

Spontaneous triplet, tubal ectopic gestation: a case report

Gravidez ectópica tubária tripla espontânea: caso clínico

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Abstract

Ectopic pregnancy occurs when the implantation of the developing blastocyst occurs at a site other than the endometrium in the uterine cavity. Few cases of spontaneous unilateral triplet ectopic gestation have been reported.

Keywords: Triplet ectopic gestation; Ectopic gestation.

INTRODUCTION

Ectopic pregnancy (EP) occurs when the developing blastocyst becomes implanted outside the endometrium in the uterine cavity¹. EP occurs in 1% of pregnancies and may seriously compromise women's health and future fertility. The prevalence of EP among women admitted at the emergency department with first trimester bleeding, pain, or both ranges from 6 to 16 percent²⁻⁵.

The maternal mortality ratio associated with EP declined 57 percent in the periods of 1980 to 1984 and 2003 to 2007, from 1.15 to 0.50 deaths per 100,000 live births⁶. Between 2003 to 2007⁶ it was 6.8 times higher in african american women than in white and 3.5 times higher in women older than 35 years as compared with those younger than 25 years.

Approximately 98% of ectopic pregnancies occur in the Fallopian tube (FT)^{2,3}. Consequently the most frightened complication of EP is tubal rupture, causing massive abdominal bleeding, infection, and possibly death^{1,2}. Risk factors for EP can be divided as high, moderate, and low (Table I).

Diagnosis and management of these pregnancies has changed dramatically over the years⁴. Early diagnosis of ectopic pregnancy is crucial to reduce the risk of rupture and improve treatment results⁵⁻¹⁰. The diagnosis is usually highly suspected based on the clinical history,

imaging (ultrasound) and blood tests (human chorionic gonadotropin, hCG). Confirmation from direct observation of the ectopic gestation, or histology, can be obtained but nowadays surgical management is not always required. In the absence of a definitive diagnosis from laparotomy, histology, or imagiology it can be difficult to distinguish between an EP and early failure of an intrauterine gestation¹⁰⁻¹³. A hemodynamically stable patient at risk to carry an EP should be attended

TABLE I. RISK FACTORS FOR ECTOPIC PREGNANCY¹¹⁻¹³

Degree of risk	Risk factors	Odds ratio
High	Previous ectopic pregnancy	9.3–47
	Previous tubal surgery	6.0–11.5
	Tubal ligation	3.0–139
	Tubal pathology	3.5–25
	In utero DES exposure	2.4–13
	Current IUD use	1.1–45
Moderate	Infertility	1.1–28
	Previous cervicitis (gonorrhea, chlamydia)	2.8–3.7
	History of pelvic inflammatory disease	2.1–3.0
	Multiple sexual partners	1.4–4.8
	Smoking	2.3–3.9
Low	Previous pelvic/abdominal surgery	0.93–3.8
	Vaginal douching	1.1–3.1
	Early age of intercourse (<18 years)	1.1–2.5

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before rupture. This can be accomplished without using invasive techniques as laparoscopy.

Several factors may explain the pathogenesis of tubal pregnancies. It is generally believed that they result from conditions that delay, or prevent, the passage of the fertilized oocyte to the uterine cavity. Also, factors inherent to the embryo may result in its premature implantation¹⁻³. Multiple ectopic gestations might result from simultaneous bilateral ovulation, or superfetation with or without trans-peritoneal migration⁷.

Only seven cases of spontaneous, unilateral, triplet ectopic gestations have been documented⁷⁻⁹. The authors report the eighth case of a spontaneous, unilateral, triplet, tubal ectopic gestation.

CASE PRESENTATION

A 27-year-old black female, gravida 5 para 4, was admitted in the emergency department at 7 weeks plus 2 days of amenorrhoea with complains of hypogastric pain, mild vaginal blood loss and fatigue for a day. Her medical history was uneventful, without no prior surgeries. Contraception was achieved using barrier methods.

At admittance, the physical examination revealed hypotension (BP 84/52mmHg) and sweating. Palpation of the abdomen was painful, with peritoneal signs and slight blood loos from the cervical os was observed on vaginal examination. The patient did not tolerate bimanual exam due to cervical tenderness.

The pelvic ultrasound scan demonstrated a gestational sac in the dependence of the left fallopian tube containing 3 embryos with cardiac activity (crown-



FIGURE 1. Gestational sac seen in the left fallopian tube containing 3 embryos (1,2,3)



FIGURE 2. Longitudinal endovaginal US image: hematoma (7.9cm) in the retrouterine space

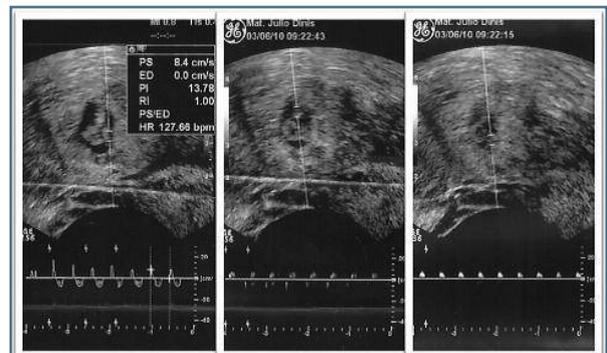


FIGURE 3. Fallopian tube containing 3 embryos with cardiac activity

-rump length 5.5mm, 5.8mm and 5.8mm) and a 7.9cm retro-uterine hematoma (Figure 1-3). Blood analysis revealed a β -hCG of 23.146 IU/L and Hb 9.1g/dL. Spontaneous ectopic triple tubal pregnancy was assumed as the patient denied any medically assisted reproduction techniques. The patient was submitted to a laparotomy. A massive hemoperitoneum was found, with approximately 1000mL of liquid blood and clots in the abdomen). A gestational sac, and active bleeding in the implantation site, was observed in the left tube (Figure 4,5). Left salpingectomy and right fimbriectomy (patient option) was performed.

EP diagnosis was confirmed by histopathological analysis. Patient was discharged at the 2nd post-surgery day.

DISCUSSION

In the developed world, 1% to 2% of all reported

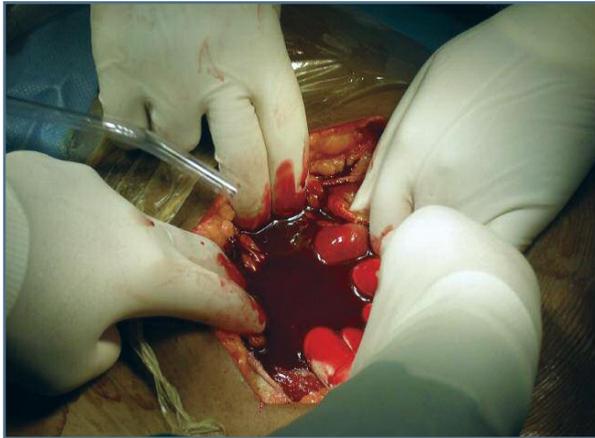


FIGURE 4. Presence of blood in the peritoneal cavity

pregnancies are ectopic, similar to the incidence of spontaneous twin pregnancy. The incidence in developing countries might be higher, but precise numbers are not known¹⁴⁻¹⁶.

In Portugal the accurate incidence of ectopic pregnancy is unknown. This is the first case of a spontaneous triplet ectopic pregnancy described.

EP is a major cause of maternal morbidity, at short and long-term, as a consequence of its acute symptomatology related to pelvic pain and fertility problems. The diagnosis of an ectopic pregnancy is based on clinical history, physical examination, ultrasound and by serial serum beta-hCG levels. Abdominal pain arises because of irritation of the peritoneum by an increasingly dilated tube or presence of a haemoperitoneum after retrograde blood flow from the tube (tubal abortion) or tubal rupture. Surely, pelvic ultrasound is the most accurate exam to detect an ectopic pregnancy with 89% and 99.8% of sensitivity and specificity, respectively¹²⁻¹⁶. In this report the diagnosis was suggested by clinical findings and confirmed by laboratory parameters and ultrasound scanning, as positive cardiac activity in the three embryonic poles was observed. Ultrasound images are variable. The most frequent aspect is an adnexal mass compatible with a gestational sac surrounded by a hyper-dense crown and a hypervascularity phenomenon during Doppler exam named as 'chapel sign'^{16,17}.

The management of an EP is dependent on the haemodynamic status of the patient, on the location, and gestational age, on the activity of the trophoblasts (β hCG), as well as the presence or not of a concomitant topic pregnancy (heterotopic pregnancy) and obs-

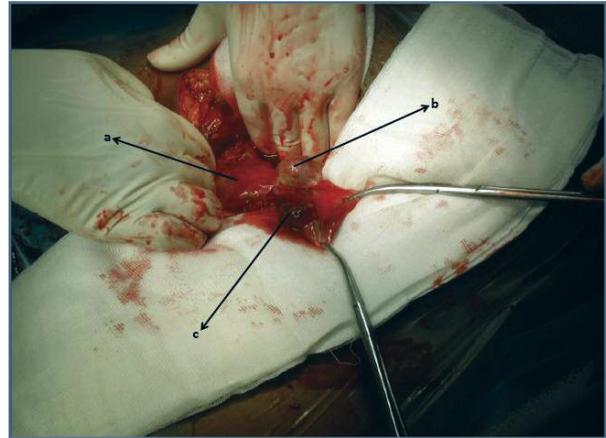


FIGURE 5. Spontaneous triplet, tubal ectopic gestation. (a) uterus; (b) left ovary; (c) tubal ectopic gestation (gestational sac)

tetric history of the patient .

Expectant management is effective in 47-82% of the cases of ectopic pregnancy. A good candidate has a decreasing β -hCG level less than 1000 U/L, an ectopic mass less than 3 cm, no detected embryonic cardiac activity, and patient agreement to comply with follow-up requirements³⁻⁵.

Methotrexate (MTX), a folic acid antagonist, is a well-studied drug to be used for medical management. Methotrexate deactivates dihydrofolate reductase, which reduces tetrahydrofolate levels (a cofactor for deoxyribonucleic acid and ribonucleic acid synthesis), thereby disrupting rapidly-dividing trophoblastic cells. Patient selection is crucial for medical management of an ectopic pregnancy. The lower the β -hCG levels at initiation of treatment, the higher the success rate of MTX therapy⁴. A concomitant viable topic pregnancy must be excluded.

In this report MTX and the expectant attitude were contraindicated: the patient presented decrease of haemoglobin, high levels of β -hCG, significant size of tubal mass and positive embryonic heart rate.

Two surgical options are possible in the presence of a tubal pregnancy: conservative surgery with salpingostomy and aspiration of tubal content or non-conservative surgery with salpingectomy¹⁴. Salpingostomy through laparoscopy has become the preferred method of surgical treatment^{14,15}. In this clinical case laparotomic surgical approach with salpingectomy was elected based on the volume of the hemoperitoneum, the patient hemodynamic instability, the presence of marked dilatation of the tube with active bleeding and on the contraceptive choice of the patient. The patient

opted for definitive sterilization and left salpingectomy and right fimbriectomy was performed.

Recurrence occurs in 5-20% of patients with a prior EP, and in 32% of those who have had two consecutive ectopic pregnancies. Prognosis is favourable in patients who receive appropriate treatment^{14,15}.

CONCLUSIONS

This case report highlights that multiple, ectopic pregnancies can occur spontaneously. Its association with medically assisted reproduction techniques may express complex events, barely known, occurring at early stages of embryonic development.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and the accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

ABBREVIATIONS

EP: Ectopic pregnancy; FT: Fallopian tube; hCG: human chorionic gonadotropin; MTX: Methotrexate; BP Blood pressure

COMPETING INTERESTS

The authors declare that they have no competing interests.

AUTHORS' CONTRIBUTIONS

L.G-M helped to prepare the case report, perform a literature search, and write the case report and discussion. DPL helped to prepare the case report and perform a literature search. JPS helped to write the case report and discussion. All authors read and approved the final manuscript.

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