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Case Report

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Molar Cornual Ectopic Pregnancy: Case Report and Literature Review

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Abstract

Ectopic molar pregnancy is very rare and of all possible locations, the uterine horn is one of the least frequent. The diagnosis represents a challenge and is made through clinical, imaging tests and serum levels of beta human chorionic gonadotropin. Uterine rupture is very frequent and is associated with significant morbidity and mortality, so early management is very important to prevent the development of complications. We present the case of a 24-year-old patient who was diagnosed with a molar cornual ectopic pregnancy, an abdominal hysterectomy was performed before it was complicated with uterine rupture.

Keywords: Hydatidiform Mole, Molar Cornual Ectopic Pregnancy

Introduction

The incidence of ectopic pregnancy has increased in the last decades as a result of the pelvic inflammatory disease, assisted reproduction techniques, ovulation inducing drugs and tubal surgeries. Now, thanks to higher sensitivity of diagnostic methods, we can diagnose and treat before complications develop [1].

Cornual ectopic pregnancy corresponds to 2% - 4% of all ectopic gestations. Because of its location between the ostium and the isthmus portion of the tube there is a greater muscle mass with abundant vascularization generating a high risk of blood loss and death in case of uterine rupture [1,2].

Hydatidiform mole is caused by abnormal fertilization, it can be of two types, partial or complete. Molar pregnancy is characterized by hydropic changes that affect placental villi [3].

Molar cornual ectopic pregnancy is extremely rare, only 40 cases of molar ectopic pregnancies have been described in the medical literature, of these only 4 were located in the cornual region, having an incidence of 0.04% or approximately 1.5 cases in one million pregnancies [3].

In the last 20 years the traditional management has been cornual resection or laparotomy hysterectomy, but thanks to better diagnostic methods and the progress of minimally invasive surgery, early diagnoses have been achieved before its rupture [2].

Case Report

A 24-year-old woman, gravida 5 para 2 miscarriage 2, with uncertain last menstrual period, who was taking combined oral contraceptives, came to the emergency department with pelvic pain and vaginal bleeding. On physical examination the abdomen was soft, with no signs of peritoneal irritation or tenderness, thick posterior closed cervix and mild vaginal bleeding was noticed, she had no fever and her vital signs were stable. Vaginal ultrasound revealed a heterogeneous echogenic mass in the right uterine horn, its diameter was about 30 mm, with multiple cysts inside and important peripheral vascularization with color-flow Doppler (Fig. 1), the serum levels of beta human chorionic gonadotropin (β-HCG) was 14922 mIU/mL. An ectopic molar pregnancy was suspected, so a diagnostic laparoscopy was performed, in which the cornual location was confirmed after visualizing a marked distention of the right uterine horn, both adnexa were normal and there was no hemoperitoneum (Fig. 2), because the patient did not want to preserve her fertility, a total abdominal hysterectomy was made without complications (Fig. 3). The day after surgery serum β-HCG was 1732 mIU/mL and it was negative within 2 weeks. The pathology reported molar gestation that invades the myometrium indicative of invasive molar pregnancy. Until the third month of follow-up, no metastatic activity has been reported.

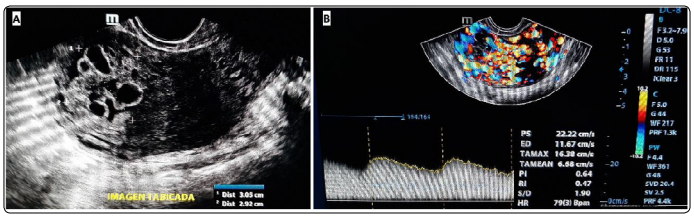

Figure 1: Vaginal ultrasound view: A, Heterogeneous echogenic multicystic mass in the right uterine horn with a diameter of 30 mm, suggestive of molar cornual ectopic pregnancy. B, Color-flow Doppler showed important peripheral vascularization, the pulsed Doppler showed a high velocity blood flow with low resistance (Resistance Index 0.47)

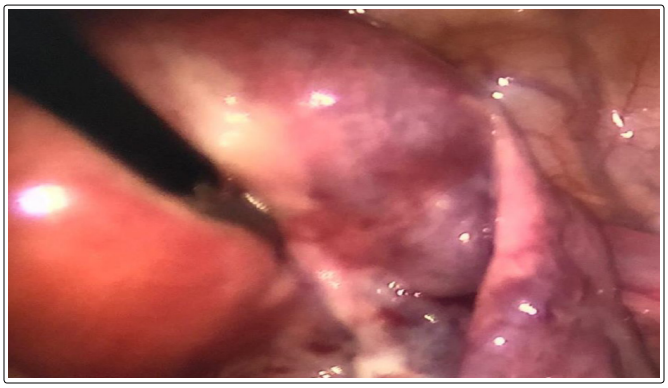

Figure 2: Diagnostic laparoscopy evidencing the marked distension of the right uterine horn, from which the uterine tube emerges without macroscopic alterations

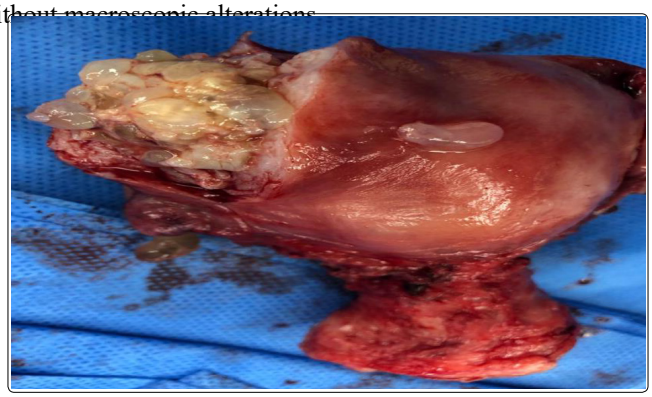

Figure 3: Surgical piece where the resection and opening of the right uterine horn allows visualization of multiple hydropic villi

LABORATORY TESTS				
	ADMISSION DAY	1 DAY AFTER SURGERY	2 WEEKS AFTER SURGERY	15 WEEKS AFTER SURGERY
serum β-HCG (mIU/mL)	14922.50	1732	40.94	1.7
White blood cells count (K/mm^3)	6.10	6.10	-	6.80
Hemoglobin (g/dl)	12.80	11.60	-	14.20
Hematocrit (%)	40.60	32.50	-	41.50
Platelets (K/mm^3)	125	123	-	162

Methodology

Several documentary sources were used to locate the bibliographic documents. A bibliographic research was performed using the descriptors: scientific writing, reviews, concept maps, critical reading. The records obtained ranged from 15 records after the combination of the different keywords.

Discussion

Brief Historical Review

Before knowing the function of the uterine tubes, Albucassi in 1063 made the first description of the ectopic pregnancy, it was Primerose in 1594 who described an abdominal pregnancy with a dead fetus. The function of the tubes was one of the discoveries of Gabriel Falopio (1523-1562), in the 17th century the ectopic pregnancy integrates a nosological picture from works by Cyprianus, Manget, Duverney and Littre. In the 19th century a successful therapy was achieved, it was Lawson Taint in 1883 who practiced salpingectomy with maternal survival. The first to report a tubal mole was Otto in 1871 [1].

Epidemiology

The incidence of ectopic pregnancy has increased to 1-2%, this has been due to three conditions, increased risk factors, assisted reproduction techniques and the use of more sensitive diagnostic methods that allow to detect cases that could have gone unnoticed. As the incidence increases, mortality decreases thanks to the early diagnosis and current therapeutic possibilities [1].

According to established data, it is estimated that the incidence of ectopic pregnancy varies according to geographical location due to multiple associated factors that are expressed differently in each country or region [1].

In Ecuador during 2011, the National Institute of Statistics and Censuses reported a total of 241 maternal deaths, of which 0.83% was due to an extrauterine pregnancy, in 2016 atotal of 166 maternal deaths occurred and 6.02% were associated with ectopic pregnancy [4].

Diagnosis

Molar cornual ectopic pregnancy is an extremely rare entity, with signs and symptoms similar to a normal ectopic pregnancy, so its diagnosis represents a challenge, with very few cases diagnosed preoperatively [5,6]. In its initial clinical presentation 70% of patients have abdominal pain, 61% vaginal bleeding and 67% hemoperitoneum [7]. In developing countries the diagnosis is usually late and with associated uterine rupture, it has been reported that the rate of rupture in ectopic molar pregnancies is higher (67%) compared to normal ectopic pregnancies (25.2% - 29.5%) this could be due to the more invasive behavior of gestational trophoblastic disease [7,8].

The initial imaging test is vaginal ultrasound with color Doppler flow, in which the characteristic finding is a heterogeneous mass with multiple small cysts located in the uterine horn, however its diagnostic efficacy for ectopic molar pregnancies is controversial, evidencing this classical ultrasound description in less than 50% of cases [5-7,9,10].

Magnetic resonance imaging (MRI) has been used as a diagnostic tool, which clearly delimits the cornual localization of the mass with cystic aspect, isointense in T1-weighted, hyperintense in T2-weighted and showed significant gadolinium contrast enhancement.

On the other hand, if the mass has poorly defined margins and central vascularization in the MRI, it is suggestive of an invasive mole [7].

It is established that intrauterine molar pregnancies have much higher serum levels of β-HCG than normal pregnancies. However, the serum β-HCG values of ectopic molar pregnancies vary from 6,642 to 15,678 mIU/mL in partial moles and from 7,920 to 24,733 in complete moles; being similar to those described in normal ectopic pregnancies (1,256 to 13,494 mIU/mL). This is why serum values of β-HCG alone are not useful to distinguish between an ectopic molar pregnancy and a normal ectopic pregnancy, the explanation for this behavior could be that the ectopic implantation of the mole does not allow adequate vascularization producing lower serum levels of β-HCG [5,7].

Treatment

Early diagnosis and prompt treatment of molar cornual ectopic pregnancy play an important role because they reduce morbidity and mortality. Traditional treatments for cornual pregnancy have been cornuectomy and hysterectomy, nevertheless both limits future reproductive function [11]. Laparoscopic surgery has replaced open surgery in this type of approach, and should be the treatment of choice in suitably selected and hemodynamically stable patients [2].

Laparoscopic Cornuostomy, unlike the cornual resection, is a less aggressive surgery, which generally produces less bleeding, but must be performed by surgeons with experience in endoscopic sutures and knots, otherwise there is a high risk of bleeding with catastrophic results for the patient. For this technique the diameter of the uterine horn should be less than 4 cm, since in the case of subsequent pregnancies, the probability of uterine rupture is significantly lower [2].

Conclusion

Molar cornual ectopic pregnancy is extremely rare. The diagnosis is difficult because clinical and serum values of β-HCG are similar to those of a normal ectopic pregnancy, and the characteristic ultrasound pattern is observed in less than half of the cases. The treatment of choice is laparoscopic cornuostomy in suitably selected and hemodynamically stable patients.

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