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Not All Swelling Is Edema in Eclampsia: A Rare and Life-threatening Potential Complication of Eclamptic Seizures

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SUBCUTANEOUS emphysema (SE) with pneumomediastinum or pneumothoraces is a rare but potentially lethal complication of second-stage labor. This condition was first described as *Hamman syndrome* in 1945 and is thought to occur secondary to abnormally high intrathoracic pressure from the repeated forceful Valsalva maneuvers during parturition.[1,2] To our knowledge, Hamman syndrome has not been reported in or associated with eclampsia or parturients without active labor. We report a first case of SE and pneumothoraces presenting as facial–cervical swelling in an eclamptic patient undergoing cesarean delivery without active labor.

Case Report

A 24-yr-old, 65-kg, 152-cm primigravida woman with severe preeclampsia and a history of asthma was admitted at 38 weeks' gestation for magnesium administration and oxytocin induction of labor. Her laboratory studies and physical examination were within normal limits except for proteinuria, hypertension, and moderate generalized edema without obvious facial edema at admission. Because of her preeclampsia, an epidural catheter was easily inserted early before onset of active labor or pain. Three hours later, the patient had a witnessed tonic–clonic eclamptic seizure, which subsided shortly after administration of 4 mg intravenous lorazepam and assisted mask ventilation. Because of recurring fetal heart rate deceleration, an urgent cesarean delivery was planned. However, the patient was combatively uncooperative despite a bilateral T4 anesthetic level achieved epidurally while her oxygen saturation remained 100% and blood pressure was 140/85 mmHg. The patient was noted to have a Mallampati class I airway with moderate generalized edema and mild facial edema. Rapid sequence general anesthesia induction was performed with successful atraumatic intubation on the first attempt, followed by verification of bilateral symmetrical breath sounds. Maintenance of anesthetic consisted of volume-controlled mechanical ventilation, 50% oxygen, 50% nitrous oxide, 1–2% sevoflurane, midazolam, and opioid without the need for additional neuromuscular blocker other than succinylcholine at induction. Intraoperative blood pressure and heart rate were maintained within 20% of preoperative values while oxygen saturation was maintained at 99–100%, end-tidal carbon dioxide between 30–35 mmHg, and airway pressures less than 25 cm H_2O. Cesarean surgery was completed without complications, and a healthy infant was delivered. On emergence from anesthesia, the patient was hemodynamically stable with adequate spontaneous ventilation while maintaining an oxygen saturation of 99–100% and an end-tidal carbon dioxide of 35–45 mmHg. However, she did not respond purposefully and had increased facial edema extending to the neck. The decision was made to transfer her to the recovery room to remain intubated on an Ayres T-piece until she followed commands. During transport, the patient might have pushed the oral airway out of her mouth. Upon arrival in the recovery room, she developed another tonic–clonic eclamptic seizure lasting less than 1 min, and she bit her tongue and the endotracheal tube momentarily for less than 10 s, during which time no obvious voluntary respiratory effort was observed. On further examination, her edema was noted to be present on the anterior chest and upper abdomen with crepitus on palpation. Crackling sounds synchronous with cardiac auscultation were noted, as well as symmetrical but decreased breath sounds in bilateral upper lung fields, suggestive of pneumothoraces. A chest x-ray revealed a pneumothorax of 30% on the right side and 15% on the left, in which chest tubes were inserted by a cardiothoracic surgeon to prevent potential acute deterioration. She was extubated the first day, and both chest tubes were removed 2 days later. Follow-up chest x-ray showed resolution of pneumothoraces, and she was discharged home on her fourth postoperative day without any permanent sequelae.

Discussion

Subcutaneous emphysema by itself may be benign, but the underlying etiology may be a life-threatening emergency. SE with or without pneumomediastinum or pneumothoraces is a rare complication of active second-stage labor with an estimated incidence as low as 1:100,000.[3] It was first referenced in 1618, when Louise Bourgeois, midwife to the French Queen, wrote, "I saw that she tried to stop crying out and I implored her not to stop for fear that her neck might swell,"[4] and was subsequently better described and named as Hamman syndrome in 1945.[2]

The initiating cause of the SE and pneumothoraces in our case was likely the result of complications of eclamptic seizures and/or anesthesia. During seizures, pulmonary complications such as aspiration and neurogenic pulmonary edema are well described,[5] but pneumothoraces, pneumomediastinum, and SE are rarely reported and poorly described if reported.[6] The likely mechanism was related to increased intraalveolar pressure generated by expiratory effort against a closed glottis during seizure activity leading to the rupture of marginally situated alveoli into the perivascular tissue planes, then with air dissecting to the mediastinum and subsequently to the tissue planes of the neck, face, and anterior chest presenting as SE.[6] The presence of SE might actually reduce

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the pressure buildup in the mediastinum that could lead to life-threatening cardiac tamponade or impediment of thoracic venous return from pneumomediastinum. Oral tissue trauma, which can occur during seizures as well as in dental surgery, has also been reported to cause similar complications.[7] The presence of airspace disease in combination with the eclamptic seizures might have predisposed our patient to this rare complication. The contributory factor from general anesthesia would most likely be the result of an abnormally high intrathoracic pressure, a previously well-described mechanism,[7] either from positive-pressure ventilation or from coughing or expiring against an occluded endotracheal tube. Other anesthesia-related causes of pneumothoraces and SE such as esophageal or hypopharyngeal perforation from traumatic intubation/laryngoscopy or the use of air in epidural techniques[8] seemed unlikely in our case because the patient's inductions were accomplished without difficulty.

Our presumption of the facial–cervical swelling to be the usual edema of eclampsia illustrates the importance of avoidance of preconceived bias in making a diagnosis, attention to details, and examining patients after eclamptic seizures more closely than just monitoring for hemodynamic, respiratory functions, and mental status. If we had better examined and recognized the crepitus earlier, we would have avoided general anesthesia, positive-pressure ventilation, nitrous oxide, and Valsalva maneuvers.

For pneumothoraces with or without SE or pneumomediastinum, awake patients may report pleuritic chest pain and shortness of breath, cough, and hemoptysis, all of which may be confused with pulmonary edema or embolism in the puerperium. Closer examination in both anesthetized or awake patients may reveal hyperresonance or decrease of breath sounds, distant heart sound, crepitus on palpation of swelling areas, or the Hamman sign, a fine auscultatory crepitation synchronous with heartbeat in the presence of air in the mediastinum. SE localized over the clavicles or anterior neck may indicate pneumomediastinum, whereas SE localized over the neck and anterior chest may suggest pneumothoraces. Tracheal deviation with hyperresonance in the lung fields, decrease or absence of breath sounds, and distention of neck veins with compromising cardiopulmonary functions may suggest tension pneumothoraces in which emergent needle decompression followed by tube thoracostomy may be needed. A lateral decubitus chest x-ray could aid and allow better visualization of smaller amount of air than upright or supine film in the diagnosis of pneumothoraces. Fortunately, SE and pneumomediastinum without pneumothoraces are usually benign and self-limiting, necessitating only conservative observatory management.

If Hamman sign occurs during active labor, general anesthesia and positive-pressure ventilation should be avoided if possible and epidural analgesia should be used to reduce a laboring patient's urges to have Valsalva maneuvers. A forceps-assisted second-stage delivery would seem appropriate to reduce maternal expulsive and Valsalva maneuvers. Management for subsequent delivery is less clear, but the evidence shows that 95% of the reported cases of Hamman syndrome were among primigravidas, with no recurrences reported to our knowledge.[1,9] Recurrence of the complication in our patient seems unlikely without a seizure and/or general anesthesia. In conclusion, this first case report of SE with pneumothoraces in association with eclamptic seizures and/or general anesthesia without active labor reminds us of this rare and life-threatening potential pulmonary complication of eclamptic seizures and the importance of its early diagnosis with closer physical examination in all patients after eclamptic seizures.

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