Medico Research Chronicles

ISSN No. 2394-3971

Original Research Article

SPONTANEOUS UTERINE RUPTURE RELATED TO EXCEPTIONAL RARE "TWIN PREGNANCY" - TROPHOBLASTIC PATHOLOGY AND ONGOING PREGNANCY

***Luciana Rotaru[1], Cristian Boeriu[2]**

1. Associate Professor, Department of Emergency Medicine and First Aid – University of Medicine and Pharmacy Craiova, ED

2 Associate Professor, Department of Emergency Medicine and First Aid – University of Medicine and Pharmacy Tirgu – Mures, ED

Submitted on: July 2016
Accepted on: August 2016
For Correspondence
Email ID:
lucianarotaru@yahoo.com

Abstract

Background: The peritoneal bleeding in intrauterine ongoing pregnancy is a serious event that poses problems of identifying and stopping the source of bleeding, long-term impact of hypovolemia, anemia, operative, anesthetics, imaging acceptance.

The purpose of the study -to highlight the very special circumstances and therapeutic strategies to treat a complicated "twin pregnancy"

Matherial and methods: Case presentation, 25 years old women (14-16 weeks ongoing pregnancy),arriving ED accusing abdominal pain, vomiting and a diarrheal stool from 3-4hours. Initially, hemodynamically stable, without fewer, with seemingly normal development of pregnancy, without free intraperitoneal fluid or vaginal bleeding, but uterine hypertonia. During ED surveillance gradually develops of an active intraperitoneal hemorrhage syndrome, uterine painful contractions and tenderness in the lower abdominal floor. No family agreement for abdominal invasive imagistic. At 2h extreme fetal bradycardia, progressive hemorrhagic shock, peritoneal fluid.. Laparatomy decided - extended uterine rupture with fetus expelled in peritoneal cavity and massive hemoperitoneum. Uterine structure modified by placental-site trophoblastic tumors. Balloon tamponade and hysteroraphyis.

Conclusions:

- A "twin pregnancy"(a GTD coexisting with a fetus) is exceptional for placental-site trophoblastic tumors type of GTD
- Spontaneous uterine rupture at this age of pregnancy is exceptional but evolving dramatic gravity, and fetus compromised

- In case of uncompliance about CT/angiography usage, alternate means, as MRI, should be setted
- Elucidation of type of trophoblastic gestational disease and treatment choices remains important
- A combined courageous therapeutic strategy applied because a further pregnancies are wished for the family, successfully - an other pregnancy obtained

Keywords: hemorrhagic shock, uterine rupture, "twin pregnancy", gestational trophoblastic disease

Introduction

The peritoneal bleeding in intrauterine ongoing pregnancy is a serious event that poses problems of identifying and stopping the source of bleeding, long-term impact of hypovolemia, anemia, anesthetics and operative act on fetus, imaging reasonable choices. But additionally, it poses questions about subsequently pathology generating bleeding, genetic or tumoral circumstances, further therapeutic goals and means and further pregnancy obtaining and development. All of these are usually sensible subjects to discuss with medical team involved in management equally with patient and her family, especially when it's about the first pregnancy, late pregnancy or artificially obtained pregnancy.

When suspected cause of bleeding is gestational trophoblastic disease problems can be very complicated, but a "twin pregnancy" (a GTD coexisting with a fetus) are an exceptional event with dramatic perspectives in case of severe, unexpected bleeding and uterine rupture.

Material and Methods

We are presenting a 25 years old pregnant women (14-16 weeks), takes ED accusing abdominal pain, vomiting and a diarrheal stool a few hours after a festive table. The pregnancy obtained in complex condition, after 4 in vitro fertilization attempts. Initially, hemodynamically stable, no fewer, with seemingly normal development of pregnancy, without free intra peritoneal fluid or vaginal bleeding, but uterine hypertonia with discrete defense in the lower abdominal floor. The placenta normally inserted, without retro placental hematoma. Elevated serum hCG level, according with pregnancy age. Increased spleen. During the 90','army'' survey gradually develops of an active intra peritoneal hemorrhage syndrome that shall not be reversed by the aggressive fluid and hematological resuscitation, associated with uterine painful contractions and tenderness in the lower abdominal floor. Abdominal CT/ arteriography [20], proposed but no agreement is received from the family. At 2-hour extreme fetal bradycardia, then intrauterine death, progressive hemorrhagic shock, free peritoneal fluid. FAST did not detect any new spleen abnormalities. Emergency surgical intervention decided, first intention being laparoscopic operative exploration. During preparations for this, dramatic abdominal pain occurred, and severe hemorrhagic shock rapid installed and progressed, so, large laparatomy practiced. At abdominal cavity exploration an extended uterine rupture on anterior median uterine area, with fetus expelled in peritoneal cavity (fig. 1) and approximately 3500 ml. blood in the peritoneum. Uterine

structure modified by placental-site trophoblastic tumors.

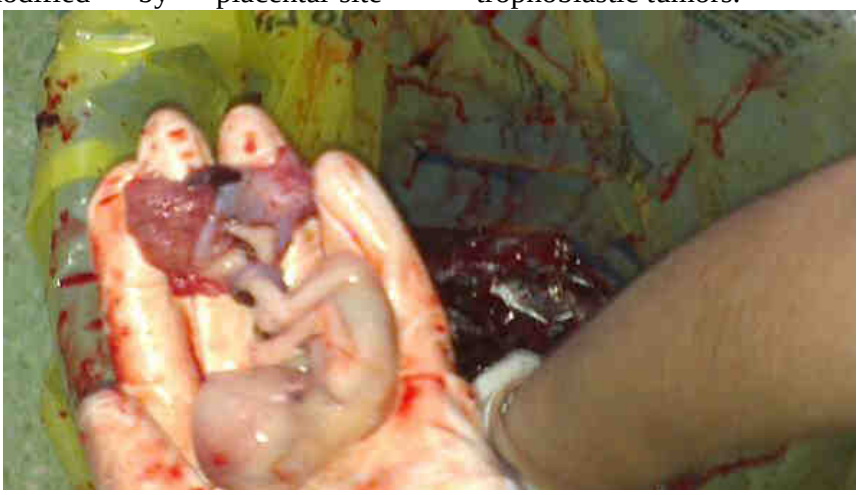

Figure 1 – fetus and placental fragment (intraoperative image)

Immediate balloon tamponade and hysteroraphyis practiced. Active fluid resuscitation, inotropic agents used. After mechanical tamponade of parietal disruption rapid hemodynamic compensation observed, encouraging the conservative procedure continuation.

After 24 hours the patient conscious, alert, hemodynamically stable, ongoing haematological compensation. No further events during the hospitalization. Discharged at 8 days.Systemic (intravenous) methotrexate (for 1 year) therapy started.

After three other years, another pregnancy obtained, either in vitro fertilization, finished through cesarean section birth.

Results & Discussion:

Some direction of debate identified in evolution of the case, consisting in key points of evaluation and management.

Suspecting the gestational trophoblastic disease was one of the first line on debate, having a lot of arguments for that: age of the women (not precisely typical but around the border line of risk which are cited lower than 20 years of age, or over 35 years of age[1,2]. In contrast with literature, no previous GTD [3] detected because not others previously pregnancy s, but multiple hormone therapy in attempts of pregnancy obtaining,

Gestational trophoblastic (jeh-STAY-shuh-nul troh-fuh-BLAS-tik) disease (GTD) is a group of rare tumors that involve abnormal growth of cells inside a woman's uterus, starting in the cells that would normally develop into the placenta during pregnancy.Most GTDs are benign non-invazive.

GTD begins in the layer of cells called the *trophoblast* (troh-fuh-BLAST) that normally surrounds an embryo. Early in normal development, the cells of the trophoblast form tiny, finger-like projections known as *villi*. The villi grow into the lining of the uterus. In time, the trophoblast layer develops into the placenta, the organ that protects and nourishes the growing fetus.

The main types of gestational trophoblastic diseases are. [4, 5]: hydatidiform (HY-duh-TIH-dih-form) mole (complete or partial), invasive mole, choriocarcinoma (KOR-ee-oh-KAR-sih-NOH-muh), placental-site trophoblastic tumor, epithelioid (ep-ih-THEE-lee-oyd) trophoblastic tumor

Diagnosis, excluding other causes of bleeding

Previously ultrasound tests realized from the personal obstetrician of the patient (the last, 2 days before the event), did not reveal any unconformities between placental implantation of fetal evolution. Vaginal bleeding, enlarged uterus, pelvic pain or discomfort, and hyperemesis are the most common symptoms of GTD, but only at the moment of emergency event the women

affirmed some of these symptoms, and their evolution already suggested a serious emergency situation developing, not only a dysgravidia Elevated serum hCG level was all the time accorded with pregnancy evolution. Since pregnancy is obviously the leading cause of elevated serum hCG, and fetal evolution under apparently normal progress, it is really hard for emergency consultants to suspect a complication such is a concomitance of GTD.

GTD should be confirmed histologically.[4], so, after uterus evacuation[5], placental highly vascular fragments remaining attached to the fetus (fig. 2) where elevated serum hCG level annalized. Placental-site trophoblastic tumors as a type of GTD confirmed.

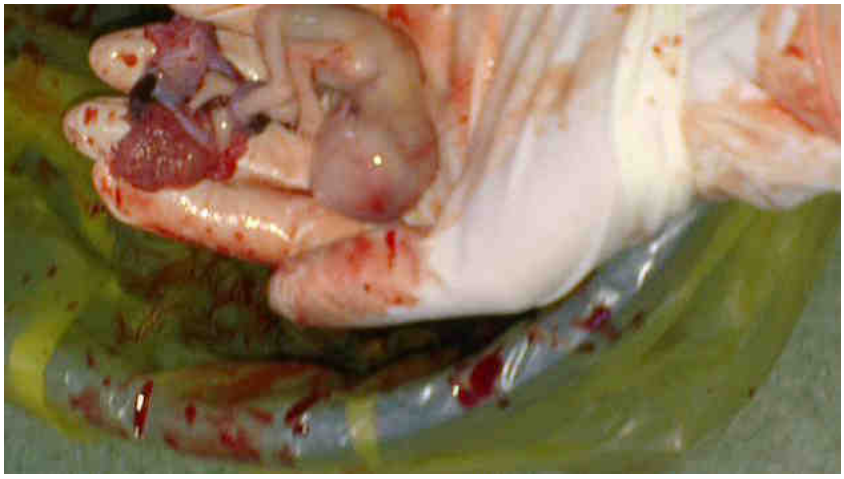

Figure 2 – modified placenta, placental haematoma, fetus (intraoperative piece)

Placental-site trophoblastic tumor (PSTT) is a very rare form of gestational trophoblastic disease (GTD) that develops where the placenta attaches to the lining of the uterus. This tumor most often develops after a normal pregnancy or abortion, but it may also develop after a complete or partial mole is removed. PSTT have a tendency to invade the layer muscle of the uterus. Some others such as exaggerated placental site, placental site nodules are not GTD [6], but could coexist.

At the first instant it was a strong suspicion of an other cause of peritoneal bleeding, such splenic rupture as a consequence of a splenic infaction, or traumatic event, but, even large, the spleen

Aspect where homogeneous, and the first space of free peritoneal fluid were into the lower abdomen spaces and between intestinal loop, associated with diarrhea, and peritoneal irritation syndrom

The "twin pregnancy"

It is a particularity of the case, which is very rare situation, a GTD coexisting with a normal fetus, called a "twin pregnancy". Evan if in theory and some specific situation, successful term delivery might be possible, the pregnancy should be allowed to proceed if the mother wishes, and following appropriate counseling. If this situation has been identified and only the pregnancy has a normal evolution, medical team should focus on the risks and opportunity's between continuing the pregnancy (without guarantees for the baby health or life) or to secure the mothers life. [7, 8]. The probability of achieving a healthy baby is approximately 40%, but there is risks of complications, pulmonary embolism, pre-eclampsia, retroplacental abcess,

hemorrhagic, general malignancy. The good news for the mothe is that, compared with women who simply had a GTD in the past, there is no increased risk of developing persistent GTD after such a twin pregnancy. [9, 10, 11, 12]

Therapeutic strategies choises

The standard treatment for hydatidiform mole consists of the evacuation of pregnancy.[13,14,15,16,17] Suction curettage is the preferred method of evacuation for this type of GTD. Hysterectomyis could be an alternative if no further pregnancies are wished for by the female patient.

Most forms of GTD are very sensitive to chemotherapy drugs, but PSTTs are not. Instead, they are treated with surgery, aimed at completely removing disease.

In this case particular surgical decision made, immediate balloon tamponade and hysteroraphyis as an alternate way for hysterectomyis, because further pregnancies are wished by the women. This technique are inspired by balloon tamponade technique of hemorrhage after uterine curettage for gestational trophoblastic disease[18,19], but the risks for a subsequently uterine rupture remains high, and this technique still remains reserved to a very narrow lot of cases, strictly under permanently monitoring and surgical facilities

Paraclinical methods of diagnostic. Imagistic resources usage

Usually, some imagistic procedures may be done to help find out the stage of the disease or the source of bleeding, the situation of placenta, differentiation of placental hematoma vs. retro placental hematoma uterus: chest x-ray, CT scan (CAT scan), MRI / (NMRI with gadolinium) , and in specific cases a lumbar puncture. Some of these techniques are invasive and harmful for the baby, and the patient and her family refused.

Arterioghaphy [20], proposed for two beneficial goals: to identify the bleeding source and attempt of selective embolisation, or other adequate interventional vascular techniques but no agreement is received from the family.

In this circumstances only MRI possible, but in this case it was no time to performed it because of the worsening condition, situation in that emergent surgery become the main priority.

Conclusions

The main conclusions deriving from this case is that any pregnancy with emergency problems is a multiple emergency case situation, with risks for mother and risks for baby, equal with chances for mother and chances for baby, but in most cases the best interest of the mother are not accordingly with the fetus one. The priorities should be stratified and an appropriate choice should be taken and applied at the right moment to preserve the most beneficial value which is no doubt about that, the mother's life.

The specific conclusions of the case are considered:

- A "twin pregnancy" (a GTD coexisting with a fetus) is a extremely rare circumstances of GTD, but exceptional for placental-site trophoblastic tumors type of GTD, and for this reason it does not exist some standard management strategies for emergency approach
- Myriad possible sources of intra peritoneal hemorrhage poses identification problem, taking into account that initially pregnancy seemed normal evolution, so proposed angiography could have equally therapeutic indications
- Spontaneous uterine rupture at this age of pregnancy is exceptional but evolving dramatic gravity, and fetus compromised
- The chance of the patient in this situation already complicated was to be right in the ED, in a high competence county hospital with complete advanced staff, so is important to precocious identify and complex monitories a suspicion of a twin pregnancy, to have the chance of early detection of the uterine rupture risk, fetal sufferance, metastasis.

- A particular issue which should be taken into account is the compliance of the patient and her family about imagistic invasive methods usage as CT or angiography, so, alternate means, as MRI, should be taken into attention.
- Elucidation of type of trophoblastic gestational disease and treatment choices remains important in the perspective of uterine suture for a possible next pregnancy.
- A heroic combined therapeutic surgical strategy applied (balloon tamponade and hysteroraphyis) as an alternate way for hysterectomyis because further pregnancies are wished for the family. After that, 1 year of systemic (intravenous) methotrexate. Follow – up at 2 years – another pregnancy obtained and no others incidents occurred - caesarean section for delivery, with no complications.

Aknowledgements:
All the authors have had equal contribution. The authors, Luciana Teodora Rotaru and Cristian Marius Boeriu declares no any commercial and financial interests.

References

1. Kohorn EI (June 2002). Negotiating a staging and risk factor scoring system for gestational trophoblastic neoplasia. A progress report. *J Reprod Med* **47** (6): 445–50.PMID 12092012.
2. Kohorn EI (2001). The new FIGO 2000 staging and risk factor scoring system for gestational trophoblastic disease: description and critical assessment. *Int. J. Gynecol. Cancer***11** (1): 73–7. doi:10.1046/j.1525-1438. 2001. 011001073 \.x.PMID 11285037.
3. Gestational Trophoblastic Tumors Treatment - National Cancer Institute. Retrieved 2010-03-21.
4. Sebire NJ (2010). Histopathological diagnosis of hydatidiform mole: contemporary features and clinical implications. *Fetal Pediatr Pathol* **29** (1): 1–16.
5. Lee SW, Kim MY, Chung JH, Yang JH, Lee YH, Chun YK (February 2010). Clinical findings of multiple pregnancy with a complete hydatidiform mole and coexisting fetus. *J Ultrasound Med* **29** (2): 271–80. PMID 20103799.
6. Gerulath AH, Ehlen TG, Bessette P, Jolicoeur L, Savoie R (May 2002). Gestational trophoblastic disease. *J Obstet Gynaecol Can* **24** (5): 434–46. PMID 12196865.
7. True DK, Thomsett M, Liley H, et al. (September 2007).Twin pregnancy with a coexisting hydatiform mole and liveborn infant: complicated by maternal hyperthyroidism and neonatal hypothyroidism. *J Paediatr Child Health* **43** (9): 646–8. doi:10.1111/j.1440-1754.2007.01145.x.PMID 17688651.
8. Behtash N, Behnamfar F, Hamedi B, Ramezanzadeh F (April 2009). Term delivery following successful treatment of choriocarcinoma with brain metastases, a case report and review of literature. *Arch. Gynecol. Obstet*. **279** (4): 579–81.doi:10.1007/s00404-008-0753-x. PMID 18726607.
9. Suri S, Davies M, Jauniaux E (2009). Twin pregnancy presenting as a praevia complete hydatidiform mole and coexisting fetus complicated by a placental abscess. *Fetal. Diagn. Ther*. **26** (4): 181–4.doi:10.1159/000253272. PMID 19864876.
10. Dolapcioglu K, Gungoren A, Hakverdi S, Hakverdi AU, Egilmez E (March 2009). Twin pregnancy with a complete hydatidiform mole and co-existent live fetus: two case reports and review of the literature. *Arch. Gynecol. Obstet*.**279** (3): 431–6. doi:10.1007/s00404-008-0737-x.PMID 18679699.
11. Vandenhove M, Amant F, van Schoubroeck D, Cannie M, Dymarkowski S, Hanssens M (May 2008). Complete hydatidiform mole with co-existing healthy fetus: a case report. *J. Matern. Fetal. Neonatal. Med*. **21** (5): 341–

4.doi:10.1080/14767050801925156. PMID 18446663.
12. Shih IM, Seidman J D, Kurman RJ (June 1999). Placental site nodule and characterization of distinctive types of intermediate trophoblast. *Hum. Pathol*. **30** (6): 687–94.doi:10.1016/S0046-8177(99)90095-3. PMID 10374778.
13. Lurain JR (January 2011). Gestational trophoblastic disease II: classification and management of gestational trophoblastic neoplasia. *Am. J. Obstet. Gynecol*. **204** (1):11–8. doi:10.1016/j.ajog.2010.06. 072. PMID 20739008.
14. Sebire NJ, Seckl MJ (2008). Gestational trophoblastic disease: current management of hydatidiform mole. *BMJ* **337**: a1193. doi:10.1136/bmj.a1193. PMID 18708429.
15. Berkowitz RS, Goldstein DP (April 2009). Clinical practice. Molar pregnancy. *N. Engl. J. Med*. **360** (16): 1639–45.doi:10.1056/NEJMcp0900696. PMID 19369669.
16. Basow, DS (Ed). Gestational trophoblastic disease: Management of hydatidiform mole. Garner EIO. In: UpToDate [Textbook of Medicine]. Massachusetts Medical Society, Waltham Wolters Kluwer Publishers, Amsterdam, The Netherlands. 2010.
17. Molina, Vanessa Ventura; Ochoa, Minerva Lazos; Céspedes, Danny Soria. Coriocarcinoma gestacional. Una causa de hemorragia de tubo digestivo alto. Informe de un caso. *Revista Medica del Hospital General de Mexico*. 2007;70(3): 130
18. Belfort, Paulo; bueno, Luiz Gustavo; novaes, Carlos Eduardo and rezende, Jorge de. Doença trofoblástica gestacional complicada por hemorragia**.** *Rev. Bras. Ginecol. Obstet.* [online]. 2004;26 (7):551-6. ISSN 0100-7203. http://dx.doi.org/10.1590/S0100-72032004000700007
19. Kolomeyevskaya NV; Tanyi JL; Coleman NM; Beasley AD; Miller HJ; Anderson ML. Balloon tamponade of hemorrhage after uterine curettage for gestational trophoblastic disease.*Obstet Gynecol; 113(2 Pt 2): 557-60, 2009 Feb.*
20. Keepanasseril A; Suri V; Prasad GR; Gupta V; Bagga R; Aggarwal N; Dhaliwal LK; Khandelwal N. Management of massive hemorrhage in patients with gestational trophoblastic neoplasia by angiographic embolization: a safer alternative. *J Reprod Med; 56(5-6): 235-40, 2011 May-Jun.*