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CASE REPORT

Ovarian ectopic pregnancy in adolescence

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Key Clinical Message

Ovarian pregnancy is one of the rarest types of extrauterine pregnancy. Its preoperative diagnosis remains a challenge since it presents quite similarly to tubal pregnancy and complicated ovarian cysts. Although in most cases, histology is necessary to confirm the diagnosis, we present an ovarian pregnancy in a teenager, correctly diagnosed during ultrasound examination.

Keywords

Laparoscopy, ovarian pregnancy.

Introduction

An ectopic pregnancy occurs when the implantation and development of the embryo occurs outside the uterine cavity [1]. Primary ovarian pregnancy is defined as a pregnancy implanted within the ovary and, although its incidence is increasing in the last couple of decades [2, 3], it remains a rare type of extrauterine pregnancy (0.5–3.0% of all ectopic pregnancies with an incidence ranging from 1 in 7000 to 1 in 2100 pregnancies) [3].

Since its clinical presentation (symptoms and ultrasound characteristics) is very similar to that of tubal pregnancy or a complicated ovarian cyst, its preoperative diagnosis remains a challenge and most of the cases are diagnosed during surgery.

In this article, we describe a case of an asymptomatic ovarian pregnancy in a teenager, which was correctly diagnosed by ultrasound scan and positively confirmed during laparoscopy and histology.

Case Report: A 16-year-old, nulliparous woman with an uneventful past gynecological history and regular menses presented to the unintended pregnancy appointment after a positive urine-pregnancy test. She had an amenorrhea of 6 weeks. She used a barrier contraceptive method irregularly.

An endovaginal ultrasound examination was performed to correctly date the pregnancy and found a trilaminar endometrium without any gestational image; in the left adnexial area a heterogeneous vascularized mass was identified, with 25 mm, contiguous to the left ovary, suggesting an ovarian pregnancy (Fig. 1). The patient was asymptomatic, hemodynamically stable with a normal physical and gynecological examination (no pain during examination). On the same day, hemoglobin (13.1 g/dL), and serologic βhCG were determined (4555 IU/dL).

According to the protocol used in the institution (*Fernandez* Score) there were conditions to start medical therapy with methotrexate (MTX) (score 11) however, since outpatient follow-up was not possible due to uneasy access to the emergency room, surgical treatment was chosen.

Laparoscopy was performed and confirmed the diagnosis previously suspected. During the procedure a heterogeneous/necrotic mass with 3 cm of diameter was identified

Figure 1. Ultrasound showing an heterogenous mass 25 mm adjacent to the ovary (OEP, ovarian ectopic pregnancy; Ov, ovary).

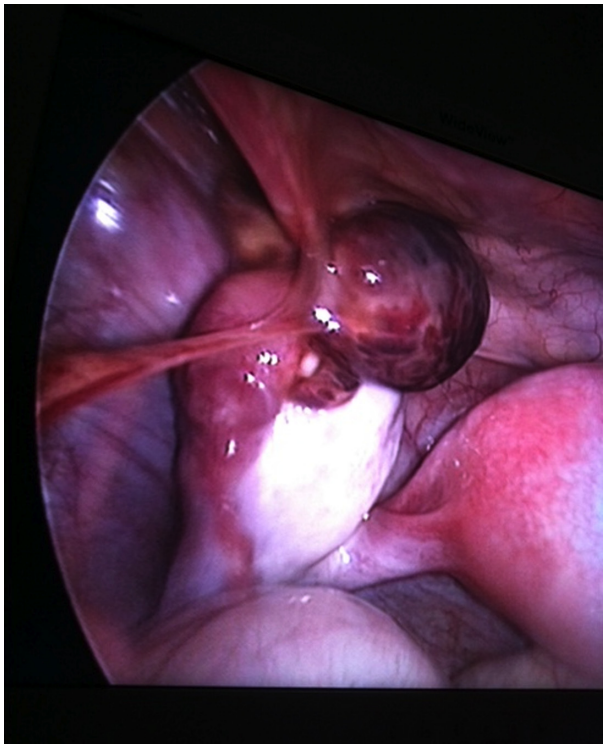

Figure 2. Initial laparoscopic view of left ovarian pregnancy. The tube is intact and the mass is adjacent to the ovary.

in the left iliac fossa, adherent to the anterior abdominal wall. There were neither signs of rupture or blood within the pelvic cavity. After adhesiolysis, it became clear that the whole structure was located contiguous to the ovarian tissue leaving the left tube intact (Figs. 2 and 3). The ectopic pregnancy was removed using bipolar energy preserving as much ovarian tissue as possible.

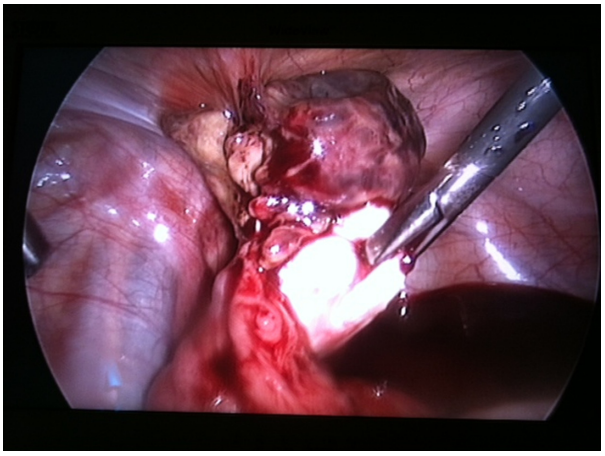

Figure 3. Laparoscopic view after dissection. It is clear that the tube remains intact, the gestation still ocupies the space of the normal ovary to which it is connected.

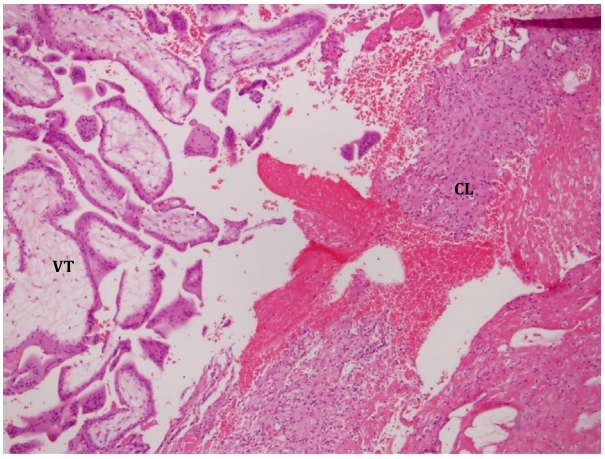

Figure 4. Histology: villous tissue (VT, Left side of the image) adjacent to corpus luteum (CL, Right side of the image).

The patient was discharged in the second day after surgery and βhCG was determined until it was negative (day 14).

The histological result confirmed an ovarian pregnancy, namely villous tissue adjacent to corpus luteum (Fig. 4).

Discussion

Ovarian pregnancy is a rare event first reported in 1682 by Saint Maurice [4]. Moreover, its true incidence may be underestimated owing to the fact that some ovarian pregnancies are diagnosed as tubal pregnancies and treated medically without surgical inspection and confirmation of its true location [5]. However, the incidence of ovarian pregnancy is rising in the last decades, which can

be explained by a higher diagnostic accuracy [6, 7] and a change in the prevalence of risk factors.

The only risk factor accurately related to ovarian pregnancy is the use of an intrauterine device [8–10]. Assisted reproductive techniques (ART) may also be a risk factor for ovarian pregnancy which may happen in 0.3% of ART pregnancies [3, 11].

Although the exact mechanism which explains ovarian pregnancy remains unknown, possible hypothesis include interference with the ovum release from the ruptured follicle, fallopian tubes malfunction or inflammatory thickening of the *tunica albuginea* [12]. Ovarian pregnancies can be classified either as primary, if the ovum is fertilized while it is still in the folicule, or as secondary, if fertilization takes place in the tube with posterior regurgitation of the conceptus back to the ovarian stroma [13].

In a review of 49 ovarian pregnancies, abdominal pain and vaginal bleeding were the most frequent symptoms. A history of amenorrhea, elevated βhCG level, and an empty uterus on ultrasound scan increase the suspicion. These features are shared with other types of ectopic pregnancies, namely tubal pregnancies. Since the ovary is a highly vascular organ, these patients are at an increased risk of having severe hemorrhage if ovarian surface rupture occurs, and may present in hypovolemic shock.

Ultrasound examination is an important diagnostic tool for ovarian pregnancy. Comstock identified a wide echogenic ring with a small internal echolucent area as the most frequent ultrasound feature [7]. Free blood or clots in the pelvis are also quite usual. Since most of these pregnancies have a low gestational age, identification of a yolk sac or an embryo is quite rare [14], however, when present, they increase the diagnostic accuracy of ultrasound. The type of transducer used (10 MHz vs. 7 MHz) may be a cornerstone to the ultrasound diagnosis [15].

Preoperative diagnosis may be extremely difficult and tube pregnancy, functional ovarian cyst, or tubo-abdominal abortion of a tube pregnancy is important differential diagnosis [14, 16]. The preoperative diagnosis of ovarian pregnancy can only be achieved in 5.3–25% of cases [17, 18].

The definite diagnosis of ovarian pregnancy can only be made during surgery. The Spiegelberg criteria, although not always easy to meet, are widely accepted and include: (1) An intact ipsilateral tube, clearly separated from the ovary; (2) A gestation occupying the normal position of the ovary; (3) A gestational sac connected to the uterus by the útero-ovarian ligament; (4) Ovarian tissue in the wall of the gestational sac.

Laparoscopic surgery is the gold standard approach in ovarian pregnancy treatment and the Practice Committee of the American Society for Reproductive Medicine recommends that ovarian pregnancy should be definitively diagnosed by surgical exploration, so that medical therapy is not a first-line option for this condition [19].

The surgical technique used depends on the individual features of the pregnancy, but special attention should be taken toward preserving as much ovarian tissue as possible, especially in reproductive aged women [20]. Hence we have moved from an era where laparotomy and ovariectomy were the most frequent procedures to an era where cystectomy or wedge-resection are the cornerstone, particularly in hemodynamically stable patients. It has also the advantage of a reduced postoperative morbidity, shorter hospitalization, and recovery time.

As stated before, a conservative approach is of outmost importance particularly in young patients who desire to bear children. In tubal pregnancy, the use of MTX is now a well established and, in selected cases, a safe mode of conservative therapy [21]. The use of MTX in ovarian pregnancy is still sparse but some case reports have been published. Kudo et al. [22] were the first to successfully treat an ovarian pregnancy with MTX, followed by Shamma et al. who used a single- intramuscular dose of MTX (50 mg/m^2) [23]. Mittal was the first to report an MTX injection directly in the gestational sac [24].

After laparoscopy, there is a low risk of recurrence of ectopic pregnancy, and only one case of repeated ovarian pregnancy has been reported to date [25]. On what future pregnancy outcomes are concerned Koo et al. [17] found that 46.4% of women had a successful intrauterine pregnancy, 10.7% had tubal pregnancies, and only 3.6% (1 woman) were diagnosed secondary infertility.

To our knowledge, this is the youngest patient ever reported to have an ovarian pregnancy. Smoking, which is often related to tubal pregnancy [26], is the only possible risk factor identified. In this case, the ultrasound features raised the suspicion that this might be an ovarian pregnancy, which was positively confirmed by surgery (intact tube, pregnancy occupying the normal position of the ovary) and lastly by histological evaluation (ovarian tissue next to the gestational sac) – Spielberg criteria were met (see Figs. 2–4). It is important to note that because of the young age of the patient, a conservative approach was attempted and was well succeeded.

In conclusion, an accurate ultrasound diagnosis and high levels of suspicion are essential for preoperative diagnosis of ovarian pregnancy. An expeditious approach, where laparoscopic surgical treatment remains the gold standard, is important to lessen morbimortality and confirm the diagnosis of this rare condition.

Conflict of Interest

None declared.

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