

## ORIGINAL ARTICLE

# Spontaneous air leak in a Pediatric Emergency Department: An 11-year experience

## Síndrome de fuga de ar espontâneo num Departamento de Emergência Pediátrica: Experiência de 11 anos

Telma Luís<sup>1</sup> , Andreia Lomba<sup>1</sup> , Maria Miguel Almiro<sup>1</sup> , Sílvia Almeida<sup>1</sup> , Carla Valente<sup>2</sup> 

### ABSTRACT

**Introduction:** Due to lack of data, the management of air leaks in children and adolescents is based on protocols for adults. In this study, the authors review and report their institutional experience in the area.

**Methods:** Retrospective and descriptive study of spontaneous air leak cases diagnosed in a Portuguese Pediatric Emergency Department (PED) between January 2007 and December 2018.

**Results:** Twenty-one episodes of spontaneous air leak were diagnosed in 16 patients (87.5% male), with a mean ( $\pm$  standard deviation) age of 14.3 ( $\pm$  5.1) years. Eighteen cases of spontaneous pneumothorax were reported, fifteen of which primary (eleven first-time episodes and four recurrences) and three secondary (two first-time episodes and one recurrence) to asthma. Three cases of pneumomediastinum with subcutaneous emphysema were identified, two of which related to infection. Smoking habits and/or recent physical activity were major triggers. Pleuritic chest pain was the most frequent symptom on admission. Conservative treatment was the only approach used in six cases. Most cases required thoracic drainage, five of which required surgical intervention.

**Discussion:** This study shows similar demographic features, triggers, and clinical presentation to those reported in the literature for these cases. The management of the condition was based on recommendations established for adults.

**Conclusion:** Although spontaneous air leak is an uncommon condition, it is a reality in PED. Prospective studies in pediatric age are required to develop adequate recommendations for children and adolescents.

**Keywords:** air leak; Pediatrics; pneumomediastinum; pneumothorax

### RESUMO

**Introdução:** Devido à escassez de dados em idade pediátrica, a abordagem da síndrome de fuga de ar em crianças e adolescentes baseia-se em protocolos estabelecidos para adultos. Neste estudo, os autores descrevem a sua experiência institucional na área.

**Métodos:** Estudo retrospectivo descritivo de episódios espontâneos de síndrome de fuga de ar diagnosticados num Serviço de Urgência Pediátrica (SUP) em Portugal entre 1 de janeiro de 2007 e 31 de dezembro de 2018.

**Resultados:** Foram diagnosticados vinte e um episódios de síndrome de fuga de ar em dezasseis crianças, 87.5% das quais do género masculino, com uma idade média ( $\pm$  desvio padrão) de 14,3 ( $\pm$  5,1) anos. Foram reportados 18 casos de pneumotórax espontâneo, quinze dos quais primários (onze primeiros episódios e quatro recorrências) e três secundários (dois primeiros episódios e uma recorrência) a asma. Foram contabilizados três episódios de pneumomediastino com enfisema subcutâneo, dois dos quais secundários a infeção. A exposição ativa

1. Department of Pediatrics, Centro Hospitalar do Baixo Vouga. 3810-193 Aveiro, Portugal. telmaluismarques@gmail.com; andreiaflcorreia@gmail.com; mariamiguelaa@gmail.com; smsalmeida@sapo.pt  
2. Department of Pulmonology, Centro Hospitalar do Baixo Vouga. 3810-193 Aveiro, Portugal. carlavalente77@hotmail.com

a fumo de tabaco e/ou esforço físico recente foram fatores etiopatogênicos preponderantes na maioria dos casos. Dor torácica pleurítica foi o sintoma mais frequente na apresentação clínica. A terapêutica conservadora foi a única abordagem utilizada em seis casos, com os restantes a necessitar de drenagem transtorácica. Cinco episódios exigiram intervenção cirúrgica.

**Discussão:** Este estudo evidencia fatores demográficos e etiopatogênicos e apresentação clínica semelhantes aos descritos na literatura para estes casos. Relativamente à abordagem, foram seguidas as recomendações existentes para a idade adulta.

**Conclusão:** Apesar de pouco frequente, a síndrome de fuga de ar é uma realidade no SUP. São necessários estudos prospetivos especificamente em idade pediátrica para permitir o desenvolvimento de recomendações adequadas às particularidades desta faixa etária.

**Palavras-chave:** Pediatria; pneumomediastino; pneumotórax; síndrome de fuga de ar

## INTRODUCTION

Air leak is a clinical phenomenon characterized by leakage of air from a body cavity into spaces where, under normal circumstances, it should not be present.<sup>(1,2)</sup> This may generate pressure on contiguous structures and have clinical consequences.<sup>(3-5)</sup> In cases of pulmonary air leak, cardiac or pulmonary compression may occur, resulting in respiratory distress and hemodynamic compromise.<sup>(6-8)</sup>

Air leak can be classified as traumatic or spontaneous. Traumatic air leak can be caused by blunt or penetrating trauma or injury from medical procedures or be a consequence of mechanical ventilation. Spontaneous air leak occurs in absence of any identified trauma.<sup>(9,10)</sup> Spontaneous pulmonary air leak can be categorized as primary (if occurring in patients without underlying lung disease) or secondary (if occurring in patients with underlying lung disease that predisposes to the occurrence of air leak).<sup>(9-11)</sup>

Excluding the neonatal period, air leak is more frequent in children with underlying chronic pulmonary disease under ventilatory support and in adolescent males with tall, thin body habitus and smoking habits.<sup>(10,12-19)</sup> The clinical presentation may be atypical in younger children and include irritability, vomiting, or cough, usually together with tachypnea and respiratory distress, making the diagnosis challenging.

Although air leak is uncommon in pediatric age, its recurrence rate is higher than in adults (50-60% vs 30% in the case of pneumothorax).<sup>(12,20,21)</sup> The lack of data in pediatric age, together with absence of specific management guidelines, lead to controversial approaches for the management of the condition in this population, currently mainly based on protocols established for adults.

The aim of the study was to review the authors' institutional experience in the management of air leak in pediatric age over 11 years, including the assessment of risk factors, clinical presentation, initial and additional studies, therapeutic approach, and follow-up.

## MATERIAL AND METHODS

This was a retrospective, descriptive, observational study of air leak cases diagnosed at the Pediatric Emergency Department (PED) of a Portuguese hospital between January 2007 and December 2018. Patients admitted to PED with a diagnosis of air leak according to the International Classification of Diseases-9 (pneumothorax, pneumomediastinum, subcutaneous emphysema, pneumopericardium, pneumoperitoneum) were included. Cases of air leak secondary to trauma or developing in the neonatal period were excluded.

Data were retrieved from patients' clinical records and included demographics, risk factors, clinical presentation, laboratory and imaging studies, classification, approach, recurrence, and follow-up. Statistical analysis was performed using *Microsoft Excel® 2010*.

## RESULTS

Twenty-one cases of air leak were identified in sixteen patients, 87.5% (14/16) of whom male and 87.5% (14/16) adolescents. The mean ( $\pm$  standard deviation [SD]) age of the cohort was 14.3 ( $\pm$  5.1) years and median age was 16 years. The youngest patient was 17 months old.

Smoking habits and/or recent physical activity were major triggers in 57.9% of patients. Available data did not allow to establish an association between air leak and passive smoking.

Regarding clinical presentation, sudden onset of symptoms was reported in 61.9% (13/21) of patients. Pleuritic chest pain was the most frequent symptom on admission (90.5%), followed by dyspnea (47.6%), cough (19.1%), and fever (14.3%). Diminished breath sounds were the most common physical finding (87.7%), followed by respiratory distress (9.5%), hypoxemia (9.5%), and palpable crepitation (9.5%). No patient presented hemodynamic instability on admission. All patients performed a posteroanterior chest X-ray that confirmed the diagnosis. Eighteen spontaneous pneumothorax

events occurred, eleven of which on the left side (61.1%). Eleven patients were diagnosed with primary spontaneous pneumothorax (PSP), four of which recurred, making a total of fifteen PSP episodes. Two patients were diagnosed with secondary spontaneous pneumothorax (SSP) related to asthma, one of which recurred, making a total of three SSP episodes. Neither of these two patients was under regular treatment for asthma and both were exposed to passive smoking.

Three cases of pneumomediastinum with subcutaneous emphysema were identified, two of which secondary to infectious diseases (acute bronchiolitis and pneumonia) and occurring in children under the age of five years. No recurrences were observed in this subgroup. No cases of pneumopericardium or pneumoperitoneum were reported.

All patients were hospitalized with a mean ( $\pm$  SD) hospital stay of 5.9  $\pm$  3.3 days (maximum of 14 days).

Conservative treatment was the only approach used in six cases (28.6%), three of which concerned pneumomediastinum with subcutaneous emphysema. In three other cases, while this treatment option was chosen as initial management strategy, subsequent thoracic drainage was also required (14.3%).

Thoracic drainage (using 14 Fr chest tube and trocar technique) was selected as first approach in twelve cases (57.1%) of moderate-to-large (volume >20% at Light index) and/or recurrent pneumothorax. Chest tube was removed after a minimum of 48 hours when there was no evidence of air leak on the drainage system and complete radiographic pulmonary re-expansion occurred. Needle aspiration was not used in any episode. Two relapses (one of PSP and one of SSP) occurred during hospitalization after initial thoracic drainage.

In PSP, thoracic drainage was removed within two days and a large pneumothorax ensued four days later, unresponsive to new drainage and requiring video-assisted thoracoscopic surgery (VATS) with mechanical pleurodesis. In SSP, thoracic drainage was removed within three days and relapse occurred two days later, with complete recovery on the third day after new thoracic drainage. Five cases required surgical intervention, three of which (two recurrent PSP and one relapsing PSP) with the need of transfer to a tertiary hospital for urgent VATS with pleurodesis. The remaining two cases (one recurrent PSP and one recurrent SSP) required elective procedures (pleurodesis).

Four patients with PSP presented ipsilateral recurrence and one patient with SSP presented contralateral recurrence. The maximum time between recurrences was 15 months.

Ten patients (76.9%) with pneumothorax were followed at the Department of Pulmonology. Most underwent laboratory and/or imaging studies to exclude lung conditions that could predispose to higher risk of air leak. Eight patients (80%) performed chest computed tomography (CT) scan, nine (90%) were tested for  $\alpha$ 1-antitripsin deficiency, and three (30%) underwent spirometry. While no newly-diagnosed lung disease was identified, subpleural bulla was identified in four cases. Two performed CT scan during hospitalization due to urgent need of surgical intervention. Bulla resection was only performed in these patients. Patients submitted to surgical intervention were also followed at the Department of Thoracic Surgery of the tertiary hospital where interventions were performed. **Table 1** shows the detailed description of each case.

**Table 1** - Detailed description of air leak cases managed at the Pediatric Emergency Department

Age (years)	Gender (F/M)	Associated diseases	Smoking habits (Y/N)	First episode				Second episode				Follow-up by Pneumology (Y/N)
				Air leak type	Clinical presentation	Initial treatment	Subsequent treatment	Air leak type	Clinical presentation	Initial treatment	Subsequent treatment	
15	M	N	N	PSP Volume >20%	Pleuritic chest pain, cough	Thoracic drainage	NP	PSP Volume >20%	Pleuritic chest pain	Thoracic drainage	Surgery	Y
15	M	N	N	PSP Volume <20%	Pleuritic chest pain	Conservative	Thoracic drainage	PSP Volume <20%	Pleuritic chest pain	Conservative	NP	Y
16	M	Asthma	Y	SSP Volume <20%	Pleuritic chest pain	Conservative	Thoracic drainage	SSP Volume >20%	Pleuritic chest pain	Thoracic drainage	Elective Surgery	Y
16	M	N	N	PSP Volume <20%	Pleuritic chest pain	Conservative	Thoracic drainage	PSP Volume >20%	Pleuritic chest pain, dyspnea	Thoracic drainage	Elective Surgery	Y
17	M	N	N	PSP Volume >20%	Pleuritic chest pain, dyspnea	Thoracic drainage	NP	PSP Volume >20%	Pleuritic chest pain	Thoracic drainage	Surgery	N

15	M	N	N	PSP Volume >20%	Pleuritic chest pain	Thoracic drainage	NP						N
16	M	N	Y	PSP Volume <20%	Pleuritic chest pain, dyspnea	Conservative	NP						N
16	F	N	N	PSP with relapse Volume >20%	Pleuritic chest pain, dyspnea, fever	Thoracic drainage	Surgery						Y
16	M	N	N	PSP Volume >20%	Pleuritic chest pain, dyspnea, cough, fever	Thoracic drainage	NP						Y
17	M	N	Y	PSP Volume >20%	Pleuritic chest pain	Thoracic drainage	NP						Y
17	M	Asthma	N	PSP with relapse Volume >20%	Pleuritic chest pain, dyspnea	Thoracic drainage	Thoracic drainage						Y
17	M	N	Y	PSP Volume >20%	Pleuritic chest pain, dyspnea	Thoracic drainage	NP						Y
17	M	N	Y	PSP Volume >20%	Pleuritic chest pain, dyspnea	Conservative	NP						Y
1	M	Acute bronchiolitis	N	PMD with subcutaneous emphysema Secondary to acute bronchiolitis	Dyspnea, cough	Conservative	NP						N
3	F	Pneumonia	N	PMD with subcutaneous emphysema Secondary to pneumonia	Fever, cough	Conservative	NP						N
16	M	N	N	PMD with subcutaneous emphysema	Pleuritic chest pain, dyspnea, neck pain	Conservative	NP						N

**Legend:** F – Female; M - Male; N - No; NP – Not performed; PMD – Pneumomediastinum; PSP – Primary spontaneous pneumothorax; SSP – Secondary spontaneous pneumothorax; Y – Yes

## DISCUSSION

This study identified similar demographic features, triggers, and clinical presentation in adolescents with spontaneous air leak to those reported in the literature, including male predominance, smoking habits and recent physical activity as major triggers, and typical symptoms at presentation.<sup>(22-24)</sup>

Most cases concerned primary spontaneous air leak, which is the most common type in pediatric age. However, pneumothorax recurrence rates were lower (27.8%) than reported in the literature.<sup>(20)</sup> One of these episodes was a late recurrence (>12 months), which is unusual. Pneumomediastinum recurrence is typically observed in less than 5% of cases, which was confirmed in this study.<sup>(16-18)</sup>

Posteroanterior chest X-ray is the gold standard for diagnostic

confirmation and was performed in all cases. The use of chest CT scan in first-time air leak episodes is controversial and should be reserved for patients with unclear diagnosis or recommendations for surgical intervention.<sup>(10)</sup>

In this study, the management of air leaks was based on recommendations established for adults, which foresee conservative treatment for clinically stable patients with non-complicated pneumomediastinum with subcutaneous emphysema, first-time small PSP episodes, or small SSP related to asthma; and invasive treatment for PSP recurrences, first-time large PSP episodes, large SSP related to asthma, or any SSP due to another lung disease, regardless of size.<sup>(25-27)</sup>

Thoracic drainage was the most used invasive technique, achieving favorable outcomes. Only two cases of relapse were identified

in this group of patients. Although adult guidelines recommend needle aspiration as first-line approach for clinically stable patients with large PSP and for some cases of SSP, data in pediatric age is limited and insufficient to state formal recommendations.<sup>(27)</sup> Some studies in Pediatrics suggest advantages of this technique, such as shorter hospitalization time and reduced pain, but its effectiveness is controversial for many authors.<sup>(10,28,29,33)</sup> In addition, the need for repeating the procedure in cases of relapse hinders its use in pediatric age. Further studies are required to evaluate the efficacy of this therapeutic option in this age group and develop appropriate recommendations. Although these procedures are routinely performed in adults, sedation or even general anesthesia may be required in pediatric patients (depending on their age and tolerability).<sup>(33)</sup>

Few cases in this study were managed only with conservative treatment, what can be explained by the high incidence of large pneumothorax cases. Surgical intervention is recommended in adults with PSP that does not remit after five to seven days of thoracic drainage, in cases of recurrent spontaneous pneumothorax, and in first SSP episodes (depending on the underlying lung disease). In relapse cases, the same recommendations should be followed.<sup>(27)</sup> These criteria were met, except in a single case of recurrent PSP that remitted with conservative measures. In some centers, thoracoscopic surgery is being proposed as an alternative to chest drainage in first PSP episodes in adults, with lower recurrence rates and morbidity and shorter length of hospital stay.<sup>(30-33)</sup> Some studies indicate that more than 10% of patients with PSP require surgical intervention for the first episode, and up to 54% of initially successfully treated patients require surgical treatment for PSP recurrence within four years.<sup>(30,32)</sup> In pediatric age, the fact that thoracic drainage often requires general anesthesia (particularly in young children) should also be considered. Additionally, thoracoscopic intervention reduces children's exposure to repeated pleural cavity drainage or puncture.<sup>(34)</sup> However, randomized studies in pediatric age still indicate that conservative measures, needle aspiration, or chest tube drainage should be recommended as initial strategy for PSP, since several patients will never require surgical intervention and the first-line approach for the condition in this population should be little invasive. Nevertheless, thoracoscopic surgery is effective and represents the best option for refractory or recurrent cases.<sup>(32-33)</sup>

The literature recommends that patients suffering from a pneumothorax should be followed by a respiratory physician to exclude underlying lung disease and due to the high risk of recurrence with later need for surgical intervention. This recommendation was fulfilled in 76.9% of cases in the present study and is becoming standard in this study's institution. As pneumothorax recurrence risk only significantly decreases after one year, this is the minimal time of required follow-up.<sup>27</sup> Performing a chest CT and excluding  $\alpha$ 1-antitrypsin deficiency may be considered during the follow-up of patients with a history of spontaneous pneumothorax. The presence of bullae in CT scan has been suggested as an independent risk factor

for PSP recurrence. However, some studies consider that bullae may not predict recurrence, and there is still lack of evidence about using CT scan findings for risk stratification and patient selection for prophylactic surgery.<sup>(31-33)</sup> In the present study, patients with pleural bullae without reasons to undergo surgery only maintained follow-up visits. After an episode of pneumothorax, patients should permanently avoid diving and refrain from air traveling until at least one week after full resolution.<sup>(27)</sup>

Non-complicated pneumomediastinum with subcutaneous emphysema is a benign condition with good prognosis and reduced risk of recurrence. Therefore, follow-up is not recommended.

The main limitations of this study were the reduced sample size and its retrospective nature.

## CONCLUSION

While uncommon, air leak is a reality in pediatric age, requiring a multidisciplinary approach involving pulmonologists, pediatricians, and thoracic surgeons. Despite the lack of evidence in this population, the present Pediatric Emergency Department has been progressively using existing adult protocols, with good results: most patients recovered with thoracic drainage alone, and recurrence rates were lower than those reported in the literature. Prospective studies are required to improve intervention strategies and promote suitable recommendations for this patient population.

## AUTHORSHIP

Telma Luís – Designed and Conceived the study; Collected and analyzed data; Interpretation of the results; Writing – original draft  
Andreia Lomba – Collected and analyzed data  
Maria Miguel Almiro – Interpretation of the results; Writing – original draft  
Sílvia Almeida – Interpretation of the results; Writing – original draft  
Carla Valente – Interpretation of the results; Writing – original draft

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#### CORRESPONDENCE TO

Telma Luís  
Department of Pediatrics  
Avenida Artur Ravara  
Centro Hospitalar do Baixo Vouga  
3810-193 Aveiro  
Email: telmaluismarques@gmail.com

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