What Factors Predict Recurrence after an Initial Episode of Primary Spontaneous Pneumothorax in Children?

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Purpose: Recurrence is the most common complication after an initial episode of primary spontaneous pneumothorax (PSP). However, preventive surgery in children remains a controversial issue. The purpose of this study was to determine predictive factors of recurrence to better inform early surgical referrals.

Methods: We retrospectively reviewed all consecutive patients under 18 years of age who conservatively treated for an initial episode of PSP between March 2005 and September 2011. The mean follow-up period was 43.1 months. Ipsilateral and contralateral recurrence developed in 47.3% and 14.0% of patients. The risk of ipsilateral recurrence for patients with or without air-containing lesions according to high-resolution computed tomography (HRCT) was 60.3% and 31.4%. In the multivariate analysis, the presence of air-containing lesions on HRCT scans and bullae on chest X-rays were independent risk factors for ipsilateral recurrence.

Conclusion: The presence of bleb or bullae on HRCT scans or chest X-rays after an initial episode of PSP was significantly related to the ipsilateral recurrence in children. If the risk factors are clarified in further studies, hospital stays and the recurrence of PSP after the first episode could be reduced with early video-assisted thoracoscopic surgery.

Keywords: primary spontaneous pneumothorax, adolescent, recurrence, bullae/bleb, retrospective study

Introduction

Most cases of primary spontaneous pneumothorax (PSP) occur in healthy adolescents and young adults in the absence of prior lung disease.1,2 The goal in the management of PSP is to treat the acute episode and prevent recurrence with minimal morbidity. The primary therapeutic strategy for the first attack of PSP is conservative treatment. However, the average rate of recurrence after the first episode of PSP with conservative treatment was
reported at 30%1;1 the rate appears to be higher in children, ranging from 50% to 60%.2-6 Surgery represents the most effective method for preventing recurrence. However, prevention of recurrence after an initial episode of PSP in children remains controversial.5-7 Establishing a standardised approach is difficult because the data in children are based on small studies.

The purpose of this study was to determine predictive factors for recurrence that could be useful for selecting patients who may benefit from surgical treatment for the prevention of recurrence after the first episode.

Materials and Methods

After obtaining institutional review board approval from the Catholic University of Korea, a retrospective review was conducted of all consecutive patients younger than 18 years of age who were admitted for an initial episode of PSP and who underwent high-resolution computed tomography (HRCT) at the Uijeongbu St. Mary’s Hospital between March 2005 and September 2011. The record of each patient was reviewed, and their gender, age, height, weight, body mass index (BMI), smoking history, laterality of PSP, degree of lung collapse, and therapeutic procedures were collected.

The size of the pneumothorax was semiquantitated after measuring the largest distance from the chest wall to the pleural line on the chest X-ray according to the following scheme: a designation of minimal pneumothorax was assigned when the largest distance was smaller than 1 cm, small was assigned when the largest distance was between 1 cm and 2 cm, moderate to intermediate was assigned when the distance was between the distance defined for small and large, large was assigned when the largest distance was greater than the width of the remaining lung, complete was assigned when the lung mass was restricted to the hilum, and tension was assigned when a mediastinal shift to the contralateral side was observed.

An asymptomatic patient with a minimal or small pneumothorax was treated with supplemental oxygen. A small pneumothorax in a symptomatic patient and a pneumothorax of any other size, regardless of symptoms, were treated by chest tube drainage. Surgical treatment at the first episode was undertaken in patients with a prolonged air leak of longer than 4 to 5 days, haemopneumothorax, tension pneumothorax, or visible bullae on the chest X-ray or HRCT scan. In cases of visible bullae, surgical interventions were planned according to the desire of the children and parents. Patients who underwent surgical interventions were excluded from the analysis.

HRCT scans were routinely performed using a slice thickness of 0.8 mm or 1 mm and section spacing of 3 mm. All HRCT images were analysed by the same radiologist and thoracic surgeon. By convention, a bleb is defined as an air collection <1 cm in diameter, and a bullae is defined as an air collection >1 cm in diameter. Dystrophic lesions were analysed in both the affected and the contralateral lung. The type, number, distribution (by side), and largest size of air-containing pulmonary lesions detected on HRCT scans were recorded for each patient. We adopted a scoring system to better assess the severity of pulmonary lesions, and this system has been previously described.5-10 This dystrophic severity score was calculated for each patient using the HRCT scans as follows: different values were assigned according to the type (1 or 2 points for blebs or bullae), number (1 or 2 points for single or multiple lesions), and distribution (1 or 2 points for unilateral or bilateral lesions) of the air-containing pulmonary lesions. This score has a maximum of 6 points and a minimum of 3 points. Points 3 and 4 were considered together as a low grade and points 5 and 6 were considered together as a high grade.

All patients were studied and treated for the first episode of PSP at our institution. Most patients presenting with a recurrence were referred to our institution; for patients who never had recurrence or who had recurrence but were referred to another centre, follow-up information was obtained by telephone interviews. For patients who did not experience recurrence, a minimum follow-up period of 12 months was established. Patients with follow-up of less than 12 months were considered as lost to follow-up and were excluded from the analysis.

All descriptive data were expressed as the frequency and mean ± standard deviation. Frequencies were compared using the \( \chi^2 \) or Fisher exact test for categorical variables, and continuous variables were compared using the independent two-sample t-test or the Mann–Whitney U test. Cox regression was performed to identify variables independently associated with the recurrence of pneumothorax. Differences were considered to be statistically significant when \( P <0.05 \).

Results

We reviewed 249 consecutive patients who were admitted with first time PSP to Uijeongbu St. Mary’s Hospital between March 1, 2005, and September 30, 2011. Patients who had undergone surgical interventions, who were lost
to follow-up, who transferred to another hospital immediately after receiving the diagnosis of PSP, and who refused a HRCT scan were excluded. After exclusion, 114 patients were identified. Of these 114 patients, ipsilateral recurrence occurred in 54 patients (47.4%). Of these, 10 patients (18.7%) also experienced contralateral recurrence after or before ipsilateral recurrence. Contralateral recurrence occurred in 16 patients (14.0%). Overall recurrence rate was 52.6%.

The mean age at the time of the first episode of pneumothorax was 16.9 ± 1.3 years (range, 11.4–18.7 years), and the patients were predominantly male (93.0%). The mean follow-up was 43.1 ± 20.4 months (range, 12 to 90 months). Initial episodes were treated with supplemental oxygen (n = 42) and chest tube drainage (n = 72). The mean hospital stay in the supplemental oxygen group was 4.5 ± 1.0 days (range, 3–12 days), and the mean duration of the chest tube drainage was 3.6 ± 1.0 days (range, 2–7 days).

The characteristics of the study population are listed in Table 1. The mean age was younger (16.5 ± 1.4 years vs. 17.1 ± 1.1 years, P = 0.012) and the mean BMI was lower (19.2 ± 2.0 kg/m² vs. 18.4 ± 2.0 kg/m², P = 0.043) in patients with ipsilateral recurrence compared to patients without recurrence. Contralateral recurrence was more common in patients with ipsilateral recurrence, but this difference was not statistically significant. The mean time to ipsilateral recurrence was 7.9 ± 9.8 months (range, 1 to 39 months), and the mean time to contralateral recurrence was 14.6 ± 11.1 months (range, 1 to 36 months).

Among the 114 patients, 14 (12.3%) presented with visible bullae on plain chest X-rays. Ipsilateral recurrence was more common in the patients with visible bullae on chest X-rays compared to those patients without bullae (22.2% vs. 3.3%, P = 0.003). Of the patients with bullae on the chest X-rays, 12 patients (12/14, 85.7%) experienced ipsilateral recurrence. Therefore, the risk of ipsilateral recurrence for patients with bullae on the chest X-ray was 85.7%; this finding indicates that the positive predictive value of the chest X-ray was 85.7%. The risk of ipsilateral recurrence for patients without visible bullae on the chest X-ray was 42.0% (42/100), indicating a negative predictive value of 58.0% for ipsilateral recurrence.

The risk of recurrence was significantly related to the presence of air-containing lesions on the HRCT scan. Of the 54 patients experiencing ipsilateral recurrence, 38 (70.4%) had air-containing lesions. Of the 63 patients with air-containing lesions, recurrence developed in 38 patients, while 25 patients did not experience recurrence. This result suggests that the risk of ipsilateral recurrence for patients with air-containing lesions on HRCT scans was 60.3%. However, the risk of ipsilateral recurrence for patients without air-containing lesions was 31.4%. Therefore, the positive predictive value of air-containing lesions was 60.3%, and the negative predictive value was 68.6%.

The size of the pneumothorax was not associated with the recurrence; however, the type, number, and distribution

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**Table 1** Comparison of clinical characteristics of patients with or without ipsilateral recurrence

<table>
<thead>
<tr>
<th></th>
<th>Patients without ipsilateral recurrence (n = 60)</th>
<th>Patients with ipsilateral recurrence (n = 54)</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, years</td>
<td>17.1 ± 1.1</td>
<td>16.5 ± 1.4</td>
<td>0.012</td>
</tr>
<tr>
<td>Male gender, n (%)</td>
<td>58 (96.7%)</td>
<td>48 (88.9%)</td>
<td>0.147</td>
</tr>
<tr>
<td>Height, m</td>
<td>1.73 ± 0.07</td>
<td>1.73 ± 0.07</td>
<td>0.958</td>
</tr>
<tr>
<td>BW, kg</td>
<td>57.7 ± 8.2</td>
<td>55.3 ± 7.3</td>
<td>0.107</td>
</tr>
<tr>
<td>BMI, kg/m²</td>
<td>19.2 ± 2.0</td>
<td>18.4 ± 2.0</td>
<td>0.043</td>
</tr>
<tr>
<td>Right side, n (%)</td>
<td>17 (28.3%)</td>
<td>16 (29.6%)</td>
<td>0.879</td>
</tr>
<tr>
<td>Smoking, n (%)</td>
<td>12 (20.0%)</td>
<td>6 (11.1%)</td>
<td>0.194</td>
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<tr>
<td>Therapeutic procedures</td>
<td></td>
<td></td>
<td>0.530</td>
</tr>
<tr>
<td>Oxygen supply or observation, n (%)</td>
<td>24 (40%)</td>
<td>18 (33.3%)</td>
<td></td>
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<tr>
<td>Chest tube drainage, n (%)</td>
<td>36 (60%)</td>
<td>36 (66.7%)</td>
<td></td>
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<tr>
<td>Contralateral recurrence, n (%)</td>
<td>6 (10%)</td>
<td>10 (18.5%)</td>
<td>0.191</td>
</tr>
<tr>
<td>Visible bullae on plain X-ray, n (%)</td>
<td>2 (3.3%)</td>
<td>12 (22.2%)</td>
<td>0.003</td>
</tr>
<tr>
<td>Presence of air-containing lesions on HRCT scans, n (%)</td>
<td>25 (41.7%)</td>
<td>38 (70.4%)</td>
<td>0.003</td>
</tr>
<tr>
<td>Size of the largest air-containing lesion, mm</td>
<td>4.17 ± 5.92</td>
<td>8.28 ± 8.07</td>
<td>0.002</td>
</tr>
<tr>
<td>Dystrophic severity score</td>
<td>2.07 ± 2.47</td>
<td>3.43 ± 2.38</td>
<td>0.004</td>
</tr>
<tr>
<td>Duration of follow-up, months</td>
<td>42.4 ± 21.4</td>
<td>43.9 ± 19.2</td>
<td>0.689</td>
</tr>
</tbody>
</table>

BW: body weight; BMI: body mass index; HRCT: high-resolution computed tomography
of air-containing lesions were associated with ipsilateral recurrence (Table 2).

The ipsilateral recurrence rate was higher in patients with high-grade scores (63.4% in patients with a high-grade vs. 54.5% with a low-grade vs. 31.4% with a score of 0, P = 0.007) (Table 2).

Risk factors for ipsilateral recurrence were identified by Cox regression analysis. The presence of air-containing lesions on the HRCT scans (Hazard ratio [HR] = 2.0; 95% confidence interval [CI]: 1.1–3.7; P = 0.030) and visible bullae on the plain chest X-rays (HR = 2.0; 95% CI: 1.0–4.0; P = 0.049) were independent risk factors for ipsilateral recurrence (Table 3).

Contrary to the findings for ipsilateral recurrence, contralateral recurrence was unrelated to the presence of visible bullae on the chest X-rays, air-containing lesion on the HRCT scans, or the dystrophic severity score (Table 2).

Discussion

The results of the present study have confirmed significant risk factors for ipsilateral recurrence present at the first attack of PSP in children. Ipsilateral recurrence was
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independently associated with the presence of visible bullae on chest X-rays and air-containing lesions on HRCT scans.

Although recurrent pneumothorax is not fatal, it is the most common complication after an initial episode of PSP. Moreover, recurrent attacks require rehospitalisation and additional hospital costs. Therefore, identification and documentation of risk factors for recurrence may reduce these additional burdens. The currently accepted treatment strategy for PSP in children is observation or tube thoracostomy at the first presentation, followed by video-assisted thoracoscopic surgery (VATS) for children in whom recurrent pneumothorax develops. However, current guidelines do not clearly describe the appropriate management of an initial episode of PSP in children, and the reason for the higher recurrence rate in children remains unclear.

The average rate of recurrent pneumothorax after the first episode of PSP with conservative treatment was reported at 30%; this rate appears to be higher in children, ranging from 50% to 60%.

Surgery is the most effective method for preventing recurrence. Many surgeons now regard VATS as the gold standard for the surgical management of PSP. In fact, VATS is associated with lower morbidity, decreased pain, and shorter hospital stays. Considering the higher rate of recurrence, especially in paediatric PSP, determining the predictive factors of recurrence could be useful for selecting patients who may benefit from early surgical referral to prevent recurrence and reduce hospital stays.

Although the pathophysiology of PSP remains unclear, ruptured apical bullae or blebs are generally accepted as the cause of PSP. HRCT has been recommended after initial presentation to identify bullae or blebs, which would then serve as an indication for VATS. Bullae or blebs are the aetiology of PSP. However, the role of bullae or blebs in the development of recurrent pneumothorax is unclear, and some authors proposed that the decision for surgical intervention should be based on documented recurrence only.

The incidence of blebs/bullae on HRCT scans in paediatric population studies was reported to range from 30.8% to 100%, and the recurrence rate after conservative treatment in patients with air-containing lesions was reported to range from 50% to 100% (Table 4). These results are consistent with the results of our study of an incidence of blebs/bullae on HRCT scans of 55.3% and the 60.3% recurrence rate associated with blebs/bullae on HRCT scans. However, prior studies in the paediatric population were limited by the small number of enrolled patients, which mitigates the significance of the association between air-containing lesions on CT scans and the recurrence of paediatric PSP. The adult literature has reported rates of air-containing lesions on CT scans of 47% to 88%. Moreover, some adult series documented that the air-containing lesions on CT scans were an independent risk factor.

The sensitivity of the plain chest X-ray for detecting the underlying cause of PSP is very low (approximately 15%). In this study, the presence of bullae detected by chest X-ray had a high predictive value for ipsilateral recurrence (Tables 2 and 3). Despite its low sensitivity, chest X-rays may be useful in decision making. If bullae are detected on an initial chest X-ray, then an early VATS may be considered to prevent recurrence. However, we did not find a significant difference between the groups with and without bullae detected on the chest X-rays.

During the follow-up period, 54 of 114 patients (47.4%) experienced ipsilateral recurrence. Air-containing lesions

<table>
<thead>
<tr>
<th>Table 4 Results of CT findings of primary spontaneous pneumothorax in children</th>
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<tbody>
<tr>
<td>No. of patients who underwent CT</td>
</tr>
<tr>
<td>---------------------------------------------</td>
</tr>
<tr>
<td>Our data</td>
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<tr>
<td>Seguier-Lipszyc (2011)</td>
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<tr>
<td>Laturu (2011)</td>
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<tr>
<td>Nathan (2010)</td>
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<tr>
<td>Bialas (2008)</td>
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<tr>
<td>O’Lone (2008)</td>
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<td>Guimaraes (2007)</td>
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<td>Choundhary (2005)</td>
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*: recurrence rate in patients treated conservatively. CT: computed tomography; NA: not available
were found on the HRCT scans of 63 patients (55.3%). The presence of these lesions was an independent risk factor for ipsilateral recurrence (Table 3).

Recent adult literature and this current study have shown that the risk of ipsilateral recurrence gradually increases with increasing dystrophic severity scores. However, this scoring system lacks clinical validation. If this score is validated in further studies, it would be a useful tool in selecting patients for early VATS.

Patients with ipsilateral recurrence had a smaller BMI (18.4 ± 2.0 kg/m² vs. 19.2 ± 2.0 kg/m², P = 0.043) and younger age (16.5 ± 1.4 years vs. 17.1 ± 1.1 years, P = 0.012) compared to patients without recurrence. Previous studies have suggested that growth during adolescence causes a rapid increase in the vertical dimension of the thorax compared to the horizontal dimension; this rapid increase causes an increase in negative pressure at the apex of the lung, which may lead to the formation of bullae and may cause PSP upon rupture. However, we cannot clearly state whether a smaller BMI and younger age affected contralateral recurrence.

Smoking has been shown to pose a 9-fold increased risk of pneumothorax in women and a 22-fold increase in men. However, this factor does not appear to be a major contributor in the paediatric series because most paediatric patients are non-smokers.

The prediction of a contralateral recurrence is another controversial issue. Some authors suggested the presence of air-containing lesions on the contralateral lung was an independent predictive factor for contralateral pneumothorax and suggested considering single stage bilateral VATS for PSP. Our data did not confirm these previous findings (Table 2). Bilateral air-containing lesions or other variables were not positively or negatively predictive of contralateral recurrence. In addition, only 16 patients (14.0%) had contralateral recurrence. Therefore, based on our results and the available reported data, the presence of bilateral air-containing lesions does not appear to be sufficient justification to recommend single stage bilateral VATS for PSP. VATS to prevent recurrence should be considered only for the affected side.

The main limitation of this study was its retrospective review of data that included 100 patients who underwent surgery at the initial presentation. Therefore, there was an inevitable selection bias. However, the large number of patients, the long term follow-up period, and the strict inclusion criteria enabled us to collect useful data. Most patients or their parents prefer not to have prolonged hospital stays to wait for air leaks to stop. Even after success of conservative treatment, there is a 30% to 60% risk of recurrence during an unpredictable amount of time. Therefore, most patients with air-containing lesions on HRCT scans or chest X-rays request early VATS to achieve early discharge and a lower chance of recurrence. In our previous study, the mean postoperative pleural drainage duration was 3.2 days, which was similar to the duration of conservative treatment. VATS is safe and is the most effective treatment method for PSP in children. Some authors have recommended that preventive VATS may be considered if air-containing lesions are detected on initial HRCT scans. However, this surgical approach for air-containing lesions to prevent recurrence has two major problems. First, the positive predictive value of air-containing lesions on HRCT scans in this study was 60.3%. This result indicates that if all patients with air-containing lesions were to have surgical interventions, approximately 40% would undergo an unnecessary surgical procedure. Second, postoperative recurrence in children was reported to be slightly higher than in adults.

Conclusion

The presence of air-containing lesions detected on HRCT scans or plain chest X-rays after an initial episode of PSP was significantly related to the development of an ipsilateral recurrence. In particular, the presence of bullae on chest X-rays may be useful for decision making. If the risk factors are clarified in further studies, hospital stays and the recurrence of PSP after the first episode could be reduced with early VATS.

Disclosure Statement

None.

References

5) Qureshi FG, Sandulache VC, Richardson W, et al.


