Spontaneous pneumomediastinum: a rare presentation of diabetic ketoacidosis in a pregnant woman

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Summary: Pneumomediastinum (PM) or mediastinal emphysema is defined as the presence of free air around mediastinal structures. Spontaneous (or atraumatic) pneumomediastinum (SPM) is a rare complication during pregnancy. Primary or spontaneous PM can arise due to increased intra-alveolar pressure. Secondary PM is due to direct trauma, intrathoracic infections or violation of the aerodigestive track. This case report describes a pregnant woman newly diagnosed with diabetes presenting with an SPM due to vigorously vomiting and Kussmaul's breathing caused by diabetic ketoacidosis. Appropriate management of SPM and its underlying cause is required to reduce the risks for both mother and child.

Keywords: diabetes, maternal–fetal medicine, thoracic medicine

INTRODUCTION

Pneumomediastinum (PM) or mediastinal emphysema is defined as the presence of free air around mediastinal structures. PM that arises spontaneously is due to increased intra-alveolar pressure. Secondary PM is due to direct trauma, intrathoracic infections or violation of the aerodigestive track.¹

To our knowledge, this is the first report of a pregnant patient presenting with a spontaneous (or atraumatic) pneumomediastinum (SPM) due to diabetic ketoacidosis (DKA). Until 2008, only 58 cases of PM in association with DKA have been reported, but never during pregnancy or labour.² Although SPM is a rare event during pregnancy, it is important to recognize it and to treat the underlying condition.

CASE

A 15-year-old girl visited her general practitioner with complaints of acute breathlessness and pleuritic chest pain after an episode of vigorous vomiting. In the diagnostic work-up, the urinary pregnancy test was positive and she was referred to the emergency department of our hospital. She was otherwise healthy.

Physical examination showed a respiratory rate of 30–40 per minute and oxygen (O₂) saturation was between 75% and 99%. The pulse rate varied from 80 to 126 beats per minute, blood pressure was 120/80 mmHg and temperature was 36.8°C. Inspection, palpitation and auscultation of the chest showed no abnormalities. Abdominal examination revealed an enlarged

uterine size corresponding with a pregnancy of 28 weeks, which was confirmed by ultrasound. The extremities showed no signs of oedema and/or deep venous thrombosis.

Initially, a high suspicion of pulmonary embolism arose because of high oxygen requirements (15 L O₂/minute) in combination with a normal electrocardiogram and chest X-ray. A spiral computed tomography (CT) scan of the chest showed no pulmonary embolus, but demonstrated a PM with stasis of fluid in the oesophagus (Figure 1).

Laboratory tests showed hyperglycaemia, metabolic acidosis and ketosis. Reassessment of the chest X-ray confirmed mediastinal air that outlined the cardiac silhouette and extended along the aorta (Figure 2).

The differential diagnosis at that time included Boerhaave's syndrome because of the PM after her episode of vigorous vomiting caused by DKA. The patient was admitted to the intensive care unit and was intubated because of respiratory failure and suspicion of Boerhaave's syndrome. Antibiotic treatment was prescribed to prevent a mediastinitis.

A gastroduodenoscopy and a spiral CT scan of the abdomen demonstrated no macroscopic ruptures or obstructions of the oesophagus. After excluding the possibility of an oesophageal rupture, a diagnosis of SPM caused by DKA was made in the pregnant woman with newly diagnosed diabetes. DKA was treated with intravenous insulin, fluid and potassium supplementation. After stabilization of the patient, a stillbirth was diagnosed.

Two days after the admission, the patient was extubated and induction of labour with misoprostol was started. One day later, a boy with a weight of 890 g (p30–40) was born. The placental weight was 180 g and histological analysis demonstrated multiple infarcts and fibrin depositions (40%).

Ten days after the admission, the patient was discharged on subcutaneous insulin therapy. Two years after the

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Figure 1 CT scanning of the thorax: (1a) transverse view, (1b) sagittal view: both anterior and posterior of the mediastinum, there is an air configuration (pneumomediastinum)

delivery, the patient died of DKA due to lack of medication compliance.

DISCUSSION

In pregnancy SPM usually occurs during the second stage of labour in healthy primiparous patients who have undergone a prolonged and protracted course of labour and have macrosomic babies. Its occurrence is related to the Valsalva manoeuvre during the expulsive phase of labour, which leads to an acute rise in intra-alveolar pressure. This Valsalva manoeuvre also occurs with strong coughing, vigorous vomiting, screaming or pushing during labour.^{1,3,4}

Other causes of SPM may be the use of inhalational drugs, physical activity, playing wind instruments, asthma exacerbation, spirometry, defecation, seizure and DKA. In DKA

increase in alveolar pressure is believed to be caused by vomiting and Kussmaul's breathing.^{1-3,5}

Chest pain is the most common presenting symptom of SPM and other symptoms can include dyspnoea (usually mild), dysphonia, throat pain, dry cough, anxiety, haemoptysis and palpitations.^{1,3,5}

A classical physical finding in SPM is Hamman's crunch. This sound is synchronous with cardiac systole and is composed of crepitus, crunching and popping noises heard best at the apex of the heart. Other physical findings include subcutaneous emphysema, neck swelling, tachycardia, decreased cardiac dullness and hyperresonance over the sternum. A low-grade fever is frequently noted in patients with DKA-associated SPM even without an identified infection.^{1,4,5}

The differential diagnosis of an obstetric patient with chest pain includes pulmonary embolism, amniotic fluid embolism, myocardial infarction, pneumothorax, toxic effect of injected drugs like β -adrenergic tocolytic therapy, aortic dissection, cardiac tamponade, pericarditis, angina pectoris and mediastinitis.^{1,4,6}

In some patients mild to moderate elevation of white blood cell count is observed, which results from reactive inflammation associated with air dissection in the mediastinum. Depending on the severity of the underlying respiratory compromise, the blood gas analysis can be normal or may reveal hypoxia, hypocarbia or hypercarbia. Other laboratory findings like hyperglycaemia can be found, depending on the underlying cause.^{1,5,7}

Electrocardiographic changes may occur in some cases of SPM and include non-specific ST and T wave abnormalities, which may be in part related to physical displacement of mediastinal structures.^{4,5}

The diagnosis of SPM is revealed by radiographic examination. CT has become the gold standard for diagnosing PM.^{1,3}

Because of the highly unfavourable outcome, if not promptly diagnosed, secondary PM, like oesophageal tears (Boerhaave's syndrome) and mediastinitis, must be sought and excluded. Therefore, if emesis or retching is the precipitating event, an oesophagogram should be obtained. It can be necessary to use CT or endoscopy, when the patient is comatose or the patient's mental status is altered.^{2,3,5}

SPM requires conservative treatment, including sedation, analgesics and oxygen supplementation. The patient should be observed closely. The air may build up and enter the

Figure 2 Chest X-ray. The mediastinal air is outlining the cardiac silhouette and extends along the aorta

pleural or pericardial space, which could lead to (tension) pneumothorax or pneumopericardium. When severe cardiovascular symptoms are present, a mediastinotomy may be indicated to relieve a symptomatic PM.

When the diagnosis of PM is made during labour, prevention of hypoxia and fetal monitoring are indicated, to prevent unfavourable outcome for the baby. Some authorities recommend shortening of the second stage of labour by instrumental delivery to prevent an increase of mediastinal pressure.^{3–5} If caesarean section is required, the use of general anaesthesia is considered contraindicated, because it is likely to cause further expansion of PM with the risk of developing a (tension) pneumothorax. Therefore, epidural anaesthesia is preferred.⁴

The maternal risks of DKA are the same as in non-pregnant patients. (Severe) hypokalaemia leads to cardiac arrhythmias. Severe hyperglycaemia can result in dehydration with loss of kidney function and ketoacidosis due to insulin deficiency. Dehydration and acidosis ultimately can result in multiple organ dysfunction. Also hypotension and acidosis cause decreased placental blood flow with eventually life-threatening hypoxia and acidosis of the fetus.

The treatment of DKA is primarily focused on the maternal condition. An emergency caesarean delivery before stabilization of the pregnant woman leads to deterioration of her condition. The fetal condition will recover after improvement of the maternal condition following adequate treatment.⁸

Both the SPM and any underlying or associated condition such as DKA must be carefully managed to ensure the best

outcome for both mother and baby. Fortunately, the recurrence of SPM is very rare.^{1,4}

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