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SPONTAN PNÖMOMEDİASTİNUM: 17 OLGUNUN KLİNİK ANALİZİ

SPONTANEOUS PNEUMOMEDIASTINUM: CLINICAL ANALYSIS OF 17 CASES

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ÖZET

AMAÇ: Spontan Pnömomediastinum (SPM), kendiliğinden veya tetikleyici faktörlerin etkisi ile oluşan alveoler rüptür sonucu havanın trakeobronşial ağaç komşuluğu yolu ile mediastende birikimi ile oluşan nadir görülen bir hastalıktır. Bu çalışmanın amacı, spontan pnömomediasten tanı ve tedavisindeki deneyimlerimizi bildirmektir.

GEREÇ VE YÖNTEM: 3.5 yıllık süreçte SPM tanısı ile tedavi edilen 17 hasta retrospektif olarak çalışmaya alındı. Olguların; yaş, cinsiyet, sigara kullanımı, ek hastalık durumu, klinik prezentasyonları, radyolojik görüntüleme, tedavi süreçleri ve nüks durumları incelendi. Sekonder pnömomediasten olguları çalışmaya alınmadı.

BULGULAR: SPM olgularının 14'ü erkek ve 3'ü kadın hastaydı. Ortalama yaş 19.2 ±2.8 ve ortalama BMI 18.5 ±6.3 kg/m^2 olarak bulundu. SPM oluşumu için tetikleyici faktör 13 (%76.5) olguda bulunmaktaydı. 4 (%23.5) olguda astım tanısı mevcuttu. 9 (%52.9) olgu sigara kullanmaktaydı. En sık görülen semptom 14 (%82.4) olguyla göğüs ağrısıydı. Subkutan amfizem 5 (%29.4) ve Hamman bulgusu 2 (%11.8) olguda vardı ve Toraks tomografisinde 3 (%17.6) olguda Macklin effect tespit edildi. Olguların hastanedeki yatış süresi ise ortalama 4.5 ±2 gündü. Olguların hiçbirisine fiberoptik bronkoskopi, üst GIS endoskopi ve pretrakeal fasya açılması yada mediastene drenaj katateri yerleştirilmesi gibi girişimsel işlem uygulanmadı. Antibiyoterapi oranları incelendiğinde ise 13 (%76.5) olguya antibiyoterapi verilmediği, ve hiçbir olguda mediastinit gibi ileri komplikasyon gelişmediği görüldü.

SONUÇ: SPM göğüs ağrısı ve dispne ile prezente olan benign bir durumdur. Tanıda bilgisayarlı tomografi altın standarttır ve genelde konservatif yollarla tedavi edilir. Nüks nadir olmakla birlikte tanı anında sekonder bir nedenin SPM'ye yol açıp açmadığının belirlenmesi hayati önem taşımaktadır.

ANAHTAR KELİMELER: Antibiyotik proflaksisi, Mediastinal amfizem, Subkütan amfizem

ABSTRACT

OBJECTIVE: Spontaneous pneumomediastinum (SPM) is a rare disease caused by the accumulation of air in the mediastinum through the tracheobronchial tree neighborhood due to alveolar rupture, which occurs spontaneously or with the effect of precipitating factors. The aim of this study is to report our experience in the diagnosis and treatment of SPM.

MATERIAL AND METHODS: Seventeen patients treated with the diagnosis of SPM in a 3.5 year period were included in the study retrospectively. Age, gender, smoking, co-morbid disease status, clinical presentations, radiological imaging, treatment processes, and recurrence status were analyzed. Secondary pneumomediastinum cases were not included in the study.

RESULTS: Fourteen of the patients were male, and three were female. The mean age was 19.2 ±2.8, and the mean Body Mass Index (BMI) was 18.5 ±6.3 kg/m^2. The precipitating factor for SPM was found in 13 (76.5%) cases. There was a diagnosis of asthma in 4 (23.5%) cases. 9 (52.9%) cases were smokers. The most common symptom was chest pain, with 14 cases (82.4%). Subcutaneous emphysema was present in 5 (29.4%) and Hamman's sign in 2 (11.8%) cases, and the Macklin effect was detected in 3 (17.6%) cases in thorax tomography. The mean hospital stay of the cases was 4.5 ±2 days. No interventional procedures were performed, such as fiberoptic bronchoscopy, upper gastrointestinal endoscopy, pretracheal fasciotomy, or mediastinal drainage catheterization. When the antibiotic therapy rates were examined, it was seen that 13 (76.5%) cases were not given antibiotics, and no advanced complications such as mediastinitis developed in any case.

CONCLUSIONS: SPM is a benign condition presenting with chest pain and dyspnea. Computed tomography is the gold standard in diagnosis, and SPM is usually treated conservatively. Although recurrence is rare, it is vital to determine the presence of an underlying secondary cause at the time of initial diagnosis.

KEYWORDS: Antibiotic prophylaxis, Mediastinal emphysema, Subcutaneous emphysema

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INTRODUCTION

Pneumomediastinum (PM) or mediastinal emphysema is the presence of free air between the supporting tissues in the mediastinum.

Hamman published the first pneumomediastinum case series in 1939 (1). The hypothesis proposed by Macklin in 1944 that the air released as a result of alveolar rupture caused by the sudden increase in intrathoracic pressure reaches the mediastinum through the peribronchial support tissue remains valid (2). While the concept of Spontaneous Pneumomediastinum (SPM), which was first defined, included PMs triggered by underlying diseases, this definition started to be defined for Primary Spontaneous Pneumomediastinum (PSPM) in the following years occurred as a result of non-pathological precipitating factors in healthy young adults without an underlying disease (3). In the literature, it is seen that the terms SPM and PSPM are used interchangeably. Secondary PM is defined as air in the mediastinum caused by trauma, surgery, medical treatments (iatrogenic PM), or infections (4, 5). The incidence of SPM has been reported to be 1/800-42.000 cases in different series (6, 7).

There is a generally accepted approach for the diagnosis of SPM, but treatment management is still not optimized. This study it is aimed to analyze the presence of typical triggering factors in SPM cases, the results of our diagnostic methods, and treatment management.

MATERIALS AND METHODS

This study was designed as a retrospective and observational study. The approval of the local ethics committee was obtained. The authors confirmed compliance with the World Medical Association Declaration of Helsinki on the ethical conduct of research involving human subjects. Seventeen SPM cases treated in a single center between November 2018 and December 2021 were included in the study retrospectively. Patients with underlying diseases such as Interstitial Lung Disease, Chronic Obstructive Pulmonary Disease, and patients with tumors in the thorax or neck region were excluded from the study. However, the study did not include all iatrogenic PM cases, such as trauma and medical procedures (upper gastrointestinal endoscopy, fiberoptic bronchoscopy, etc.). PM cases in patients with bronchial asthma were included in our study because they were evaluated as SPM in the literature (3, 4).

Patient data were obtained from physical and digital archive files. Age, gender, height, weight, Body Mass Index (BMI), family history, presence of co-morbid disease, smoking, triggering event, length of hospital stay, symptoms, physical examination findings (subcutaneous emphysema and Hamman's sign), radiological findings (Macklin effect), antibiotic therapy, length of hospital stay, and recurrence data were recorded. Patients diagnosed with spontaneous pneumomediastinum were hospitalized for close follow-up in the thoracic surgery inpatient service **(Figure 1)**.

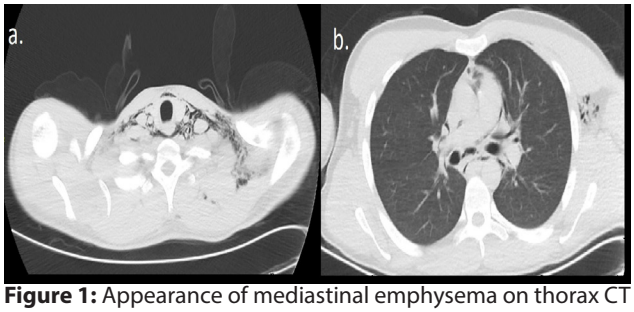

Figure 1: Appearance of mediastinal emphysema on thorax CT **(a)** image of free air around the trachea, thyroid, esophagus, and both carotid sheaths in the lower cervical region, **(b)** Free air around both main bronchi and truncus pulmonalis in the axial section of the subcarinal space.

Daily chest X-ray, hemogram, and C-reactive protein (CRP) tests were requested from the patients. Nasal oxygen (4lt/min) therapy was initiated in all patients diagnosed with spontaneous pneumomediastinum, and antibiotic prophylaxis was administered to patients with elevated inflammatory markers.

Ethical Committee

The Ordu University Ethics Committee approved this study. (Approval number and date: 2022/73, 25.03.2022)

Statistical Analysis

Statistical analysis was performed using the "IBM SPSS Statistics for Windows. Version 22.0 (Statistical Package for the Social Sciences, IBM Corp., Armonk, NY, USA)" program. Descriptive statistics of the research; frequency and percentage for categorical variables, mean and standard deviations for numerical variables.

RESULTS

Of the 17 cases, 14 (82.4%) were male, 3 (17.6%) were female, and the mean age was 19.2 ± 2.8 (15-24 years). The mean height of the cases was 180.8 ± 6.3 cm (170-191 cm), the mean weight was 60.6 ± 10 kg (42-80 kg), and the mean BMI was calculated as 18.5 ± 6.3 kg/m^2 (14.2-24.4 kg/m^2). The mean hospital stay of the cases was 4.5 ± 2 days (3-10 days). When the cases'co-morbid diseases were examined, it was noticed that 4 (23.5%) had asthma, and 1 case each had type 1 diabetes mellitus, migraine, and acute rheumatic fever in childhood. When the presence of previous SPM or Pneumothorax was questioned in the family history, it was learned that there was a history of Primary Spontaneous Pneumothorax in the elder brother and mother of 1 patient. When the smoking histories of the cases were questioned, it was determined that 9 (52.9%) cases did not smoke, and 8 (47.1%) cases did **(Table1)**.

Table 1: Demographic data of spontaneous pneumomediastinum patients

Baseline Characteristic	n mean	% SD
Gender		
Female	3	17.6
Male	14	82.4
Age	19.2	2.8
Height	**180.8**	6.3
Weight	60.6	10
BMI	18.5	6.3
Co-morbidities		
Asthma	4	23.5
Type I DM	1	5.9
Migraine	1	5.9
Acute rheumatic fever	1	5.9
Smoking		
Yes	8	47.1
No	9	52.9

Note. DM: Diabetes Mellitus

It was observed that there was a precipitating factor for SPM in 13 (76.5%) cases and it was absent in 4 (23.5%) cases. When the precipitating factors are examined; 4 (23.5%) cases had asthma, but only 1 patient had an asthma attack during the development of SPM. 3 (17.6%) cases occurred during physical activity (lifting weights in the gym, playing football and running to catch the bus). SPM developed in 2 (11.8%) patients following severe cough after aspiration while eating, in 1 (5.9%) patient with severe vomiting after alcohol intake, 1 (5.9%) patient developed sobbing. It was observed that SPM developed in one case each due to severe retching (regurgitation) caused by stress before the school exam, after climbing and descending to a high altitude on the same day, and after loud shouting **(Table 2)**.

Table 2: Precipitating factors of spontan pneumomediastinum

Variable	n	%
Non-apperent	4	23.5
Apperent	13	76,5
Physical activity	3	17.6
Cough	2	11.8
Vomitting	1	5,9
Asthma attack	1	5.9
Sobbing	1	5.9
Retching	1	5.9
Altitude change	1	5.9

The most common symptom was chest pain, described in 14 (82.4%) cases. Other symptoms were neck pain, dysphagia or odynophagia, and dyspnea. Subcutaneous emphysema was detected in 5 (29.4%) and Hamman's sign in 2 (11.8%) cases. All patients were diagnosed with SPM by thorax computed tomography (CT) taken in the emergency department. The Macklin effect was detected in 3 (17.6%) cases when the thorax CTs were examined **(Table 3), (Figure 2)**.

Table 3: Presenting symptoms, physical examination and radiological findings of spontaneous pneumomediastinum patients

Variables	n	%
Symptom		
Chest pain	14	82.4
Pain in the neck	3	17.6
Dyspnea	2	11.8
Dysphagia	2	11.8
Physical examination finding		
Subcutaneous emphysema	5	29.4
Hamman sign	2	11.8
Macklin effect	3	17.6

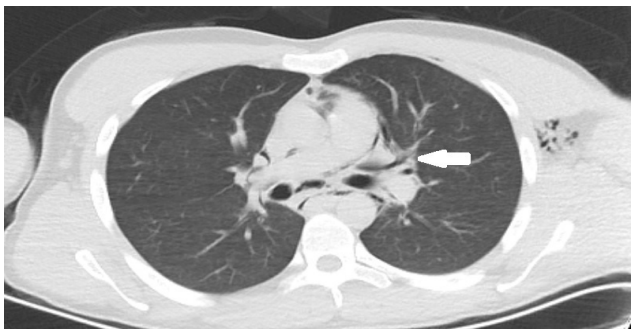

Figure 2: The Macklin effect as depicted on a thorax CT scan Air tracking along the peribronchovascular sheaths (white arrow) towards the hilum and concomitant pneumomediastinum in pulmonary interstitial emphysema

No interventional procedures such as fiberoptic bronchoscopy, upper gastrointestinal endoscopy, pretracheal fasciotomy, or mediastinal drainage catheterization were performed. The cases were followed up and treated with con-

servative methods, especially nasal oxygen support therapy. While oral intake of 13 (76.5%) cases continued with a regular diet, oral feeding of 4 (23.5%) cases was stopped temporarily (2-7 days). In the anamnesis of these cases, retching, vomiting, episode of hiccups, and severe cough after food aspiration were present as precipitating factors. Since there were no complications in their clinical follow-up, they were discharged after switching to normal nutrition. When the rates of antibiotic therapy given due to the development of SPM of the patients were examined, it was seen that 13 (76.5%) cases were not given, and 4 (23.5%) cases were given antibiotics. It was determined that leukocytes and c-reactive protein (CRP) levels were elevated in patients given antibiotics and none of the 17 cases developed complications related to SPM. Recurrent SPM that developed two years later was seen in only 1 (5.9%) case with asthma, and this case was also treated conservatively.

DISCUSSION

SPM is a rare clinical condition. The treatment approach for secondary PM secondary to the underlying disease or interventional procedure is generally straightforward. However, there is no consensus on the follow-up and treatment algorithm for SPM. We think that this is due to the studies with a small number of cases published in the literature on SPM. The fact that the centers exhibit different approaches to the follow-up and treatment of SPM prevents the formation of consensus on this issue. We think that preventing unnecessary diagnostic and interventional procedures by sharing our approach to SPM with results is critical in developing countries such as Turkey.

SPM is generally seen in males around the age of 20 (8 - 12). The mean age of the cases in our study was 19.2, and 82.4% were male, compatible with the literature data. In studies where the BMI of the cases was evaluated, values in the range of 19.56-20.8 kg/m^2 were published (6 - 8). The mean BMI value of our cases was 18.5 ± 6.3 kg/m^2. The fact that the literature data is close to the lower limit supports the idea that thin and tall people are a factor in their susceptibility to SPM, just like in Primary Spontaneous Pneumothorax patients. When the inpatient follow-up periods in the published studies are examined, it is seen that the hospitalization period is reported in the range of 1.8-8.5 days (11 - 19). The mean hospitalization period of our cases was 4.5 ± 2 days (3-10 days), and it was seen to be in parallel with the studies above. SPM recurrence is infrequent. The 0-3% rate of recurrence rates have been reported in published data (4, 8, 11, 13, 17). Recurrence developed two years later in 1 (5.9%) case with an asthma diagnosis. The low number of cases in our study, compared to the literature, may explain the greater rate of SPM recurrence.

Precipitating factors for SPM are mainly related to the Valsalva maneuver. Events such as intense coughing, excessive shouting or singing loudly, vomiting, and strenuous physical activity, such as intrathoracic pressure changes in the Valsalva maneuver, are trigger factors for SPM (15-20). Studies in the literature have reported that it is a precipitating factor at 30-69%. In addition, different types of triggering factors have been reported in the literature. A precipitating factor was discovered in 76.5% of the cases in our investigation. In addition, although the rate of SPM triggered by drug abuse is around 15% in the literature, this finding was never detected in our study (13). We think that detailed anamnesis is essential in revealing the triggering factor in SPM cases.

The presenting symptoms, physical examination (subcutaneous emphysema and Hamman's sign), and radiological findings (Macklin effect) in SPM cases are all reported at various rates. Chest pain was the most common symptom in the literature, with a rate of 72-82% (4 - 8). Similarly, the most common symptom in our study was chest pain, with a rate of 82.4%. Other symptoms were listed as neck pain, dysphagia, and dyspnea, and these symptoms were similar to the literature (21, 22). Subcutaneous emphysema and Hamman sign, two physical examination findings specific for SPM, are reported in different ranges such as 29-92% and 0-52%, respectively (8, 18). Hamman's sign is the heartbeat accompanied by the crepitation sound during auscultation. It was described by the scientist who gave it its name in 1939 and is pathognomonic for SPM (1). In our study, subcutaneous emphysema was found to be 29.4%, and Hamman's sign was 11.8%. In a systema-

tic review study, Dajer-Fadel et al. reported an average of 40.3% subcutaneous emphysema and 13.8% Hamman's sign, including 27 studies with spontaneous pneumomediastinum cases (13). We think that studies with more extensive case series should be conducted to determine the true incidence of these findings. Diagnostic use of thorax CT is increasingly seen in current studies and has reached approximately 85% (18, 21). In our study, the diagnosis of all cases was made by thoracic CT without applying intravenous contrast. Thorax CT is considered the gold standard radiological examination for the diagnosis of SPM. The Macklin sign, defined as the air appearance between the supporting tissue of the bronchovascular area, has now taken its place in the literature as a thorax CT finding. It has been reported at very different rates, such as 15-83% in a few studies (2, 4, 11, 13, 22). In our study, the Macklin sign was detected with 17.6%. We think that the overall incidence could not be determined because this specific finding was not addressed in many SPM studies.

There are many studies on prophylactic antibiotic therapy in conservative treatment. In these studies, it is seen that prophylactic and then therapeutic antibiotics are given in a significant difference, such as 15-100%, and there are also different approaches to ceasing oral intake for certain periods (11, 13, 19). Our study revealed that prophylactic antibiotic therapy and ceasing oral intake were applied in the same 4 (23.5%) cases. Oral intake was ceased in these cases due to signs of suspected esophageal perforation, such as vomiting and sobbing, and the presence of inflammatory indicators like CRP and leukocyte elevation. In the presence of anamnesis, triggering factors, and laboratory findings suggesting possible mediastinitis in SPM cases, we recommend stopping oral intake and starting antibiotic therapy. Since these conditions are not present, we would like to state that unnecessary antibiotic therapy contributes negatively to the increasing prevalence of antibiotic-resistant microorganisms.

We would like to point out that esophageal graphics with contrast material, upper gastrointestinal endoscopy, fiberoptic bronchoscopy, advanced blood tests, and high-cost prophylactic antibiotic therapy for the diagnosis, etiology, and treatment of SPM are all economically unfavorable issues, especially in developing countries like ours. Although none of those mentioned above high-cost procedures were performed in our series, no advanced complications such as mediastinitis were observed in any patient with close follow-up and clinical judgment.

Our study has several limiting factors. Initially, the study was conducted in a single center and included a limited number of cases. Secondly, our study is a retrospective study similar to the studies in the literature. Finally, we would like to state that it may have been detected at low rates since the consideration of subjective criteria such as physical examination and Hamman's sign depends on the personal diagnostic perception of the clinician who performed the first examination.

Primary spontaneous pneumomediastinum is a rare pathology of thoracic surgery emergencies. It should be considered in patients with acute chest and/or neck pain in the differential diagnosis. Computed tomography is the gold standard in diagnosis and is generally treated conservatively. Antibiotic prophylaxis and oral intake discontinuation are not required in all cases. Although recurrence is rare, it is vital to determine the presence of an underlying secondary cause at the time of initial diagnosis.

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