Clinical Manifestations of Primary Spontaneous Pneumothorax in Pediatric Patients: An Analysis of 78 Patients

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Key Words
pediatric; primary spontaneous pneumothorax (PSP)

Background: The aims of this investigation were to explore primary spontaneous pneumothorax (PSP) in pediatric patients and to evaluate the clinical manifestations and outcomes of the PSP.

Methods: Seventy-eight patients diagnosed with PSP between January 2004 and December 2009 was retrospectively studied. The clinical data on demographics, diagnostic imaging, therapeutic approach, and outcomes were collected and analyzed.

Results: The sex ratio of 78 PSP patients was 7.7:1 (male:female = 69:9), and the age distribution concentrated between 15 years and 18 years (66 patients, 84.6%). The most common presenting symptom was chest pain (69 patients, 88.5%). The average body mass index was 18.2 ± 1.6 (n = 66). Autumn was the more likely attack season for PSP in this study (p = 0.005). Twenty-eight patients (35.9%) had tension pneumothorax. Only nine (11.5%) patients had a past history of cigarette smoking. All 21 outpatients received supportive treatment. Out of 57 inpatients, 10 (17.5%) received oxygen therapy, 39 (68.4%) received closed-tube drainage, and 6 (10.5%) received video-assisted thoracoscopic surgery. Apical bleb and subpleural bullae formation were common pathological findings (21 patients, 91.3%). Twenty-four (42.1%) patients experienced a second attack, and six (10.5%) patients had a third attack.

Conclusion: Pediatric PSP occurred mainly in boys of the late teenage group with lower body mass index. Autumn was the most likely attack season. There was only a small portion of the patients who smoked. There was no evidence to find a correlation between smoking and pediatric PSP attacks. Length of stay was shorter in supportive treatment and closed-tube drainage patients than that in video-assisted thoracoscopic surgery—treated patients. The outcomes were satisfactory.

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Clinical manifestations of PSP in 78 patients

1. Introduction

Primary spontaneous pneumothorax (PSP) is defined as the accumulation of air in the pleural space in healthy people without apparent lung disease being found. This is a common problem, with an incidence of 8/100,000 persons per year.\(^1\) Many reports stated that PSP tends to occur in tall thin boys and men between 10 years and 30 years.\(^2\) PSP is most often associated with the rupture of subpleural bleb or bullae on the apical portion of the upper lobes. However, the real pathogenesis is unknown.

Although cigarette smoking is an important factor for PSP in adults, the issue of children with PSP who are smokers has rarely been investigated.\(^3,4\) Furthermore, there is still no consensus about the seasonal distribution of prevalence of PSP.\(^5,6\) One recent study showed that neither particular months nor seasons of the year were significantly associated with the incidence of spontaneous pneumothorax.\(^7\) However, there were some limitations in the study. The authors or investigators did not separate pediatric patients from adult patients. Pediatric PSP, typically, is buried within adult series in the literature, with few purely pediatric series available.

The purpose of this investigation was to explore the demographic data, such as gender, age groups, BMI, smoking status, seasonal change, and outcomes of pediatric PSP patients.

2. Materials and Methods

2.1. Study population

This retrospective study was conducted between June 2004 and December 2009 in the Pediatric Division of Cheng Ching General Hospital. The medical records of 89 patients with a diagnosis of PSP (the International classification of diseases, 9th Revision, clinical modification = 512.8) were reviewed and discussed by three pediatricians and two internists. The confirmation of cases was based on radiological images and reports by radiologists, admission notes, internists. The confirmation of cases was based on radiological images and reports by radiologists, admission notes, internists.

Eleven patients, including five with chest pain symptom alone, two with pneumonia, two with diagnosis of pneumomediastinum, and the remaining two with premature condition, were excluded.

2.2. Measurements of variables

To measure the size and severity of pneumothorax, the method suggested by Rhea et al.\(^8\) was adopted. Severity was divided into two classes: (1) mild: involved area less than 25% of hemithorax and with mild signs or symptoms and (2) severe: greater than 25% of involved area and tension pneumothorax. Tension pneumothorax was defined as accumulating air in the pleural cavity to the degree that it compressed the heart, mediastinum, and trachea, and caused them to move away from the midline.

Treatment modalities included oxygen inhalation (OI), two types of chest drainage (chest tube and pigtail catheter), and video-assisted thoracoscopic surgery (VATS) with wedge resection surgical intervention. The three kinds of treatment options included (1) bed rest, with or without OI; (2) closed-tube drainage (CTD); (3) VATS, which was applied in case of recurrence or failure of treatments with OI and simple chest tube insertion. Chest tube and pigtail catheter were assigned as a single treatment modality because of their same draining function, except that the former had a better caliber than the latter.\(^9,10\)

BMI was calculated with the BMI table provided by the National Center for Health Statistics in collaboration with the National Center for Chronic Disease Prevention and Health Promotion (2000), USA.\(^11\)

The other study variables, such as smoking status, month of first onset (seasonal pattern or trend), length of stay, and recurrence rate were collected from medical records and confirmed by telephone survey. Monthly incidence was defined as the total monthly first-time attack of PSP.

2.3. Statistical analysis

All data were collected and statistical analyses were performed by using SPSS software (SPSS Traditional Chinese Version 12.0; Sinter Information Corp, SPSS Inc., Taipei, Taiwan). Data were reported as means ± standard deviation.

3. Results

3.1. Demographic characteristics

The 78 patients (57 inpatients and 21 outpatients) who were enrolled in this study had a PSP attack from June 2004 to December 2009. Table 1 summarizes their demographic characteristics, severity of pneumothorax, BMI, and others.

The male:female ratio was 7.7:1 (69:9), and the mean age was 16.76 ± 0.16 years. There was no patient younger than 11 years at our hospital in the past 5 years. The major presenting symptoms of PSP were chest pain (69, 88.5%); dyspnea (23, 29.5%); chest tightness (11, 14.1%); and cough (8, 10.3%). Among these 78 patients, in 43 (55.1%), the attack occurred at the left side of lungs; in 34 (43.6%), at the other side; and in only one patient (1.3%), a bilateral attack occurred simultaneously. Thirty-two (41.0%) patients were classified into mild degree (involved area, <25%), whereas 46 (59.0%) patients were assigned to "severe degree."

The mean BMI was 18.24 ± 1.63 kg/m\(^2\) (n = 66). Nine (11.5%) patients admitted to having smoking experiences. The mean seasonal case number of PSPs showed fluctuation and peak attacks in autumn (p = 0.005) during this 6-year period (Figure 1).

All of the 21 outpatients received supportive treatment. Out of 57 inpatients, 10 (17.5%) received oxygen therapy, 39 (68.4%) received CTD (24 with chest tube and 15 with pigtail catheter), and 6 (10.5%) received VATS (Table 2). Among 23 episodes of patients receiving VATS, 21 (91.3%) episodes’ pathologies were bleb(s) over their upper lobe of lung, and two episodes (8.7%) had blebs at the lower lobe.
Pneumothorax recurred in 28 patients (35.9%), including four outpatients and 24 inpatients. Twenty-eight patients had tension pneumothorax (35.9%). During their first attack, seven patients received OI therapy, 16 patients received close-tube drainage, and the other three patients received VATS; two patients’ data were incomplete. There were six patients who experienced a third attack. No mortality or morbidity was found in any of the patients. The lengths of stay of the three groups of treatment modalities (OI, CTD, and VATS) were 3.5 (0.85, SD) days, 7.95 (3.53, SD) days, and 13.67 (2.42, SD) days, respectively.

4. Discussion

PSP is rarely encountered in young children and is mostly found in teenagers.8,13,14 It is a male predominance, with a male-to-female ratio ranging from 1.6:1 to 8:1, and the ratio is 7.7:1 in this study.

In our patients, the mean BMI was 18.24 ± 1.63 kg/m², which is classified as underweight (<18.5 kg/m²).12 This is in accordance with the report of Cook et al,15 which showed that their patients had a median BMI of 18 kg/m² (their normal range is 20–25 kg/m²). It may reflect that PSP is prone to occur in slender children, probably because they have a higher transpulmonary pressure in their lung apex, or their rapid growth rate relative to pulmonary vasculature results in relative ischemia and bleb formation.16,17 There is no consensus regarding the seasonal distribution of the occurrence of PSP. Primrose2 reported that significantly fewer admissions were noted during May, June, and July. Celik et al5 reported that pneumothorax episodes were observed most commonly in June, August, and November, out of all the 12 months. On the contrary, some authors stated that the seasons of the year did not significantly influence the occurrence of PSP.6,7

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The seasonal variation in the incidence of spontaneous pneumothorax in Taiwan had been studied in a nationwide population-based study.7 The result did not support the involvement of seasonal factors in precipitating spontaneous pneumothorax. This result was quite different from that of our study. They did not confine their subjects to pediatric patients. They also did not include outpatients or separate primary from secondary spontaneous pneumothorax. In our study, PSP tended to occur in autumn.

Although many reports on PSP revealed that most of the adult PSP patients were smokers and there was an increased risk of PSP in smoking patients with a dose–response relationship,2–4 our study reveals only a few patients (only 9 patients, 11.5%) who are smokers. This may

Table 1 Demographic data of primary spontaneous pneumothorax patients

<table>
<thead>
<tr>
<th>Demographics and variables</th>
<th>All (n = 78)</th>
<th>Severe (n = 46)</th>
<th>Mild (n = 32)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Gender*</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>69 (88.5)</td>
<td>41 (89.1)</td>
<td>28 (87.5)</td>
</tr>
<tr>
<td>Female</td>
<td>9 (11.5)</td>
<td>5 (10.9)</td>
<td>4 (12.5)</td>
</tr>
<tr>
<td>Age (yr)*</td>
<td>16.76 ± 0.16</td>
<td>16.54 ± 1.25</td>
<td>17.07 ± 1.64</td>
</tr>
<tr>
<td>BMI (kg/m²)*,y</td>
<td>18.24 ± 1.63</td>
<td>18.41 ± 1.62</td>
<td>17.87 ± 1.62</td>
</tr>
<tr>
<td>Tension pneumothorax</td>
<td>28 (35.9)</td>
<td>28 (60.9)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Smoking</td>
<td>9 (11.5)</td>
<td>3 (6.5)</td>
<td>6 (18.8)</td>
</tr>
<tr>
<td>Location</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Left</td>
<td>43 (55.1)</td>
<td>24 (52.2)</td>
<td>19 (59.4)</td>
</tr>
<tr>
<td>Right</td>
<td>34 (43.6)</td>
<td>21 (45.7)</td>
<td>13 (40.6)</td>
</tr>
<tr>
<td>Bilateral</td>
<td>1 (1.3)</td>
<td>1 (2.1)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Presenting symptoms</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chest pain</td>
<td>69 (88.5)</td>
<td>40 (87.0)</td>
<td>29 (90.6)</td>
</tr>
<tr>
<td>Dyspnea</td>
<td>23 (29.5)</td>
<td>15 (32.6)</td>
<td>8 (25.0)</td>
</tr>
<tr>
<td>Chest tightness</td>
<td>11 (14.1)</td>
<td>7 (15.2)</td>
<td>4 (12.5)</td>
</tr>
<tr>
<td>Cough</td>
<td>8 (10.3)</td>
<td>4 (8.7)</td>
<td>4 (12.5)</td>
</tr>
<tr>
<td>Back pain</td>
<td>2 (2.6)</td>
<td>0 (0)</td>
<td>2 (6.3)</td>
</tr>
<tr>
<td>Shoulder pain</td>
<td>1 (1.3)</td>
<td>1 (2.2)</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>

* p Value of χ² test and t test did not reach a significant level of 0.05;

BMI = body mass index.

Figure 1 Seasonal pattern of first primary spontaneous pneumothorax attacks.
be because the prevalence of smoking in children is far lower than that in adults. According to Wen et al’s study, the prevalences of smokers in Taiwanese male and female adults were 46.8% and 4.3%, respectively, whereas in underage teenagers, they were 14.3% and 4.0%, respectively. Wang and Chou also found that the prevalence of smoking in Taiwanese students was only 5.7%, with male and female student percentages being 11.5% and 0.4%, respectively. This may be because smoking is prohibited by school authorities and social culture in Taiwan, and it implies that children’s PSP is smoke related, but not necessarily.

Apical bleb and subpleural bullae formation are the most common findings of PSP in children and in adults. Pneumothorax may occur when these peripheral bullae or bleb become distended and rupture into the pleural space. Although patients with PSP do not have apparent pulmonary disease, actually, subpleural bleb or bullae (usually on the apical portion of the upper lobes) are found in 76–100% of patients during VATS and in nearly all patients during thoracotomy. We have a similar result: among the 23 patients during VATS and in nearly all patients during thoracotomy.13 We have a similar result: among the 23 episodes of operation, upto 21 (91.3%) episodes were found to have bleb in their lung apex, and two (8.7%) episodes had bleb other than upper lobe.

Recurrence is a common and serious problem of PSP. In a report including 11 studies of PSP in which patients were treated by observation, needle aspiration, or chest tube drainage, the average rate of recurrence was 30%, with a range of 16–52%. In our study, 28 patients had a recurrence (20 had it once and 8 had it 2 or more times), with a total recurrence rate of 35.9%. Some authors reported an even higher recurrence rate (>50%) after conservative treatment (bed rest/oxygen therapy, CTD, or both) for pediatric PSP.

Surgical treatment is more invasive, but its recurrence rate is much less than that of the conservative treatment. Many authors asserted that patients who received VATS had a lower recurrence rate than those who received CTD or conservative treatment. The role of VATS in decreasing the recurrence rate of PSP has been getting attention in recent decades. According to a retrospective study of Hirai et al., the recurrence rates of their three treatment modalities (tube drainage, VATS, and limited thoracotomy) were 42%, 13%, 0%, respectively. Another study also reported a much lower recurrence rate in VATS group compared with the tube insertion group (10.3% vs. 54%). Their average recurrence rate of VATS was around 11.6%. In our study, there were 23 recurrences of VATS during the investigation period and five recurrences post—VATS therapy. The VATS recurrence rate was 21.7%. Some reports suggested that the probable factors of recurrence may include newly grown bullae, neglected bullae during previous operation, and failed pleurodesis, and pointed that it could be prevented by more extensive pleurectomy and further bullectomy during the subsequent session of VATS.

We conclude that pediatric PSP occurs mainly in boys of lower BMI in the late teenage group. Pediatric patients with PSP should be treated according to their severity of symptoms and size of the pneumothorax. There was only a small portion of the patients who smoked in this study. We did not find a correlation between smoking and pediatric PSP attacks. The PSPs were found to occur more during the autumn in the midst of Taiwan. We believe that more significant results could be observed if broader studies are conducted, and a better understanding of the clinical manifestations will assist us in managing the PSP.

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