Detection of unruptured ovarian pregnancy subsequently successfully treated by conservative laparoscopic surgery: a case report and review of the literature

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Summary

Early detection of ovarian pregnancy (OP) is essential for successful laparoscopic conservative surgery. However, early preoperative ultrasonography-based diagnosis is often difficult when fetal cardiac activity or the yolk sac is absent. The authors report a case of OP diagnosed at eight weeks gestational age in a natural pregnancy. The patient presented with amenorrhea and transient vaginal bleeding, and slight tenderness in the right ovary was noted during vaginal ultrasonography. Furthermore, ultrasonography showed a gestational sac (GS) without fetal cardiac activity or yolk sac, consistent with OP, and an adjacent compressible lutein cyst. The uterus, fallopian tubes, and left ovary were normal, and no cul-de-sac blood or ascites were found. Laparoscopy showed a two-cm mass partially covering the right ovary, which contained an unruptured GS. Subsequently, the mass was removed, and OP was histologically confirmed

Key words: Laparoscopic surgery; Ovarian pregnancy; Ultrasonography.

Introduction

Ovarian pregnancy (OP) is a rare form of ectopic pregnancy, constituting approximately three percent of all ectopic pregnancies [1]. Assisted reproductive technologies have been associated with an increased incidence of ectopic pregnancy. In cases of ectopic pregnancy following in vitro fertilization-embryo transfer (IVF-ET), the prevalence of OP has been reported to be six percent [2]. Early diagnosis of OP is mandatory to ensure the success of life-saving laparoscopic conservative surgery.

Several reports of unruptured OP in IVF-ET patients attributed successful treatment by laparoscopic surgery to early ultrasonography diagnosis, close follow-up, and awareness of the high-incidence of ectopic pregnancy, including OP, compared with natural pregnancy [3]. Diagnosing OP in the case of natural conception is difficult, especially when the date of conception is not known. Although a low serum human chorionic gonadotropin (HCG) level facilitates early recognition of abnormal implantation [3], repeat measurement is usually needed.

Transvaginal ultrasonography (TVUS) is an important tool for early detection of OP. Comstock et al reported that an echolucent ovarian area with a wide echogenic ring was a diagnostic sonographic finding of OP [4]. Here, the authors present a case where a lesion with this typical ring appearance compressed an adjacent echolucent sol of a corpus luteum cyst in the ovary.

Case Report

A 31-year-old nulligravida woman presented to the present hospital with amenorrhea and transient vaginal bleeding. She was undergoing prednisolone therapy (three mg, daily) for treatment of rheumatoid arthritis but had no history of pelvic inflammation, sur-

gery, or infertility. The chief complaint was a scant brownish vaginal discharge; there were no other symptoms. A rapid urine pregnant test yielded positive results, and the HCG level on the following day was 400 mIU/ml. The menstrual age was eight 4/7 weeks. Vaginal ultrasonography showed no intrauterine gestational sac (GS) and a normal left ovary without a lutein cyst (Figure 1a). There was no evidence of cul-de-sac fluid or pelvic adhesions. The right ovary was slightly enlarged, with a maximum diameter of 47 mm, and the patient complained of slight tenderness when it was pushed by the ultrasonography probe. In the right ovary, there was a echolucent area (diameter, 20 mm) surrounded by an echogenic ring. The ring was more echogenic than the ovarian stroma or adjacent corpus luteum, an appearance consistent with GS. No fetal cardiac activity or yolk sac was evident. A 25-mm corpus luteum cyst was compressed by the adjacent ovarian mass (Figure 1b). The findings suggested a provisional diagnosis of OP, and the patient elected surgical treatment.

During laparoscopy, the uterus and both fallopian tubes appeared normal (Figure 2a), and no ascites or adhesions were seen. The right ovary was enlarged because of a lutein cyst and a dark bluish mass with a smooth external surface (Figure 2b). Wedge resection was performed with monopolar electroscissors (Figure 2c), and the mass was removed through a ten-mm trocar using a retrieval bag. The resected tissue was cut in half (Figure 2d). Macroscopic examination revealed chorionic villi within the mass and negative tissue margins. The lutein cyst was confirmed by cutting the surface. The remaining right ovary was not oversewn. Uterine curettage showed no chorionic villi in the endometrium. The operative time was 80 minutes, and intraoperative blood loss was minimal. The patient had an uneventful recovery and was discharged on postoperative day five. Subsequent pathological diagnosis confirmed the diagnosis of OP. She became pregnant four months later and has experienced no complications.

Discussion

Spiegelberg published the first report describing the diagnostic criteria of OP. He stated that the fallopian tube on the affected side must be intact and separate from the ovary, the gestational sac must occupy the position of the ovary, the ovary must be connected to the uterus by the utero-ovar-

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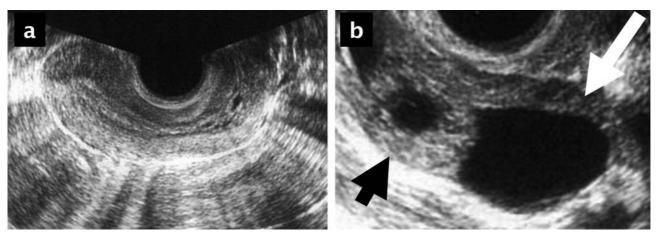


Figure 1. — Transvaginal ultrasonography scan. (a) Normal uterus with an empty endometrial cavity. (b) Coexisting well-defined GS (arrowhead) and lutein cyst (arrow); GS compressed an adjacent lutein cyst.

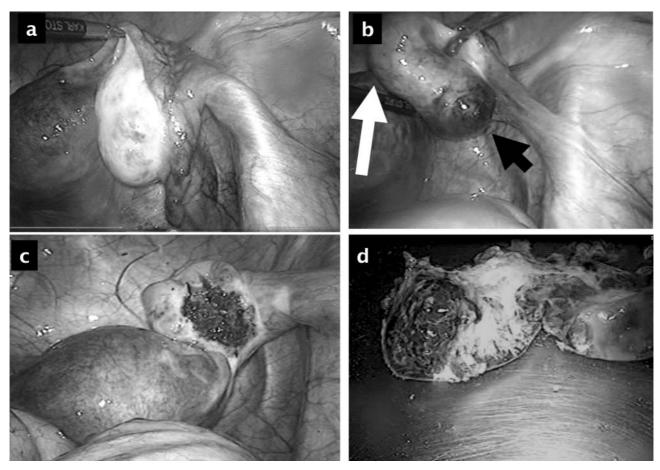


Figure 2. — Photograph of the right ovarian pregnancy during laparoscopic surgery. (a) Normal right fallopian tube. No adhesions were seen. In this view, the right ovary appears normal. (b) Right ovary (reverse side), showing the GS (arrowhead) and lutein cyst (arrow). (c) Wedge resection of GS. Cut surface, confirming lutein cyst. (d) Clos-eup view of chorionic villi within the resected GS.

ian ligament, and that ovarian tissue must be found in the gestational sac wall [5]. The risk factors for OP include history of prior gynecologic surgery, use of intrauterine contraceptive devices, assisted reproduction, or endometriosis [6]. The present case did not have known risk factors. Mar-

cus and Brinsden suggested that implantation in the ovary occurs after reverse migration of the fertilized egg [3]. According to this theory, the fertilized egg may have adhered to the ruptured follicle and thus remained in the ovary. Laparoscopic resection of the GS and preservation of remain-

Authors	n	Study period	Risk factors			Before surgery			At surgery	
			IUD use	Prior surgery or endometriosis	Post IVF follow-up	No symptom	OP diagnosed by US	Embryo or yolk sac detected by US	Un- ruptured	Wedge resection
Comstock et al. [4]	6	1990-2003	0	NA	0	0	NA	2 (33%)	4 (67%)	NA
Odejinmi et al. [7]	12	2003-2008	2 (17%)	NA	NA	NA	9 (75%)	NA	0	11 (92%)
Var et al. [10]	2	NA	0	0	0	0	1	1	2	2
Priya et al. [11]	2	2001-2005	0	0	2	1	2	2	0	1
Tobiume et al. [12]	3	NA	0	1	0	0	0	0	0	3
Choi et al. [6]	49	1996-2009	2 (4%)	NA	5 (10%)	9 (18%)	8 (16%)	NA	NA	39 (80%)
Koo <i>et al.</i> [13]	28	1996-2009	2 (7%)	18 (64%)	5 (18%)	3 (11%)	ΝA	17 (61%)	NA	26 (93%)

Table 1.— Summary of findings in reports of OP. Studies published since 2000, excluding single case reports. Data reported by Choi and Koo for patients in the same institute during the same period. n, number of patients; IUD, intrauterine contraceptive device; OP, ovarian pregnancy; US, ultrasonography; NA, not available.

ing ovarian tissue is the preferred treatment for ovarian pregnancy [7-15].

Early diagnosis of OP is vital to prevent emergency invasive procedures, serious complications, or death. Raziel *et al.* reported that OP was diagnosed after laparoscopic examination or direct laparotomy in 20 patients from 1971 to 1989 [1]. Odejinmi et al and Choi *et al.* reported that 75% and 16% cases of OP, respectively, were diagnosed before surgery [6, 7]. These findings are summarized in Table 1.

Technical advances in ultrasonography and the development of more sensitive methods for HCG detection facilitate earlier and more accurate non-invasive diagnosis of ovarian pregnancies. The finding of a round echogenic ovarian mass on ultrasonography in a pregnant patient suggests the diagnosis of OP [4, 7]. However, once rupture occurs, the GS may resemble a hemorrhagic corpus luteum sonographically and even macroscopically at surgery. In this present case, the coexistence within the ovary of an unruptured GS without fetal cardiac activity or yolk sac and of a lutein cyst made the ultrasonography-based diagnosis relatively easy. The coexistence of GS and a lutein cyst within the ovary suggested OP. Compressibility also helped to distinguish between a GS and lutein cyst. The authors believe that the "coexistence and compression" sign might be useful for the early diagnosis of OP.

This conservative procedure avoids excessive removal of healthy ovarian tissue and allows young patients to maintain their reproductive capability. In the present patient, laparoscopic surgery was successfully conducted before rupture of OP. To avoid misdiagnosis, frozen section analysis of specimens obtained at the time of surgery is advisable [16] in order to detect chorionic villi in the surgical specimen.

Methotrexate is an effective therapeutic option for the medical management of unruptured ectopic pregnancy. However, in cases where diagnostic laparoscopy is necessary, definitive surgical management is easily performed at the same time [17-19]. Therefore, surgery is currently the mainstay for management of OP.

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