Acute onset shortness of breath in labour: haemothorax secondary to pulmonary arteriovenous malformation

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Summary

We report the case of a 31-year-old female presenting with haemothorax secondary to rupture of pulmonary arteriovenous malformation following induction of labour. This caused fetal compromise that necessitated immediate delivery. Emergency embolisation was performed after inter-hospital transfer to a regional centre with interventional radiology facilities. The postoperative course was complicated by large volume pulmonary emboli and persistent infection which required video assisted thorascopic surgery.

Introduction

Arteriovenous malformations are abnormal communications between arteries and veins. When present in the pulmonary vasculature they can be asymptomatic or cause significant right-to-left shunt with associated cardiovascular compromise and a high risk of mortality if they bleed. They may be part of a multisystem disease or occur individually. The normal physiological changes of pregnancy can increase the risk of progression of a previously asymptomatic arteriovenous malformation.

Report

A 31-year-old para 1, gravida 2 female presented at 40\textsuperscript{th} weeks for induction of labour following 3 episodes of reduced fetal movements. Her past history consisted of emergency lower segment caesarean section for fetal distress 4 years previously. She smoked 10 cigarettes a day and had a booking BMI of 36. Induction of labour was commenced with normal observations and cardiotocography (CTG).

Ninety minutes after commencing syntocinon CTG changes were noted and the patient reported severe left sided chest pain, worse on inspiration and radiating to her back. Blood pressure was recorded at 149/111 mmHg. Heart rate was 110 beats.min\textsuperscript{-1}, respiratory rate 35 breaths.min\textsuperscript{-1} and
SpO₂ 94% breathing room air. Heart sounds were normal but she had reduced air entry at the left lung base. Oxygen was administered via a facemask at 5 l.min⁻¹ and SpO₂ improved.

The CTG showed persistent fetal bradycardia; vaginal examination demonstrated a fully dilated cervix but the patient was too distressed to facilitate vaginal delivery. Spinal anaesthesia was administered after portable chest radiograph showed a normal mediastinum with some shadowing at the left lung base only and arterial blood gas sampling (ABG) demonstrated mild hypoxaemia. A live baby girl was delivered by forceps. Umbilical cord gases showed pH 6.95. The baby was admitted to the neonatal unit briefly and made a good recovery.

During delivery SpO₂ were maintained at 98% with 6 l.min⁻¹ oxygen; tachycardia and hypertension persisted. Urgent computed tomography pulmonary angiography (CTPA) performed immediately post-delivery showed a left sided haemothorax with an actively bleeding pulmonary arteriovenous malformation (AVM) (Figure 1). A contralateral pulmonary AVM was also noted. She was referred to a regional centre for emergency embolisation using interventional radiology.

Following uneventful transfer general anaesthesia was administered in theatre; two 8 mm Amplatzer plugs were inserted into the feeding artery, successfully embolising the pulmonary AVM. Blood pressure intra-operatively required vasopressor support and 1500 ml intravenous crystalloid fluid. An acute decrease in haemoglobin from 113 g.l⁻¹ to 85 g.l⁻¹ occurred. A wide bore intercostal drain (ICD) was inserted post embolisation and drained 2100 ml blood. Following the procedure the patient became increasingly hypotensive (systolic pressure 65 mmHg) and remained tachycardic. Two units of packed red cells were transfused which improved her clinical condition. She was transferred to the HDU. The ICD was removed after 2 days but a residual effusion and moderate pneumothorax was noted on chest radiograph; a further ICD was sited and subsequently removed following radiological resolution of the pneumothorax. Over the following days the patient had an ongoing oxygen requirement and rising inflammatory markers. She was treated with piperacillin-tazobactam and the possibility of an infected haemothorax was considered. A further CTPA showed large volume pulmonary emboli in the contralateral lung and a heparin infusion was commenced. Owing to the high risk of performing evacuation by video assisted thorascopic surgery (VATS) a further wide bore ICD was inserted into the pleural collection and 1300 ml blood-stained fluid drained. Ongoing oxygen requirement necessitated transfer to the regional cardiothoracic centre for VATS drainage of persistent infected haemothorax. The patient was discharged 20 days after admission.

**Discussion**

Arteriovenous malformations (AVMs) are abnormal thin walled communications between arteries and veins that can occur anywhere within the vascular system but are most commonly found intracranially. Pulmonary AVMs are rare with an incidence of 2-3 per 100,000 and a higher frequency in women [1]. Between 48% and 80% of patients with pulmonary AVMs have a parallel or subsequent diagnosis of Hereditary Haemorrhagic Telangiectasia (HHT), also known as Osler-Weber-Render disease [1]. This is an autosomal dominant condition that affects between 1 in 5,000 and 1 in 8,000 people with variable phenotypes. Symptoms can include recurrent nosebleeds, telangiectasia and gastrointestinal bleeding, all of which can cause a chronic anaemia [2]. AVMs are a common feature that can be present in the pulmonary (50%), hepatic (30%), cerebral (10%) and, occasionally, spinal (1%) vasculature, but most patients remain asymptomatic [2]. The recommended treatment for a pulmonary AVM is radiological embolisation as well as dental hygiene advice with antibiotic prophylaxis for procedures with a high risk for bacteraemia [3]. This is due to reduced exposure to the capillary bed reticuloendothelial cell system by virtue of the right-to-left shunt, and co-amoxiclav (clindamycin for penicillin allergic patients) has been recommended allowing broad-spectrum cover for the common organisms [4]. If left untreated mortality from pulmonary AVM has been reported to be as high as 11% [5].

There are multiple reports of pulmonary AVMs presenting in pregnancy [6,7,8], however presentation during labour is very unusual. All AVMs are likely to enlarge and are at higher risk of rupture during pregnancy due to the normal changes that occur in antenatal physiology. Increased...
Anaesthesia Cases

blood volume, increased cardiac output and hormonal changes causing smooth muscle relaxation contribute to an increased risk of bleeding [5]. Mortality may occur from either catastrophic haemorrhage or marked right-to-left shunt resulting in cardiovascular collapse.

Anaesthetic management of obstetric patients with known pulmonary AVMs related to HHT has been previously discussed within the literature [9]. Key management focuses on reducing pulmonary vascular resistance and maintaining systemic vascular resistance to minimise the right-to-left shunt [9]. Regional anaesthetic techniques with strict blood pressure control may be preferable to general anaesthesia requiring positive pressure ventilation. The risk of systemic embolism across the AVM necessitates antibiotic prophylaxis regardless of mode of delivery. AVMs may also affect the airway and therefore the risk of bleeding from instrumentation associated with tracheal intubation need to be recognised [9].

Pre-pregnancy diagnosis of pulmonary AVM and management with embolisation is ideal, although often not possible in cases such as ours. Antenatal embolisation may be undertaken, however, concern regarding ionising radiation exposure during pregnancy will contribute to decision making. Despite successful embolisation there will always remain a significant risk of re-canalisation [10] and vigilant antenatal care is imperative.

Sudden onset of chest pain, shortness of breath and oxygen desaturation in a previously well parturient in labour is more commonly associated with pulmonary or amniotic fluid embolus. However our patient's presentation was unclear and differential diagnoses initially included aortic dissection and pneumothorax. A decision to perform spinal anaesthesia was made as it was felt beneficial to avoid general anaesthesia with the potential to further increase blood pressure during direct laryngoscopy.

Our patient was initially cardiovascularly stable and able to tolerate spinal anaesthesia. Owing to the location of her pulmonary AVMs it is likely that lying more supine intra-operatively will have reduced blood flow through the AVM and improved right-to-left shunt. It is probable that the sustained fetal bradycardia was secondary to reduced apparent maternal circulating volume and deteriorating oxygen levels.

The post embolisation period was complex. Recurrent ICDs have significant risk of infection but were necessitated by ongoing cardiovascular and respiratory symptoms. Many risk factors for venous thromboembolism were present (pregnancy, increased BMI, smoker, instrumental delivery, reduced mobility and critical care admission) however pharmacological thromboprophylaxis was withheld due to the high risk of bleeding.

Embolisation of the right sided pulmonary AVM was undertaken as a day case procedure following discharge after her acute admission. All patients with pulmonary AVMs should be referred to the Hereditary Haemorrhagic Telangiectasia clinic for genetic testing and counselling.

Previously asymptomatic pulmonary AVMs are commonly revealed in pregnancy and are associated with a high risk of mortality, thus anaesthetic management of these patients can be complex and require coordinated multidisciplinary care. The preferred management is embolisation in the antenatal period with awareness that re-canalisation can occur. Anaesthesia should focus on reducing pulmonary vascular resistance whilst maintaining systemic vascular resistance.

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Competing Interests

No external funding and no competing interests declared.
CTPA showing marked left sided effusion (haemothorax) and contrast highlighting the AVM.

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