Case Report

Spontaneous Pneumomediastinum and Macklin Effect: Three Rare Case Reports with Computed Tomography Findings

M Serindere, M Ersen[1], U Balyemez[2]

Department of Radiology, Hatay Education and Research Hospital, Hatay, Turkey, [1]Department of Radiology, Burdur Bucak State Hospital, Burdur, Turkey, [2]Department of Radiology, Faculty of Medicine, Near East University, Nicosia, Cyprus

Received: 22-Dec-2022;
Revision: 26-Feb-2023;
Accepted: 08-Mar-2023;
Published: 15-May-2023

Abstract

Spontaneous pneumomediastinum (SPM) is defined as free air or gas in the mediastinum that is not associated with an obvious cause such as chest trauma. The SPM results from acutely elevated intra-alveolar pressure: The high-pressure gradient between the distal alveoli and the pulmonary interstitium leads to alveolar rupture. This causes free gas to separate through the peribronchovascular fascial sheaths (interstitial emphysema) into the hilum and then into the mediastinum. Once the gas is in the mediastinum, it can travel up to the cervical soft tissues (even the retroperitoneum) producing subcutaneous emphysema. The Macklin effect appears on thoracic computed tomography (CT) as linear air collections adjacent to bronchovascular sheaths. This case report presents CT findings of SPM due to the Macklin effect in three cases and a brief literature review on this subject.

Keywords: *Computed tomography, emphysema, Macklin effect, spontaneous pneumomediastinum*

Introduction

Spontaneous pneumomediastinum (SPM) is defined as free air in the mediastinum that is not associated with any apparent causes such as chest trauma, intrathoracic infections, surgery, other organ ruptures, or mechanical ventilation.[1] The mechanism of spontaneous mediastinum was firstly suggested by Charles Macklin in 1937. The pathogenesis of this entity is air leakage into the peribronchovascular sheaths, interlobular septa, and mediastinum via the visceral pleura due to alveolar rupture because of pressure increase.[2,3] SPM often develops in young adults and usually resolves spontaneously within a few days of treatment, including rest and analgesics.[4] However, esophageal perforation is the most serious gastrointestinal perforation and is associated with high morbidity and mortality. Previously, pneumomediastinum was reported in 83.5% of patients with esophageal perforation.[5] Therefore, it is important to distinguish SPM from esophageal perforations. The Macklin effect is useful in distinguishing it from esophageal perforation.[6]

The purpose of this case report was to present three rare cases of SPM with computed tomography (CT) findings that provide important information in differentiating the Macklin effect from esophageal perforation.

Case Report

Case 1

A 20-year-old male patient presented to the emergency department with a complaint of chest pain. Body temperature was 36.5°C. Oxygen saturation was 98%. Laboratory and electrocardiogram (ECG) findings were normal. He underwent a ventriculoperitoneal shunt operation five years ago due to hydrocephalus. Air density adjacent to the right atrium of the heart was observed on the chest X-ray image. The mediastinal window of the axial thorax CT image

Address for correspondence: Dr. M Serindere, Hatay Education and Research Hospital, Department of Radiology, Hatay, Turkey.
E-mail: drserindere@hotmail.com

Access this article online

Quick Response Code:

Website: www.njcponline.com

DOI: 10.4103/njcp.njcp_901_22

How to cite this article: Serindere M, Ersen M, Balyemez U. Spontaneous pneumomediastinum and Macklin effect: Three rare case reports with computed tomography findings. Niger J Clin Pract 2023;26:528-30.

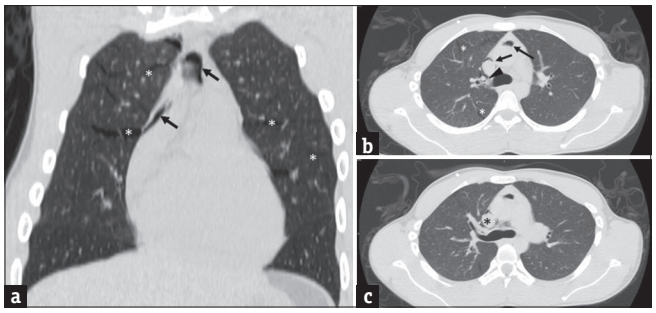

Figure 1: Coronal (a) reconstructed and axial (b and c) unenhanced CT images showing low attenuation areas along the bronchovascular sheaths (white and black asterisks) and the mediastinum (black arrows) due to mediastinal and interstitial emphysema

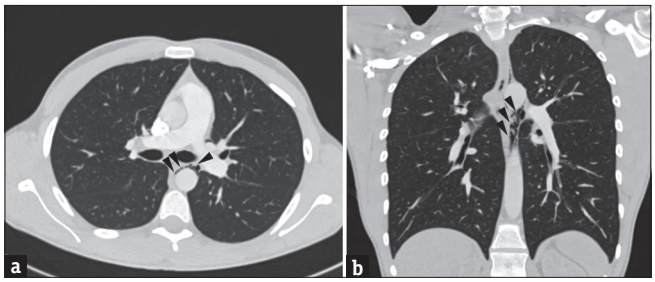

Figure 2: Axial (a) and coronal (b) reconstructed unenhanced CT images showing pneumomediastinum in the preaortic and subaortic spaces, respectively (white arrows)

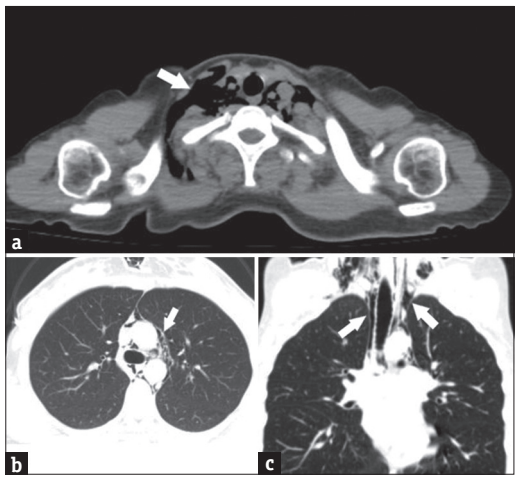

Figure 3: (a) Axial unenhanced CT image with soft tissue window showing soft tissue emphysema (white arrow). Axial (b) and coronal (c) unenhanced CT images with parenchyma window showing pneumomediastinum (white arrows)

demonstrated the presence of air in the mediastinum, suggesting the Macklin effect. In the lung window of CT, peribronchovascular interstitial emphysema was detected [Figure 1a-c]. There was no evidence of soft tissue emphysema. No surgical treatment was required. The patient was followed and had no complications.

Case 2

A 25-year-old male patient presented with a sudden onset of stabbing chest pain. He had no trauma history. The ECG was performed on the patient because his father had a history of sudden cardiac death at an early age. His ECG revealed normal findings. Oxygen saturation was 99%. The complete blood count (CBC), creatine kinase-MB, and troponin values were normal. A thorax CT was performed because the patient had mild shortness of breath, and no cardiological etiology could be found. Free air values in mediastinal spaces were observed in a thorax CT [Figure 2a and b]. No rupture was observed in the trachea and main bronchi. In esophageal endoscopic evaluation, no signs of perforation were detected. The patient was diagnosed with the Macklin effect, and conservative treatment, including oxygen therapy and analgesics, was applied.

Case 3

A 29-year-old female patient presented to the emergency department with stabbing chest pain that had been present for one day, and the symptoms were similar to those of a panic attack. She had no trauma history. Oxygen saturation was 97%. Laboratory and ECG findings were normal. Symptomatic treatment was applied to the patient who was under psychiatric follow-up for panic attacks, and she was discharged. One day later, a thorax CT was performed when the patient came to the emergency department complaining of similar symptoms and additionally crepitation in the inferior cervical region. In a thorax CT, air values in the mediastinum and soft tissue emphysema in the lower cervical region were revealed [Figure 3a-c]. Tracheal and esophageal defects were not observed. These findings suggested the Macklin effect as a diagnosis. She was discharged because her symptoms regressed when she was hospitalized.

Discussion

SPM is predominantly seen in young men and is a rare condition.[2,4] Similarly, our cases 1 and 2 were young males. The incidence of SPM has not been clearly established because available published reports are only case studies or small case series and the incidence of this entity is probably underestimated because it can easily be ruled out when the diagnostic index of suspicion is not high; moreover, the symptoms of SPM are not very specific, some symptoms may go unnoticed, and some radiographic findings are difficult to identify.[4]

The most common symptoms described in the literature are chest pain, dyspnea, and neck pain or discomfort.[4] In Macia *et al*.'s[4] case series of 41 patients, the three most common clinical manifestations of SPM were chest pain, dyspnea, and subcutaneous emphysema of

the neck. All three patients had severe chest pain, Case 3 had also subcutaneous emphysema. Kaneki *et al*.[7] reported that up to 30% of patients with SPM present with a normal radiograph, and therefore, the authors recommend a chest CT scan. A chest CT scanning is considered the gold standard of imaging tests that can detect pneumomediastinum in patients with small amounts of mediastinal air or even when the Macklin effect is present.[8] Sakai *et al*.[8] also reported that the Macklin effect may often be demonstrated by CT in patients with SPM due to nontraumatic respiratory causes. A CT-proven Macklin effect may be useful in distinguishing respiratory from other causes of pneumomediastinum. Given these results, chest CT scans should be reserved for cases where the diagnosis is unclear.[4]

SPM is sometimes associated with pneumorrhachis, the presence of air in the spinal epidural space.[2] A literature review of 48 patients with pneumorrhachis revealed that only one case had neurological symptoms and signs.[9] No such complication was encountered in all three patients during the follow-up period.

However, the radiological appearance of pneumomediastinum includes a band of hyperlucency parallel to the left side of the cardiac silhouette and a thin radiopaque line pointing to the elevated mediastinal pleura; radiolucent lines in the mediastinum extending toward the neck; and mediastinal structures surrounding the air, such as the aorta, trachea, esophagus, or thymus gland. The presence of subcutaneous emphysema of soft tissues (especially in the neck and less often in the chest) is associated with the abovementioned symptoms in a high percentage of patients.[4] Subcutaneous emphysema was detected only in case 3.

In conclusion, CT plays an important role in the differential diagnosis of etiological factors such as esophageal and tracheal perforation, which cause pneumomediastinum and progress with more severe clinical findings. Particularly in young patients, in cases with chest pain and mild dyspnea, SPM should be considered and supported by radiological findings.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Macklin MT, Macklin CC. Malignant interstitial emphysema of the lungs and mediastinum as an important occult complication in many respiratory diseases and other conditions: Interpretation of the clinical literature in the light of laboratory experiment. Medicine 1944;23:281-358.
2. Murayama S, Gibo S. Spontaneous pneumomediastinum and Macklin effect: Overview and appearance on computed tomography. World J Radiol 2014;6:850-4.
3. Kim KS, Jeon HW, Moon Y, Kim YD, Ahn MI, Park JK, *et al*. Clinical experience of spontaneous pneumomediastinum: Diagnosis and treatment. J Thorac Dis 2015;7:1817-24.
4. Macia I, Moya J, Ramos R, Morera R, Escobar I, Saumench J, *et al*. Spontaneous pneumomediastinum: 41 cases. Eur J Cardiothorac Surg 2007;31:1110-4.
5. Sohda M, Kuwano H, Sakai M, Miyazaki T, Kakeji Y, Toh Y, *et al*. A national survey on esophageal perforation: Study of cases at accredited institutions by the Japanese Esophagus Society. Esophagus 2020;17:230-8.
6. Kumeda H, Saito G. Spontaneous pneumomediastinum diagnosed by the Macklin effect. J Surg Case Rep 2022;2022:rjab634.
7. Kaneki T, Kubo K, Kawashima A, Koizumi T, Sekiguchi M, Sone S. Spontaneous pneumomediastinum in 33 patients: Yield of chest computed tomography for the diagnosis of the mild type. Respiration 2000;67:408-11.
8. Sakai M, Murayama S, Gibo M, Akamine T, Nagata O. Frequent cause of the Macklin effect in spontaneous pneumomediastinum: Demonstration by multidetector-row computed tomography. J Comput Assist Tomogr 2006;30:92-4.
9. Belotti EA, Rizzi M, Rodoni-Cassis P, Ragazzi M, Zanolari-Caledrerari M, Bianchetti MG. Air within the spinal canal in spontaneous pneumomediastinum. Chest 2010;137:1197-200.