Spontaneous pneumomediastinum with severe subcutaneous emphysema secondary to prolonged labor during normal vaginal delivery

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A healthy 33-year-old primiparous woman developed sudden central, nonradiating chest pain and right-sided neck and facial swelling during a prolonged second stage of labor. The main finding was subcutaneous emphysema involving the neck and face. Spontaneous pneumomediastinum (SPM) is a rare cause of chest pain and can lead to subcutaneous emphysema during labor. It is probable that SPM occurred during the second stage, due to prolonged and strenuous valsalva maneuvers. Subcutaneous emphysema can accumulate, enlarge, and cause upper-airway obstruction in the neck.

Case report
A 33-year-old Caucasian gravid 1 para 0 was admitted at 40 weeks to the birthing unit in early labor. She had no significant medical history and (apart from pregnancy-induced hypertension) had an uneventful antenatal course. Twelve hours after onset of labor, she progressed to full dilation, with a vertex presentation, in the left-occiput-anterior position. Her second stage of labor was prolonged, and minimal progress was documented at two hours. It was at this point that the patient complained of central chest pain.

On examination, swelling and crepitus was noted over the right side of her face and neck, extending over the anterior chest wall. Her chest was not hyper-resonant to percussion. Her voice was normal, and there was no tracheal deviation or stridor. Blood pressure, pulse rate, and oxygen saturations remained within normal limits. The patient continued to push with contractions, though progress remained minimal.

An initial mobile chest x-ray (CXR) (Fig. 1A) identified bilateral subcutaneous emphysema of the neck with no evidence of pneumothorax or pneumomediastinum. Due to SPM, the patient subsequently underwent an urgent

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Figure 1A. 33-year-old woman with spontaneous pneumomediastinum. Chest X-ray done during labor, before emergency cesarean section, showing subcutaneous emphysema in the neck (arrows).
cesarean section under spinal anesthesia. A postdelivery double-contrast CT of the neck and chest (Fig. 2) on day 1 detected a pneumomediastinum with tracking of subcutaneous emphysema extending from her face to the anterior and posterior thoracic walls bilaterally. There were no pulmonary lesions, bullae, or pneumothorax. There was no extravasation of oral contrast to demonstrate suggest esophageal perforation, and a gastro-graffin swallow performed day 1 after cesarean section confirmed that there was no esophageal breach. Laryngoscopy with a flexible bronchoscope did not reveal mucosal lacerations of the endolarynx.

The patient was monitored closely after delivery. Conservative management (bedrest and analgesia) led to a gradual resolution of subcutaneous emphysema without complications. She was discharged on day 3 after cesarean section.

**Discussion**

SPM in the postpartum period is rare (1). In one case study, SPM developed three hours postpartum (1) and 12 hours postpartum in a further case study (2). SPM in the setting of labor was first described by Louis Hamman in 1939 (3). He noted an audible crackling sound occurring with each heartbeat. This condition is extremely rare and has an incidence between 1 in 29,670 (4) and 1 in 44,511 (5). Hamman's Syndrome (postpartum pneumomediastinum associated with vaginal deliveries) is rare, with approximately 200 cases reported worldwide (6). Common presenting symptoms of SPM include retrosternal chest pain, dyspnoea, pleurisy, facial and neck pain, odynophagia, or dysphagia (which can be secondary to pressurized air from alveolar rupture tracking between tis-
Chest pain in labor and pregnancy may be due to a number of causes. Since SPM is extremely rare, it is essential to exclude pulmonary embolism, amniotic-fluid embolism, myocardial infarction, pneumothorax, and aortic dissection before making this unusual diagnosis.

Risk factors for pneumomediastinum include smoking and cocaine use (2, 5). Actions causing prolonged and strenuous valsalva such as coughing, sneezing, defecating, birthing, and vomiting have led to SPM, and patients with respiratory diseases such as asthma or chronic obstructive airways disease are particularly at increased risk. Being primiparous and having a prolonged second stage of labor increase the likelihood of prolonged valsalva and hence SPM. Boorhaeve’s Syndrome (esophageal rupture from vomiting during pregnancy) has been associated with SPM. This syndrome presents with severe retching and vomiting leading to excruciating retrosternal chest pain, upper abdominal pain, odynophagia, tachypnea, dyspnea, cyanosis, fever, and shock. The patient had none of the symptoms that would suggest Boorhaeve’s Syndrome as the cause of her SPM.

To our knowledge, this is the first documented case of SPM with severe subcutaneous emphysema diagnosed in the second stage of labor and requiring an urgent cesarean section. It is likely that this patient developed a pneumomediastinum following prolonged and strenuous valsalva during her extended second stage of labor.

A thorough history and clinical examination and CXR is often enough to diagnose SPM (8). 30% of SPM cases may not be easily seen on routine CXR. Double-contrast CT imaging may be helpful, particularly if the air-leak involves the posterior mediastinum (1, 9). Interestingly in the described case, the initial CXR showed subcutaneous emphysema only of the neck regions, whereas the subsequent double-contrast CT of the neck and chest showed extensive subcutaneous emphysema of the anterior thoracic wall, neck, and face. The followup CXR (Fig. 1B) reported evidence of a pneumomediastinum, suggesting postdelivery progression of SPM.

SPM is often a benign and self-limiting condition. It is managed conservatively with bedrest, observation, and analgesia. Other complications of SPM such as tension pneumomediastinum and pneumothorax are rare. There is currently no reliable data to suggest the rate of recurrence of SPM in pregnancy (1). The use of nitrous oxide increases the risk of gas trapping and may increase the risk of SPM (10). In patients with a history of SPM, it may therefore be prudent to avoid the use of nitrous oxide. While the second stage of labor is generally shorter for multiparous patients, epidural analgesia may be discussed as a method of avoiding premature or prolonged pushing in the late first or early second stages of labor. If SPM is recognized early, it is felt that delivery should be hastened (for example, by Ventouse, forceps, or urgent cesarean section) to minimize prolonged or strenuous valsalva and progression of mediastinal emphysema.
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