Rare disease

Spontaneous subcutaneous emphysema and pneumomediastinum during second stage of labour

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Summary

A 27-year-old healthy primigravida presented with spontaneous labour at 40 weeks gestation and proceeded to an unassisted vaginal delivery of a 3.2 kg infant. She immediately developed swelling in the neck and facial area associated with pleuritic chest pain. There was palpable crepitus throughout her anterior chest wall and neck. CT confirmed pneumomediastinum. She was managed with oxygen and analgesia in the intensive care unit and subsequently discharged with resolution of her symptoms. Postpartum pneumomediastinum with subcutaneous emphysema is treated conservatively after cardiac and embolic causes have been excluded; however, there is no consensus on the management of subsequent pregnancies.

BACKGROUND

Pneumomediastinum is a rare occurrence during pregnancy, when it is eponymously known as Hamman's syndrome. Pushing during labour is implicated as a cause; others include oesophageal rupture, trauma, acute asthma or any condition which increases intrathoracic pressure.

It commonly presents with sudden onset of chest pain and shortness of breath: a sinister collection of symptoms in these patients. Therefore, after swiftly excluding embolic or cardiac causes, this rare diagnosis should be considered.

It carries a good prognosis and usually resolves without intervention.¹ Close monitoring is required as theoretically cardiac tamponade can occur secondary to tension pneumomediastinum; however, this has never been reported in humans.² In addition, pneumothorax has also been associated with the syndrome in a small number of cases.³

We report a case of spontaneous subcutaneous emphysema and pneumomediastinum occurring during the second stage of labour.

CASE PRESENTATION

A 27-year-old healthy primigravida Caucasian, with uneventful antenatal care, presented at 40 weeks of gestation to the labour ward of a district general hospital in spontaneous labour. She had no medical or surgical history. She was an ex-smoker of 3 years, with a 2 pack-year history. The first stage of labour lasted 6 h and she pushed for 1 h and 30 min, during which there were no concerns regarding foetal well being. She delivered a healthy 3.2 kg male infant.

Post partum, she immediately developed facial swelling extending to the neck and upper chest wall with dysphonia. Initially there was no chest pain, cough or shortness of breath, although subsequently she reported mild pleuritic chest pain which was relieved by non-opioid analgesia.

On examination, she appeared comfortable with a respiratory rate of 18 breaths per minute and oxygen saturation

of 98% on room air. Her pulse rate was 72 beats per minute and her blood pressure was 118/72 mm Hg. There was significant neck and lower facial swelling, with palpable crepitus, which extended down to her thorax involving the upper abdomen and arms. On auscultation, transmitted crepitus was heard but air entry appeared equal in both lungs. The trachea was central and apex beat was not deviated.

INVESTIGATIONS

ECG showed sinus rhythm with a normal axis and no evidence of ischaemia. She underwent routine laboratory investigations which were unremarkable. An urgent chest radiograph (figure 1) revealed marked subcutaneous emphysema throughout the soft tissues of the chest wall and unremarkable lung fields. CT scan of the thorax (figure 2) confirmed significant pneumomediastinum.

TREATMENT

She was managed conservatively with analgesia and oxygen in the intensive care unit. Daily chest radiograph revealed no worsening of her pneumomediastinum and no progression of her subcutaneous emphysema.

OUTCOME AND FOLLOW-UP

Her symptoms resolved and she was discharged on the 3rd day post partum. She was followed up 2 weeks later, with complete resolution of her symptoms.

DISCUSSION

Spontaneous pneumomediastinum is a rare condition in pregnancy with about 200 cases reported worldwide since Simmons first described it in 1783.⁴ The midwife to the Queen of France may, however, have been the first to have noted the problem in 1617. The incidence of pneumomediastinum associated with pregnancy is between 1 in 2000

Figure 1 Chest radiograph showing marked bilateral subcutaneous emphysema advancing into the soft tissues of the neck and upper limbs. The lung fields are clear and heart size normal.

Figure 2 CT slice through lung views showing extensive bilateral subcutaneous emphysema, significant pneumomediastinum but no oesophageal rupture or pneumothorax. There is significant cardiac compression with reduced right ventricular filling due to the pneumomediastinum.

and 1 in 100 000 deliveries.^{5 6} Primiparous women appear to be more affected.

This phenomenon most likely occurs when intrathoracic pressure increases, causing alveolar rupture during multiple valsalva-type manoeuvres.² Air, under pressure, leaks into the perivascular connective tissue planes and then into the mediastinum, retropharyngeal and subcutaneous spaces of the neck;⁷ from here air can track throughout the body. The clinical presentation depends on the amount of air in the free spaces, and often involves chest pain, breathlessness and facial swelling. The typical Hamman's sign of 'crackles or churning sounds heard in the lateral decubitus position during systole'⁷ is commonly missed or not looked for because of lack of awareness by clinicians. Indeed, in this case, Hamman's sign was not elicited because of unfamiliarity.

Theoretically, women with prolonged second stage of labour or with cephalopelvic disproportion should be at higher risk because of presumed increased intrathoracic pressures during more pronounced valsalva manoeuvres. However, our patient was a healthy primigravida with a normal second stage of labour and average-sized baby for our population. Indeed, a review by Reeder suggests it often occurs with an average labour length and normal infant birth weights.³ Most cases occur during labour or immediately post partum; however, the syndrome has been described up to 2 months post partum.¹ The mechanism for this is unclear.

Spontaneous subcutaneous emphysema with pneumomediastinum is a self-limiting condition, rarely complicated, unless there are underlying respiratory conditions. Although four maternal deaths have been reported in patients with Hamman's syndrome,³ these were all before 1908 and it is unclear whether this may be due to a complication of the syndrome or the lack of obstetric facilities in that period of time. Cardiac, infective and embolic causes of chest pain must be excluded and the diagnosis is confirmed with a CT scan of the thorax. Echocardiography may also be useful if available.

The treatment is mainly supportive with oxygen and analgesia, involving obstetricians, physicians and cardiothoracic surgeons. If the syndrome occurs before the fetus is delivered, it may be necessary to convert to caesarean section to prevent the pneumomediastinum from expanding. Follow-up radiography is reasonable to exclude any

progression or complications; however, it may not be needed if the patient is improving clinically.

The risk of recurrence is low¹ and there is no consensus on the management of subsequent pregnancies; however, we recommend reassurance with increased maternal monitoring during labour and the immediate post partum period. We do not recommend prophylactic forceps delivery as a means to reduce the risk of recurrence.

Learning points

- ▶ Spontaneous subcutaneous emphysema with pneumomediastinum is a rare complication of pregnancy and the postpartum period.
- ▶ Cardiac, infective and embolic causes of chest pain must be excluded and the diagnosis is confirmed with a CT scan.
- ▶ It should be managed conservatively by a team of specialists, ideally in a high dependency setting and when one suspects pneumomediastinum, look for the elusive Hamman's sign.
- ▶ The best management in subsequent pregnancies is unknown; however, we recommend reassurance with increased maternal monitoring during labour and the immediate postpartum period.

Competing interests None.

Patient consent Not obtained.

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Please cite this article as follows (you will need to access the article online to obtain the date of publication).

McGregor A, Ogwu C, Uppal T, Wong MG. Spontaneous subcutaneous emphysema and pneumomediastinum during second stage of labour. *BMJ Case Reports* 2011;10.1136/bcr.04.2011.4067, date of publication

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