Asthmatic versus Non-Asthmatic Spontaneous Pneumomediastinum in Children

Chih-Yung Chiu^{1,2}, Kin-Sun Wong², Tsung-Chieh Yao² and Jing-Long Huang^{1,2}

SUMMARY The aim of this study was to analyze the clinical spectrum and seek potential curable causes of spontaneous pneumomediastinum (SPM) in children in order to minimize respiratory morbidity. Medical records from 1986 to 2003 were retrospectively reviewed at a tertiary pediatric facility in northern Taiwan. Sixteen cases of SPM were identified. There were eleven boys and five girls (M:F = 2.2:1) and ages ranged from 2 to 17 years (average, 10 years). Cough (81%), dyspnea (75%) and chest pain (56%) were the predominant symptoms and expiratory wheezing (63%) and neck crepitus (50%) were the most common physical findings. The specific sign of Hamman's crunch was noted in only one child initially. A coughing-related Valsalva maneuver (13 patients/81%) was the most common cause of pneumomediastinum in these children. The most common underlying medical causes were asthma (8 patients/50%) and idiopathic origin (5 patients/31%). Acute gastroenteritis, foreign body aspiration and mycoplasmal pneumonia were each found in one patient respectively. All patients had subcutaneous emphysema on initial chest radiographs. Two patients were complicated by pneumothorax and required intensive respiratory therapy. The average hospital stay was 4 days (range 1-9 days). Rapid resolution of symptoms without long-term sequelae was common except for one patient who had hypoxic-ischemic encephalopathy with epilepsy after foreign body removal. We conclude that in young teenagers, who suffer from cough, dyspnea, chest pain and associated discomfort of throat or neck, the diagnosis of SPM should be considered and chest radiography including posterioranterior and lateral projections should be performed to verify the diagnosis. Because of the high prevalence of asthma related SPM, children of idiopathic SPM should undergo diagnostic pulmonary function tests after the acute episode, to establish whether the child has asthma. Targeted investigations of the underlying causes of SPM might decrease respiratory morbidity and avoid further complications.

Pneumomediastinum, also known as mediastinal emphysema, is defined as the presence of air in the mediastinum. Spontaneous pneumomediastinum (SPM) occurs in patients who have non-traumatic mediastinal air leaks,¹ and is usually benign and selflimited. Although SPM is a well-known complication of children with asthma,^{2,3} patients usually benefit from early appropriate therapy when the true causes are identified. SPM can be serious and requires specific medical therapy. The purpose of this study was to analyze the clinical spectrum and seek potential curable causes of SPM in children in order to minimize respiratory morbidity.

MATERIALS AND METHODS

A computer search for the diagnosis of SPM in the hospital record of Chang Gung Children's Hospital was performed. Sixteen patients under 18 years of age were recruited from January 1986 to January 2003. Our children hospital, located in

Correspondence: Kin-Sun Wong

E-mail: pedchest@adm.cgmh.org.tw

From the ¹Department of Pediatrics, Chang Gung Memorial Hospital, Keelung, Taiwan, ²Department of Pediatrics, Chang Gung Children's Hospital and Chang Gung University, Taoyuan, Taiwan.

northern Taiwan, serves both as a primary care facility and a tertiary pediatric referral center with a surrounding population of over 2 million and 10,000 admissions annually. Pneumomediastinum was diagnosed by chest radiographs. Pneumomediastinum was defined as spontaneous if positive-pressure ventilation or trauma injuries were absent. Intubated patients and patients who had undergone cardiac or thoracic procedures were excluded. Patients who developed pneumomediastinum in the setting of a Valsalva maneuver were also considered "spontaneous". Presenting symptoms and signs, physical findings on presentation, precipitating factors, underlying medical causes, diagnostic investigations, associated complications and clinical outcome were reviewed. A medical cause was the diagnosis considered to most likely have led to the pneumomediastinum. Asthma related SPM (ARSPM) was defined as pneumomediastinum attributed to coughing during an acute asthmatic episode. Idiopathic SPM applied to those patients in whom the cause of pneumomediastinum was not identified. Hamman's sign which has a "crunching" or "clicking" noise synchronous with the heart beat was specifically sought in the chart review. Associated complications including pneumothorax, pneumopericardium, pneumoperitoneum and other associated long-term sequelae were analyzed. Radiographic features at presentation and the time of resolution were also recorded. The Student's t test was used to compare continuous variables. The χ^2 or Fisher's exact test was used to compare nominal data in unmatched groups. Statistical analysis was performed using the Statistical Program for Social Sciences (SPSS 10.0 for windows, SPSS Inc., Chicago, IL, USA). A *p* value of less than 0.05 was considered statistically significant.

RESULTS

Sixteen patients satisfying the criteria for SPM were identified and retrospectively reviewed. All these patients had previous admissions to our hospital and complete medical records were available for analysis. There were 11 boys and 5 girls, with ages ranging from 2 to 17 years and a mean age of 10 years. At the time of initial evaluation, all patients had one or more symptoms that could be related to the presence of a pneumomediastinum. Cough (81%), dyspnea (75%) and chest pain (56%) were the predominant symptoms. Sore throat and neck pain, and symptoms of discomfort referred to the throat or neck, were noted in six (37.5%) and four patients (25%), respectively. Dysphagia was noted in one child. Expiratory wheezing (63%) and

	ARSPM (%) (n = 8)	NARSPM (%) (n = 8)	Total (%) (n = 16)	р
Age (years)	9.6 ± 3.1	11.8 ± 3.7	10.7 ± 2.4	0.393
Symptoms				
Cough	8 (100)	5 (65)	13 (81)	0.200
Dyspnea	7 (88)	5 (65)	12 (75)	0.569
Chest pain	6 (75)	3 (38)	9 (56)	0.315
Sore throat	3 (38)	3 (38)	6 (38)	1.000
Neck pain	2 (25)	2 (25)	4 (25)	1.000
Cough & dyspnea	7 (88)	3 (38)	10 (63)	0.119
Cough & chest pain	6 (75)	1(13)	7 (44)	< 0.05
Dyspnea & chest pain	5 (65)	1(13)	6 (38)	0.119
Signs				
Wheezing	8 (100)	2 (25)	10 (63)	< 0.05
Neck crepitus	2 (25)	6 (75)	8 (50)	0.132
Hospital stay (days)	3.3 ± 1.6	4.8 ± 1.5	4.0 ± 1.1	0.198

 Table 1
 Comparison of demographic data, presenting symptoms/signs and length of hospital stay in ARSPM versus NARSPM patients

ARSPM, asthma related spontaneous pneumomediastinum; NARSPM, non-asthma related spontaneous pneumomediastinum

neck crepitus (50%) were the most common physical findings. Hamman's crunch was noted in only one child initially. In many cases, the physical findings such as neck crepitus and Hamman's crunch were not recognized initially until the radiographic findings of pneumomediastinum were noted.

Precipitating factors could be identified in 14 of the 16 cases. Coughing-related Valsalva maneuver was the most likely cause of pneumomediastinum in the majority of these children (13 patients/81%). Vomiting-related Valsalva maneuver and no apparent precipitating factors were determined in one and two patients, respectively. The most common underlying medical causes were asthma exacerbations (8 patients/50%) and idiopathic origin (5 patients/31%). Among the other three patients having SPM, one had acute gastroenteritis with forceful vomiting, one had a foreign body in the airways and one had mycoplasma pneumoniae pneumonia with forceful coughing.

All patients had subcutaneous emphysema on initial chest radiographs. One asthmatic patient had a small pneumothorax at the time of admission. One patient developed pneumothorax on the second day of hospitalization due to air trapping by a foreign body. Both of them required intensive therapy due to progressive respiratory failure. No subsequent pneumopericardium or pneumoperitoneum developed in our patients. Seven patients received serial chest radiographs and required an average of 4 days to resolve all radiographic changes. The average hospital stay was 4 days with a range of 1 day to 9 days. Rapid resolution of symptoms without long-term sequelae was common except for one patient who complicated with epilepsy due to hypoxic-ischemic encephalopathy after foreign body removal.

A study to differentiate non-asthma related SPM (NARSPM) from asthma related SPM (ARSPM) regarding demographic features, presenting symptoms/signs and length of hospital stay was performed. There were no significant differences between age, sex and duration of hospital stay in NARSPM versus ARSPM patients (p > 0.05). Further comparisons of presenting symptoms and signs are shown in Table 1. There were no significant differences in the frequency of these complaints alone between NARSPM and ARSPM. However, the co-

existence of cough and chest pain was significantly higher in ARSPM patients, especially those having wheezing episodes.

DISCUSSION

Spontaneous pneumomediastinum (SPM) usually occurred in the setting of a Valsalva type maneuver.¹ Clinically, SPM is frequently not diagnosed and is probably overlooked because the clinical signs and radiographic findings are subtle.⁴ Cough, dyspnea and chest pain were the most frequent complaints in children with SPM in this series as reported previously.^{5,6} However, SPM may be missed if patients have uncommon symptoms and signs.⁷⁻⁸ In our series, the unusual presenting symptoms of sore throat, neck pain and dysphagia were found in 6, 4 and 1 patients, respectively. An awareness of these uncommon symptoms that refer to the neck and throat should lead first-line physicians to early suspicion of SPM. Accordingly, neck crepitus or a Hamman's crunch which characterizes the presence of a SPM should vigilantly be looked for.⁵ However, neck crepitus and Hamman's crunch were noted initially only in seven (37.5%) and one patients (6.3%), respectively. Increased awareness of these pertinent findings should be emphasized to decrease the number of missed or delayed diagnoses of SPM.

The diagnosis of pneumomediastinum is ultimately confirmed by radiological findings.^{4,9} However, radiographic findings of SPM can be subtle and are easily overlooked by inexperienced primary care physicians. Lucent streaks or bubbles of gas that outline mediastinal structures confirm pneumomediastinum. Gas outlines the inner surface of the mediastinal pleura, creating a visible pleural line lateral to the main pulmonary artery and the aortic arch.¹⁰ The mediastinal gas is often seen above the left cardiac border and better visualized on the lateral view than posterior-anterior projection.⁸ More than 50% of pneumomediastinum may be missed if only a standard posterior-anterior film is examined.^{4,11} In case of doubt, the lateral projection of the chest may offer additional information to confirm a diagnosis of pneumomediastinum.

The majority of children with SPM had precipitating factors or underlying diseases. In our study, 81 percent of SPM were preceded by cough and half of the patients with SPM had asthma. The high prevalence of bronchial asthma or cough (or both) in these patients emphasizes the importance of recent respiratory obstruction in the pathogenesis of SPM. For this reason, we suggest that diagnostic pulmonary function tests should be performed after acute episodes, which should be designed to establish whether children with idiopathic SPM have asthma.

SPM generally resolves spontaneously within a few days, meaning that conservative treatment including bed rest, analgesics and oxygen is usually appropriate. Although precipitating factors should be identified and controlled to prevent recurrence, treatment of the underlying cause remains the mainstay of therapy. In children, SPM is most commonly associated with asthma and patients recover rapidly without sequelae.² Although the further complication of a pneumothorax may appear, a review of the literature showed that subsequent tension pneumothorax is extremely rare as in our series.^{4,12} In NARSPM patients, early recognition and therapy of the underlying cause, such as airway foreign body, should be able to avoid unnecessary life-threatening events and decrease morbidity. However, it is difficult to differentiate NARSPM from ARSPM patients by historical or demographic features. Nonetheless, the co-existence of cough and chest pain may be a very useful prognostic factor for ARSPM patients, especially in those patients having wheezing episodes.

The major limitations of our study are the retrospective nature of this review and the small sample size. Many patients who may have had SPM did not have it included in the discharge diagnosis. Furthermore, the finding of Hamman's crunch may have been missed by the first-line pediatricians due to unfamiliarity of this specific physical sign of SPM. Because of the limited number of cases reviewed, our study may not exactly represent the true manifestations of SPM in the general population. However, because the findings in this study are highly consistent with a review of the recent reports on this subject, we believe our findings or recommendations are generally applicable.

In conclusion, in young teenagers who present with cough, dyspnea, chest pain and associated discomfort of throat or neck, a diagnosis of SPM should be considered and the physical findings of neck crepitus or a Hamman's crunch should be specifically looked for. Chest radiography including posterior-anterior and lateral projections should be performed to verify the diagnosis. Because of the high prevalence of asthma related SPM, children of idiopathic SPM necessitate diagnostic pulmonary function tests after the acute episode, to establish whether the child has asthma. Although SPM is generally benign and self-limited, conservative management is usually appropriate; treatment of the underlying cause remains the primary goal of therapy. Targeted investigation of the underlying cause of SPM might decrease respiratory morbidity and avoid further complications.

REFERENCES

- 1. Chalumeau M, Le Clainche L, Sayeg N, *et al.* Spontaneous pneumomediastinum in children. Pediatr Pulmonol 2001; 31: 67-75.
- Stack AM, Caputo GL. Pneumomediastinum in childhood asthma. Pediatr Emerg Care 1996; 12: 98-101.
- 3. Dekel B, Paret G, Szeinberg A, Vardi A, Barzilay Z. Spontaneous pneumomediastinum in children: clinical and natural history. Eur J Pediatr 1996; 155: 695-7.
- 4. Versteegh FG, Broeders IA. Spontaneous pneumomediastinum in children. Eur J Pediatr 1991; 150: 304-7.
- Panacek EA, Singer AJ, Sherman BW, Prescott A, Rutherford WF. Spontaneous pneumomediastinum: clinical and natural history. Ann Emerg Med 1992; 21: 1222-7.
- Abolnik I, Lossos IS, Breuer R. Spontaneous pneumomediastinum. A report of 25 cases. Chest 1991; 100: 93-5.
- Werne C, Ulreich S. An unusual presentation of spontaneous pneumomediastinum. Ann Emerg Med 1985; 14: 1010-3.
- Ralph-Edwards AC, Pearson FG. Atypical presentation of spontaneous pneumomediastinum. Ann Thorac Surg 1994; 58: 1758-60.
- Bejvan SM, Godwin JD. Pneumomediastinum: old signs and new signs. AJR Am J Roentgenol 1996; 166: 1041-8.
- Zylak CM, Standen JR, Barnes GR, Zylak CJ. Pneumomediastinum revisited. Radiographics 2000; 20: 1043-57.
- Maunder RJ, Pierson DJ, Hudson LD. Subcutaneous and mediastinal emphysema. Pathophysiology, diagnosis, and management. Arch Intern Med 1984; 144: 1447-53.
- Damore DT, Dayan PS. Medical causes of pneumomediastinum in children. Clin Pediatr 2001; 40: 87-91.