

# TREATMENT OF PRIMARY SPONTANEOUS PNEUMOTHORAX IN PEDIATRIC PATIENTS: 15-YEAR EXPERIENCE AT A SINGLE-INSTITUTION

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## ABSTRACT

**Background:** Primary spontaneous pneumothorax (PSP) is a relatively uncommon condition in children. Due to the lack of pediatric-specific guidelines the treatment strategy varies among different centers. This study demonstrates a single-institution experience in the treatment of primary spontaneous pneumothorax in pediatric patients.

**Materials and methods:** Retrospective review of 49 patients with the diagnosis of primary spontaneous pneumothorax between 2003 and 2018 who were treated conservatively or invasively at the surgical department of Children's Hospital Zagreb.

**Results:** During the period of 15 years there were 49 patients noted with spontaneous pneumothorax at the surgical department of Children's Hospital Zagreb. The patient age ranged from 11 to 18 years (mean 15.28). 36 patients were male and 13 female, with a male to female ratio of 2.77:1. Pneumothorax occurred on the left side in 31 patients, on the right side in 15 patients, and on both sides in 3 patients. Clinical presentation in all patients was sudden, powerful chest pain. Other symptoms include dyspnea, cough. In all patients, a plain x-ray was made, and later the vast majority of patients underwent computed tomography (CT) scan. The CT scans detected 13 cases of apical bullae, 2 large bullae, 1 bulla in 6. segment and 1 parenchymal inflammation. Eight patients with stable clinical presentation and small pneumothorax underwent hospital observation. Eighteen patients were successfully managed with chest tube drainage without recurrence. Video-assisted thoracoscopic surgery (VATS) was performed on 19 patients with only two recurrences. Open thoracotomy was performed on 4 patients.

**Conclusion:** Due to the variation of diagnostic and therapeutic approaches from different centers, the creation of guidelines and standardized practice for the pediatric patient is necessary.

## Keywords:

Primary spontaneous pneumothorax, Pediatric, Video-assisted thoracoscopic surgery

## INTRODUCTION

Pneumothorax is defined as a collection of air in the pleural space with secondary lung collapse [1]. Pneumothorax is divided into two main groups: spontaneous and nonspontaneous. Spontaneous pneumothorax is classified into primary spontaneous pneumothorax (PSP), which occurs in healthy patients without underlying lung disease, and secondary spontaneous pneumothorax (SSP) which occurs with preexisting lung diseases such as cystic fibrosis, asthma, congenital cystic adenomatoid malformation, connective tissue disease, malignancies, and other disorders [2,3]. Nonspontaneous pneumothorax may be iatrogenic or caused by trauma [4].

Primary spontaneous pneumothorax is a relatively uncommon condition in the pediatric population, with an incidence of 3.4 per 100,000 children [4]. The disease most commonly occurs in tall, thin adolescent males between 15-17 age, with a male to female ratio of 4:1 [5,6]. There are no large studies showing a combined ratio PSP to SSP [5]. Smaller studies on 162 patients suggest that PSP is more common than SSP, with the presentation of PSP in 120 and SSP in 42 patients [7]. Also in the same studies small PSP (66%) were more common than large ones (34%) [7].

The size of the pneumothorax is divided into small and large pneumothorax. According to the British Thoracic Society (BTS) small pneumothorax is defined as the presence of a visible rim less than 2 cm between the lung margin and the chest wall (at the level of the hilum), where in the large pneumothorax rim is more than 2 cm [8].

The first line in the diagnostic approach is chest X-ray, but the use of computed tomography (CT) scan in the initial presentation of PSP is controversial. Some centers perform routine CT scan in patients with PSP to detect abnormal lung findings, while in other CT is performed in recurrent PSP or in a patient with an atypical presentation [2,3,6,9,10,11].

The management strategy varies between different centers due to the lack of evidence-based guidelines [3,6,9,12]. Treatment of the spontaneous pneumothorax varies from a conservative observation approach to the tube thoracostomy, video-assisted thoracoscopic surgery (VATS) and thoracotomy. Most often the first line of treatment in

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**DOI:** 10.5281/zenodo.4010895

small pneumothorax is intensive observation while in large pneumothorax the tube thoracostomy with observation is indicated [2,3,8,6,9]. The nonoperative approach was associated with higher recurrency rate > 40% [2,6,9,13]. The first method of choice in surgical intervention is VATS [2,3,8]. Video-assisted thoracoscopic surgery has proven to be safe and effective compared to open thoracotomy, with decreased postoperative pain and morbidity. Low recurrence rates (10%–13%) were conceted to VATS [2,6,9]. Open thoracotomy remains a method of choice in the case of giants bullas and blebs that are not positioned in the apical part of the lung [2,14].

## MATERIAL AND METHODS

Paper is a retrospective review of all patients (49 patients) with a diagnosis of spontaneous pneumothorax between the years 2003. and 2018. who were treated conservatively or invasively at the surgical department of Children's Hospital Zagreb.

The data was analyzed using descriptive statistics to demonstrate demographic and baseline patient characteristics (age, sex), initial clinical presentation, hospital course, procedures, imaging, outcomes, length of hospitalization and follow up period. Results are expressed as the means, medians, ranges, percentages and proportions.

All patients under 18 years old with confirmed diagnoses of PSP between 2003–2018 were included. Patients with secondary spontaneous and nonspontaneous pneumothorax were excluded. PSP was classified as small if the visible rim was less then 2cm between the lung margin and the chest wall on the x-ray scan, where the large PSP is more than 2cm [8]. Depending on the side of PSP we distinguish left, right and simultaneous bilateral in which bilateral PSP includes patients with PSP on one side and second presentation on the contralateral side, and patients with a simultaneous presentation on both sides. Recurrent PSP patients are described as the occurrence of new ipsilateral PSP. Indication for CT scan was confirmed PSP with x-ray imaging [2,7,12]. CT scan was noted as positive if CT scan has abnormal findings.

The patient was described as undergoing hospital observation if they did not undergo any procedure or operation, and only observation of vital signs, oxygen saturation, and control plan x-ray was done. Hospital observation was indicated in patients with small PSP and minimal symptoms [2,8,9].

Patients were described as undergoing thoracostomy if they underwent thoracostomy tube placement, aspiration, and observation without any additional intervention. Tube thoracostomy was indicated in unstable symptomatic patients and in large PSP [2,8,9]. The size of the tube and location of thoracostomy was choosed depending on the age and size of the patient. In patients younger than 14 years old small chest

drain (No.14) was inserted, while in 14 years and older patients larger tube were used (No.20) [2]. In all patients, thoracostomy was done with low-pressure suction with pressures between -10 and -20cm H<sub>2</sub>O [2]. The most common placement of the drain was between 4<sup>th</sup> and 6<sup>th</sup> intercostal space [2, 8,9].

Patients were described as undergoing operative management if they underwent VATS or open thoracotomy. VATS were indicated in the case of a persistent air leak (rang 5–6 days) after chest tube drainage, recurrence and in all patients with apical bullae [2, 8, 9]. VATS was performed with a stapler wedge resection of the apical part of the lung with an endostapler device (articulating endoscopic linear cutter with 45mm staple line) and mechanical pleurodesis or pleurectomy [2]. In children with giants bullas and blebs that are not positioned in the apical part of the lung, open thoracotomy with resection of bullas was indicated. The incision was adjusted to the localization and size of the lesion. Upper posterolateral open thoracotomy was most commonly applied [2,14].

## RESULTS

There were 49 patients with spontaneous pneumothorax, and recurrence was noted in 14 patients. The patient age ranged from 11 to 18 years (mean 15.28) with the majority between 15 and 17 years (81.6%). Thirty-six patients were male and 13 female with a male to female ratio of 2.77:1. Pneumothorax occurred on the left side in 31 patients, on the right side in 15 cases and it was bilateral in 3 patients. The follow-up period was from 4 months to 5 years (median 3.6 years). Length of hospital stay ranged from 4 - 29 days with a median of 11 days. Clinical presentation in all patients was sudden, powerful chest pain. Other symptoms included dyspnea and cough. In all patients, we made a plain X-ray, followed by CT scan. Chest CT scans were indicated in all patients, but were performed on 35 patients (71%). In 13 cases CT has shown apical bulla, 2 large bullae in the lower part of the lung, 1 bullae in 6. segment, and 1 parenchymal inflammation.

Eight patients with stable clinical presentation and small pneumothorax underwent hospital observation. Eighteen patients with unstable symptomatic presentation and with large PSP underwent thoracostomy. The mean duration of the chest drainage was 5 days. In 18 patients with tube drainage, the resolution of the pneumothorax and air leak with re-expansion of the lung were followed (Figure1.). VATS was performed in 19 patients. Three patients had pneumothorax on both sides. One case of simultaneous presentation of small PSP on both sides underwent hospital observation. Two had pneumothorax on both sides, but not simultaneously, and 2 separate VATS procedures were done in each case (Table 1).

We had only 2 recurrences after VATS, in one case with small pneumothorax recurrence, the intensive

observation was indicated and in other case open thoracotomy was indicated. There was no complication related to the VATS approach.

Four patients underwent open thoracotomy, from which one after VATS recurrence and other 3 had pneumothorax with lung collapse. After a CT scan investigation in patient with lung collapse, lung bulla was found. A fourteen-year-old boy had 1,5cm bulla in 6. lung segment, a 17-year-old boy had a bulla of 9 cm diameter, and a 12-year-old girl had a bulla of 5 cm diameter. In these children, VATS wasn't performed because the location of pneumothorax was not on the apical part of the lung and we considered that bullas were too large for endostapler wedge resection. There was no complication related to the open thoracotomy approach.

## DISCUSSION

Spontaneous pneumothorax is a very serious medical condition, and intensive care with hospitalization is necessary for all patients [2]. The generally accepted cause of PSP is the rupture of subpleural bulla [10,7]. Some studies suggest that bullas are cause of PSP in 60% [7]. The demographic and baseline characteristics of PSP patients were similar to other studies with characteristic male to female prevalence and with similar age interval presentations with the majority between 15-17 years [5].

Due to the lack of pediatric-specific guidelines, the diagnostic approach and treatment strategy varies from different centers. The initial treatment of PSP is based on clinical presentation and the degree of lung collapse on imaging. Management recommendations of PSP in the pediatric population are based on adult British Thoracic Society (BTS) guidelines [7].

Indication for CT scan in the first episode remained controversial. While some studies suggest CT scan at initial presentation [2,7,12,13], others are reserving it for atypical presentation and recurrences [10]. In our center, a CT scan was indicated at the initial presentation of PSP with aim to identify the presence or absence of abnormal pulmonary findings [2,7]. Studies have suggested that only a few pediatric cases of PSP were without identifiable pathology and that in pediatric patients there is a higher rate of bulla than it is in adults [7,15]. Early detection of the bullae or blebs can improve the operative management of PSP and eventually reduce the recurrence rate [7]. Other studies detected that CT has low sensitivity (36%) for the detection of blebs and therefore limited diagnostic value, that why CT scan is an option only in recurrent and atypical patients with the aim to avoid potentially unnecessary ionizing radiation [10,12].

The majority of surgeons treat the first episode of PSP nonoperatively. Patient with small pneumothorax and stable clinical presentation require only an observation

and short-interval follow-up, while those who are unstable or those with a large pneumothorax requires chest tube drainage with suction and observation [8,9,2]. The nonoperative approach was associated with higher recurrency rate > 40% [6, 9].

Regardless of the imaging, surgeons most often relay on clinical presentation, post drainage persistent air leak, incomplete lung re-expansion, and recurrence rate as the main indication for surgical intervention. Majority of surgeons recommend VATS intervention in persistent air leak in initial hospitalization, but number of days before recommending operation varies from 3 to 5 days. Also VATS is mainly indicated at the first episode of PSP with bullas in CT scan, and in patients with recurrence [8,9,2]. Nowadays surgical management of PSP is apical wedge resection of blebs/bullae and pleurodesis or pleurectomy typically performed using VATS. Video-assisted thoracoscopic surgery has been a safe and effective way of treating PSP, it has many advantages over open thoracotomy [8,9,2], and it's associated with low recurrence rates (10%–13%) [2,9]. Our studies also detected a lower recurrence rate in the VATS approach.

Open thoracotomy is performed in the case of giant bullae and bullae that are not positioned in the apical part of the lung [2]. Open thoracotomy was done in 4 patients.

In Children's Hospital Zagreb over a period of 15 years there were 49 patients with PSP. In 8 patients with stable clinical presentation and small pneumothorax, only short-interval follow-up was done. 18 patients were successfully managed with chest tube drainage without recurrence. Chest-tube suction was recommended for at least 5 days. VATS were performed on 19 patients with only two recurrences. Open thoracotomy was done in 4 patients. VATS is the gold standard in surgical management of PSP due to its many advantages compared to open thoracotomy.

## CONCLUSIONS

The use of computed tomography at the initial presentation remains controversial due to the lack of reliable results. While in management of patients with initial presentation of small stable PSP there is consensus, management of patients with large unstable PSP varies between different centers. Due to the variation of diagnostic and therapeutic approaches at the initial presentation from different centers, the creation of guidelines and standardized practice for the pediatric patient are necessary.

## CONFLICT OF INTEREST:

**The authors declare that there is no conflict of interest.**

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## FIGURES AND TABLES



**Figure 1.** Left- chest tube drainage of pneumothorax. Right- post drainage resolution of the pneumothorax and air leak with re-expansion of the lung.

**Table 1**

Patient demographics, clinical characteristics, and management	
<b>Patient demographics</b>	All patients (n=49)
Gender (M/F)	36/13
Age (mean, range), years	15.28 (11-18)
<b>Clinical characteristics</b>	
PSP location	31 left 15 right 3 bilateral
<b>Management</b>	
CT scen:	35
-apical bullae	13
-large bullae	2
-not apical bullae	1
-parenchymal inflammation	1
Hospital observation	8
Tube drainage	18
VATS	19
Open thoracotomy	4
PSP, primary spontaneous pneumothorax, CT, computed tomography, VATS, video-assisted thoracoscopic surgery	