Rare disease

Postpartum pneumomediastinum manifested by surgical emphysema. Should we always worry about underlying oesophageal rupture?

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Summary

Spontaneous pneumomediastinum during labour is a rare, usually benign and self-limiting condition. It often presents with chest or neck pain and surgical emphysema. The latter sign is easy to demonstrate but often missed during clinical assessment if the condition is not included in the differential diagnosis of chest pain and dyspnoea in peripartum. The authors describe a case of 20-year-old primigravida who developed surgical emphysema following prolonged vaginal delivery. The chest x-ray revealed pneumomediastinum, and small left apical pneumothorax. She was investigated with CT of the chest and contrast swallow, both of which excluded oesophageal perforation. The management was conservative and she made a complete recovery. Spontaneous oesophageal rupture is a potential cause of pneumomediastinum and leads to high morbidity and mortality if not diagnosed early. However, it is extremely uncommon in labour, especially without a preceding history of vomiting. Unless a strong clinical suspicion exists, routine investigations and or treatment of suspected oesophageal perforation are unnecessary.

BACKGROUND

Subcutaneous emphysema originating in the mediastinum has an estimated incidence of 1 in 100 000 deliveries.¹ Approximately 200 cases have been reported worldwide.^{2 3}

Despite its rarity and usually benign course it requires vigilance and prompt diagnosis. Therefore, both obstetricians and physicians should be familiar with this condition and understand its pathophysiology.

It is often of concern to the attending clinician whether the pneumomediastinum has arisen from oesophageal rupture, as the latter carries high morbidity and mortality. Therefore, patients with spontaneous pneumomediastinum of various aetiologies are often subjected to further investigations and treatment for suspected oesophageal perforation. We review this approach and present a case where pneumomediastinum coincided with pneumopericardium and a small unilateral apical pneumothorax which has not previously been described.

CASE PRESENTATION

A 20-year-old primigravida developed mild retrosternal chest and neck pain associated with neck swelling 5 h after a prolonged, assisted vaginal delivery.

Her medical history included eczema and mild asthma; she was a non-smoker and her pregnancy had been uneventful.

She presented at 41 weeks and 3 days' gestation in early spontaneous labour. She had a strong contractions throughout; however, her progress was slow and she required augmentation by artificial rupture of the membranes. Seven hours after presentation full dilatation was reached and active pushing began. She received 50 mg of pethidine and

50 mg of cyclizine. No nausea or vomiting was reported. She used entonox intermittently. An episiotomy was performed, but labour continued to progress slowly and at approximately two and a half hours of active pushing signs of fetal distress were detected. This prompted an immediate attempts at vacuum-assisted delivery which were unsuccessful and the baby was finally delivered using forceps.

The baby girl was live, weighed 3225 g but required immediate resuscitation. Placenta and membranes were delivered by controlled cord traction. No postpartum haemorrhage occurred.

Five hours later, the patient reported mild retrosternal chest pain associated with discomfort and swelling in the neck region. Clinical assessment revealed no obvious cause of the symptoms and simple analgesia was administered for the discomfort.

Chest and neck pain occurred again on the following day and was accompanied by mild, transient breathlessness associated with a tachycardia of 130 beats per minute and mild tachypnoea at 20 breaths per minute. Blood pressure remained stable at 125/81 mm Hg and saturation were 96% on room air.

A chest x-ray was requested and clinical review by the acute medical team instituted.

At this time she was noted to have surgical emphysema palpable over the anterior neck, both supraclavicular fossae and the anterior chest wall. Lung fields were otherwise clear on auscultation with good bilateral air entry.

INVESTIGATIONS

ECG revealed sinus tachycardia with no other abnormalities and arterial blood gas analysis was normal.

Figure 1 AP chest x-ray demonstrating surgical emphysema, small left apical pneumothorax (1) and pneumomediastinum (2).

A chest x-ray confirmed pneumomediastinum and suggested a small left apical pneumothorax. The view was obscured by overlying surgical emphysema (figure 1).

The patient was placed nil by mouth and the following morning a CT thorax and barium contrast swallow were performed in order to exclude a spontaneous oesophageal rupture as the cause of pneumomediastinum.

A CT confirmed chest x-ray findings and in addition revealed a small pneumopericardium. There was no evidence of free fluid in the mediastinum, pleural effusion or any other sign of oesophageal rupture (figure 2). Barium oesophagogram was normal (figure 3).

DIFFERENTIAL DIAGNOSIS

Causes of pneumomediastinum include; Hamman's syndrome (spontaneous pneumomediastinum due to alveolar rupture), oesophageal rupture, pneumothorax with pleuro-mediastinal communication, intra-abdominal hollow viscus perforation with an associated hiatal defect, tracheo-oesophageal fistula with an air leak and mediastinal blunt force trauma with an airway injury.

TREATMENT

No treatment was required.

OUTCOME AND FOLLOW-UP

Normal oral intake was re-established and she was regularly clinically reviewed. Her pain settled, and tachycardia resolved. The white cell count fell from 17.2 after labour to 15.7. Her chest and neck discomfort improved and the subcutaneous emphysema remained stable. Transfer to another hospital, where her baby was receiving additional care was facilitated.

She was reviewed at 5 weeks and complete recovery was noted.

DISCUSSION

Since it was first formally reported by Simmons⁴ in 1793 and then described by Hamman⁵ in 1945, spontaneous pneumomediastinum remains an unusual phenomenon in labour. It usually occurs in the second stage of labour^{1 4 6} and is much uncommon in the first stage.⁷ The signs and symptoms usually develop some time after the delivery.

Clinical manifestation include retrosternal chest pain often radiating to the neck, mild dyspnoea and anxiety. Dysphagia, tachycardia, transient leucocytosis and fever may occur.³ Surgical emphysema and Hamman's sign (a loud harsh sound over the precordium in systole) are pathognomonic.

Figure 2 CT thorax demonstrating surgical emphysema (1), pneumomediastinum (2) and pneumopericardium (3).

In this case the presenting symptoms were typical, but recognition was delayed. The diagnosis was established over 24 h after the delivery. However, the patient already reported first symptoms indicating development of surgical emphysema 5 h following the childbirth and it is very likely that pneumomediastinum occurred much earlier in the second stage of labour.

There is no consensus on the predisposing factors leading to spontaneous pneumomediastinum in labour. Gordon,⁶ who published first review of 130 cases stated that most often the patient was a healthy primigravida with a long labour. Reader *et al*² observed that, although 95% of women were primiparus the duration of labour was normal and the babies were of average size.

Our case is consistent with the view that first pregnancy and prolonged, difficult labour carry a higher risk of this complication.

The chest x-ray is usually diagnostic. However, the interpretation may be occasionally difficult in cases of extensive subcutaneous emphysema.

In this instance, despite overlying surgical emphysema the chest x-ray clearly demonstrated both pneumomediastinum and tiny left apical pneumothorax. We acknowledge that CT has not contributed further to the management and in retrospective could have been avoided.

The pathophysiology of spontaneous pneumomediastinum relates to Valsalva manoeuvre repeatedly generated during labour. Increased intra-alveolar pressure causes rupture of peripheral alveoli into perivascular tissue planes, with tracking of air into the mediastinum or pericardium.^{5 6} Air subsequently dissects through facial planes in the neck and chest wall. This mitigates rises in intramediastinal pressure which could compromise venous return in the great veins and result in haemodynamic instability. Therefore, tension pneumomediastinum or tension pneumopericardium in Hamman's syndrome is very rare.

Only a single case manifesting as a tension pneumomediastinum with cardiovascular instability in the second stage of labour has been reported.⁸ The patient recovered following intravenous fluid resuscitation and no surgical intervention was required. Nevertheless, this rare, but potentially fatal complication should be noted.

Pneumothorax can originate either from an independent rupture of peripheral alveoli or result from extension of free air from mediastinum to the pleural space.^{2 3}

Coexistence of spontaneous pneumomediastinum, pneumopericardium and pneumothorax is uncommon and to our knowledge has not been previously reported in labour. However, we suspect this is because of underdiagnosis as

Figure 3 Normal barium oesophagogram.

pneumopericardium may not always be apparent on the plain chest x-ray and CT is not necessarily undertaken.

Although, the pathogenesis of pneumomediastinum and surgical emphysema outlined above has been widely accepted, establishing the final diagnosis for the clinician unfamiliar with the condition often poses a clinical dilemma.

The most common concern is missing an oesophageal rupture, a condition which itself can lead to pneumomediastinum and carries a high morbidity and mortality if not diagnosed early. Many case reports reflect this and often oesophageal contrast studies and occasionally CT have been performed.^{8 9}

A management strategy we followed also confirms this practice.

Interestingly our observations seem to be shared by others who investigated the occurrence of spontaneous pneumomediastinum in different cohorts of patients. Chapdelaine¹⁰ found no oesophageal perforation in a group of 53 paediatric patients and advised caution when requesting further radiological investigations. Similarly, Abolnik¹¹ who studied an older group of patients with a mean age of 18.8 years found no incidence of oesophageal perforation, but 24% of patients were investigated with contrast swallow.

Some investigators believe that the oesophagus in young patients, unlike the alveoli, is more tolerant of sudden pressure changes.¹²

Spontaneous oesophageal rupture is a rare event in labour. If it occurs it is usually a consequence of repeated vomiting and only two confirmed cases of oesophageal perforation in labour are reported both of which describe preceding vomiting.^{13 14}

Investigations in this area can be difficult. A contrast study particularly in a case of small oesophageal tear may be normal. Endoscopy may be useful in the management of confirmed tears but may not be sufficiently sensitive to make the diagnosis.¹⁵

We would recommend plain posteroanterior and lateral chest radiographs which can strongly suggest the diagnosis. Effusion in the mediastinum, a pleural effusion, hydropneumothorax, basal consolidations, emphysema located posteriorly and inferiorly along the lateral edge of the aorta and beneath the parietal pleura over the left hemidiaphragm are more suggestive of oesophageal perforation.^{15 16} In contrast, surgical emphysema or pneumomediastinum alone are present in only 27% of patients with oesophageal rupture, whereas the chest x-ray was abnormal in 97% of cases.¹⁷ Persistent fever, leucocytosis and systemic sepsis would strongly indicate acute mediastinitis

while a transient leucocytosis alone may be a feature of surgical emphysema and pneumomediastinum.³

In conclusion, surgical emphysema and spontaneous pneumomediastinum are very rare events in labour. Oesophageal rupture is exceedingly unusual particularly in the absence of a history of vomiting when plain chest radiograph is otherwise normal and there are no signs of sepsis. Current evidence does not support the routine use of contrast enhanced CT or a contrast oesophagography. However, careful clinical assessment including a thorough history of the circumstances leading to a pneumomediastinum should be taken. We suggest that the risks of radiation exposure in a childbearing age woman should be carefully considered and the routine use of contrast enhanced CT with or without contrast swallow studies is not necessarily indicated in Hamman's syndrome.

Learning points

- ▶ Spontaneous pneumomediastinum during labour is a rare condition. However, both physicians and obstetricians should be familiar with its pathophysiology and management.
- ▶ It is usually caused by a rupture of the alveoli.
- ▶ Spontaneous oesophageal rupture in labour is extremely rare.
- ▶ Unless strong clinical suspicion exists, routine investigations and or treatment of suspected oesophageal tear are unnecessary.

Competing interests None.

Patient consent Obtained.

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