CASE REPORTS  
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Case report

EARLY PRIMARY ABDOMINAL PREGNANCY IMPLANTED IN THE VESICOOUTERINE POUCH – A CASE REPORT

RANA PRIMARNA ABDOMINALNA TRUDNOĆA IMPLANTIRANA U VEZIKOUTERUSNOM ŠPAGU – PRIKAZ SLUČAJA

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Introduction

An abdominal pregnancy is a rare form of ectopic pregnancy and potentially life-threatening condition. It is difficult to make an early diagnosis of abdominal pregnancy. Case Report. We present a case of early primary abdominal pregnancy, diagnosed at 6th gestational week, located in the vescicouterine pouch and treated laparoscopically. Despite the rapidly decreasing serum β-human chorionic gonadotropin levels, the presence of the intraperitoneal blood allowed neither expectant management nor medical treatment, although the patient was hemodinamically stable at that moment. The absence of significant bleeding during the surgery and histopathological finding of placental villi with necrosis confirmed that, in this case, the abdominal pregnancy was already the subject of spontaneous involution. Conclusion. High index of suspicion and carefully interpreted clinical and ultrasound findings are crucial for timely diagnosis of early abdominal pregnancy before the occurrence of massive and potentially fatal intraperitoneal bleeding.

Summary

Introduction. An abdominal pregnancy is a rare form of ectopic pregnancy and potentially life-threatening condition. It is difficult to make an early diagnosis of abdominal pregnancy. Case Report. We present a case of early primary abdominal pregnancy, diagnosed at 6th gestational week, located in the vescicouterine pouch and treated laparoscopically. Despite the rapidly decreasing serum β-human chorionic gonadotropin levels, the presence of the intraperitoneal blood allowed neither expectant management nor medical treatment, although the patient was hemodinamically stable at that moment. The absence of significant bleeding during the surgery and histopathological finding of placental villi with necrosis confirmed that, in this case, the abdominal pregnancy was already the subject of spontaneous involution. Conclusion. High index of suspicion and carefully interpreted clinical and ultrasound findings are crucial for timely diagnosis of early abdominal pregnancy before the occurrence of massive and potentially fatal intraperitoneal bleeding.

Key words: Pregnancy, Abdominal; Pregnancy, Ectopic; Laparoscopy; Early Diagnosis; Douglas' Pouch; Ultrasonography; Pregnancy Complications

Sažetak

Uvod. Abdominalna trudnoća je retka forma ektopične trudnoće i stanje koje potencijalno ugrožava i život. Rana dijagnoza abdominalne trudnoće se teško postavlja. Prikaz slučaja. Prikazujemo slučaj rane primarne abdominalne trudnoće, dijagnostikovane u šestoj nedelji gestacije, koja se nalazila u vescicouterinom špagu i bila tretirana laparoskopski. Upkros brzom padu serumskog β-humanog horionskog gonadotropina, prisustvo krvi u peritonealnoj duplji nije dopuštalo ni ekspektativni pristup niti medicinski tretman, mada je pacijentkinja bila hemodinamički stabilna u datom trenutku. Odstupstvo signifikantnog krvarenja tokom intervencije i patohistoloških nalaza placentnih čupica sa nekrozom potvrdili su da je, u ovom slučaju, abdominalna trudnoća već bila subjekat spontane involucije. Zaključak. Za blagovremeno postavljanje dijagnoze rane abdominalne trudnoće, pre nego što nastupi mastišno i potencijalno fatalno intraperitonealno krvarenje, od suštinskog je značaja da se ima na umu ova mogućnost i pažljivo interpreta klinički i ultrazvučni nalazi.

Ključne reči: Abdominalna trudnoća; Ektopična trudnoća; Laparoskopija; Rana dijagnoza; Duglasov špag; Ultrasonografi; Komplikacije trudnoće

Introduction

The primary abdominal pregnancy is defined as an ectopic pregnancy developing on the serosal surface within the peritoneal cavity excluding tubal, ovarian, intraligamentous pregnancies and secondary implantation of primary tubal implantation [1]. Original Studford’s diagnostic criteria are: 1. normal tubes and ovaries with no evidence of recent or remote injury; 2. no evidence for uteroperitoneal fistula; 3. the pregnancy related solely to the peritoneal surface and young enough to eliminate the possibility of secondary implantation following a primary nidation in the tube; 4. no evidence of secondary implantation following initial primary tubal nidation [1, 2].

Abdominal pregnancies are very rare, accounting for only 1.3% of all ectopic pregnancies [3]. It is estimated that the primary abdominal pregnancy occurs in 10.9 per 100,000 pregnancies and in 9.2 per 1,000 ectopic pregnancies [4]. The maternal mortality rate is 5.1 per 1000 pregnancies; 7.7 times higher than in tubal ectopic pregnancies and 90 times higher than in an intrauterine pregnancy [1,4]. Here we present the case of early primary abdominal pregnancy located in the vescicouterine pouch and partly on the peritoneal surface of the bladder.
Case Report

A 35-year old gravida 0, para 0 with a 3-year long history of unexplained infertility presented to our Department for treatment. The patient had no history of pelvic inflammatory disease, use of intrauterine devices or gynecological surgery other than diagnostic laparoscopy performed two years before, which demonstrated patent Fallopian tubes and no pathological findings. She was otherwise healthy. Menarche occurred in her thirteenth year and menstrual cycles were regular. Her last menstrual period was 38 days before and in that cycle she did not receive any infertility treatment. On admission, the patient suffered from constant lower abdominal pain, she was pale, but hemodynamically stable. The examination of her cardiac and respiratory systems was unremarkable. Her abdomen was soft, but with mild suprapubic tenderness. The speculum examination indicated the presence of a single cervix, without pathological findings and no bleeding from cervical channel. The bimanual pelvic examination revealed the slightly enlarged soft uterus and tender palpable mass, about 4 cm in diameter in front of the uterus. There were no palpable pathological findings on adnexal regions. The transvaginal ultrasound examination [Toshiba Nemo XG, 6 MHz] showed an empty uterus with 5 mm endometrial strip. A cystic mass, 3 cm in diameter, filled with dense content, was seen in front of the uterus on the left side (Figure 1). Color Doppler examination revealed only scarce vascularisation on the periphery of described mass. Both ovaries appeared normal on ultrasound examination, with corpus luteum on the right ovary. Intrapерitoneal pooling of fluid in pouch of Douglas was also seen. Her laboratory results were as follows: white blood cells (WBC) 9.54x10^9/l, neutrophil granulocytes (Ne) 5.25x10^9/l, red blood cells (RBC) 3.44x10^12/l, hemoglobin (Hb) 105 g/l, hematocrit (Ht) 0.322; platelets (PLT) 396x10^9/l. Serum electrolytes, coagulation profile and liver function tests were all within physiological limits. A day before the admission, her serum β-human chorionic gonadotropin (β-HCG) level [Abbott test; Architect-Total-β-HCG] was 98 mIU/ml and the next day quantitative β-HCG level decreased to 60.54 mIU/ml. Ectopic gravidity was suspected and diagnostic laparoscopy was performed after the written informed consent had been obtained.

Surgery was conducted under general anesthesia, which was induced by means of propofol as an induction agent, fentanyl as an analgesic and rocuronium as a muscle relaxant. Anesthesia was maintained with 1 – 1.5% end-tidal sevoflurane in 50%:50% O_2/N_2O mixture at 6 l/min flow. The lungs were ventilated to maintain end-tidal carbon dioxide concentration 30 – 35 mm Hg. Laparoscopy revealed that there were about 200 ml liquid and coagulated blood in the abdomen. The omentum was in slight adhesions with the anterior abdominal wall and adhesiolysis was immediately undertaken. The uterus was slightly enlarged. On the left side in the vesicouterine pouch and partly on the vesical peritoneum, there was a round hemorrhagic mass, about 3 cm in diameter, with grayish ruptured capsule, filled with almost black coagulated blood (Figure 2). Behind that formation, there were dark

Figure 1. Ultrasound image of an early primary abdominal pregnancy in vesicouterine pouch in 6th gestational week: hemorrhagic mass in front of the uterus. The uterine cavity is empty.

Slika 1. Ultrazvučni prikaz rane primarne abdominalne trudnoće u vezikouterusnom špagu u šestoj nedelji gestacije: hemoragična masa ispred uterusa; kavum uterusa je prazan.

Abbreviations

β-HCG – β-human chorionic gonadotropin
IUD – intrauterine device
An abdominal pregnancy is a very rare, but potentially life threatening condition and very difficult to diagnose in early stages, moreover it is commonly misdiagnosed. It is also difficult to differentiate between primary and secondary peritoneal implantation. Therefore, the original Studdiford's criteria were modified by Friedrich and Rankin in 1968: 1. the presence of pregnancy less than 12 weeks of histologic gestational age whose trophoblastic attachments are related solely to a peritoneal surface; 2. grossly normal tubes and ovaries; and 3. the absence of uteroperitoneal fistula [5, 6]. The case reported hereby fulfills the original as well the modified criteria.

The most frequent locations for early abdominal pregnancies are pouches surrounding uterus (24.3%), primarily located in the pouch of Douglas (87%) [1]. Early abdominal pregnancies are located on the serosal surface of the uterus and tubes in 23.9%, and multiple organs are included in 12.8%. The serosal surface of the bladder (as in the case reported here) is a rare location: in the cited review of 225 reported cases of early abdominal pregnancies from 1965 to 2009, there were only 3 cases located on the serosal surface of the bladder [1]. The other reported locations are: omental [7], bowel [8], hepatic [9], splenic [10,11], retroperitoneal [12-18], (even in close association of major vessels) [19], at intra-abdominal surface of the diaphragm [20], even after hysterectomy [21].

An ectopic abdominal pregnancy could be a result of the primary peritoneal implantation, or the secondary one, following the primary tubal or ovarian pregnancy that subsequently implants somewhere in the abdomen [1, 22]. There are several attempts trying to explain the occurrence of primary abdominal pregnancies. If there was a delayed ovulation, retrograde menstrual flow could reverse the fertilized ovum through the Fallopian tubes into the abdomen [23]. There is also a possibility that fertilization may occur in the pouch of Douglas and intraperitoneal fluid flow may carry the zygote to some other intraperitoneal location [22]. A retroperitoneal ectopic pregnancy is explained by spontaneous migration of the embryo from the uterus to the retroperitoneal space, possibly through the fistula after bilateral salpingectomy [12] or along the lymphatic channels.

The theory of spontaneous migration of the embryo from the uterus to the retroperitoneal space along lymphatic channels was based on findings of trophoblast surrounded by the lymphatic tissue [16]. The contrast-enhanced computed tomography was used to demonstrate the route of embryo migration in a retroperitoneal ectopic pregnancy providing further evidence in support of the proposed embryo migration mechanism via the lymphatic vessels [17].

There is still a possibility of iatrogenic direct placement during the embryo transfer in cases of retroperitoneal pregnancies after in vitro fertilization (IVF) [14]. An abdominal pregnancy after total hysterectomy could be explained by a fistulous tract from the vaginal apex to the peritoneal cavity or by the prolapse of the Fallopian tube into the vagina, which enable passage for spermatozooa [21]. Intraterine device (IUD) in situ was found in 4.2% of all ectopic pregnancies [24] and in 8% of abdominal pregnancies, so it is speculated that IUD could be a factor contributing to the development of abdominal pregnancy [1].

Maternal morbidity and mortality associated with abdominal pregnancies could be reduced by early diagnosis. Transvaginal ultrasound examination is the main tool in diagnostics of early abdominal pregnancy. The proposed criteria are: 1) the absence of an intrauterine gestational sac; 2) the absence of tubal...
dilatation or a complex adnexal mass; 3) a gestational sac surrounded by loops of bowel and separated from the uterus; and 4) a wide mobility of the gestational sac [25]. In fact, sonographic appearance of early abdominal pregnancy depends of its location. It is usually fixed deep within the pelvis [26] and not mobile as pregnancy in the non-communicating horn of a unicorneate uterus (cornual pregnancy) [27]. If the early abdominal ectopic pregnancy is located in the vesicouterine pouch, it was in the case reported here, transvaginal ultrasound examination detects a gestational sac or complex cystic mass in front of the uterus, but separated from the uterus with no communication between the gestational sac and endometrial cavity. In such a location, the loops of bowel around it are not necessarily found (Figure 1). The absence of communication between the gestational sac and the endometrial cavity differentiates the abdominal pregnancy from the pregnancy in non-communicating horn of a unicorneate uterus (cornual pregnancy) and interstitial ectopic pregnancy [27]. The absence of myometrial layer around the abdominal pregnancy differentiates it from the interstitial pregnancy, which could be demonstrated on ultrasound examination (Figure 1). If the early abdominal pregnancy is located outside the pelvis, transvaginal ultrasound examination is helpless, and other diagnostic tools, such as magnetic resonance imaging and other imaging techniques, must be applied.

Management of early abdominal pregnancy could be surgical, medical, combination of surgical and medical management, and surgery with additional postoperative selective transcatheter arterial embolization for hemostatic failure [1, 28]. Surgical management of early abdominal pregnancies involves laparotomy for the patients with significant intra-abdominal bleeding and laparoscopy for hemodynamically stable patients. It is difficult to diagnose an early abdominal pregnancy and the majority of the cases are presented with intra- abdominal bleeding, so surgical management is more frequent (87.8%) than medical [1]. The patients with the early abdominal pregnancy diagnosed before bleeding are candidates for primary medical treatment with systemic methotrexate [29], laparoscopic embryo methotrexate injection [11] or intracardiac potassium chloride injection (KCl) (in cases with fetal cardiac activity). There is also the combination of these treatments: ultrasound guided feticide and systemic methotrexate, followed by laparotomy for removal of the fetus and placenta [30, 31] or without laparotomy [32, 33]. In later reports there is a tendency to treat an ectopic pregnancy medically after the initial laparoscopic diagnosis. Surgical treatment is used for cases that do not respond to medical treatment [1]. An early abdominal pregnancy, if diagnosed on time - before the occurrence of significant intra-abdominal hemorrhage, could be safely removed by laparoscopy [26, 34, 35]. There are also reports of successful laparoscopic treatment of abdominal pregnancies in second trimester [34]. Immediate surgery is indicated for pregnancies prior to 23 to 24 weeks because of the high incidence of maternal morbidity and a poor prognosis for the fetus. If the patient is stable, more conservative approach could be applied after 24 weeks with better chances for fetus to survive [23]. There are many reports of abdominal pregnancies with viable fetuses and normal-course pregnancies advanced to term [36], but the risk for mother is still very high.

The suspicion is crucial for the timely diagnosis of an abdominal ectopic pregnancy. Rising HCG levels without identification of uterine or ectopic (tubal) pregnancy should raise suspicion of an abdominal ectopic pregnancy. In the case reported here, the diagnosis of ectopic pregnancy was made when the patient was still hemodynamically stable, so we opted for laparoscopic treatment during which the definitive diagnosis of abdominal pregnancy was confirmed. According to the macroscopically normal Fallopian tubes, the absence of extensive pelvic adhesions and the presence of the corpus luteum on the right ovary, it seemed likely that after the fertilization, which could have occurred in the pouch of Douglas, the zygote was carried by intraperitoneal fluid and implanted into the vesicouterine pouch. The possibility of postoperative uterine fistula was excluded in this case: there were no uterine or tubal surgeries in the patient’s medical history. The full content of early abdominal pregnancy was removed during laparoscopy without significant bleeding. Decreasing serum β-HCG levels (which were slightly elevated at the beginning) can explain the absence of significant bleeding: in this case, the pregnancy was the subject of spontaneous involution. This statement was confirmed by histopathological report: the presence of the necrosis of chorionic villi and necrosis and bleeding in the samples from the peritoneal surface of the bladder. Nevertheless, the presence of the intraperitoneal blood allowed neither expectant management nor medical treatment in this case, although the patient was hemodynamically stable. We speculate that spontaneous resolution is probably a destiny for the most of early primary abdominal pregnancies. It is crucial to establish the adequate vascularisation for the pregnancy to survive. If it fails, the majority of early primary abdominal pregnancies may undergo spontaneous involution, staying asymptomatic and undiscovered. The wide use of transvaginal ultrasound, determination of serum β-HCG levels and laparoscopy enable diagnosis of early abdominal pregnancy before the intra-abdominal bleeding occurs.

The possibility of gestational trophoblast disease in ectopic pregnancy is small [37], with prevalence of 0.16:1000 deliveries [38]. In the case reported here, there were no clinical suspicions of gestational trophoblast disease, which was also excluded by histopathological findings.

**Conclusion**

A primary abdominal pregnancy is a very rare and potentially life-threatening condition. A high index
of suspicion, combined with carefully interpreted clinical and ultrasound findings, are crucial for timely diagnosis, before the occurrence of severe intra-abdominal bleeding. We have presented the case of early primary abdominal pregnancy, diagnosed in the sixth gestational week, located in the vesicouterine pouch and treated laparoscopically. Despite the rapidly decreasing serum β-human chorionic gonadotropin levels, the presence of the intraperitoneal blood allowed neither expectant management nor medical treatment in this case, although the patient was hemodynamically stable at that moment. The absence of significant bleeding during the surgery and histopathological finding of placental villi with necrosis confirmed that this abdominal pregnancy was already the subject of spontaneous involution.

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