Original Article

Clinical experience of spontaneous pneumomediastinum: diagnosis and treatment

Kyung Soo Kim[1], Hyun Woo Jeon[2], Youngkyu Moon[1], Young Du Kim[2], Myeong Im Ahn[3], Jae Kil Park[1], Keon Hyun Jo[1]

[1]Department of Thoracic and Cardiovascular Surgery, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Republic of Korea; [2]Department of Thoracic and Cardiovascular Surgery, Bucheon St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Bucheon, Republic of Korea; [3]Department of Radiology, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Republic of Korea

Contributions: (I) Conception and design: KS Kim; (II) Administrative support: KH Jo; (III) Provision of study materials or patients: YD Kim, JK Park, KH Jo; (IV) Collection and assembly of data: HW Jeon, Y Moon; (V) Data analysis and interpretation: KS Kim, HW Jeon, Y Moon; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

Correspondence to: Keon Hyun Jo, MD. Department of Thoracic and Cardiovascular Surgery, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Banpo-daero 222, Seocho-gu, Seoul 137-701, Republic of Korea. Email: khjo@catholic.ac.kr.

Background: Spontaneous pneumomediastinum (SPM) is a benign disease with a variety degree of severity but definite treatment modality is not clearly identified with its rarity. The purpose of this study was to review our experience and discuss the management of SPM according to the severity of disease.

Methods: From March 1996 to December 2012, total 64 patients were enrolled and classified as mild, moderate and severe groups and subsequent clinical courses were analyzed retrospectively.

Results: Fifty-one were males and 13 were females (M:F =3.9:1) with a mean age of 18 years old (range: 10-30 years old). Thirty-six patients were in mild, 22 in moderate and 6 in severe group. Chief complaints were chest pain (50 cases; 78.1%), neck pain (35 cases; 54.7%), dyspnea (18 cases; 28.1%), odynophagia (9 cases; 14.1%) and precipitating factors were coughing in 12 cases, feeding problems in 9 cases, and vomiting in 7 cases; however, 34 patients (53.1%) had no precipitating signs. All patients received oxygen therapy (100%), prophylactic antibiotics in 57 patients (89.1%), and pain medications in 47 patients (73.4%). The mean hospital stay was 4.6 days (range: 1-10 days). There was an increased linear trend according to time to visit (P=0.023) but clinical course demonstrated no significant trend between groups.

Conclusions: These data demonstrated that there was no difference in symptom, clinical course and SPM was adequately treated with conservative management regardless of the degree of severity of SPM.

Keywords: Pneumomediastinum; mediastinal emphysema; chest pain; subcutaneous emphysema

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Introduction

Spontaneous pneumomediastinum (SPM) is defined as the presence of interstitial air in the mediastinum. It is a benign disease entity, which rarely occurs but responds well to conservative treatment (1). SPM is diagnosed with presenting symptoms, physical signs with air in the mediastinum, radiologic confirmation, and the absence of specific pathologic causes that are present in secondary pneumomediastinum (2,3). SPM was first described by Louis Hamman in 1939 (4) and published reports have described the nature of SPM and its clinical significance in only a small number of cases and definite treatment modality is not clearly identified yet. The purpose of this study was to review our clinical experience and detail the clinical approach on the management of SPM according to

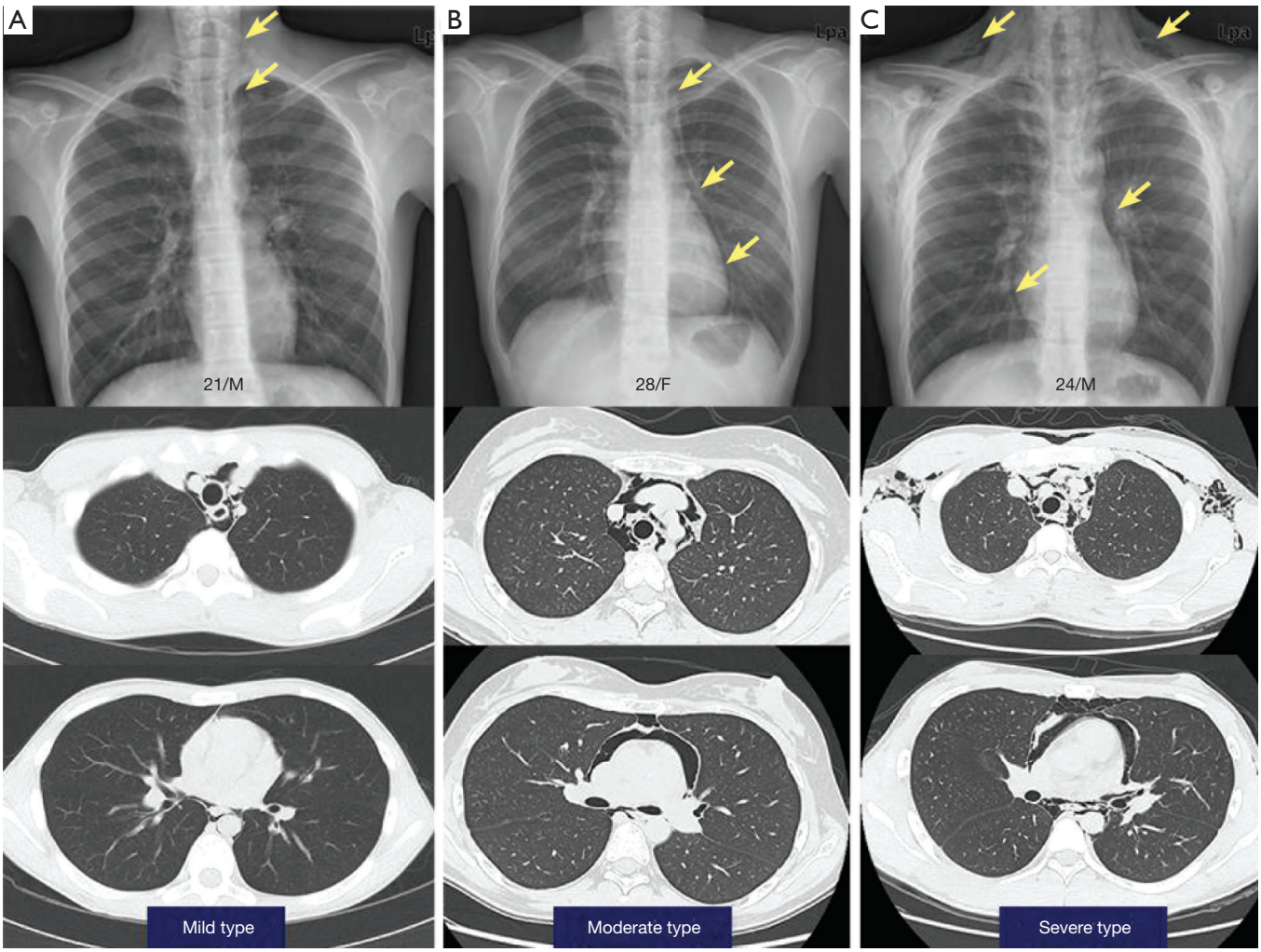

Figure 1 Representative appearance of spontaneous pneumomediastinum in initial chest X-ray and chest computed tomography by simple classification. Mild type (A), moderate type (B) and severe type (C).

the severity of the disease with a literature review.

Methods

From March 1996 to December 2012, we retrospectively reviewed the data of patients admitted for treatment of SPM as an initial primary diagnosis in two institutions. We analyzed demographic data, presenting symptoms and signs, radiologic and interventional evaluation studies, and treatment courses. A precipitating factor was defined as a related recent event, which could trigger the development of SPM. Secondary pneumomediastinum due to specific causes were excluded: including trauma, esophageal perforation, recent intervention or surgery involving aero-digestive organs, abnormal findings in tracheobronchial tree, intrathoracic infections by gas forming organisms, mechanical ventilator or tracheostomy related conditions, or combined pneumothorax or emphysematous lung disease.

Sixty-four cases were diagnosed with SPM who were older than 10 years of age. We classified the patients three groups according to the extent of disease (mild: presence of minimal air shadow from larynx to carina level plus restricted cervical subcutaneous emphysema; moderate: mild criteria plus involvement of peribronchial tree and pericardial area with subcutaneous emphysema; and severe: moderate criteria plus extensive chest wall subcutaneous emphysema) by initial radiologic evaluation (*Figure 1*) and clinical course, including lab findings were analyzed. Institutional Review Board approved this study and individual informed consents were waived.

Table 1 Demographics of patients (n=64)

Variables	Groups, n (%)				P value
	Mild, n=36	Moderate, n=22	Severe, n=6	Total, n=64	
Age, mean ± SD (years)	19.06±4.28	17.81±3.67	18.00±4.05	18.53±4.04	0.505
Male	31 (86.1)	15 (68.2)	5 (83.3)	51 (79.7)	0.262
Height, mean ± SD (cm)	171.36±8.11	167.68±10.83	173.50±7.97	170.30±9.20	0.227
Weight, mean ± SD (kg)	58.31±10.21	56.30±12.09	53.17±16.47	57.14±11.43	0.550
BMI, mean ± SD	19.78±2.58	19.79±2.40	17.35±4.71	19.56±2.81	0.130
Smoking					0.679
Non-smoker	30 (83.3)	21 (95.5)	6 (100.0)	57 (89.1)	
Smoker	5 (13.9)	1 (4.5)	0 (0.0)	6 (9.4)	
Ex-smoker	1 (2.8)	0 (0.0)	0 (0.0)	1 (1.6)	
Season					0.244
Spring	11 (30.6)	2 (9.1)	2 (33.3)	15 (23.4)	
Summer	16 (44.4)	10 (45.5)	2 (33.3)	28 (43.8)	
Fall	8 (22.2)	7 (31.8)	1 (16.7)	16 (25.0)	
Winter	1 (2.8)	3 (13.6)	1 (16.7)	5 (7.8)	
Predisposing factor					
Asthma	2 (5.6)	2 (9.1)	0 (0.0)	4 (6.3)	0.753
History of pneumothorax	2 (5.6)	0 (0.0)	0 (0.0)	2 (3.1)	0.607
Interstitial lung disease	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	N/A
Steroid use	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	N/A
Subcutaneous emphysema	13 (36.1)	9 (40.9)	5 (83.3)	27 (42.2)	0.123

Categorical variables are reported as frequencies (%), and continuous variables are reported as mean ± SD. Chi-square test or Fischer's exact test was used for categorical variables. SD, standard deviation; BMI, body mass index; N/A, not applicable.

Statistical analysis

Categorical variables are expressed as percentages and continuous variables are reported as mean ± standard deviation (SD). Chi-square test or Fisher's exact test was used for categorical variables. Patients were divided into three groups (mild, moderate, and severe type) and analyzed with one-way analysis of variance (ANOVA), and performed test for linear trend. The P values <0.05 were considered to indicate statistical significance. All statistical analyses were performed with SAS (version 9.3; SAS Institute, Cary, NC) or SPSS (version 19.0; SPSS Inc., Chicago, IL).

Results

After exclusion of secondary pneumomediastinum cases, 64 patients were enrolled as a SPM. There were 36 (56.3%) patients in mild type, 22 (34.4%) in moderate type and 6 (9.4%) in severe type. Fifty-one were males and 13 were females (M:F =3.9:1); the mean age was 18.5±4 years old (range, 10-30 years old) and the mean body mass index (BMI) was 19.6±2.8 kg/m^2 (range, 9.0-24.9 kg/m^2). There were 57 (89.1%) non-smokers, 6 (9.4%) current smokers and 1 (1.6%) ex-smoker, but no patients had received steroids or other drugs. There was an increased prevalence in the summer season (28 cases; 43.8%) and 2 (3.1%) with history of pneumothorax patients and 4 (6.3%) with asthmatic patients. The demographic data shows no difference between groups (*Table 1*). The most frequent chief complaint at presentation was chest pain (50 cases, 78.1%), followed by neck pain (35 cases, 54.7%), dyspnea (18 cases, 28.1%), odynophagia (9 cases, 14.1%). Subcutaneous emphysema was presented by initial assessment in 27 cases (42%). Precipitating factors were coughing (12 cases, 18.8%), feeding difficulties (9 cases, 14.1%), vomiting (7 cases, 10.9%), exercise (4 cases, 6.3%), and playing wind instrument (1 case,

Table 2 Presenting complaints and precipitating factors (n=64)

Variables	Groups, n (%)				P value
	Mild, n=36	Moderate, n=22	Severe, n=6	Total, n=64	
Presenting complaints					
Chest pain	26 (72.2)	18 (81.8)	6 (100.0)	50 (78.1)	0.317
Neck pain	20 (55.6)	12 (54.5)	3 (50.0)	35 (54.7)	1.000
Dyspnea	9 (25.0)	7 (31.8)	2 (33.3)	18 (28.1)	0.774
Odynophagia	5 (13.9)	4 (18.2)	0 (0.0)	9 (14.1)	0.765
Coughing	0 (0.0)	1 (4.5)	0 (0.0)	1 (1.6)	0.438
Voice change	0 (0.0)	0 (0.0)	1 (16.7)	1 (1.6)	0.094
Precipitating factors					
No precipitating factor	21 (58.3)	11 (50.0)	2 (33.3)	34 (53.1)	0.490
Coughing	6 (16.7)	5 (22.7)	1 (16.7)	12 (18.8)	0.888
Feeding	4 (11.1)	4 (18.2)	1 (16.7)	9 (14.1)	0.566
Vomiting	2 (5.6)	2 (9.1)	3 (50.0)	7 (10.9)	0.015
Exercise	2 (5.6)	1 (4.5)	1 (16.7)	4 (6.3)	0.535
Playing wind instrument	1 (2.8)	0 (0.0)	0 (0.0)	1 (1.6)	1.000

Categorical variables are reported as frequencies (%). Chi-square test or Fischer's exact test was used for categorical variables.

1.6%); however, 34 patients (53.1%) had no precipitating signs. Notably, vomiting and disease severity showed significant relationship (P=0.015, *Table 2*).

Plain chest radiography was initially done in all patients. The plain radiography of the neck was concomitantly checked in 37 (57.8%) patients. Fifty-four (84.4%) chest computed tomography (CT) and 7 (10.9%) neck CT scans were performed. Additional studies of 10 (15.6%) esophagograms, 4 (6.3%) endoscopies and 1 (1.6%) bronchoscopy was conducted; however, they revealed otherwise unremarkable findings. One patient with an abnormal endobronchial tumor was finally diagnosed as secondary pneumomediastinum and was excluded from the study. The mean duration between onset of symptoms and hospital presentation was 14.0±18.6 hours (range, 0.5-96 hours). Conservative management, consisting of bed rest with oxygen inhalation (64 patients; 100%), prophylactic antibiotics (57 patients; 89.1%), and analgesic medications (47 patients; 73.4%) were applied. The mean duration of hospitalization was 4.6±1.8 days (range: 1-10 days). Feeding restriction was conducted in 39 (60.9%) patients. There were no mortality or morbidity cases, such as mediastinitis, tension pneumomediastinum, or tension pneumothorax during hospitalization; furthermore, recurrence of SPM developed in only 1 (1.6%) patient in the moderate type after 12 months, which was also managed conservatively. Primary spontaneous pneumothorax developed 6 and 17 months later in 2 (3.1%) patients in the mild type who had no previous history of pneumothorax. There was no significant difference in clinical course and management between groups (*Table 3*).

The mean white blood cell (WBC) count was 10,226.6±3,473.2/μL (range, 3,440-19,160/μL) with a mean absolute neutrophil count (ANC) of 7,000.0±3,107.7/μL (range, 1,307-14,774/μL), and an elevated WBC (>10,000) was observed in 29 cases (45.3%). Erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were checked in 39 (60.9%) cases and the mean ESR was 6.18±5.43 mm/h (range, 2-24 mm/h) and the mean CRP count was 2.44±8.91 mg/L (range, 0.02-54.72 mg/L). Time to visit has a significant linear trend (P=0.023); however, there was no statistically significant linear trend in hospital days, smoking history, WBC count, ANC count, ESR and CRP (*Table 4*).

Discussion

Pneumomediastinum is a benign disease entity, defined as the presence of free air in the mediastinum and is classified as spontaneous or secondary type (1,2). A secondary pneumomediastinum has several causes: for example, recent interventions, history of aero-digestive organ injury, cervico-

Table 3 Study evaluation and clinical course (n=64)

Variables	Groups, n (%)				P value
	Mild, n=36	Moderate, n=22	Severe, n=6	Total, n=64	
Radiologic evaluation					
Chest X-ray	36 (100.0)	22 (100.0)	6 (100.0)	64 (100.0)	N/A
Neck film	23 (63.9)	11 (50.0)	3 (50.0)	37 (57.8)	0.597
Chest CT	28 (77.8)	20 (90.9)	6 (100.0)	54 (84.4)	0.332
Neck CT	4 (11.1)	2 (9.1)	1 (16.7)	7 (10.9)	0.714
Both	3 (8.3)	1 (4.5)	1 (16.7)	5 (7.8)	0.487
Aero-digestive organ study					
Esophagography	3 (8.3)	6 (27.3)	1 (16.7)	10 (15.6)	0.143
Endoscopy	1 (2.8)	3 (13.6)	0 (0.0)	4 (6.3)	0.300
Bronchoscopy	1 (2.8)	0 (0.0)	0 (0.0)	1 (1.6)	1.000
Treatment modality					
Initial nothing by mouth	22 (61.1)	13 (59.1)	4 (66.7)	39 (60.9)	1.000
Oxygen inhalation	36 (100.0)	22 (100.0)	6 (100.0)	64 (100.0)	N/A
Empirical antibiotics	31 (86.1)	21 (95.5)	5 (83.3)	57 (89.1)	0.392
Pain medication	27 (75.0)	15 (68.2)	5 (83.3)	47 (73.4)	0.768
Clinical course					
Time to visit, mean ± SD (hours)	12.06±14.15	12.68±21.36	30.67±25.97	14.02±18.60	0.068
Hospital days, mean ± SD	4.11±1.55	5.23±2.00	5.17±1.33	4.59±1.76	0.043
Recurrence	0 (0.0)	1 (4.5)	0 (0.0)	1 (1.6)	0.438
Spontaneous pneumothorax	2 (5.6)	0 (0.0)	0 (0.0)	2 (3.1)	0.607

Categorical variables are reported as frequencies (%), and continuous variables are reported as mean ± SD. Chi-square test or Fischer's exact test was used for categorical variables. N/A, not applicable; CT, computed tomography; SD, standard deviation.

Table 4 Comparison results according to the classification by disease severity

Variables	Groups, mean ± SD				P value[#]
	Mild, n=36	Moderate, n=22	Severe, n=6	Total, n=64	
Hospital days	4.11±1.55	5.23±2.00	5.17±1.33	4.59±1.76	0.164
Time to visit (hours)	12.06±14.15	12.68±21.36	30.67±25.97	14.02±18.60	0.023
Smoking history, n (%)[§]	6 (16.7)	1 (4.5)	0 (0.0)	7 (10.9)	0.099
WBC count (/μL)	9,561.1±3,169.1	10,984.5±3,629.7	11,440.0±4,344.6	10,226.6±3,473.2	0.220
Segmented neutrophils (%)	63.89±11.20	68.06±9.49	75.28±10.05	66.39±10.95	0.039
ANC count (/μL)	6,351.2±2,823.3	7,603.5±3,220.5	8,679.5±3,782.8	7,000.0±3,107.7	0.089
ESR (mm/h)*	5.95±4.74	7.29±6.74	2.67±1.15	6.18±5.43	0.402
CRP (mg/L)*	3.42±11.80	1.30±1.96	0.63±0.35	2.44±8.91	0.621

[#], P values for differences were determined by trend test; [§], smoking history includes smoker plus ex-smoker; *, ESR and CRP were checked in 39 (60.9%) cases (i.e., numbers of cases in mild, moderate and severe groups were 22, 14 and 3, respectively). SD, standard deviation; WBC, white blood cell; ANC, absolute neutrophil count; ESR, erythrocyte sedimentation rate; CRP, C-reactive protein.

thoracic infections, pneumothorax, or mechanical ventilator related injury. These cases may require invasive intervention (3,5,6). On the contrary, SPM is diagnosed without any definite cause. Pneumomediastinum was first reported by René Laennec in 1819 and spontaneous mediastinal emphysema was described by Louis Hamman in 1939 (4). The pathophysiologic mechanism of SPM is uncertain; but Macklin *et al.* in 1944 suggested that increased intrathoracic pressure induces alveolar rupture with passage of air into the interstitium or bronchovascular tissues of the tracheobronchial tree (7,8). This uncommon disease has been described in several case series but diagnostic and therapeutic guidelines for SPM remain debatable. Kaneki *et al.* suggested detailed classification could be made by the presence of subcutaneous emphysema on chest X-ray and the degree of free air in the mediastinum imaged by a chest CT (9). We also theorized that the mediastinal air shadow in a radiologic exam and subcutaneous emphysema is indicative of a pneumomediastinum; therefore, we suggested detailed classification made by the presence of subcutaneous emphysema on chest X-ray and the degree of free air in the mediastinum imaged by the chest CT scan.

The most common symptom at presentation is chest pain, as it was found in our study. The other symptoms including dyspnea, sore throat or dysphagia were observed (5,9-13). Possible precipitating events include coughing, vomiting, defecation, parturition, or other physical activity that is associated with increased intrathoracic pressure (e.g., Valsalva maneuver, playing wind instrument); however, it can develop without a triggering event. Many western reports have described a relationship between smoking or drug abuse and the development of SPM. However, we found that the majority of patients were non-smokers (89%) and none used steroids or were drug abusers. According to the literature, precipitating events ranged from 39-100% (2,12). Our study also revealed that chest pain (78%) was the most common symptom; however, more than half of the patients did not experience a precipitating event.

Several studies reported that SPM occurs predominantly in young males, and that minimal, undetectable bulla can be the cause of the pneumomediastinum (2,9-14). Our results also revealed that SPM developed with high prevalence in young males, and it has an ectomorphic appearance similar to that of patients with pneumothorax. However, concomitant pneumothorax cases were excluded initially and definite bullae were not found in all patients who underwent a CT scan. This undefined relationship results in SPM being classified as a unique disease entity; thus, the treatment methodology should be different from that of spontaneous pneumothorax.

A simple diagnostic tool to detect SPM is either a plain chest radiograph or chest CT scan. Previous reports have noted that SPM can be confirmed by chest radiography alone in about 70% of cases; and the other 30% of patients can be diagnosed with a chest CT scan (9,14). In our study, all patients were suspected of suffering from a SPM by an initial chest X-ray; however, chest CT (84.4%) and neck CT (10.9%) scans were also conducted. We performed a CT scan to confirm the diagnosis as well as to exclude secondary pneumomediastinum; moreover, a diagnostic accuracy was achieved up to 100% with chest radiography, including additional CT scans. Further evaluation studies such as an esophagogram, bronchoscopy and endoscopy were performed in several cases to determine another obvious origins; however, as was the case in similar studies (15,16), all but one patient with an incidental endobronchial tumor, exhibited no abnormal finding. Thus, we demonstrated that a plain chest X-ray including a CT scan is an adequate evaluation procedure after a thorough history and physical examination to exclude secondary pneumomediastinum. Further invasive interventional studies such as bronchoscopy or esophagogram should be performed in highly suspicious cases of aero-digestive organ injury, such as Boerhaave's syndrome or tracheobronchial tree rupture.

Oxygen therapy has been recommended in most previous reports because it is considered that the consumption of oxygen increases the diffusion pressure of nitrogen in the interstitium, promoting absorption of free air in the mediastinum. In our data, oxygen inhalation was applied in all 64 patients (100%). Although an elevated WBC (>10,000) was observed in 29 cases (45.3%) but most of patients (89.1%) were prescribed empirical antibiotics to prevent possible infection or disease aggravation such as mediastinitis. It remains debatable whether antibiotic treatment is essential. We suggest that empirical antibiotics should be minimized and applied in selected patients showing leukocytosis with fever and subsequent infection is likely. Moreover, oral feeding was restricted in 39 patients (60.9%) but a normal diet was given within 24-48 hours in most of cases. Neither evaluation nor clinical course documented digestive organ injury or abnormality in all cases including seven patients with a history of vomiting. We recommend that dietary restriction is unnecessary and early feeding is reasonable, except in the cases of highly suspected patient of Boerhaave's syndrome. Discharge was

determined by relief of subjective symptoms, with regressed pneumomediastinum on follow-up plain radiography. The duration of hospitalization was shorter in the mild type (P=0.043) and it is likely that the early disappearance of subjective symptoms and visible subcutaneous emphysema in a follow-up radiologic examination results in the clinical decision for early discharge without further evaluation.

Recurrence of SPM is rarely reported (5,12). One recurrence (1.6%) occurred in our study after 12 months but it responded well to conservative management. Two other male patients developed a spontaneous pneumothorax; one patient who developed blebs underwent thoracoscopic surgery 6 months later and another patient was managed with a closed thoracostomy 17 months later. We reviewed the chest CT scans in these recurrence and pneumothorax cases just after the SPM was diagnosed but we could not find any evidence of bulla lesion or emphysema. Most published reports demonstrated a very low incidence of recurrence, which responded well to conservative management; similar to our case in which routine outpatient follow-up was not recommended.

According to the literature, the presence of subcutaneous emphysema ranges from 40% to 100% (10,15); 27 cases (42.2%) showed subcutaneous emphysema in this study. There was no difference in demographics, evaluation studies, treatment modality and clinical courses between the groups (*Tables 1-3*). We also analyzed the relationship between disease groups by classification and laboratory markers of inflammation by test for linear trend. The trend analysis revealed significant result in time to visit only (P=0.023). Progression of the pneumomediastinum according to the increased time interval may show in a radiologic examination as a disease progression. However, there was no significant trend in hospital days, smoking history, WBC count, ANC count including ESR and CRP (*Table 4*)

Our retrospective review showed good results from conservative management, similar to that described by previous studies (13,17-21). Thus, we suggest that a chest X-ray and CT scan is adequate for diagnostic evaluation and that routine dietary restriction is unnecessary. Empirical antibiotics should be minimized and bed rest with oxygen inhalation or supportive pain control is adequate for the most of cases diagnosed with SPM.

Conclusions

SPM is adequately treated with conservative management regardless of the degree of severity of SPM. Thus, invasive evaluation studies and intensive treatment should be reserved for highly suspicious, selective cases of aero-digestive organ injury.

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Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Consents: Written informed consent was obtained from all the patients. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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