagnostic performance and confidence when adnexal masses are classified using pattern recognition. Gynecol Obstet Invest 2010; 69: 160-168.
13. Smorigick N, Herman A, Schneider D, Halperin R, Pansky M. Paraovarian cysts of neoplastic origin are underreported. JSLS 2009; 13: 22-26.
14. Savelli L, Ghi T, De Iaco P, Ceccaroni M, Venturoli S, Cacciatoire B. Paraovarian/paratubal cysts: Comparison of transvaginal sonographic and pathological findings to establish diagnostic criteria. Ultrasound Obstet Gynecol 2006; 28: 330-334.
15. Terek MC, Sahin C, Yeniel AO, Ergenoglu M, Zekioğlu O. Paratubal borderline tumor diagnosed in the adolescent period: a case report and review of the literature. J Pediatri Adolesc Gynecol 2011; 24: e115-e116
16. Puig F, Crespo R, Marquina I, Serous cystadenoma of borderline malignancy arising in a parovarian paramesonephric cyst Eur J Gynaecol Oncol 2006; 27: 417-418.
17. Persaud V, Anderson MF. Endometrial stromal sarcoma of the broad ligament arising in an area of endometriosis in a paramesonephric cyst. Case report Br J Obstet Gynaecol 1977; 84: 149-152.
18. Korbin CD, Brown DL, Welch WR. Paraovarian cystadenomas and cystadenofibromas: Sonographic characteristics in 14 cases. Radiology 1998; 208: 459-62.
19. Honore LH, O’Hara KE. Serous papillary neoplasms arising in paramesonephric parovarian cysts. A report of eight cases. Acta Obstet Gynecol Scand 1980; 59: 525-528.
20. Altaras MM, Jaffe R, Corduba M, Holtzinger M, Bahary C. Primary paraovarian cystadenocarcinoma: clinical and management aspects and literature review. Gynecol Oncol 1990; 38: 268-272.
21. Gupta A, Gupta P, Manaktala U, Khurana N. Clinical, radiological, and histopathological analysis of paraovarian cysts. J Midlife Health 2016; 7: 78-82.
22. Kishimoto K, Ito K, Awaya H, Matsunaga N, Outwater EK, Siegelman ES. Paraovarian cyst: MR imaging features. Abdom Imaging 2002; 27: 685-689.
23. Arhey PA, Cooper NB, Sonographic features of parovarian cysts AJR Am J Roentgenol 1985; 144: 83-86.
24. Kishimoto K, Ito K, Awaya H, Paraovarian cyst: MR imaging features Abdom Imaging 2002; 27: 685-689.
25. Güterriero S, Alcazar JL, Pascual MA, Ajossa S, Gerada M, Bargellini R, Virgilio B, Melis GB. Intraobserver and interobserver agreement of grayscale typical ultrasonographic patterns for the diagnosis of ovarian cancer. Ultrasound Med Biol 2008; 34: 1711-1716.
26. Van Holsbeke C, Daemen A, Yazbek J, Holland TK, Bourne T, Mesens T, Lannooy L, De Moor B, De Jonge E, Testa AC, Valentin D, Timmerman D. Ultrasound methods to distinguish between malignant and benign adnexal masses in the hands of examiners with different levels of experience. Ultrasound Obstet Gynecol 2009; 34: 454-461.
27. Faschingbauer F, Benz M, Haberrele L, Goecke TW, Beckmann MW, Renner S, Muller A, Wittenberg T, Munzenmayer C. Subjective assessment of ovarian masses using pattern recognition: the impact of experience on diagnostic performance and interobserver variability. Arch Gynecol Obstet 2012; 285: 1663-1669.
28. Dotters-Katz SK, James AH, Jaffe TA. Paratubal/Paraovarian Masses: A Study of Surgical and Non-Surgical Outcomes. Med J Obstet Gynecol 2014; 2: 1019-1024.
29. Pérez-López FR, Chedraui P, Troyano-Luque JM. Peri- and post-menopausal incidental adnexal masses and the risk of sporadic ovarian malignancy: new insights and clinical management. Gynecol Endocrinol 2010; 26: 631-643.
30. Ulusoy S, Akbayir O, Numanoglu C, Ulusoy N, Odabas E, Gulkilik A. The risk of malignancy index in discrimination of adnexal masses. Int J Gynaecol Obstet 2007; 96: 186-191.

Received: 19 December 2016
Accepted: 24 January 2017
Correspondence: Vincenzo De Sanctis MD, Pediatric and Adolescent Outpatient Clinic, Quisisana Hospital, 44100 Ferrara, Italy
Tel: 39 0532 770243
E-mail: vdesanctis@libero.it