Original Article

Clinical features of recurrent spontaneous pneumomediastinum

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Background: Spontaneous pneumomediastinum recurrence is rare, and its clinical presentation is unclear. We investigated the clinical features of and predisposing factors for spontaneous pneumomediastinum recurrence.

Methods: We retrospectively investigated 30 consecutive patients treated for new-onset spontaneous pneumomediastinum at Shinshu Ueda Medical Center between 2012 and 2021. We evaluated the patient background characteristics, trigger activity, radiological findings, and clinical course of spontaneous pneumomediastinum, including those of recurrent cases. Predisposing factors for spontaneous pneumomediastinum recurrence were evaluated by comparing patients with and without recurrence.

Results: Most patients were male (87%). The median age of the patients was 16 years (range, 12–26 years). Among the 30 patients, five experienced at least one recurrence of spontaneous pneumomediastinum. All recurrences occurred within 1 year after new-onset spontaneous pneumomediastinum. Clinical presentations associated with spontaneous pneumomediastinum recurrence, including vital signs, laboratory data, length of hospital stay, and radiological extent of spontaneous pneumomediastinum, were similar to or less aggressive than those associated with new-onset spontaneous pneumomediastinum. Patients with recurrence were more likely to have a medical history of preexisting lung diseases, such as asthma, than those without recurrence (60% *vs.* 8%; P=0.022). Only one of five patients with recurrence had trigger activity at spontaneous pneumomediastinum onset (20%); however, 60% of patients without recurrence had trigger activity (P=0.15).

Conclusions: Spontaneous pneumomediastinum recurrence may have a similar or less aggressive clinical presentation than new-onset spontaneous pneumomediastinum. The presence of preexisting lung diseases may increase the risk of spontaneous pneumomediastinum recurrence.

Keywords: Spontaneous pneumomediastinum; subcutaneous emphysema; recurrence

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Introduction

Spontaneous pneumomediastinum (SPM) is a rare entity defined as the presence of free air in the mediastinum that is not associated with any noticeable cause such as chest trauma. It has been proposed that the pathophysiology of SPM involves rupture of the alveoli caused by a rapid increase in alveolar pressure, which has been referred to as the Macklin effect (1). The Macklin effect refers to the phenomenon of a large pressure gradient between the alveoli and lung interstitium inducing alveolar rupture

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and the subsequent release of free air along the pulmonary vessels and bronchus in the limited area of the lungs coursing toward the mediastinum. Studies have suggested that SPM frequently presents with the sudden onset of symptoms, such as chest pain, in young adults with or without events, such as physical exercise, that can trigger a rapid increase in intrathoracic pressure (2,3). SPM is generally considered to be associated with a relatively benign course. However, data pertaining to long-term clinical outcomes, such as SPM recurrence, are limited.

The recurrence of SPM is considered rare, with a reported incidence of 1.2% (4). Because of its rarity, predisposing factors for SPM recurrence have not been investigated. In this study, we retrospectively investigated 30 consecutive patients with new-onset SPM who had been treated at our institution for 10 years, including five patients who subsequently experienced SPM recurrence. To assess the predisposing factors for SPM recurrence, we specifically evaluated patient background characteristics including body habitus and preexisting lung diseases, clinical presentation including trigger actions, radiological extent of SPM, and inflammatory response to SPM evaluated by laboratory data. We present the following article in accordance with the STROBE reporting checklist (available at https://jtd.amegroups.com/article/view/10.21037/jtd-22-1141/rc).

Methods

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This single-institution, retrospective study was approved by the institutional review board of the National Hospital Organization Shinshu Ueda Medical Center (IRB No: 03-06). We utilized an opt-out approach instead of obtaining written informed consent from each patient. Using the institutional database of the National Hospital Organization Shinshu Ueda Medical Center, we identified 30 patients who were diagnosed with and treated for SPM between January 2012 and December 2021. Patient demographic information and data regarding clinical variables, including age, smoking history, body mass index (BMI), medical history, trigger action, symptoms, and radiological and blood test findings, were obtained from the medical records to perform this study. SPM was defined as the presence of free air in the mediastinum that was not associated with chest trauma, mechanical ventilation, surgical or medical procedures, or specific diseases known to cause pneumomediastinum, such as esophageal perforation. The radiological extent of SPM was assessed using a previously reported grading system (5). Using this grading system, the extent of subcutaneous emphysema was primarily evaluated based on chest radiography (CXR) findings (CXR grades 0–4), and the extent of mediastinal air was evaluated based on chest computed tomography (CT) scans (CT grades A-C) (*Figure 1*). CXR grading was performed as follows: grade 0, no abnormal findings; grade 1, air space present only in the mediastinum; grade 2, air space in the mediastinum plus mild subcutaneous emphysema that was only identifiable by careful observation using the magnification function; grade 3, subcutaneous emphysema clearly revealed without using the magnification function; or grade 4, marked subcutaneous emphysema with outlined muscle fibers of the pectoralis major. The CT grading was performed as follows: grade A, free air confined to the superior mediastinum that was defined as the mediastinum superior to the level of the tracheal carina; grade B, free air extending from the superior mediastinum to the middle mediastinum that was defined as the mediastinum between the level of the tracheal carina and that of the caudal end of the inferior pulmonary vein; and grade C, free air extending from the superior to the inferior mediastinum that was defined as the mediastinum inferior to the inferior pulmonary vein.

We evaluated the inflammatory response to SPM by assessing the maximum body temperature, white blood cell (WBC) count, and serum C-reactive protein (CRP) level during hospitalization, including the time in the emergency department. All patients were hospitalized and treated

Highlight box

Key findings

- Recurrence of spontaneous pneumomediastinum may have a similar or less aggressive clinical presentation than its new onset.
- The presence of preexisting lung diseases may increase the risk of spontaneous pneumomediastinum recurrence.

What is known and what is new?

- Spontaneous pneumomediastinum recurrence is rare, and its clinical presentation is unclear.
- Clinicoradiological findings in patients with spontaneous pneumomediastinum recurrence revealed its relatively mild clinical presentation and potential association with respiratory comorbidities.

What is the implication, and what should change now?

- In patients with spontaneous pneumomediastinum and preexisting lung diseases, care should be taken about recurrent spontaneous pneumomediastinum.

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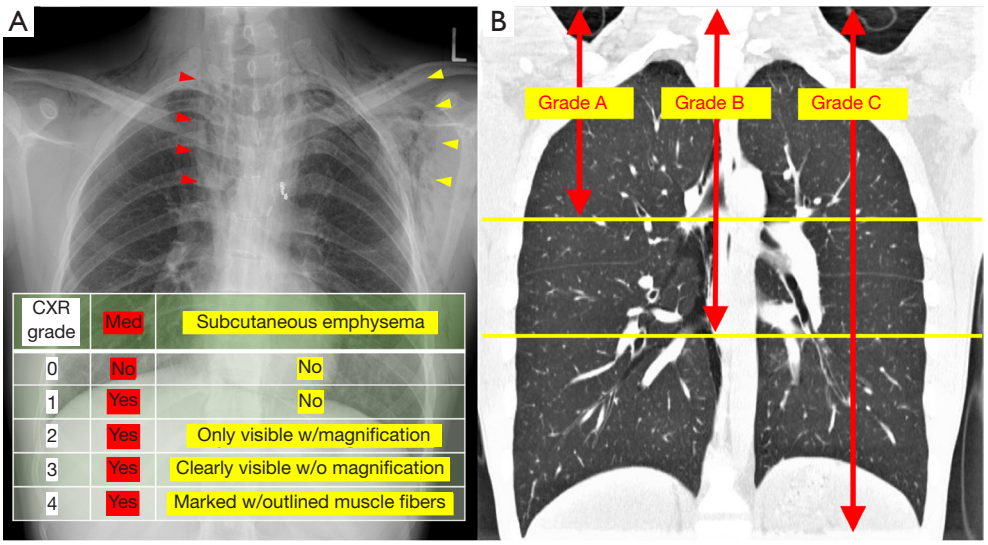

Figure 1 The radiological grading system of SPM. (A) Representative case of grade 3 in the grading system based on CXR grading of SPM showing air space in the mediastinum (red arrowheads) plus subcutaneous emphysema (yellow arrowheads) clearly. The table in the figure demonstrates each CXR grade with radiological findings of the mediastinum and subcutaneous emphysema. (B) Schema of computed tomography-based grading system of SPM. Grade A: free air confined to the mediastinum superior to the level of the tracheal carina. Grade B: free air confined to the mediastinum superior to the caudal end of the inferior pulmonary vein. Grade C: free air extending from the superior to the inferior mediastinum. CXR grade, grade based on chest radiography; Med, mediastinum emphysema; SPM, spontaneous pneumomediastinum.

conservatively. If the general condition of a patient was stable, the blood test showed acceptable results, and the CXR did not show worsening of the pneumomediastinum, then the patient was discharged. We usually followed-up patients at our outpatient clinic after hospitalization, but we did not perform periodic follow-up thereafter. Instead, we contacted all patients via telephone to ask them whether they had been diagnosed and treated for recurrent SPM specifically for the purpose of this study.

Statistical analyses

Data are shown as median [interquartile range (IQR)] or number (%). Comparisons between groups were evaluated using the chi-square test for categorical variables and the Mann-Whitney U test for continuous variables. Significant differences in clinical data for cases of recurrence and non-recurrence of SPM were analyzed using the Mann-Whitney U test. All statistical analyses were conducted using IBM SPSS Statistics 27 (IBM, Armonk, NY, USA). All statistical tests were two-sided, and $P<0.05$ was considered statistically significant.

Results

Patient characteristics and clinical course of SPM

Table 1 shows the demographic data of the 30 patients who were diagnosed with and treated for SPM. Most of these patients were male (87%). The median age of the patients was 16 years (range, 12–26 years; IQR, 13–18 years). The median BMI was 17.9 kg/m^2 (IQR, 16.6–20.1 kg/m^2). Five patients had a relevant medical history. Of these five patients, three had preexisting lung diseases (two patients had bronchial asthma and one patient had congenital pulmonary atresia). The remaining two patients had a history of pneumothorax. All patients had experienced at least one symptom during the first visit. The acute onset of chest pain (70%), dyspnea (23%), and throat symptoms (neck and pharyngeal pain) (50%) were the most frequently reported symptoms. In terms of trigger factors, 16 patients (53%) had performed physical activity that could have increased the intrathoracic pressure at the initiation of SPM. The most common trigger was a ball game exercise (9 patients), followed by swimming/diving (2 patients), other exercise (2 patients), vomiting (2 patients), and using a

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Table 1 Characteristics of patients with and without recurrent spontaneous pneumomediastinum

Patient characteristics	Recurrent SPM			
	All (n=30)	No (n=25)	Yes (n=5)	P value
Background				
Age (years)	16 [13–18]	15 [13–19]	16 [16–18]	0.59
Sex				1
Male	26 (87%)	21 (84%)	5 (100%)	
Female	4 (13%)	4 (16%)	0	
BMI (kg/m^2)	17.9 [16.6–20.1]	18.0 [16.7–20.1]	17.6 [16.6–19.4]	0.83
Smoking	2 (7%)	2 (8%)	0	0.51
MH	5 (17%)	2 (8%)	3 (60%)	0.022
CPA	1 (3%)	0	1 (20%)	
Asthma	2 (7%)	0	2 (40%)	
Pneumothorax	2 (7%)	2 (8%)	0	
Bullae or blebs on CT	9 (30%)	6 (24%)	3 (60%)	0.14
Triggers[a]				
Without trigger	14 (47%)	10 (40%)	4 (80%)	0.15[b]
With trigger	16 (53%)	15 (60%)	1 (20%)	
Ball games	9 (30%)	9 (36%)	0	
Football	6 (25%)	6 (24%)	0	
Volleyball	2 (7%)	2 (8%)	0	
Basketball	1 (3%)	1 (4%)	0	
Other triggers	7 (23%)	6 (24%)	0	
Swimming/diving	2 (7%)	2 (8%)	0	
Other exercise	2 (7%)	2 (8%)	0	
Vomiting	2 (7%)	2 (8%)	0	
Shouting	1 (3%)	0	1 (20%)	
Symptoms				
Any symptoms	30 (100%)	25 (100%)	16 (100%)	-
Chest pain	21 (70%)	18 (72%)	3 (60%)	
Back pain	2 (7%)	2 (8%)	0	
Dyspnea	7 (23%)	6 (24%)	1 (20%)	
Pharyngeal pain	7 (23%)	6 (24%)	1 (20%)	
Neck pain	8 (27%)	7 (28%)	1 (20%)	
Hoarseness	1 (3%)	1 (4%)	0	

Data are shown as the number (%) or median [25–75 percentiles]. [a], Physical activity with the potential for increased intrathoracic pressure; [b], comparison between patients with and without triggers. SPM spontaneous pneumomediastinum; BMI, body mass index; MH, medical history; CPA, congenital pulmonary atresia; CT, computed tomography.

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Table 2 Radiologic extent and clinical data during hospitalization of patients with and without recurrent spontaneous pneumomediastinum

Clinicoradiologic findings	All (n=30)	Recurrent SPM		
		No (n=25)	Yes (n=5)	P value
Radiologic extent of SPM				
Subcutaneous emphysema				
Negative	12 (40%)	9 (36%)	3 (60%)	0.36
Positive	18 (60%)	16 (64%)	2 (40%)	
Chest radiography grade				
Grade 0	6 (20%)	5 (20%)	1 (20%)	0.49
Grade 1	6 (20%)	4 (16%)	2 (40%)	
Grade 2	6 (20%)	6 (24%)	0 (0%)	
Grade 3	12 (40%)	10 (40%)	2 (40%)	
Grade 4	0 (0%)	0 (0%)	0 (0%)	
Chest CT grade				
Grade A	1 (3%)	1 (4%)	0 (0%)	0.29
Grade B	5 (17%)	3 (12%)	2 (40%)	
Grade C	24 (80%)	21 (84%)	3 (60%)	
Clinical data during hospitalization				
Initial oxygen saturation (%)	98 [97–99]	98 [97–99]	98 [97–100]	0.78
Oxygen requirement				
Yes	8 (27%)	8 (32%)	0 (0%)	0.28
No	22 (73%)	17 (68%)	5 (100%)	
Maximum body temperature (℃)	37.2 [37.0–37.3]	37.2 [37.0–37.3]	37.3 [37.0–37.3]	0.87
Maximum WBC count (10^3/μL)	9.7 [6.3–12.3]	10.1 [6.6–12.5]	7.0 [5.6–10.9]	0.35
Maximum CRP count (mg/dL)	0.2 [0.1–0.7]	0.4 [0.2–0.7]	0.1 [0.1–0.6]	0.20
Length of hospital stay (days)	8 [6–9]	8 [6–8]	8 [6–9]	0.77

Data are shown as the number (%) or median [25–75 percentiles]. SPM, spontaneous pneumomediastinum; CT, computed tomography; WBC, white blood cell; CRP, C-reactive protein.

loud voice (1 patient). In contrast, 14 patients (47%) had no specific triggers for the initiation of SPM.

Table 2 shows the radiological extent of SPM and clinical data during hospitalization. Twenty-four patients had radiographic signs suggestive of subcutaneous or mediastinum emphysema (CXR grade 1–3). However, SPM in six patients could not be diagnosed by CXR alone, and pneumomediastinum was confirmed by chest CT (CXR grade 0). Nine patients had bullae or blebs on chest CT. None of the patients underwent bronchoscopy, esophagogastroduodenoscopy, or esophagoscopy. All patients were admitted to the hospital because of the initial SPM. The median duration of hospitalization was 8 days (range, 6–9 days). The vital signs of all patients were evaluated, and all patients underwent blood tests during hospitalization and their time in the emergency department. The median maximum values were 37.2 ℃ (range, 37.0–37.3 ℃) for body temperature, 9.7×10^3/μL (range, 6.3×10^3–12.3×10^3/μL) for the WBC count, and 0.2 mg/dL (range, 0.1–0.7 mg/dL) for the CRP level. The patients did

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Table 3 Clinical features of five patients with recurrent spontaneous pneumomediastinum

Case	Age/sex	BMI	MH	Bullae or blebs on CT	Interval (days)[a]	Clinical presentation at the initial SPM					Clinical presentation at the recurrence				
						Trigger factor	Symptom	CXR grade	CT grade	LOS (days)	Trigger factor	Symptom	CXR grade	CT grade	LOS (days)
1	18/male	19.4	Asthma	Yes	32	None	Chest pain	1	2	6	None	Chest pain	1	3	6
2	16/male	16.6	CPA	Yes	39	None	Chest pain	3	3	8	None	Chest pain	0	3	Outpatient
3	16/male	16.6	None	No	96	None	Chest pain, dyspnea	3	3	9	None	Neck pain	1	2	6
4	18/male	17.6	None	Yes	126	None	Neck pain	1	3	9	None	Neck pain, dyspnea	1	3	8
5[b]	14/male	20.5	Asthma	No	315	Loud voice	Pharyngeal pain	0	2	5	None	Neck pain	0	2	5

[a], interval between the date of onset of the initial SPM and that of recurrence; [b], this case involved multiple recurrences; however, the data shown in this table are those of the first recurrence. BMI, body mass index; MH, medical history; SPM, spontaneous pneumomediastinum; CXR, chest radiography; CT, computed tomography; LOS, length of hospital stay; CPA, congenital pulmonary atresia.

not show worsening of the general condition or emphysema; therefore, they were discharged from the hospital.

Clinical features of five patients with SPM recurrence

Table 3 presents the clinical features of the five patients who experienced SPM recurrence. All these patients were male and had experienced at least one symptom at the time of recurrence. The interval between the date of the initial SPM and that of recurrence was 32 to 315 days. One patient experienced two episodes of recurrence 315 days and 372 days after the initial onset. The median patient age was 16 years (range, 16–18 years). The median BMI was 17.6 kg/m^2 (range, 16.6–19.4 kg/m^2). Among the five patients, two (40%) had a history of asthma and three (60%) had bullae or blebs on chest CT. In terms of trigger factors, only one patient had performed strenuous physical activity (shouting) at the time of the initial SPM episode, and none of the patients reported a specific trigger at the time of recurrence. The extent of pneumomediastinum at the time of recurrence was similar to or less aggressive than that at the time of the initial SPM for all patients except for one patient with the same CXR grade but an increased CT grade (case 1) (*Figure 2*).

Predisposing factors for SPM recurrence

A comparison of patients with and without SPM recurrence (*Tables 1*,*2*) showed no statistically significant difference in terms of patient background characteristics, triggers for SPM, symptoms, radiologic extent of SPM, and clinical data during hospitalization; however, there was a statistically significant difference in medical history, with preexisting lung diseases (congenital pulmonary atresia and bronchial asthma) only observed in patients with recurrent SPM (P=0.002). In terms of triggers for SPM, only one of the five patients with SPM recurrence (20%) had an apparent trigger at the onset of the initial SPM, whereas the majority of patients without recurrence (15/25; 60%) had performed trigger activities at the onset of SPM (no statistically significant difference; P=0.157). Ball game exercise was the most frequent trigger activity (9/16; 56%); however, it was only observed in patients without recurrence (*Table 2*).

Representative image of SPM and the suggested Macklin effect

Figure 3 shows the representative chest CT images at the onset of SPM, with free air spreading widely to the mediastinum. However, lung parenchymal free air was only seen in a limited area of the lungs; in this case, it was in the lower lobe. This finding suggests that the Macklin effect is a potential mechanism underlying SPM. The mechanism of the Macklin effects involves a large pressure gradient between the alveoli and lung interstitium inducing alveolar rupture, which is followed by the release of free air along

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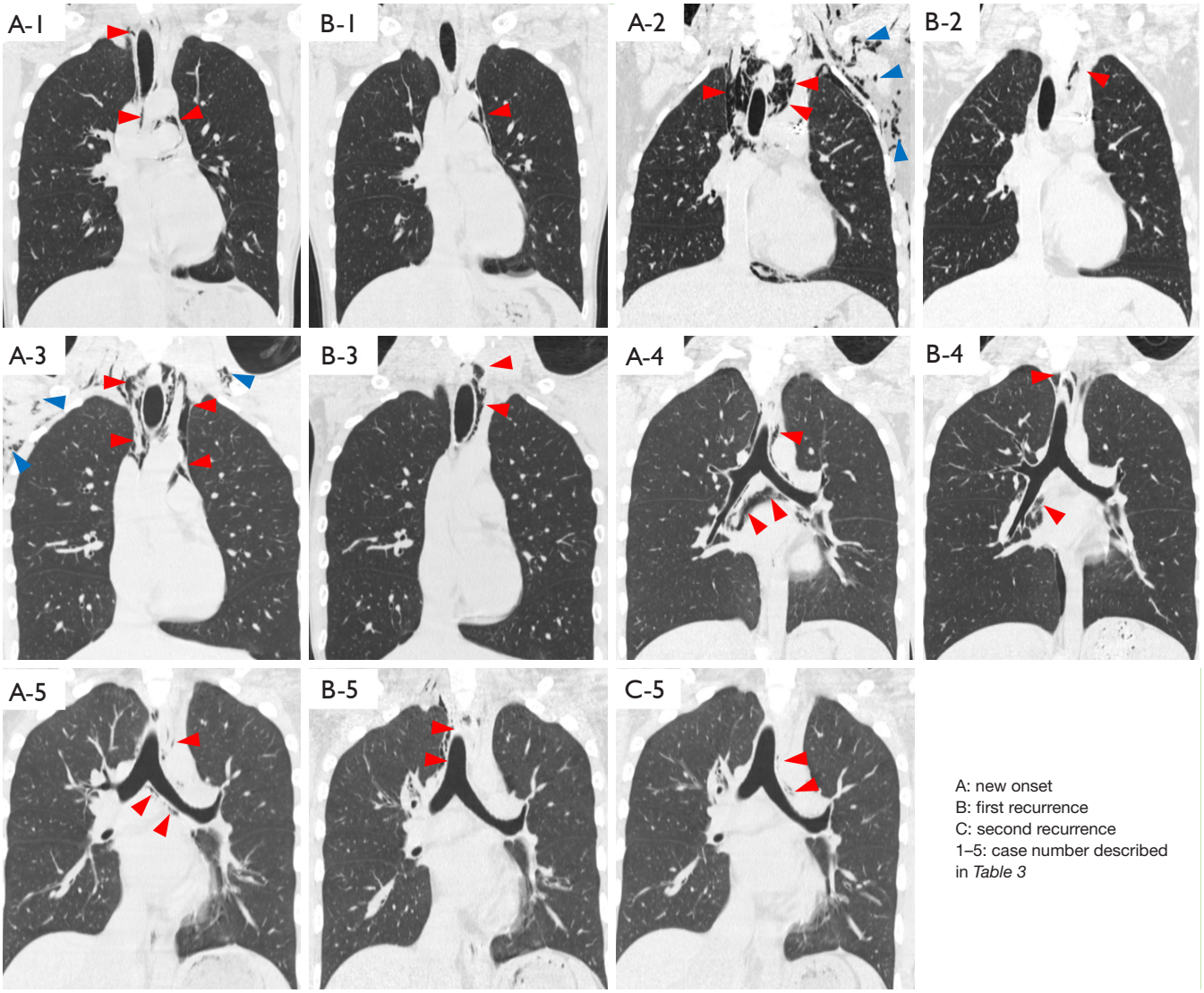

Figure 2 Coronal images of chest computed tomography scans at the onset and recurrence of SPM. The number corresponds to the case number described in *Table 3*. (A) Coronal image of new-onset SPM. (B) Coronal image of the first recurrence. (C) Coronal image of the second recurrence (only case 5). All images showed free air accumulation in the mediastinum (red arrowheads). In cases 2 and 3, subcutaneous emphysema (blue arrowheads) was seen only with new-onset SPM (A); it was not seen with recurrence (B). In all five cases, the degree of free air accumulation was similar to or less aggressive during recurrence (B) than that observed with new-onset SPM (A). SPM, spontaneous pneumomediastinum.

the pulmonary vessels and bronchus in the limited area of the lungs coursing toward the mediastinum (1,6).

Discussion

In this study, we examined the clinical features of 30 patients with SPM, including five patients with SPM recurrence. This novel study had some strengths. For example, this is the first study to provide comparative data and radiological images of the initial SPM and SPM recurrence (*Figure 2*, *Table 3*), thereby demonstrating that the clinical presentation during recurrence is similar to or less aggressive than that at the time of the initial onset. Furthermore, this is the first study to statistically analyze

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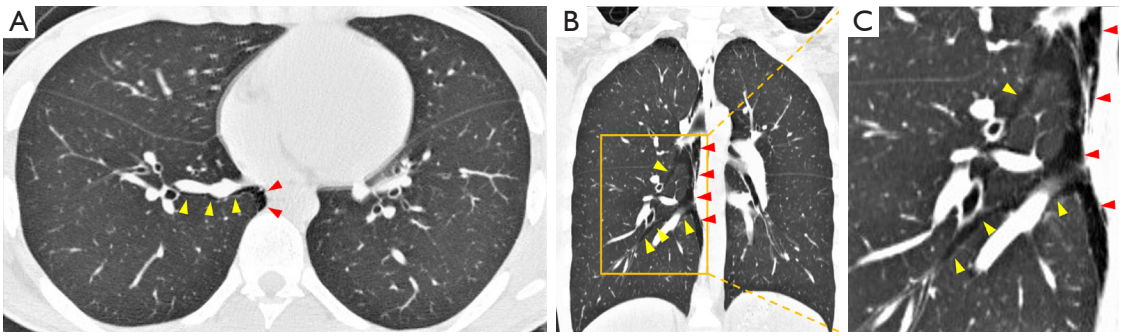

Figure 3 Representative chest computed tomography images of the onset of spontaneous pneumomediastinum in a 15-year-old boy with no trigger factors at onset. (A) Axial image, (B) coronal image, and (C) magnified image of the yellow rectangle in (B) show free air in the mediastinum (red arrowheads) and in the lung parenchyma with the pulmonary vessels and bronchus (yellow arrowheads). Free air had diffusely spread to the wide area of the mediastinum (A,B). However, free air was not seen in the lung parenchyma, except for the limited area of the right lower lobe (B,C). These findings suggest that free air spread from the limited area of the right lower lobe to the mediastinum, which further suggests that the Macklin effect was one of the mechanisms underlying spontaneous pneumomediastinum. The Macklin effect refers to the phenomenon of a large pressure gradient between the alveoli and the lung interstitium inducing alveolar rupture and the subsequent release of free air along the pulmonary vessels and bronchus in the limited area of the lungs coursing toward the mediastinum.

the predisposing factors for SPM recurrence by using various factors, including the radiologic extent of SPM and laboratory data, suggesting that the presence of preexisting lung diseases, such as bronchial asthma, is a potential predisposing factor for recurrence (*Tables 1*,*2*).

SPM was first described by Hamman in 1939 (7). Since then, many case reports and literature have been published, and the potential causes of SPM have been discussed. Childhood asthma is one of the known predisposing conditions for SPM (8-10). Among patients with childhood asthma visiting emergency departments, the reported incidence of SPM was 0.3% (8), which was higher than the general incidence of SPM of 0.001% to 0.01% (11). However, many reports suggest that SPM recurrence is rare. According to a literature review of SPM, only 5 (1.2%) of the 389 cases were identified as recurrences (4). However, many of these studies did not clearly document the clinical characteristics of patients with recurrent SPM (3,12-15). In our study, SPM recurrence was observed in five patients (16.7%) within 1 year of the initial onset. Regarding trigger factors, none of the patients with SPM recurrence performed strenuous exercise at the onset of the initial or at the onset of recurrent SPM. However, patients with SPM recurrence frequently had bullae or blebs on chest CT and a history of preexisting lung diseases including asthma. These findings indicate that SPM recurrence could be related to intrinsic factors such as alveolar fragility rather than extrinsic factors, which increase intrathoracic pressure, such as strenuous exercise.

In this study, the median length of the hospital stay was 8 days, which was relatively longer than that reported by other studies. During the study period, we treated patients in the hospital setting until we confirmed that their emphysema mostly disappeared, which might have contributed to the relatively longer hospital stay. Additionally, the relatively lower medical care costs in Japan compared with those in other developed countries might have contributed to the relatively longer hospital stay. However, none of the patients had any exacerbating conditions, including emphysema, during hospitalization. Moreover, none of the patients required invasive treatments such as surgery, radiological interventions, or chest tube drainage. Therefore, outpatient treatment or short-term hospitalization for 1 or 2 days should be sufficient unless the patients have coexisting pneumothorax or are suspected of having organ rupture, such as that associated with Boerhaave syndrome. Moreover, because recurrent SPM demonstrated similar or less aggressive clinical presentations compared with the initial SPM, hospitalization might not be required for SPM recurrence.

This study had several limitations. First, this was a single-center, retrospective investigation with a relatively small sample size. Second, we did not have a unified treatment algorithm for SPM, and the treatment policy

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and evaluations of clinical presentations were dependent on the attending physicians, which may have affected our retrospective findings. Third, we did not perform regular follow-up evaluations after the initial SPM. Although we conducted telephone interviews of all patients who did not have SPM recurrence to assess the potential for SPM recurrence, we may have missed some patients with SPM recurrence that was not recognized and healed spontaneously. Fourth, according to a published literature review of spontaneous pneumomediastinum, the mean age range is generally between 18 and 27 years (4). In a report with the largest number of cohort (n=62), the reported median age was 30 years (range, 18–84 years) years old. In comparison, the range of age of our patient cohort (median, 16 years; and range, 12–26 years) is relatively lower. Additionally, despite their young ages, 17 % of the patients had a medical history of respiratory-related illness and 30% of patients had radiological findings of bulla or bleb on CT scan (*Table 1*). Patient characteristics in our cohort mentioned above might have affected the higher recurrence rate in our study cohort compared with previously published studies.

Conclusions

In comparison with the initial onset of SPM, recurrent SPM had a similar or less aggressive clinical presentation. The extent of pneumomediastinum, triggers, and the clinical presentations of the initial SPM were not associated with recurrence; however, preexisting lung diseases may affect the risk of SPM recurrence.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://jtd.amegroups.com/article/view/10.21037/jtd-22-1141/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the institutional review board of National Hospital Organization Shinshu Ueda Medical Center (IRB No. 03-06). We utilized an opt-out approach instead of obtaining written informed consent from each patient.

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