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Research Letter

Unusual maternal hemoglobin elevation before delivery as a rare presentation of massive fetomaternal hemorrhage



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Dear Editor,

We present a case of unusual maternal hemoglobin elevation before delivery as a rare presentation of massive fetomaternal hemorrhage (FMH). Ethical approval was obtained from National Taiwan University Hospital, code: 201404050RINA.

A 36-year-old woman, gravida 3, para 1, ectopic 1, without other systemic disease, was admitted to our hospital for the first time at 37 and 4/7 completed gestational weeks. The blood group of both the patient and her husband was O type and Rh positive. She underwent regular prenatal examination, and neither any major fetal anomaly nor any medical complications of the mother was found. No relevant family history was noted. She was admitted due to regular labor pain every 3-5 minutes. Uterine contractions every 3 minutes with 80-100% amplitude were detected by cardiotocogram. Fetal heart beat tracing was Category I. Bishop score was 6/10 (cervix: posterior presentation, intermediate consistency, 60% effacement, 2 cm dilatation, and Stage 0). However, uterine contractions weakened and ceased 1 day after admission. Induction with oxytocin (5 IU) in Ringer solution (500 mL) at a rate of 8 mL/ hour with titration was prescribed. Augmentation of labor failed, so she was discharged 3 days later.

Five days after the first discharge, decreased fetal movement was noticed for 1 day. Fetal cardiotocogram showed nonreassuring fetal tracing (baseline: 170 beats/minute, minimal variability, and variable deceleration >50% contraction), so she was admitted to our hospital for continuous monitoring. Complete blood count test was performed routinely in our hospital. Maternal hemoglobin level was elevated from 14.0 g/dL to 15.3 g/dL, compared with that 5 days

ago. No fever, no chills, and no other transfusion reaction symptom were noted. No trauma was found or no invasive procedure was performed between these two admissions. Other laboratory tests are shown in Table 1.

Fetal heartbeat deceleration to an undetectable level was noted 6 hours after admission, and emergent cesarean section was performed. One mature female baby weighing 3250 g was delivered by vertex extraction at 38 and 2/7 completed gestational weeks. The first and fifth Apgar scores were 0 to 0.

Neonatal resuscitation was performed immediately and return of spontaneous circulation at the 50th minute. On examination, the neonate was very pale without obvious external bleeder. A neonatal hemoglobin level of 3.6 g/dL, an arterial cord pH of 7.07, and a base deficit of 23.7 mmol/L were reported. Other laboratory test results are shown in Table 2. The Kleihauer—Betke test was performed and it showed 159 fetal red blood cells in 2000 cells.

Massive FMH was diagnosed, and the neonate was admitted to the neonatal intensive care unit. Component therapy was given immediately. Brain magnetic resonance imaging showed extensive intraventricular hemorrhage and diffuse cerebral ischemic changes (Figure 1). Electroencephalogram reported low-amplitude and no epileptic spikes. Severe hypoxic-ischemic encephalopathy was diagnosed, and hypothermia therapy was given. After head-cooling therapy and supportive care at the neonatal intensive care unit for 1 month, the baby was discharged with stable vital signs and severe

Table 1 Maternal laboratory data.

Variable	First admission	Second admission (5 d later)	Reference range
Erythrocyte	4,850,000	5,170,000	3,780,000
count (/mm³)			-4,990,000
Hemoglobin (g/dL)	14.0	15.3	10.80-14.90
Hematocrit (%)	39.8	43.6	35.60-45.40
Mean corpuscular volume (μm³)	82.1	84.3	80-100
Platelet count (/mm ³)	255,000	225,000	150,000-361,000
White-cell count (/mm³)	11,030	10,760	3540-9060

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Table 2 Neonatal laboratory data.

Variable	Initially	Reference range (age adjusted)
Erythrocyte count (/mm³)	950,000	4,100,000-5,740,000
Hemoglobin (g/dL)	3.6	12-20
Hematocrit (%)	11.4	36-60
Mean corpuscular volume (μm³)	120.0	91.3-120
Reticulocyte (%)	6.95	2-5.4
Reticulocyte number (/mm³)	196,700	22,400-82,900
Platelet count (/mm³)	148,000	144,000-450,000
White-cell count (/mm ³)	52,740	4940-27,480
рН	7.074	7.35-7.45
pCO2 (mmHg)	22.4	35-45
pO2 (mmHg)	33.9	83-108
HCO ₃ (mmol/L)	6.6	21-28
Base excess (mmol/L)	-23.7	−2 to 3
Direct: Coombs test	Negative	Negative
Indirect: Coombs test	Negative	Negative

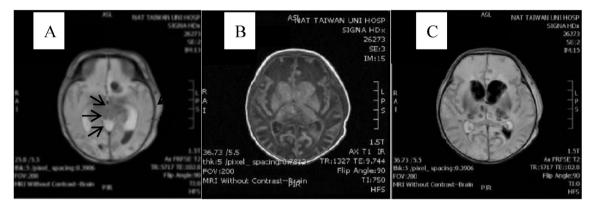


Figure 1. Brain MRI of the neonate. (A) Intraventricular hemorrhage within both frontal horns and occipital horns found at T2 phase (arrow). Hypoxic-ischemic injury to gray matter results in (B) the characteristic T1 hyperintensity () and (C) variable T2 intensity. Injury to white matter results in the T1 hypointensity and T2 hyperintensity due to ischemia-induced edema.

neurological impairment. The maternal postpartum course was smooth and she was discharged 5 days after cesarean section.

FMH refers to the entry of fetal blood into the maternal circulation before or during delivery. Fetal blood likely enters the maternal circulation during all pregnancies [1], and the volume lost is typically small, usually <15 mL [2]. An earlier summary of cases of hemorrhage of >30 mL, as detected by the Kleihauer—Betke test, reported a similarly high risk of perinatal morbidity and mortality [2]. Massive FMH was defined as more than 80 mL fetal blood loss because neonatal anemia appeared at this level [3]. The incidence of FMH was found to be 22—182 per 1000 anemic neonates [4]. Perinatal mortality due to massive FMH occurs in about one in 1000 deliveries [5]. It was reported that nine in 18 neonates diagnosed with massive FMH and a hemoglobin level of below 7.5 g/dL exhibited poor outcomes, such as cerebral palsy, mental retardation, attention deficit/hyperactivity disorder, and epilepsy [6].

Antenatal diagnosis requires a high index of suspicion, as the presenting signs are often nonspecific [7]. In previous research, the presenting signs and symptoms were described as follows: neonatal anemia: 35.0%; decreased or absent fetal movement: 26.7%; stillbirth: 12.5%; hydrops fetalis: 7.5%; nonreassuring fetal tracing: 6.7%; and other signs and symptoms: intrauterine growth restriction, sinusoidal fetal tracing, fetal atrial fibrillation, and maternal transfusion reaction [8]. FMH could be presented by chronic fetal anemia, acute fetal anemia, or both acute and chronic clinical features [9].

In our case, besides decreased fetal movement and neonatal anemia, one unique sign of FMH was elevated maternal hemoglobin level. Several formulas for estimating fetal blood loss volume had been published [7]. Assuming an average maternal blood volume of 5000 mL, the estimated fetal blood volume lost ranges from 397 mL to 1520 mL, using different formulas [7]. In our case, a significant elevation of hemoglobin level from 14.0 g/dL to 15.3 g/dL in 5 days came into notice for the suspicion of massive FMH.

Erythrocytosis during pregnancy is uncommon and may be due to chronic hypoxia environment, cigarette smoking, polycythemia vera, or neoplasm [10,11]. In this case, unusual maternal hemoglobin elevation was a rare presentation of massive FMH. In conclusion, in case of pregnant woman with unexplained elevation of maternal hemoglobin, examination of FMH may be considered.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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