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Case report

Postmortem imaging identified pneumomediastinum in two cases of diabetic ketoacidosis

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ABSTRACT

Pneumomediastinum is defined as the presence of extraluminal gas within the mediastinum. Hamman described a diabetic ketoacidosis (DKA) patient with subcutaneous emphysema and spontaneous pneumomediastinum, a case known in current times as Hamman's syndrome and now pneumomediastinum is known to be one of the accompanying findings of DKA. We describe two cases with diabetes mellitus in which postmortem computed tomography (CT) could detect pneumomediastinum prior to autopsy. One was a woman in her 40 s, who had type II diabetes mellitus with bronchopneumonia in the middle and lower lobes of the right lung (Case 1) and the other also aged in her 40 s with type I diabetes mellitus (Case 2). In these cases with diabetic mellitus, the finding of pneumomediastinum at postmortem CT gave differential diagnostic considerations of DKA in addition to common cause such as decomposition or cardiopulmonary resuscitation. Autopsy with histopathological and biochemical examination suggested the cause of death in both cases were fatal DKA. The main cause of pneumomediastinum in Case 1 was pulmonary emphysema, and DKA was an exacerbation factor of pneumomediastinum. In contrast, DKA could have caused pneumomediastinum in Case 2, so called Hamman's syndrome. Pneumomediastinum is an obvious morphological abnormality, and thus we should pay attention to the information of pneumomediastinum from postmortem CT scanning.

1. Introduction

Pneumomediastinum, also described as mediastinal emphysema, is defined as the presence of extraluminal gas within the mediastinum, which is usually identifiable on chest radiography or computed tomography (CT) images. Although pneumomediastinum is a rare entity with estimated incidence in 1/100,000–1/44,500, it is believed that the occurrence of pneumomediastinum is more frequent than reported due to underdiagnosis, with many patients refraining from medical help [1,2].

Pneumomediastinum was first accurately described in 1819 by Laënnec et al. in the setting of trauma injuries [3]. On the other hand, spontaneous pneumomediastinum refers to the presence of gas in mediastinum in otherwise healthy subjects without an obvious causative factor such as an operation, air following a viscous perforation, presence of infection or trauma [4]. Spontaneous pneumomediastinum is also known as one of the accompanying findings of diabetic ketoacidosis (DKA) [5,6]. Hamman originally described a diabetic patient with ketoacidosis who had subcutaneous emphysema and spontaneous pneumomediastinum, a case known in current times as Hamman's syndrome [7,8]. Although the pathophysiology was not elucidated, a recent hypothesis states that acidotic 'Kussmaul breathing' associated with DKA might induce trans-alveolar pressure swings and cause alveolar rupture, resulting in air leakage along perivascular sheaths toward the mediastinum [6,9].

Here, we reported two cases with diabetes mellitus in which pneumomediastinum could be detected on postmortem CT scanning prior to autopsy. The presence of pneumomediastinum gave differential

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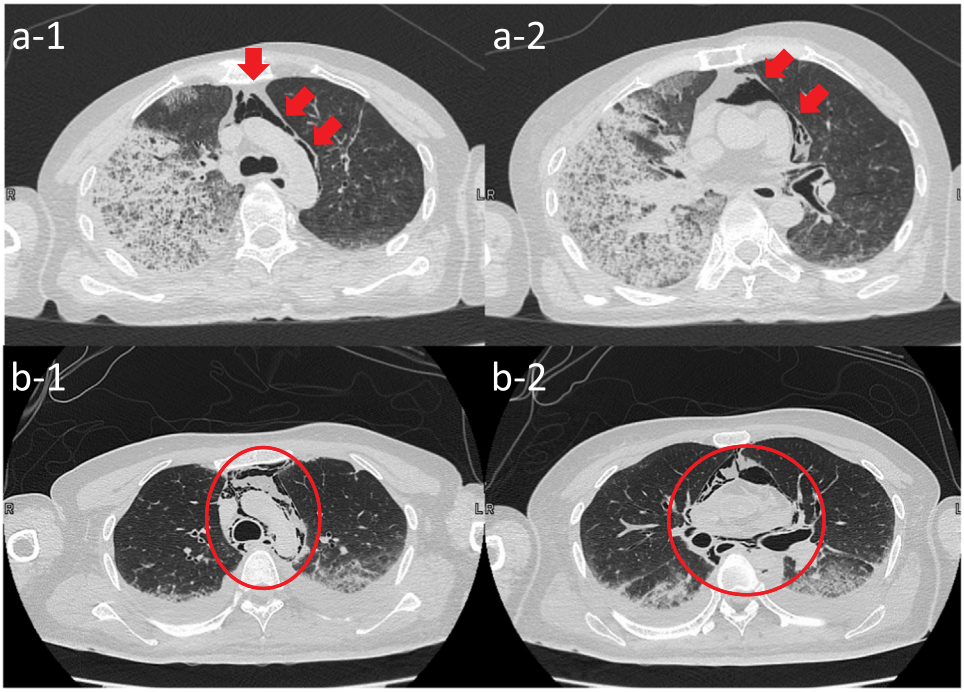

Fig. 1. Representative postmortem CT findings in Case 1 (a) and Case 2 (b). CT shows pneumomediastinum (a, arrows, b, circles). Chest CT also shows consolidation in the middle and lower lobes of the right lung (a-1).

diagnostic consideration of DKA in addition to common cause such as decomposition or cardiopulmonary resuscitation. Since to diagnose functional diseases including DKA as a cause of death is usually difficult, this morphological abnormality arising after DKA would be useful in the forensic environment.

2. Case presentation

2.1. Case 1

A woman in her 40 s with type II diabetes mellitus, alcoholic liver disease and chronic pancreatitis was found dead in her house. Cardiopulmonary resuscitation was not attempted. According to her caregiver, she had a poor appetite and had hardly eaten for a few days before her death. She has no habit of smoking and has no clinical history of pulmonary emphysema. Postmortem CT findings at about 14 h postmortem revealed gas within the mediastinum, that is, pneumomediastinum (Fig. 1a) and no gas in other organs or spaces such as the cranial cavity. Consolidation in the middle and lower lobes of the right lung and emphysema in the both lung were also identified (Fig. 1a).

An autopsy revealed mild abrasions in the right forearm, but no signs of trauma to the chest or abdomen. The postmortem CT findings were consistent with a diagnosis of pneumomediastinum (Fig. 2a). The emphysematous left lung was accompanied by interlobular sub-pleural emphysema (Fig. 2b) and microscopy confirmed bronchopneumonia in the middle and lower lobes of the right lung. The heart contained a large amount of dark-red blood mixed with fat-like and soft coagulated blood. Esophageal or gastric rupture was not evident despite a sliding esophageal hiatal hernia. Microscopy revealed many fat droplets at the basal side of the cytoplasm in the proximal tubular epithelium of the kidney.

Table 1 shows the postmortem biochemical findings. A toxicological screen of the blood and urine detected none and 0.2 mg/mL of ethanol, respectively. Drug screening of a urine sample using Triage (Biosite Diagnostics, San Diego, CA, USA) was negative.

2.2. Case 2

A woman aged in her 40 s was found dead at home and cardiopulmonary resuscitation was not attempted. She has been undergoing type I diabetes mellitus ever since she was a child. Postmortem CT was performed at 16 h after she was pronounced dead. The major findings of CT were pneumomediastinum (Fig. 1b) and brain edema. None of the organs including the brain, liver, and spleen or spaces such as the cranial, thoracic and abdominal cavities contained gas.

Consistent with the postmortem CT findings, brain edema (1424 g) and pneumomediastinum (Fig. 3) were also determined at autopsy. Signs of trauma to the chest and abdomen were absent. The heart contained a large amount of dark-red blood mixed with fat-like and soft coagulated blood. The esophagus was intact with no signs of rupture or laceration. Microscopy revealed diffuse diabetic glomerulosclerosis and many fat droplets in the basal side of the cytoplasm in the proximal tubular epithelium of the kidney. Autolysis without fibrosis was evident in the pancreas. The only remarkable findings in other organs were congestion.

Table 2 shows the postmortem biochemical findings. A toxicological screen did not detect ethanol in the blood or urine. A urine sample tested negative for drug use after screening using Triage (Biosite Diagnostics).

3. Discussion

The presence of gas, especially inside organs, is one of the common findings of postmortem CT, and is caused ordinarily by decomposition and cardiopulmonary resuscitation [10]. In our two cases where pneumomediastinum was detected on postmortem CT (Fig. 1), the absence of gas in other organs and spaces can exclude putrefactive modification, and neither woman underwent cardiopulmonary resuscitation obviously. Pneumomediastinum can arise as a result of many conditions such as trauma, recent medical interventions, a history of aero-digestive organ injury, pneumothorax, mechanical ventilator related injury, or diseases including Boerrhaave syndrome, respiratory disease, cervicothoracic infections or childbirth [1,2,4,11]. The pathophysiological mechanism of pneumomediastinum is advocated to be barotrauma by Macklin et al. [12]. Increased intrathoracic pressure causes the alveolar rupture, which results in air leakage from the alveolus to the mediastinum via the pulmonary interstitium and pulmonary hilum [4,11].