CASE REPORT

Unresponsive primipara after rupture of membranes

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SUMMARY

Amniotic fluid embolism, also called anaphylactoid syndrome of pregnancy, is a rare but severe problem in obstetrics. It occurs in 8/100 000 births and the maternal mortality is up to 90%. We report the case of a patient with amniotic fluid embolism who was transferred to our hospital. The initial presentation was an unresponsive patient after spontaneous rupture of the membranes. The massive hypotension and coagulopathy as well as fetal bradycardia of 60 bpm led, after stabilisation of the mother, to an emergency caesarean section. The neonate expired hours later, despite neonatological intensive care. During the operation, we had to deal with massive bleeding due to the coagulopathy. Through interdisciplinary teamwork including Bakri postpartum balloon insertion through the obstetrics team, uterine artery embolism by the interventional radiologists and transfusion of blood products, the maternal life was saved and the patient was discharged 9 days after admission.

BACKGROUND

Amniotic fluid embolism (AFE) is a well-known but fortunately rare occurrence for obstetricians. Therefore, most practitioners will never see a patient with this severe diagnosis. In its sudden and life-threatening presentation, it is of utmost important to recognise this problem immediately and correctly deal with it in order to save the patient's life.

CASE PRESENTATION

A 29-year-old primipara at 38 weeks and 5 days gestation was admitted to the emergency room of the University Hospital of Basel, Switzerland, by ambulance, 1 h after spontaneous rupture of membranes, followed by syncope at home. She had delivered 5 years before by ventouse and had an unremarkable pregnancy. She was otherwise healthy and had not been taking any medications. She had no known allergies.

On admission, the patient was unresponsive, with a blood pressure of 74/45 mm Hg, a heart rate of 120 bpm and oxygen saturation of 88% on room air. The fetus was in a cephalic position, and at first ultrasound examination the heart rate was >100 bpm. There was no free liquid visible in the abdomen.

At examination, there was no vaginal bleeding and no uterine contractions were palpable.

INVESTIGATIONS

Blood samples were sent to the laboratory for analysis and patient stabilisation with two large-bore venous catheters and volume therapy was started. Fifteen minutes after admission, fetal heart rate

dropped to 60 bpm and an emergency caesarean section was performed.

The first blood results showed a coagulation profile with a haemoglobin value of 92 mg/L, thrombocytes of $135 \times 10^9/L$, international normalised ratio >12, activated partial thromboplastin time >180 s and fibrinogen <0.3 g/L.

DIFFERENTIAL DIAGNOSIS

At admission, we considered the differential diagnosis for syncope in pregnancy. As pre-eclampsia/eclampsia may cause loss of consciousness and is a fairly common problem in pregnancy, we thought of this first and prepared a magnesium sulfate infusion as soon as we were notified of this patient. This diagnosis was discarded after her arrival in the emergency room and hypotension with normal liver enzymes was found. Hypotension as a consequence of massive peripartum haemorrhage was another idea, but neither intra-abdominal fluid (eg, due to uterine rupture) nor severe vaginal bleeding was identified.

Massive placental abruption is also known for being associated with acute severe coagulopathy. However, ultrasound examination did not show any sign of abruption.

In the patient's history, we found no suggestions pointing towards pulmonary embolism, anaphylactic shock, myocardial infarction, aspiration or sepsis.

Therefore, our suspected diagnosis for this patient presenting with spontaneous rupture of the membranes with acute hypotension, acute hypoxia and coagulopathy, was AFE, despite its rare occurrence.

TREATMENT

With the suspected diagnosis of AFE, we decided to perform the emergency caesarean section. The patient underwent general anaesthesia with thiopental, succinylcholine and fentanyl, and caesarean section was performed 40 min after admission to our hospital.

Following uterine incision, the amniotic fluid was noted to be bloody due to the disseminated intravascular coagulopathy. A non-reactive, floppy female infant was delivered. The weight of the neonate was 3030 g, with an APGAR score of 0/0/0 and umbilical arterial pH of 6.85. Intensive care for the baby was provided by the on-site neonatology team.

The placenta was manually removed. For enhanced uterine contraction, an infusion with sulprostone was started. A Bakri postpartum balloon was inserted into the uterus to try to lessen the severe bleeding and the abdominal wall was closed

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as per standard procedure after insertion of an intra-abdominal Blake drain.

The anaesthesiological team corrected the coagulopathy and massive blood loss, by aggressive blood and blood-component replacement with 13 erythrocyte units, 4×200 mL fresh frozen plasma, 2000 E factor VIII/vWF (Haemate), 2500 E Prothromplex, 2 g calcium gluconate, 2000 E Kybernin, 4×2 g fibrinogen, 1 unit of thrombocytes and 2 g Cyklokapron. A central venous catheter was placed in the internal jugular vein as well as a catheter in the femoral artery.

Owing to uncontrollable bleeding, massive blood loss of approximately 5000 mL, a uterine artery embolisation was performed. After successful embolisation and cessation of bleeding, the patient was transferred to the intensive care unit. We removed the Bakri balloon 24 h later and transferred the patient to the postpartum ward in stable condition. The Blake drain was removed on postpartum day 2. The nadir of the haemoglobin levels was 46 g/L.

OUTCOME AND FOLLOW-UP

Despite maximal neonatal intensive care unit support, the baby's life could not be saved. All resuscitative and life-preserving efforts were stopped 2.5 h after delivery and the baby died in the father's arms.

On the first day, the patient showed transient global amnesia, which was reversible. In the following days, she did not show any neurological deficits. Echocardiography was performed and showed a dilated right ventricle, highly suggestive of an embolic event. CT did not demonstrate a pulmonary embolism. Nine days after admission, the patient was discharged from the hospital.

Four weeks after discharge, another transthoracic echocardiography was performed, and showed normal cardiac function with a normally sized right ventricle.

Histological examination of the placenta did not show any suspicion for placental abruption.

DISCUSSION

AFE is a rare, unpredictable and life-threatening complication in obstetrics. First described by Meyer¹ in 1926, Steiner and Lushbaugh² established it as a clinical entity in 1941 with a series of 32 cases and named it such based on findings from autopsies. The name was based on the hypothetical mechanism that amniotic fluid is forced into the maternal circulation, thus triggering an obstruction of pulmonary arterial blood flow leading to hypoxia, right heart failure and death.

In the 1980s, another pathophysiological explanation, which involved an abnormal maternal response to fetal tissue in maternal circulation, was established.³ Basically, there is an abnormal inflammatory response with activation of proinflammatory mediator systems, known as a systemic inflammatory response syndrome. This is the reason why some authors call the syndrome 'anaphylactic syndrome of pregnancy',⁴ although one author of many papers on AFE states that "amniotic fluid embolism seems too deeply embedded in the language of medicine to be changed despite the availability of more precisely descriptive terms".^{3,5}

Diagnosis is based on clinical observations. Symptoms associated with AFE are hypotension, dyspnoea, cyanosis, fetal heart rate abnormalities, loss of consciousness, cardiac arrest, uterine bleeding and uterine atony. There are also many cases in which the components of the triad hypoxia, hypotension and coagulopathy, are absent.⁶ The biggest problem in our case was the coagulopathy and hypotension, whereas dyspnoea and cyanosis

were not significant. To support our suspected diagnosis of AFE, echocardiography on the day of admission and 4 weeks later demonstrated a dilated right ventricle, and CT of the chest showed that the ventricular dilation was not due to a pulmonary embolism.

The other potential cause of severe coagulopathy, massive placental abruption, was found neither during caesarean section nor in the histopathological examination of the placenta.

Rath *et al*⁷ performed a PubMed literature review in 2014 in which they found that the incidence of AFE is about 2–8/100 000 births, depending on the country and on diagnostic criteria. Other publications showed a rate of about 1/40 000, but as the prospective risk of AFE is very low, determination of the exact incidence is unlikely.³ In spite of the rare occurrence, AFE is a leading cause of maternal mortality, with 5–15%.⁸

Prognosis and outcome are dependent on rapid intervention. Hypoxia and dyspnoea are treated with oxygen administration and, if necessary, with intubation. Coagulopathy and haemorrhage need aggressive blood and component replacement. If the fetus is still in utero, prompt delivery is indicated.

For counselling regarding subsequent pregnancies, it is worth knowing that a total of 12 cases of successful pregnancy after survival from AFE have been reported in the literature, while no case of recurrence of confirmed AFE has been seen.⁹ Therefore, we think that our patient may not be at higher risk for another AFE in a following pregnancy.

Learning points

- ▶ Amniotic fluid embolism (AFE) is a rare complication during labour or within 30 min postpartum, during caesarean section or with abortion.
- ▶ AFE is an anaphylactic syndrome of pregnancy.
- ▶ AFE is a life-threatening clinical diagnosis of exclusion when other reasons for the symptoms can be found.
- ▶ No specific therapy is available. The aim is correction of hypoxaemia, hypotension and coagulopathy by intubation, volume therapy, blood and blood component replacement, and delivery of the baby.
- ▶ The maternal mortality is high. The fetal outcome depends on time delay until delivery can be performed.

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