CEOG Clinical and Experimental Obstetrics & Gynecology

Letters to the Editor

The circumvallate placenta as a possible culprit of fetomaternal hemorrhage

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Dear Editor,

We read with interest the article, "Idiopathic massive fetomaternal hemorrhage (FMH) in the third trimester of pregnancy causing neonatal death" by Peng *et al.* [1]. They concluded: "A pregnant woman at late pregnancy with complaints of unspecific signs such as decreased fetal movement (DFM) should arouse a high index of clinical suspicion of idiopathic FMH". Another recent study demonstrated that 44% of stillbirth cases due to FMH showed DFM [2]. It also showed that in two-thirds of stillbirth due to FMH, risk factors of FMH remained unidentified [2], which Peng *et al.* referred to as "idiopathic FMH". Many FMH are deemed to be unpreventable. Thus, identifying conditions underlying FMH may increase obstetricians' level of concern for FMH occurrence.

Recently, a new concept of FMH was reported [3]: FMH was more likely to occur in placentas with "pathological permeability", in which fetal blood is more likely to transfer to maternal blood. This is reasonable. An "abnormal trophoblastic invasion" underlies this pathological permeability: preeclampsia, placental abruption, and placenta previa were listed [3]. Briefly describing a case with DFM, we suggest that circumvallate placenta, an "abnormal trophoblast invasion"-related condition, may be associated with FMH.

At the 32nd week, a 28-year old primiparous woman complained of DFM, showing sinusoidal pattern on cardiotocogram (Figure 1a) and high middle-cerebral-artery peak systolic velocity (2.32 MoM). Cesarean section yielded a 1,486-gram infant (hemoglobin 2.0 g/dL; Apgar score 4/5 [1/5 minutes]) with maternal hemoglobin F 4.3%, confirming the diagnosis of FMH. Placental hematoma was present at the periphery of the circumvallate plate (Figure 1b). With transfusion, the infant was healthy without sequelae. An abnormally shallow trophoblastic invasion is considered to underlie circumvallate placenta [4] similar to preeclamptic placenta, and may increase the permeability of the placental barrier, causing FMH. In addition, circumvallate placenta frequently accompanies "peripheral hematoma" [4, 5] as observed here. Pregnant women with circumvallate placenta sometimes bleed, which was shown to be of mixed maternal and fetal origin [4]. Hematoma, destroying the placental barrier there, at least partly, also may contribute to FMH.

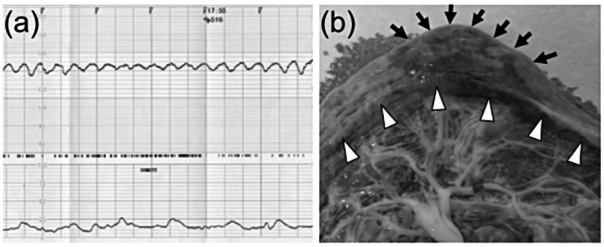

Figure 1. — Cardiotocographic finding (a) and placental gross finding (b).
(a): Sinusoidal pattern on cardiotocogram at the 32nd week.
(b): Arrowheads indicate circumvallate plate and arrows indicate peripheral hematoma.

Circumvallate placenta was reported to be associated with various disorders including preterm delivery, placental abruption, or fetal growth restriction [4, 5]. To our knowledge, circumvallate placenta was not associated with FMH. This may be due to: 1) circumvallate placenta may be frequently unrecognized, as suggested by the wide range of its reported incidences (0.62-18.3%) [4], or 2) even if recognized, significant proportion of obstetricians may consider the circumvallate placenta as non-pathological, and thus consider concomitant occurrence of circumvallate placenta and FMH as insignificant, and therefore unreported. If we focus on severe circumvallate placenta, severe to the

Revised manuscript accepted for publication December 1, 2016

Clin. Exp. Obstet. Gynecol. - ISSN: 0390-6663
XLIV, n. 5, 2017
doi: 10.12891/ceog4022.2017

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extent that it causes some perinatal morbidities, FMH may be present in a significant proportion.

Six decades ago, when FMH was unrecognized, Mitchell *et al.* [6] stated that circumvallate placenta may be associated with "neonatal anemia". A direct evidence was lacking whether permeability actually increased in this patient. However, since abnormal placental trophoblastic invasion underlies FMH, and circumvallate placenta has this placental pathology, we assume that circumvallate placenta, at least partly, may have contributed to FMH, and not a mere coincidence.

Peng *et al.* [1] concluded that obstetricians should be suspicious of "idiopathic FMH" at DFM. Circumvallate placenta may be hidden among "idiopathic FMH". Cautious antenatal ultrasound may reveal circumvallate placenta [5]. Thus, a pregnant woman with antenatally diagnosed circumvallate placenta may be asked to pay much attention to fetal movement. Further study is needed to confirm our suggestion.

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Reply by Peng *et al.*

Most cases of FMH are idiopathic or "silent" and they may remain undiagnosed until delivery. The real reason is unknown but it was speculated that fetal placental blood vessels have higher blood pressure than the intervillous space; if there is disruption of the maternal-fetal barrier, hemorrhage will occur from the fetus to the maternal circulation. It is meaningful to refer that FMH is more likely to occur in placentas with "pathological permeability", an "abnormal trophoblastic invasion" underlies this process, in which fetal blood is more likely to transfer to maternal blood. We cannot agree more that it is reasonable. The risk factors causing FMH include blood type incompatibility, abdominal trauma, amniocentesis, the external cephalic inversion, hypertensive disorders such as preeclampsia, placental abruption or placental, and umbilical cord abnormalities, such as choriocarcinoma and chorioangiomas. The author presented one rare FMH case with circumvallate placenta, suggesting that circumvallate placenta, an "abnormal trophoblast invasion"-related condition, may be associated with FMH. It is reasonable and remarkable. We also speculated that "potential placental abnormalities" might be the main cause of idiopathic FMH. Thus, obstetricians should be suggested to pay much attention to a pregnant woman with any placental pathology.

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