CASE REPORT

Intrapartum spontaneous pneumomediastinum and surgical emphysema (Hamman's syndrome) in a 30-year-old woman with asthma

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SUMMARY

The authors report a case of a 30-year-old nulliparous woman with a prior medical history of asthma who developed intrapartum pneumomediastinum with surgical emphysema during the second stage of labour. She was managed conservatively with supportive care and there was complete resolution of symptoms within 4 days. Clinical and radiological findings were consistent with Hamman's syndrome (spontaneous pneumomediastinum). This case reiterates the possibility of childhood asthma or a family history of asthma as being an associated risk factor for Hamman's syndrome and reminds us of the need to distinguish such clinical findings from more important and potentially life-threatening conditions such as oesophageal rupture, pulmonary embolism and aortic dissection that can present with similar findings.

BACKGROUND

Intrapartum subcutaneous emphysema is a rare phenomenon. This case presents an interesting learning opportunity for clinicians to refresh their understanding of the presentation and pathophysiology of pneumomediastinum causing surgical emphysema during labour by analysis of the patient's presenting clinical history and pertinent prior medical history. We note the patient's prior medical history of asthma, and reiterate a strong likelihood of association between asthma and Hamman's syndrome.

CASE PRESENTATION

A 30-year-old primigravid woman with a prior medical history of childhood asthma and a strong family history of asthma was induced at 41 weeks. There was no progression of the second stage of labour and the patient had a forceps-assisted delivery approximately 13 hours later. During the initial phases of the second stage, while straining the patient heard a 'pop' sound, followed by periorbital swelling which swiftly spread over her face (figure 1), neck up to the level of T4. She noticed her voice changed and had a nasal twang to it and heard a 'crunching' sound on movement. On examination, crepitations could be heard on auscultation of the chest and felt on palpation.

After 13 hours of labour, the patient delivered a healthy male baby, weighing 3.8 kg. Her symptoms began to resolve approximately half an hour after

delivery. She was managed conservatively and there was complete resolution of symptoms 4 days after delivery.

Except for pregnancy-induced hypertension, she had no other medical history of note. Her highest recorded blood pressure during labour was 130/92 mm Hg, which did not warrant treatment. She gave a history of occasional inhaler use up to the age of 7 and a strong family history of asthma.

INVESTIGATIONS

Her immediate postpartum vitals were blood pressure 124/79 mm Hg, oxygen saturation 94%, heart rate 102 and respiratory rate 16. The Apgar score was 9/10 and after 5 min it was 10/10. Her cardiotocography was normal and reassuring.

Chest x-ray, 1 September 2016, 11:44

There is bilateral subcutaneous emphysema and some bronchial wall thickening that could be related with inflammatory disease. No evidence of pneumothorax and no clear features of pneumomediastinum (figure 2).

CT, 1 September 2016, 13:04

There is dramatic appearance of subcutaneous emphysema extending all the way from the scalp to the lowest visualised section of the upper abdomen (figure 3 and 4). There is also pneumomediastinum (figure 5 and 6). No evidence of mediastinal collection seen. No significant pneumothorax. No mediastinal shift. No evidence of intra-abdominal free air. The lungs appear unremarkable. No evidence of cysts or bullae. Findings could be part of Hamman's syndrome.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis at the onset of symptoms included pulmonary embolism, oesophageal rupture, pneumomediastinum, aortic dissection, pneumothorax, pneumopericardium and myocardial infarction. Initial chest X-ray radiograph was inconclusive, but a definitive diagnosis of pneumomediastinum with surgical emphysema was made after CT scan.

TREATMENT

Labour was induced and assisted by forceps. The patient was administered oxygen via nasal cannulae and was closely monitored in the postpartum

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Figure 1 Puffiness of the face and eyes due to surgical emphysema in Hamman's syndrome.

Figure 2 A chest X-ray showing bilateral subcutaneous emphysema and some bronchial wall thickening that could be related with inflammatory disease. No evidence of pneumothorax and no clear features of pneumomediastinum.

Figure 3 A CT showing dramatic subcutaneous emphysema and pneumomediastinum.

period by both obstetric and general surgical teams. Her pain was controlled through a pudendal block and Entonox.

OUTCOME AND FOLLOW-UP

Symptoms began to resolve half an hour after delivery and complete resolution was achieved within 4 days with conservative

Figure 4 A CT showing dramatic appearance of subcutaneous emphysema extending all the way from the scalp to the lowest visualised section of the upper abdomen.

Figure 5 A CT showing subcutaneous emphysema with pneumomediastinum extending up to the lowest visualised section of the upper abdomen. No evidence of mediastinal collection, no significant pneumothorax. No evidence of intra-abdominal free air. No evidence of cysts or bullae.

management and supportive care. She was discharged after 4 days and at outpatient clinic follow-up 6 weeks later, she reported that she was completely asymptomatic.

DISCUSSION

Spontaneous pneumomediastinum during labour was first described by Simmons¹ in 1793. It describes a condition where air is found within the mediastinal space, due to trauma or underlying pathological process. Pneumomediastinum with accompanying surgical emphysema, however, was first described by Louis Hamman of Johns Hopkins Hospital in 1945.² The condition is very rare, and has an approximate incidence of 1 in 100 000 cases.³ Approximately 200 cases have been thus far reported in the literature.

Hamman's syndrome is thought to most commonly affect nulliparous women, and pathophysiology is thought to occur during the second stage of labour, when intra-alveolar pressures are the highest.¹ However, clinical presentation is often delayed and usually occurs in latter stage of labour or in the postpartum period. This is not always the case however, as illustrated by our case report, where the onset of symptoms occurred during the second stage of labour. Typical presenting symptoms include chest pain, dyspnoea, dysphonia and swelling of the skin with associated crepitations on palpation or movement.

It is of vital clinical importance, when interpreting these symptoms, to exclude more serious differential diagnoses when considering the usually benign and self-limiting Hamman's syndrome. Spontaneous oesophageal rupture (distinguished from Boerhaave syndrome, reserved for cases of oesophageal rupture due to intense vomiting or retching), pneumothorax, aortic dissection, myocardial infarction and pulmonary embolism can often present with the same spectrum of symptoms, and

Figure 6 A CT showing pneumomediastinum and subcutaneous emphysema.

are potentially life-threatening conditions that need immediate medical and/or surgical attention. It is also noted that facial asymmetry due to extensive surgical emphysema may mandate the need to exclude cerebrovascular accident as a cause of symptoms.⁴

There are different theories which explain the pathophysiology of this condition. It is most likely that rupture of the peripheral alveoli due to high intra-alveolar pressures in the second stages of labour causes interstitial emphysema. This can be secondary to coughing and bronchospasm associated with asthma and the Valsalva manoeuvre as a result of pushing and vomiting during labour. This free air subsequently traverses along the perivascular connective tissue down a pressure gradient created by excess intrathoracic pressure into the mediastinum.¹ The air then spreads into subcutaneous tissues,⁵ causing the typical appearance of swelling of the face, neck and chest seen in this condition.

Learning points

- ▶ Pneumomediastinum and accompanying surgical emphysema (Hamman's syndrome) is a very rare complication of labour.
- ▶ Hamman's syndrome is usually a benign and self-limiting condition, although must be differentiated from more complex and serious conditions such as oesophageal rupture.
- ▶ Chest X-ray is the first line imaging modality for definitive diagnosis, although chest CT is required when X-ray is inconclusive.
- ▶ Where diagnosis is confirmed radiologically, treatment is usually conservative (analgesia, oxygen, monitoring).
- ▶ A prior medical history of asthma indicates a possible higher likelihood of the incidence of Hamman's syndrome during labour, likely due to bronchospasm contributing to the rupture of peripheral alveoli.

Also of importance is malignant mediastinum, a very rare complication of massive pneumomediastinum involving very high mediastinal pressures resulting in shock. This condition requires surgical intervention often involving splitting of the sternum in order to release trapped air. Without surgical intervention, this condition can lead to fatality.⁶

Chest X-ray is deemed the most important radiological investigation in confirming diagnosis, however, as illustrated in this case, often cannot rule out a diagnosis of spontaneous pneumomediastinum. In such cases, chest CT is required for more detailed imaging and is of greater utility in detecting surgical emphysema.

Treatment of Hamman's syndrome is usually conservative. Supportive management with oxygen therapy, analgesia, reassurance and bed rest is indicated to manage the chest pain, dyspnoea and anxiety that is often experienced. Symptoms usually resolve within a few days without further complication. Recurrence is uncommon, although not formally studied due to the rare nature of this complication of labour. There is no substantive evidence of appropriate management in subsequent pregnancies.⁷

Contributors JM helped in the planning and conduct of the article; he along with MS identified the diagnosis. DBN was behind the reporting, obtaining the data, conception, design of the article and interpretation of data. MDR worked on the analysis, reporting, interpretation and helped draft the article. MS made the radiological diagnosis, procured the images and provided analysis of the images.

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