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Case Report

Hamman's Syndrome after Vaginal Delivery: A Case of Postpartum Spontaneous Pneumomediastinum with Subcutaneous Emphysema and Review of the Literature

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Abstract: Hamman's syndrome is a rare condition that mostly affects young males, often with a predisposition of asthma. It includes the presence of free air in the mediastinum and subcutaneous emphysema with no other underlying cause such as trauma or infection. It occurs spontaneously and often in association with prolonged Valsalva maneuver. This might explain why there are some cases of Hamman's syndrome among young females giving birth. Here we present a case report of a 24-year-old primigravida with this syndrome. She presented with symptoms a few hours after an uncomplicated vaginal delivery at 40+1 pregnancy week. The symptoms resolved spontaneously after a few days of observation. We also give a systemic review of reported cases since 2000 to provide overview on pathomechanism, symptoms, diagnostics, treatment and management of this condition. Hamman's syndrome is a rare, usually benign, but potentially serious complication that can occur during the second stage of labor. Diagnostics include inquiring about typical symptoms, clinical examination and chest x-ray or CT scan. Treatment is usually conservative with oxygen, bronchodilators and pain relief. The recurrence rate is low and there is no contraindication to vaginal delivery in future pregnancies. However, it is suggested that physicians and midwives be cautious and consider low threshold for instrumental delivery or caesarean section to avoid excessive Valsalva maneuver.

Keywords: delivery; Hamman's syndrome; pneumomediastinum; pregnancy

1. Introduction

Spontaneous pneumomediastinum (SPM) during labor is a rare event, occurring in about 1 in 100 000 deliveries. Together with subcutaneous emphysema it is called Hamman's syndrome [1]. The condition was named after Louis Hamman (1877-1946), the physician who described it in several case reports from 1939 to 1945 in postpartum women [2,3].

SPM is defined as the presence of free air in the mediastinum, with no underlying trauma and mostly affects young males and pregnant females. It may be associated with a pulse-synchronous crunching sound, referred to as the « Hamman's sign» best heard when the patient is lying in the left lateral decubitus position [4,5].

Hamman's syndrome may occur during prolonged labor, usually in the second stage, after prolonged Valsalva maneuver. Other predisposing events may be intensive coughing, retching/vomiting or physical activity [6].

The condition is usually benign and self-limiting, but in rare cases there may be complications such as significant dyspnea and chest pain, and even development of malignant pneumomediastinum, which requires surgical intervention [7]. In this paper, we report a case of Hamman's syndrome in a 24-year-old primigravida with underlying asthma, and we are reporting on the pathomechanism and management of this condition with regards on overview of 42 other published cases in the last two decades.

2. Case Presentation

A 24-year-old primiparous woman, with a normal pregnancy, presented to the maternity ward in spontaneous labor, with regular uterine contractions at 40+1 weeks. She was a non-smoker and denied drinking alcohol or using illicit drugs. Her body mass index was 27.1, and her past medical history was significant for depression, ADHD (Attention Deficit Hyperactivity Disorder) and childhood asthma, without need for medical treatment. There was no history of any heart condition.

In the latent phase of delivery, she received morphine for analgesia. The water broke spontaneously, and the amniotic fluid was discolored. The patient had normal temperature and there was no fetal tachycardia or other signs of fetal distress son CTG. The active phase of labor lasted for three hours with normal progress. After one hour of passive descent, she started pushing, and the second stage lasted for 30 minutes. It was a spontaneous vaginal delivery of a healthy baby with normal Apgar score (9-10-10) weighting 4170 g. There was a normal expulsion of the placenta, and there was normal bleeding.

Eight hours after delivery, she complained of pain in the right ear, swelling and pain in the neck, chest tightness, shortness of breath, dysphagia, odynophagia and pain in the upper thorax on the right side. Her vital signs were stable (BP 128/67 mmHg, pulse 91/min., temperature 36.0, respiratory rate 14/min., oxygen saturation 99% on room air). ECG was unremarkable. Blood gas showed normal values with pH 7.45, pCO₂ 4.2. PCR- test from nasopharynx was negative for viral infections. There were no findings on otoscopy. On palpation, there were subcutaneous crepitations in the neck, parasternal region, right axillary fossa, clavicle and over the chest. Auscultation of the heart and lungs was normal. A chest X-ray (Figure 1) was taken immediately, revealing subcutaneous emphysema extending bilaterally to the neck, but more prominent on the right side, and suspicious for pneumomediastinum. There were no signs of pneumothorax or esophageal or skeletal pathology. The heart configuration was normal. The patient received 1 g of paracetamol and 2.5 mg of morphine intravenously. The situation was clinically stable. The next day, a CT scan (Figure 2) without contrast was performed, confirming pneumomediastinum, with air extending from diaphragm up to the thoracic apexes and across larynx. There were discrete amounts of pleural fluid bilaterally, and subcutaneous emphysema from the base of the skull to the neck and upper thorax.

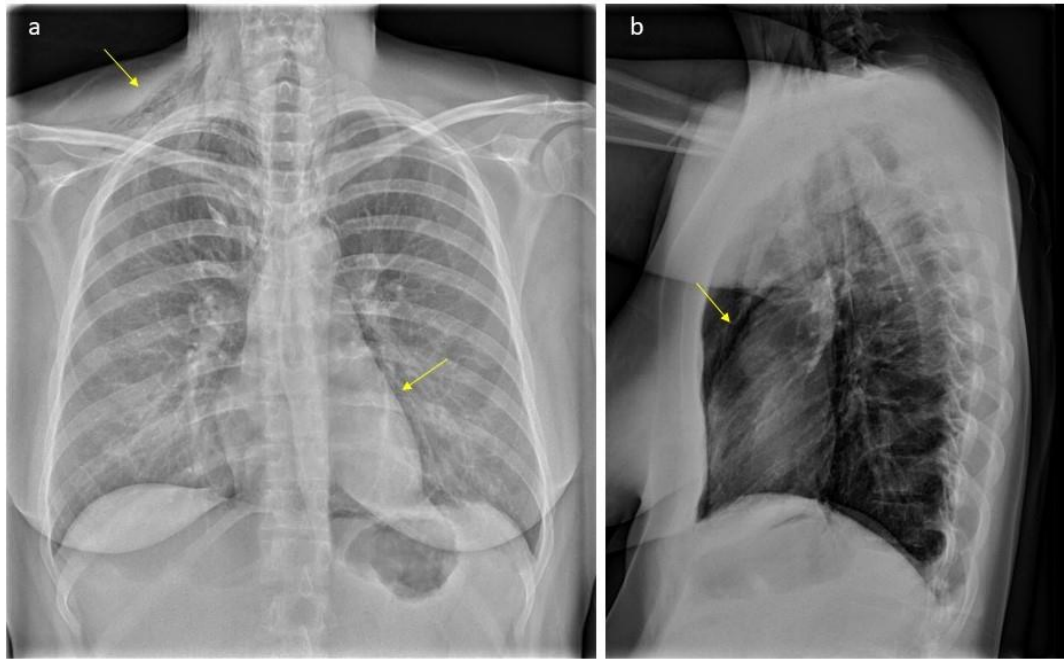


Figure 1. Plain radiograph scan showing typical gas accumulations seen subcutaneous and with pneumomediastinum (arrows, part A): There is significant subcutaneous emphysema, more pronounced on the right side (arrow, part A). We can see an outlining of the pericardium both in lateral projection (arrow, part B) and with «continuous diaphragm sign» on frontal projection (arrow, part A). You can also see continuous lucencies along upper mediastinum to the neck, through upper thoracic aperture.

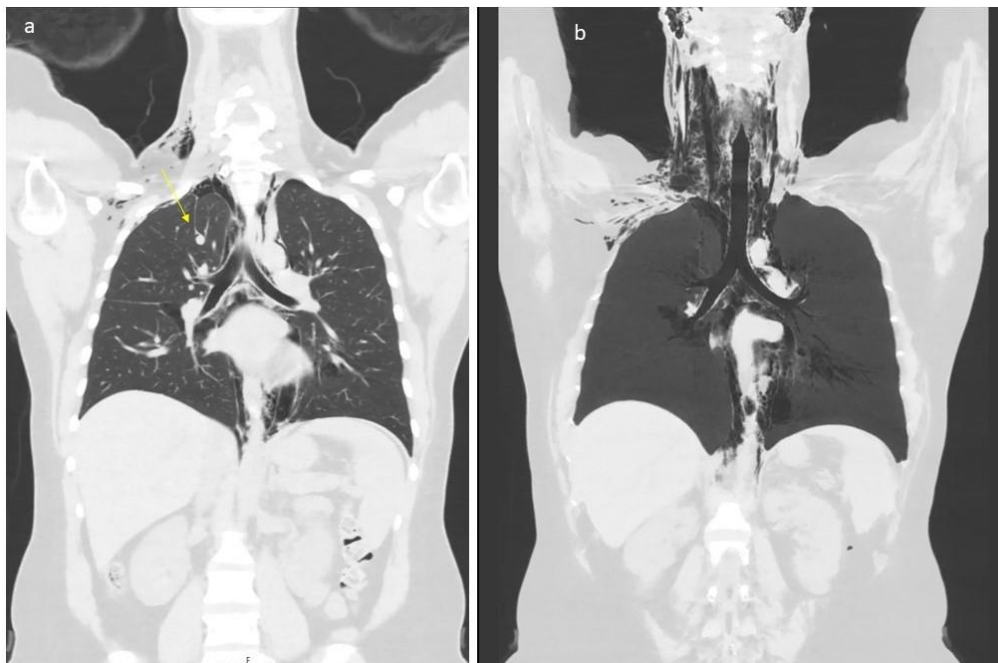


Figure 2. Low dose CT scan showing air within the mediastinum starting caudally in the level of hiatus aorticus, surrounding the pericardium, throughout the mediastinum. In the neck level, air was seen subcutaneously (part A), along the great vessels (carotid space) and in the retropharyngeal space (part B). The patient has an «azygos lobe», a normal variant with the vena azygos running laterally with a pleural fissure surrounding it (arrow, part A). Air bubbles can be seen along the vein and small amounts of air within the pleural cavity apically on the right side. Apart from this, no signs of

pneumothorax. Small amounts of pleural effusion are seen bilaterally. The upper abdomen was included in the low-dose scan, without any signs of air below the diaphragm.

The patient was reviewed by the medical team (radiologist, gynecologist and surgeon), and diagnosed with Hamman's syndrome. As the patient was hemodynamically stable, she was managed conservatively with observation and analgesics. Her symptoms resolved gradually over the next three days and she was discharged home on her third day postpartum. She was advised to avoid strenuous physical activity for the next four weeks. On follow up at the 6-weeks postnatal, she was in wellbeing and completely without symptoms. Follow-up correspondence was also done over the phone at five months. The patient had recovered well, and was now practicing normal physical activity.

3. Discussion

Hamman's syndrome is a rare clinical entity. Its incidence is 1:100 000 women giving birth. The incidence is higher in the case of accidents of emergencies and has a male predisposition, accounting for 76% of cases [8]. It is believed to be a result of a sudden increase in intra-alveolar pressure. Mostly it is associated with Valsalva maneuver, extensive vomiting, or coughing, all of which can occur in pregnancy and labor.

There have been several cases on Hamman's syndrome occurring in labor, but it has also been reported in association with other medical conditions, such as diabetic ketoacidosis with repeated vomiting or Kussmaul breathing [9,10] and bronchial asthma, with vomiting and coughing as common precipitating factors [11,12]. There has also been reports of SPM occurring after intense coughing during strenuous physical activity [13] or hyperemesis gravidarum [14].

In our case, extensive breathing during the first stage and intensive Valsalva maneuver in the second stage of labor in a patient with an underlying bronchial asthma, led to the development of symptoms of Hamman's syndrome after delivery. The CT scan of the chest was taken to exclude other severe diseases like pulmonary embolism, amniotic fluid embolism, myocardial infarction and Boerhaave syndrome.

The pathomechanism of Hamman's syndrome is usually explained as follows: during the Valsalva maneuver within expulsion phase of the delivery, the lung's intra-alveolar pressure is highly increased, what is leading to the rupture of small marginal alveoli adjacent to surrounding blood vessels. The liberated free air from ruptured alveoli spreads along parabronchial vascular structures towards the lung's hilum. From there, it extends proximally and may spread within the mediastinum and pericardium, upwards along the neck up to the head basis, or to the subcutaneous tissue in the upper thorax and sometimes downwards to the retroperitoneum. The anatomical absence of transverse fascial structures in the mediastinum allows the passage of air along tissue planes into the intern neck parts and around the larynx. The trapped air between the visceral and parietal pleura, may cause the pneumothorax. Fortunately, the pressure level of the interstitial air is not so high to cause severe respiratory compromise [1]. Intense screaming, vomiting, coughing, and stimulated pushing during labor, all together, can increase the intrathoracic pressure.

The most common symptoms of Hamman's syndrome are retrosternal and chest pain radiating to the neck or back, intense dyspnea, and swelling of neck, upper thorax and face. The typical pathognomonic sign of the condition is subcutaneous crepitus palpable in the neck and face [4,15]. Other symptoms include cough, sore throat, change of voice (dysphonia), dysphagia, hemoptysis and tachycardia. A characteristic sign is the bubbling or crunching sounds over the heart, synchronous with the cardiac cycle known as Hamman's sign or murmur. The occurrence of the symptoms, time onset after/during delivery, severity of the condition and management can be very variable, as it is described in our summarized overview on the reported cases over the last two decades (Table 1).

Table 1. An overview on 42 previously published cases of pregnancy associated Hamman's syndrome (database Pubmed.gov from 2000 to 3/2024).

Author	Age y/o	Parity	When symptoms developed	Duration of labor	Week of gestation	Treatment
Sutherland et al. 2002 [24]	32	Para 1	Postpartum	8h	N/A	None
Sutherland et al. 2002 [24]	22	Para 1	13h postpartum	N/A	N/A	None
Miguil et al. 2004 [25]	19	Para 0	N/A	N/A	40	Oxygen & analgesics, C-section
Duffy 2004 [26]	19	Para 0	2h	90 min 2 nd stage	40	Oxygen v analgesics
Bonin et al. 2006 [27]	27	Para 0	2 nd stage	6h	38	Lorazepam for anxiety & anxiolytics for dyspnea
Norzilawati et al. 2007 [28]	21	Para 0	12h postpartum	4h, 100 min 2 nd stage	40	None
Yadav et al. 2008 [29]	21	Para 0	2 nd stage	2 nd stage 1,5h	N/A	Oxygen & analgesics
Mahboob et al. 2008 [30]	24	Para 0	18h postpartum	N/A Normal	39	Oral antibiotics, iv fluids & analgesics
Zapardiel et al. 2009 [31]	29	Para 0	Postpartum	N/A	39	Oxygen
Revicky et al. 2010 [32]	32	Para 0	3h	14h	40	None
Beynon et al. 2011 [33]	18	Para 0	8h postpartum	4h	39	Antibiotics & analgesics
Wozniak et al. 2011 [34]	20	Para 0	5h postpartum	9h	41	Observation
Shrestha et al. 2011 [35]	19	Para 0	N/A	N/A	36	None
Kuruba et al. 2011 [1]	32	Para 1	2 nd stage	1.5h	40	None
McGregor et al. 2011 [36]	27	Para 0	2 nd stage	7,5h	40	Oxygen & analgesics
Houari et al. 2012 [37]	21	Para 0	Postpartum	N/A	40	Conservative management

Kandiah et al. 2013 [38]	25	Para 0	2 nd day postpartum	2 nd stage 3h, 16 min. Ending in a C-section	40	Observation
Kandiah et al. 2013 [38]	30	Para 0	2 nd stage	6h	38	Observation
Kouki et al. 2013 [39]	23	Para 0	2 nd stage	9h	40	Oxygen & analgesics and sedatives
Khoo et al. 2015 [40]	33	Para 0	2 nd stage	12h	40	Analgesics & bed rest
Cho et al. 2015 [7]	28	Para 0	2 nd stage	5h	36	Oxygen & analgesics
Wijesuriya et al. 2015 [41]	24	Para 0	N/A	N/A	N/A	N/A
Khurram et al. 2015 [4]	24	Para 1	2h postpartum	2 nd stage prolonged	40	None
Scala et al. 2016 [42]	30	N/A	2 nd stage	N/A	40	None
Elshirif et al. 2016 [43]	27	Para 0	4h postpartum	19h 2 nd stage 3h	41	Analgesics, oxygen & antibiotics
Berdai et al. 2017 [44]	22	Para 0	2 nd stage	2h	40	Oxygen
Lou et al. 2017 [45]	29	Para 0	2 nd stage	Prolonged	At term	Supportive
Sagar et. al 2018 [46]	22	Para 0	3h postpartum	4,5h	37	None
Khan et al. 2018 [47]	30	Para 0	N/A	N/A	N/A	Antibiotics, oxygen & bronchodilators
Jakes et al. 2019 [48]	23	Para 0	40 min postpartum	2 nd stage 2h	38	Oxygen
Madhok et al. 2019 [49]	21	Para 0	2h postpartum	3h	39	None
Lee et al. 2019 [50]	31	Para 0	2 nd stage	8,4h	41	IV antibiotics, hydrocortisone & Loratadine
Chavan et al. 2019 [51]	33	Para 0	10h postpartum	90 min 2 nd stage	38	Oxygen & analgesics

Opstelten et al. 2019 [52]	25	Para 0	2 nd stage	N/A	N/A	N/A
Oshovskyy et al. 2020 [53]	34	Para 4	2 nd stage	4,5h	39	Pigtail catheter
Badran et al. 2020 [54]	N/A	Para 0	4h postpartum	N/A	Full term	Nil by mouth
Zethner-Møller et al. 2021 [55]	35	Para 1	2 nd stage	N/A	36	Oxygen
Mullins et al. 2021 [56]	17	Para 0	postpartum, prolonged second stage	N/A	39	Oxygen & opioids
La Verde et al. 2022 [17]	23	Para 0	2 nd stage	5h	41	None
Gomes et al. 202 [27]	21	Para 0	2 nd stage	N/A	40	C-section & observation
Peña-Vega 2023 [57]	18	Para 0	30h postpartum	12h	39	Oxygen
Chooi et al. 2023 [58]	22	Para 0	2 nd stage	3h 2 nd stage	39	None
Hülsemann et al. 2023 [59]	21	Para 0	2 nd stage	Prolonged	N/A	N/A
Inesse et al. 2023 [60]	29	Para 0	1h postpartum	2 nd stage lasted 2h, 40 min active pushing	40	None
Chen et al. 2023 [61]	20	Para 0	immediately after delivery	Prolonged	43	Analgesics & antibiotics iv

Abbreviations: N/A - not available; IV – intravenous; h - hours.

In our case, the woman presented immediately after delivery with pain around the ear and the feeling of plugged ear. A few hours later she reported swelling of the neck, chest tightness and shortness of breath. On examination, the crepitus on the neck and thorax was obvious, but there was no typical Hamman's murmur.

Hamman's syndrome is usually a benign and non-recurrent condition, however, in rare cases it may be life threatening and lead to cardiac tamponade with significant hemodynamic compromise. Such situation requires surgical intervention [16]. Prior to any intervention, it is important to exclude other serious, potentially life-threatening conditions such as esophageal rupture (cancer related), Boerhaave syndrome (rupture of the esophagus due to forceful vomiting), pharyngeal rupture, pulmonary embolism, amniotic fluid embolism, aortic dissection, myocardial infarction, pneumopericardium, or pneumothorax of any cause [17,18].

Apart from the clinical picture, a CT scan is the gold standard in diagnosing pneumomediastinum. In a systematic review, it was found that about 30% of cases of

pneumomediastinum were poorly detected by chest x-ray, but were easily detected on a CT-scan [19]. The CT also provides more accurate information on the extension of subcutaneous emphysema and other thoracic pathologies. An additional tool that can be considered is diagnostic endoscopy (bronchoscopy or esophagoscopy) or esophagography [20].

Initial management is supportive treatment with sedatives, analgesics and oxygen, as needed. The purpose of oxygen therapy is to maintain oxygen saturation and to increase the resolution rate. The role of oxygen therapy is very important as high flow oxygen therapy, by breathing 90-100% oxygen using nasal cannula at 2–4 L/min, reduces the partial pressure of nitrogen in the alveolus compared with the pleural cavity, and a diffusion gradient for nitrogen accelerates resolution of pneumomediastinum by increasing the gas absorption gradient [21].

In severe cases, the treatment with antibiotics and bronchodilators, along with oxygen support may added. The patient should be reassured about good prognosis and expected spontaneous resolution within (3-14 days) [22]. Patients can be discharged if they are in good general condition and do not have a significant pneumothorax. There is no recommended routine follow up.

There have been a few reports of cases with spontaneous pneumomediastinum occurring in the setting of hyperemesis gravidarum in early pregnancy [14] or spontaneously in the third trimester. Here, the operative delivery should be considered to prevent worsening/recurrence of this condition [23].

4. Conclusion

Postpartum pneumomediastinum (Hamman's syndrome) is a rare complication however its timely diagnosis is necessary for patient safety and management. The most cases in pregnant women occur in the second stage of labor, as a result of excessive straining and Valsalva maneuver. The recurrence rate in subsequent pregnancies is low, and there are no established guidelines on the management. It is suggested that measures which can be implemented are to minimize barotrauma with the low threshold for instrumental or operative delivery, and approaches limiting the duration of the second stage of labor. However, this is not evidence based and may require meta-analytical approach.

Authors contributions: KOB: original draft preparation, clinical management; MMK: retrieving clinical data, database search, clinical management; KK: methodology, conceptualization; LJR: clinical management, original draft preparation; JV: writing, review and editing; PZ: writing and original draft preparation, clinical management, conceptualization, review and editing, supervision. All authors have read and agreed to the published version of the manuscript.

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Informed consent statement: Written informed consent was obtained from the patient. Under the all-management steps and in case presentation the principles of the Helsinki Declaration were followed.

Institutional review board statement: The presented study was carried out following the rules of the Declaration of Helsinki of 1975, revised in 2013. According to local IRB principles, ethical approval from IRB for a single case is not required, as long as data are kept anonymous and subject signed informed consent with the publishing.

Data availability statement: Data sharing nor applicable to this article as no datasets were generated or analyzed during the current study. The data presented in this study are available on request.

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