



Successful laparoscopic management of uterine serosal pregnancy

Gee Hee Seo¹, Hyun Jung Lee¹, Ji-Hyun Jang¹, Min Chul Choi², Chan Lee², Gwangil Kim³

¹Department of Obstetrics and Gynecology, ²Comprehensive Gynecologic Cancer Center, Department of Obstetrics and Gynecology, ³Department of Pathology, CHA Bundang Medical Center, CHA University, Seongnam, Korea

Uterine serosal pregnancy is an extremely rare form of ectopic pregnancy. This is a report of a 35-year-old primigravida woman who was diagnosed with uterine serosal pregnancy via laparoscopic intervention. A 35-year-old woman (gravida 1, para 0) was referred from a local clinic for a ruptured left tubal pregnancy at amenorrhea 5+0 weeks with elevated serum beta human chorionic gonadotropin (16,618 mIU/mL). A pregnancy on the left posterior wall of the uterine serosa was diagnosed during the operation and successfully treated with laparoscopic surgery as a conservative management strategy to enable fertility preservation. With the advantages of ultrasonography and laparoscopy, an early diagnosis of a primary abdominal pregnancy located on the left posterior wall of the uterine serosa was made, prior to the occurrence of severe intra-abdominal massive hemorrhage, which was then treated laparoscopically as a conservative management strategy enabling the preservation of fertility.

Keywords: Pregnancy, abdominal; Pregnancy, ectopic; Uterine serosal pregnancy

Introduction

Abdominal pregnancy is a very rare form of ectopic pregnancy where implantation occurs in the peritoneal cavity. While ectopic pregnancy accounts for 2% of all pregnancies, abdominal pregnancy accounts for 1% to 4% of all ectopic pregnancies [1]. Early diagnosis of an abdominal pregnancy is often delayed since it cannot be diagnosed until there is a sign of ectopic mass rupture, which results in high maternal morbidity and mortality due to massive bleeding [1]. The associated maternal mortality rate of abdominal pregnancy is 7.7 times greater than that of tubal pregnancy, and about 90 times higher than that of normal pregnancy [2]. Although recent advances in imaging modalities, such as ultrasonography and magnetic resonance imaging, have enabled the early diagnosis of abdominal pregnancy, definitive diagnosis is based on surgery [3]. Therefore, diagnostic surgical intervention should be considered as a first-line intervention if there is any sign of suspected abdominal ectopic pregnancy.

Uterine serosal pregnancy is an extremely rare form of abdominal ectopic pregnancy, wherein the fetus is implanted within the uterine serosa, without a connection to the endometrial cavity, fallopian tubes, or round ligament [4]. Neither the exact mechanism nor the cause of uterine serosal

pregnancy is well understood, and only few cases have been reported thus far [4-6]. Here, we report a case of 35-year-old primigravida woman who was diagnosed with uterine serosal pregnancy via laparoscopic intervention.

Case report

A 35-year-old woman (gravida 1, para 0) with the chief complaint of lower abdominal pain was referred from a local clinic. The presumed diagnosis was ruptured left tubal pregnancy

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Corresponding author: Min Chul Choi
Comprehensive Gynecologic Cancer Center, Department of
Obstetrics and Gynecology, CHA Bundang Medical Center, 59
Yatap-ro, Bundang-gu, Seongnam 13496, Korea
Tel: +82-31-780-6191 Fax: +82-31-780-6194
E-mail: oursk79@cha.ac.kr
<http://orcid.org/0000-0001-5858-4748>

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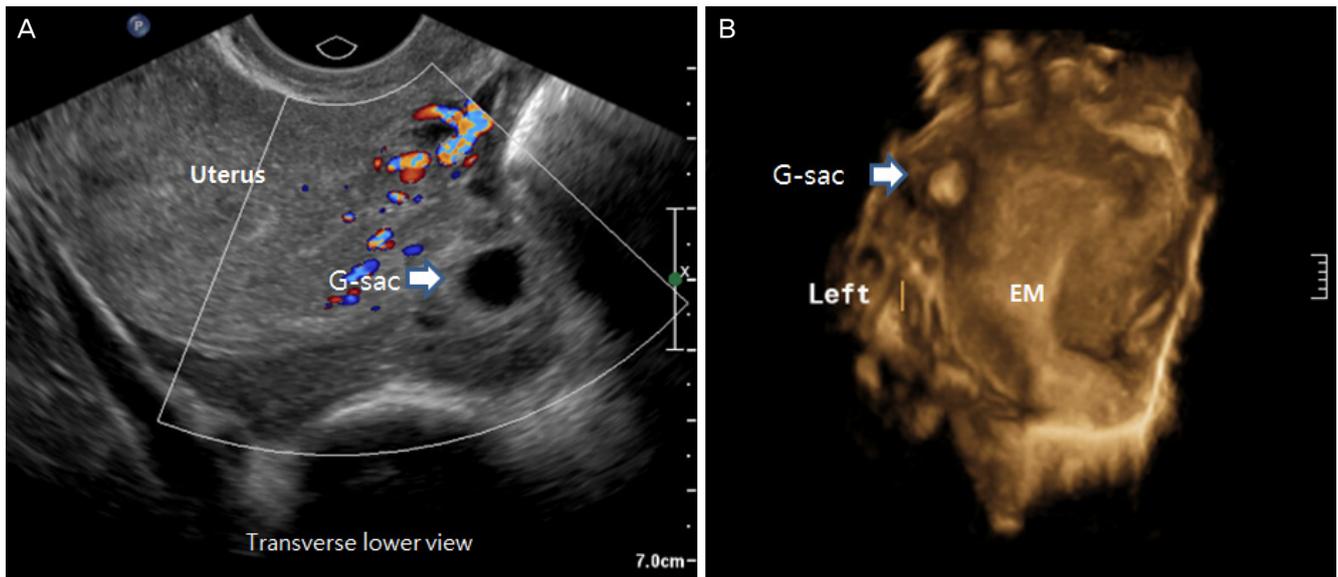


Fig. 1. (A) Preoperative two-dimensional transvaginal sonogram revealed a gestational sac (G-sac) that was separated from the uterus, with a certain amount of hemoperitoneum, suggesting the presence of an ectopic pregnancy. (B) Three-dimensional transvaginal sonogram revealed a 1.8-cm mass located adjacent to the left cornus, clearly outside the endometrium (EM).

at amenorrhea 5+0 weeks with elevated serum beta human chorionic gonadotropin (16,618 mIU/mL). The patient had no prior medical or surgical history, no history of intrauterine device use, and no history of any assisted reproductive techniques or medications.

Her vital signs were stable and her abdomen was somewhat rigid with tenderness and rebound tenderness noted in the left lower quadrant and suprapubic area. On pelvic exam, minimal cloudy discharge at the cervical os was noted with mild cervical motion tenderness. There was no sign of vaginal bleeding. The laboratory studies were within normal limits including a hemoglobin level of 12.7 g/dL, and serum beta human chorionic gonadotropin level of 17,143 mIU/mL.

The result of a two-dimensional (2D) transvaginal ultrasound (TVS) revealed a gestational sac that was separated from the endometrium along with fluid in the cul-de-sac, suggesting the presence of an ectopic pregnancy. No embryonic pole or yolk sac was seen in the gestational sac. Further investigation by three-dimensional (3D) TVS demonstrated a 1.8-cm mass located adjacent to the left cornus (Fig. 1). With the preoperative impression of left cornual pregnancy with a partially ruptured status, the patient underwent emergent diagnostic laparoscopy.

Entering the abdominal cavity, a hemoperitoneum of approximately 300 mL was noted, and a 2x2 cm ruptured ectopic mass was dangling from the left posterior wall of the

uterine serosa that was distinct from both fallopian tubes, causing active bleeding (Fig. 2). Grossly, the ectopic mass was confined within serosa exclusively and lacked myometrium; the mass was visible through a thin layer of serosal coverage only. There was no bleeding from either fimbria. The left adnexa seemed to be normal except for one small paratubal cyst. The right ovary was adherent the round ligament and three paratubal cyst was noted. All other pelvic organs were grossly normal.

After the diagnosis of uterine serosal pregnancy, surgery was performed with monopolar diathermy scissors, forcing the removal of gestational tissue on the uterine serosa. The ectopic mass was successfully removed via scraping after confirming no remnant chorionic villi tissue. Visible vessels were coagulated with bipolar diathermy, and the uterine scar was meticulously sutured using Vicryl 2-0 (Fig. 2). The specimen was placed in a bag and completely removed from the pelvic cavity. In addition, paratubal cystectomy was performed on the right and left adnexa, along with adhesiolysis on the right ovary that was adherent to the round ligament. Tubal patency was confirmed by passing methylene blue dye through the bilateral fallopian tubes. Dilation and evacuation (D&E) was done to rule out possibility of intrauterine or heterotrophic pregnancy. The operating time was 50 minutes, and the estimated blood loss was minimal. The postoperative recovery was uneventful.

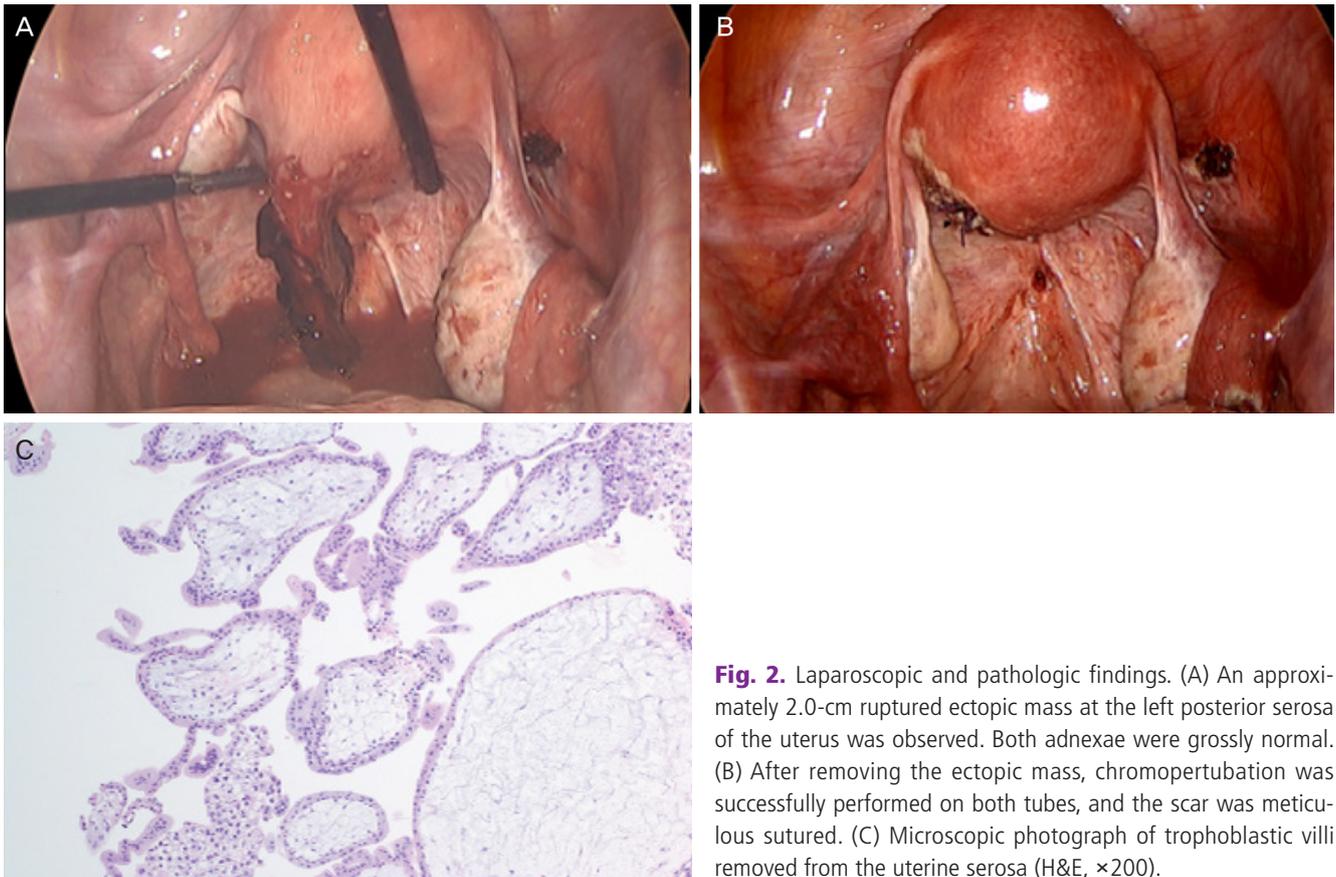


Fig. 2. Laparoscopic and pathologic findings. (A) An approximately 2.0-cm ruptured ectopic mass at the left posterior serosa of the uterus was observed. Both adnexae were grossly normal. (B) After removing the ectopic mass, chromopertubation was successfully performed on both tubes, and the scar was meticulous sutured. (C) Microscopic photograph of trophoblastic villi removed from the uterine serosa (H&E, ×200).

The pathologic reports confirmed an ectopic pregnancy implanted in the serosa of the uterine posterior wall as the trophoblastic villi were seen microscopically from the removed uterine serosal ectopic mass (Fig. 2). Uterine endometrium from dilation and evacuation confirmed to be gestational endometrium without trophoblast. The serum beta human chorionic gonadotropin level gradually decreased to within the normal range (1,429 mIU/mL at postoperative day 7, 79.47 mIU/mL at postoperative day 14, and 4.87 mIU/mL at postoperative day 28).

Discussion

Abdominal pregnancy accounts for 0.6% to 4% of all ectopic pregnancies [7,8]. Given its rarity, abdominal pregnancy is commonly misdiagnosed, and delayed diagnosis leads to high mortality and morbidity due to massive bleeding.

Abdominal pregnancy is classified as either primary or secondary abdominal pregnancy. Primary abdominal pregnancy is defined if implantation occurs initially in the abdomen with

normal adnexae and no evidence of injury or uteroplacental fistula [9]. Secondary abdominal pregnancy is defined if implantation first occurs in the tube, ovary, or uterus, then leads to tubal rupture, and eventually results in subsequent implantation in the abdominal cavity [3,9,10]. Most reported cases are the secondary type, and primary abdominal pregnancy is very rare [8]. However, it is difficult to differentiate between primary and secondary abdominal pregnancy since the pathogenesis and etiology of abdominal pregnancy remain unclear.

Considering that all other pelvic organs and both adnexae were macroscopically normal in the present case, with only the ectopic mass in the serosa causing active bleeding, we could conclude that the uterine serosa was the primary implantation site of the pregnancy. We do not exactly know whether the pregnancy confined within serosa or involved myometrium layer microscopically, based on the pathological finding, as the tissue was not viable for further pathologic evaluation. The free fluid observed on ultrasonography, and the intraoperative findings of a ruptured ectopic mass in the left posterior wall of the uterine serosa, with grossly normal adnexae and other

pelvic organs, were highly suggestive of a primary abdominal pregnancy. However, these findings could not completely confirm that it was a primary abdominal pregnancy, since they were based on intraoperative findings only, without any clear evidence of the pathogenesis or etiology leading to the implantation of a gestational sac on the uterine serosa.

According to the definition of primary abdominal pregnancy, two possible hypotheses of etiology were made for this case. First, the gestational sac was initially implanted in the endometrium, invaded into the myometrium through a microscopic tract (usually formed by previous uterus trauma or focus of adenomyosis), and eventually implanted on the uterine serosa [11]. The possible risk factors for abdominal pregnancy include prior uterine surgery history in vitro fertilization, adenomyosis, pelvic inflammation history, low socioeconomic class, endometriosis, and intrauterine devices, among others [4]. Previously reported uterine serosal pregnancy in 2006 [4] and 2012 [5], had several risk factors that have been associated with uterine serosal pregnancy. Among these possible risk factors for uterine serosal pregnancy, there was no clearly identified risk factor in our case. There was a case report of uterine serosal pregnancy without any risk factor that could cause serosal pregnancy in 2005 [6], similar to our case. Therefore, the first hypothesis does not correlated with the present patient clinic-surgical condition; therefore, the possibility that the gestational sac had invaded through a microscopic tract could be excluded.

Second, the retrograde passage of a fertilized embryo via the fallopian tubes to the intra-abdomen region with subsequent migration through the lymphatic channels [12,13]. The second hypothesis is supported by the study of Liang et al. wherein a contrast-enhanced computer tomography was used to demonstrate the route of embryonic migration in a retroperitoneal ectopic pregnancy via the lymphatic vessels [12]. The study suggested that the dissemination of the embryo occurred through the lymphatic channels, involving a mechanism similar to that of endometrial cancer metastasis via the periaortic lymphatic channels [12]. In our case, the intraoperative finding of grossly normal adnexae supports that the migration of the gestation sac could have resulted from retrograde menstrual flow, derived from delayed ovulation, which could send the fertilized ovum through the fallopian tubes into the abdomen [13]. Then, the zygote might have been carried by intraperitoneal fluid or by the lymphatic channels and then implanted on the uterine serosa [13]. If

pathologic finding of present case showed a trophoblast surrounded by lymphatic tissue, we could have had strong evidence for this hypothesis [13].

The most common diagnostic tool for ectopic pregnancy is 2D TVS, but it may also require 3D TVS to enable more accurate localization of the gestational sac [11]. In our case, 2D TVS showed a gestational sac outside the endometrium with fluid in the cul-de-sac, which prompted the initial suspicion of ectopic pregnancy. Previously reported uterine serosal cases mostly used 2D TVS as diagnostic tool. Despite initial diagnosis of cornual pregnancy, we tried to use 3D TVS for more accurate diagnosis. 3D sonographic images revealed that the gestational sac was located outside the endometrium at the left cornus. 3D TVS provided a distinct view of the gestational sac, as it can quasi-simultaneously display multiple scan planes, which offers superior visualization of the endometrial cavity and myometrium than that of 2D TVS [14,15].

In the past, laparotomy was performed in most cases of abdominal pregnancy [2]. These days, laparoscopy is the standard management strategy as it can be easily converted to laparotomy if needed. In the present case, the diagnosis of ectopic pregnancy was made at early gestational stage when the patient was still hemodynamically stable; therefore, we opted for laparoscopic treatment. Since the patient was nulliparity, fertility sparing was especially considered important factor. In order to preserve fertility, the early diagnosis and minimal invasiveness of the procedure are crucial. The factors enabling fertility sparing surgery mostly depends on patient condition, including the implantation site of ectopic mass (amount of vascular supply involved and depth of invasion). Gestational ages above 20 weeks or implantation sites with high vascular supply are difficult to preserve uterus as it lead to massive hemorrhage [4]. Fortunately, the present case was not difficult to remove the ectopic mass and preserve the uterus as the diagnosis was made at early gestation age, with the mass surrounded by serosal layer only without massive hemorrhage, which allows conservative surgical treatment [4].

Uterine serosal pregnancy is a rare form of ectopic pregnancy, and the diagnosis is seldom made prior to ectopic mass rupture or surgery. Herein we report a case of uterine serosal pregnancy treated laparoscopically with successful fertility sparing without intra-operative massive hemorrhage.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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