Correlation between diaphragmatic sagittal rotation and pulmonary dysfunction in patients with ankylosing spondylitis accompanied by kyphosis

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Abstract

Objective: This study was performed to investigate the correlation between pulmonary dysfunction patterns and diaphragmatic sagittal rotation in patients with ankylosing spondylitis accompanied by kyphosis.

Methods: Thirty patients (27 male, 3 female) with kyphotic deformity secondary to ankylosing spondylitis underwent pedicle subtraction osteotomy and were retrospectively reviewed. All patients had undergone preoperative computed tomography with three-dimensional reconstruction, full-length spine radiographs, and pulmonary function tests. The diaphragmatic angle in the median sagittal plane (DA), pulmonary function test results, and radiological parameters were studied.

Results: Correlation coefficients were used to present the correlation between the DA and pulmonary function and the global kyphosis (GK), respectively. The data analysis presented positive correlations between the DA value and vital capacity (VC), forced vital capacity (FVC), expiratory reserve volume (ERV), inspiratory reserve volume (IRV) and peak expiratory flow (PEF). There was likewise a negative correlation between DA value and the global kyphosis (GK). Additionally, there were further significantly statistical improvements for DA, ERV, IRV, FVC, and VC, PEF, postoperatively.
Conclusions: Except for the restriction of the chest wall motion and the abnormalities of lung parenchyma, the diaphragmatic sagittal rotation is also an influencing factor of pulmonary dysfunction in patients with ankylosing spondylitis accompanied by kyphosis.

Keywords
Kyphosis, ankylosing spondylitis, osteotomy, diaphragm, pulmonary function, diaphragmatic sagittal rotation

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Introduction
Ankylosing spondylitis (AS) is a common inflammatory rheumatic disease that predominantly affects the sacroiliac joints and spine and causes characteristic spinal deformities such as flattening of the normal lumbar lordosis, which can lead to structural and functional impairments and a decreased quality of life. In severe cases, the spinal kyphotic deformity results in muscle fatigue, activity-related pain due to continuous strain on the spinal muscles during erect standing, the inability to look straight ahead, intra-abdominal complications, and impaired respiratory function. Restriction of chest wall motion and abnormalities of the lung parenchyma are major factors associated with pulmonary dysfunction. Liu et al. demonstrated that the diaphragm exhibited rotation in the sagittal plane in patients with AS-related kyphotic deformity. However, whether such rotation influences pulmonary function in these patients has not been investigated.

Materials and methods
Patients
Patients in our department who had AS-related kyphotic deformity and had undergone pedicle subtraction osteotomy were retrospectively reviewed. The inclusion criteria were as follows: (1) the diagnosis of AS fulfilled the modified New York criteria; (2) the types of AS-related kyphosis were lumbar and thoracolumbar kyphosis; (3) all patients were nonsmokers, free of cardiopulmonary disorders, and experienced in performing respiratory maneuvers; (4) preoperative and postoperative computed tomography scans included the pubis and sacrum; and (5) preoperative pulmonary function was evaluated. All patients provided verbal informed consent for inclusion in the study. This study was approved by the medical ethics committee of Suzhou Kowloon Hospital.

Cobb’s angle measurement
Global kyphosis (GK) was measured from the superior endplate of the T1 vertebra to the superior endplate of the S1 vertebrae.

Computed tomographic measurement
The diaphragmatic angle (DA) in the median sagittal plane was measured to quantify the degree of diaphragmatic rotation in the sagittal plane. The DA was defined as negative when the E-F line was in front of the P line (Figure 1(a)), and the DA was defined as positive when the P line
was in front of the E-F line (Figure 1(b)). When the DA was smaller, the degree of diaphragmatic rotation in the sagittal plane was more severe.

**Pulmonary function test results**

All patients in this study underwent pulmonary function tests to evaluate their preoperative physical condition. An experienced physician's assistant took each measurement using a digital spirometer (Renaissance II; Puritan Bennett, Boulder, CO) with the patients in the standing position. The following pulmonary function parameters were evaluated: vital capacity (VC), forced VC (FVC), forced expiratory volume in 1 s (FEV1), FEV1/FVC, maximal voluntary ventilation (MVV), expiratory reserve volume (ERV), inspiratory reserve volume (IRV), and peak expiratory flow (PEF). All values are presented as the percentage of the measured value / the predicted value.

**Statistical analysis**

The data analyses were performed with SPSS version 16.0 for Windows (SPSS Inc., Chicago, IL). Correlation coefficients were utilized to present the correlations of the DA with the pulmonary function values and GK. A p-value of $<0.05$ was considered statistically significant in all analyses.

**Results**

Thirty patients (27 male, 3 female) with an average age of 38.6 years (range, 26–54 years) who satisfied the aforementioned inclusion criteria were enrolled in this study. The data analysis revealed positive correlations between the DA and VC, FVC, ERV, IRV, and PEF. No significant correlation was found between the DA and the other pulmonary function parameters (Table 1). These results suggest that
Diaphragmatic rotation in the sagittal plane of patients with AS can reduce the VC and FVC by reducing the IRV and ERV, and these parameters would theoretically be improved by correction surgery. Data analysis demonstrated a negative correlation between the DA and GK (Table 1). This result indicates that the degree of diaphragmatic rotation in the sagittal plane becomes more severe as the GK increases.

### Discussion

Pulmonary dysfunction is prevalent in patients with AS-related kyphotic deformity. The most prevalent pattern of pulmonary dysfunction is restrictive ventilatory impairment. Costovertebral and sternoclavicular joint involvement result in chest wall restriction, and lung parenchymal abnormalities cause the pulmonary dysfunction that develops in patients with AS.\(^\text{11-14}\) Because chest expansion is limited in patients with AS, ventilation becomes more dependent on the diaphragm, and respiration becomes compensated by the diaphragm, especially during hyperventilation.\(^\text{15-17}\) During ventilation, the diaphragm acts mainly as a flow generator.\(^\text{18}\) However, none of the patients in previous studies had kyphotic deformity, and diaphragmatic rotation in the sagittal plane was not observed in those patients. However, all patients with AS-associated kyphotic deformity exhibit diaphragmatic rotation in the sagittal plane.\(^\text{10}\) Therefore, the present study was performed to investigate whether diaphragmatic sagittal rotation influences pulmonary function in patients with AS-related kyphotic deformity.

In this study, all patients exhibited complete fusion of the costovertebral and sternoclavicular joints and limited rib cage motion. The maximal anterior attachment point of the diaphragm is the xiphoid process, and the maximal posterior attachment point is the T12 vertebra. The xiphoid process is approximately in the same coronal plane as the T9 vertebra. Diaphragmatic rotation leads to a change in the orientation of the diaphragm (Figure 1(a)). The diaphragmatic attachment point is irregular; therefore, to show the moving orientation in the sagittal plane, the A-B line between the xiphoid process and the anteroinferior edge of T12 was adopted to represent the diaphragmatic plane in the median sagittal plane (A-B line in Figure 1(a), (b)). The DA was assessed to quantify the degree of diaphragmatic rotation in the sagittal plane.\(^\text{10}\) In Figure 1(a) and (b), the P line perpendicular to the A-B line between the xiphoid process and the anteroinferior edge of T12 was adopted to represent the diaphragmatic plane in the median sagittal plane (A-B line in Figure 1(a), (b)). The DA was assessed to quantify the degree of diaphragmatic rotation in the sagittal plane.\(^\text{10}\) The DA was the angle between the P line and the E-F line in Figure 1(a) and (b). The E-F line is the line between the midpoints of the following two lines: 1) the midpoint E of the A-B line between the xiphoid process and the anteroinferior edge of T12 and 2) the midpoint F of the C-D line between the superior edge of the pubis and the anterosuperior corner of the sacrum. Because the xiphoid process

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**Table 1. Pearson coefficient analysis results**

| Parameters       | r    | p     |
|------------------|------|-------|
| DA and ERV       | 0.3893 | 0.033* |
| DA and IRV       | 0.6184 | 0.000**|
| DA and PEF       | 0.4966 | 0.005**|
| DA and FVC       | 0.3633 | 0.048 |
| DA and FEV1      | 0.0387 | 0.839 |
| DA and FEV1/FVC  | 0.1064 | 0.576 |
| DA and MVV       | 0.1249 | 0.511 |
| DA and VC        | 0.3732 | 0.042 |
| DA and GK        | -0.5337 | 0.002**|

DA, diaphragmatic angle in the median sagittal plane; ERV, expiratory reserve volume; IRV, inspiratory reserve volume; PEF, peak expiratory flow; FVC, forced vital capacity; FEV1, forced expiratory volume in 1 s; FEV1/FVC: forced expiratory volume in 1 s/forced vital capacity; MVV, maximal voluntary ventilation; VC, vital capacity; GK, global kyphosis. *p < 0.05, **p < 0.01
is approximately in the same coronal plane as the T9 vertebra, the E-F line is in front of the P line in normal people. The DA is thus positive in normal people. In patients with AS-associated kyphotic deformity (Figure 1(a)), the kyphotic deformity causes the P line to be located behind the E-F line before surgery. The preoperative DA was negative in patients with kyphotic deformity. Liu et al. noted that the diaphragm was more severely rotated in patients with greater GK. A positive correlation was found between the DA and IRV. The correlation showed that the IRV was reduced because of diaphragmatic rotation in the sagittal plane. The IRV is the extra volume of air that can be inspired with maximal effort at the end of a normal inspiration. During the inspiration process, the diaphragm pushes the viscus downward, thus expending the anterior abdominal wall, increasing the thoracic volume, and pumping air into the lung. Because of the diaphragmatic rotation in the sagittal plane, the diaphragmatic orientation is more backward in patients with than without AS-associated kyphotic deformity, and the diaphragm pushes the viscus further backward. Additionally, relative to the anterior abdominal wall, the posterior abdominal wall cannot be stretched. The posterior abdominal wall cannot compensate for the decreased abdominal volume, especially during hyperventilation, and the range of diaphragmatic motion is decreased due to diaphragmatic rotation in the sagittal plane in patients with AS-related kyphotic deformity. In the present study, the IRV was reduced due to the change in the diaphragmatic motion orientation resulting from diaphragmatic rotation in the sagittal plane in patients with AS-associated kyphotic deformity.

The ERV is the extra volume of air that can be exhaled with maximal effort at the end of a normal expiration. During the expiration process, contraction of the anterior abdominal wall muscle reduces the abdominal volume, squeezing the viscus, which pushes the diaphragm upward and compresses the lungs, pushing the air out. In patients with AS-related kyphotic deformity, however, the change in the diaphragmatic motion orientation and the shortened anterior abdominal wall reduces the effects of the anterior abdominal wall and diaphragm during the expiration process. The present study showed a positive correlation between the DA and the ERV and PEF. The positive correlations suggested that the ERV and PEF decrease with diaphragmatic rotation in the sagittal plane.

The VC comprises the TV, ERV, and IRV. Therefore, diaphragmatic rotation in the sagittal plane reduces the VC and FVC by reducing the ERV and IRV. This study showed positive correlations of the DA with the FVC and VC.

Spinal osteotomy is the only way to restore the sagittal balance in patients with AS by improving GK, lumbar lordosis, and thoracolumbar kyphosis. In most patients with AS, pedicle subtraction osteotomy is performed at the lumbar vertebrae, and neither thoracic kyphosis nor the thoracic cage is significantly changed. Because the DA needs to be improved as the GK is improved, the pulmonary function in patients with AS-associated kyphosis is also theoretically improved. Fu et al. demonstrated that the postoperative pulmonary function in patients with AS-associated kyphosis was significantly improved at the 2-year follow-up. In our pilot study, we also found statistically significant improvements in the DA, ERV, IRV, FVC, VC, and PEF postoperatively. Hence, rotation of the diaphragm in the sagittal plane reduced the VC and FVC by reducing the ERV and IRV, and corrective surgery could improve pulmonary function in patients with AS-associated kyphosis by reducing the rotation of the diaphragm in the sagittal plane.
In summary, in addition to restriction of chest wall motion and abnormalities of the lung parenchyma, diaphragmatic sagittal rotation is an influential factor of pulmonary dysfunction in patients with AS-related kyphosis.

Declaration of conflicting interest
The authors declare that there is no conflict of interest.

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References
1. Van Royen BJ, De Gast A and Smit TH. Deformity planning for sagittal plane corrective osteotomies of the spine in ankylosing spondylitis. *Eur Spine J* 2000; 9: 492–498.
2. Van Royen BJ, De Kleuver M and Slot GH. Polysegmental lumbar posterior wedge osteotomies for correction of kyphosis in ankylosing spondylitis. *Eur Spine J* 1998; 7: 104–110.
3. Braun J and Sieper J. Ankylosing spondylitis. *Lancet* 2007; 369: 1379–1390.
4. Goie The HS, Steven MM, van der Linden SM, et al. Evaluation of diagnostic criteria for ankylosing spondylitis: a comparison of the Rome, New York and modified New York criteria in patients with a positive clinical history screening test for ankylosing spondylitis. *Br J Rheumatol* 1985; 24: 242–249.
5. Ragnarsdottir M, Geirsson AJ and Guðbjörnsson B. Rib cage motion in ankylosing spondylitis patients: a pilot study. *Spine J* 2008; 8: 505–509.
6. Chang KW. Quality control of reconstructed sagittal balance for sagittal imbalance. *Spine (Phila Pa 1976)* 2011; 36: E186–E197.
7. Suk KS, Kim KT, Lee SH, et al. Significance of chin-brow vertical angle in correction of kyphotic deformity of ankylosing spondylitis patients. *Spine (Phila Pa 1976)* 2003; 28: 2001–2005.
8. Kim KT, Suk KS, Cho YJ, et al. Clinical outcome results of pedicle subtraction osteotomy in ankylosing spondylitis with kyphotic deformity. *Spine (Phila Pa 1976)* 2002; 27: 612–618.
9. Cho H, Kim T, Kim TH, et al. Spinal mobility, vertebral squaring, pulmonary function, pain, fatigue, and quality of life in patients with ankylosing spondylitis. *Ann Rehabil Med* 2013; 37: 675–682.
10. Liu C, Song K, Zhang YG, et al. Changes of the abdomen in patients with ankylosing spondylitis kyphosis. *Spine (Phila Pa 1976)* 2015; 40: E43–E48.
11. Hunninghake GW and Fauci AS. Pulmonary involvement in the collagen vascular diseases. *Am Rev Respir Dis* 1979; 119: 471–503.
12. Tanoue LT. Pulmonary involvement in collagen vascular disease: a review of the pulmonary manifestations of the Marfan syndrome, ankylosing spondylitis, Sjogren’s syndrome and relapsing polychondritis. *J Thorac Imaging* 1992; 7: 62–77.
13. Wiedemann HP and Matthay RA. Pulmonary manifestations of the collagen vascular diseases. *Clin Chest Med* 1989; 10: 677–722.
14. Cerrahoglu L, Unlu¨ Z, Can M, et al. Lumbar stiffness but not thoracic radiographic changes relate to alteration of lung function tests in ankylosing spondylitis. *Clin Rheumatol* 2002; 21: 275–279.
15. Grimby G, Fugl-Meyer AR and Bloomstand A. Partitioning of the contribution of rib cage and abdomen to ventilation in ankylosing spondylitis. *Thorax* 1974; 29: 179–184.
16. Hauge BN. Diaphragmatic movement and spirometric volume in patients with ankylosing spondylitis. *Scand J Respir Dis* 1973; 54: 38–44.
17. Ünlü E, Pamuk ÖN, Erer B, et al. Diaphragmatic movements in ankylosing spondylitis patients and their association with clinical factors: an ultrasonographic study. *Rheumatol Int* 2012; 32: 435–437.
18. Sanna A, Bertoli F, Misuri G, et al. Chest wall kinematics and respiratory muscle......
action in walking healthy humans. *J. Appl. Physiol* 1999; 87: 938–946.

19. Willems KF, Slot GH, Anderson PG, et al. Spinal osteotomy in patients with ankylosing spondylitis: complications during first postoperative year. *Spine (Phila Pa 1976)* 2005; 30: 101–107.

20. Fu J, Zhang G, Zhang Y, et al. Pulmonary function improvement in patients with ankylosing spondylitis kyphosis after pedicle subtraction osteotomy. *Spine (Phila Pa 1976)* 2014; 39: 1116–1122.