Case Report

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Persistent low-level elevation of serum human chorionic gonadotropin after termination of pregnancy: a rare case of peritoneal trophoblastic implant

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Peritoneal trophoblastic implant can occur after treatment of ectopic pregnancy. Similarly, after termination of intrauterine pregnancy, trophoblastic implants are rare but can be a complication of perforation during dilatation and curettage. We report an extremely rare case of trophoblastic implant on the myometrium, ovarian surface, and peritoneal wall 4 months after uncomplicated dilatation and curettage. To the best of our knowledge, this is the first case of peritoneal trophoblastic implant following dilatation and curettage without uterine perforation. Knowledge of this case is useful for the management of patients with persistent low-level elevation of serum human chorionic gonadotropin after termination of pregnancy.

Keywords: Chorionic gonadotropin; Trophoblast; Dilatation and curettage

Introduction

Persistent low-level elevation of serum β-human chorionic gonadotropin (β-hCG) is associated with several conditions. Indeed, β-hCG can be elevated in response to benign conditions and can return to the normal range spontaneously or with treatment [1-3]. However, elevated β-hCG can also occur under active malignant conditions such as gestational trophoblastic neoplasia (GTN) [4]. It is not currently possible to distinguish between innocuous and severe conditions based only on β-hCG level.

One condition that is associated with persistent low-level elevation of β-hCG is trophoblastic implant in the abdominal cavity, which is a rare finding after treatment of ectopic pregnancy [5] and an extremely rare event following termination of intrauterine pregnancy. To date, only 3 cases of peritoneal trophoblastic implant have been reported; these implants occurred in the myometrium and sigmoid colon after surgically-induced first trimester termination of pregnancy, and all of these cases were associated with uterine perforation during dilatation and curettage [6-8].

We report a rare case of trophoblastic implant on the myometrium, ovarian surface, and peritoneal wall 4 months after uncomplicated dilatation and curettage for a blighted ovum. To the best of our knowledge, this is the first case of peritoneal trophoblastic implant following dilatation and curettage without uterine perforation.

Case report

A 39-year-old woman was referred to our clinic for persistent elevated β-hCG after surgical evacuation of a blighted ovum. She complained of amenorrhea combined with intermittent vaginal spotting and lower abdominal discomfort. She had an obstetric history of 2 full-term vaginal deliveries and one

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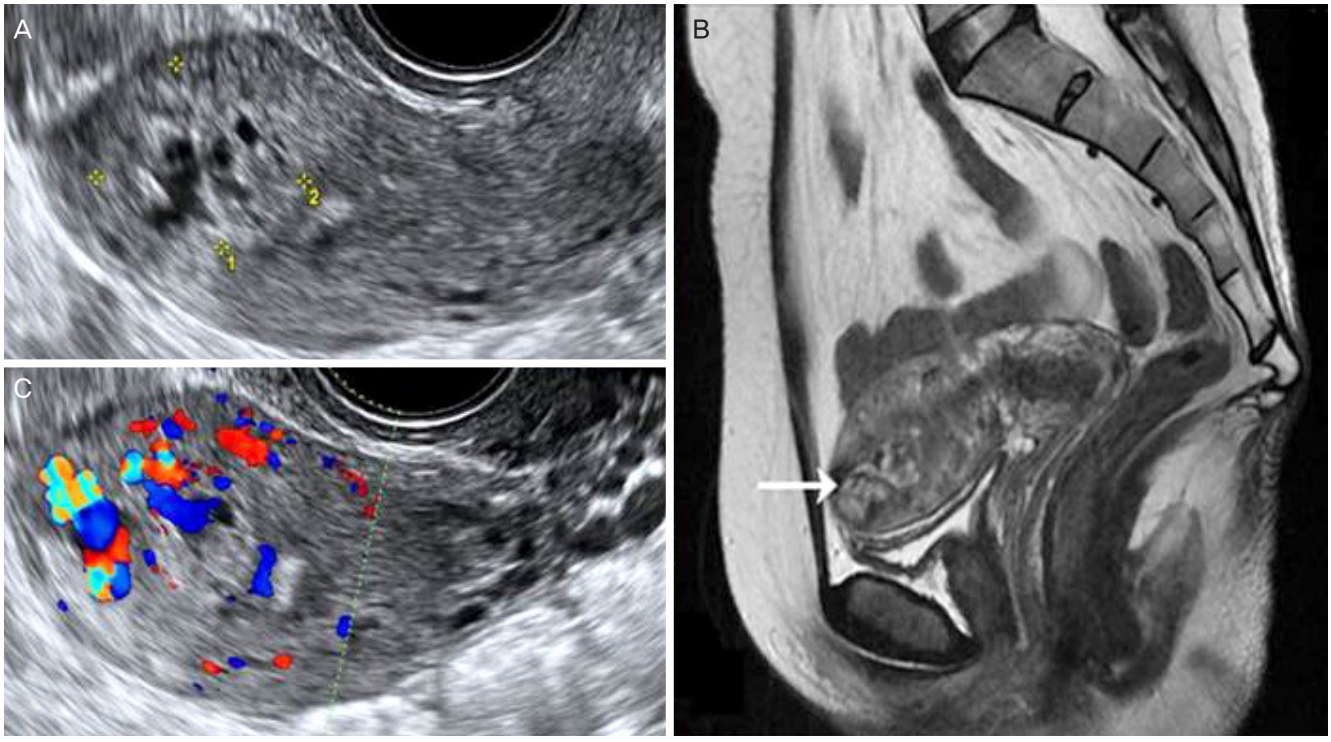

Fig. 1. (A) Transvaginal ultrasound image showing a 2.00×2.18 cm mixed echoic mass in the myometrium of the uterine fundus. (B) Color Doppler examination reveals increased flow to the mass. (C) Sagittal T2-weighted pelvic magnetic resonance imaging showing a 2.3×1.3 cm mass with heterogeneous signal intensity in the uterine fundus.

induced abortion. Fourteen weeks earlier, she had undergone surgical curettage for a blighted ovum at 7 weeks of gestation at a local clinic. Physical examination was normal except mild low abdominal tenderness, and vital signs were normal. Her serum β-hCG was 95 mIU/mL. A review of her previous sonography, performed before curettage, revealed a gestational sac with a yolk sac in the endometrial cavity, a diagnosis compatible with a blighted ovum. We telephoned the obstetrician who had performed the curettage and confirmed that the previous procedure was uneventful with no suspicious uterine perforation. Transvaginal sonography revealed an empty uterus and a 2-cm mixed echoic mass in the uterine fundus (Fig. 1A). Color Doppler evaluation demonstrated abundant arterial and venous flow on the mass (Fig. 1B). Magnetic resonance imaging (MRI) showed a 2.3×1.3 cm mass lesion of heterogeneous signal intensity with many tortuous tubular signal voids in the uterine fundus. The endometrial-myometrial junction was intact (Fig. 1C); the mass was heterogeneously well enhanced after contrast material infusion. Also, multiple small cysts in left ovary were noted with MRI.

After conservative care including analgesics for 2 weeks, her β-hCG level had slightly decreased to 73 mIU/mL but her abdominal discomfort was persistent, and the mass on ultrasonography had increased to 2.7×2.0 cm. At this time, the possible differential diagnosis included placenta site trophoblastic tumor (PSTT), heterotopic pregnancy, and remnant placenta. Diagnostic laparoscopy was planned in order to acquire samples for histology. We suggested resection of the mass, but the patient opted for total hysterectomy due to fear of malignancy and further pregnancy.

Laparoscopy revealed a 3-cm brown and bluish mass on the fundus and multiple small bluish round cystic implants of 5–10 mm on the left ovary and left broad ligament (Fig. 2). We performed a laparoscopic total hysterectomy and excised all cystic implants. Histologic examination of the bluish masses showed normal trophoblastic tissue with degenerated and necrotic chorionic villi in the hemorrhagic background. This mass was located within left fundus without disruption of endometrial-myometrial junction. The postoperative course was uneventful, and β-hCG was <5 mIU/mL at 13 days after surgery.

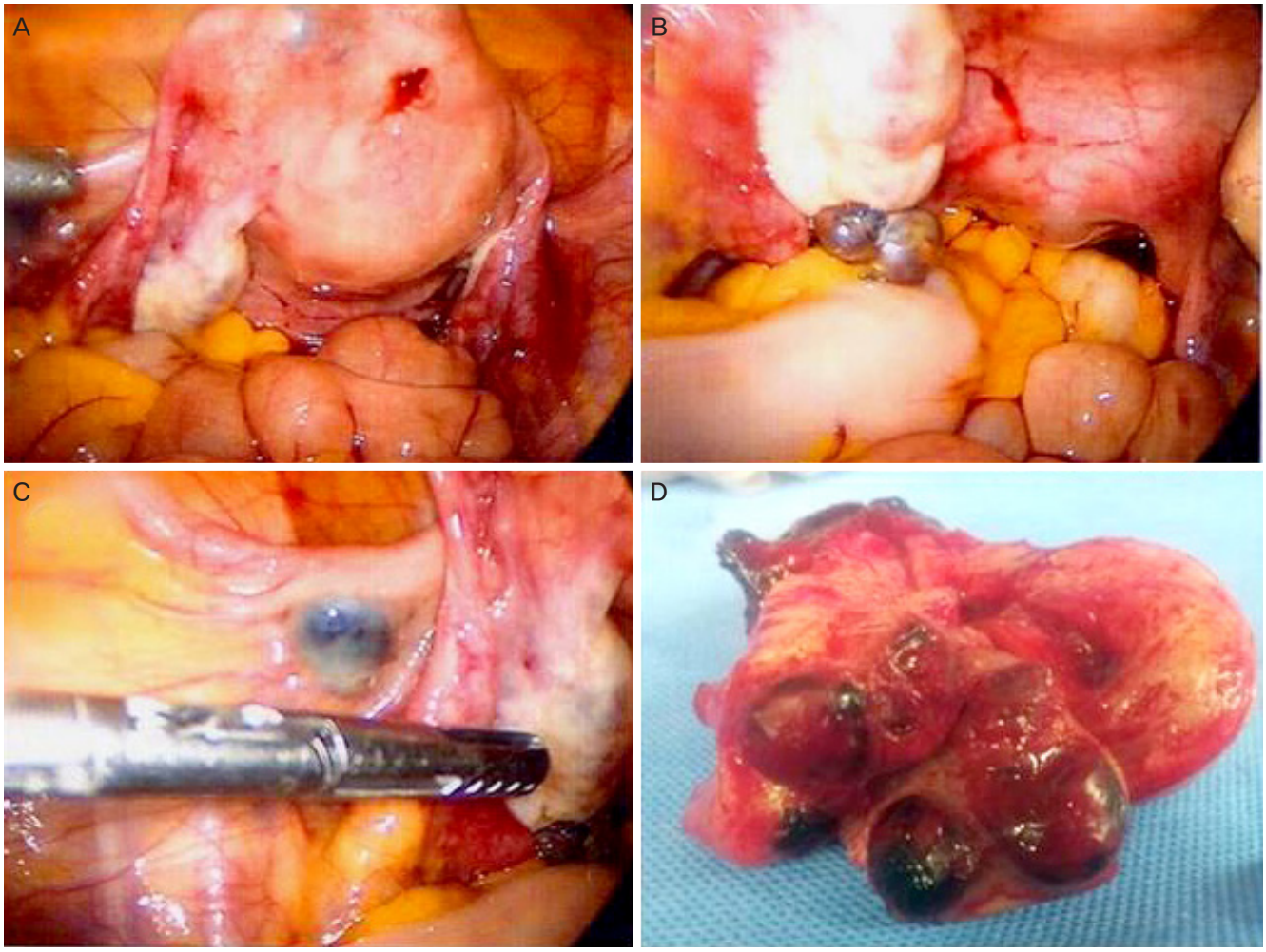

Fig. 2. Laparoscopic findings showing brown and bluish masses on the fundus (A), left ovary (B), and left broad ligament (C). A cross-section of the uterus showing a hemorrhagic cystic mass in the fundal myometrium from the endometrial cavity (D).

Discussion

Persistent peritoneal trophoblastic implants are not suspected after dilatation and curettage. Indeed, this is a rare complication that occurs mainly after treatment of ectopic pregnancy, and most cases occur in the original tubal site of the ectopic pregnancy after salphingostomy [5]. It is also possible after salphingectomy or at different peritoneal and visceral sites [5,9]. Herein, we present an extremely rare case of trophoblastic implant after termination of an intra-uterine pregnancy. A few cases of peritoneal trophoblastic implants after termination of pregnancy have been reported, and all of these reports were associated with uterine perforation during dilatation and curettage [6-8]. After the procedure, the trophoblastic tissue spread along the perforated tract into the myometrium and the peritoneal cavity.

In this case, there was no uterine perforation or false cannulation of the endometrial cavity that could be recognized by the operator during uterine curettage. After the procedure, the patient's recovery process was typical. However, the finding of trophoblastic tissue implanted in the ovary, pelvic wall, and myometrium after 4 months shows that peritoneal trophoblastic implant should be considered in cases with persistent elevated β-hCG, even after uncomplicated surgical curettage.

This case of peritoneal trophoblastic implant after uncomplicated curettage might have occurred through the salphinx. Considering that the peritoneal implant occurred on the left ovary surface and the left pelvic wall, the possibility of trophoblast spread through the left salphinx is possible. The

pathogenesis of the myometrial implant is more difficult to deduce, and the postoperative gross examination of the uterus showed that the uterine lesion was contiguous with the endometrial cavity but completely separate; it also protruded toward the serosa. That is to say, the possibility of spreading from the pelvic cavity to the uterine serosa cannot be excluded. Of course, there is a possibility that the trophoblast spread to the myometrium and pelvic cavity through a minor crack in the endometrial junctional zone, which could have occurred during dilatation and curettage without recognition. More cases will be needed to clarify the pathophysiology of trophoblastic peritoneal implants.

Persistent low-level elevation of β-hCG after termination of a pregnancy is associated with benign and malignant conditions [1-3]. Persistent low-level elevation of β-hCG is empirically defined as less than 250 IU/L for more than 3 months with the possibility of tumor or abandoned pregnancy [2,10]. GTN is a malignant condition that can be classified as an invasive mole, choriocarcinoma, PSTT, or epithelioid trophoblastic tumor (ETT) [11]. Approximately 7.6% of patients with persistent low-level elevation of β-hCG have active malignancies [2], and it is very important to identify when low-level elevation of β-hCG is due to malignancy.

Among GTNs, invasive moles and choriocarcinoma are straightforward to diagnosis as they have a relatively high level of β-hCG [11]. On the contrary, PSTT and ETT are associated with a low level of β-hCG combined with a nonspecific hypervascular or hypovascular tumor in the myometrium, which is similar to the findings in our case [11,12]. When a tumor is present, MRI can be efficient for the evaluation of the vasculature, location, and extent of the tumor. However, imaging findings are non-specific, and it is difficult to discriminate tumors from benign condition such as remnant of conception or ectopic pregnancies [13].

In this case, we selected surgical treatment instead of methotrexate treatment or observation, as the patient had a myometrial mass requiring pathologic examination accompanied with elevated β-hCG. Management of patients with persistent low-level elevation of β-hCG is still controversial, and a few studies are investigating management strategies. According to analysis of 99 patients at the β-hCG Reference Service Center in the USA, chemotherapy is unnecessary and ineffective in the early stage of persistent low-level elevation of β-hCG because of the possibility of spontaneous cure [2]. However, if there is radiologic evidence of a mass, treatment is recommended, though there are no specific guidelines on whether to perform surgery or chemotherapy [1-3]. Generally, unless there is evidence of a definite malignancy, it can be helpful to try chemotherapy first and see if β-hCG returns to a normal level. As PSTT and ETT are chemoresistant, the response to chemotherapy is more associated with a benign condition than malignancy [11,12].

In previous reports of peritoneal trophoblastic implant after termination of pregnancy, surgical resection of implants was performed in all 3 cases [6-8]. Also, most cases of trophoblastic implant after ectopic pregnancy were treated with surgical excision because of sonographic evidence of an abnormal tumor in the pelvic cavity or a symptom of hemoperitoneum [5]. However, there is a possibility that several cases of trophoblastic implant have been treated with methotrexate chemotherapy or resolved spontaneously without recognition.

We report a rare complication of peritoneal trophoblastic implant following dilatation and curettage. In order to identify other cases like this, it is important to follow-up with a β-hCG titer after dilatation and curettage, even if it is an uneventful procedure. For patients with persistent low-level elevation of β-hCG combined with a hypervascular mass in the pelvic cavity, diagnosis of trophoblastic implant and methotrexate treatment should be considered.

Conflict of interest

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

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