GYNAECOLOGY

Diagnostic Dilemma: Acute Abdomen from Ruptured Corpus Luteum Requiring Surgical Intervention in Young Women

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ABSTRACT

Aim: Rupture of corpus luteum can occur in women of reproductive age, and is usually asymptomatic. Rarely, there is hemoperitoneum leading to circulatory shock requiring surgical intervention. This presents a diagnostic dilemma, with differential diagnoses of appendicitis, ectopic pregnancy and torsion of adnexal mass to consider. This study was undertaken to define characteristics and risk factors for corpus luteum rupture, to improve diagnostic capability and optimize management. There is a lack of local data on the incidence, and we hope to provide updates on management, given the ubiquity of laparoscopic surgery now.

Materials and Methods: A retrospective study was conducted on patients treated surgically for ruptured corpus luteum at National University Hospital, Singapore, from September 1, 2004 to March 31, 2011, looking at demographics, clinical features, operative findings and follow-up.

Results: 47 cases were analyzed; 80.8% in peak reproductive age of 15-34 years, 25.5% in first trimester pregnancy. Most are healthy, but two patients were newly diagnosed with idiopathic thrombocytopenic purpura on this admission. All patients presented with severe abdominal pain, while some also had signs of peritoneal irritation. 55.4% of such patients were initially misdiagnosed. Sonography revealed free fluid in 84.1%. Diagnostic laparoscopies were performed in 78.7%; with the rest undergoing laparotomies. Hemoperitoneum below 1L was seen in 85.1% of patients, above 1L in seven patients, of which three went onto require blood transfusions. All patients recovered well, with return of normal menses. None of the patients required further surgical treatment.

Conclusion: Increasing awareness of ruptured corpus luteum is recommended, especially for cases with right-sided pain, pregnancy and adnexal masses. High index of suspicion is required for timely intervention for securing hemostasis. Laparoscopy is used first-line, except cases better managed by laparotomy. Underlying bleeding diatheses can be a predisposing factor. Recurrence may be prevented with ovulation suppression.

Keywords: corpus luteum, hemoperitoneum, laparoscopy

Introduction

Ovulation can be associated with hemorrhage at several junctions. Where the ovum is extruded from the Graafian follicle, there can be bleeding into the peritoneal cavity. As the follicle develops into a corpus luteum, there is spontaneous bleeding into the central cavity forming the corpus hemorrhagicum, which could then rupture, resulting into hemoperitoneum. The rupture of the corpus luteum is more likely with cyst formation secondary to abnormal follicle maturation. In most women, these events are associated with minimal trauma and bleed and are rarely noticed. However. these events can also cause abdominal pain that is mostly self-limited requiring observation and analgesia. Uncommonly, this abdominal pain is severe enough to present as acute abdomen from hemoperitoneum causing circulatory collapse that warrant surgical intervention to achieve hemostasis and relief. Such cases present a diagnostic challenge; with differential diagnoses of acute appendicitis in cases with right iliac fossa pain, ectopic pregnancy, and torsion or rupture of pre-existing ovarian cysts. Scans are often not able to distinguish between the possible causes and surgical intervention becomes necessary as patients continue to be symptomatic. This leads to potential problems, as different presumptive diagnoses would mean diverse approaches during surgery.

This study was undertaken to recognize the profile of ruptured corpus luteum requiring surgical intervention, in our local population. 47 cases of ruptured corpus luteum requiring surgical intervention diagnosed and treated at our institution in the stated period were analyzed. The results serve to define characteristics in terms of risk factors, associations, clinical features, operative findings and events in subsequent follow-up. The aim is to optimize diagnostic capability and therapeutic management of such cases, to enhance patient care. Diagnostic laparoscopy has also become the standard of care as a first-look into the abdomen, as compared to previous data.

Materials and Methods

At our institution, all surgeries are recorded in an

electronic logbook database known as "Computerised Patient Support System". To begin our retrospective study, key words "cystectomy", "corpus luteum", "diagnostic laparoscopy", "therapeutic laparoscopy", "exploratory laparotomy" were used to pick out cases of ruptured corpus luteum managed surgically from September 1, 2004 to March 31, 2011.

Cases with confirmed diagnosis of hemorrhagic corpus luteum were then recruited and their medical records reviewed in depth. Specific data collected included demographic factors (age, race, marital status), medical, surgical, obstetric and gynecological history, clinical presentation, investigation results, imaging studies, intra-operative findings, histology and follow-up at 1 year (except for patients admitted after March 2010 where follow-up is less than a year). The protocol for collection, storage and data retrieval is under compliance with our institutional review board.

Results

In this period, there were 47 cases of ruptured corpus luteum requiring surgical intervention. The majority of patients (80.8%) were in the peak reproductive age group of 15-34 years (mean age 30.0 years, range 15-50 years). (Table 1)

All patients were sexually active except for two adolescent girls who had no sexual intercourse. In the sexually active, parity was not protective against a ruptured corpus luteum. A significant proportion of women (25.5%) were found to be pregnant at the event. They were in the first trimester of pregnancy with the largest gestational age at 9 weeks and 5 days. Only one knew of the pregnancy prior to the presentation. Four patients, including two with intra-uterine gestational sacs seen on sonography, proceeded to have successful pregnancies, three underwent dilation and curettage together with laparoscopy, and the remaining five were failed pregnancies.

Table 1. Patient characteristics

1. Age (years)	n=47	%
15-24	15	31.9
25-34	23	48.9
35-44	7	14.9
45-54	2	4.3
2. Sexual activity		
Active	45	95.7
Nulliparous	25	55.6
Parous	20	44.4
Inactive	2	4.3
3. Pregnancy at presentation		
Pregnant	12	25.5
Not pregnant	35	74.5
4. Phase of menstrual cycle		
Follicular	8	17.1
Luteal	23	48.9
Unknown (irregular menses)	9	19.1
Amenorrhea > 4 weeks	7	14.9
5. Antecedent to onset of symptoms		
Sexual intercourse	4	8.5
Exercise/Trauma	0	0
None significant	43	91.5
6. Gynecological problems		
Dysmenorrhea/Menorrhagia	6	12.8
Inter-menstrual bleed	1	2.1
Subfertility (on treatment)	3	6.4
7. Medical problems		
ITP (diagnosed post-event)	2	4.3
8. Initial diagnosis		
Appendicitis	6	12.8
Ectopic pregnancy	7	14.9
Cyst accident	11	23.4
OHSS	2	4.3

Regarding the menstrual period phase during which rupture occurred, it is more likely to occur during the luteal phase (48.9%) than in the follicular phase (17.1%), with remaining patients having irregular menses (19.1%) or amenorrhea longer than four weeks (14.9%). Oral contraceptive pills were not used in any

of the patients in the cycle during which the rupture occurred. Vast majority of patients did not recall any precipitating event prior to the onset of symptoms; with only 8.5% of patients reporting sexual intercourse.

Most patients were healthy with no medical issues, other than a small proportion (14.9%) with

dysmenorrhea, menorrhagia or inter-menstrual bleed. Three patients had subfertility and were on Clomiphene. None were known to have bleeding diatheses or on anticoagulants, but two patients were diagnosed with idiopathic thrombocytopenic purpura after this presentation.

Clinical features and imaging findings are depicted in Table 2. All patients presented with abdominal pain, abdominal tenderness was more common in the right iliac fossa (36.2%) than left (14.9%) on examination. This correlates well with intra-operative findings of 57.4% having ruptured right corpus luteum.

(Table 3) It has been thought to be more common in the right ovary with the rectosigmoid acting as cushioning of the left ovary from trauma. Peritoneal irritation with guarding and rebound tenderness was common (40.4%), along with excitation pain on pelvic examination (25.5%). Non-specific symptoms of nausea, vomiting or diarrhea were seen in fourteen patients (29.8%). The two patients with vaginal bleeding were pregnant. In particular, the initial diagnosis for 26 patients (55.4%) was not that of ruptured corpus luteum, but other presumptive

Table 2. Clinical features and imaging findings

Signs and symptoms	n=47	%
Abdominal pain (generalised)	31	66.0
Right iliac fossa	11	23.4
Left iliac fossa	5	10.6
Nausea/Vomiting/Diarrhea	14	29.8
Vaginal bleeding	2	4.3
Abdominal tenderness (lower abdomen)	23	48.9
Right iliac fossa	17	36.2
Left iliac fossa	7	14.9
Guarding/Rebound tenderness	19	40.4
Cervical excitation	12	25.5
Ultrasound imaging	n=44	%
Size of cyst (largest dimension-mm)		
<50	25	56.8
51-70	7	15.9
Not measured	10	22.7
Multiple	2	4.6
2. Free fluid		
Present	37	84.1
Absent	7	15.9
3. Other features		
Intra-uterine gestational sac	2	4.6
Tubo-ovarian mass	16	36.4
Uterine fibroid	2	4.6

Table 3. Surgical treatment and findings

Surgical Treatment	n=47	%
Laparoscopy	37	78.7
Laparoscopy converted to Laparotomy	2	4.3
Laparotomy	8	17.0
Intra-operative findings:	n=47	%
Ruptured corpus luteum		
Right	27	57.4
Left	18	38.3
Bilateral	2	4.3
Hemoperitoneum (ml)		
<500	26	55.3
500-1000	14	29.8
>1000	7	14.9
Hemostasis		
Cautery	37	78.7
Suturing	10	21.3

Discussion

Ruptured corpus luteum causing severe acute abdominal pain is a diagnostic challenge for the clinician. In particular, ruptured corpus luteum in pregnancy made up a significant proportion of our study cohort, 25.5% out of forty-seven subjects, as compared to 11% in a similar study of Asian women(1). An earlier study done by Hallatt et al in 1984 had 15.6% of patients pregnant, and they suggested the increase in ruptured corpus luteum in pregnancy could be due to increase in corpus luteum cysts(2). Although the majority of ruptured corpus luteum occurs in regular menstrual cycles, our findings indicate that it could still happen in pregnancy, since the corpus luteum persists until about 12 weeks to support the pregnancy. None of the ruptures were in second trimester pregnancies. Hence, in patients with acute abdominal pain in first trimester pregnancies, ruptured corpus luteum should also be considered besides ectopic pregnancy.

All patients in this study presented with acute abdominal pain. The pain in ruptured corpus luteum is usually of sudden onset with patients able to tell the exact time it started. This is in contrast to the pain in torsion that is more of a "waxing and waning" nature.

An accurate history in non-pregnant women is an important pointer to the diagnosis. If patients are in the luteal phase of the menstrual cycle, ruptured corpus luteum is likely. It may also be useful to enquire regarding history of preceding sexual intercourse or strenuous physical activity, as previous studies have noted the association with ruptured corpus luteum⁽¹⁻⁴⁾.

Other symptoms appear to be non-specific. Ultrasound is the imaging modality of choice for such patients, but signs are not specific, and often the only clue is presence of free fluid in Pouch of Douglas. However, free fluid can be present right after ovulation in up to 26% of asymptomatic women with no contraception pill usage, as a result of estradiol-induced increase in capillary permeability $^{(5)}$. As such, the amount of free fluid is more important than its mere presence. In general, raised inflammatory markers and leukocytosis have been thought to favor infective pathologies, but they do not seem to be good differentiating factors here, with 53.2% of our patients having leukocytosis. The absence of elevated β -hCG levels can help in ruling out ectopic pregnancy $^{(6)}$.

Due to diagnostic difficulty, six patients were managed surgically as acute appendicitis that

subsequently resulted in intra-operative consults. This is less than ideal, as some hospitals may not have Gynecology units. Hence, we are of the opinion that increasing awareness of ruptured corpus luteum in clinicians involved in acute care would optimize patient management.

Case reports have been made of ruptured corpus luteum being the first presentation of underlying bleeding diathesis^(7,8). In our study, two patients with cyst size of 15mm and 20mm, and hemoperitoneum of 2L and 1L respectively, had a diagnosis of idiopathic thrombocytopenic purpura (ITP) made during the admission. ITP can predispose to hemorrhage, and massive hemoperitoneum with rupture of the corpus luteum^(7,8). Therefore it is worthwhile to perform a full blood work-up for apparently healthy patients diagnosed with a ruptured hemorrhagic corpus luteum, especially if the amount of hemoperitoneum is large.

Ovarian hyperstimulation syndrome (OHSS) can be an iatrogenic complication in women undergoing fertility treatment, and they present with abdominal pain too. Hemoperitoneum in the pelvis could be mistaken for ascites on sonography. Although hemoperitoneum is differentiated from ascites by echogenic material or blood blots, in the very early or late stages, hemoperitoneum can be anechoic, making diagnosis difficult⁽⁹⁾. With regards to our study, two patients undergoing treatment with Clomiphene had bilateral multiple corpus luteum cysts with hemoperitoneum. They were initially managed conservatively as OHSS, but the pain persisted and worsened, and surgery was performed. Thus, worsening pain and decreasing hemoglobin levels are indications for laparoscopy to secure hemostasis.

Accepted management for hemodynamically stable patients includes conservative therapy of observation and analgesia. If ultrasound findings show a large amount of free fluid, along with severe abdominal pain, laparoscopy should be performed on admission. In circulatory collapse, direct laparotomy is mandatory⁽¹⁰⁾. Our study shows that patients who underwent prompt surgery before deterioration of symptoms gained pain relief and also recovered well post-operatively. Some

initially stable patients received conservative therapy, but continued to experience pain or deteriorated, at which point, there was surgical intervention.

With regards to recurrence, none required further surgical treatment. One patient was admitted two months after surgery with the same complaint, but recovered with analgesia and observation. Another complained of occasional mild abdominal pain well controlled by oral analgesia. As rupture can occur with any ovulatory cycle, the lifetime risk of recurrence may be considerable, and prevention is desirable to avoid life-threatening bleeds and other complications⁽⁸⁾. Eight patients were put on long-term oral contraceptive pills, and have remained symptom-free with ovulation suppression. The rest of our patients returned to having normal menses as before, and remained well.

To conclude, ruptured corpus luteum causing severe abdominal pain is indeed a diagnostic challenge. It takes a clinician with a high index of suspicion to diagnose, our study highlights these characteristics to aid diagnostic capability and optimize therapeutic management for patients. We recommend taking a targeted history with regards to the type of pain, phase of cycle, and preceding events. This diagnosis should also be considered in women in first trimester pregnancies. Physical examination and biochemical markers are largely non-specific. However, findings of free fluid on sonography are useful. In patients whose pain is progressively worse and causing hemodynamic instability, timely surgical intervention, with laparoscopy as first-line, unless cases of massive hemoperitoneum better managed by laparotomy, is called for and a definitive diagnosis made then. This can also be the first presentation of bleeding diatheses, requiring a full work-up. Ovulation suppression can prevent recurrences.

Acknowledgements

National University Health System, Department of Obstetrics and Gynecology for the database and help rendered.

Disclosure

Conflict of interest: The authors have no financial

interest to declare in relation to the content of this article.

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