

Case report



Chronic tubal ectopic pregnancy with a negative serum betahuman chorionic gonadotropin test: A case report

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Abstract

Early measurements of serum beta-human chorionic gonadotropin (β -hCG) levels and transvaginal ultrasound constitute the mainstay of timely ectopic pregnancy diagnosis. The current case highlighted that an ectopic pregnancy may occur even with an undetectable initial serum β -hCG level and may go ignored if taken quietly. Difficulties in diagnosing chronic ectopic pregnancy make this clinical case a challenge in gynecology and are not commonly described in the literature. This case presents a 31-year-old female patient, Gravid 5 Para 3, who presented to the Emergency Department with a two-week history of spotting along with recent worsening associated with pelvic pain. The gynecological examination displayed a blackish bleeding of endocavitary origin and adnexal tenderness especially on the right. The serum β -hCG level was negative and it was checked twice in two different biomedical laboratories. An urgent transvaginal pelvic ultrasound showed a right heterogeneous tubal mass with a double component of 65x57 mm. Laparoscopy revealed a heterogeneous ampulla mass of 4 cm related to an encysted hematocele in the right Fallopian tube. The treatment consisted of a right salpingectomy. The final pathological analysis confirmed right tubal pregnancy. In the absence of typical clinical symptoms or characteristic ultrasound signs for this entity, a diagnostic laparoscopy was urgently indicated to prevent the complications of a ruptured pregnancy and to avoid a poor prognosis for the patient.

Keywords: Ectopic pregnancy, Negative serum βHCG level, Laparoscopy, Case Report.

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1. Introduction

The ectopic pregnancy prevalence is 1-2% worldwide [1]. The fear of a ruptured ectopic pregnancy can cause great maternal morbidity and mortality [1]. In South Africa, an ectopic pregnancy occurs in 11 of 1000 reported pregnancies, with an estimated mortality rate of 2% [2]. Chronic ectopic pregnancy represents a variant of ectopic pregnancy characterized by low or imperceptible serum beta-human chorionic gonadotropin (β -hCG) levels, resistance to methotrexate (MTX), and an adnexal mass with fibrosis, necrosis, and blood clots. Delay in the diagnosis of chronic ectopic pregnancy makes this clinical case a challenge in gynecology and not commonly reported in the literature.

2. Case report

We describe a case of a 31-year-old female patient, blood group A positive, Gravid 5 Para 3, with a past medical history of two instrumental abortions and three living children. She was a non-smoker. Her menstrual cycle was regular and lasted 28 days without delay. The patient was not using any form of contraception. She had also no history of sexually transmitted infections. Her last menstrual period was seven days prior to her admission.

*Correspondence: Dr Skander Abid, Department of Obstetric Gynecology, Farhat Hached University Hospital of Sousse, Sousse, Tunisia. Email: <u>Abid.skander1992@outlook.com</u> The patient described the notion of spotting for two weeks with recent aggravation associated with the appearance of pelvic pain without any other associated signs. The patient was apyretic with an accurate blood pressure. A tenderness was identified in the right iliac fossa on the abdominal examination. There was a dark uterine bleeding on per speculum test. On the bimanual vaginal examination, there was an adnexal tenderness, especially on the right. The hemoglobin concentration was 10.9g/dl. White blood cells were 10,000. The C-reactive protein (CRP) was 3mg/L. In addition, there were negative serum β -hCG level results which were assessed two times in distinct biomedical laboratories.

Transvaginal ultrasound (TVUS) showed an empty uterus with a hematometra (Fig.1), effusion of great abundance right latero-uterine and at the level of Douglas (Fig.2). The two ovaries were identified with the normal size. Additionally, we discovered a heterogeneous right mass with a double component of 65x57mm independent of the ovary and the uterus drawing the path of the Fallopian tube (Fig.3).

The diagnosis of an ectopic pregnancy in its encysted form or a tumor of the Fallopian tube were evoked. Hence, a laparoscopy was indicated as a diagnostic and operative approach. The diagnostic laparoscopy demonstrated a hemorrhagic effusion of great abundance with clots. The right Fallopian tube was dilated at its distal end of an ampullar mass of 40mm related to an encysted hematocele. The treatment was a total right salpingectomy (Fig.4). The postoperative followup was uneventful. The patient left the hospital on the second postoperative day.



Fig.1. An empty uterus with a hematometra (yellow arrow) soaked in blood.



Fig. 2. Effusion of great abundance right latero-uterine and at the level of Douglas.



Fig. 3. A heterogeneous right mass with a double component of 65 mm x 57 mm independent of the ovary and the uterus drawing the path of the fallopian tube.

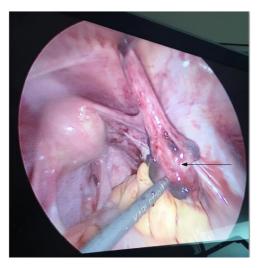


Fig.4. Right fallopian tube presented an adnexal ampullar mass of 40mm (black arrow) related to an encysted hematocele.

The histopathology examination confirmed a right tubal pregnancy (Fig.5). As highlighted by the current case, an undetectable β -hCG serum level does not absolutely reject the diagnosis of an ectopic pregnancy. The patient did not provide her perspective for this case study.

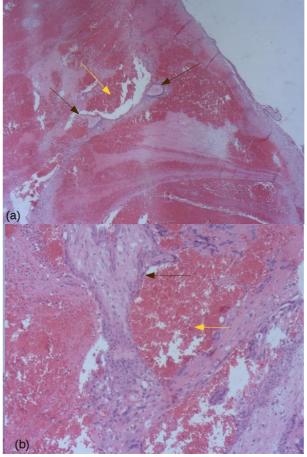


Fig. 5. Dilated tubal lumen with luminal hemorrhage (yellow arrow) and including chorionic villus (black arrow).

3. Discussion

An ectopic pregnancy is a pregnancy arising separately to the uterine cavity. In most cases, it is confined in the Fallopian tube and mostly in the Ampulla area [3]. Early measurement of serum β -hCG level and TVUS constitute the basis of an appropriate ectopic pregnancy diagnosis. With extremely sensitive β -hCG assays available, all pregnancies, including ectopic pregnancies, are presumed to exhibit measurable serum β -hCG levels [4]. The diagnostic performance of B-mode ultrasound is 74.5% in a Tunisian study. The coupling of the suprapubic route and the endovaginal route as well as the experience of the sonographer are the main factors to improve the sensitivity of the ultrasound and to increase the chances of an early diagnosis of ectopic pregnancy [5].

As reported in our case, an ectopic pregnancy may occur even with an imperceptible initial serum β -hCG level and may go ignored if taken quietly. An ectopic pregnancy associated with an absent serum β -hCG level is a rare situation estimated only in 1% of ectopic pregnancies [6]. There have been two hypotheses proposed to explain this phenomenon. The first is the cessation of its production by the degenerated trophoblastic tissue. The second is the presence of low-mass chorionic villi that secretes this hormone and increases its removal from blood circulation [6]. Chronic ectopic pregnancy is considered an independent clinical entity in which a blood collecting is ordered. This bleeding is spontaneously interrupted by blood sets or blocked by the omentum, the sigmoid colon, and the intestines creating a real diaphragm. Certainly, when the bleeding is rare (tubo-abdominal abortion, a cracked hematosalpinx), the blood is restricted to the lowest point of the peritoneal cavity, namely the Douglas inducing a retrouterine encysted hematocele as well as a latero-uterine or peri-tubal hematocele [7]. More recently, Tempfer et al. [8] showed in their systematic review of the literature that the serum β -hCG levels were perceived for 124 patients and were undetectable in 32% of samples.

The most frequent exhibiting symptom of patients with chronic ectopic pregnancy was abdominal pain (71%), followed by irregular vaginal bleeding (55%) and fever (5%). However, only 18% of cases showed an asymptomatic presentation. Moreover, 48% of cases displayed an adnexal mass at the time of the initial diagnosis [8]. In the absence of typical clinical symptoms or characteristic ultrasound signs for this entity, it is always necessary to think about it and not to hesitate to indicate a diagnostic laparoscopy, as it was performed in our case, in order not to end up with a ruptured pregnancy putting at risk the vital prognosis of the patient.

Recent recommendations for medical treatment with MTX in ectopic pregnancy include single-dose or multipledose regimens for hemodynamically stable patients with no medical contraindications to MTX. Monitoring includes serial β -hCG serum levels. Treatment failure was defined as a failure to reduce serum β -hCG level by at least 15% between days 4 and 7 after MTX. Surveillance includes weekly serum β -hCG level monitoring until negativity [9]. Otherwise, it is evident that in this case with the initial serum β -hCG level being negative, the follow-up would have been based only on ultrasound control and monitoring of clinical signs.

The most common treatment of chronic ectopic pregnancy previously reported was surgery with laparoscopic salpingectomy practiced as the first-choice approach. This treatment policy is highly efficient. According to Clemens et al. [8], 97% of patients with chronic ectopic pregnancy were effectively managed with this strategy and the remaining patients required a second-line therapy. Interestingly, based on our case report, we support the recommendation indicating the laparoscopic salpingectomy for patients diagnosed with chronic ectopic pregnancy.

In conclusion, our findings support that, even though scarce, an absent serum β -hCG does not entirely exclude the diagnosis of an ectopic pregnancy. Specialists should be careful not to miss a latero-uterin image to avoid a hypovolemic shock and to improve the vital prognosis of the patient.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Conflict of Interest Disclosures

All authors declare that they have no conflict of interest.

Authors' contributions

All authors have read and agreed to the published version of the manuscript.

References

- [1] Nyakura M, Mhlanga FG, Madziyire M, Matshalaga S. Spontaneous bilateral tubal ectopic pregnancy : a case report. Pan Afr Med J. 2021;38:395. <u>https://doi.org/ 10.11604/pamj.2021.38.395.28771</u>.
- [2] Nzaumvila DK, Govender I, Ogunbanjo GA. An audit of the management of ectopic pregnancies in a district hospital, Gauteng, South Africa. Afr J Prim Health Care Fam Med. 2018;10(1):e1-e8. <u>https://doi.org/10.4102/</u> phcfm.v10i1.1757.
- [3] Lau S, Tulandi T. Conservative medical and surgical management of interstitial ectopic pregnancy. Fertil Steril. 1999;72(2):207-15. <u>https://doi.org/10.1016/s0015</u> -0282(99)00242-3.
- [4] Irani M, Elias RT, Pereira N, Gunnala V, Rosenwaks Z. Abdominal ectopic pregnancy with undetectable serum β-human chorionic gonadotropin 9 days following blastocyst transfer. J Obstet Gynaecol Res. 2016; 42(12):1886-8. <u>https://doi.org/10.1111/jog.13127</u>.
- [5] Mathlouthi N, Slimani O, Fatnassi A, Ben Temime R, Makhlouf T, Attia L, et al. Diagnostic échographique de la grossesse extra-utérine : étude prospective à propos de 200 cas [Ultrasound diagnosis of ectopic pregnancy: prospective study of 200 cases]. Tunis Med. 2013; 91(4):254-7.
- [6] Daniilidis A, Pantelis A, Makris V, Balaouras D, Vrachnis N. A unique case of ruptured ectopic pregnancy in a patient with negative preg-nancy test - a case report and brief review of the literature. Hippokratia. 2014;18(3):282-4.
- Clark JF, Bryant W. Chronic ectopic pregnancy. J Natl Med Assoc. 1975 Mar;67(2):118-21.
- [7] Tempfer CB, Dogan A, Tischoff I, Hilal Z, Rezniczek GA. Chronic ectopic pregnancy: case report and systematic review of the literature. Arch Gynecol Obstet. 2019;300(3):651-60.<u>https://doi.org/10.1007/s00404-19-05240-7</u>.
- [8] Hughes M, Lupo A, Browning A. Ruptured ectopic pregnancy with a negative urine pregnancy test. Proc (Bayl Univ Med Cent). 2017;30(1):97-8. <u>https://doi.org/ 10.1080/08998280.2017.11929547</u>.

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