



RESEARCH ARTICLE

MASSIVE PLURAL EFFUSION CAUSED BY HELLP SYNDROME COMPLICATED COMPLETE MOLAR PREGNANCY IN 16 WEEKS OF GESTATION, IN A 14 YEARS OLD WOMAN

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ABSTRACT

HELLP syndrome is one of the life-threatening situations in pregnancy and has a tendency to multi organ insufficiency in some particular cases. We discuss about a 14 years old woman with molar pregnancy who complicated by the syndrome in 16 weeks of gestation, 24 hours after evacuation of conception products. She was one of the missed molar pregnancies with management delays. After 24 hours of her evacuation, the patient developed severe plural effusion and returned to a normal condition after 7 days. It was an exceptional situation implies a high risk of mortality. Based on our knowledge, there are very rare reports as such HELLP syndrome complicates complete molar pregnancy in 16 weeks of gestation. Notably we will report massive plural effusion caused by the syndrome in molar pregnancy as a very scare manifestation of HELLP syndrome. Early diagnosis of molar pregnancies is very important. When HELLP syndrome complicated molar pregnancy, treatment is similar to non-trophoblastic pregnancy with only conservative closed management. Respiratory distress due to sudden onset massive pleural effusion is not as a common symptom of HELLP syndrome but our assertion is, it's possible and only conservative management may be sufficient for this situation too.

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INTRODUCTION

HELLP syndrome that is known by hemolysis, elevated liver enzyme level and a low platelet count usually occurs in the third trimester of pregnancy but in molar pregnancy can be arisen sooner than 20 weeks of gestation. Early HELLP syndrome in molar pregnancy is very rare (Joshi, 2010). Such fulminate presentation before 20 weeks has been described few up to now. Only 3 cases. Triploid karyotype was confirmed in all of them (Stefos, 2002; David, 2006 and Sebire, 2008). HELLP syndrome is extremely uncommon prior to 24 weeks. In early presentation of HELLP syndrome the risk of considerable maternal and prenatal morbidity and mortality is high (Guillaume Vogin, 2016). The most common form of gestational trophoblastic disease is complete hidatiform molar pregnancy (David, 2006).

Immediate suction curettage is mandatory as a routine treatment option. Herein we have present an unusual molar pregnancy which had not been diagnosed until 16 weeks of gestation within 24 hours after evacuation her condition got worse significantly with the development of severe HELLP syndrome. To days based on our knowledge, there are extremely rare reports which HELLP syndrome complicates complete molar pregnancy in 16 weeks of gestation.

Case Presentation

A 14 years old woman referred to our urgency ward of Emam Khomeini Hospital of Tehran University of Medical Sciences, Tehran, Iran with vaginal bleeding, in 16 weeks of pregnancy. her past medical history was unremarkable and she got married 6 months ago. Her gestational age by last menstrual period date was 16 weeks. On admission, she was in stable situation but seemed to be little bit pale. She complained of spotting from 1 day ago and did not have any significant symptoms. Patient blood pressure was 100/60 mmHg, Temperature was

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38 oral degrees and her pulse rate was 85 per min. BMI was 16/6. She had fundal height in size 24 cm and closed cervix as such long and mid. Deep tendon reflexes were normal and there was no peripheral edema. Her physical examination was otherwise normal. She had not performed any sonography until that time. In the first transvaginal sonography which was performed on her hospitalization time, there were heterogeneous cystic spaces confirmed as complete molar pregnancy and bilateral theca lutein ovarian cysts. Her laboratory tests were as below:

Platelet count (plt):143000(150000-400000), Hematocrit (htc):25(37-48), Hemoglobin (Hg):8/2(11-14), Creatinine(Cr):0/6(0/5-0/9) mg/dl, Aspartate aminotransferase activity (AST): 25(<20 U/L), Alanine aminotransferase activity (ALT): 30(<20 U/L), Bhcg level was 48,000,000. We had not found any remarkable cause for her low grade fever. Due to immediate suction curettage which was necessary, we had not performed more diagnostic evaluation for her low grade fever and we prescribed combination Clindamycin 600 mg plus Gentamycin 80 mg every 8 hours' infusion after sending several blood and urine cultures. 2 units packed red blood cell was infused before operation. Our concept was perhaps the old clots in the large uterus as a result of hemorrhage from product of molar conception could be the reason of low grade fever. We performed suction curettage after ripening of the cervix by misoprostol 400 microgram vaginally. The uterus was very large about 24 cm, so we started Syntocinon 30 unit in 500 cc normal saline at the beginning of the procedure. The amount of bleeding was very high, about 2500cc or may be more. Our patient BMI was 16/6 and she was very thin. So we started 3000cc ringer lactate crystalloids plus 3 units packed red blood cells plus 2 units Fresh Frozen.

Plasma (FFP) inter operation which was continued during recovery time after evacuation. Then she became stable but on the second day of her admission her general medical condition deteriorated and she became oliguric, hematuria was detected too. There were not any other significant symptoms on examination except icteric sclera and skin. Her blood pressure was normal and laboratory tests were as below:

PLT: 60000, Hg: 10 mg/ dl, Cr: 0/7mg/dl, her total Bilirubin (Bill):8, direct Bill: 4.8 Lactate

dehydrogenase (LDH):1100 u/l.AST:358 u/l, ALT: 324 u/l. Our differential diagnosis included the following: Sepsis, Thrombotic Thrombocytopenic Purpura (TTP), Systemic lupus Erythematosus (SLE), viral hepatitis, HELLP syndrome and Antiphospholipid syndrome. She did not have any poor sign of sever preeclampsia. Her mean blood pressure was 100/80 mm Hg during the monitoring. At this time her abdomen was distant and respiratory distress was developed. So abdominal sonography was done, there is not any abnormality just moderate free fluid was shown in abdominal cavity. We performed an Abdomino pelvic CT scan, to rule out rupture of uterus which was normal also. For the treatment of respiratory distress, we started oxygen therapy by mask under semi setting position and salbutamol inhalator. In the chest radiography, bilateral plural effusion was detected. Thorax CT scan also confirm it. The fluid was tapped under ultra sound guided for diagnostic laboratory tests. Result confirmed to be transudate effusion not exudative. There was no sign of trophoblastic embolism pattern on imaging. Fever was low grade and urine and blood culture were negative after 48 hours. So the

antibiotic infusion discontinued. Cr was normal and schizocyte count was lower than %1 in peripheral blood smear therefore the probably of TTP was less. Viral hepatic tests and lupus diagnostic markers and Antiphospholipid antibodies were also negative, and HELLP syndrome was the best diagnosis for her, lead to select conservative management. The Figure 1, 2 show the curve of laboratory test. After 48 hours' urine output came back to normal range 30cc/hour and was less hematuric. Platelet count which decreased to 50000 during 4 days, normalized after 3 days' riches to 95000. As showing in Figure the liver enzyme level and billirubin came back to normal ranges also within 7 days. And the pathological examination confirmed complete molar pregnancy. We continued to check BHcg weekly. The Figure 3 shows the curve of BHcg titrage.8 weeks after evacuation serum level BHcg came back to negative range. Consent form was filled by the patient for reporting her presentation.

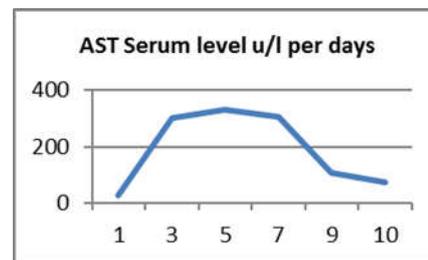


Figure 1. Amino aspartate transferase (AST) serum level per days was showed which declined to base after 7 days

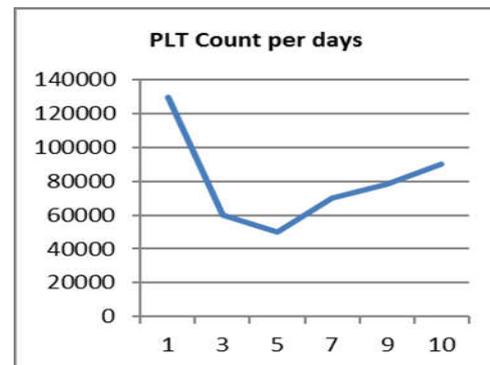


Figure 2. The platelets count per days was showed which raised to base after 10 days

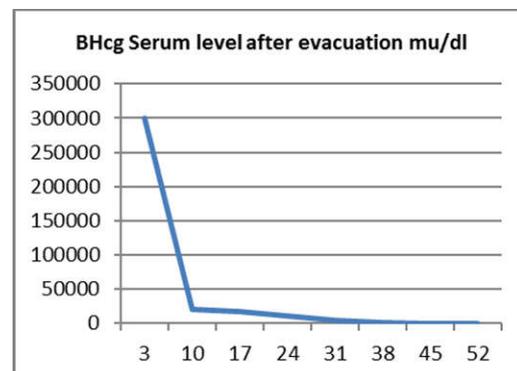


Figure 3. The level of Bhcg after evacuation8 weeks after evacuation serum level BHcg returned to negative range under weekly and then monthly surveillances

DISCUSSION

HELLP syndrome is one of the life threatening complications of pregnancy that occurs in third trimester of pregnancy (Joshi,

2010). In molar pregnancy because of abnormal placental function early preeclampsia may be arised, but up to now concomitant HELLP syndrome and molar pregnancy has been reported rarely. To days according to available ultrasound imaging, early diagnosis of molar pregnancy is possible before complication. (Guillaume Vogin *et al.*, 2016) Even a patient does not have any possibility to prenatal care, vaginal bleeding or hyper emesis gravid arum which are the most symptoms of complete molar pregnancy, causes the patient to go to hospital and the Diagnosis dose not delay (Steigrad, 2003). But our patient was one of the illegal immigrants from Afghanistan, who was living suburbs, was not under insurance and her condition causes to poor prenatal care. In addition to this, she did not have any chance to be diagnosed since she had no symptoms until 16 weeks of pregnancy. Even she was glad to her abdomen enlargement, as a normal pregnancy until she was symptomatic in 16 weeks of gestation. To know the cause of fever, we reviewed all of her body system. Neither universal examination nor laboratory tests could help us to know the reason of fever; pathological analysis had not shown any certification for inflammation. Only a few poly morph nuclear cells (PMN) has been appeared which was unspecific. Early HELLP syndrome in molar pregnancy is very rare (David M. Sherer *et al.*, 2006). Such fulminate presentation before 20 weeks has been described few up to now. Only 3 cases which were triploid (Stefos *et al.*, 2002; David M. Sherer *et al.*, 2006; Sebire and Seckl, 2008). Therefore, it is necessary to early diagnosis of pregnancy with ultrasound to avoid missed complications of molar pregnancy and hospitalization with closed observation is recommended in case of gestational trophoblastic disease. On early presentation of HELLP syndrome the risk of considerable maternal morbidity and mortality is high (Joshi *et al.*, 2010). Immediate suction curettage is mandatory as a rutting treatment option. The cause of molar pregnancy is unclear. For teenagers there is 1/3-fold increase compared with women over 40 years of age 10 fold increased risk. Other factors which are responsible for increasing the risk of hidatiform molar pregnancy are, rice, gravity, diet and contraception (Steigrad, 2003; Fawaz Khaza'leh and Kareem Haloub, 2015). Atypical hyper plastic trophoblasts and hydropic villi can cause ischemia in the placental arteries lead to preeclampsia or HELLP syndrome. (Camille E. Powe *et al.*, 2011) Placental ischemia and wide spread maternal endothelial dysfunction is the same condition which occurs in HELLP syndrome. Reports have confirmed that this endothelial dysfunction is widely causes inhibition of the endothelial growth factors vascular endothelial growth factor and placental growth factor which lead to life threatening complications such as preeclampsia or HELLP syndrome (Camille E. Powe *et al.*, 2011) Except laboratory characteristic, oliguria, hematuria, ascites and icterus are common manifestation in HELLP syndrome and such an intense plural effusion is unusual. although trophoblastic pulmonary emboli or pulmonary edema are more common differential diagnosis of sudden onset respiratory distress but in chest radiography and spiral tomography scan imaging both of them were ruled out and the accumulation of fluid in the third space as the disease entity found to be the fundamental etiology.

Conclusion

It is important to early diagnosis of molar pregnancy to avoid such complications. When HELLP syndrome complicated molar pregnancy, treatment is as the same as in non-trophoblastic pregnancy with only Conservative closed management.

Respiratory distress due to massive pleural effusion is not as a common symptom of HELLP syndrome but our assertion is it's possible and only closed monitoring as well as conservative management may be sufficient for this situation too.

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Conflict of interest

All authors have not any conflict of interest to declare.

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