Korean J Thorac Cardiovasc Surg 2018;51:254-259 □ CLINICAL RESEARCH □
ISSN: 2233-601X (Print) ISSN: 2093-6516 (Online) https://doi.org/10.5090/kjtcs.2018.51.4.254

Thoracic Scoliosis in Patients with Primary Spontaneous Pneumothorax

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Background: Primary spontaneous pneumothorax (PSP) affects patients without clinically apparent lung disorder found in tall and thin young male. Scoliosis refers to curves exceeding 10° Cobb angle observed through chest X-ray and affects 2% to 4% of adolescents. Both conditions are commonly encountered in primary health care setting. The aim of this study is to access the correlation of thoracic scoliosis and PSP in adolescent. **Methods:** A retrospective analysis was conducted for patients diagnosed for PSP in Konyang University Hospital between January 2010 and March 2017. Chest X-rays of 222 patients and 155 normal control (NC) cases were reviewed to measure the Cobb angle. Greater than 10° of Cobb angle is diagnosed as scoliosis. **Results:** Scoliosis in patient with PSP has higher incidence than that of NC group ($p<0.001$). Median value of Cobb angle is 12.9° in PSP group and 14.7° in NC group. Directional relationship between scoliosis and pneumothorax in PSP group is also observed; 40.5% cases are ipsilateral and 59.5% are contralateral. **Conclusion:** PSP patients tend to have thoracic scoliosis more commonly compared with normal healthy adolescent. Scoliosis may contribute to heterogeneity of alveolar pressure which exacerbates subpleural bleb formation that can cause pneumothorax. The causal relationship is unclear and further studies are needed in the future.

Key words: 1. Thorax
2. Pneumothorax
3. Scoliosis

Introduction

Spontaneous pneumothorax is a thoracic disorder defined as abnormal accumulation of air in the pleural space which occurs in the absence of precipitating factors such as thoracic trauma, surgery, or diagnostic procedure [1]. Primary spontaneous pneumothorax (PSP) affects patients with no clinically apparent lung or pleural disorders. It is generally accepted that PSP is found frequently in young male patients who are tall and thin. Scoliosis is defined as a lateral curvature of the spine greater than 10° as measured using the Cobb method on a standing radiography [2]. It is also commonly encountered clinical condition in adolescent. Upon careful observation, we have suspected that PSP patients tend to present thoracic scoliosis on chest X-ray (CXR). Therefore, we designed this study to access the cor-

Received: October 19, 2017, Revised: October 26, 2017, Accepted: November 6, 2017, Published online: August 5, 2018

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relation of thoracic scoliosis and PSP in the adolescent.

Methods

1) Study population

A retrospective analysis was conducted for patients diagnosed and treated for PSP in Konyang University Hospital between January 2010 and March 2017. The clinical records and CXR of 222 patients were retrospectively reviewed. Patients who underwent thoracic surgery due to chest wall deformity (i.e., pectus excavatum) and median sternotomy were excluded from this study since the operations of chest wall might affect structures of both chest wall and spine. Normal control (NC) group was selected by 2 ways; clinical data base of orthopedic surgery department of our institution collected from student health medical examination in 2016 and patients aged between 18 and 25 years old who underwent surgery at our institution from the same period also were selected as NC group.

2) Evaluation and definition

Pneumothorax was diagnosed by CXR and/or chest computed tomography. CXRs were taken twice for each PSP patient: when the patient was diagnosed as pneumothorax and when he/she first visited at outpatient department for regular follow-up after discharge. For scoliosis assessment, standing posteroanterior direct CXR is used to measure the Cobb angle. To measure the Cobb angle, the most tilted vertebrae (end-vertebrae) at the top and bottom of the spinal curve are identified. The Cobb angle can be measured manually be determining the angle between the intersecting lines, respectively, drawn tangentially along the upper endplate of the superior end-vertebra and the lower endplate of the inferior end-vertebra (Fig. 1). To minimize measurement error, 2 different assessors measured the Cobb angle independently. We stipulated a measurement discrepancy of $\geq 5°$ between the 2 assessors for re-assessment of the image. Additionally, to increase measurement quality, we used the average value of the 2 assessors.

Symptoms of pneumothorax such as pain and dyspnea could hinder patients from upright position and make pseudo-scoliosis on X-ray. Therefore, we selected the PSP patients with thoracic scoliosis if only both first and follow-up CXR images showed more than 10° in Cobb angle measurement with axial rotation.

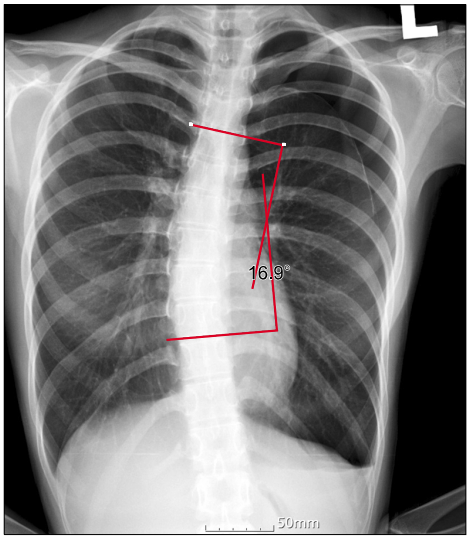

Fig. 1. Posteroanterior chest X-ray of patient with pneumothorax and scoliosis. The Cobb method is used to measure the degree of scoliosis.

For NC group, X-ray images of whole spine and/or CXR images were taken for each case. We used thoracic part of whole spine X-ray to evaluate thoracic scoliosis. Cobb angle for NC group was measured with the same method.

3) Statistical analysis

Statistical analysis was performed using IMM SPSS ver. 20.0 for Windows (IBM Corp., Armonk, NY, USA). The normality of the distribution of continuous variables was analyzed using the Shapiro-Wilk test. Differences in continuous variables of demographic data were analyzed using one-way analysis of variance. Continuous variables were analyzed using the Kruskal-Wallis test followed by the Mann-Whitney U-test when $p<0.05$. Categorical variables were analyzed using the χ^2 test or Fisher's exact test, as appropriate. A 2-tailed $p<0.05$ was considered statistically significant.

Results

Between January 2010 and March 2017, a total of 224 patients with PSP were treated at the Konyang

Table 1. Characteristics of PSP group and NC group

Characteristic	PSP (n=222)	NC (n=155)	p-value
Age (yr)	19 (18–25)	20 (18–25)	0.104
Gender			<0.001
Male	206 (92.8)	85 (54.8)	
Female	16 (7.2)	70 (45.2)	
Height (cm)	175 (151–188)	170 (150–185)	<0.001
Weight (kg)	60 (42–85)	65 (40–100)	<0.001
Laterality of pneumothorax			
Right	90 (40.5)	NA	
Left	132 (59.5)	NA	
No. of scoliosis	37 (16.7)	5 (3.2)	<0.001

Values are presented as median (range) or number (%).
PSP, primary spontaneous pneumothorax; NC, normal control; NA, not available.

Table 2. Characteristics of patients with scoliosis in PSP group and NC group

Characteristic	PSP (n=37)	NC (n=5)	p-value
Age (yr)	19 (18–25)	18 (18–23)	0.548
Gender			0.063
Male	35 (94.6)	3 (60)	
Female	2 (5.4)	2 (40)	
Height (cm)	176.8 (162–188)	171 (162–180)	0.034
Weight (kg)	60.5 (45–80)	68 (51.5–78.0)	0.363
Laterality of PNX			
Right	13 (35.1)	NA	
Left	24 (64.9)	NA	
Cobb angle	12.9 (10.6–28.5)	14.7 (12.1–17.0)	0.459
Direction of scoliosis			0.141
Right	33 (89.2)	3 (60)	
Left	4 (10.8)	2 (40)	
Directional relation between scoliosis & PNX			
Ipsilateral	15 (40.5)	NA	
Contralateral	22 (59.5)	NA	

Values are presented as median (range) or number (%).
PSP, primary spontaneous pneumothorax; NC, normal control; PNX, pneumothorax; NA, not available.

University Hospital. Among them 2 patients were excluded: 1 patient was undergone Nuss operation due to pectus excavatum and the other was undergone median sternotomy.

The characteristics of PSP group (n=222) and NC group (n=155) is summarized in Table 1. Median ages of PSP group and NC group are 19 and 20 years, respectively, with range of 18–25 years. Two hundred and six patients (92.8%) were male in PSP group while 85 (54.8%) were male in NC group ($p<0.001$). Median height and weight of PSP group are 175 cm (range, 151–188 cm) and 60 kg (range, 42–85 kg), respectively, which are significantly different from those of NC group ($p<0.001$). Number of patients diagnosed as scoliosis (Cobb angle greater than 10°) in PSP group is 37 (16.7%) which is statistically significant compared with those in NC group (3.2%, $p<0.001$).

The characteristics of patients with scoliosis in PSP group and NC group are summarized in Table 2. Median value of the Cobb angle is 12.9° (range, 10.6°–28.5°) in PSP group and 14.7° (range, 12.1°–17.0°) in NC group with no statistical significance (p=0.459). Laterality of pneumothorax shows predominance of

left side (64.9%) while direction of scoliosis (i.e., convex side of spinal curve on X-ray) shows predominance of right side in both PSP group and NC group (89.2% and 60%, respectively). Directional relationship between scoliosis and pneumothorax in PSP group is observed: 15 cases (40.5%) are ipsilateral and 22 cases (59.5%) are contralateral.

Discussion

This study demonstrates that the association between PSP and scoliosis in adolescent. Scoliosis was significantly common in PSP group compared to NC group (16.7% and 3.2%, respectively).

Pneumothorax is a thoracic disorder defined as abnormal collection of air or gas in the pleural cavity of the chest. Spontaneous pneumothorax which occurs in the absence of thoracic trauma, surgery, or diagnostic intervention, is further classified as primary or secondary [1]. PSP affects patients with no parenchymal lung disease while secondary spontaneous pneumothorax involves underlying lung or pleural diseases. PSP is relatively common condition in young adults as the reported incidence of PSP ranges between 8 and 28 cases per 100,000 per year for men and 1.2 to 6 cases per 100,000 per year for women [3,4]. PSP is not a rare disease, but a commonly observed disease in a community.

Scoliosis is a 3-dimensional deformation of spine defined as a lateral curve to the spine that is greater than 10° with vertebral rotation [2]. It is classified as congenital, neuromuscular, or idiopathic. Adolescent idiopathic scoliosis is found between age 10 years and skeletal maturity with no clear underlying cause [5]. The adolescent form accounts for the majority of cases of idiopathic scoliosis; approximately 85% of cases are idiopathic. Idiopathic scoliosis is commonly encountered in the primary care setting, affecting roughly 2% to 4% of adolescents [5-7]. This study also reveals 4.2% of scoliosis in NC group which is similar to other previous studies.

Primary physicians easily diagnose both pneumothorax and scoliosis by simple CXR which is the most economical imaging modality till date. Upon close observation of CXR and plentiful clinical experience, we have noticed that patients with PSP show more scoliosis features than those without pneumothorax, although the causal relationship is unclear.

Considering chest wall deformity and scoliosis, there are several studies that have revealed relationship of 2 clinical conditions found commonly in adolescent. Pectus excavatum which is one of the most frequently observed congenital deformity of chest wall has high percentage of association with scoliosis [8]. Johnson et al. [9] reported that about more than 20% of pectus excavatum patients had scoliosis. Moreover, other subclinical chest wall deformities such as flat chest or asymmetric chest had been revealed some correlation with PSP [10]. However, there have been no comprehensive analyses between pneumothorax and scoliosis. In this study, we observed statistically significant difference in incidence of scoliosis between PSP group and NC group ($p < 0.001$); patient with PSP shows much higher incidence of scoliosis than normal young adult.

The etiology of PSP still remains uncertain. The most common cause of PSP is rupture of an apical subpleural bullae or blebs [11]; however, the underlying etiology of such a bleb remains unclear either. According to the common consensus, PSP is frequently found in tall and thin young male patients, during their late adolescence or early adulthood. We re-confirmed these physical characteristics as well. In addition to these physical characteristics, we observed one additional characteristic of the patients with PSP which is thoracic scoliosis.

In this study, we excluded past history of thoracic surgery due to other chest wall deformities and applied strict criteria for selection of scoliosis in PSP group. Since pain and shortness of breath could affect the chest wall configuration we diagnosed scoliosis only if the measured Cobb angle showed more than 10° both before and after treatment of pneumothorax. We believed that this considerate selection of patients group might be helpful to exclude any pseudo-scoliosis.

There are some reports that have postulated a difference in alveolar pressure in upright human, between the base and the apex of lung, as a cause of bleb formation in the apical area which causes pneumothorax [10,12]. The rib cage in idiopathic scoliosis patients is narrower than in the non-scoliotic counterparts. Consequently, if the chest cannot develop normally during growth, there is insufficient space available for pulmonary alveolar growth [13,14].

Therefore, thoracic scoliosis observed in the PSP group may contribute to alveolar pressure heterogeneity and may exacerbate subpleural bleb formation.

Directional relationship between laterality of pneumothorax and direction of scoliosis was also analyzed in this study. In 15 out of 37 patients with PSP, scoliosis occurred at ipsilateral side of pneumothorax while contralateral cases were 22 out of 37 (40.5% and 59.5%, respectively). We hypothesized that different volume of thoracic cage affected by scoliosis might contribute to more heterogeneity in alveolar pressure gradient which could make contralateral predilection of pneumothorax. Our study showed slightly more incidence of contralateral relationship between pneumothorax and scoliosis. With a larger number of cases for the further study, this may show some statistically significant correlation.

Studies have attempted to uncover the pathophysiologic process underlying idiopathic scoliosis. Multiple abnormalities have been bound, yet none has been conclusively linked to all cases [5]. Up to date, the etiology of adolescent idiopathic scoliosis is believed to be multifactorial, including genetic factors. Muscular, nervous system, hormonal, and connective tissue defects have been noted in subgroups of patients with scoliosis.

Several connective tissue diseases as result of inherited genetic disorder are found in patients with pneumothorax and scoliosis. Patients with Marfan syndrome, one of the well-known major connective tissue disorders, tend to have very asymmetric thoraxes and frequently display spontaneous pneumothorax [15,16]. Scoliosis is also frequently observed in patients who are carriers of connective system diseases such as osteogenesis imperfect, Marfan syndrome or the Ehlers-Danlos syndrome [17-19]. Therefore, we assume that patients with PSP and scoliosis might be affected by subclinical and unidentified minor connective tissue disorders, although it remains still obscure whether an asymmetry in thoracic wall or spine due to scoliosis causes the heterogeneous alveolar pressure gradient that may contribute to subsequent bleb formation, or whether an unknown underlying connective tissue disorder is the common cause of thoracic and/or spine deformity and bleb formation. If further genetic studies were to be conducted in the future, a minor connective tissue disorder caused by the same specific gene may be identified in patients with PSP and scoliosis.

This study has several limitations. First, this study is a retrospective, observational and single-institutional-based analysis. Second, the statistical power may be weak because the number of study population is small. Therefore, a larger number of cases of both spontaneous pneumothorax and idiopathic scoliosis with well-designed prospective, randomized, controlled trials should be investigated in the future.

In conclusion, we found the association between pneumothorax and scoliosis. The incidence of scoliosis in patient with PSP is relatively higher than that of normal healthy adolescent. Scoliosis could cause asymmetry of thoracic cage or abnormal growth of pulmonary alveolar which exacerbate heterogeneity of alveolar pressure gradient and cause the formation of subpleural bleb. However, the causal relationship is unclear and further studies including genetic analysis are required.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Acknowledgments

This study was supported by a Grant of the Samsung Vein Clinic Network (Daejeon, Anyang, Cheongju, Cheonan; Fund no. KTCS04-108).

References

1. Baumann MH, Strange C, Heffner JE, et al. *Management of spontaneous pneumothorax: an American College of Chest Physicians Delphi consensus statement*. Chest 2001;119: 590-602.
2. Kane WJ. *Scoliosis prevalence: a call for a statement of terms*. Clin Orthop Relat Res 1977;(126):43-6.
3. Melton LJ 3rd, Hepper NG, Offord KP. *Incidence of spontaneous pneumothorax in Olmsted County, Minnesota: 1950 to 1974*. Am Rev Respir Dis 1979;120:1379-82.
4. Henry M, Arnold T, Harvey J; Pleural Diseases Group, Standards of Care Committee, British Thoracic Society. *BTS guidelines for the management of spontaneous pneumothorax*. Thorax 2003;58 Suppl 2:ii39-52.
5. Reamy BV, Slakey JB. *Adolescent idiopathic scoliosis: review and current concepts*. Am Fam Physician 2001;64: 111-6.
6. Horne JP, Flannery R, Usman S. *Adolescent idiopathic sco-*

liosis: diagnosis and management. Am Fam Physician 2014;89:193-8.
7. Lonstein JE. *Adolescent idiopathic scoliosis*. Lancet 1994;344:1407-12.
8. Jaroszewski D, Notrica D, McMahon L, Steidley DE, Deschamps C. *Current management of pectus excavatum: a review and update of therapy and treatment recommendations*. J Am Board Fam Med 2010;23:230-9.
9. Johnson WR, Fedor D, Singhal S. *Systematic review of surgical treatment techniques for adult and pediatric patients with pectus excavatum*. J Cardiothorac Surg 2014;9:25.
10. Saita K, Murakawa T, Kawano H, Sano A, Nagayama K, Nakajima J. *Chest wall deformity found in patients with primary spontaneous pneumothorax*. Asian Cardiovasc Thorac Ann 2013;21:582-7.
11. Getz SB Jr, Beasley WE 3rd. *Spontaneous pneumothorax*. Am J Surg 1983;145:823-7.
12. West JB. *Distribution of mechanical stress in the lung, a possible factor in localisation of pulmonary disease*. Lancet 1971;1:839-41.
13. Tsiligiannis T, Grivas T. *Pulmonary function in children with idiopathic scoliosis*. Scoliosis 2012;7:7.
14. Leong JC, Lu WW, Luk KD, Karlberg EM. *Kinematics of the chest cage and spine during breathing in healthy individuals and in patients with adolescent idiopathic scoliosis*. Spine (Phila Pa 1976) 1999;24:1310-5.
15. Wood JR, Bellamy D, Child AH, Citron KM. *Pulmonary disease in patients with Marfan syndrome*. Thorax 1984;39: 780-4.
16. Hall JR, Pyeritz RE, Dudgeon DL, Haller JA Jr. *Pneumothorax in the Marfan syndrome: prevalence and therapy*. Ann Thorac Surg 1984;37:500-4.
17. De Seze M, Cugy E. *Pathogenesis of idiopathic scoliosis: a review*. Ann Phys Rehabil Med 2012;55:128-38.
18. Hadley-Miller N, Mims B, Milewicz DM. *The potential role of the elastic fiber system in adolescent idiopathic scoliosis*. J Bone Joint Surg Am 1994;76:1193-206.
19. Miller NH, Mims B, Child A, Milewicz DM, Sponseller P, Blanton SH. *Genetic analysis of structural elastic fiber and collagen genes in familial adolescent idiopathic scoliosis*. J Orthop Res 1996;14:994-9.