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Case Report

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PARTIAL MOLAR PREGNANCY WITH LIVE FETUS COMPLICATED BY INTRAUTERINE GROWTH RESTRICTION AND SEVERE PREECLAMPSIA: CASE REPORT

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ABSTRACT

Objective: To report the case of a partial molar pregnancy with live fetus and conduct a review of the literature regarding maternal and fetal complications associated to this condition. **Materials and methods**: Case report of a partial mole with a 33 weeks live fetus complicated by intrauterine growth restriction, oligohydramnios and severe preeclampsia. We report satisfactory maternal and neonatal outcomes and 1-year follow-up. A search was conducted in the Medline via Pubmed, LILACS, Ovid, Uptodate and Google Scholar databases using the following MESH terms: hiditadiform mole, partial mole, live fetus, coexisting live fetus. Case series and case reports of pregnant women with coexisting partial mole and live fetus at the time of diagnosis were selected and information regarding maternal and fetal prognosis was extracted. **Results:** Initially, 129 related titles were identified. Of these, 29 met the inclusion criteria, and 4 articles were excluded due to failed access to the full text. Overall, 31 reported cases were included; 9 ended in miscarriage, 8 in fetal demise or perinatal death, and 14 (45%) resulted in a live neonate. The most frequent maternal complication was preeclampsia in 6 (19.35%) cases. **Conclusion:** The coexistence of a partial mole with a live fetus poses a high risk of adverse perinatal outcomes and preeclampsia. The volume of information regarding this rare condition must be increased in order to better determine potential interventions in cases of euploid fetuses and to provide adequate counseling in clinical practice. Therefore, reporting these cases is important to build sufficient evidence about the natural course of this condition.

INTRODUCTION

Hydatidiform mole is one of the presenting forms of gestational trophoblastic disease, along with choriocarcinoma, invasive mole and placental site trophoblastic tumour.[1] placental site trophoblastic tumour.[1] Hydatidiform mole hydatidiform mole is a placental pathology characterised by hyperproliferation of the placental villi and hydroproliferative and hydropic degeneration.[2]

The mole is classified as complete or partial.[3] Complete hydatidiform mole is characterised by a placental abnormality that has a diploid set of chromosomes of diploid set of chromosomes of paternal origin, with no fetal tissue present.[4] presence of fetal tissue.[4] From a cytogenetic point of view cytogenetically, it develops after an egg, whose genetic material has been lost or damaged, is fertilised by one or two sperm fertilised by one or two spermatozoa, resulting in a diploid zygote. results in a diploid zygote of androgenetic origin.[3] The karyotype is 46 XX in 90% of cases and 46 XY in 10%. and 46 XY in 10% of cases,[1] Partial mole is characterised by placental disruption plus the presence of fetal tissue.[2] plus the presence of foetal tissue.[5] Candelier indicates that, according to Golubovsky's theory, it is caused by fertilisation of a normal ovum by two spermatozoa, giving rise to a triploidy in most cases, so that in the majority of cases it triploidy in most cases, so it is usually a polymalformed foetus polymalformed foetus that ends in miscarriage.[2] Without However, there are reported cases of euploid fetuses fetuses that can go on to viable pregnancies.[6]

The incidence of complete mole varies in different regions; the highest incidence is in Southeast Asia, with an incidence of 13 per 1,000 pregnancies, and the lowest in South America, with an incidence of 0.3 per 1,000 pregnancies.[3] of 0.3 per 1,000 pregnancies.[3] The incidence of partial mola is 3 per 1,000.[1] and the incidence of the coexistence of live foetus with partial mole is 1 in 22,000 to 1 in 100,000.[6,7]

The coexistence of mola and live foetus may be due to the presence of a pregnancy. be due to the presence of a multiple pregnancy, where one pregnancy is normal, with a normal foetus and placenta, and the other is a normal normal, and the other is a molar pregnancy, either

a full or partial mola. The other possibility, much less frequent, as mentioned above, is that it is a is a partial mole with a living eupliode foetus, of which there are only a few cases reported in the literature.[6,8] Literature.[6,8]

There are three major reviews of the coexistence of molar pregnancy and living foetus, the first by Vejerslev in 1991.[7,8]

by Vejerslev in 1991, which analyses the literature from 1903 to 1989, and reports 113 cases of complete molar with coexistent of complete molar pregnancy with coexisting live foetus; of these, 3 cases of these, 3 cases corresponded to partial mole with a live foetus.[6] live foetus.[6] The second review is the one by Wee and Jauniaux in 2005, in which they report 174 cases of complete mole and live foetus cases of complete mole and live foetus.[9] The third is by Lin et al. in 2017, who report a series of 72 cases of multiple series of 72 cases of multiple pregnancies with complete molas and live foetuscomplete mole and live foetus, and 9 articles are reviewed, which together together add up to a further 173 cases.[10]

The prenatal diagnosis of molar pregnancy and live foetus is based on elevated beta HCG levels and ultrasonographic findings.[9] ultrasonographic findings.[9] It has been described that ultrasound that ultrasound has been reported to have a sensitivity of 44% and a 74% specificity in the first trimester, and that the differential diagnosis is incomplete abortion incomplete abortion and anembryonic egg.[11]

weekly monitoring of the beta-human chorionic gonadotrophin (B HCG) beta-human chorionic gonadotrophin (B HCG) fraction after the end of until it is negative.[14]

It has been reported that the presence of a twin pregnancy with mola and a live foetus is associated with an increase in maternal complications such as increased maternal complications such as pre-eclampsia in 20-30%.[6,9] vaginal bleeding in 46%, hyperemesis gravidarum in 17%.[6]

From the foetal point of view, wide variability in survival data is reported. variability in newborn survival data is reported, ranging from 25%.[9,10,11] ranging from 25%,[9] to nearly 60%.[6,10] 60%[6,10] In these newborns, a significant presence of growth restriction is reported significant presence of intrauterine growth restriction intrauterine growth restriction and preterm delivery.[6]

As can be seen, there are several publications on complete molas and live foetus, however, the information is However, there is limited information on cases of partial molas specifically on cases of partial molas with live foetus, in terms of treatment according to live foetus, in terms of treatment according to the gestational gestational age at diagnosis, and fetal and maternal prognosis.

and maternal prognosis, so our aim is to make a case report and review the literature, focusing on the maternal outcome literature, focusing on the maternal and fetal outcome of this condition. fetal outcome of this condition.

CASE REPORT

We present the case of a 19-year-old woman, primigravid, 33 weeks gestation, resident of a rural area in the department of Meta, in the Meta region of the country rural area in the department of Meta, in the Eastern Plains region of of the Eastern Plains region of Colombia, with a one-week clinical of generalised oedema and blood pressure of 154/94, and was referred to the city of Bogota with a diagnosis of pre-eclampsia. diagnosis of pre-eclampsia. She had a negative medical and surgical history and a family history of hypertension. hypertensive mother. She had started her prenatal check-up since week 16, when a weight of 53 kg and a height of 160 cm was recorded. height 160 cm. Mandatory laboratories of the prenatal control and thyroid stimulating hormone (TSH) within normal limits. TSH) within normal limits. She had three ultrasounds from her prenatal check-up. Ultrasound. week 16: fetus with normal anatomy, normal fluid and placenta. placenta normal. Ultrasound 22nd week: fetus with hepatic cyst, normal amniotic fluid, and placental lesion placental lesion suspicious of placental accretism. 32nd week ultrasound: intrauterine growth restriction: biometry for 28 weeks, oligohydramnios (ILA 4 cm). Placenta with suspected placental placental accretism. In March 2018, the patient was admitted to the maternal and child the maternity and infant ward of the La Victoria hospital, a third level institution located in the city of Bogotá, which attends patients belonging to the state-subsidised subsidised by the State in the General Social Security System in Social Security System in Colombia. The patient is admitted alert, alert, hydrated, weight 61 k, blood pressure 150/100, uterine height 150/100, uterine height of 28 cm, oedema grade II of the grade II oedema of the lower limbs and the rest of the physical examination without alterations. Laboratory tests and detailed ultrasound detailed ultrasound was requested. The anatomical detail obstetric ultrasound yields the following results: estimated foetal weight, 830 g; growth below the 3rd percentile for gestational age. gestational age. In the assessment of the placenta an image measuring 90 x 167 mm was found, with multiple hypoechoic multiple hypoechoic images in a honeycomb pattern, suggestive of a suggestive of gestational trophoblastic disease (figures 1 and 2). gestational trophoblastic disease (figures 1 and 2). Assessment of amniotic fluid amniotic fluid assessment confirms oligohydramnios (ILA 3). Doppler evaluation ruled out placental accreta. The Doppler fetal haemodynamics shows increased resistance in the umbilical artery. Normal resistances in middle cerebral artery and venous

ductus. Fetal assessment shows a female foetus with a focal hepatic lesion.female foetus, with focal hepatic lesion in the right lobe of 25 x 26 mm, hypoechoic 25 x 26 mm, irregular hypoechoic, with small septa, suggestive of a small septa and suggestive of hepatic mesenchymal hamartoma (Figure 3); the rest of the foetal anatomy was normal. normal foetal anatomy. A diagnostic impression was made of 33 weeks' gestation, pre-eclampsia without criteria of severity, trophoblastic disease with coexistence of a live foetus, intrauterine growth restriction, oligohydramnios and intrauterine growth restriction, oligohydramnios, foetal liver cyst: suspicion of hepatic hamartoma. As for the the haemoglobin, 14 g/dl; haematocrit, 41%; leucocytes, 12,300/mm3, 14 g/dl; haematocrit, 41%; leukocytes, 12,300/mm3 platelets, 166,000/mm3 Creatinine, 0.6 mg/dl; transaminases: AST, 32 IU, ALT, 34 IU; 24-hour proteinuria, 400 mg. 24-hour proteinuria, 400 mg. No gonadotropin was taken was not taken due to the advanced stage of gestation.

The patient went into labour spontaneously 6 hours after admission. spontaneous onset of labour 6 hours after admission, at 33 weeks of gestation. After 6 hours of labour, vaginal delivery was attended and the newborn was delivered without complications. vaginal delivery with a female newborn weighing 900 g, APGAR 8-9-10, at 1, 5 and 10 minutes. minutes. Placenta Shultze type complete.

After delivery, in the delivery room, she presented haemorrhage in the immediate puerperium secondary to uterine atony. uterine atony, with a heart rate of 114 beats/min. min, blood pressure 148-95 mm/Hg. Thepostpartum haemorrhage protocol, known as code red in code red in Colombia (15); the haemorrhage stopped with pharmacological with pharmacological management with oxytocin. Uterine revision under anaesthesia uterine revision under anaesthesia, transfusion of 2 U of red blood cells were transfused. Laboratory reports taken during the application of code red: haemoglobin, 11 g/dl; hematocrit, 34%; platelets, 183,000 / mm3 Fibrinogen, 775 mg/dl; D-dimer, D 30 ng/ml.

In the early puerperium, she presented blood pressure figures with blood pressure with criteria of severe pre-eclampsia (184/111 mm/Hg) which were not controlled with the administration of nifedipine and administration of nifedipine and clonidine, so she was transferred to the transferred to the intensive care unit and managed with labetalol in labetalol by continuous infusion at 1 g/h; transient loss of vision was reported and a computerised axial tomography computerised axial tomography (CAT) scan of the brain was performed and reported as normal. An increase in proteinuria increase in proteinuria over 24 h to 6.3 g, with the rest of the totemic profile within normal limits. After 72 h the patient was discharged from the Intensive Care Unit and after 15 days the patient was discharged from hospital, with a formula of losartan 150 mg/day, nifedipine 90 mg/day and clonidine 900 mcg/day. The final report of placental pathology shows monochorial placenta monoamniotic, 550 g, in which chorionic villi with hydropic degeneration are chorionic villi with hydropic degeneration, reported as partial mole type gestational trophoblastic disease.

The newborn received two doses of surfactant and required ventilatory support for 5 days. and required ventilatory support for 5 days. She presented She had neonatal sepsis due to Kluyvera ascorbata BLEE, managed with ertapenem, with a positive evolution. During The following tests were performed during hospitalisation: total abdominal ultrasound showing a cystic image compatible with hamartoma cystic image compatible with hamartoma, normal transfontanelar normal transfontanelar ultrasound, normal echocardiogram, normal renal ultrasound and normal karyotype,normal renal ultrasound and normal 46XX karyotype. She is assessed by paediatric surgery who considered outpatient control. She was hospitalised for 62 days and she was discharged with home oxygen supplementation due to for moderate pulmonary dysplasia. Follow-up mother and child at one year, with no evidence of persistent trophoblastic persistent trophoblastic disease, with negative b-HCG negative and without antihypertensive drug requirements. The girl had a satisfactory evolution. Ethical considerations. The patient was informed of the diagnosis, signed informed consent for each procedure performed and signed an express consent for the publication of this case. Precautions were taken to ensure confidentiality of information and anonymity. confidentiality of information and anonymity of the patient. anonymity of the patient; the photographic record was taken by the authors.

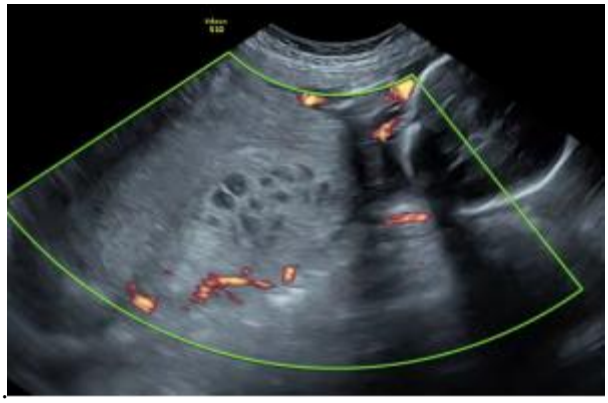

Figure 1: Ultrasound image, with normal placenta area and altered placenta area with normal placenta and altered placenta with hydropic degeneration. Patient with live foetus pregnancy and partial mole, Bogotá (Colombia), 2018

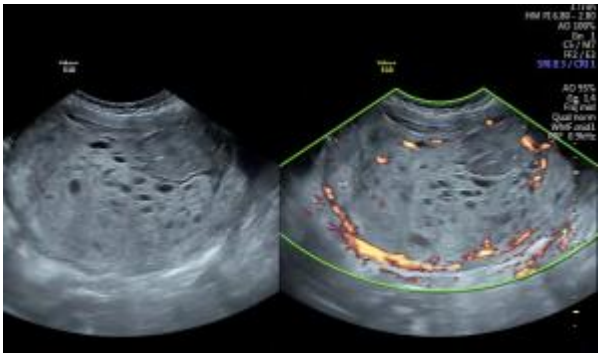

Figure 2: Close-up of the placenta with classic honeycomb pattern, suggestive of trophoblastic honeycomb pattern, suggestive of gestational trophoblastic disease. gestational trophoblastic disease. Patient with live foetus pregnancy and partial mole, Bogotá (Colombia), 2018.

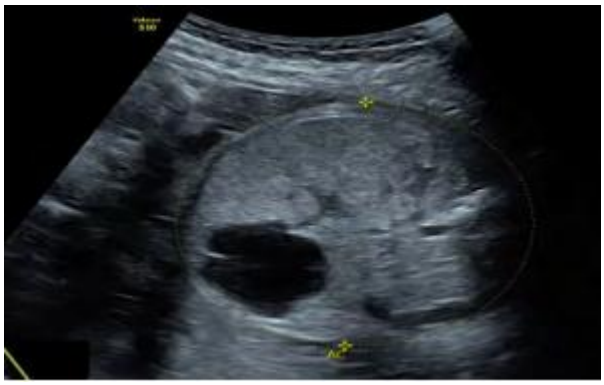

Figure 3: Fetal intrahepatic cystic image, suggestive of hamartoma of hamartoma. Patient with a live foetus pregnancy and partial mola partial, Bogotá (Colombia), 2018.

MATERIALS AND METHODS

To answer the question of maternal and foetal complications associated with cases of partial mole and live foetus, a search was conducted partial mole and live foetus, a literature search was carried out in the literature search was performed in the following databases: Medline via PubMed, google scholar and LILACS, with the following terms MESH: "hydrocephalus following MESH terms: "hiditadiform mole", "partial mole", "live fetus" and "live fetus", "partial mole", "live fetus", "coexisting live fetus", in English and Spanish, with no time limit. Criteria Inclusion criteria: reports or case series; population type type of population: pregnant women diagnosed with incomplete mole with live fetus. We used the snowball" method and a search was carried out based on the references from the references cited in each article.

The selection and extraction of data and review was carried out by one author and by one author and verified by the other. Information on the following variables was taken from information was collected on the following variables: country where the case occurred, maternal age, gestational age, diagnosis, method of diagnosis, type of management (expectant management, pregnancy termination), type of delivery (caesarean section or vaginal delivery), fetal outcome (foetal fetal outcome (fetal death, live newborn, intrauterine growth retardation, congenital malformations), maternal complications (antepartum, intrapartum or postpartum), newbornpostpartum), newborn complications. The results are presented in a narrative form.

RESULTS

Initially, 128 titles and abstracts were identified; of these, a total of 29 studies met the inclusion criteria, of which 25 met the inclusion criteria, of which 25 met the inclusion criteria. met the inclusion criteria, of which 25 had access to the full text and only access to the full text and in 4 studies only the abstract was the abstract, which is why they were discarded from the review.[16-19] from the review.[16-19] The 25 included studies corresponded to 25 case reports.

In total 31 cases of coexistence of incomplete mole and live foetus were found.

Study site: A total of 5 studies were conducted in the United States.[13,20] conducted in the United States.[13,20-23] three in India.[24-26] two in Mexico.[27,28] two in China.[29,30] and two in Turkey.[29,30] two in Turkey.[34,41] and the other reports were in were in: Australia,[31] the Netherlands,[32] England,[33] Pakistan,[35] Nepal,[36] Poland,[42] Bangladesh,[37] Chile,[38] Malaysia,[39] Italy,[43] and Canada.[40]

Maternal age was found to be in the range of 17 to 29 years in 25 cases (13.5 to 13.5 years). 29 years in 25 cases[13,22-29,32-37,39-42] from 30 to 39 years in 6 cases.[20,21,23,30,31,43] and older or equal to 40 years in no cases. 40 years of age in none of the cases. Regarding the gestational age gestational age at diagnosis was before 22 weeks in 17 cases 22 weeks in 17 cases[20-23,25,28,30,30,33-35,40,23,35,40] and 23 to 40 years of 35,40), from 23 to 27 weeks in 4 cases.[29,32,38,41] from 28 to 37 weeks in 6 cases.[29,32,38,41] from 28 to 37 weeks in 6 cases[24,27,36,37,39] and 37 weeks or more in 4 cases (13,38,41). weeks or more in 4 cases (13,26,31). The method of diagnosis was prenatal ultrasound in 21 cases,[20,21,23-25,27,28,30,32-35,37,40,42-43] casos[20,21,23-25,27,28,30,32-35,37,40,42-43] and at delivery or due to pathology in 10 cases.[13,26,29,31,36,38,13,26,29,31,36,38,39,41,43] Termination of pregnancy was offered in 4 cases.[20,23]

Gestational age at the time of termination of pregnancy ranged pregnancy ranged from 15 to 40 weeks: 22 weeks or less in 9 cases.[20,23]

22 weeks or less, 9 cases[20,22,23,28,40] from 23 to 27 weeks, 4 cases (29,29,28,40). to 27 weeks, 4 cases[29,32,34,38] from 28 to 37 weeks, 13 cases.[21,24- 25- 26-28,30,33,36,39,41,42- 43] and from 38 to 42 weeks, 5 cases.[13,26,31,35,37]

Regarding the type of delivery: caesarean delivery, 12 cases,[8,21,26,27,30-34,39,42,43] vaginal delivery, 10 cases.[13,25,28,29,29,39,42,43,13,25,28,29,35-37,41,42]

Fetal outcome: 9 cases ended in miscarriage.[20,22,23,28,40] 8 cases had early neonatal death or stillbirth,[24,28,29,32,34,38,39,41] Intrauterine growth restriction, 6 cases,[24,25,27,37,39,43] fetal malformations, 2 cases; one case of Dandy Walker,[26] and one case with myelomeningocele,[38] In 14 cases a live, viable foetus was live, viable foetus.[13,21,25,26,27,30,31,33,35-37,42,43]

Maternal complications were reported as follows: pre-eclampsia in 6 cases,[6,23,28,31,34,40] one case of postpartum haemorrhage and one case of case of postpartum haemorrhage requiring hysterectomy,[27] persistent trophoblastic disease in 3 cases.[4,23] and in 3 cases,[4,23] one case with hyperthyroid crisis,[41] there were no cases of sepsis or maternal mortality. maternal mortality.

Information on the case reports

CONCLUSION

The coexistence of partial mole with a live foetus presents a high risk of adverse perinatal outcome and pre-eclampsia. There is a need to increase the volume of information on this rare condition is required to determine possible interventions in cases of euploid fetuses and to provide adequate euploid foetuses and to provide adequate counselling in clinical practice, so it is important to report these cases in order to build sufficient is important in order to build sufficient evidence on the natural behaviour of the disease. the natural behaviour of the disease.

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