Management of the pediatric spontaneous pneumothorax: is primary surgery the treatment of choice?

Monica E. Lopez, M.D.*, Sara C. Fallon, M.D., Timothy C. Lee, M.D., J. Ruben Rodriguez, M.D., M.M.Sc., Mary L. Brandt, M.D., Mark V. Mazziotti, M.D.

Division of Pediatric Surgery, Michael E. DeBakey Department of Surgery, Baylor College of Medicine, Houston, TX, USA

**KEYWORDS:**  
Primary spontaneous pneumothorax; Pediatric; Video-assisted thoracoscopic surgery; Blebectomy; Mechanical pleurodesis

**Abstract**  
**BACKGROUND:** Surgery as the primary management strategy for pediatric primary spontaneous pneumothorax is controversial. This study aims to evaluate the outcomes and effectiveness of management approaches for pediatric spontaneous pneumothorax.

**METHODS:** Outcomes of pediatric patients undergoing initial nonoperative treatment versus video-assisted thoracoscopic surgery with blebectomy and mechanical pleurodesis were compared via a retrospective review.

**RESULTS:** We identified 96 patients with 108 pneumothoraces. Of 98 pneumothoraces with initial nonoperative management, 37% had surgery during their initial hospitalization for persistent air leak. Of those discharged home without video-assisted thoracoscopic surgery, 40% recurred. Initial nonoperative management resulted in more total hospital days (median: 11 vs 5 days, \( P \), .001). No significant predictors of recurrence were identified on multivariate analysis. Sixty-three percent of all patients ultimately required surgery.

**CONCLUSIONS:** Fewer than 40% of primary spontaneous pneumothorax patients are definitively treated with nonoperative management. A prospective study is needed to determine whether primary surgery with blebectomy/mechanical pleurodesis is a more effective treatment strategy.

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Primary spontaneous pneumothorax (PSP) is a relatively rare condition in the pediatric population, found in 3.4 per 100,000 children.1 The disease most commonly occurs in tall, thin teenage men, with a 4:1 male predilection.1,2 Children with connective tissue disorders and asthma are at an increased risk for developing PSP.3 Initial management typically consists of supplemental oxygen with timely evacuation of the PSP with needle aspiration, percutaneous placement of a drainage catheter, or tube thoracostomy. Hemodynamic compromise is rare as part of the initial presentation.

The initial management of a first episode of a pediatric spontaneous pneumothorax is a matter of controversy. Historically, all patients were managed nonoperatively...
with supplemental oxygen and tube thoracostomy, reserving surgical management via an open thoracotomy for recurrent or refractory cases. However, with the advent of video-assisted thoracoscopic surgery (VATS), definitive operative management of this disease can be achieved with decreased pain, length of hospital stay, and morbidity.5 As recurrence rates in the pediatric population are reported to be higher than 50%, some surgeons advocate for early VATS aiming for definitive disease resolution and earlier resumption of usual daily activities.4,5 Others recommend reserving surgery for those who fail nonoperative management because of either persistent communication with the pleural space or recurrent disease to avoid unnecessary surgery in a proportion of patients.6,7

The purpose of this study was to evaluate whether primary or delayed surgery is the preferable treatment for the pediatric patient with a spontaneous pneumothorax regarding complications after surgery, recurrence rates, total number of days of hospitalization, and cost of treatment. We hypothesize that in children with spontaneous pneumothorax, primary VATS blebectomy with mechanical pleurodesis is a more effective management strategy than nonoperative treatment, leading to lower complication and recurrence rates, as well as decreased total duration of hospitalization.

Methods

Patient population

After institutional review board approval at Baylor College of Medicine (H-29248), patients diagnosed with a PSP at our institution from 2005 to 2011 were included in the study. All the patients were initially diagnosed with a chest radiograph. Patients with a pneumothorax secondary to underlying lung parenchymal disease (cystic fibrosis), trauma, iatrogenic injury, or infection were excluded. Contralateral pneumothoraces were treated as distinct events. Charts were reviewed for data concerning demographics, initial management, imaging (computed tomography) use, operative details, complications, hospital course, and recurrence rates.

Patient management

The non-VATS approach consisted of administration of supplemental oxygen, insertion of percutaneous catheter, or tube thoracostomy. Percutaneous catheters were placed in the emergency department under conscious sedation. All tube thoracostomies were performed in the operating room. Chest computed tomography (CT) scan was obtained in some patients based on previous history of pneumothorax or physician preference. These were reviewed for the documented presence of unilateral or bilateral blebs. Operative management consisted of VATS, apical blebectomy, and mechanical pleurodesis. Chemical pleurodesis was reserved for management of persistent air leaks. Persistent air leak was defined as persistent bubbling of air within the Pleur-Evac water chamber upon eliciting positive pressure lasting for more than 4 days. Synchronous bilateral VATS, during the same anesthetic event, with therapeutic or prophylactic blebectomy took place for those presenting with bilateral disease in the form of either active bilateral pneumothoraces or unilateral disease with blebs detected contralaterally on CT scan.

Study design

The primary outcomes in this retrospective study were the recurrence rate, the surgical complication rate, the cumulative hospital length of stay, and the variable direct cost of treatment. Direct measurement of variable costs incurred, as opposed to charge-based or reimbursement-based accounting, was chosen as the most accurate measure of cost related to the 2 surgical treatment strategies.8,9 We compared patients who had a recurrence with those who did not based on demographic characteristics and initial management, which included supplemental oxygen only, tube thoracostomy, pigtail catheter, or primary surgery. A recurrent pneumothorax was defined as the presence of a new ipsilateral pneumothorax on chest radiograph after complete resolution of the previous pneumothorax episode. Complications after surgery were defined as a recurrence, a persistent air leak (>4 days) treated with surgery, wound infection, or empyema.

Chi-square and Student t tests were used to compare normally distributed data between these groups. Mann–Whitney U tests were used for nonparametric data. A time to event analysis using a Kaplan–Meier survival curve with a log rank test was performed to evaluate for factors influencing the time to recurrence. A logistic regression analysis was performed to assess for independent predictors of recurrence. Clinically relevant determinants of recurrence were included as variables in the multivariate model independently of reaching statistical significance in univariate analyses. All analyses were performed using SPSS Statistics for Windows, Version 19.0 (IBM Corp, Armonk, NY).

Results

During the study time period from 2005 to 2011, 96 patients had 108 pneumothoraces. The majority of patients were male (79%; n = 76), with a median age of 16.4 years (range 8.5 to 20.9) and median body mass index (BMI) of 18.8 (range 14.4 to 34.7). Associated comorbidities were noted in 13% (n = 12) and included asthma, pectus excavatum, Marfan syndrome, and immunosuppression. Smoking history was not routinely recorded in the medical records. The median initial length of hospital stay was 5.0 days (range 1 to 43), and the cumulative number of hospital days was 7.0 (range 1 to 57).
The initial management consisted of supplemental oxygen only in 23% \((n = 25)\), pigtail catheter in 22% \((n = 24)\), tube thoracostomy in 45% \((n = 49)\), and primary surgery in 9% \((n = 10)\) of patients. These 4 groups were similar with respect to sex, presence of an associated disease, age, and BMI (Table 1). The indications for surgery in patients managed initially with primary surgery included immunosuppression, history of contralateral disease, large extensive bullae on CT scan, remote history of Wilms tumor, and hemopneumothorax. CT scans were obtained in 45% of patients during their initial hospitalization. Sixty percent of patients were found to have blebs on CT scan, and of these, 60% had evidence of contralateral disease (17% of the total patient cohort). Eleven patients underwent synchronous bilateral VATS based on the presence of active bilateral pneumothoraces at presentation, or unilateral disease with contralateral disease, large extensive bullae on CT scan, remote history of Wilms tumor, and hemopneumothorax. CT scans were obtained in 45% of patients during their initial hospitalization. Sixty percent of patients were found to have blebs on CT scan, and of these, 60% had evidence of contralateral disease (17% of the total patient cohort). Eleven patients underwent synchronous bilateral VATS based on the presence of active bilateral pneumothoraces at presentation, or unilateral disease with contralateral asymptomatic blebs detected on CT scan. Among those with CT scans obtained during initial hospitalization or at the time of recurrence who also had surgical intervention, 98% \((59 \text{ of } 60)\) had a bleb identified during the operation.

Of the 98 pneumothoraces initially managed nonoperatively, the median number of days with a drainage catheter (before an operation or discharge) was 3 (range 1 to 10). Over one third (37%) of the patients failed nonoperative management and required surgery before discharge because of a persistent air leak. There was no difference in failure rates among the 3 nonoperative management groups (Table 1). Initial nonoperative management resulted in an increased median total hospital length of stay (11 vs 5 days, \(P < .001\)) compared with primary operative management.

Overall, 63% of patients underwent surgery for treatment of a PSP. There were no surgical complications (intraoperative injury, wound infection, empyema) in the 89 operations. After VATS, 12/89 (14%) patients had a persistent air leak for greater than 4 days. Ten of these patients (11%) returned to the operating room for a repeat VATS with repeat blebectomy and/or chemical pleurodesis with doxycycline. Two patients were successfully managed with doxycycline pleurodesis under conscious sedation. When comparing patients who had primary VATS with those who had surgery after initial nonoperative management, there was no difference in recurrence post-operation \((P = .15)\) or surgical complications \((P = .5)\).

The overall recurrence rate in either the ipsilateral or contralateral lung in our patient population \((n = 96)\) was 46%. After initial nonoperative management, 40% \((39/98)\) experienced an ipsilateral recurrence at a median time of 2.7 months (range 0.1 to 18.9). Using Kaplan–Meier log-rank analysis, there was no difference in the time to recurrence between the supplemental oxygen only, pigtail, and chest tube groups \((P = .27)\) (Fig. 1). The ipsilateral recurrence rate after surgical management was 14.6% \((13/89)\) at a median time of 3.0 months (range 0.67 to 37.2). There was a statistically significant difference \((P = .0003)\) between recurrence after nonoperative management (40%) and surgery (14.6%). On logistic regression analysis, neither age, sex, BMI nor comorbidity proved to be significant preoperative predictors of recurrence.

We evaluated the variable direct cost related to different treatment strategies. The median cost for patients treated with nonoperative management successfully (no recurrence or surgery for persistent air leak) was $2,443 (range $393 to $5,511). The median total variable direct cost of patients treated with initial surgery was $9,133 ($3,380 to $19,702). This is in comparison with patients treated with nonoperative management at their initial hospitalization who experienced a recurrence (median cost $9,349, range $3,733 to $25,040) and patients treated with nonoperative management who had a persistent air leak and required surgery at their initial hospitalization (median cost $9,246, range $4,124 to $44,015). The median overall variable direct cost of initial nonoperative management was less than that for initial surgery ($5,445 vs $9,133, \(P < .001\)).

**Comments**

Among a typical population of adolescent patients with spontaneous pneumothorax treated at a single center, initial nonoperative management was successful less than half of the time. Overall, initial nonoperative management was associated with an increased total length of stay but decreased variable direct costs. The majority of our patients were initially managed nonoperatively, mostly with insertion of a chest tube or percutaneous drain. Those

<table>
<thead>
<tr>
<th>Clinical/Outcome Variable</th>
<th>Primary surgery ((n = 10))</th>
<th>Oxygen only ((n = 25))</th>
<th>Pigtail catheter ((n = 24))</th>
<th>Chest tube ((n = 49))</th>
<th>(P) value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>70%</td>
<td>84%</td>
<td>71%</td>
<td>82%</td>
<td>.571</td>
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<tr>
<td>Comorbidity</td>
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<td>12%</td>
<td>21%</td>
<td>20%</td>
<td>.326</td>
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<tr>
<td>Age (mean)</td>
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<td>16.6</td>
<td>15.7</td>
<td>16.1</td>
<td>.330</td>
</tr>
<tr>
<td>BMI (mean)</td>
<td>18.8</td>
<td>20.1</td>
<td>18.6</td>
<td>20.4</td>
<td>.239</td>
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<tr>
<td>Surgery during initial hospitalization</td>
<td></td>
<td>32%</td>
<td>25%</td>
<td>45%</td>
<td>.216</td>
</tr>
<tr>
<td>Recurrence rate (%)</td>
<td>40%</td>
<td>40%</td>
<td>48%</td>
<td>27%</td>
<td>.313</td>
</tr>
</tbody>
</table>

*BMI = body mass index.*
Surgery was required in the management of PSP in 63% of our patients. The procedure was well tolerated without significant surgical complications. Our recurrence rate after surgery (14%) is comparable with other published reports (11% to 28%).

Various patient-based and technical factors have been proposed as contributors to the increased recurrence rate after surgery in children. These include the inability to discover blebs at operation, incomplete resection without demonstration of normal lung tissue at the margin, coexisting severe asthma, and formation of new bullae.

While some of the surgical technical factors could be addressed by changes to the operative technique, such as the extent and type of pleurodesis, type of endoscopic stapling device, and use of fibrin glue or mesh to reinforce the staple line, some hypothesize that the ectomorphic physical appearance of those children and their rapid growth may continue to affect the recurrence rates more so than surgical technique owing to the formation of de novo blebs after surgery.

The rate of postoperative air leak after VATS for PSP ranges in the literature from 2.7% to 13%.

However, in the majority of these studies, persistent air leak was defined as lasting for longer than 7 days. In our series, 11% of patients required reoperation during the same hospitalization for persistent air leak, defined as longer than 4 days. This lower cut-off in our definition of persistent air leak could explain the greater detection and earlier surgical treatment of these patients, leading to the increased direct variable costs observed in this group.

Our lower cut-off for persistent postoperative air leak (>4 days) parallels data from a consensus statement by the American College of Chest Physicians recommending surgery for adults with air leaks lasting longer than 4 days and for recurrent spontaneous pneumothorax.

The same expert panel further recommended limited continued observation for resolution of an air leak, as more prolonged delays may decrease the effectiveness of thoracoscopy and increase the cost of care. A pediatric evidence-based review has supported the American College of Chest Physicians recommendations, and other pediatric studies have yielded corroborative data suggesting that refractory air leaks are best treated surgically, because these are unlikely to close spontaneously after 3 days, and that the persistent need for thoracic catheter drainage indicates underlying lung pathology that is less likely to heal in children than in adults, also pointing to the benefits of early surgical intervention.

Beyond recurrence and adverse event rates, other important patient-centered measures should be considered to determine which treatment is superior for children with PSP. We found that primary surgery leads to a decreased total number of hospital days, as also reported by Qureshi...
et al. Primary surgery allows for early resumption of normal physical activity and decreased time spent in the hospital. However, few studies have formally examined the effects of different treatment strategies on quality of life in the pediatric patient. Using a Markov model, others have found that primary VATS for the management of PSP in young adults resulted in an increased quality-adjusted life expectancy 1 year after treatment compared with drainage catheter placement.

Cost effectiveness has been addressed in only a few studies. Cook et al. determined that surgical treatment of a recurrence, as opposed to continued nonoperative management, was a more cost-effective strategy. Others have favored secondary VATS after initial tube thoracostomy as the most cost-effective treatment in pediatric spontaneous pneumothorax. However, both these studies used total treatment charges as a measure of calculating cost. Charges and costs are frequently erroneously used interchangeably. It is well established that charges do not accurately reflect costs or reimbursements and should not be used as a proxy for costs. While it is known that charges overestimate cost, this discrepancy varies across hospitals and surgical procedures. This inherent variability makes them a poor marker of actual cost of care and can lead to unwarranted conclusions regarding economic efficacy. Our data did not demonstrate a significant direct variable cost difference in patients who were treated with initial versus delayed surgery. This is an expected finding because variable direct costs related to surgical care, in comparison to those associated with length of stay (ie, room and board), contribute more to the overall treatment costs within our institutional cost allocations system. Although initial surgery reduced resource utilization in terms of length of stay, this strategy did not confer overall cost savings, likely because of the costs of surgical care. Taheri et al. previously demonstrated that decreases to length of stay by as much as 1 full day have a minimal impact on overall costs, and that efforts aiming to reduce costs should be focused in the earlier stages of admission when resource consumption is most intense and involves expensive diagnostics and therapeutic interventions. Thus, implementing definitive surgical therapy early on in the course of disease while identifying patients most likely to respond to nonoperative management are plausible examples of future efforts to deliver cost effective care.

We acknowledge that the limitations of our study are related to its retrospective design, small cohort size, and potential problems estimating true recurrence rates as some patients could have been lost to follow-up or obtained further treatment at other institutions.

Nevertheless, the majority of patients were not successfully treated with a nonoperative approach, either because of a persistent air leak at initial presentation or a recurrence. The benefits of definitive primary surgery in terms of lower recurrence rate and decreased total hospital time can potentially mitigate its higher cost. This treatment strategy should be studied prospectively with attention to patient-centered outcomes such as quality of life. Identifying risk factors for adverse outcomes could help direct decision making and patient counseling. Clinical outcomes data presented in this study have helped shape our management algorithm and standardize family and patient information with regards to each treatment modality. Our findings support initial management with pleural catheter drainage, reserving CT scan for evaluation of recurrent disease, and early surgical intervention in the setting of failure of conservative management to achieve full resolution of persistent air leaks in pediatric patients with spontaneous pneumothorax. Larger prospective studies are needed to recommend whether primary or delayed VATS is the more effective treatment strategy for children with PSP.

References


