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Pre-operative halo-gravity traction in severe neurofibromatosis type 1 and congenital scoliosis with thoracic rotatory subluxation

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ABSTRACT

Objectives: To evaluate the efficacy and safety of pre-operative Halo-gravity traction in the treatment of severe neurofibromatosis type 1 and congenital scoliosis patients with thoracic rotatory subluxation.

Patients and methods: Patients with neurofibromatosis type 1 and congenital scoliosis undergoing Halo-gravity traction were reviewed. Radiographic parameters were measured at pre-, post-traction and post-operation. The forced vital capacity and forced expiratory volume in 1 s were recorded at pre- and post-traction. The neurologic function were assessed according to the Frankel score. The complications during Halo-gravity traction, operation and post-operative follow-up were recorded.

Results: A total of 35 patients (21M and 14F) with rotatory subluxation including 18 neurofibromatosis type 1 and 17 congenital scoliosis patients were included, of whom the average age was 14.9 ± 4.8 years. The average duration of Halo-gravity traction was 72.3 ± 11.2 days, during which the average Cobb angle improved from 105.4 ± 34.2° to 81.7 ± 32.6° ($P < 0.001$), and the global kyphosis decreased from 79.2 ± 22.5° to 59.7 ± 23.0° ($P = 0.003$). At pre-traction, the values of coronal and sagittal rotatory subluxation were 9.3 ± 5.2 mm and 7.5 ± 3.5 mm, which significantly improved to 6.7 ± 3.6 mm ($P < 0.001$) and 4.9 ± 2.3 mm ($P < 0.001$), respectively. The average improvement in forced vital capacity and forced expiratory volume in 1 s were from 43.6% to 54.2% predicted and from 40.4% to 48.8% predicted, respectively. After Halo-gravity traction, the Frankel scores improved from C to D in 3 patients, from D to E in 2 patients.

Conclusion: Halo-gravity traction can improve the coronal and sagittal curvature, and the rotatory subluxation in neurofibromatosis type 1 and congenital scoliosis patients. The pre-operative Halo-gravity traction is a safe option for severe neurofibromatosis type 1 and congenital scoliosis patients with rotatory subluxation.

1. Introduction

Surgery has been widely recognized as the only effective strategy for patients with severe scoliosis or kyphoscoliosis, of whom the neurofibromatosis type 1 (NF1) [1] and congenital scoliosis (CS) [2] are important etiologies. The NF1 patients with kyphoscoliosis are usually accompanied by multiple abnormalities including vertebral rotatory subluxation (RS), vertebral scalloping, dysplastic pedicles and spinal tumors. [3,4] The CS patients are often characterized with RS, thoracic insufficiency syndrome, neurologic deterioration and cardiopulmonary dysfunction [5]. These concomitant abnormalities in NF1 and CS truly led to plenty of challenging for the correction surgery.

Recently, RS has been proven to be an independent risk factor for severe 3-dimentional deformities. [6] For NF1 and CS patients with severe kyphoscoliosis, RS seems to be rather common. Hu et al [7] investigated the morphological difference in the vertebrae of scoliosis secondary to NF1, reporting that the incidences of RS were 35.0% in patients without paraspinal neurofibromas and 66.7% in those with paraspinal neurofibromas. Similarly, RS was often observed in CS patients though related studies was rarely seen in the literatures. The occurrence of RS in these cohorts brought more surgical challenges due to the relatively rigid and severe deformity and the high risks of neurological complications.

In order to minimize the risks of correction surgery in severe NF1 and CS patients with RS, the pre-operative Halo-gravity traction (HGT) was usually applied in our center. The HGT could achieve an increase in curve flexibility, a reduction in neurologic risks through gradual traction on a chronically tethered cord and an improvement in pre-operative pulmonary function. [8,9] However, little was known with respect to the efficacy of pre-operative HGT in severe NF1 and CS with RS. Therefore, the purposes of the current study were: (1) to investigate the radiographic and clinical outcomes after HGT in severe NF1 and CS

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patients with RS; and (2) to evaluate the efficiency and safety of HGT in these cohorts.

2. Materials and methods

2.1. Subjects

Ethical approval of this study was obtained from the Clinical Research Ethics Committee of our university hospital. Those NF1 and CS patients treated surgically between June 2009 and September 2016 were reviewed. The inclusion criteria were as follows: (1) patients suffered from severe scoliosis or kyphoscoliosis (Cobb angle in coronal or sagittal plane more than 90°) with RS in thoracic spine; (2) undergoing pre-operative HGT followed by one-stage posterior spinal instrumentation and fusion; (3) HGT duration more than 2 months; (4) with a minimum of 2 years post-operative follow-up; (5) with integrated radiographic data at pre-traction, post-traction, post-operation and the last follow-up. The following exclusion criteria were applied: (1) patients undergoing 3-column osteotomy at RS level; and (2) with any previous history of spinal surgery.

2.2. HGT strategy

Generally, HGT was recommended to those patients with severe and rigid deformities, progressive neurologic deficit due to curvature, necessary of pre-operative adaptation of the cord to corrective stress, presence of intraspinal anomalies might cause acute tension during surgery and poor cardiopulmonary function. The protocols of HGT were in accordance with previous studies. [10,11] The individualized halo ring with 4–6 pins was applied with local anesthesia for all patients in the current study. Pins were tightened appropriately based on the skull size and bone density. The traction weight was initially 3–4 kg, which was added by 2 kg per day. The target weight was 30–50% of the body weight depending on patients' tolerance and should be no more than 15 kg. The traction should be performed more than 12 h each day and the weight was decreased by half at night. The pulmonary training and daily exercises were highly recommended to maximumly reduce the effects of deconditioning. Patients received daily neurologic checks in case of any HGT-related complications. If complications such as upper or lower extremity numbness occurred, the traction weight should be decreased immediately. The duration of HGT was mainly decided by the improvement of coronal and sagittal curves, pulmonary function, and neurologic function. The cease of HGT would be recommended for those with improved or plateaued pulmonary function, curvature, and neurologic deficit. Fig. 1

2.3. Radiographic measurements

On standing whole spinal x-rays, the coronal Cobb angle and sagittal global kyphosis (GK) were measured, respectively. The C method [12] was used for the RS measurement, which was the distance between 2 diagonal lines within the cephalad and caudal vertebral bodies from each superior corner to the contralateral inferior corner. The RS measured on coronal plane (CRS) and on sagittal plane (SRS) were recorded accordingly. The axial rotation (AR) of the RS level was measured on spinal axial CT scans, which was defined as the angle between 2 bisector lines of the cephalad and caudal vertebrae of RS level. The measurements of the parameters were illustrated in Fig. 2.

2.4. Evaluation of pulmonary function

At pre- and post-traction, the forced vital capacity (FVC) and forced expiratory volume in 1 s (FEV1) were recorded, respectively.

2.5. Clinical assessment

The neurologic function at pre-traction, post-traction and post-operation were assessed and recorded according to the Frankel score. The complications during HGT, operation and post-operative follow-up were recorded for each patient.

2.6. Statistical analysis

SPSS software version 17.0 (SPSS, Inc.) was applied for the statistical analysis. Data were shown as Mean ± Standard Deviation (SD). The comparison between pre-traction, post-traction and post-operation was performed using paired samples *t*-test Statistical significance was set at P value < 0.05.

3. Results

3.1. Demographics

A total of 35 patients (21 males and 14 females) ere included in the study, of whom the average age was 14.9 ± 4.8 years. There were 18 NF1 patients (51.4%) and 17 CS patients (48.6%), respectively.

3.2. Comparison between pre- and post-traction

The comparison analysis between pre-traction and post-traction were shown in Table 1. The average duration of HGT was 72.3 ± 11.2 days, during which the average Cobb angle improved from 105.4 ± 34.2° to 81.7 ± 32.6° (P < 0.001), and GK decreased from 79.2 ± 22.5° to 59.7 ± 23.0° (P = 0.003), respectively. At pre-traction, the CRS and SRS values were 9.3 ± 5.2 mm and 7.5 ± 3.5 mm, which significantly improved to 6.7 ± 3.6 mm (P < 0.001) and 4.9 ± 2.3 mm (P < 0.001), respectively. In addition, the correction rates by RS levels after HGT were summarized in Table 2. A demo case was illustrated in Fig. 3.

3.3. Comparison between pre- and post-operation

At post-operation, the average Cobb angle and GK were 55.8 ± 27.4° and 39.0 ± 18.8°, with the total post-operative correction rates being 31.2 ± 8.5% and 44.6 ± 10.2%. In terms of CRS and SRS, the average post-operative values were 5.1 ± 1.8 mm and 3.3 ± 1.9 mm, respectively. The values of AR were 44.5 ± 11.7° at pre-traction and 29.5 ± 9.4° at post-operation, and significant difference was observed (P < 0.001). Table 1

3.4. Change in pulmonary function after traction

The average FVC values were 0.822 L at pre-traction and 0.976 L at post-traction, and no significant difference was found (P = 0.102). The FEV1 was 0.736 L at pre-traction, which slightly improved to 0.766 L after traction (P = 0.791). The average improvement in FVC and FEV1 were from 43.6% to 54.2% predicted and from 40.4% to 48.8% predicted, respectively.

3.5. Clinical outcomes

A total of 9 patients were found to be with neurologic deficit at pre-traction, including 3 NF1 patients and 6 CS patients. The Frankel scores were C in 5 patients and D in 4 patients. After HGT, the Frankel scores improved from C to D in 3 patients, from D to E in 2 patients. The Frankel scores of 2 patients with C and 2 patients with D were not significantly improved, and no deterioration in neurologic function was observed during HGT. Table 3

Traction pin loosening occurred in 2 patients and superficial infection around pin in 3 patients during traction. No deep venous

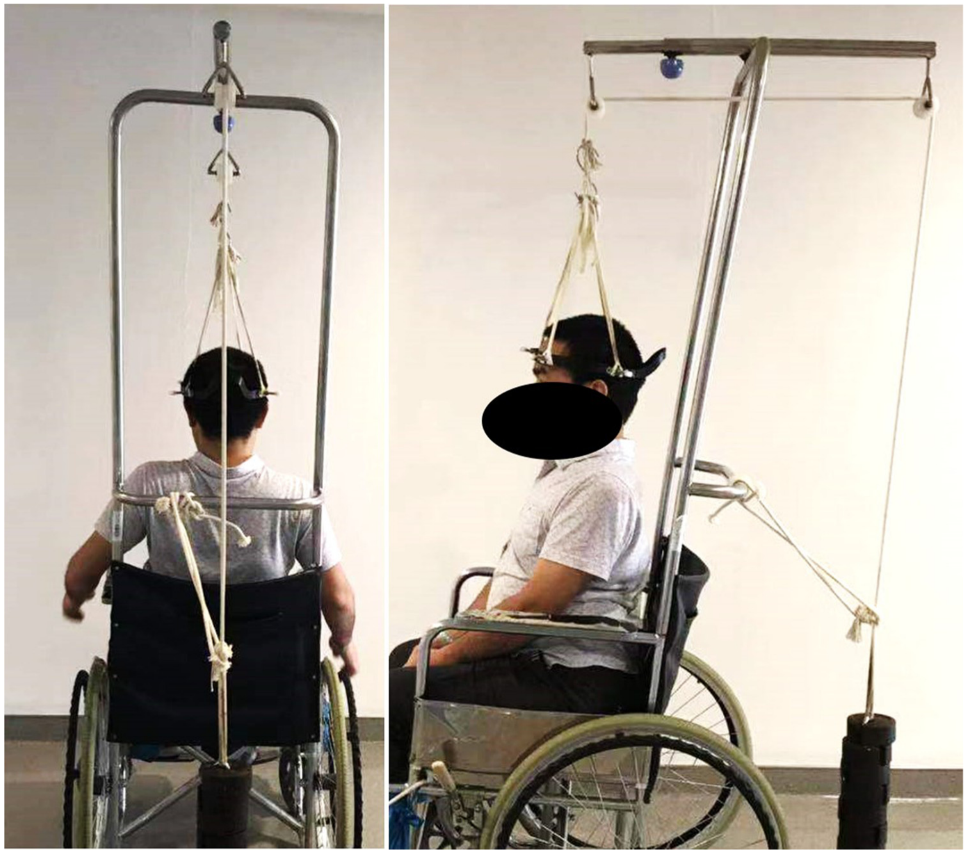

Fig. 1. An illustration of Halo-gravity traction in wheelchair.

thrombosis or brachial plexus injury was found. The intra-operative complications included misplacement of pedicle screws in 4 patients, massive haemorrhage in 1 patient, and transient change in neurologic monitoring in 1 patient. The Frankel scores improved from C to D in 1 patient and from D to E in 1 patient after correction surgery, respectively. At the last follow-up, there was no implant-related complications except for 1 patient suffering from trunk shift. No revision was recommended until the last follow-up.

4. Discussion

The RS was a triaxial deformity mainly consisting of axial rotation and lateral translation toward the convexity of the curve, firstly described by Trammell et al [13] in 1988. In clinical practice, RS was significantly associated with a various of clinical symptoms such as radicular and low back pain, and foraminal stenosis [6,14]. More importantly, RS leaded to the instability of spinal alignments and seemed to be a high risk factor for deterioration in the deformity [15]. For severe patients with RS, surgery seemed to be the only effective strategy in spite of more intra-operative difficulties secondary to RS. In order to maximumly improve the safety of spinal correction surgery, the pre-operative HGT has been widely applied by spinal surgeons. According to previous studies, HGT benefited the pulmonary function, neurologic function, and clinical outcomes in patients with severe spinal deformities. [11,16] The radiographic and clinical efficacies of pre-operative HGT in severe NF1 and CS patients with RS, however, has not been thoroughly investigated.

The improvement in spinal deformity after HGT has been reported

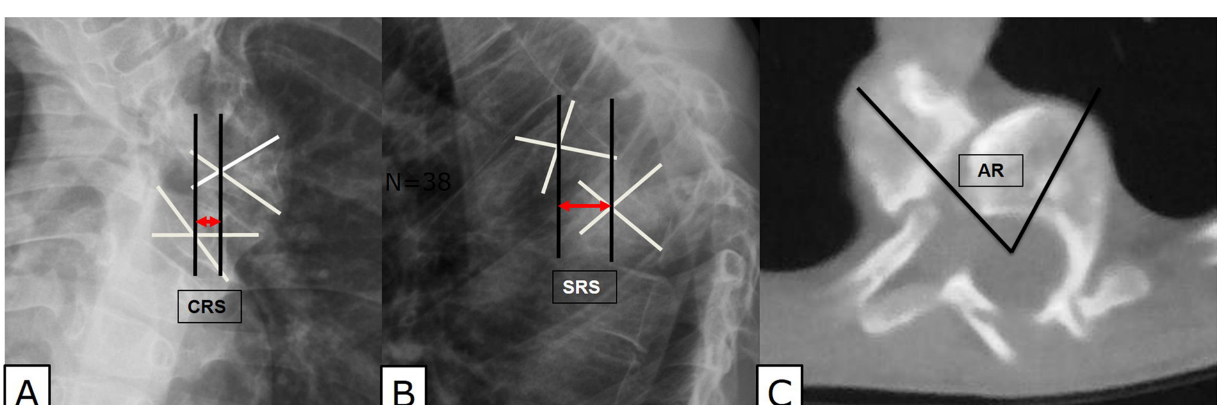

Fig. 2. The rotatory subluxation measured on coronal plane (CRS, Fig. A) and on sagittal plane (SRS, Fig. B) using the C method. The axial rotation (AR, Fig. C) of the rotatory subluxation level was measured on spinal axial CT scans.

Table 1
Comparison between pre-traction, post-traction and post-operation.

	Pre-traction	Post-traction	Post-operation	P (pre- vs post-traction)	P (pre- vs post-op)
Cobb angle (°)	105.4 ± 34.2	81.7 ± 32.6	55.8 ± 27.4	< 0.001	< 0.001
GK (°)	79.2 ± 22.5	59.7 ± 23.0	39.0 ± 18.8	0.003	< 0.001
CRS (mm)	9.3 ± 5.2	6.7 ± 3.6	5.1 ± 1.8	< 0.001	< 0.001
SRS (mm)	7.5 ± 3.5	4.9 ± 2.3	3.3 ± 1.9	< 0.001	< 0.001
AR (°)	44.5 ± 11.7	–	29.5 ± 9.4	–	< 0.001

GK: global kyphosis; CRS: rotatory subluxation on coronal plane; SRS: rotatory subluxation on sagittal plane; AR: axial rotation.

Table 2
Summary of correction rates after HGT by RS levels.

RS Level	Case No.	Cobb angle	GK	CRS	SRS
T2/3	1	9.24%	27.85%	42.84%	70.09%
T3/4	2	37.63%	30.90%	39.40%	43.94%
T4/5	2	65.13%	44.46%	38.19%	68.50%
T5/6	4	12.14%	30.48%	23.87%	44.38%
T6/7	11	13.17%	11.79%	23.89%	30.21%
T7/8	5	13.46%	19.00%	19.11%	17.09%
T8/9	4	23.72%	35.02%	19.32%	31.93%
T9/10	3	29.20%	28.77%	44.84%	35.89%
T11/12	3	43.24%	35.96%	36.10%	34.47%

GK: global kyphosis; CRS: rotatory subluxation on coronal plane; SRS: rotatory subluxation on sagittal plane; AR: axial rotation.

with various degrees in previous studies. Han et al [10] revealed that the mean correction rates after HGT in coronal curve and GK were 35.98% and 33.27% in severe kyphoscoliosis patients with an average duration of 77.9 days. Koller et al [11] reported that the average correction rates of scoliosis and kyphosis after 30 days of HGT were 15.1% and 16.5%. A total of 35 severe NF1 and CS patients with RS were included in the current study. After 72.3 days of HGT, the average correction rate of coronal Cobb angle and sagittal kyphosis were 22.5% and 24.6%, respectively. The results of the current study was lower than that of Han et al [10] but higher than that of Koller et al [11]. We assumed that the relatively short HGT duration should be responsible for the low correction rate of HGT in the study of Koller et al [11]. Therefore, enough HGT duration until the plateau of improvement in pulmonary function, curvature, and neurologic deficit was recommended for this cohort. On the other hand, it should be mentioned that longer duration of traction indicated higher incidences of traction-related complications. We believed that the duration of traction might be decided individually, and minimal duration of traction with satisfactory radiographic outcomes was the initial goal of HGT. In addition, a great deal of variation in the correction rate after HGT was found in our study (Table 2), which should be mainly attributed to the difference in flexibility and severity of the curvature. A higher correction rate after HGT usually indicated a better post-operative realignment of the spine.

Moreover, the change in RS after HGT in severe kyphoscoliosis was not systematically investigated by previous studies. According to the current study, the average CRS decreased from 9.3 mm to 6.7 mm, and the average SRS decreased from 7.5 mm to 4.9 mm. Significant difference was observed. Since severe RS could significantly increase both the difficulty of implanting pedicle screws and the risk of neurologic impairment, our study firstly proved that the pre-operative HGT was of great clinical significance in reducing RS for severe NF1 and CS patients. Additionally, the values of CRS, SRS and AR were found to be significantly improved after the correction surgery. Therefore, with the help of pre-operative HGT, the correction surgery could further improve the RS along with the improvement in curvature.

The changes in pulmonary function after HGT have been investigated in literatures, and the expected inspiring results were revealed. Bogunovic et al [17] showed that HGT improved pulmonary function results in 19 of 22 patients with severe pediatric spinal deformities. After HGT, the overall improvement in pulmonary function was approximately 20% from baseline values with the FVC improved from 45.4% to 53.1% predicted and FEV1 from 43.7% to 52.7% predicted. Koller et al [11] reported that a total of 54% severe patients positively responded to the HGT in pulmonary function and the average FVC% significantly improved from 41.8% to 47.7% after HGT. In the current study, the average improvement in FVC was from 43.6% to 54.2% predicted and the FEV1 was from 40.4% to 48.8% predicted, respectively. Though the difference was not statistically significant due to the relatively more severe deformity of the cohort, we could still presume the potential benefit of pulmonary function from HGT in severe NF1 and CS patients with RS.

According to previous studies, one-stage posterior spinal correction and fusion would be challenging with plenty of intra-operative complications for the severe cases. [18,19] In a Scoli-RISK-1 study evaluating the complications for complex spinal deformity, Kelly et al [19] reported that the rates were 49.3% for all complications and 8.7% for new neurologic deficits. Papadopoulos et al [20] showed that the intra-operative monitoring changes occurred in 22% severe and rigid patients undergoing single posterior approach correction surgery, with 1 patient progressing to complete spinal cord injury. In the current study, only 1 patient was found to have transient change in neurologic monitoring, who recovered at post-operation. A total of 2 patients were identified to have improved Frankel scores (1 case from C to D, and 1 case from D to E). One patient suffered from trunk shift but no revision surgery was recommended during follow-up. Therefore, our results demonstrated that HGT could significantly reduce the difficulty in placement of pedicle screws by improvement in the severity of RS, and further enhanced the safety of correction procedure by gradual traction on a chronically tethered cord.

The limitation of the current study is the relatively small sample size. The measurements of CRS and SRS based on the standing whole spinal x-rays, especially CRS and SRS of upper thoracic spine, are influenced by the image resolution though reliable results has been proved by previous study. [12] Further studies with EOS 3D assessment of RS are highly recommended. In addition, the changes in AR are only analyzed between pre-traction and post-traction because the post-traction CT is not taken due to the ethical consideration.

5. Conclusions

Pre-operative HGT can significantly correct the coronal curve and sagittal kyphosis, improve the neurologic deficit, and correct the rotatory subluxation in NF1 and CS patients. Therefore, the pre-operative HGT can be considered as an optimal and safe option for severe NF1 and CS patients with rotatory subluxation in thoracic spine.

Declaration of competing interest

The manuscript submitted does not contain information about medical device(s) /drug(s). No relevant financial activities occurred outside the submitted work.

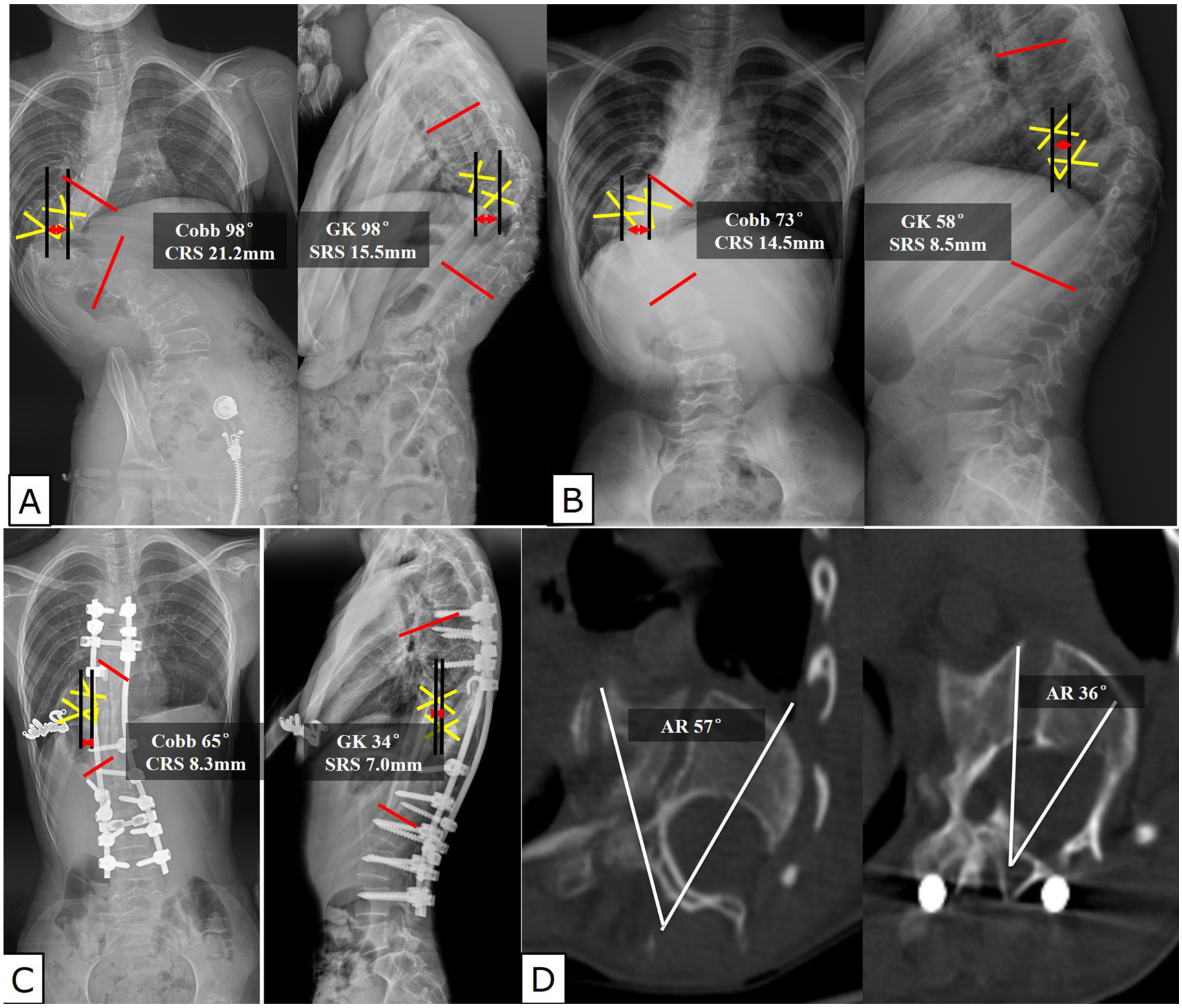

Fig. 3. A 11 years old girl with NF1. The patient showed severe kyphoscoliosis and RS at T9/10 level (A) at initial visit. The curvature and RS showed significant improvement after HGT (B), which further improved after the correction surgery (C). The AR measured on spinal axial CT scan also significantly improved after the correction surgery (D).

Table 3
Change of Frankel scores of 9 patients with neurologic deficit at pre-traction.

No. of patients	Etiology	Pre-traction	Post-traction
3	2 with NF1, 1 with CS	C	D
2	CS	C	C
2	1 with NF1, 1 with CS	D	E
2	CS	D	D

Acknowledgements

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