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CASE REPORT HAMMAN SYNDROME SECONDARY TO SEVERE ACUTE RESPIRATORY SYNDROME CORONAVIRUS 2 INFECTION: A CASE REPORT

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Abstract: Introduction: Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) infection is a public health emergency. A rare complication of this infection is spontaneous pneumomediastinum, also known as Hamman syndrome. **Case report:** A 58-year-old hypertensive and diabetic male presented with flu-like symptoms and respiratory failure and was subjected to orotracheal intubation and mechanical ventilation. Extensive subcutaneous emphysema and chest asymmetry were observed. Chest computed tomography also revealed a pneumomediastinum with extensive subcutaneous emphysema. The patient progressed with reduced ventilatory capacity and hemodynamic deterioration before subsequently dying. **Discussion:** Hamman syndrome is a marker of poor prognosis denoting worse outcomes among patients with SARS-CoV-2 infection.

INTRODUCTION

The coronavirus disease 2019 (COVID-19), caused by severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2), was declared a global public health emergency by the World Health Organization (WHO) on March 11, 2010.

Despite the predominance of mild cases, COVID-19 may lead to severe acute respiratory syndrome (SARS) in approximately 5% of diagnosed cases. In fact, as of September 20, 2020, a total of 30,675,675 cases and 954.417 deaths have been reported worldwide[1].

Several studies have described the computed tomography (CT) features of COVID-19, the temporal evolution of these features, and its differential diagnosis with other viral infections. In particular, COVID-19 produces an imaging pattern that resembles organizing pneumonia, including peripheral ground-glass opacities, nodular images, and bilateral and multilobar consolidations[2].

Other imaging findings include linear, curvilinear, and perilobular pulmonary opacities, as well as focal and diffuse ground-glass opacities, which mimic other diseases, including infections, inhalation exposure, and drug toxicity. Pneumothorax have also been described in some cases[3].

Interestingly, a possible but rare complication of COVID-19 is spontaneous pneumomediastinum, also known as Hamman syndrome, which has an estimated prevalence of 0.001% in the general population[4]. Furthermore, a previous study has reported a relationship between this condition and SARS-CoV-2 infection.

Therefore, assistant physicians should pay attention to physical examination and ultrasound imaging findings[5], as well as understand the pathogenesis of this syndrome to avoid unnecessary therapies, thereby minimizing risk to patients.

CASE REPORT

A 58-year-old hypertensive and diabetic male presented with fever, headache, myalgia, fatigue, and intense cough, which resulted to reduced respiratory capacity, prompting admission to an emergency care unit five days after the onset of symptoms. The patient did not present with odynophagia, anosmia, ageusia, diarrhea, and vomiting. He was suspected to have pneumonia and transferred to a tertiary hospital for initial screening and treatment.

The patient progressed to type 1 respiratory failure and was referred to the intensive care unit (ICU) to receive non-invasive ventilation. Oxygen delivery through a high-flow nasal cannula and dexamethasone treatment for hypoxemia correction did not improve his clinical condition. Afterwards, he underwent orotracheal intubation 10 days after the onset of symptoms.

The patient remained under mechanical ventilation (MV) with protective parameters (driving pressure, peak inspiratory pressure, and plateau pressure). On Day 12 of MV, extensive subcutaneous emphysema with associated left-sided chest asymmetry was observed during physical examination, extending from the xiphoid appendix to the pectoral muscles. Percussion and auscultation of the chest were unremarkable, and plain chest radiography did not show pneumothorax. CT scan performed on Day 13 of MV showed bilateral pulmonary involvement characterized by peripheral and basal ground-glass opacities; extensive bilateral subcutaneous emphysema affecting the chest, intercostal, and paravertebral muscles; pneumomediastinum occupying the anterior, posterior, and middle mediastinum; and pleura without obvious signs of lesions or effusion.

Extubation failed in this patient, and he was tracheostomized seven days after. However, despite undergoing tracheostomy, his ventilatory capacity decreased, forgoing the possibility of mechanical ventilator disconnection, and hemodynamic stability subsequently deteriorated. The patient eventually died from refractory septic shock on Day 26 of ICU admission.

DISCUSSION

Given the high morbidity and mortality of this disease, the COVID-19 pandemic has brought several challenges to the clinical management of affected patients, especially in severe cases.

The virus has caused approximately thousands of deaths in a short period of time due to its high transmissibility and consequent organ system complications[6], especially in the respiratory system. Among these complications, spontaneous pneumomediastinum, also known as Hamman syndrome, is a benign and self-limiting syndrome that has been reported to be a marker of poor prognosis and worse clinical outcomes in SARS-CoV-2 patients[7].

Triggering factors for Hamman syndrome include exercise, birth delivery, diabetic ketoacidosis, drug inhalation, coughing, and vomiting. Notably, the first event underlying its pathophysiology is alveolar rupture due to high intra-alveolar pressure, low perivascular pressure, or both. Although rare, the association between pulmonary function tests and air leak syndrome has also been increasingly reported in the literature, of which lung diseases, including interstitial lung diseases, involve structural changes that facilitate the development of this complication[8].

Following the initial event, air freely enters the mediastinum during the respiratory cycle to balance the pressure gradients, which is known as the Macklin effect as first described in 1939. Studies have shown that pressure gradients seem to be related to the heterogeneous involvement of the lungs, in cases where the normal parenchymal tissue is adjacent to diseased tissue[9].

Physical examination findings of the present case report were consistent with pneumothorax, which has a high morbidity and requires immediate and invasive therapeutic interventions in most cases[7]. Although spontaneous pneumomediastinum is usually self-limiting, and its pathophysiological mechanism remains incompletely understood, this syndrome can potentially cause serious circulatory and respiratory complications, thereby serving as an indicator of clinical deterioration in COVID-19 patients[5].

Furthermore, the differential diagnosis of Hamman syndrome in the intensive care setting is broad. In addition to the lesser effectivity of MV in critically ill COVID-19

patients with other pathologies, these patients usually remain in the ICU for extended periods and may undergo invasive procedures, which then increases the risk of lung injury.

The objective of this case study was to demonstrate an atypical progression of SARS-CoV-2 infection associated with Hamman syndrome, focusing on its detection, natural history, and differential diagnosis, as a reference for future similar cases.

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