J Anesth (1995) 9:370–373

Journal of
Anesthesia

A case of pulmonary arteriovenous fistula in which venous air embolism during cesarean section may have caused postoperative subendocardial infarction

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Key words: Cesarean section, Venous air embolism, Pulmonary arteriovenous fistula

Introduction

A high incidence of venous air embolism (VAE) has been reported during cesarean section, but serious cases of this complication are comparatively few [1–4]. In the present report, we describe a case of pulmonary arteriovenous fistula complicated by possible postoperative subendocardial infarction due to VAE during cesarean section.

Case report

The patient was a 30-year-old woman in her 38th week of pregnancy, who was 156 cm tall and weighed 58 kg. She had shown cyanosis since 6 years of age. At the age of 8, dyspnea on exertion developed, and cardiac disease was suspected. At the age of 14, she was examined by cardiac catheterization, but no abnormalities were found. No special therapy was performed. In the present pregnancy period, polycythemia [red blood cell count (RBC) 533 × $10^4 \cdot \mu l^{-1}$, hemoglobin concentration (Hb) 17.1 $g \cdot dl^{-1}$, and hematocrit (Ht) 51.6%] were observed. Although she underwent further examinations such as computed tomography (CT), magnetic resonance imaging (MRI), radioisotope (RI)-angiography, and chest X-ray, no abnormal findings other than hypoxemia and right aortic arch were observed. No abnormalities of hemoglobin were found either. She was diagnosed as having secondary polycythemia of unknown origin. Because of intrauterine growth retardation, cesarean section was scheduled. Upon physical examination, blood pressure was 96/66 mmHg and the pulse rate was 104 beats·min^{-1} and regular. Cyanotic lips, clubbed fingers, and pretibial edema were observed. The results of peripheral blood examination were: white blood cell (WBC) 7300·μl^{-1}, RBC 476 × $10^4 \cdot \mu l^{-1}$, Hb 11.6 $g \cdot dl^{-1}$, Ht 39.3%, and platelets (Plt) 16.8 × $10^4 \cdot \mu l^{-1}$. No polycythemia was observed because of dilution anemia in pregnancy. Although no abnormalities were seen in hepatic or renal function, decreases in serum cholinesterase (ChE) 0.34 ΔpH, plasma total protein (TP) 5.2 $g \cdot dl^{-1}$, and albumin (Alb) 3.1 $g \cdot dl^{-1}$ were observed. Pulmonary function and electrocardiogram (ECG) were normal, and only the right aortic arch was found to be abnormal by chest X-ray. The values of arterial blood gas analysis under breathing room air were: pH 7.437, partial arterial pressure of CO_2 ($Paco_2$) 25.3 mmHg, and O_2 (Pao_2) 56.2 mmHg, and base excess (BE) −4.5 mM. Since the data indicated hypoxemia, oxygen was supplied at 4 l·min^{-1} through a nasal cannula. However, Pao_2 increased to only 73.2 mmHg.

Anesthetic procedure

The anesthesia record is shown in Fig. 1. The patient was premedicated with atropine sulfate 0.5 mg intramuscularly 30 min prior to arrival in the operating room. The radial artery was cannulated under local anesthesia for continuous monitoring of arterial blood pressure. Pao_2 in the supine position while breathing room air was 59.7 mmHg and was raised to 100.4 mmHg by oxygen inhalation at 6 l·min^{-1} using a mask. Anesthesia was induced with thiamylal and muscle relaxation was obtained with succinylcholine. After induction of anesthesia and tracheal intubation, a probe of transesophageal

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Received for publication on January 26, 1995; accepted on June 8, 1995

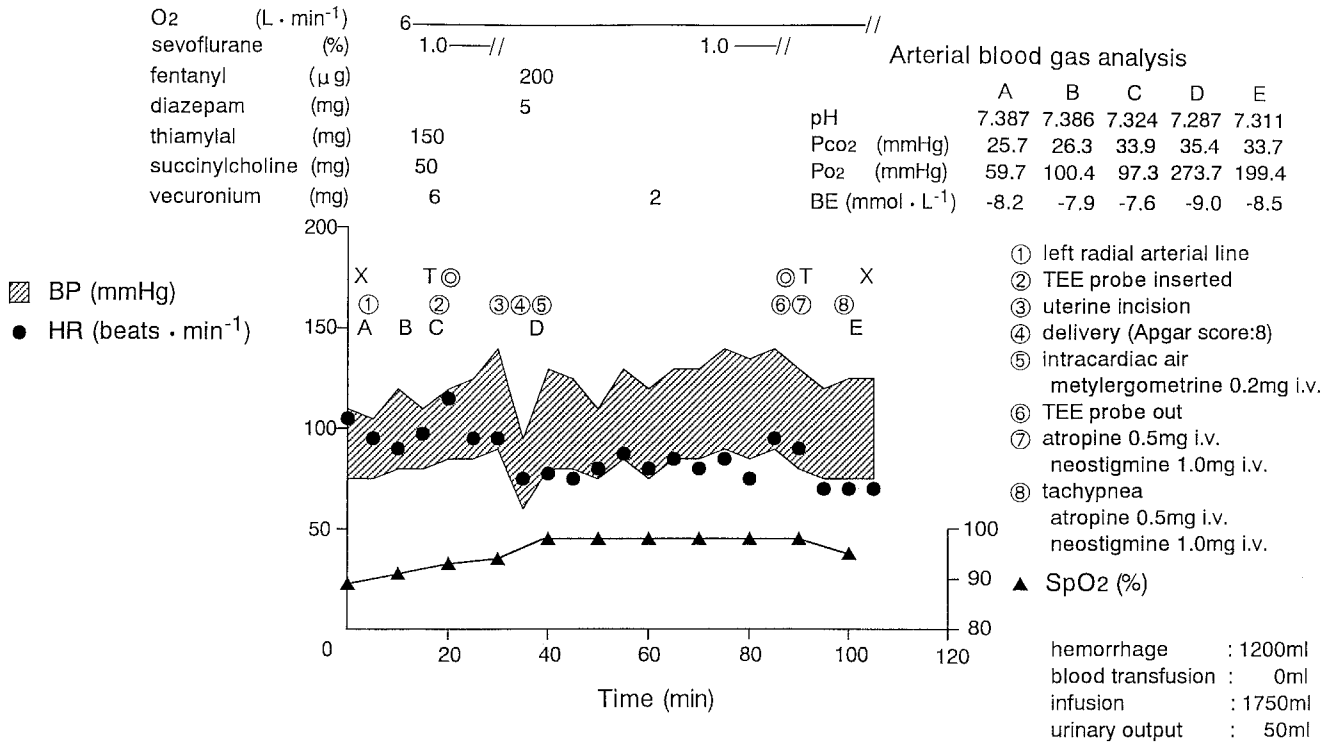

Fig. 1. Anesthesia record

echocardiography (TEE) for child (5 MHz, UST-5234S-5, Aloka, Tokyo, Japan) was inserted into the esophagus and attached to a color Doppler imaging system (SSD-830, Aloka). Anesthesia was maintained with oxygen and sevoflurane, and muscle relaxation was obtained with vecuronium. After delivery of the neonate (1 min after, the Apgar score was 8 points), marked VAE, rated grade 2 by the scoring system of Rodigas et al. [5], was observed with the TEE (Fig. 2). Since air emboli were also detected in the left atrium and ventricle through the left upper pulmonary vein, the presence of a shunt in the left lung was suspected. VAE was transient and was not accompanied by hemodynamic changes. After delivery, anesthesia was maintained with fentanyl and diazepam. After operation, oxygen saturation was good with a pulse oximeter, then extubation was performed and anesthesia was concluded.

Postoperative procedure

After transfer into the intensive care unit, no marked changes in the patient's general condition were observed; nevertheless she complained of chest pain the next day. Although no ST changes were detected on the ECG, creatine kinase (CK) and isoenzyme CK-MB were increased (526 IU·l^{-1} and 125 IU·l^{-1}, respectively). Echocardiography showed no wall motion abnormality, therefore, subendocardial infarction caused by VAE was suspected. Later, CK and CK-MB decreased gradually, and on the 4th day after operation, returned to normal levels of 121 IU·l^{-1} and 15 IU·l^{-1}, respectively. Two weeks later, two-dimensional echocardiography was performed. When 10 ml of normal saline was rapidly injected into the left cubitus vein, contrast echo appeared in the right ventricle followed by the left atrium with a delay of 1.5–2.0 s. Hence, arteriovenous fistula was diagnosed. Although cyanosis developed during physical exercise thereafter, no significant changes in the ECG and no symptoms such as chest pain were observed. The mother and the child were discharged without other complications.

Discussion

The incidence of VAE during cesarean section is high, and a 25% incidence was observed in our study using TEE [1]. Various incidences from 11% to 65% have been reported elsewhere [2,3], and a high incidence of 97% has been observed by monitoring the expired nitrogen concentration [4]. Generally, VAE is slight and its effects on respiration and circulation are limited. However, hypoxemia [3,6] or hemodynamic changes could be induced at times, and serious complications such as cardiac arrest have been reported [7]. The risk is especially high in patients with a right-to-left shunt as in

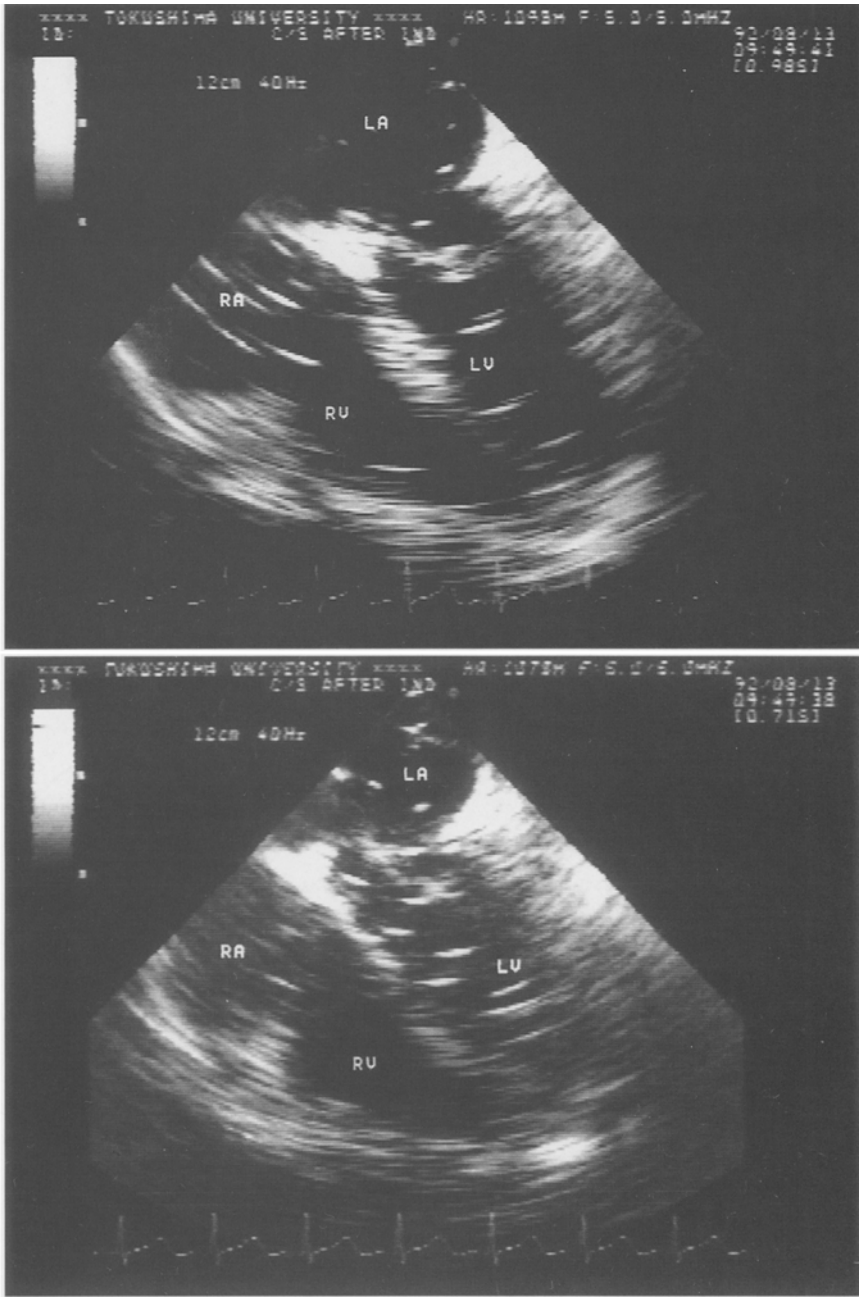

Fig. 2. **a** Air emboli were detected in the right atrium (*RA*) and right ventricle (*RV*) by transesophageal echocardiography. **b** Air emboli were also detected in the left atrium (*LA*) and left ventricle (*LV*) through left upper pulmonary vein

the present case [8]. In the present patient, subendocardial infarction seems to have been caused by air emboli. It has been reported that no statistical difference related to the type of anesthesia existed in the incidence of VAE [2], but in this case we selected general anesthesia suitable for the inhalation of oxygen with a high concentration.

Arteriovenous fistula is a relatively rare disease, and shows no abnormalities in routine examinations for cardiac or respiratory function and, therefore, can easily be overlooked. Although pulmonary angiography is usually required for a definite diagnosis, the recent development of contrast echocardiography makes it possible to diagnose definitively. In comparison with the conventional M-mode contrast echocardiography [9], two-dimensional contrast echocardiography can achieve more accurate diagnosis and is useful for postoperative follow-up examinations [10]. The present case is a very rare one in which intraoperative diagnosis was made by TEE. Our results indicate the usefulness of TEE monitoring during operation.

It has been reported that about 50% of arteriovenous fistula cases are complicated with Rendu-Osler-Weber disease [11]. This disease, a so-called hereditary hemor-

rhagic telangiectasis (HHT), is attributed to a dominant gene located on a autosome. A characteristic of this disease is thinning of the skin, conjunctiva, and fine veins and capillaries in various organs. Since only the endothelium is found in the affected regions, hemorrhage occurs easily by an even weak external force [12]. In Japan, 141 family lines of this disease have been reported [13]. Although no hemorrhagic tendency was observed in the present patient, her sister showed similar symptoms suggesting an intra-family disease. Consequently, it is possible that she is suffering from Rendu-Osler-Weber disease.

In summary, we reported a case of arteriovenous fistula complicated with possible postoperative subendocardial infarction caused by VAE during cesarean section. For patients with a right-to-left shunt, prevention and early diagnosis of VAE during operation are of great importance. TEE was useful in the detection of VAE during cesarean section and the diagnosis of pulmonary arteriovenous fistula.

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