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A Case Report On 29-Year-Old Pregnant Female, 36-Week Gestation, Presented with Sudden Onset of Severe Back Pain, Followed by Dyspnea, Shock, Hemothorax and Sudden Death: Very Rare Cause of Maternal Mortality During the Covid-19 Pandemic

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Abstract

A 29-year-old female with 36 weeks pregnancy, previously healthy, presented with severe back pain at interscapular region followed by dyspnea, shock and, hemothorax. She had sudden cardiac arrest during diagnostic pleural aspiration. Post-mortem examination revealed normal descending aorta and massive hemothorax probably due to rupture of arteriovenous malformation of intercostal artery or pulmonary vasculature.

Keywords: pregnancy, back pain, dyspnea, shock, hemothorax, cardiac arrest, arteriovenous malformation, intercostal artery.

Supplementary information The online version of this article (https://doi.org/xx.xxx/xxx.xx) contains supplementary material, which is available to authorized users.

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Background

According to WHO, an estimated 810 women are dying each day due to complications of pregnancy and childbirth - mostly from preventable or treatable causes, such as infectious diseases and complications during or after pregnancy and childbirth.The study from Mexico highlighted that the maternal mortality rate were higher than pre-COVID era due to both COVID-19 infection and COVID-19 non-related causes like poor access to treatment center/hospital (Mendez-Dominguez et al., 2021). Three-quarters of maternal deaths are in women with coexisting medical complications which are already known like diabetes mellitus, hypertension or unknown. It is also difficult to differentiate symptoms of normal pregnancy from pathological symptomatology; thus, the awareness of acute medical problems in pregnancy is important (Neuberger & Nelson-Piercy, 2015).

The differential diagnosis of dyspnea both in pregnancy and during the postpartum period in COVID-19 era are pulmonary embolism, spontaneous hemothorax, COVID-19 pneumonia, acute exacerbation of bronchial asthma and heart failure either due to cardiomyopathy or Beri beri; spontaneous hemothorax is very rare among them. The reports on massive spontaneous hemothorax due to arteriovenous malformation (Doshi et al., 2009) (Md Noh et al., 2018) (Sood et al., 2011) and therapeutic measures, therapeutic embolization, open surgical exploration and resection were mentioned (Md Noh et al., 2018). Some cases of them had underlying connective tissue/collagen disorder like hereditary hemorrhagic telangiectasia (Di Crescenzo et al., 2015)(Raiya et al., 2017)(Mourad et al., 2016), Ehlers-Danlos syndrome, Osteogenesis imperfecta and neurofibromatosis (Hashimoto et al., 2021a) or idiopathic (Dimitriou et al., 2016).

Case presentation

A 29-year-old female with term pregnancy, 36 weeks by gestation, was attending antenatal clinic regularly; and, she was in good health apart from breech presentation. She had abortion one time, 2 years ago. She took combined contraceptive pill for one year. There was no history or physical signs suggestive of Ehlers-Danlos syndrome, neurofibromatosis, osteogenesis imperfecta or hypermobility of joints.

She experienced sudden onset of severe back pain at mid-scapular region while she was having shower (11:00 hour). It was associated with sweating and vomiting for one time; and not related with breathing or coughing or body movements. There was no fever or sore throat. Twenty minutes later, the pain was gradually better; thus, she had to rest in bed to avoid further pain. Two hours later (13:00 hour), the pain came back again; thus, she was brought to hospital. At A&E, she was not dyspneic, SaO2 on air was 98%, afebrile, blood pressure 130/70 mmHg, pulse rate 84/ minutes. The abdomen was soft and not tender. Obstetric examination revealed 36-week-sized uterus with single fetus, longitudinal lie, breech presentation; there was no definite uterine contraction nor uterine tenderness. Fetal heart sound was 140 / min, strong and regular. Therefore, the initial impression was premature labor pain causing back pain. Thus, she was given anti-emetics, dexamethasone; and, she was kept under observation for both maternal and fetal monitoring.

Then, the pain was relived temporarily; but recurred at 16:00 hour. It was associated with vomiting. Blood for complete picture showed neutrophil leukocytosis (Total WBC 13.0 X 10^9/L; Neutrophils 85%, Lymphocyte 14%, Monocyte 1%), normochromic normocytic anemia (Hemoglobin 9.1 gm%) with normal platelet count (228 X 10^9/L). Nasopharyngeal swab for COVID-19 was negative. Blood for D dimer and serum amylase were pending.

At 20:00 hour, she suffered dyspnea. At 21:00 hour, the patient became restless; the blood pressure suddenly dropped to 100/40 mmHg and pulse rate rose to 110/min. The SaO2 was 96% on air and rose to 99% on oxygen 2L/minutes. The extremities became cold. The whole on call team: obstetric and gynecologist, on-call physician, surgeon and chest physician reviewed the case.

Respiratory system examination revealed dramatic changes; reduced vesicular breath sound with dullness on left lower lung. Chest radiograph showed left-sided moderate pleural effusion though mediastinal shift and tracheal shift were difficult to interpret because of positioning. Figure (1) ECG revealed sinus tachycardia. In USG abdomen, a single viable 36-week fetus with breech presentation; there was no features of placenta abruption. Cardiotocography was reactive pattern; no uterine contraction. Figure (3) USG left chest was

consistent with fluid in left pleural cavity. Figure (2) Thus, USG guided pleural aspiration was done; and, 10 cc of bloody fluid was aspirated. While doing diagnostic aspiration, the patient had excruciating pain and cardiopulmonary arrest on 13:45 hour. Cardio-pulmonary resuscitation was done for 30 minutes; and it was without success. The patient expired at 00:30 hour.

Post-mortem cause of death was massive left-sided hemothorax 3.0 liter. (Figure 4) The aorta was dissected; there was no dissection or aneurysm. (Figure 5,6,7& 8) There was no aneurysm in main branches of aorta or intercostal arteries. Therefore, the possible site may be one of pulmonary arteriovenous malformation or small intercostal arteries; anterior intercostal artery. The weakened wall site, either aneurysm or arteriovenous malformation, became collapsed after rupture and invisible in postmortem; it was cut during thoracic wall dissection. The cut sections of lungs were normal. Heart and pericardium were normal. Dissection along abdominal aorta was normal. There was no evidence of neurofibromatosis, Ehlers Danlos syndrome, osteogenesis imperfecta or hereditary hemorrhagic telangiectasia in external appearance.

Discussion

The common causes of severe back pain at interscapular region in term pregnancy are acute pulmonary embolism, acute myocardial infarction, pleurisy, aortic dissection, acute cholecystitis, acute pancreatitis and acute pyelonephritis. In this patient, absence of fever excluded infective causes like acute cholecystitis, acute pancreatitis and acute pyelonephritis. Acute myocardial infarction was unlikely as she was young and there was no risk factor like obesity, diabetes mellitus, smoking and family history; ECG was normal. Regarding aortic dissection, it was also low possibility as the prevalence was very low. Acute pulmonary embolism was the only provisional diagnosis. There were several reports on pregnancy with pulmonary embolism presenting with acute chest pain, dyspnea and hypotension.

The possibilities of severe pain for 8 hours followed by sudden onset of dyspnea and shock were acute pulmonary embolism, acute myocardial infarction, acute pancreatitis and rupture aortic aneurysm/dissection. As ECG was normal, acute myocardial infarction was excluded. As the abdomen was soft and non-tender, acute pancreatitis was unlikely. Pulmonary embolism and rupture aortic aneurysm/dissection were the two remaining possibilities.

New physical signs only 10 hours after onset of pain showed presence of fluid in left pleural cavity; confirmed by chest radiograph and bedside ultrasound. It highlighted the importance of clinical observation, repeating and reviewing the case; also, the value of bedside ultrasound and chest radiograph.

There were several reports on hemothorax particularly in late pregnancy. One case report on neurofibromatosis invading intercostal arteries which rupture during pregnancy; hemorrhagic shock may be overlooked in late pregnancy (Hashimoto et al., 2021). Blood volume in late pregnancy is 40–50% higher; and, the blood pressure may drop after 40% of circulating blood volume (1.5 liter) is lost. In this case, initial pain at 11:00 hour was due to impending rupture; it was followed by relatively reduced intensity of pain probably due to sealed by normal defense mechanism. Development of hypotension at 21:00 hour, 10 hours after initial pain, with dyspnea and fall in SaO2 on air was owing to complete rupture; hemothorax, massive exsanguination and hypovolemic shock. The whole event from onset of pain to shock took 10 hours; and, from onset of pain to cardiac arrest was 12 hours. Unfortunately, we could not save the patient; cardiopulmonary arrest coincided with the time of diagnosis of hemothorax.

Several reports mentioned therapeutic measures on etiology of hemothorax in pregnancy (Zimmer, E. Z., 1988) like coiling, therapeutic arterial embolization and lobectomy. They could save their patients as the whole event, from onset of pain to rupture, lasted several days; not like this patient- 12 hours. Most frequent timing for rupture of either aneurysm or arteriovenous malformation was in late pregnancy; it may be due to increasing blood volume in last trimester.

The likely cause of acutely developed left sided pleural effusion and bloody pleural aspirates revealed acute hemothorax; it may be due to rupture of artery or vein which has weaken walls either due to arteriovenous malformation or aneurysm. Post-mortem cause of death was massive left-sided hemothorax 3.0 liters. The aorta was dissected; there was no dissection or aneurysm. There was no aneurysm in main branches of aorta or intercostal arteries. Therefore, the possible site may be one of pulmonary arteriovenous malformation or small

intercostal arteries; anterior intercostal artery. The weakened wall site, either aneurysm or arteriovenous malformation, became collapsed after rupture and invisible in postmortem; it was cut during thoracic wall dissection.

The reported cause of spontaneous hemothorax in late pregnancy was due to rupture of arterial aneurysm or arteriovenous malformation (Doshi et al., 2009) (Md Noh et al., 2018) (Sood et al., 2011). Few reports mentioned the etiology of arterial abnormalities/arteriovenous malformation (Sood, N., 2011) as a result of neurofibromatosis (Hashimoto et al., 2021), Ehlers Danlos syndrome, osteogenesis imperfecta or hereditary hemorrhagic telangiectasia (Di Crescenzo et al., 2015) (Raiya et al., 2017)(Mourad et al., 2016); however, there was no evidence of them in both physical examination and external appearance in this patient. Therefore, the cause of spontaneous hemothorax in this patient was probably due to idiopathic arteriovenous malformation; it was seen in some report (Dimitriou et al., 2016).

Conclusion

This case highlighted the value of repeating physical examination which pointed the diagnosis though we could not save the patient. If the course of hemothorax was not too short, we could have time to arrange CT pulmonary angiogram and radiological intervention. Hemothorax due to either aneurysm or arteriovenous malformation is a rare and potentially devastating disease that may present during pregnancy particularly in last trimester; awareness is important.

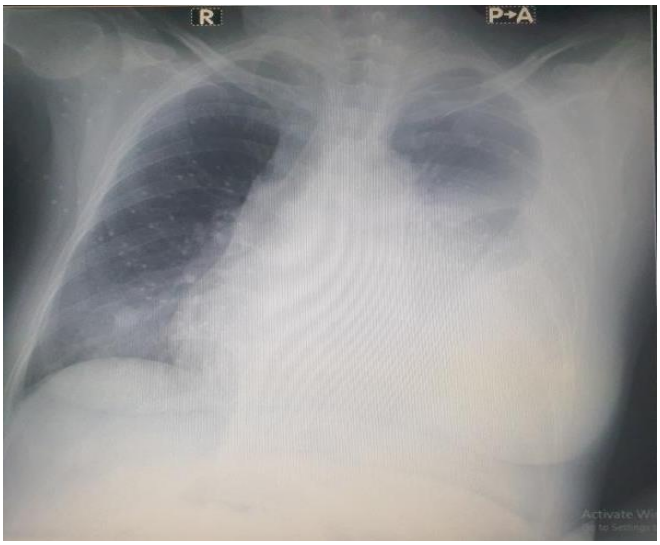

Figure (1) Chest radiograph showing left pleural effusion

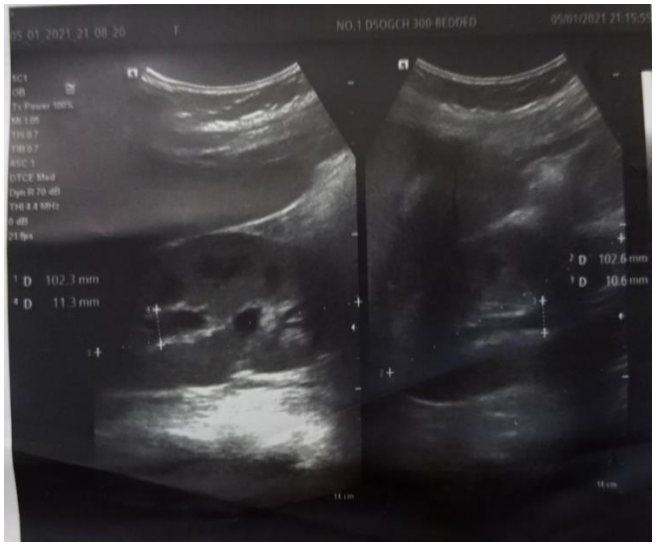

Figure (2) USG left chest showing pleural effusion

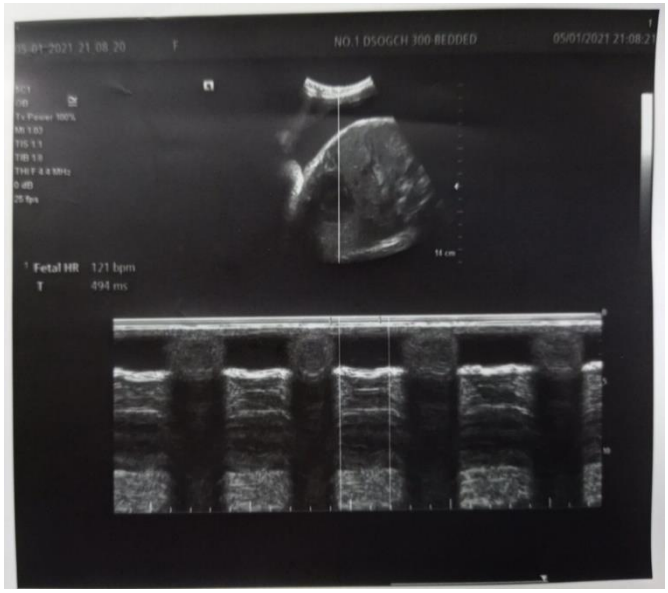

Figure (3) Cardiotocography showing fetal heart activity

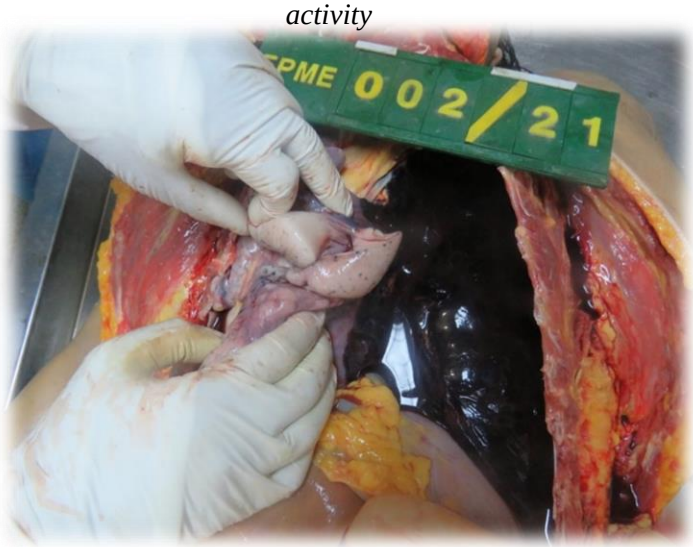

Figure (4) Left hemothorax occupying nearly the whole chest

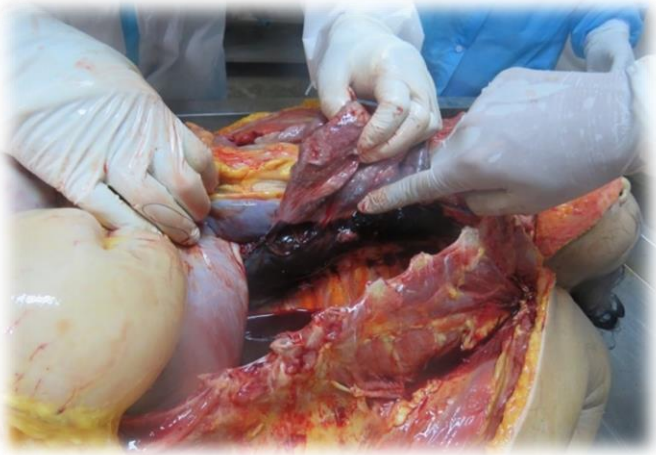

Figure (5) The hematoma near inferior vena cava and descending aorta, after removing the blood in Left chest

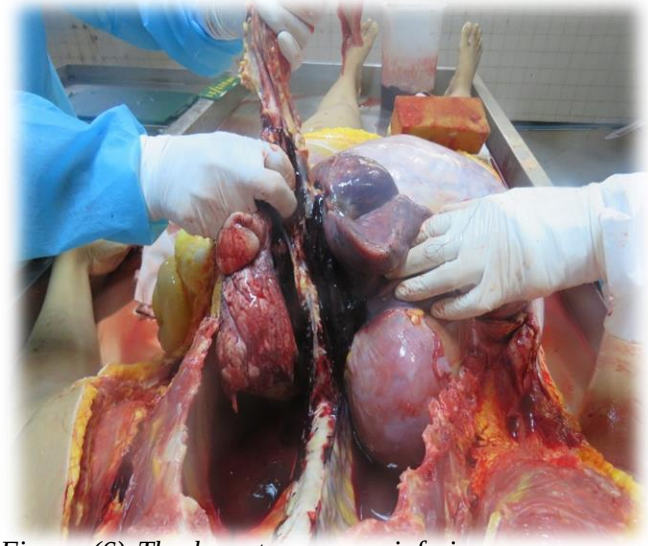

Figure (6) The hematoma near inferior vena cava and descending aorta, after removing the blood in Left chest

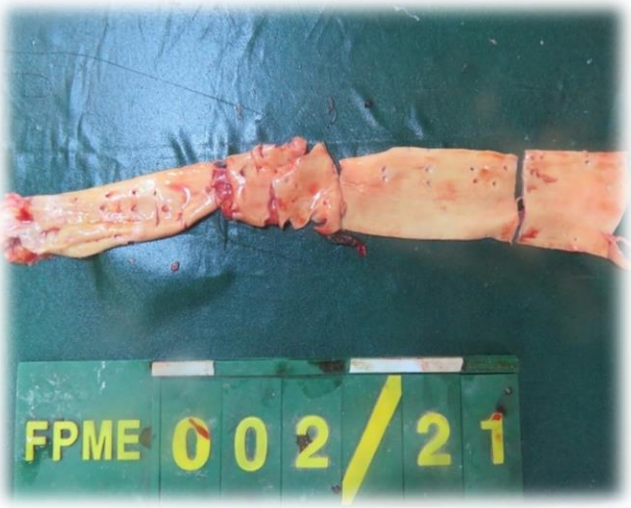

Figure (7) The interior wall of the descending aorta showing no aneurysm or dissection

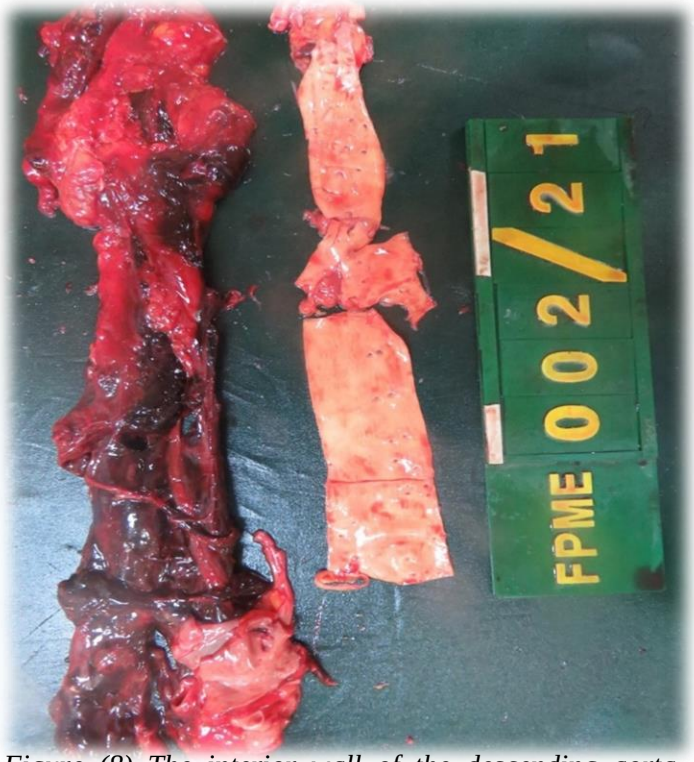

Figure (8) The interior wall of the descending aorta showing no aneurysm or dissection and the hematoma attached to oesophagus

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Declaration of conflict of interest

The authors declared no potential conflicts of interests with respect to authorship and publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting cases.

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Informed consent

The informed consent for publication in this article was obtained from husband of patient.

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