Distal Adding On In Lenke 2AR Adolescent Idiopathic Scoliosis: risk Factors Analysis and Clinical Assessment

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Abstract

Background: Risk factors and clinical outcome associated with distal adding on in Lenke 2AR adolescent idiopathic scoliosis (AIS) are not fully elucidated.

Methods: Radiographic data of Lenke 2AR AIS patients treated by posterior pedicle screws in a single institution were retrospectively analyzed. Patients who developed distal adding on were compared with patients who did not. Clinical assessment were carried out by SF-36 and SRS-22 questionnaire. Risk factors of distal adding on were explored by binary logistic regression analysis.

Results: A total of 88 patients completed an average of 2.9 (2-12.9) years of follow-up were included. 18 (20.5%) patients met the criterion of distal adding on while the rest of 70 cases did not at follow up. Clinical parameters were comparable before surgery and became lower at follow up in adding on group in dimensions of vitality (63.1±11.4Vs.67.5±11.8,P=0.026), mental health (73.9±9.2Vs.77.8±10.2,P=0.039), self-image (3.9±0.3Vs.4.2±0.3,P=0.021) and satisfaction (4.0±0.3Vs.4.5±0.4,P=0.035). The proportion of selection lowest instrumented vertebra (LIV) higher than last touching vertebra (LTV) was significantly higher in adding on group. (6/18vs 4/70, P=0.004) Binary logistic regression analysis revealed that preoperative main thoracic curve magnitude (OR=1.118,P=0.041), preoperative lumbar lordosis (OR=1.091,P=0.029), and the gap between LIV and LTV(OR=6.123,P=0.043) were independent risk factors associated with adding on.

Conclusions: In Lenke 2AR AIS cases, the selection of LIV higher than LTV is closely correlated with the development of adding-on. Patients with minor main thoracic curve and lumbar lordosis are more likely to develop distal adding on. Adding on may cause discontent of patients.

Introduction

Adding-on is described as progression or extension of the major thoracic curve after selective fusion of thoracic curve in Lenke 1 and Lenke 2 adolescent idiopathic scoliosis (AIS) patients. 1 Risk factors of distal adding on in Lenke 1 AIS such as young age and less fusion segments have been extensively studied, 1,2,3 data about Lenke 2 cases was much less. A proximal thoracic (PT) curve in Lenke 2 scoliosis may cause shoulder imbalance. The left shoulder is often elevated by the proximal thoracic curve before surgery and could be further elevated after surgery. 4,5 In Lenke 2 scoliosis, the mechanism of distal adding on is even more complicated because of the underlying compensation of shoulder imbalance and little literature has explored the association between shoulder imbalance and adding on. 6 Lenke 1A was divided into two different lumbar sub-types according to the tilt direction of L4. 7 Lenke 1AR was reported to have higher incidence of adding on. 8 Thus Lenke 2AR was inferred to have high risk of adding on. The risk factors of adding on in Lenke 2AR haven’t been fully reported. 9,10 Although adding on has been widely discussed radiographically, 11 the psychological and physical influences of adding on to patients haven’t been clearly reported. We reviewed a series of consecutive cases of Lenke 2AR AIS treated by posterior pedicle screws instrumentation. We sought to answer the following three questions in
this study: 1. What was the risk factors of adding on in Lenke 2AR scoliosis? 2. Is there an association between shoulder imbalance and adding on? 3. Does adding on phenomenon have a negative effect on patients?

**Materials And Methods**

**Patients**

The institutional review board of our hospital approved all aspects of this study. A total of 121 patients diagnosed with Lenke 2AR AIS treated with pedicle screws in our institution were screened, finally 88 patients who completed a minimum of 2-year follow up were retrospectively analyzed clinically and radiographically.

**Inclusion and Exclusion Criteria**

Lenke 2 type AIS with lumbar modifier A and superior endplate of L4 vertebra tilting to the right were included in the study. If the direction of L4 was horizontal and difficult to judge, L3 was selected. (see figure 1) Cases which underwent osteotomy or anterior release were excluded. Revision surgery or scoliosis of other causes was also excluded.

**Surgical Technique**

All the surgical procedures were performed by senior spine surgeons in our center. All patients were treated using posterior segmental spinal instrumentation with monitoring of motor-evoked potentials (MEP). Lamina hooks may be occasionally used in upper thoracic as a result of failure in pedicle screw placement. Preoperative traction was not performed. The upper instrumented vertebra (UIV) was chosen based on the preoperative shoulder balance on AP film. If the patient presented with right shoulder elevation, T4 would be chosen as UIV. If the patient presented with level shoulders or left shoulder elevation, T1 or T2 would be chosen as UIV. Usually, the lowest instrumented vertebra (LIV) was selected at last touching vertebra (LTV) or last substantially touching vertebra (LSTV). Sometimes, the LIV may not be comply with the aforementioned standards according to the specific conditions during the operation. For example, if the curve was more flexible than anticipated, shorter fusion (LTV-1) may be performed. Rod derotation and direct vertebral rotation were applied according to the surgeon's preferences. After correction, a fluoroscopy would be taken to check the T1 tilt and shoulder balance. If the left shoulder was elevated, a “level by level” compression of the PT on the left side and the distraction on the right side would be performed.

**Clinical assessment**

The patients were asked to complete SF-36 and SRS-22 questionnaires before the surgery and at each follow up (3 months, 6 months, 1 year, 2 years and 5 years after surgery until 18 years old). These data were prospectively collected and retrospectively reviewed. The SRS-22 Outcomes Instrument has been a widely used HRQL questionnaire to evaluate the perception of patients suffering from scoliosis. A
simplified Chinese version was used in mainland China.\textsuperscript{12} The Chinese (main land) version of the SF-36, quite similar to the original American population, was also tested in reliability, convergent, and discriminant validity.\textsuperscript{13} The questionnaires were assigned and collected by a senior nurse who did not participate in the study.

**Radiographic Measurements**

A standing full-length anterior-posterior (AP), lateral and side-bending (SB) radiographs were taken before operation. Standing full-length AP and lateral film were taken at immediate postoperative (before discharge from the hospital) and last follow up. Radiographic measurements were taken by the Digimizer (MedCalc Software bvba, Belgium) software.

The Cobb angle of the PT and main thoracic(MT) were measured to evaluate the correction rates. Parameters related to should balance include T1 tilt, first rib angle (FRA), clavicle angle (CA), trapezial angle (TA), and shoulder height difference (SHD) were also measured.\textsuperscript{14,15} T1 tilt was defined by the angle of the upper end plate of T1 vertebrae and the horizontal line. First rib angle was defined by the angle of the horizontal line and the tangential line that connects the superior border of the first ribs. Clavicle angle was defined by the angle between the horizontal line and the tangential line that connects the upper end of each clavicle. The trapezial angle was defined by the angle between the horizontal line and the line connecting the intersections of sternocleidomastoid muscle and trapezius muscle profiles. The shoulder heights difference (SHD) was defined by the height discrepancy between the lateral ends of the clavicle. All of the above parameters were defined positive with left shoulder up and right shoulder down. Coronal balance was defined by the distance between C7 plumb line and center sacral vertical line (C7P-CSVL) on the standing AP film. The stable vertebra (SV), last touching vertebra (LTV) and last substantially touching vertebra (LSTV) were determined in standing AP film. (Figure 1) Distal adding on was defined as a progressive increase in the number of vertebrae included within the distal curve, with either more than 10mm deviation from CSVL of lower instrumented vertebra, more than 5 mm deviation of the first vertebra from the CSVL below instrumentation or an increase of more than 5° in the angle of the first disc below the instrumentation.\textsuperscript{16}

All the parameters were measured twice by 2 fellows to improve the accuracy and the mean value was used for statistical analysis.

**Statistical Analysis**

Statistical analysis was performed using SPSS version 16.0 (SPSS, Inc., Illinois, USA). All continuous variables were written as a mean ± standard deviation. Independent T-tests and chi-square test were carried out to assess the differences of clinical and radiographic parameters between the adding on and non-adding on group. Factors related to the development of adding-on were identified by binary logistic regression analysis. Statistical significance was considered when P< 0.05.
Results

A total of 88 cases of AIS completed an average of 2.9 (2-12.9) years of follow-up. 18 (20.5%) patients met the criterion of distal adding on while the rest of 70 cases did not at last follow up. The preoperative parameters were shown in table 1. Age and Risser sign were significantly lower in adding on group compared with non-adding on group. The Cobb angle of PT and MT were also smaller in adding on group. In sagittal plane, thoracic kyphosis was slightly lower and lumbar lordosis was significantly lower in adding on group. Shoulder balance and coronal balance were comparable between the two groups.

|                         | Adding on(18) | Non adding on(70) | P  |
|-------------------------|---------------|-------------------|----|
| age                     | 12.8±2.5      | 14.7±2.3          | 0.047 |
| Risser sign             | 2.6±1.7       | 3.5±1.1           | 0.010 |
| Pre-PT                  | 32.1±8.4      | 38.1±11.1         | 0.035 |
| Pre-MT                  | 42.0±9.4      | 51.8±13.8         | 0.006 |
| Pre-TK                  | 11.6±9.1      | 18.2±15.6         | 0.089 |
| Pre-LL                  | 37.0±11.7     | 47.6±12.3         | 0.002 |
| Pre-T1 tilt             | 6.9±5.8       | 8.4±7.2           | 0.392 |
| Pre-FRA                 | 5.5±5.2       | 6.3±5.8           | 0.627 |
| Pre-CA                  | 0.7±2.2       | 0.4±2.1           | 0.587 |
| Pre-TA                  | 2.2±3.0       | 2.7±4.1           | 0.683 |
| Pre-SHD                 | -0.05±1.1     | 0.1±1.3           | 0.643 |
| Pre-C7PL-CSVL           | 1.5±1.0       | 1.4±1.4           | 0.647 |
| Pre-PT bending          | 21.9±11.6     | 27.4±13.0         | 0.124 |
| Pre-DT bending          | 18.4±14.3     | 23.2±15.9         | 0.251 |

Table 1. Comparison of preoperative parameters between patients with adding on and without adding on.

*PT, proximal thoracic; MT, main thoracic; TK, thoracic kyphosis; LL, lumbar lordosis; FRA, first rib angle; CA, clavicle angle; TA, trapezial angle; SHD, shoulder height difference; C7PL-CSVL, C7 plumb line-center sacral vertical line

Clinical assessment before surgery and at last follow up between patients with adding on or not was shown in table 2. The preoperative clinical score was comparable between the two groups. At follow up, the scores of cases with adding on were lower in vitality and mental health dimensions. Self-image, mental health and satisfaction were also lower in adding on group by SRS-22 questionnaires.
Table 2. The comparison of clinical assessment between patients with adding on and without adding on by SF-36 and SRS-22 questionnaire before surgery and at last follow up.  

| Questionnaire | preoperative | follow up | P value | P value |
|---------------|--------------|-----------|---------|---------|
|               | Adding on    | Non-adding on | Adding on | Non-adding on |
| SF-36         |              |            |         |          |
| PF            | 81.0±5.7     | 83.0±6.4   | 0.254   | 83.1±4.9 | 84.2±5.9 | 0.173   |
| RP            | 76.4±16.3    | 77.5±17.1  | 0.219   | 73.3±15.4 | 74.3±16.5 | 0.309   |
| BP            | 92.1±5.8     | 91.7±6.0   | 0.371   | 92.5±5.5 | 90.5±5.5 | 0.072   |
| GH            | 64.2±12.4    | 66.8±13.6  | 0.581   | 70.5±10.3 | 73.1±12.4 | 0.058   |
| VT            | 65.7±11.5    | 67.7±12.0  | 0.106   | 63.1±11.4 | 67.5±11.8 | 0.026   |
| SF            | 78.6±10.4    | 79.1±11.4  | 0.232   | 83.6±9.9 | 84.6±10.2 | 0.232   |
| RE            | 69.4±23.5    | 68.6±24.7  | 0.554   | 85.2±15.5 | 86.4±16.3 | 0.107   |
| MH            | 78.3±11.7    | 77.1±10.4  | 0.239   | 73.9±9.2 | 77.8±10.2 | 0.039   |
| SRS-22        |              |            |         |          |
| FA            | 4.1±0.1      | 4.0±0.2    | 0.837   | 4.0±0.2 | 4.1±0.2 | 0.586   |
| Pain          | 4.1±0.2      | 4.2±0.2    | 0.428   | 4.1±0.1 | 4.2±0.2 | 0.323   |
| Self-image    | 3.5±0.2      | 3.4±0.3    | 0.335   | 3.9±0.3 | 4.2±0.3 | 0.021   |
| MH            | 3.8±0.2      | 3.9±0.1    | 0.440   | 3.9±0.3 | 4.1±0.3 | 0.018   |
| Satisfaction  | NA           | NA         | 4.0±0.3 | 4.5±0.4 | 0.035   |

After operation, the average SHD was larger in adding on group, and the fusion segments were significantly shorter in adding on group. The proportion of selection LIV higher than LTV was significantly higher in adding on group. The average LIV was higher than LTV in adding on group while the average LIV was lower than LTV and higher than LSTV in non-adding on group (Table 3).
Table 3. Comparison of parameters at immediate postoperative between patients with and without adding on at follow up

|                         | Adding on(18) | Non adding on(70) | P    |
|-------------------------|---------------|-------------------|------|
| Male percentage         | 5/18          | 13/70             | 0.512|
| Post-PT                 | 13.5±7.6      | 15.0±7.7          | 0.475|
| Post-MT                 | 10.7±7.6      | 11.3±7.9          | 0.750|
| Post-TK                 | 16.0±7.2      | 16.9±6.7          | 0.602|
| Post-LL                 | 40.8±8.4      | 44.4±9.2          | 0.137|
| Post-T1 tilt            | 5.8±4.9       | 7.7±4.8           | 0.157|
| Post-FRA                | 5.1±3.5       | 6.3±3.8           | 0.251|
| Post-CA                 | 2.6±1.7       | 2.5±2.6           | 0.186|
| Post-TA                 | 3.4±2.7       | 3.7±3.1           | 0.794|
| Post-SHD                | 1.8±1.1       | 1.1±1.4           | 0.012|
| Post-C7PL-CSVL          | 1.5±0.7       | 1.0±0.8           | 0.125|
| Fusion segments         | 11.2±1.5      | 12.4±1.4          | 0.001|
| UIV                     | 2.8±1.7       | 2.5±1.2           | 0.312|
| LIV= LTV-1              | 6             | 4                 | 0.004|
| LIV≥LTV                 | 12            | 66                |      |
| LIV-LTV                 | -0.056±0.80   | 0.464±0.72        | 0.009|
| LIV-LSTV                | -0.778±0.73   | -0.304±0.79       | 0.024|

*PT, proximal thoracic; MT, main thoracic; TK, thoracic kyphosis; LL, lumbar lordosis; FRA, first rib angle; CA, clavicle angle; TA, trapezial angle; SHD, shoulder height difference; C7PL-CSVL, C7 plumb line-center sacral vertical line; LIV, lowest instrumented vertebra; LTV, last touched vertebra; LSTV, last substantially touched vertebra.

At follow up, the T1 tilt, FRA, and SHD was significantly lower in adding on group (Table 4).
Table 4. Comparison of parameters between patients with and without adding on at last follow up

*PT, proximal thoracic; MT, main thoracic; TK, thoracic kyphosis; LL, lumbar lordosis; FRA, first rib angle; CA, clavicle angle; TA, trapezial angle; SHD, shoulder height difference; C7PL-CSVL, C7 plumb line-center sacral vertical line

Binary logistic regression analysis revealed that pre-operative main thoracic curve magnitude, preoperative lumbar lordosis, and the gap between lower instrumented vertebra and last touched vertebra were independent risk factors associated with adding on (Table 5).

Table 5. independent risk factors associated with adding on

*Pre-PT, preoperative proximal thoracic; Pre-MT, Preoperative main thoracic; Pre-LL, Preoperative lumbar lordosis; Post-SHD, postoperative shoulder height difference; LIV,
lowest instrumented vertebra; LTV, last touched vertebra; LSTV, last substantially touched vertebra

Representative case was shown in Figure 2 and 3. Two patients received revision surgery by extending fusion segments. No implant failure or other serious complications were found in these patients.

Discussion

Distal adding on is a common phenomenon in the correction of Lenke 1 and 2 type adolescent idiopathic scoliosis. The incidence of distal adding on was reported between 2-51%. The exact mechanism is unclear. According to literature, Lenke 1AR curves were 2.2 times more likely to experience adding-on than 1AL curves. The treatment of Lenke 2 type scoliosis was more complicated than Lenke 1 type because the proximal thoracic curve may cause shoulder imbalance. Shoulder imbalance may cause adding on. Therefore, Lenke 2AR type scoliosis may be at highest risk of adding on and little literature has specifically focus on this topic. Moreover, as far as we know, the clinical impact of adding on was seldom reported.

Shoulder imbalance can be an obvious manifestation in Lenke 2 type AIS both preoperatively and postoperatively. Smyrnis et al reported shoulder asymmetry of 2 cm or greater postoperatively was a potential cause of dissatisfaction. It is challenging for Lenke 2 AIS for the correction of major right thoracic curve could worsen the shoulder imbalance. Whether distal adding on was a compensation for the shoulder imbalance or it was a newly developed distal curve? Cao et al investigate the relationship between postoperative shoulder balance and adding on in Lenke type 2 adolescent idiopathic scoliosis and found that the occurrence of adding on was significantly lower in the shoulder imbalance group at follow-up. Qin followed a total of 104 patients with a minimum of 2 years after selective posterior thoracic instrumentation and found that the radiographical shoulder height (RSH) was significantly smaller in patients with adding on at last follow up. Our conclusion was similar with that. Postoperative shoulder height difference was significantly higher at first erect, after an average of 3 years of follow up, the shoulder height difference was significantly lower in adding on group. The left shoulder was often elevated by the proximal thoracic curve and over-correction of the more flexible main thoracic curve may even worsen the situation. If this happened, the distal adding on tend to develop in lumbar spine to compensate for the shoulder imbalance. However, logistic regression analysis failed to recognize SHD as an independent factor, this could be explained that the compensation mechanism took place only with the presence of short fusion. If the fusion segments were long enough, adding on would not develop even with shoulder imbalance.

Several study revealed that skeletal maturity was a risk factor of developed distal adding on. However, some authors concluded otherwise. Cho et al classified 1A into two different subtype (1AR and 1AL )based on L4 vertebral tilt. They concluded that Lenke 1AR curves were 2.2 times more likely to experience adding-on than 1AL curves. In 1AL curves, younger and less skeletally mature patients based on Risser grading were more likely to experience adding-on. In 1AR curves, selection of LIV instead of age
and skeletal maturity was identified as risk factor. In our series, although the age and Risser sign were significantly lower in adding on group at the initial surgery, they were not recognized as independent risk factor. We supposed that young cases tended to have high flexibility and surgeons preferred shorter fusion segments in order to preserve more mobile segments in young adolescents.

The selection of LIV was considered as an important determinant in preventing distal adding on. For Lenke 1A type AIS, Wang conduct a study and recommend choosing DV (the first vertebra in cephalic direction from sacrum with deviation from CSVL of more than 10 mm) as LIV may provide the best outcome as it not only prevents adding-on but also conserves more lumbar motion. Cho et al recommended fusing distally to 1 level above the neutral vertebra or 1 to 2 levels above the stable vertebra in 1A-R. Matsumoto et al reported that the residual apical translation of the main thoracic curve and the lowest instrumented vertebra more cranial to the last touching vertebra were significantly associated with adding-on in Logistic regression analysis, which was quite similar with ours. For Lenke 2AR, the LIV should not be more cranial to the last touching vertebra to avoid the risk of distal adding on. In a very recent published study, Qin concluded 2AR had a higher incidence of adding on compared with 2AL (26.1% vs. 21.8%). They suggested choosing LIV at LSTV in 2AR and LSTV+1 in 2AL to reduce the risk of adding on. However, one must balance the risk of adding on and mobile segments preservation. LSTV+1 may be too long for adolescent patients.

Our study revealed smaller preoperative main thoracic curve and preoperative lumbar lordosis were risk factors for adding on. A meta-analysis has drawn similar result. Small major thoracic curve often means high flexibility, afterwards, shorter fusion segments may be performed. Moreover, over correction of the major curve in flexible Lenke 2 cases may worsen the shoulder imbalance thus further increase the risk of adding on. Smaller lumbar lordosis was also recognized as independent risk factor of adding on, this may be attributed to the bio-mechanical characteristics of the spine. The rotation axis of the spine lies in front of the thoracic spine and behind the lumbar spine. The rotation force of the spine can be more easily transmitted into the lumbar spine if the lumbar lordosis was small and thus adding on phenomenon developed.

Few reports have been published about the clinical outcome about adding on. In our series of cases, the scores were lower in vitality and mental health dimensions in SF-36. Self-image, mental health and satisfaction were also lower in adding on group by SRS-22 questionnaires. However, lower scores may be result of treating physician making patients aware of the imbalance, rather than truly patients themselves noticing the difference. Although adding on is quite common, few cases undergo revision surgeries. In our series of cases, two cases of adding on insisted on revision surgery by extending fusion segments. More cases and longer follow up are needed to evaluate the influences of residual lumbar curve in patients with distal adding on both physically and psychologically.

The strengths of the study include a relatively large cohort of homogeneous adolescent idiopathic scoliosis patients and strict inclusion criteria. Multiple clinical and radiographic parameters were
evaluated. The drawback of the study was that the cosmetic parameters were not measured as we didn’t take photograph routinely. Lastly, our study was limited by its retrospective nature.

**Conclusion**

In Lenke 2AR AIS cases, the selection of lowest instrumented vertebrae (LIV) higher than LTV is closely correlated with the development of adding-on. Patients with minor main thoracic curve and lumbar lordosis are more likely to develop distal adding on. The shoulder height difference was larger in adding on group just after operation and smaller at follow up. Adding on may cause discontent of patients.

**Declarations**

**Ethics approval and consent to participate:** yes

**Consent for publication:** yes

**Availability of data and material:** yes

**Competing interests:** NA

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**Authors’ contributions:** DYL performed measurements and manuscript preparation; TN performed measurements and manuscript preparation; WSR performed statistical analysis and manuscript preparation; ZJG performed study design and follow up of cases; ZH performed study design and follow up of cases

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**List Of Abbreviations**

AIS adolescent idiopathic scoliosis

AP anterior-posterior

C7P-CSVL C7 plumb line and center sacral vertical line

CA clavicle angle

FRA first rib angle

LIV lowest instrumented vertebra

LSTV last substantially touching vertebra
LTV last touching vertebra
MEP motor-evoked potentials
MT main thoracic
PT proximal thoracic
RSH radiographical shoulder height
SB side-bending
SHD shoulder height difference
SV stable vertebra
TA trapezial angle
UIV upper instrumented vertebra

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Figures
The measurement methods of shoulder balance parameters were shown in figure 1. T1 tilt, First rib angle, clavicle angle and trapezial angle were the angle between the line and the horizontal line. The stable vertebra (SV), last touching vertebra (LTV) and last substantially touching vertebra (LSTV) were showed in figure 1. Lenke 2AR means a double thoracic curve (PT and MT) with a lumbar modifier A and the direction of superior end plate of L4 tilts to the right.

Representative case of Lenke 2AR AIS without adding on. This was a 10 year-old girl with a preoperative PT and MT 37 degree and 63 degree. The preoperative TK and LL were 25 degree and 62 degree. (panel A,B) After posterior correction and instrumentation from T2-L2 (LIV was LTV also the LSTV), the patient got satisfying correction (PT 4 degree, MT 3 degree) and shoulder balance. (panel C,D) At 7 years of follow up, the PT and MT were 5 degree and 6 degree and good balance was maintained. (panel E,F)
Figure 3

Representative case of Lenke 2AR AIS with adding on. This was a 11 year-old girl with a preoperative PT and MT 40 degree and 38 degree. The preoperative TK and LL were 15 degree and 30 degree. (panel A,B) After posterior correction and instrumentation from T1-L1 (LIV was LTV-1), the patient got satisfying correction (PT 0 degree, MT 5 degree) and a trunk shift to the right. (panel C,D) At 5.6 years of follow up, the PT and MT were 0 degree and 8 degree and distal adding on developed. The shoulder height difference was 1.6cm before, 2.5cm postoperatively and 0 at last follow up. (panel E,F)