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Spontaneous Pneumomediastinum in Children: The Experience of a Pediatric Tertiary Center in Antalya

Çocukluk Çağında Spontan Pnömomediastinum: Antalya' daki Üçüncü Basamak bir Merkezin Deneyimi

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This study was presented as a poster presentation at the 4th Congress of Pediatric Chest Diseases 9-11 October 2019, İstanbul, Turkey.

ABSTRACT

Objective:

To evaluate the demographic data, clinical findings, complications and prognosis of children with spontaneous pneumomediasinum.

Material and Methods: This was a retrospective chart review including demographic characteristics, trigger factors, underlying diseases, symptoms, signs, length of hospital stay, complications, treatments, and outcomes of 15 children aged <18 years with a discharge diagnosis of pneumomediastinum, between January 2009 and August 2019, at the University of Health Sciences Antalya Training and Research Hospital.

Results:

The mean age of the 12 boys (80%) and three girls (20%) with spontaneous pneumomediastinum was 11.33 ± 7 years, with a range of 4 months to 17 years. The most common trigger factor was severe cough and/or bronchospasm due to respiratory tract infection. In 8 patients with pneumomediastinum seen radiologically on the chest X-ray, thoracic computed tomography was also taken. Pneumorrhachis was determined on the thoracic computed tomography of 3 (20%) patients. Pneumomediastinum was accompanied by pneumothorax in 7 (46.6%) cases. The mean length of hospital stay was 8.4 ± 4.5 days (3-18 days). All patients except one recovered and were discharged and followed up for 1-72 months. Hospital records showed no recurrence during the follow-up period.

Conclusions:

Spontaneous pneumomediastinum is a rare, self-limiting condition in childhood and the prognosis depends on the underlying disease. The problem of whether or not routine chest computed tomography scanning should be performed in children with spontaneous pneumomediastinum has not yet been resolved, but if chest computed tomography scanning is performed in every patient with spontaneous pneumomediastinum, pneumorrachis might be detected more frequently.

Key Words: Spontaneous pneumomediastinum, Pneumorrhachis, Pneumomediastinum, Pneumothorax

ÖZ

Amaç: Spontan pnömomediastinumlu çocuk olguların demografik verilerini, klinik bulgularını, komplikasyonlarını ve prognozlarını değerlendirmek.

Gereç ve Yöntemler:

Ocak 2009- Ağustos 2019 tarihleri arasında Sağlık Bilimleri Üniversitesi Antalya Eğitim ve

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Araştırma Hastanesinden pnömomediastinum tanısıyla taburcu edilen 15 çocuk olgunun demografik özellikleri, tetikleyici faktörleri, altta yatan hastalıkları, semptomları, bulguları, hastanede kalış süreleri, komplikasyonları, tedavileri ve sonuçları içeren verileri geriye dönük incelendi.

Bulgular:
Spontan pnömomediastinumu olan 12 erkek (% 80) ve üç kızın (% 20) ortalama yaşları 11.33 ± 7 yıldı (4ay-17 yıl). En sık tetikleyici faktör solunum yolu enfeksiyonuna bağlı şiddetli öksürük ve / veya bronkospazmdı. Sekiz olguya akciğer grafisinde pnömomediastinum saptanmasına rağmen toraks bilgisayarlı tomografi de çekilmişti. Üç (% 20) olgunun toraks bilgisayarlı tomografisinde pnömoraşisi mevcuttu. Yedi (% 46.6) olguda pnömomediastinuma pnömotoraks eşlik etmekteydi. Ortalama hastanede kalış süreleri 8.4 ± 4.5 (3-18) gündü. Bir hasta dışında tümü iyileşerek taburcu edildi ve 1-72 ay boyunca takip edildi. Takip süresi boyunca olgularda nüks saptanmadı.

Sonuçlar:
Spontan pnömomediastinum çocukluk çağında nadir görülen, kendi kendini sınırlandıran bir durumdur ve prognozu altta yatan hastalığa bağlıdır. Spontan pnömomediastinumlu çocuk olgulara rutin akciğer tomografisi çekilip çekilmeyeceği henüz net değildir. Tüm spontan pnömomediastinumlu olgulara akciğer tomografisi çekilirse pnömoraşi daha sık tespit edilebilir.

Anahtar Sözcükler: Spontan pnömomediastinum, Pnömoraşi, Pnömomediastinum, Pnömotoraks

INTRODUCTION

Spontaneous pneumomediastinum (SPM) is defined as the presence of interstitial air in the mediastinum, without any obvious trigger factors such as chest trauma, invasive procedure or positive pressure ventilation (1). It is rarely seen in children and mainly occurs in male adolescents (2).

The pathogenesis of SPM is described as secondary to a sudden increase in intrathoracic pressure that causes an increase in intra-alveolar pressure. This causes alveolar ruptures and air leakage from ruptured alveoli, escapes and may reach the hilium and spread into the mediastinum. If leaked air spreads into subcutaneous tissues in the chest wall and neck, and through facial planes, subcutaneous emphysema occurs (3, 4).

Clinically, patients usually present with dyspnea, chest pain, subcutaneous emphysema, tachypnea, cough, or pharyngalgia. In most patients, physical examination and chest X-ray are sufficient for the diagnosis of PM. It is thought to be a benign condition and usually resolves without any complications. The aim of this study was to evaluate the demographic data, clinical findings, complications and prognosis of children with SPM.

MATERIAL and METHODS

This was a retrospective chart review of 15 children aged <18 years with a discharge diagnosis of PM, during the 10-year period of January 2009 to August 2019, at University of Health Sciences Antalya Training and Research Hospital. Approval for the study was granted by the Clinical Research Ethics Committee of University of Health Sciences Antalya Training and Research Hospital (decision no:2020-017, dated: 23.01.2020). This study was conducted in accordance with the Declaration of Helsinki.

The criteria for inclusion of the patients in the study were a clinical history consistent with PM, the presence of interstitial air in the mediastinum on chest X ray and/or thoracic computed tomography (CT), the absence of a clearly defined triggering cause, and age <18 years. Patients were excluded from the study if they were younger than one month old, or if they had evidence of a clear trigger factor for the PM, such as chest trauma, invasive procedure, or positive pressure ventilation. The demographic data, trigger factors, underlying diseases, symptoms, signs, length of hospital stay, complications, treatments, and outcomes were analyzed.

Statistical Analysis

Data obtained in the study were analyzed statistically using SPSS 23.0 software. Descriptive statistics were presented as number (n) and percentage (%), mean ± standard deviation (SD), minimum and maximum values. In the analysis of categorical data, Fisher's Exact Test was used if the expected value was <5 in >20% of cells and if it was smaller, the Pearson Chi-Square Test was used. Normality assumption was evaluated with the Shapiro Wilks Test. The Mann-Whitney U test was used to compare the age and length of hospital stay of the two groups, since the data did not conform to normal distribution. A value of $p < 0.05$ was considered statistically significant.

RESULTS

The mean age of the 12 boys (80%) and three girls (20%) with SPM was 11.33 ± 7 years, with a range of 4 months to 17 years. The presence of PM was confirmed radiologically in all cases. The demographic data, trigger factors, symptoms, signs, complications, treatments, and outcomes are summarized in Table I.

The most common trigger factor was severe cough and/or bronchospasm due to a respiratory tract infection (n=6, 40%) (pneumonia in 4 cases, bronchiolitis in 1, croup in 1) followed by lifting a heavy object (n=1, 6.7%), convulsion (n=1, 6.7%), and sudden movement (n=1, 6.7%). Six patients were idiopathic (n=6, 40%). Three patients (20%) had asthma.

The most frequent symptoms at presentation were chest pain (n=7, 46.6%), dyspnea (n=4, 26.7%), cough (n=3, 20%), back pain (n=2, 13.3%) and sore throat (n=2, 13.3%). Upon physical examination, the most common sign was pneumoderma in 11 (73.3%) cases. Neck and facial swelling was observed in 9 patients and dysphonia in one. A chest X-ray was performed in all patients and thoracic CT was performed in 12 (80%) patients. Chest X-rays showed pneumoderma in 9 cases, vertical lucent sign and continuous diaphragm in 3 cases, and pneumopericardium in 1 case (Figure 1. a, b, c).

Table I. Demographic data, predisposing factors, symptoms, signs, complications and treatments in children with sponta neous pneumomediastinum

Parameter	Value, n (%) or mean ± standard deviation
Male / female gender	12 (80) /3 (20)
Age (years)	11.3 ± 7
Predisposing Event	
Cough/Upper-Lower Respiratory Tract Infections	6 (40)
Lifting heavy object	1 (6.7)
Convulsion	1 (6.7)
Sudden movement	1 (6.7)
None (at rest)	6 (40)
Symptom	
Chest pain	7 (46.6)
Dyspnea	4 (26.6)
Cough	3 (20)
Back pain	2 (13.3)
Pharyngalgia	2 (13.3)
Signs	
Pneumoderma	11 (73.3)
Face or neck swelling	9 (60)
Dysphonia	1 (6.6)
Complications	
Pneumothorax	7 (46.6)
Pneumorrhachis	3 (20)
Pneumoperitoneum	1 (6.6)
Pneumopericardium	1 (6.6)
Treatments	
Oxygen therapy	15 (100)
Analgesics	11 (73.3)
Bronchodilators	9 (60)
Antibiotics	9 (60)
Length of hospital stay (days)	8.4 ± 4.5
Mortality	1 (6.6)

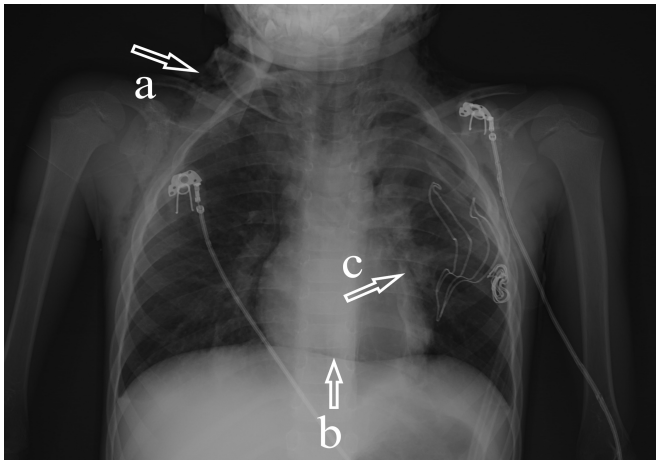

Figure 1. (a) Pneumoderma, (b) Continuous diaphragm and (c) Vertical lucent sign on the left side of the heart on chest x-ray.

PM was observed radiologically on the chest X-ray of 8 patients, and thoracic CT was also taken. Pneumorrhachis was determined on thoracic CT of 3 (20%) patients (Figure 2a). No statistically significant difference was determined between patients with and without pneumorrachis accompanying SPM in terms of age (p = 0.53), gender (p =0.08), accompanying pneumothorax (p = 0.99), subcutaneous emphysema (p =0.51), need for intensive care unit (p =0.2) and duration of hospital stay (p =0.18). PM was accompanied by pneumothorax in 7 (46.6%) cases.

The primary treatments offered were management of the underlying disease and primary analgesics. Oxygen therapy was given to all patients, 11 patients received analgesics and 9 patients received bronchodilators and antibiotics. Chest tube drainage was required by 5 patients because of the associated pneumothorax. The mean length of stay was 8.4 ± 4.5 days (range, 3-18 days) and all patients except one, recovered and were discharged. The remaining patient was 3 years old and presented with a fever, convulsions and dyspnea. Pneumoperitoneum, PM and pneumothorax were detected on the chest X-ray (Figure 2b).

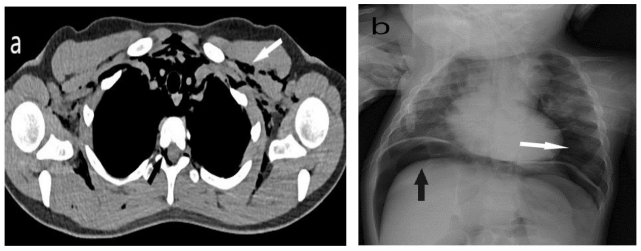

Figure 2. (a) Pneumoderma (white arrow) and pneumorrhachis (black arrow) on thoracic CT. (b) Pneumoperitoneum (black arrow), pneumomediastinum and pneumothorax (white arrow) on chest x-ray.

This patient was hospitalized in the intensive care unit and underwent exploratory laparotomy because of the poor general condition and suspected intra-abdominal perforation. No perforation was detected. Acute respiratory distress syndrome developed due to pneumonia and sepsis and the patient died. All discharged patients were followed up with outpatient visits. The median follow-up time was 7 months (range, 1-72 months) and hospital records showed no recurrence during the follow-up period.

DISCUSSION

SPM is a rare, self-limiting condition in childhood and the prognosis depends on the underlying disease. After transient worsening of symptoms, it usually resolves spontaneously within 3-15 days (5). In the pediatric population younger than 18 years presenting at the emergency department, the incidence is 1/800 to 1/42,000 (2). While SPM is in itself benign, the main concern is the risk of missing associated complications. Interestingly, according to the current study, pneumorrhachis may be more common than previously thought in children with SPM.

Most publications related to SPM include small case series of children and adults, pediatric case series and case reports. Most publications focus on patient presentation and only a few small studies have reported complications associated with SPM. Perna et al. reported a case series of 47 patients with SPM and one new case, where surgical intervention was necessary (6). Bakhos et al. (7) also reported a case series of 49 patients with SPM and only one patient requiring intubation for a severe asthma attack (PM was diagnosed before intubation). However, no mortality was recorded in those studies. Furthermore, in the literature, a case of laryngeal compression with stridor has been reported in a child (8). Other rare complications are pneumothorax, pneumopericardium, pneumoperitoneum, pneumorrhachis, gas embolism and respiratory distress.

Pneumorrhacis is defined as the presence of air in the epidural space. The mechanism of entry of air from the mediastinum to the epidural space is uncertain. Balachandran et al. (9) stated that the air in the retropharyngeal space has a connection with the epidural space through the neural foramina, and it was suggested that this is the probable pathway through which SPM causes epidural pneumatosis. Pneumorrhachis presents with vague clinical findings and is difficult to diagnose on chest radiographs. If abnormal neurological findings are detected, an additional CT is required for further neurological evaluation and the treatment plan. This rare complication was observed in 3 of the 15 cases in the current study. Recently, Yaginuma et al. reviewed the relevant literature regarding children with pneumorrhachis accompanying PM and retrieved 25 articles, reporting 32 cases (10). In literature there is only one study about the incidence of pneumorrhachis accompanying SPM. Kono et al. (11) reported pneumorrhachis in 4 (9.5%) of 42 cases with SPM. The problem of whether or not routine CT scanning should be performed in children with SPM has not yet been resolved, but if CT scanning is performed in every patient with SPM, pneumorrachis might be detected more frequently. In the current study, 20% of the cases were accompanied by pneumorrhachis and none had neurological deficits. This finding can be considered to be due to the high proportion of the study patients undergoing CT scanning.

Although SPM may occur spontaneously in children, a trigger can be found in 70-90% of cases (12). The most frequent triggers in children are asthma, vomiting, situations reproducing the Valsalva maneuver (e.g. coughing, shouting) and intense sport activities (2). In the current study, trigger factors were found in 60% of the cases with SPM and excessive coughing and/or bronchospasm due to respiratory tract infection was the major cause. Bronchopneumonia was the key trigger factor among infectious diseases.

Clinically SPM should come to mind in children with prevalent acute chest pain, dyspnea and subcutaneous emphysema. This combination has been reported in literature to be found in 40% of cases. Chest pain, present in 50-90 % of cases, is often retrosternal, is worsened by postural changes and deep inspiration, and radiates to the arms, shoulders or back. Additionally, some extra-thoracic signs, such as neck swelling or pain, dysphonia, or dysphagia, may also be present (2). In the current study, chest pain, the most frequent symptom at presentation, was observed in 46.6% of cases, similar to the literature. Face or neck swelling was seen in 9 patients and one patient had dysphonia as extra-thoracic signs.

History, physical examination, and direct radiography are usually sufficient for diagnosis and further investigations are rarely needed. Specific findings on frontal and lateral chest radiographs are the spinnaker sail sign, the continuous diaphragm sign, a vertical lucent sign, pneumoderma and finding air in the retrosternal space (13). In the current study population, chest X-rays showed pneumoderma in 9 cases, and the vertical lucent sign and continuous diaphragm sign in 3 cases. Most studies have reported that thoracic CT and/or esophagography are frequently used for routine evaluation of patients with SPM. Thoracic CT is superior to chest X-rays for the detection of subcutaneous air, subdiaphragmatic air, esophageal or tracheal injury, pulmonary interstitial opacities, interstitial fibrosis, or pulmonary interstitial emphysema. However, there is a higher risk of radiation and it is more expensive (14). Evidence of esophageal perforation must be investigated with further investigative tools (e.g. esophagography, thoracic CT) when PM occurs after violent vomiting and is associated with pleural accumulation of fluid or fluid and gases on chest X ray. Bakhos et al.7 reported the need to restrict further imaging tests to SPM patients with clinical presentation concerning esophageal injury, as additional imaging tests are often negative. In two recent studies including high numbers of pediatric cases with SPM, Noorbakhsh et al. (15) and Abbas et al. (16) reported that there was potentially no added benefit from further studies such as thoracic CT or esophagography. Chest computed tomography was applied to 68 (37%) of 183 patients with SPM in the Noorbakhsh et al. study (15) and to 32 (24.8%) of 129 patient in the Abbas et al. study (16). In the current study, thoracic CT was taken of 12 (80%) of 15 patients, which was a much higher rate than previously reported in the literature. In 8 of the current study patients, although SPM was diagnosed on chest X-rays, thoracic CT was also taken and no treatment changes were made in any of these cases. Considering the benign nature of this condition, further investigations should be used only if there is a suspicion in the diagnosis or if there is a specific treatment option for the underlying cause.

Treatment for SPM includes bed rest, oxygen therapy, analgesics, bronchodilators for wheezing and asthma attack, and management of underlying conditions. Rarely, PM may be extensive, where a large volume of entrapped air interferes with adequate circulation and respiration, and would require surgical treatment. Cervical mediastinostomy with a suprasternal incision and dissection with a small bore catheter insertion may be necessary in these patients. Even the issue of whether to hospitalize patients with SPM is contradictory. Outpatient treatment and follow up is suitable for stable patients with no complications (17, 18), but if the child is young and the history of triggering events cannot be clearly established, a short in-hospital observation may be required

(19). For inpatients with a good general condition, if the treatment for the underlying disease is finished, complete resolution of the symptoms is sufficient for discharge on condition of close follow up after discharge. The length of hospital stay in previous studies varied between 1 and 53 days (20). In the current study, the mean length of stay was longer at 8.4 ± 4.5 days, which could be attributed to the high incidence of associated complications, such as pneumothorax (46.6% of the patients) and underlying conditions, such as lower respiratory tract infections.

The major limitations of this study were the small sample size and the retrospective design. The data used in this study were obtained from retrospective hospital records, and many clinical details may not have been complete, such as oxygen flow rate during oxygen treatment and long-term follow-up pulmonary function test results.

CONCLUSION

Considering the benign nature of SPM, further investigation tools should be used only if there is a specific treatment option or if there is suspicion in the diagnosis of the underlying cause. The problem of whether or not routine CT scanning should be performed in children with SPM has not yet been resolved, but if CT scanning is performed in every patient with SPM, pneumorrachis might be detected more frequently.

Ethics Committee Approval: Approval for the study was granted by the Clinical Research Ethics Committee of University of Health Sciences Antalya Training and Research Hospital (decision no:2020-017, dated: 23.01.2020). This study was conducted in accordance with the Declaration of Helsinki.

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