Hemorrhagic Corpus Luteum Cysts: An Unusual Problem for Pediatric Surgeons

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Abstract. Study Objective: Hemorrhagic corpus luteum cysts (HCLC) constitute a common disorder in pediatric subjects undergoing surgical intervention. HCLCs especially develop in the early period after menarche, and they are commonly associated with dysfunctional ovulation.

Design: Retrospective analysis of surgery outcome of HCLC patients.

Setting: Pediatric Surgery Unit, S. Chiara University Hospital.

Participant: 13 girls with HCLC diagnosis.

Interventions: Surgical treatment of HCLCs.

Main Outcome Measures: We reviewed the clinical presentation and outcome of 13 post-menarcheal girls surgically treated for HCLCs in the Pediatric Surgical Unit from 2002 to 2006.

Results: Primary presentation was persistent abdominal pain in 84.6% and acute abdominal pain in 15.4% of patients, respectively. Ultrasound examination showed complex ovarian masses in 77.23% cases and simple ovarian masses in 33.7% cases, respectively. Although laparoscopic excision of HCLC was performed in more than 45% cases, laparotomic approach was commonly required. After conservative surgery, ovarian size and viability were normal, as assessed by 6-month ultrasound scan. No recurrences of disease and regular menses were reported at 2 years follow-up.

Conclusions: In pediatric subjects with HCLC that required surgical intervention, no complications or disorder recurrence were reported. In order to preserve ovarian function, conservative surgery has to be performed whenever feasible.

Key Words. Hemorrhagic corpus luteum cysts—Ovarian surgery—Tumor markers—Pediatric gynecology—Pediatric surgery

Introduction

Clinical presentation and outcome of ovarian disorders differ from those in adults requiring different diagnostic and management approaches.1,2 In particular, hemorrhagic corpus luteum cysts (HCLC) are usually unilocular and lined by luteinized theca and granulosa cells.3 Although hemorrhagic lesions can occur at any time of life, HCLCs develop during puberty especially in the early period after menarche, and they are commonly associated with dysfunctional ovulation.3–5

Patients with these cysts may present with abdominal pain or signs of peritoneal irritation that can be confused with acute appendicitis.3,6 After thorough diagnostic work-up, it is compulsory to decide the appropriate management for these patients. The HCLC spontaneously tend to regress on follow-up ultrasound few weeks later, and only a minority cause complications such as intracystic hemorrhage, rupture, and adnexal torsion.3,4,7,8

The HCLC treatment depends on the symptoms and the size of the cyst; when surgery is necessary, enucleation is performed whenever possible in order to conserve ovarian tissue.3 In fact ovarian surgery during childhood and adolescence could result in compromising future fertility, either from removal of normal ovarian tissue or from formation of adhesions.1

This study was undertaken to gain more knowledge on the HCLC in the pediatric age and to optimize their diagnostic and therapeutic management.

Patients and Methods

In the present study, we retrospectively reviewed pediatric subjects who underwent surgical intervention in Pediatric Surgical Unit from January 1, 2002, to August 31, 2006. After reviewing the pathology reports, the medical records of the patients were retrospectively reviewed. Specific data collected on each patient included clinical presentation, age at
operation, imaging studies (ultrasonography (US), computerized tomography (CT) and magnetic resonance imaging (MRI) scans), serum data (CA-125, α-fetoprotein and β-human chorionic gonadotropin), surgical route and outcome. The data have been reported as mean (± SD). After the local Institutional Review Board approval, the informed consent was obtained from all parents prior to the study.

Results

In the 5-year period, 67 patients were surgically evaluated for ovarian lesions (e.g. mature cystic teratoma, cystoadenoma, and yolk-sac tumor). Of them, 13 (19.4%) girls were affected by HCLC (Table 1).

At first surgery evaluation, they were 17.2 (±1.6) years (range 13–18 years) with menarcheal age of 12.3 (± 1.7) years. Four patients (30.8%) presented with a personal history of disruption of the menstrual cycle or dysmenorrhea. One patient presented with a family history of HCLCs: her mother was surgically treated for the same disease. No patients presented with clinical characteristics of precocious puberty, abnormal skin pigmentation, or other known syndrome markers at physical examination. Height, weight, and body mass index were 0.7 (± 0.41), 0.6 (± 0.28) and 0.4 (± 0.26) SDS, respectively.

Abdominal US was performed as the first diagnostic test for all patients to characterize ovarian diseases as simple, complex, 9 or solid lesions. US examination showed unilateral HCLCs in all our patients: complex ovarian masses in 10 (77.0%) and simple ovarian masses in 3 cases (23.0%), respectively. An additional abdominal CT scan was also performed in 7 patients (53.8%) to better define the complex lesions, such as to exclude associated pathologies (Fig. 1).

Serum levels of tumor markers CA-125, α-fetoprotein, and β-human chorionic gonadotropin were within in our normal ranges for all cases, respectively. The primary presentations that led to operation were persistent abdominal pain in 11 patients (84.6%) and acute abdominal pain in 2 patient (15.4 %), respectively.

Eleven patients (84.6%) were surgically treated after an ongoing observation of 5.3 ± 1.2 weeks (range 4–8 weeks), with several US scans. Two patients (15.4%) were treated urgently, one with a cyst complicated by ovarian torsion and the other with a suspected rupture of the lesion, in whom we found intrapelvic blood at the surgery view (Fig. 2A, B).

Six patients (46.2%) underwent operative laparoscopy: excision of the luteal cyst was performed in all cases. Seven patients (53.8%) underwent laparotomy: simple resection of the luteal cyst in 2 cases, oophorectomy in 2 cases, and salpingo-oophorectomy in one case. Incidental appendectomy was performed
in 5 patients (38.5%) with no associated complications. The decision of a laparoscopic or laparotomic surgical approach was based on the combination of two different parameters: the size of the lesion and its echographical aspects. Bigger lesions, especially if associated with a complex US aspect, have been treated with laparotomy due to technical difficulties, the higher risk of cyst rupture, and concern about the malignant potential of the lesions.

The histology of the ovarian lesions revealed the following: HCLC lesion was detected in the right ovary in 8 patients (61.5%) and in the left in 5 patients (38.5%); one girl also had an ipsilateral serous cystoadenoma another girl also had a contralateral paraovarian cyst.

After conservative surgery, ovarian size and viability, as assessed by 6-month ultrasound scan, has been normal. No recurrences of disease and regular menstrual cycles of 28–31 days were reported after 24.5 ± 12.8 months (range 9–53 months) of follow-up.

Discussion

Pediatric ovarian lesions, although rare, span a spectrum of pathology from functional nonneoplastic ovarian cysts to ovarian torsion, and from benign tumors to advanced neoplasms.1,2,6,10 Functional ovarian cysts constitute from 17.1% to 43.6% (mean 29.5 ± 9.6) of all surgically treated ovarian lesions in pediatric age; corpus luteum cysts constitute from 14.2% to 26.4% (mean 17.6 ± 5.1) (Table 2).1,2,6,11,12

The early period following menarche is commonly associated with dysfunctional ovulation, so ovulatory cysts are often seen as the result of aborted ovulations or persistence of the corpus luteum.3–5 Ovarian cysts in teenage years, as we have seen, may be asymptomatic or associated with menstrual irregularities, abdominal pain, urinary frequency, constipation, or pelvic discomfort.4,13,14. In adolescents, it is essential to elicit a full history including details of the menstrual and sexual history.4,14 The differential diagnosis widens to include a variety of acquired reproductive disorders such as pregnancy, sequelae of sexually transmitted diseases or endometriosis.13,14

The preoperative diagnostic work-up of ovarian pathologies in adolescents includes US scan and blood samples for tumor markers.8,14 The diagnosis is greatly aided by the use of imaging; the widespread availability and use of US has resulted in higher detection rate of functional cysts.13,14 US is non-invasive and has proven to be very valuable not only in diagnosis, but also in allowing observation of these lesions over time.

Hemorrhage in a functional cyst can lead to a diagnostic dilemma; in fact it may appear ultrasonographically complex or solid.9,12,15,16 Balan reported a patient in whom an ovarian lesion was considered as highly suspicious of malignancy at US but it was found to be an HCLC after surgery.9 Ten patients (76.9%) enrolled in our study presented with complex lesions. In the presence of complex or solid lesions, other investigations such as CT or MRI scan must be performed, because of the malignancy risk.9,15,17
The overall accuracy of US, CT, and MRI scans in the assessment of pelvic pathology are 77%, 87%, and 97% respectively. CT, used to clarify images, is helpful in distinguishing ovarian disorders from other diseases such as acute appendicitis, appendix abscess, tubo-ovarian abscess, or hydrocolpos/hematocolpos. The difficulty in identifying cyst histology before surgery is due to the fact that a histological lesion corresponds to several US pictures and vice versa. Moreover, sometimes, even macroscopically, it may be difficult to define the exact nature of a lesion, as reported by Doret and Raudrant. Indeed, imaging modalities may present false positive and false negative findings for a presumed functional ovarian cyst. Only surgery and histopathological analysis allow precise diagnosis of a solid or a complex lesion.

Emergency presentations should be considered first. In fact these may be misleading and may compromise the long-term outcome, especially in cases of acute rupture, torsion, or hemorrhage. Two patients came to our attention with acute abdominal pain; one presented with a cyst complicated by ovarian torsion.

The HCLC management depends on clinical symptoms, lesion size, and US appearance. These cysts, as they are hormonally driven, tend to resolve spontaneously in few weeks (4–6 on average) and only a minority will require surgical treatment. In the presence of asymptomatic cysts under 5 cm in diameter and normal US-based and biochemical parameters, ongoing surveillance is a reasonable option. Surgical indications, although not absolute, include cysts greater than 5 cm in diameter, a failure of the cyst to resolve or decrease in size spontaneously, complex or solid cysts indicative of suspected malignancy, severe, or persistent abdominal pain and complications of simple cysts greater than 5 cm in diameter. The cutoff of 5 cm for surgery indication is controversial; some authors, in fact, suggest that in a stable patient with an appearing benign, cystic, unilocular lesion under 10 cm in diameter, simple surveillance can be considered an acceptable option.

US and CT may be helpful in differentiating functional ovarian cysts from other surgical conditions characterized by abdominal pain but, as confirmed in our experience, a certain diagnosis may be made only at the time of surgery. Finally, the goal of surgical management of pediatric HCLC is to remove the lesion preserving the underlying ovary in order to optimize the conservation of steroidogenesis and fertility.

References


Table 2. Incidence of pediatric ovarian cysts: a review of the literature

<table>
<thead>
<tr>
<th></th>
<th>Freud et al, 1999²</th>
<th>Templeman et al, 2000¹⁰</th>
<th>Cass et al, 2001⁶</th>
<th>Deligeoroglou et al, 2004¹</th>
<th>Skiadas et al, 2004¹¹</th>
<th>Bristow et al, 2005⁹</th>
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<tr>
<td>Surgically treated ovaries</td>
<td>34</td>
<td>140</td>
<td>106</td>
<td>47</td>
<td>40</td>
<td>129</td>
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<td>Functional cysts, n (%)</td>
<td>14 (41.2)</td>
<td>61 (43.6)</td>
<td>26 (24.6)</td>
<td>12 (25.5)</td>
<td>10 (25.0)</td>
<td>14 (17.1)</td>
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<tr>
<td>Follicular</td>
<td>-</td>
<td>24 (17.2)</td>
<td>11 (10.4)</td>
<td>5 (10.6)</td>
<td>4 (10.0)</td>
<td>-</td>
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<tr>
<td>Corpus luteum</td>
<td>-</td>
<td>37 (26.4)</td>
<td>15 (14.2)</td>
<td>7 (14.9)</td>
<td>6 (15.0)</td>
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