Original Article

Management and recurrence of spontaneous pneumothorax in children

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Abstract

Objective: No guideline clearly prescribes an approach to management of spontaneous pneumothorax in children. The objectives of this study were to evaluate practice variation in the management of spontaneous pneumothorax in children and its probability of recurrence.

Methods: This study was a retrospective chart review followed by a phone follow-up that included all children who had visited a tertiary care paediatric hospital for a first episode of spontaneous pneumothorax between 2008 and 2017. The primary outcomes were the management of pneumothorax (observation, oxygen, needle aspiration, intercostal chest tube, surgery) and the probability of recurrence. All charts were evaluated by a rater using a standardized report form and 10% of the charts were evaluated in duplicate. All children/families were contacted by phone to assess recurrence. The primary analyses were the proportions of each treatment modalities and recurrence, respectively.

Results: During the study period, 76 children were deemed eligible for the study. Among them, 59 had a primary spontaneous pneumothorax while 17 were secondary. The most common first therapeutic approaches were chest tube insertion (31), oxygen alone (27), and observation (14). A total of 54 patients were available for follow-up among whom a recurrence was observed in 28 (37% of the total cohort or 52% of available children).

Conclusion: Chest tube insertion was the first line of treatment in about 40% of children with a first spontaneous pneumothorax. In this population, the recurrence probability is established between 37 and 52% and the majority occurs in the following months.

Keywords: Children; Emergency medicine; Pneumothorax

Spontaneous pneumothorax affects approximately 4 per 100,000 children/year, with a male to female ratio of 2 to 4 to 1 (1–8). It is mostly seen in adolescents (2,5,7–9). While secondary spontaneous pneumothoraces occur in children with an underlying lung problem such as asthma, Marfan syndrome, or cystic fibrosis (3), primary spontaneous pneumothorax are of unknown etiology. The latter is most likely caused by the formation and rupture of a bleb/bubble which could be linked to the rapid growth of the pulmonary tissue (3,9).

The potential therapeutic approaches for primary and secondary spontaneous pneumothorax are diverse: (1) observation with or without high-flow oxygen (conservative treatment), (2) needle aspiration followed or not by the insertion of a pigtail catheter, (3) insertion of an intercostal catheter (chest tube), and (4) surgery (10). Successful treatment is defined as a positive lung re-expansion visible on a chest radiography (9). As opposed to adults, there is no generally accepted guideline or international consensus on the management of spontaneous

pneumothorax for children (2,3,6,11). Given the lack of recognized procedure and paediatric data, studies have shown that it is standard practice to extrapolate adult guidelines to children for their management (2,3,12).

In order to better understand the factors associated with success and recurrence, it is important to evaluate how children diagnosed with a spontaneous pneumothorax are currently managed. The first objective of this study was to evaluate the therapeutic approach used for the management of a first episode of spontaneous pneumothorax in children. The second objective was to measure the probability of recurrence. As an exploratory analysis, the performances of the various therapeutic approaches in terms of success, recurrence, hospitalization, and length of stay were compared.

METHODS

Study design and setting

This was a retrospective chart review followed by a prospective follow-up survey within a tertiary care, paediatric university-affiliated hospital for a 10-year period from January 1, 2008 to December 31, 2017. The emergency department had an average annual census varying between 70,000 and 84,000 with approximately 15 cases of pneumothorax per year. All children seen in this setting were initially managed by a physician with experience in paediatric emergency medicine.

Participants

All children below 18 years of age with a final diagnosis of first spontaneous pneumothorax at the emergency department were eligible, including transferred patients. For children with multiple episodes, only the first one was eligible. Exclusion criteria were: malignancy as the identified cause of the pneumothorax, traumatic pneumothorax, tension pneumothorax, surgical or iatrogenic pneumothorax, mechanically ventilated pneumothorax and neonates (postgestational age less than 1 month).

Outcomes

The primary outcome was the therapeutic approach used. Possible therapeutic approaches were observation with or without oxygen, needle aspiration followed or not by a pigtail insertion, intercostal chest tube insertion, and surgery. In cases of multiple treatments, all were documented, and the analysis was performed with the first-line and last therapy separately. The other primary outcome was recurrence defined by any new pneumothorax diagnosed at any setting and at any time following the successful treatment of a first episode. Secondary outcomes included success/failure, hospitalization, and the length of stay. Failure was defined as the need for a second intervention, failure of lung re-expansion, re-accumulation of air immediately after the chest tube removal or an air leak for more than 5 days.

Independent variables

The first independent variable was the type of pneumothorax. This was divided as primary (children without underlying lung disease) and secondary (children with an underlying lung disease) pneumothorax. For children with a past medical history of asthma, the pneumothorax was classified as 'secondary' if it occurred during an acute asthma exacerbation and 'primary' if it was not related to an exacerbation. Other independent variables were the demographic data (age and sex) as well as other clinical information such as the past medical history, pneumothorax side, physical exam including respiration rate and pulse oximetry, and size of the pneumothorax (small or large) as defined by the British Thoracic Society guideline (13). In this guideline, a large pneumothorax is defined by 'the presence of a visible rim of >2 cm between the lung margin and the chest wall (at the level of the hilum)' on chest radiography (13). The presence of blebs/bubbles was evaluated using plain radiography for most patients and thoracic CT scan for a minority of patients.

Procedure and data collection

All patients with a final diagnosis of pneumothorax on the emergency department computerized database were selected. Medical charts were evaluated for eligibility and reviewed to extract data related to the first episode of pneumothorax using a standardized case report form. To ensure reliability of the charting, a second co-author evaluated blindly a random sample of 10% of health records for inter-rater analysis.

For the recurrence evaluation, all charts were reviewed to assess any new consult for another pneumothorax following the first visit. If there was no new consult in the medical chart, the patient or his or her parents (if under 18 years old), was contacted by phone after being notified by mail. During the phone interview, the patient was asked about the occurrence of another episode of pneumothorax using a standardized questionnaire. For each participant, a maximum of three phone follow-up attempts were made at different times and days.

Data analysis

First, participants were dichotomized into either a primary or a secondary spontaneous pneumothorax. Double data entry verification as well as random verification (10% of the charts) were made to ensure the accuracy of the data abstraction. Inter-rater reliability measurement was done using the kappa score for categorical variables and intra-class correlation for continuous variables. A priori, it was decided to exclude variables having a Kappa score or an intra-class coefficient lower than 0.6.

The primary analyses were the proportions of each treatment modalities and recurrence, respectively. Then, for each therapeutic approach, the success rate, proportion of hospitalization, recurrence rate, and length of stay were reported using descriptive statistics (proportion, median, and quartiles). Secondary

analysis was conducted to evaluate the associations between therapeutic approach and (1) the success probability and (2) the size of the pneumothorax using chi-square (χ^2) statistics. Finally, secondary analyses were conducted using univariate and multivariable logistic regression to identify risk factors of recurrence. Potential predictors of recurrence were related to age, sex, size of the pneumothorax, type (primary versus secondary), presence of bubbles/blebs, and final therapeutic approach.

Ethics

The study protocol was evaluated and accepted by the Sainte-Justine Research Institute research ethics board. Owing the retrospective nature of this chart review, no written consent was required. All participants of the follow-up study received an explanatory letter prior to the phone interview. Also, verbal consent was obtained from the patient or his or her parent before the phone interview.

RESULTS

From January 1, 2008 to December 31, 2017, there were 142 diagnoses of pneumothorax made in the emergency department among 104 children. Among them, 76 were deemed eligible for this study (Figure 1). The most common reasons for ineligibility were recurrent episodes ($n=40$, 32%) and traumatic pneumothorax ($n=10$, 7%). The charts of all eligible and ineligible children were available and reviewed with no missing data. As shown in Figure 2, primary pneumothorax occurred mostly in adolescents while secondary pneumothorax occurred in younger children. Of the 76 spontaneous pneumothorax episodes, 59 (78%) were classified as primary while 17 (22%) as

secondary. In addition, 60 (79%) were adolescents between the ages of 14 to 17 years old and 61 (79%) were male (Table 1).

Fifteen eligible medical charts were evaluated by two reviewers to assess inter-rater reliability. With the exception of history of smoking, level of shortness of breath and trigger, all other characteristics were deemed reliable (Supplementary Appendix Table 1).

The two most common first therapeutic approaches used were chest tube insertion ($n=31$, 41%) and oxygen therapy alone ($n=27$, 36%). However, no patient had needle aspiration alone (Table 2). A conservative approach defined as observation with or without oxygen was the first treatment used for 32 (54%) children with primary spontaneous pneumothorax. No difference was found in the success rate of the various first therapeutic approaches for either the primary ($\chi^2=4.92$, 3 Degree of Freedom [DF], $P=0.18$) or secondary spontaneous pneumothorax ($\chi^2=2.55$, 2DF, $P=0.28$). Sixty (79%) children were hospitalized for a duration varying between 1 and 20 days distributed as follows: 44 with a primary (75% of all primary) and 16 with a secondary pneumothorax (94% of all secondary) (difference: 19%; 95% confidence interval [CI]: 4 to 33%). Eight children had a surgery as a definitive therapy (six with blebectomy and two with blebectomy combined with pleurodesis). Thirteen out of 14 (93%) children who were initially treated with a chest tube were hospitalized for more than 1 week and surgery was associated with a longer hospitalization overall.

The size of the pneumothorax was associated with the therapeutic approach for both primary ($\chi^2=13.41$; 3DF; $P=0.004$) and secondary pneumothorax ($\chi^2=7.47$; 2DF; $P=0.024$). More specifically, patients with a small pneumothorax ($n=20$) were initially observed with ($n=12$, 60%) or without oxygen ($n=6.30\%$), while chest tube insertion was the most common

Figure 1. Flow diagram of the participants.

Figure 2. Age distribution of participants according to the type of pneumothorax. Acute disease = Asthma or pneumonia. CF Cystic fibrosis; PTX pneumothorax.

Table 1. Baseline characteristics and pneumothorax information (n=76)

	PSP N=59 (%)	SSP N=17 (%)	All N=76 (%)
Median age in years of the first episode (Q1: Q3)	16 (15: 16)	4 (2: 14)	15 (14: 16)
Sex male	50 (85)	11 (65)	61 (79)
Past medical history			
Asthma	17 (27)	9 (64)	26 (34)
Marfan syndrome	0 (0)	2 (3)	2 (3)
Cystic fibrosis	0 (0)	1 (2)	1 (1)
Presenting symptoms			
Chest pain	58 (98)	0 (18)	61 (79)
Shortness of breath	30 (51)	15 (88)	45 (59)
Cough	16 (27)	15 (88)	31 (41)
Median respiratory rate (/min) (Q1: Q3)	20 (16: 20)	34 (23: 56)	20 (16: 24)
Median pulse oximetry (Q1: Q3)	100 (98: 100)	95 (91: 97)	99 (97: 100)
Side of pneumothorax			
Left	38 (64)	14 (82)	52 (68)
Right	17 (29)	3 (18)	20 (26)
Bilateral	4 (7)	0 (0)	4 (5)
Size of the pneumothorax*			
Small	11 (19)	9 (53)	20 (26)
Large	48 (81)	8 (47)	56 (74)
Presence of Bubbles or Bleb	22 (37)	3 (18)	25 (33)

PSP Primary spontaneous pneumothorax; SSP Secondary spontaneous pneumothorax.

*According to the British Thoracic Society guidelines.

primary therapeutic approach for large pneumothorax (n=29, 52%). Twenty-three (41%) children with a large pneumothorax were treated conservatively where 16 (70%) of them were successful.

Most participants were contacted by phone. However, 22 patients could not be reached either because we did not

have valid contact information, or because they did not answer after three attempts. In total, recurrence was observed in 28 (37%) patients giving a probability of recurrence between 37% (of all eligible children) and 52% (of all followed participants). Recurrences were reported between 0.5 and 83 months following the first pneumothorax and the median

Table 2. Therapeutic approach used and outcomes for children with a primary and secondary spontaneous pneumothorax

PSP (N=59)	N (%)	Success (probability of success)	Hospitalization rate (%)	Median duration of hospitalization in days (Q1: Q3)	Recurrence rate (%)
Primary therapeutic used					
Observation only	13 (22)	12 (92)	1 (8)	3	1 (8)
Oxygen	19 (32)	13 (68)	16 (84)	2.5 (1.3: 5)	9 (47)
Pigtail	4 (7)	4 (100)	4 (100)	3 (1.5: 5.3)	3 (75)
Chest tube	23 (39)	15 (63)	23 (100)	4.5 (2.8: 8.3)	12 (52)
Surgery	0	-	-	-	-
Last therapeutic used					
Observation only	12 (20)	12 (100)	0 (0)	0	1 (8)
Oxygen	13 (22)	13 (100)	10 (77)	2 (1: 2.3)	5 (38)
Pigtail	7 (12)	7 (100)	7 (100)	3 (3: 5)	5 (71)
Chest tube	20 (34)	20 (100)	20 (100)	4 (2: 6)	10 (50)
Surgery	7 (12)	7 (100)	7 (100)	11 (8: 17)	4 (57)
SSP (N=17)	N (%)	Success (probability of success)	Hospitalization rate (%)	Median duration of hospitalization in days (Q1: Q3)	Recurrence rate (%)
Primary therapeutic used					
Observation only	1 (6)	1 (100)	0 (0)	-	-
Oxygen	8 (47)	8 (100)	8 (100)	3 (2: 4.5)	1 (13)
Pigtail	0	-	-	-	-
Chest tube	8 (47)	6 (75)	8 (100)	7 (3.5: 9.8)	2 (0.75)
Surgery	0	-	-	-	-
Last therapeutic used					
Observation only	1 (6)	1 (100)	0 (0)	-	0
Oxygen	8 (47)	8 (100)	8 (100)	3 (2: 4.5)	1 (13)
Pigtail	0	-	-	-	-
Chest tube	7 (41)	7 (100)	7 (100)	7 (3.5: 9)	2 (29)
Surgery	1 (6)	1 (100)	1 (100)	10	0

PSP Primary spontaneous pneumothorax; SSP Secondary spontaneous pneumothorax.

delay was 1 month (first quartile: 0.7 month and third quartile: 7 months).

The probability of recurrence was higher for children with primary spontaneous pneumothorax (45%), and Marfan syndrome- or cystic fibrosis-related pneumothorax (66%) compared to spontaneous pneumothorax secondary to an acute illness (7%) for a difference of 36% (95% CI: 9 to 50%).

Using univariate logistic regression, we identified that males (odds ratio [OR]: 4.8; 95% CI: 1.00 to 23.3), primary spontaneous pneumothorax including Marfan syndrome and cystic fibrosis (OR: 10.0; 95% CI: 1.34 to 81.5), large pneumothorax (OR: 4.57; 95% CI: 1.20 to 17.4), and bubbles/blebs identified on radiological evaluation (OR: 5.78; 95% CI: 2.04 to 16.4) were associated with a higher risk of recurrence. Age was not a factor (Table 3). Observation was associated with a lower risk of recurrence. However, there was no association between the

last successful therapeutic approach and the risk of recurrence. On multivariable logistic regression, only the presence of bubbles/blebs remained statistically associated to the risk of recurrence (Table 3).

DISCUSSION

This study reported that a conservative approach was used in approximately half of the children diagnosed with a spontaneous pneumothorax while a chest tube was inserted in 40% of them. The therapeutic approach was associated to the size of the pneumothorax but not to the success probability. Using a prospective phone survey, we identified a probability of recurrence between 37 and 52% for children with spontaneous pneumothorax. Recurrence was most common in children with cystic fibrosis, Marfan syndrome, or idiopathic pneumothorax and in children with thoracic blebs/bubbles.

Table 3. Logistic regression for the association between predictors and risk of recurrence

	OR (95% CI) Univariate	OR (95% CI) Multivariate
Age higher than 9 years old	6.4 (0.76–53.5)	
Sex male	4.8 (1.00–23.3)	0.50 (0.08–3.2)
PSP*	10.0 (1.34–81.5)	6.78 (0.66–69.3)
Large pneumothorax	4.57 (1.20–17.4)	1.00 (0.14–7.18)
Presence of Bubbles/blebs	5.78 (2.04–16.4)	3.42 (1.03–11.3)
Last treatment		
Observation	0.08 (0.007–0.98)	0.15 (0.009–2.67)
Oxygen	0.40 (0.08–2.14)	0.88 (0.11–6.79)
Pig tail	2.50 (0.29–21.4)	2.95 (0.28–30.64)
Chest tube	0.80 (0.17–3.89)	1.31 (0.14–7.17)
Surgery	1.00 ref	1.00 ref

CI Confidence interval; OR Odds ratio; PSP Primary spontaneous pneumothorax.

*Including idiopathic, and those associated with Marfan syndrome or cystic fibrosis.

The therapeutic approaches distribution reported in our study were similar to previous research in which 83% of the children diagnosed with a first episode of spontaneous pneumothorax were treated conservatively. However, our distribution is completely different than a retrospective American study, which reported that 20% of the children hospitalized for a first episode underwent surgery (14). When analyzing the success rates with conservative approaches, our results are in concordance with the BTS guidelines, which suggest a conservative approach for small pneumothoraces and patients with minimal symptoms (13). Since 2010, the BTS guidelines suggest needle aspiration for patients with large primary spontaneous pneumothorax with incommoding symptoms and discharge for those who are asymptomatic. Small-bore intercostal chest tube insertion is recommended by the BTS solely for treatment failure by the BTS. We found no use of needle aspiration in our study population. This is surprising given that previous studies showed that needle aspiration is safe, efficient, and associated with less hospitalizations and shorter length of stay (15–18).

Our recurrence rate is in agreement with the interval of 40 to 60% reported in the medical literature (1–4,19–23). The only strong predictive factor of recurrence identified by multivariate analysis was the presence of blebs/bubbles. This result agreed with the theory stating that primary spontaneous pneumothorax is most likely caused by the formation and rupture of a bleb/bubble associated with a rapid growth of the pulmonary tissue (3,9).

Our results will impact clinical practice and help anticipatory guidance for children with spontaneous pneumothorax. Wide practice variation highlights the need for standardized paediatric guidelines to manage children with spontaneous pneumothorax. Owing to the fact that primary spontaneous pneumothorax is mainly seen in middle to late adolescents, paediatric guidelines should be derived from the adult ones.

However, this would need to be studied in a multicentre prospective research study because of the low incidence of occurrences. The high success rate for conservative approaches as first-line therapy suggests that it could be used in children with small or large pneumothorax without incommoding symptoms as suggested by adult guidelines (13). The high probability of recurrence among children with lung blebs/bubbles raises the question of whether the initial investigation of a primary spontaneous pneumothorax should include a CT scan (5) However, we would not expect to do a 'prophylactic' surgery in patients with blebs/bubbles.

Finally, the results suggest that patients with secondary pneumothorax can be divided into two groups based on the underlying lung disease. Children with pneumothorax secondary to an acute disease were mostly younger than 6 years of age, necessitated a conservative approach and had a very low probability of recurrence. However, those (only three patients) associated with a chronic disease had characteristics similar to a primary spontaneous pneumothorax. This, however, is limited by the small number of patients and should be viewed as an exploratory result.

Limitations

There are limitations to this study. First, it was conducted in a single hospital that may not reflect management strategies used in other settings. However, we identified wide practice variation and one would expect that inclusion of a single setting would decrease variability because of local practices/guidelines. While evaluating all pneumothoraces managed in a busy paediatric emergency department over 10 years, the number of participants was small. Consequently, no conclusions can be drawn for secondary spontaneous pneumothorax. In addition, statistical analysis of primary spontaneous pneumothorax was limited. Owing to its retrospective design, the study was limited

to the quality of information reported in the medical charts. The radiological evaluation of the children was not standardized and the proportion of children who had CT scan was low. Finally, we were not able to reach all participants to assess for recurrence. However, all their charts were evaluated, and one would expect that the children would have returned to the same hospital in case of recurrence. However, this cannot be assumed, especially for older children who may have been treated at an adult hospital.

CONCLUSION

This study showed wide practice variation. For small primary spontaneous pneumothorax, conservative approaches were prioritized. For larger ones, medium size chest tube insertion was the most common procedure despite the fact that is no longer recommended in adult guidelines.

The recurrence rate for children with spontaneous pneumothorax varied between 37 and 52%. A strong predictive factor of recurrence is the presence of blebs/bubbles. In the future, the impact of the therapeutic approach on recurrence risk should be evaluated prospectively.

Finally, in our sample, the characteristics of children with a spontaneous pneumothorax secondary to a chronic lung disease such as Marfan syndrome or cystic fibrosis were more similar to those of an idiopathic primary spontaneous pneumothorax rather than those secondary to an acute asthma exacerbation. This should be further evaluated in a larger study, possibly leading to a new classification of secondary pneumothorax based on the chronicity of the underlying lung disease.

SUPPLEMENTARY DATA

Supplementary data are available at *Paediatrics & Child Health* Online.

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