Early Approach to Primary Spontaneous Pneumothorax Treatment in Children

Çocuklarda Primer Spontan Pnömotoraks Tedavisinde Erken Yaklaşım

Hayriye Nihan KARAMAN AYYILDIZ[1], Ceyhan SAHIN[1], Mehmet ARPACIK[1], Zeliha AKIS YILDIZ[1], Semih MIRAPOGLU[1], Fatma Tugba GUVENC[1], Aysel YUCAK OZDEMIR[2], Zekeriya ILCE[1], Aytekin KAYMAKCI[1]

[1]Umraniye Training and Research Hospital, Department of Pediatric Surgery, Umraniye, Istanbul, Turkey
[2]Sakarya University Training and Research Hospital, Department of Pediatric Surgery, Sakarya, Turkey

ABSTRACT

Objective: Etiology, diagnosis, and treatment of primary spontaneous pneumothorax (PSP) are not well-established and subject to debate in the pediatric age group. Our study aims to clarify the subject and discuss it in the light of available literature.

Material and Methods: We performed a retrospective analysis of the age, sex, etiology, smoking habits, diagnostic methods, pneumothorax percentages, first treatment approach, and treatment results of the patients with PSP by examining the patients' records. A total of 71 patients, 65 (91.5%) male and six (8.5%) female, who were followed up and treated between 2010 and 2020 were included in the study. Descriptive statistical methods, Shapiro-Wilk test Mann-Whitney U test, Pearson chi-square test, Fisher's exact test, Fisher-Freeman-Halton exact test were used while evaluating the study data.

Results: The mean age of the patients was 16.23±0.81 (13–18 years). The etiology was not clear in most of the patients. However, among them, 14 (25.5%) patients had bullae and 23 (32.4%) patients had a smoking habit. The diagnosis was made by means of taking medical history, physical examination, and post-anterior (PA) chest X-ray. Treatment with nasal oxygen was initiated in 14 (19.7%) patients with a pneumothorax percentage <20%. For a total of 57 patients the first line of treatment was initiated with tube thoracostomy. Video-assisted thoracoscopic surgery (VATS) was performed in cases where tube thoracostomy failed.

Conclusion: Different forms of initial treatment modalities exist for spontaneous pneumothorax. However, we suggest that the first option in patients of the pediatric age group should be clinical follow-up and supportive treatment, if necessary, tube thoracostomy should be applied.

Key Words: Primary spontaneous pneumothorax, Nonoperative, Recurrence, Pediatric

ÖZ

Amaç: Çocuk yaş grubunda Primer Spontan Pnömotoraks (PSP) etyolojisi, tanı ve tedavisi henüz kesinleşmemiş ve tartışmalı bir konudur. Makalemizde konuya açıklık getirme ve literatür eşliğinde tartışılması amaçlanmıştır.

0000-0003-0180-5022 : KARAMAN AYYILDIZ HN
0000-0003-3101-3915 : SAHIN C
0000-0001-7149-5627 : ARPACIK M
0000-0001-7637-6512 : AKIS YILDIZ Z
0000-0002-0122-7294 : MIRAPOGLU S
0000-0002-9464-9737 : GUVENC FT
0000-0001-5579-5160 : YUCAK OZDEMIR A
0000-0002-3473-5051 : ILCE Z
0000-0002-6147-5566 : KAYMAKCI A

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Correspondence Address / Yazışma Adresi:
Hayriye Nihan KARAMAN AYYILDIZ
Umraniye Training and Research Hospital,
Department of Pediatric Surgery, Ümraniye İstanbul, Türkiye
E-posta: nkaraman99@hotmail.com

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Gereç ve Yöntemler: "Primer Spontan Pnömotoraks (PSP) nedeniyle takip ve tedavi ettiğimiz hastaların yaş, cinsiyet, etyoloji, sigara alışkanlıkları, tanı yöntemleri, pnömotoraks oranları, ilk tedavi yaklaşımımız ve sonuçları hastaların kayıtları incelenerek değerlendirildi. 2010-2020 yılları arasında takip ve tedavi edilen 65'i (%91.5) erkek, 6'sı (%8.5) kız, toplam 71 hasta çalışmaya alındı. Çalışma verileri değerlendirilirken tanımlayıcı istatistiksel metodlar, Shapiro-Wilk testi, Mann-Whitney U test, Pearson ki-kare test, Fisher's exact test, Fisher-Freeman-Halton exact test kullanıldı.

Bulgular: Hastaların yaş ortalaması 16.23±0.81 (13–18 years)'di. Hastaların çoğunda etyoloji belli değildi. Ancak 14 (%25.4) hastada bül saptanırken, 23 (%32.4) hastanın hikayesinde sigara içme alışkanlığı saptandı. Tanı, hikâye, fizik muayene ve PA Akciğer grafisi ile kondu. Pnömotoraks oranı %20'nin altında olan 14 (%19.7) hastaya nazal oksijen ile tedaviye başlandı. Toplam 57 hastaya ilk tedavi olarak tüp torakostomi uygulandı. Tüp torakostominin başarısız olduğu olgulara video yardımlı torakoskopik cerrahi (VATS) uygulandı.

Sonuç: Spontan pnömotoraks başlangıç tedavi yöntemleri arasında farklı uygulamalar mevcuttur. Ancak çocuklarda ilk seçenek klinik takip ve destek tedavisi gerekirse tüp torakostomi uygulaması şeklinde olmalıdır. Bu uygulamalar kolay, başarı oranı yüksek, güvenli ve etkili bir tedavi yöntemidir. İleri cerrahi uygulamalar seçilmiş olgularda yapılmalıdır.

Anahtar Sözcükler: Primer Spontan Pnömotoraks, Nonoperatif, Nüks, Çocuk

INTRODUCTION

Spontaneous pneumothorax is defined as the collection of air between parietal and visceral pleural layers resulting in the sudden onset of collapsed lung. The occurrence of a pneumothorax without any lung disease is called primary spontaneous pneumothorax (PSP), and its occurrence due to an underlying lung disease is called secondary spontaneous pneumothorax (SSP) (1).

The main cause of PSP in adults is air leakage following the rupture of subpleural bullae and blebs located at the apex of the lung. The generally accepted mechanism for the formation of blebs is the prolonged retention of high swelling pressure in the alveoli. PSP is more common in young, tall, and thin males due to high apical pleural negative pressure (1-4). SSP is caused by chronic obstructive pulmonary diseases such as asthma and cystic fibrosis (1, 5).

PSP is a rare disease in children. Its incidence in childhood is 3.4 in 100,000 cases. Except for the neonatal period, it is mostly observed in tall, thin adolescents at the ages of 13–17 (1, 4, 6). The male to female ratio is between 2:1 and 9:1 (5, 7). During physical examinations in patients who generally present with sudden onset of chest pain and shortness of breath, the hemithorax, where the pneumothorax is detected, is seen to be larger and less involved in breathing. Breathing sounds cannot be heard or detected at the corresponding side by listening alone. The diagnosis is made by visualizing the pleural line on the chest radiograph. Lateral chest radiography and computed tomography (CT) are used in the diagnosis of suspicious cases of PSP in children.

Despite the existence of appropriate guidelines for adults in the follow-up and treatment of PSP, a standard follow-up and treatment protocol has not been developed for pediatric patients (8-13). Observation, oxygen therapy, needle aspiration, tube thoracostomy, sclerosing agent administration, thoracotomy, or video-assisted thoracoscopic surgery (VATS) methods are used in the treatment of PSP in adults. The treatment modality is decided by the severity of symptoms, percentage of pneumothorax detected radiologically, recurrence, duration of drainage, and presence of bullae.

In the present study, we aimed to share our experience by evaluating the age, sex, etiology, diagnostic methods, smoking habits, rates of pneumothorax, early treatment modalities used, and treatment results of the patients that we followed up and treated for PSP.

MATERIALS and METHODS

Records of 71 patients who were followed up and treated with the diagnosis of PSP in Health Sciences University Umraniye Training and Research Hospital and Sakarya University Training and Research Hospital Pediatric Surgery Clinics from January 2010 through October 2020 were retrospectively examined upon the affirmative decision of the Clinical Research Ethics Committee of Umraniye Training and Research Hospital (B.10.1.TKH.4.34.H.GP.0.01/264, dated: 25.06.2020). Patients included in the study were evaluated in terms of age, sex, complaints at presentation, etiology, diagnostic methods, smoking habits, pneumothorax percentage, applied treatment methods, and treatment results.

Pneumothorax diagnosis was confirmed from the medical history, physical examination, and radiological examination

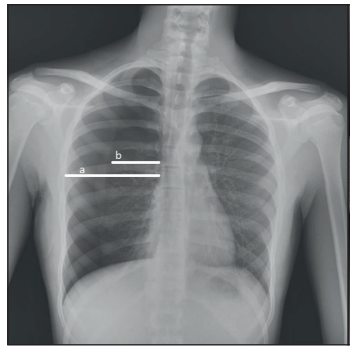

Figure 1: Post-anterior Image of Pneumothorax. **a:** Hemithorax diameter, **b:** Collapsed lung diameter.

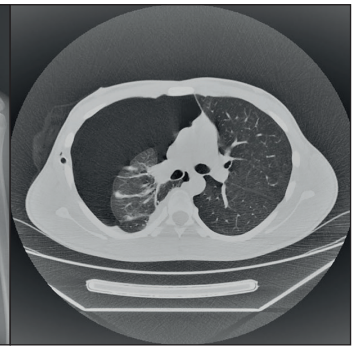

Figure 2: Appearance of Pneumothorax in CT.

findings. Posteroanterior (PA) chest radiography was performed in all patients (Figure 1). For etiology investigation, lung tomography was performed in cases of suspected PSP and those with recurrent PSP at the time of diagnosis or after the lung was fully expanded following tube thoracostomy drainage (Figure 2). Pneumothorax percentages of patients were calculated using the below formula as suggested by Light et al.(14):

Patients with a pneumothorax percentage < 20% at the time of diagnosis, nasal oxygen of 2–3 l/minute was given and were followed up.

All patients with a pneumothorax percentage > 20% and were symptomatic or with unstable vital signs were treated with tube thoracostomy and underwater seal drainage. Chest tube placement was performed in the operating room with sedation (midazolam at a dose of 0.15 mg/kg) as a standard in the 4th intercostal space from the anterior axillary line. A 16–20 F chest tube was used in such patients. Follow-up was performed clinically and based on PA chest radiography findings and the presence of air leakage through the drain. The drain was clamped for 6 hours in patients with expanded lungs as observed on PA chest radiography and with no air leak through the chest tube. Chest tube was removed upon patient's clinical complaint and if there was no air leak when the drain was opened. Failure to detect pneumothorax in control radiographic images taken 24 hours after chest tube removal was accepted as a successful treatment. Surgical treatment was performed in patients whose lungs were not expanded after chest tube was placed, in whom the air leak lasted longer than 7 days, or who had bullae on thorax CT images, and who developed recurrence.

NCSS (Number Cruncher Statistical System) 2007 (Kaysville, Utah, USA) software program was used for statistical analysis. Descriptive statistical methods (mean, standard deviation, median, frequency, percentage, minimum, maximum) were used while evaluating the study data. The conformity of the quantitative data to the normal distribution was tested with the Shapiro-Wilk test and graphical examinations. The Mann-Whitney U test was used for comparisons between two groups of quantitative variables that did not show normal distribution. Pearson chi-square test, Fisher's exact test, Fisher-Freeman-Halton exact test were used to compare qualitative data. Statistical significance was accepted as $p<0.05$.

RESULTS

Among the 71 patients included in the study, 65 (91.5%) were male and six (8.5%) were female. The average age was determined to be 16.23±0.81 years (13–18 years). Right pneumothorax was found in 33 cases (46.5%), left pneumothorax in 34 cases (47.9%), and bilateral pneumothorax in 4 cases (5.6%). The clinical characteristics of the patients are demonstrated in Table I.

Table I: Clinical Features of Primary Spontaneous Pneumothorax Cases.

	n (%)
Age range (years)*	16.23±0.81 16 (13-18)
Sex	
Female	6 (8.5)
Male	65 (91.5)
PSP location	
Left	34 (47.9)
Right	33 (46.5)
Bilateral	4 (5.6)
Bullae observed in CT (n=55)	
No	41 (74.5)
Yes	14 (25.5)
PSP percentage*	51.68±22.70 60 (10-90)
Smoking habit	
No	48 (67.6)
Yes	23 (32.4)
Treatment	
O_2	6 (8.5)
Tube thoracostomy	65 (91.5)
Drainage time (days)*	6.03±3.26 5 (318)
Length of hospital stay (days)*	7.70±3.77 7 (1-19)
Relapse	
No	63 (88.8)
Yes	8 (11.2)

***** *Mean±SD, Median (Min-Max)*

The most common symptoms observed were chest pain (n:71, 100%) and shortness of breath (n:30, 42.2%). Chest radiography was performed in all patients as a part of the first examination. In the first years, lung tomography was performed in 10 (14.1%) patients at the time of first presentation, and 45 (63.4%) patients after treatment with tube thoracostomy. However, bullae were found in the lungs of 14 (25.5%) out of 55 patients who underwent CT imaging. We noted unilateral bullae in 7 (50%) patients and bilateral bullae in 7 (50%) patients. Bilateral pneumothorax developed in 4 of the cases with bilateral bullae. Because of this low rate, CT imaging was not routinely performed during diagnosis in recently. Hence, CT imaging was not performed in 16 (22.5%) patients who presented recently.

Upon examination of the smoking habits of the patients, it was found that 23 (32.4%) patients were smokers. Whereas none of the female patients were found to be smokers, 23 (35.4%) out of the 65 male patients were smokers. The results of the comparison of the clinical features of pneumothorax between smoker and nonsmoker patients are given in Table II.

The pneumothorax percentage was <20% in 14 (19.7% of the studied cases) patients and >20% (30%–85%) in 57 (80.3%) patients. In 14 patients (19.7%) with a pneumothorax percentage

Table II: Clinical features according to smoking habit.

	Smoking habit		p
	No	Yes	
Treatment			[a]0.167
Tube thoracostomy	42 (87.5)	23 (100)	
O_2	6 (12.5)	0 (0)	
PSP percentage*	49.04±23.36 57.5 (10-90)	56.96±20.82 60 (10-85)	[b]0.198
Drainage time (days)*	5.86±3.22 5 (3-18)	6.35±3.37 5 (3-17)	[b]0.436
Length of hospital stay (days)*	7.23±3.48 6 (1-19)	8.7±4.22 7 (4-18)	[b]0.222
Bullae observed in CT (n=55)			[a]0.527
No	23 (71.8)	18 (78.3)	
Yes	9 (28.2)	5 (21.7)	
Relaps			[a]0.458
No	44 (89.8)	19 (86.4)	
Yes	5 (10.2)	3 (13.6)	

[a]Fisher's Exact Test, [b]Mann Whitney U Test, Mean±SD, Median (Min-Max)*

< 20%, administration of 2–3 l/min nasal oxygen therapy and observation were employed as the first line of treatment. While oxygen therapy and observation were sufficient in 6 of these patients (42.8%), tube thoracostomy was performed in addition to oxygen therapy in the remaining 8 (57.2%) patients due to increased respiratory distress observed during the follow-up. The average duration of stay of patients who received only oxygen therapy was 2 days. None of these patients were found to be smokers. Among the patients who needed tube thoracostomy, 3 were found to be smokers, whereas the other 5 were not.

Tube thoracostomy with sedation was performed in 8 patients with pneumothorax percentage <20% and increased respiratory distress and in 57 patients with pneumothorax percentage > 20%. Among the 65 (91.5%) patients, who underwent tube thoracostomy, no major complications were observed except subcutaneous emphysema in 2 and tube revision in 3 patients. Tube thoracostomy was discontinued in 51 patients whose air leak terminated and whose lungs were seen to expand. The mean drainage time of the patients treated with tube thoracostomy was 6.03±3.26 days (3–18 days), and the mean duration of hospital stay was 7.70±3.77 days (1–19 days). In 14 patients whose lungs were not fully expanded, the air leak continued for more than 7 days.

The success rate with nasal oxygen therapy was found to be 42.8% in patients with spontaneous pneumothorax and 78.5% in patients who underwent tube thoracostomy. The patients were followed up for an average of 22.83±17.55 months (1–74 months). Recurrence occurred in 8 (11.2%) patients during the follow-up period. The pneumothorax percentage of 3 patients who developed recurrence was < 20%. Observation was sufficient in these patients. Bullae were detected in the lung tomography images of 5 patients with recurrence and pneumothorax percentage > 20%. Among the patients who developed recurrence, 3 were smokers.

Surgical treatment was performed in 5 patients who developed recurrence after tube thoracostomy and whose lung tomography images revealed the presence of bullae, and in 14 patients with an air leak persisting for > 7 days in whom lung expansion could not be achieved during the treatment. VATS was preferred in choosing the surgical procedure.

DISCUSSION

PSP occurs as a result of the rupture of subpleural bullae and blebs located in the apex of the lung due to the high apical pleural negative pressure in young, tall, and thin male individuals without an underlying lung disease (1,2). Increased height and male sex are risk factors for spontaneous pneumothorax and smoking and atmospheric pressure changes are included in its etiology. In our study, consistent with the literature, pneumothorax was found to be more common in young men.

Although in most studies conducted in adults, subpleural blebs or rupture of the bullae at the apex of the lung was responsible for the formation of spontaneous pneumothorax, pneumothorax may also occur in children without any underlying lung disease. Subpleural blisters and blebs were found in 76%–100% of patients who underwent VATS in studies conducted in adults (2,4,15,16). In his study, Lopez et al. (4) detected bullae in 60% of the patients during CT scan and 98% of the patients during surgery. Therefore, the preferred approach for treatment in adults who have bullae is VATS.

However, there is not enough data on the occurrence of pneumothorax in the pediatric age group in the available literature. In most of the cases observed in our study, no obvious etiological factor was found. Only 14 (25.4%) of the patients had bullae. Thus, we suggest that follow-up and support should be the first treatment option in children, and if this is not sufficient, tube drainage is the appropriate method.

It has been suggested that smoking habit in adults has an important role in the development of pneumothorax. In the relevant studies, the risk of developing pneumothorax in men who smoke is 12%, whereas it is 0.1% in men who do not smoke. The risk increases 20 times in men who smoke (17). Although the smoking rate is higher in adults, Chiu et al. (5) found that 22% of adolescents smoked as well.

We found that 32.4% of our patients had a smoking habit. In our study, when smoking and non-smoking patients were compared, there was no statistically significant difference between smoker and non-smoker patient in terms of treatments, PSP percentages, tube durations, length of hospital stay, presence of bullae and recurrence rates of the cases (p>0.05). There is an increase in smoking among adolescents in our

country. We believe that the availability of data on adolescents who smoke is limited due to the fact that adolescents hide their smoking habits from their families and reply "I don't smoke" in spite of smoking. Thus, the actual smoking rate could be higher in our study.

The diagnosis of pneumothorax is made by the visualization of the pleural line in the chest radiography images taken after physical examination of the patients presenting with sudden chest pain and/or shortness of breath. In cases with insufficient PA chest radiographic findings, lateral chest radiography and thoracic CT findings can be used. All patients in this study reported chest pain, and 30 patients had shortness of breath accompanying chest pain. PA chest radiography imaging was performed in all our patients.

CT imaging is widely used in the diagnosis and etiology research of adult pneumothorax patients, and some authors advocate routine CT scanning in these patients. The reason for this is demonstrated to be the high probability of detecting an underlying pathology (bullae) (4,15).

Many authors argue that CT scanning in children should be used sparingly due to the inadequacy of the technique in the detection of bullae and blebs in pneumothorax and the high radiation dose used (3,4,5,9,11,18). Moreover, the rate detection of bullae or blebs was found to be quite low in our cases.

In our study, only 14 (25.4%) of the 55 patients who underwent thoracic CT scans had bullae. No pathology was found in the CT results of other patients. Due to the low rate of pathology detection and the high radiation dose used in CT, we recommend that CT scanning should not be performed at the first admission to hospital. However, it may be performed to investigate the underlying pathology in suspected cases at the time of diagnosis, cases with recurrence or cases where the lung is not expanded despite performing tube thoracostomy.

Despite the existence of standardized and important guidelines for adults in the follow-up and treatment of PSP, a similar standard follow-up and treatment protocol has not been developed for pediatric patients. The management of pneumothorax in children is based on various retrospective studies (11, 19). Observation, oxygen therapy, aspiration, tube thoracostomy, sclerosing agent administration, thoracoscopy, and thoracotomy are the modalities employed in the treatment of PSP in adults.

The American College of Chest Physicians (ACCP) recommends observation for small PSP in adults, chest tube application for large PSP, and surgical treatment for air leaks lasting longer than 4 days (12). The British Thoracic Society (BTS) recommends observation only for PSP without significant shortness of breath, regardless of its size. It also recommends that patients with significant dyspnea should be treated with needle aspiration or chest tube, and surgical treatment should be performed if there is an air leak that persists for 3–5 days (13).

There are different opinions about the treatment modalities and when to perform surgical treatment in children. Lee et al. (2) recommended conservative treatment in patients with a pneumothorax percentage of >15% and reported 80% success. Soccorso et al. (3) recommended tube thoracostomy as the initial treatment for children with PSP as 53% of patients with initial needle aspiration required tube thoracostomy. As a result of a survey conducted among North American Pediatric Surgeons, various treatment preferences emerged. Among the surgeons participating in the survey, 57% preferred chest tube drainage in the first instance, 29% followed with oxygen therapy alone, 3% preferred needle aspiration, and 4% preferred VATS. In this survey, there was a wide variability in the observation time when VATS decision is made. Among the participants, 40% of surgeons stated that they waited for 3 days, and 21% stated that they waited for 5 days (20).

Zganjer et al. (16) developed an algorithm for the treatment of spontaneous pneumothorax in children in their study of 16 patients in 2010. In children with chest pain, shortness of breath, and cough, they first performed chest radiography and subsequently a CT scan. Intercostal tube drainage was applied to all patients with pneumothorax, and observation was done. In the follow-up, patients with ongoing air leakage for 3–7 days underwent VATS and mechanical pleurodesis with apical bulla excision and wedge resection (16).

In our study, oxygen therapy and observation were performed in 14 patients with a pneumothorax percentage of 10–20%. While only oxygen therapy was sufficient for 6 of these patients, the other 8 patients continued to be treated with tube thoracostomy because the percentage of pneumothorax and respiratory distress increased. In total, tube thoracostomy was performed in 65 patients. The average treatment time with tube was 6.03 days (3–18 days), and average duration of hospital stay in patients treated with tube thoracostomy was found to be 7.70 days (1–19 days). We did not prefer to perform needle aspiration, which was included in adult treatment modalities, in the pediatric age group.

Although the number of patients we followed up and treated with oxygen therapy was limited, it was observed that oxygen therapy was effective in patients with a low percentage of pneumothorax and without clinical complaints. While administering oxygen therapy, tube thoracostomy was considered as an appropriate approach in cases with increased clinical complaints and pneumothorax percentage. When compared with the results of the available literature, in our study, it was found that the success rate of treatment with tube thoracostomy was higher and the duration of treatment with tube and the hospitalization period of the patients were longer.

In some studies, conducted in adults, although PSP was successfully treated with conservative treatment in the

beginning, it was reported that surgical treatment was required due to the recurrence rate of 40%–60% (2,4,5,11,19). However, recurrence after treatment (n:8) was found in 11.2% of our patients. Among the recurrent cases, the pneumothorax percentage of 3 patients was <20% while the rate of pneumothorax in 5 patients was >20%. It was observed that only 3 of the relapsed patients were smokers. There was no significant relationship among recurrence, pneumothorax percentage and smoking. While only observation was sufficient in 3 of our recurrent patients, bullae were detected in the axial CT images of the remaining 5 patients. VATS was applied to these patients.

In the treatment of pneumothorax, surgical treatment was performed in cases with air leak lasting longer than 5–7 days, non-expansion of lungs, and recurrence after tube thoracostomy treatment. Surgical treatment was applied to 5 patients who developed recurrence after tube thoracostomy and whose lung tomography images were found to have bullae and 14 patients with an air leak for more than 7 days and in whom lung expansion could not be achieved during the treatment. VATS was preferred as the surgical procedure.

CONCLUSIONS

As a result of our observations, we suggest that smoking may be an important risk factor for PSP in children as it is in adults. From our results, the underlying pathology and recurrence rate is low in children with pneumothorax, unlike in adults. Therefore, obtaining medical history and conducting physical examination and PA radiography should be sufficient for the diagnosis of PSP. CT imaging should be performed only in cases with recurrence and prolonged air leakage. As a treatment option, follow-up and supportive treatment, tube thoracostomy, and surgical treatment (VATS) can be applied in cases with prolonged air leak and recurrence. However, we believe that multi-center studies with more series and experience are needed to develop treatment algorithms in children.

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