

Case Series

Understanding Primary Spontaneous Pneumothorax Management and Outcomes in Children: A Case Series

Sara M Touzinsky, MD^{1*}; Beth Rymeski, DO²; William D Hardie, MD^{3,6}; Adam A Vukovic, MD, MEd^{4,6}; Eric J Crotty, MD⁵; Paria M Wilson, MD, MEd^{4,6}

¹Department of Pediatrics, Division of Emergency Medicine, Nationwide Children's Hospital, Columbus, OH, USA.

²Department of Surgery, Division of General and Thoracic Surgery, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA.

³Department of Pediatrics, Division of Pulmonary Medicine, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA.

⁴Department of Pediatrics, Division of Emergency Medicine, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA.

⁵Department of Radiology, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA.

⁶University of Cincinnati College of Medicine, Cincinnati, OH, USA.

***Corresponding Author: Sara M Touzinsky**

Department of Pediatrics, Division of Emergency Medicine, Nationwide Children's Hospital, 700 Children's Drive, Columbus, OH, USA.

Tel: 614-722-4385, Fax: 614-722-4380;

Email: sara.touzinsky@nationwidechildrens.org

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Abstract

Background: Recent literature describes variability in management of primary spontaneous pneumothorax (PSP), with evolving evidence that conservative management is non-inferior to intervention.

Methods: Retrospective case series of PSP cases presenting to two emergency departments (ED) between 2014 and 2021. We included all patients <21 years presenting to the ED with first time episode of PSP. We described frequency of tube thoracostomy (TT), length of stay (LOS), recurrence rates, rate of tension pneumothorax, and disposition.

Results: Of the ninety-five cases of PSP reviewed, 82 received oxygen and 48 underwent TT. LOS was seven times longer for patients who underwent TT compared to those who did not. Overall recurrence rate was 31%; 38 % in those who underwent TT and 23% in those who didn't. No patients developed tension physiology. There was significant variability in the decision to perform TT and caliber of tube placed, oxygen administration, and disposition.

Conclusions: Lower recurrence rate and shorter LOS for patients who did not undergo TT for PSP combined with the lack of tension physiology support shifting management away from TT in children. The inability to reliably calculate pneumothorax size in pediatric patients emphasizes the importance of standardizing care and limiting unnecessary procedures.

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Introduction

Primary spontaneous pneumothorax (PSP) is defined as accumulation of air in the pleural space in patients without a history of underlying lung disease [1]. Varying hypotheses exist as to why it occurs, including spontaneous rupture of blebs [2], the impact of greater distending pressures to the lung apex during growth spurts, or pleural porosity [3,4]. PSP is most prevalent in thin adolescent males [5] who typically present with acute onset unilateral chest pain and less frequently dyspnea and cough [6].

While PSP is a well-defined pathology in pediatrics, there is marked variability in management strategies, including rate and route of oxygen administration [7], use of suction [8], and placement of small versus large bore tube thoracostomies [9,10]. A recent survey of North American Surgeons demonstrated significant variability in the management of PSP [10]. Approximately 78% of pediatric patients with PSP undergo intervention [3,5,6,11-14]; however recent literature questions the need for interventional management. In 2020, Brown et al. published an open-label, multi-center non-inferiority trial of 316 patients 14-50 years of age with PSP, demonstrating that conservative management (analgesia and oxygen use) was non-inferior to placement of a small-bore chest tube when evaluating lung re-expansion within 8 weeks [15]. Furthermore, patients with conservative management experienced fewer hospitalized days, fewer adverse events and a lower likelihood of recurrence [15]. A recent meta-analysis supports conservative management by demonstrating equivocal risk of PSP recurrence when comparing conservative management to tube thoracostomy. A lower risk of adverse events was demonstrated in the conservative management group with no difference noted in the rate of PSP resolution [16].

Brown et al. provides strong evidence that conservative management is safe in the adult population, but the median age was 26 years old with a standard deviation of 8 raising concern for how this extrapolates to a pediatric population [15]. Our first aim was thus to determine the safety of translating the recommendations surrounding conservative management to the pediatric population [15]. We also hoped to fill the gap of primary literature focusing solely on the pediatric population [17]. There have been no recent pediatric studies evaluating current practice surrounding PSP and establishing baseline data prior to transition to conservative management. Specifically, the last case series of pediatric PSP were published in 2015 from Australia [5] and the United Kingdom [11], prior to the new evidence on conservative management. Given this gap surrounding management of pediatric patients, significant management variation and emerging data of non-inferiority of conservative management, we sought to describe the characteristics and outcomes of patients with PSP presenting to our pediatric emergency department (ED) from 2014-2021.

Methods

Study setting and design

We conducted a descriptive study of pediatric patients diagnosed with primary spontaneous pneumothorax who presented to the ED from 2014 to 2021. Cincinnati Children's Hospital Medical Center (CCHMC) is a 600-bed quaternary care pediatric

referral center with EDs in two freestanding children's hospitals, one urban and the other suburban. The hospital has approximately 100,000 emergency department (ED) visits and 20,000 admissions annually. The ED is staffed by pediatric and emergency medicine residents supervised by pediatric emergency medicine fellows and attendings. Additionally, clinical staff (nurse practitioners and pediatricians) see patients independently. Our institutional review board approved the study prior to commencement and granted waiver of informed consent (IRB ID 2021-0553).

Sample description and data collection

All children less than 21 years of age who presented for care to either ED were eligible for inclusion. Eligible patients were identified using International Classification of Diseases, 10th revision (ICD-10) codes for pneumothorax (93.9,93.11,93.83, 95,811,93.12,93.0). A detailed electronic medical record (EMR) review was performed to identify patients with a first-time episode of x-ray confirmed pneumothorax as read by a pediatric radiologist. Secondary pneumothorax, including trauma, predisposing underlying lung disease and infection were excluded. Underlying lung disease was defined as any lung condition that predisposed to pneumothorax, including asthma, congenital diaphragmatic hernia, tuberous sclerosis. Additionally, patients were excluded if the pneumothorax developed while inpatient or management was performed at an outside facility.

Data were collected through manual chart review. Variables extracted from the electronic medical record included patient age, sex, triage vital signs, and disposition. We subcategorized age less than 10 as the incidence of PSP is known to be very low in this age group. Chief complaint was extracted from the provider note. We utilized the initial chest x-ray (CXR) to determine pneumothorax laterality. Determination of pneumothorax size was at the discretion of the radiologist, either subjective or using objective measurements [18-20]. We defined small pneumothorax as a radiologist reading of tiny, trace, small, small to moderate or moderate pneumothorax. Large pneumothorax was defined as moderate to large or large according to radiologist read. Hypoxia was defined as an oxygen saturation less than 90% on room air. Oxygen use was defined as receipt of any amount of oxygen via nasal cannula, oxymask or non-rebreather. Patients were deemed to have tension physiology if they had hypotension associated with tachycardia [21]. The tube thoracostomy procedure note was reviewed for catheter size, use of anxiolytics or analgesics, and primary proceduralist. Small bore chest tube was defined as <14 French [22]. Length of stay (LOS) was calculated from time of hospital admission to time of discharge as noted in the encounter timeline. All imaging and procedure notes were reviewed to establish if computed tomography (CT) or video-assisted thoracic surgery (VATS) occurred during the initial encounter. All subsequent encounters were reviewed for recurrence defined as any future occurrence of pneumothorax on CXR after a CXR demonstrated complete resolution of initial pneumothorax. Patients were marked as having a follow-up CXR if an x-ray was performed at CCHMC within 8 weeks of initial presentation [15].

Statistical analysis

Descriptive statistics (counts and percentages) were used

to summarize patient characteristics and management strategies. Continuous variables were described using means and range. All analyses were performed in Microsoft Excel.

Results

A total of 381 patients were evaluated for eligibility, 95 of which had PSP and were included in the analyses (Figure 1). Two hundred and six patients were excluded for secondary pneumothoraces: 94 had recurrent disease, 9 developed a pneumothorax in the setting of cardiopulmonary resuscitation, 17 had an associated pleural effusion, 37 had traumatic processes, 30 had an underlying lung disease, and 19 had a lower respiratory tract infection at the time of pneumothorax. Table 1 displays the characteristics of the study sample.

Chief complaint consisted of chest pain (n=79, 83%), dyspnea (n=4, 4%), shoulder, arm or back pain (n=6, 6%), cough (n=2, 2%), foreign body sensation (n=1, 1%), chest palpitations (n=1, 1%), abdominal pain (n=1, 1%) and incidental pneumothorax found on x-ray of spine (n=1, 1%). None of the patients in the study sample were hypoxic, but 86% received oxygen therapy. Seven patients were discharged home after observation in the ED. The remainder were admitted to pediatric surgery (n=76, 80%), hospital medicine (n=8, 8%), pulmonology (n=2, 2%), or hematology (n=1, 1%) services. One patient required admission to the pediatric intensive care unit for sedation in the setting of significant anxiety. Thirty-eight chest tubes were placed in the ED; 17 by ED providers and 21 by surgery providers. Ninety-five percent (n=18) of patients with large pneumothorax had tube thoracostomy compared to 39% (n=27) of those with small pneumothorax. No patients had tension physiology. One patient was admitted with a plan for interventional radiology (IR) to perform a tube thoracostomy. Nine patients were initially admitted for observation but due to enlarging or unchanged pneumothorax on repeat CXR, tube thoracostomy was performed by IR. Time between initial CXR and the CXR that prompted tube thoracostomy ranged from 6 to 39 hours. All chest tubes placed by IR required general anesthesia, while the ED used ketamine (n=29, 76%), opiates (n=8, 21%), or midazolam (n=1, 3%). In the 48 patients who had a chest tube placed, 31(65%) had documented administration of local analgesia. Forty-six patients with tube thoracostomy received low wall suction. During the course of treatment, including follow-up x-rays, patients received on average 6.4 CXRs.

Overall, recurrence rate was 31% (n=29), 38 % in those who underwent tube thoracostomy and 23% in those who did not. After discharge from the hospital (n=14) or the ED (n=1), 15 (16%) patients returned to the ED within 8 weeks for pneumothorax related complaints. Return reasons included recurrence after full resolution noted on CXR (n=9), chest pain/costochondritis (n=1), post-surgical pain (n=1), enlarging pneumothorax requiring admission for observation (n=1), and enlarging pneumothorax warranting chest tube placement (n=3). Of the 4 patients who returned for enlarging pneumothorax, none had concern for tension physiology at time of re-presentation. Twenty patients had a PSP recurrence 9 weeks or greater from initial PSP. Timing of recurrence ranged from 9 weeks to 2 years. Management of recurrent PSP was variable with 7 patients (35%) undergoing conservative management, 1 patient (5%) undergoing tube thoracostomy, 11 patients (55%) having VATS, and 1 patient's (5%) management was unknown as treatment occurred outside of our institution.

Table 1: Summary characteristics of patients with first encounter for PSP (N=95).

Patient characteristics	N(%)
Presentation	
Age in years (mean, range)	16.3, 8-20
Age less than 10 years	1(1)
Male	83(87)
Pneumothorax laterality on CXR	
Right	30(32)
Left	61(64)
Bilateral	4(4)
Pneumothorax size	
Small	69(73)
Large	19(20)
No size description	7(7)
Triage Vitals (mean, range)	
Heart rate	89, 51-150
Respiratory rate	20, 10-36
Pulse oximetry	99, 92-100
Systolic blood pressure	125, 89-175
Diastolic blood pressure	75, 43-106
Management	
Received oxygen	82(86)
Tube thoracostomy	48(50)
Small bore	33(69)
Large bore	14(29)
Size not documented	1(2)
Discharged home from ED admitted	7(7)
Admitted after tube thoracostomy or planned placement	39(44)
Admitted for observation	40(45)
Converted from observation to tube thoracostomy	9(10)
Length of stay for admission (mean hours, range)	
Tube thoracostomy	118,12-407
No tube thoracostomy	16.7,7-32
Chest computed tomography scan	19(20)
VATS	22(23)
Follow-up	
Patients with 1 recurrence	29(31)
Follow-up x-ray in 8 weeks	71(75)

*CXR:chest x-rays; VATS: video assisted thoracoscopic surgery.

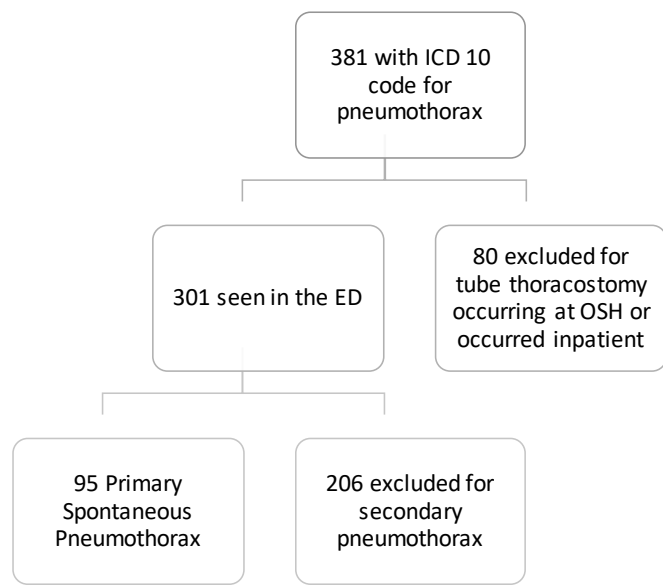


Figure 1: Overview of cases reviewed through manual chart review.

Discussion

Our retrospective case series of 95 pediatric patients with PSP is the largest in the United States and provides several insights into the presentation, management and outcomes of patients evaluated in our pediatric EDs. The predominantly adolescent male population and recurrence rate observed are consistent with prior published studies [5,23]. As predicted, there was significant variation in the management of PSP regarding oxygen administration, procedural intervention, and disposition with a resultant wide range in mean LOS. Most notably, no patients developed tension physiology.

Patients with PSP are frequently placed on oxygen due to a theoretical nitrogen washout leading to an increased rate of resolution, which was initially proposed in 1932 [24] and further explained by an animal model in 1995 [25]. Evidence on the effect of oxygen administration on rate of resorption and how it relates to pneumothorax size is contradicting, with some proposing a greater effect on small pneumothoraces [26] and others postulating that effect on large pneumothoraces [27]. Park et al. retrospectively reviewed PSP cases to evaluate rate of resorption on room air vs oxygen [7]. They found a minimally increased rate of resolution with oxygen administration, but these findings were confounded by a selection bias as patients with large pneumothoraces received oxygen while those with small pneumothoraces were managed outpatient. There have been no prospective studies to evaluate indications for oxygen therapy in PSP and no standardized recommendations for route or fraction of inspired oxygen to administer. Thus as expected, while most patients in our study were placed on oxygen, the route and fraction of inspired oxygen was inconsistent ranging from nasal cannula to 15 liters per minute via non-rebreather. The initiation and discontinuation criteria for oxygen administration were unclear, which is note-worthy given the lack of hypoxia in our patient population.

It is also important to note in our study, only a minority admitted for observation eventually required tube thoracostomy and that none of these patients developed tension physiology after deferring initial tube thoracostomy. No patients returning to the ED with post-discharge enlarging pneumothorax developed tension physiology. We speculate that the fear of development of tension physiology likely impacts clinician deci-

sion to perform tube thoracostomy. Recent evidence indicates that development of tension physiology is extremely rare in PSP and potentially not physiologically possible in a spontaneously breathing patient due to an inability for the pressure in the pneumothorax to exceed 1 standard atmospheric unit [28]. Combined, these findings support the consideration of observational management in the pediatric population [15,16].

Leading societies disagree on the preferred method for pneumothorax size calculation. The British Thoracic Society (BTS), American College of Chest Physicians, and the Belgian Society of Pulmonology all utilize unique definitions of small versus large pneumothorax that rely on single measurements from CXR [18,19]. The Collin's formula is an additional measurement strategy that requires 3 measurements but it has only been studied in a small sample of adults and not yet validated [20]. The BTS calculation differentiates small versus large pneumothorax based on less than or greater than 2 cm from chest wall to lung margin. No standardized measurement strategy has been developed for the pediatric population, as varying chest size in a growing child limits its feasibility. Pneumothorax size calculations in our study were at the discretion of the radiologist and no uniform calculations were used. Pneumothorax size is a significant factor in management decisions thus we hypothesize this led to variability in decision to pursue tube thoracostomy. The subjective nature of pneumothorax size is a major barrier to standardizing PSP care and a vital gap in literature highlighted by our case series.

Amongst the patients who received tube thoracostomy, catheter size, use of local analgesia and suction, and decisions leading to disposition were not uniform. Variability surrounding catheter size and the use of large bore catheters leads to increased patient discomfort without benefit as larger catheter size has not been shown to have improved efficacy [29].

With recent evidence regarding the safety of conservative management in PSP, it is imperative to consider standardization efforts for the vulnerable pediatric population. Standardization has been shown to decrease hospital LOS, use of CT scans, and average admission cost without altering rates of recurrence [9]. In our population, rates of recurrence were higher in the tube thoracostomy group compared to the group who underwent observation which is consistent with recent literature [15]. Furthermore, ambulatory management is cost-effective when compared to traditional treatment with tube thoracostomy and inpatient admission [30]. Standardizing indications for placement of tube thoracostomy and size considerations decreases associated complications [31]. Further research is needed to consider needle aspiration as an alternative to tube thoracostomy and failed aspiration as an indicator to proceed to VATS in the pediatric population [32].

Our study has multiple limitations related to its retrospective single center design, including lack of generalizability. However, given our ED volumes and the 7-year time span covered, we developed one of the largest case series ever studied [23]. We were limited by the documentation available in the EMR which prevented reporting factors not reliably charted, such as chest tube duration, smoking/vaping status, and family history of pneumothorax. Outside of pneumothorax size, we did not investigate factors associated with conservative versus interventional treatment. Additionally, as this was a retrospective study, follow-up data was limited to what was available in the EMR at our institution. While in an ideal situation, the same radiologist would interpret every CXR, this does not happen in clinical prac-

tice and our study reflects normal care delivery.

Conclusion

In conclusion, the significant variation in the management of pediatric PSP globally and within our institution necessitates development and implementation of evidence-driven expert consensus guidelines using quality improvement methodology. Continued investigation into reliable methods of estimating pneumothorax size in the pediatric population are necessary.

Declarations

Data availability statement: The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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Contribution statement: Dr. Touzinsky participated in conceptualization, data acquisition and analysis, interpretation of the data, drafting the manuscript and providing final approval. Dr. Rymeski, Dr. Hardie, Dr. Crotty and Dr. Vukovic assisted in conceptualizing this study, revised the manuscript and approved the final version. Dr. Crotty assisted in data analysis, revision of the manuscript and approval of the final version. Dr. Wilson designed the study, assisted with data collect, analysis and interpretation, drafted the manuscript and approved the final version.

Competing interests: The authors have no conflicts of interest to declare.

Consent statement: Consent was not required for this work.

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