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Case Report

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Unilateral Twin Tubal Ectopic Pregnancy after Intrauterine Insemination-Case Report

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ABSTRACT

Introduction: Twin ectopic pregnancy is a rare event that affects 1:125,000 pregnancies. There are around 100 reported cases in the literature, 15 cases with the presence of fetal heartbeat diagnosed by ultrasound examination. It is important to highlight that early diagnosis is essential to avoid serious complications, including maternal death.

Case Report: 36 years old anovulatory patient underwent intrauterine insemination, with a positive result, having been subsequently diagnosed with early unilateral ectopic twin pregnancy at 6 weeks. The patient underwent video laparoscopic left salpingectomy and was discharged the following day.

Discussion: Early diagnosis is essential so that treatment can be instituted as quickly as possible, as this is an event with high morbidity and mortality.

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Introduction

Unilateral twin tubal ectopic pregnancy is a very rare form of ectopic pregnancy, in which the implantation and development of the blastocysts occur outside the uterine cavity, both within the same uterine tube [1]. The first report of unilateral twin ectopic pregnancy was in 1891 by De Ott after which there were around 100 cases reported in the literature [2]. Twin ectopic pregnancy is a rare event that affects 1:125,000 pregnancies [3].

As it is a rare event, this report aims to demonstrate that technological advances and the development of more modern ultrasound devices, as well as video endoscopic devices, has allowed for more precocious diagnoses as well as effective and less invasive treatment for what is an extremely serious event which, if not managed well and in a timely manner, can lead to death. Despite this, it was only in 1994 that they were able to diagnose the first unilateral twin ectopic pregnancy on ultrasound with a fetal heartbeat present, before tubal rupture [4].

The risk factors most involved in the development of ectopic pregnancy include

a) pelvic inflammatory diseases (PID)
b) large number of IUD insertions (mainly copper)
c) endometriosis
d) increase in the number of tubal microsurgeries
e) cigarette smoking and
f) assisted reproduction techniques (artificial insemination and in vitro fertilization) [5,6].

The objective of this case report is to make the medical population aware of the care and risk of this disease, showing the most effective diagnostic measures, as well as the best therapeutic options present in the literature with the aim of reducing maternal deaths. The secondary objective is to leave our record in the literature.

Case Report

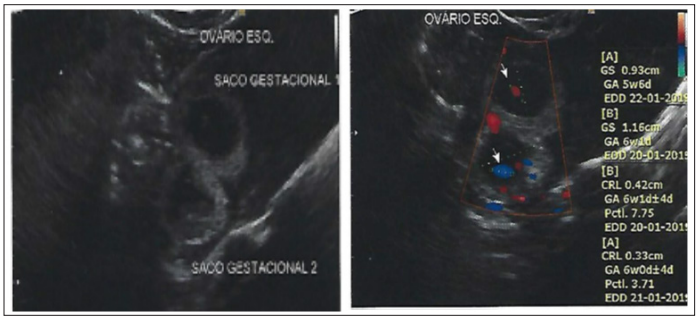
A 36 years old patient, nulligravida, with primary infertility due to an anovulatory condition (polycystic ovary syndrome, hyperinsulinism, overweight), without anatomical changes; that is, with normal uterine tubes and uterus (hysterosalpingography and negative infectious panel), and with the male partner with semen quality within normal limits. Therefore, ovarian stimulation was indicated for low complexity treatment, with two unsuccessful scheduled intercourse cycles, followed by intrauterine insemination

(IUI). For IUI, the patient was submitted to ovarian stimulation using recombinant FSH (50 IU/day) and serial ultrasounds. When the dominant follicle reached 18mm, she received hCG trigger. After 36 hours, semen was collected by masturbation, followed by density gradient centrifugation and transfer to the uterine cavity with the use of a Wallace catheter. A quantitative β-hCG test was carried out on days 12 and 14, with a positive result (2,227 IU/L). Control β-hCG measures were collected subsequently every 48 hours. Because there was no significant increase in β-hCG, and in the absence of a gestational sac image in the uterus, we initiated observation of the adnexa.

An ultrasound was performed at a specialized fetal medicine service and, at 6 weeks, two images were visualized in the left annex that were compatible with twin pregnancy, both with the presence of fetal heartbeats.

Ultrasound examination revealed the presence of 2 gestational sacs in the left tube. Fetus 1 had a gestational sac with an average diameter of 9.6 mm, compatible with 6 weeks gestational age, and a fetal heart rate of 119 bpm, with a CCN of 3.0 mm. Fetus 2 had a gestational sac with an average diameter of 11.4 mm (6 weeks gestational age), a fetal heart rate of 133 bpm and a CCN of 4.2 mm (Figure 1).

Figure 1: Ultrasound Images



(a) **(b)**

Figure 1a: Visualization of the two gestacional sacs showing a dichorionic diamniotic ectopic twin pregnancy. The left ovary can be seen in the right upper quadrant of the image. **Figure 1b**: Doppler ultrasound showing presence of fetal heartbeats in both embryos. The gestacional sac referred to as number 1 in image 1a was measuring 0.93 cm of diameter and the gestacional age according to the ultrasound was of 5 weeks and 6 days, and the one referred to as number 2 was measuring 1.16 cm of diameter and the gestacional age according to the ultrasound was of 6 weeks and 1 day.

The patient, up until then asymptomatic, began to experience severe pain in the left iliac fossa, which was accompanied by vaginal bleeding. The abdomen was distended and painful on palpation, but there were no signs of peritoneal irritation. At this point, she was taken to the hospital where she underwent urgent ultrasound that revealed the absence of blood in the abdominal cavity and confirmed the ectopic pregnancy in the left tube; the patient was hemodynamically stable, which is why she underwent laparoscopy. Laparoscopic surgery was performed under general anesthesia and the left uterine tube was removed without any complications. The patient was discharged the following day, without the need for use of replacement fluid or blood products (Figure 2).

Figure 2: Videolaparoscopy Images

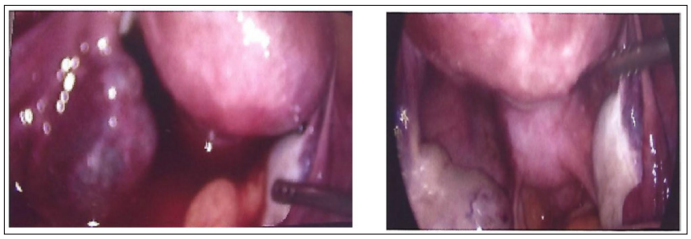


Figure 2a and Figure 2b: Overview of the uterus, ovaries and uterine tubas. The picture also shows the ectopic pregnancy located in the left tuba before the salpingectomy.

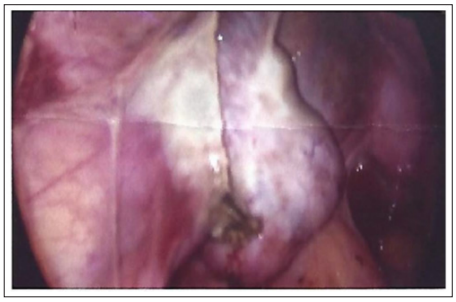


Figure 2c: Results after the exeresis of the left tuba

The material was sent for anatomopathological study and the salpingectomy product revealed
1) Twin ectopic pregnancy, partially ruptured
2) Acute, non-specific chronic salpingitis (Figure 3).

Figure 3: Anatomopathological Images
Left Tuba

Figure 3a: Left uterine tuba after salpingectomy showing signs of salpingitis

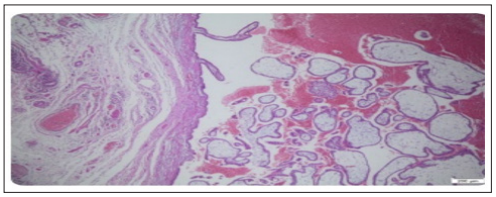


Figure 3b: Chorionic villi compatible with a 1st trimester pregnancy. Signs of decidualization in the left uterine tube

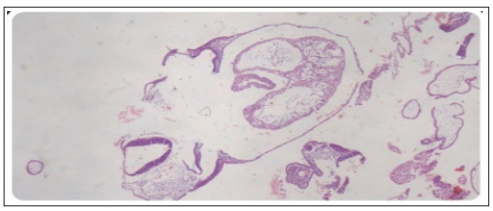


Figure 3c: Presence of fetal tissue and chorionic villi as well as fragments of the decidua

Discussion and Conclusion

Tubal twin ectopic pregnancy is a very rare disease, with a very high morbidity. In the literature there are around 100 reported cases, fifteen of which with a detected fetal heartbeat diagnosed by ultrasound examination [7].

The most important risk factors for the development of ectopic pregnancy are tubal microsurgery, cigarette smoking, pelvic inflammatory processes, endometriosis, and use of reproductive medicine techniques (artificial insemination and in vitro fertilization) [5,6]. However, its etiology is unknown. Twin ectopic pregnancy has a 30%-50% greater chance of rupture when compared to a single ectopic pregnancy and is generally unilateral [8,9].

From an anatomopathological point of view, twin tubal ectopic pregnancy presents characteristics like those of single tubal ectopic pregnancy. The embryos are implanted in the uterine tubes probably because there is a failure in the paracrine interaction mechanism between tubal epithelial cells, smooth muscle cells, immune cells, and the blastocyst [10,11]. Any changes in these signs can lead to an unfavorable tubal microenvironment and lead to inadequate and early implantation of the embryo in the fallopian tube, thus leading to the growth of fetal tissue and the formation of the gestational sac [10-13].

Over time, symptoms may manifest, such as abdominal pain, abnormal vaginal bleeding and breast tenderness [14].

Early diagnosis of twin tubal ectopic pregnancy is essential to avoid serious complications, such as rupture of the uterine tube and internal bleeding. For diagnosis, the gold standard is the association of ultrasound examination and serum levels of plasma β-hCG examination, using the discriminatory zone. Imaging tests, such as transvaginal ultrasound, can be performed to identify the location of the gestational sac and its degree of chorionicity; when plasma β-hCG levels reach values around 1500 IU/L, the imaging test provides high specificity-sensitivity [14].

However, it is important to highlight that the accurate diagnosis of twin tubal ectopic pregnancy can be challenging, as it can be confused with a multiple intrauterine pregnancy in the early stages [15].

Regarding treatment, it can be surgical (laparotomic or laparoscopic) or medication (use of methotrexate). It is important to highlight that there are criteria for using methotrexate [10].

The criteria for using drug therapy (methotrexate) are: hemodynamic stability, adnexal mass smaller than 3.5 cm, β-hCG dosage less than 5,000 mIU/mL, absence of fetal heartbeat, desire for future pregnancy, patient's agreement to undergo follow-up, and normal kidney function. It is contraindicated in intrauterine pregnancy, active lung disease, anemia, peptic ulcer, leukopenia (leukocyte count lower than 2,000/mL), and thrombocytopenia (platelet count lower than 100,000/mm) [16].

In summary, twin tubal ectopic pregnancy is an extremely rare condition in which two embryos implant in the uterine tubes. Its early diagnosis is essential to avoid serious complications, including maternal death. A biochemical pregnancy test and transvaginal ultrasound are important diagnostic tools, but in some cases invasive procedures may be necessary to confirm the diagnosis and provide appropriate treatment [6].

In this case report, it should be highlighted that twin pregnancy with cardiac activity was diagnosed on ultrasound examination, in addition to visualizing two gestational sacs and two embryos on ultrasound, being classified as dichorionic and diamniotic tubal twin ectopic pregnancy, which is an especially rare form of manifestation. This was possible thanks to serial β-hCG examination and high-resolution ultrasounds, to establish early diagnosis and institute timely and effective therapeutic measures.

Patient evolution in this case was excellent, and after 6 months the patient achieved natural pregnancy, with a visualized corpus luteum ipsilateral to the removed uterine tube (external transmigration). The pregnancy progressed to 37 weeks, with a cesarean section and the birth of a live and active newborn.

Authors Contributions to the Manuscript

José Roberto Lambert - conceptualization, data collection, statistics, visualization, validation, writing original draft, writing editing and review.
Giovanna Leite Calixto Saldanha - visualization, validation, writing original draft, writing editing and review.
Rogério Gomes dos Reis Guidoni - visualization, validation, writing editing and review.
Edilson Raymundo Martins Lira Filho - visualization, validation, writing editing and review.
Eduardo Carvalho de Arruda Veiga - visualization, validation, writing editing and review.
Luciano de Melo Pompei - conceptualization, visualization, validation, writing editing and review.

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Conflict of Interest

The authors have no conflict of interest to disclose.

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