

A low birth weight infant with unexpected hepatic artery pseudoaneurysm rupture: A case report and review of the literature

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Summary

Here, we describe an unusual case of a preterm infant presenting with massive bleeding as a result of sudden hepatic artery pseudoaneurysm rupture during exploratory laparotomy. During surgery, this low birth weight infant (1.8 kg) was administered chest compressions and a blood transfusion. However, he repeatedly experienced massive bleeding episodes and thus received a massive transfusion. The infant shows unbelievable tolerance and tenacious vitality.

Key words: Cardiopulmonary resuscitation; Hemorrhage; Low birth weight infant; Massive blood transfusion.

Introduction

Management of low birth weight infants with severe or refractory abdominal bleeding due to surgery or trauma is extremely challenging [1, 2]. Such bleeding leads to anemia, hypovolemic shock, and even cardiac arrest. Meanwhile, massive blood transfusions may cause infection, volume overload cardiac decompensation (transfusion-associated cardiac overload), acute lung injury, immunosuppression and metabolic derangement [3], coagulopathy, electrolyte disturbance, and hypothermia during the rescue process. Pediatric data on the successful management of massive transfusions (MTs) are limited [4] and the optimal transfusion approach is currently unknown. This leads to practice variability among institutions, depending on resource availability and each patient's unique situation. Here, we report an unusual case of a preterm infant with massive bleeding as a result of sudden hepatic artery pseudoaneurysm rupture during exploratory laparotomy.

Case report

The case is a 1.8 kg preterm infant boy who was born outside of the hospital setting by vaginal delivery at 36 weeks to a multiparous woman. The infant otherwise appeared normal. His Apgar Score was 9. Twenty hours after delivery, vomiting of gastric content was noted and the infant was promptly transferred to hospital. Barium meal examination of the upper digestive tract suggested duodenal ileus. Given that the infant's situation was getting progressively worse, it was necessary to identify any abdominal abnormality prior to proceeding with exploratory laparotomy.

Peripheral intravenous access (basilic vein, PICC) was

implemented 30 minutes before anesthesia induction. The patient was not pre-medicated before the surgery. In the operating room, electrocardiography, invasive arterial pressure, nasopharyngeal temperature, and pulse oximetry were monitored using routine methods (HR 140/min, SpO₂ 98%, nasopharyngeal temperature 36.7 °C, invasive arterial pressure 60/40 mmHg). Anesthesia was induced by intravenous administration of midazolam (0.1 mg/kg), penehyclidine hydrochloride (0.01 mg/kg), sufentanil (0.001 mg/kg), and cisatracurium (0.1 mg/kg). After oral tracheal intubation (ID 3.0), mechanical ventilation was established; the inspired oxygen concentration was 30-50%, tidal volume was 10 mL/kg, respiratory rate was 28 breaths/min, and the inspiration/expiration ratio was 1 : 2. Catheterization of the right internal jugular vein failed. Thus, the right radial artery was catheterized to monitor arterial blood pressure. After the surgeon opened the abdomen, annular pancreas and oppression of the duodenal descending segment were observed. Thus, intestinal resection and anastomosis were performed. During the surgery, the disorder of electrolytes (low potassium and low calcium) and dehydration were continuously corrected to maintain the body's metabolism balance and avoid abnormal bleeding. When the operation was almost complete, a hepatic artery pseudoaneurysm rupture led to massive and unexpected bleeding. In response, a massive amount of electrolyte solution was first injected through the basilica vein. Then, 1 U of red blood cells (RBCs) and 200 mL of fresh frozen plasma were rapidly injected. Because the bleeding was difficult to control, the infant suffered from hypovolemic shock. His HR dropped from 140/min to 100/min and his blood pres-

Table 1. — *The blood gas analysis results during the operation*

Blood gas analysis	Before operation (15:44)	20 min after operation (16:43)	70 min after operation (17:32)	After First CPR (17:39)	After Second CPR (18:23)	In NICU (19:02)
pH	7.365	7.378	7.202	7.173	6.84	6.976
pCO ₂ (mmHg)	30	32	47.5	58.4	89.3	58.1
pO ₂ (mmHg)	269	183	124	85	29.5	186
Hb(g/L)	124	112	84	79	66	101
Hct	38	34.4	25.8	24.1	20.1	30.9
K ⁺ (mmol/L)	3.2	3.4	2.9	3.3	5.2	3.4
Na ⁺ (mmol/L)	143	144	149	148	148	146
Ca ²⁺ (mmol/L)	0.73	0.92	0.93	0.74	1.21	1.4
Cl ⁻ (mmol/L)	110	112	113	108	103	103
Glu (mmol/L)	11.1	6.5	4.6	3.9	14.1	13.5
Lac (mmol/L)	6.6	5	3.5	2.8	5.2	4.9
ABE (mmol/L)	-7.4	-5.8	-8.5	-6.2	-16.2	-16.7
SBE (mmol/L)	-8.3	-6.3	-9.4	-7	-18.7	-18.1
SBC	18.5	19.7	17.5	19.2	11.3	11.9
SAT	100	100	100	96.9	39.4	99.1
Anion gap	16.3	13.4	18.1	17.9	29.7	30
Serum osmolality (mOsm)	297.7	294.5	303.4	299.2	310.8	305.9
p50	27.1	26.84	32.65	24.34	34.64	41.21
pO ₂ (A-a)	—	57.5	99.3	126.5	148.2	26.7
F Shunt	2.7	2.5	4.7	11.8	53.2	1.9

pH, acidity of the blood; pCO₂, the partial pressure of carbon dioxide; pO₂, the partial pressure of oxygen; Hb, hemoglobin; Hct, hematocrit; K⁺, potassium ion; Na⁺, sodium; Ca²⁺, calcium ion; Cl⁻, chloridion; Glu, glucose; Lac, lactate; ABE, actual base excess; SBE, standard base excess; SBC, standard HCO₃⁻; SAT, saturation; pO₂(A-a), alveolo-arterial oxygen gradient; p50, the partial pressure of oxygen at half saturation; F shunt, intrapulmonary shunt.

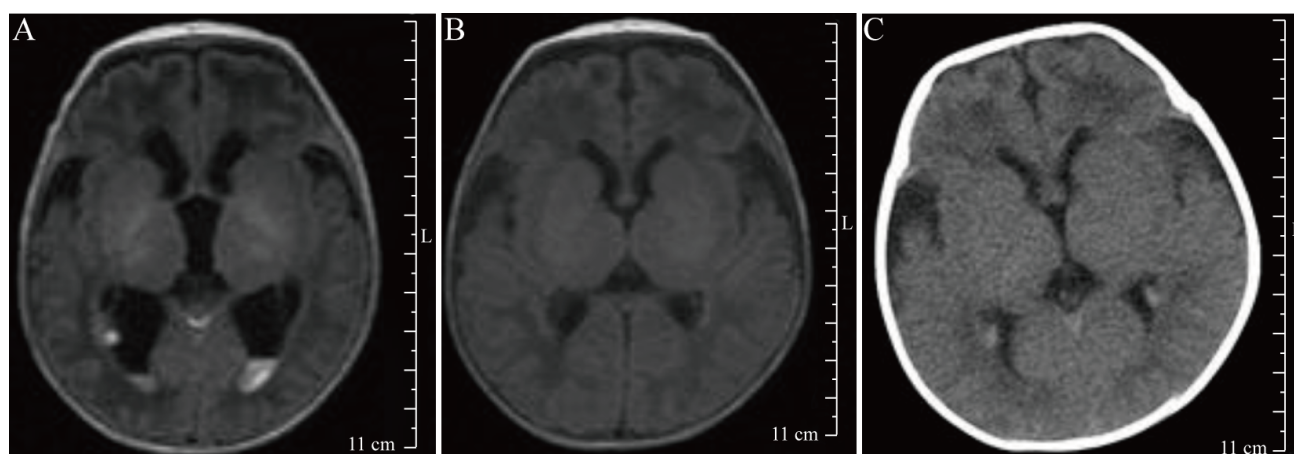


Figure 1. — Brain MRI and CT scan. A: MRI: Obvious cerebral hemorrhage and edema one day after CPR; B: MRI: Cerebral hemorrhage and edema decreased 20 days after CPR; C: CT: Cerebral hemorrhage and edema decreased 28 days after CPR.

sure dropped from 62/40 to 50/40. Thus, CPR was activated, including chest compressions (over 120/min) and epinephrine (1/10000 0.1 mL/kg). Following this, the patient's HR recovered to 130-140/min. However, the bleeding was too severe and the patient's HR kept dropping from 130/min to 50/min over a 5 min period. Thus, 1.5 U RBCs were again injected. At this time, methylprednisolone (10 mg/kg), phosphocreatine (0.5 g/kg), and ulinastatin (10000 U/kg) were also used. FiO₂ was increased to 100%. The

patient's HR recovered to 120/min after 30 min of CPR with continual correction of electrolyte and acid-base balance disorders according to the blood gas analysis results. However, 25 min later, the patient's HR again dropped from 120/min to 100/min. Thus, CPR was repeated and 1 U of RBCs, 200 mL of fresh frozen plasma, and 2 U of cryoprecipitate were injected. Five minutes later, the patient's HR recovered to 110/min. Because the bleeding was difficult to stop completely, the abdomen was closed with pressure.

Table 2. — The blood gas analysis results in neonate intensive care unit

Blood gas analysis	19:51	23:35	2:30
PH	6.96	7.24	7.4
pCO ₂ (mmHg)	50	36	30
pO ₂ (mmHg)	280	108	96
Hb(g/L)	105	119	133
Hct	31	35	39
K ⁺ (mmol/L)	3.9	3.2	2.3
Na ⁺ (mmol/L)	143	144	148
Ca ²⁺ (mmol/L)	0.55	1.55	0.9
Glu(mmol/L)	11.5	18.1	13.2
Lac(mmol/L)	6.5	11.3	11.3
ABE(mmol/L)	-18.4	-12	-6.2
SBE(mmol/L)	-18.2	-11.1	-5
SBC	10.8	16.3	21
HCO ₃ ⁻	13.5	15.4	18.6
TCO ₂	15.3	16.5	19.5
SAT	100	97	97

pH, acidity of the blood; pCO₂, the partial pressure of carbon dioxide; pO₂, the partial pressure of oxygen; Hb, hemoglobin; Hct, hematocrit; TCO₂, total carbon dioxide.

At this time, some blood was found in the endotrachea tube. Norepinephrine (0.2 mg) was injected into the trachea and the infant was sent to NICU for further treatment. The blood gas analysis results during the operation are shown in Table 1.

In NICU, 2 U of platelets and 5 g of albumin (20%; 25 mL) were transfused. During the operation (3 h and 50 mins in total), 800 mL of crystal solution, 3.5 U of RBCs (700 mL), 400 mL of fresh frozen plasma, and 2 U of cryoprecipitate were transfused. Most of the crystal solution and all of the blood products were transfused within 1.5 h. The urinary volume was 100 mL in total.

The infant recovered with spontaneous breathing and movement of the limbs on the second morning following surgery. Unfortunately, the infant's parents chose to forgo further treatment in NICU at noon of the second day because of economic problems. Subsequently, they transferred the child to a local hospital. The blood gas analysis results when the child was in the NICU are shown in Table 2.

One week later, the child had recovered to an extent, and was able to suck, cry, and produce the holding reflex; however, convulsions had been observed several times. Thus, he was again transferred to our hospital. The child was diagnosed with purulent meningitis after examination of cerebrospinal fluid (micro protein 1.61 g/L, glucose 1.96 mmol/L, Cl 121.1 mmol/L). MRI showed some hemorrhage in the bilateral ventricle (Figure 1). After treatment for almost one month, the child recovered and was discharged from hospital.

Discussion

Massive bleeding is life-threatening, especially in low birth weight neonates. The definition of massive transfusion (MT) in children is not well established and most definitions of MT in adults are not applicable to pediatric patients. Luban *et al.* suggested that pediatric MT should be defined as transfusion of > 50% of total blood volume (TBV) in 3 h, transfusion of >100% of TBV in 24 h, or transfusion support to replace ongoing blood loss of > 10% of TBV per min [1]. The child described here was 1.8 kg and the TBV was less than 200 mL. However, in 1.5 hours, he was transfused with 700 mL of RBC, 400 mL of fresh frozen plasma, 2 U of cryoprecipitate, and about 800 mL of crystal solution. Thus, this is a rare case of pediatric MT.

The child almost died with massive bleeding beyond our control; his heart rate was only maintained by chest compressions. Continuous and effective chest compressions to guarantee perfusion of the organs is the key to the success of CPR. Preoperative and postoperative myocardial enzymes, coagulation function, and routine blood examination results are shown in Table 3.

Transfusion-associated hyperkalemia is a recognized complication of massive blood transfusions [5]. However, this did not occur in our patient. The patient's highest potassium concentration was 5.2 mmol/L. The main reason for this is that we use a peripheral vessel not a central vein for the transfusion.

Continual increases in lactic acid levels are lethal [6]. However, if lactic acid levels drop quickly, there is no threat to life. Lactic acid is the resource of glucose through the lactate cycle and is necessary for brain energy. Thus, lactic acid is also beneficial to critically ill patients, even if only used as an indicator of illness severity.

Ethics Approval and Consent to Participate

All subjects gave their informed consent for inclusion before they participated in the study. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of Children's Hospital of Chongqing Medical University.

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Conflicts of Interest

The authors declare no conflicts of interest.

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Table 3. — Preoperative and postoperative myocardial enzyme, coalgulation function and blood routine examination results

Parameter	Preoperative	Postoperative	Reference value
Allergic troponin I (ug/L)	0.108↑	1.282↑	< 0.06
myoglobin(ug/L)	97.36↑	150.35↑	< 110
Creatine kinase MB Isoenzyme(ug/L)	7.09↑	2.2	0.21-5
B-type brain natriuretic peptide(pg/nL)		2866.73↑	< 100
Prothrombin time(seconds)	15.8	16.6	13.0-20.0
activated partial thromboplastin time APTT(seconds)	52.3	> 240	37.0-45.0
Fibrinogen(g/L)	2.01	1.5	1.2-2.25
Thrombin time(seconds)	16.1	41.7	14.4-22.2
D-dimer(mg/L)	2.36	1.48	< 0.55
Total bilirubin(μmol/L)	104.8↑	182.8↑	1-20.5
Conjugated bilirubin(μmol/L)	1	1	0-6.7
Unconjugated bilirubin(μmol/L)	103.7↑	181.8↑	0-19.5
Total protein(g/L)	44.3↓	36.1↓	60-83
Albumin(g/L)	31.1↓	24.3↓	37-52
Glutamic-pyruvic Transaminase(U/L)	21.1	21.1	0-50
Glutamic oxalacetic Transaminase(U/L)	76.7↑	34.2	0-55
LDH(U/L)	380↑	478.7↑	140-270
Creatinine(Umol/L)	91	67.9	15.4-90.4
BUN(mmol/L)	6.86	6.41	2.42-6.72
WBC($\times 10^9/L$)	4.71	3.26	15.0-20.0
PLT($\times 10^9/L$)	225	12	100-300
RBC($\times 10^{12}/L$)	2.53	3.67	4.8-6.2
Hb(g/L)	98	115	170-200
Average red blood cell volume(fL)	113.4	99.2	98.0-112.0
MCV(pg)	38.7	31.3	26-32
Average hemoglobin concentration(g/L)	341	316	320-360
HCT(%)	28.7	36.4	50-70

APTT, activated partial thromboplastin time; LDH, lactate dehydrogenase; BUN, blood urean nitrogen; WBC, white blood cell; PLT, platelet; RBC, red blood cell; Hb, hemoglobin; MCV, mean corpuscular volume; Hct, hematocrit.

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