Hybrid versus Total Sublaminar Wires in Patients With Spinal Muscular Atrophy Undergoing Scoliosis Surgery

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Research Article

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Abstract

Background

Early versions of spinal muscular atrophy (SMA) scoliosis correction surgeries often involved sublaminar devices. Recently the utilization of pedicle screw is gaining much popularity. Pedicle screw generally believed to provide additional deformity correction, but pedicle size and rotational deformity limit the application of pedicle screw in the thoracic spine, resulting in a hybrid construct of the pedicle screw and sublaminar wire. Studies of the efficacy of hybrid instrumentation in SMA scoliosis is often limited by the scarcity of the disease itself. In this study, we aimed to compare the surgical outcome of using hybrid constructs of the pedicle screw and sublaminar wire and that of sublaminar wire alone in patients with SMA scoliosis.

Methods

We retrospectively reviewed the clinical records and radiographic assessments of patients with SMA scoliosis who underwent corrective surgery between 1993 and 2015. The radiographic assessments included the deformity correction and the progressive change of major curve angle, pelvic tilt (PT) and coronal balance (CB). The correction of deformities was observed postoperatively and at the patient's 2-year follow-up to test the efficacy of each type of constructs.

Results

Thirty-three patients were included in this study. There were 14 and 19 patients in the wiring and the hybrid construct groups, respectively. The hybrid construct demonstrated a higher major curve angle correction (50.5° ± 11.2° vs. 36.4° ± 8.4°, p < 0.001), a higher apical vertebral rotation correction (10.6° ± 3.9° vs. 4.8° ± 2.6°, p < 0.001), and reduced the progression of major curve angle after the 2-year follow-up (5.1° ± 2.9° vs. 8.7° ± 4.8°, p < 0.001). A moderate correlation was observed between the magnitude of correction of apical vertebral rotation angle and major curve (r = 0.528, p = 0.002).

Conclusion

This study demonstrated that hybrid instrumentation can provide a greater magnitude of correction in major curve and apical rotation, as well as less major curve progression in comparison with sublaminar wire in patients with SMA scoliosis.

Level of evidence III

Introduction

Spinal muscular atrophy (SMA) is an autosomal recessive neuromuscular disease characterized by a progressive course of muscular weakness and atrophy. Scoliosis is the most common orthopaedic manifestation in patients with SMA and its incidence is positively correlated with the severity of SMA. Individuals with type 1 or type 2 SMA are almost presented with scoliosis and only about 50% of individuals are affected in type 3 SMA.

The deformities involved are often located in both thoracic and lumbar spine with a collapsing "C" shaped curve with associated obliquity of the pelvis. Loss of coronal and sagittal balance makes it difficult to maintain an upright posture and the rapid deterioration of pulmonary function greatly affecting the quality of life for both the patient and the caregiver. Conservative management with bracing is rarely used as a definitive treatment because of its minimal efficacy and potential constrictive effects on the already compromised respiratory system. Surgical treatment is usually indicated in the early stage of life, even before skeletal maturity, to control the progression of the deformity and to preserve the cardiopulmonary function.

Surgical correction of neuromuscular scoliosis was first performed through Harrington's distraction rod and there has been considerable advancement since then. Modern scoliosis surgeries are mostly based on the concept of segmental instrumentation and fusion described by Luque and uses in combination with various pelvic fixation. Early versions of
posterior instrumentation often involved the use of sublaminar devices e.g. wires, bands, and hooks. Recent iterations often involve pedicle screws in place of or in combination with wiring for segmental fixation. There are few comparative studies on the surgical outcome in neuromuscular scoliosis surgery. Reportedly, the full pedicle screw method provides a better correction of the major curve compared with the hybrid method in scoliosis surgery of cerebral palsy patients. Comparable surgical results between sublaminar wire, hybrid and pedicle screw methods in scoliotic surgery of cerebral palsy and Duchenne muscular dystrophy patients had also been reported. However, study on the comparison of deformity correction of SMA using different methods still scarce. In this study, we aimed to retrospectively compare the clinical and radiological outcomes of sublaminar wire and those of hybrid constructs which consist of pedicle screws and sublaminar wire in patients with SMA undergoing surgery for scoliosis.

**Methods**

**Participants**

After Institutional Review Board approval was obtained, the medical records and radiographic assessments of patients with SMA scoliosis underwent surgical correction using either total sublaminar wire or hybrid constructs including sublaminar wire in the high thoracic spine and pedicle screws in the low thoracic and lumbar spine between 1993 and 2015 were retrospectively analyzed by two pediatric orthopaedic surgeons. All patients had their diagnosis of SMA and the associated neuromuscular scoliosis confirmed based on clinical manifestations and radiographic examinations by pediatric neurologists. All the surgeries were performed by the same pediatric orthopaedic surgeon.

**Surgical Procedure**

The surgical indications for SMA scoliosis included pulmonary function deterioration, progressive scoliosis (major curve angle >40°), and difficulty sitting. All the reviewed patients underwent surgery using a segmental spinal construct either with the total sublaminar wire or with the hybrid construct (sublaminar wire and pedicle screw) as described in our previous report. The construct extended from T2 or T3 to the sacrum and the Galveston pelvic fixation technique was used in all cases. In the hybrid construct group, the insertion of pedicle screws was attempted until the pedicle size reached the screw limitation. Cross-link systems were used for augmentation in every case. Pedicle screws of apical vertebrae were inserted if feasible.

**Postoperative course**

Every patient was transferred to the pediatric intensive care unit for intensive care by a pediatric cardio-pulmonologist immediately after the surgery. Patients were observed for 24 hours after extubation and then transferred to the general ward for further postoperative care and rehabilitation. Boston brace was used for at least three months for supporting successful bone fusion. Once bone fusion occurred, especially in the lumbar-pelvic area, bracing was no longer recommended.

**Assessments**

Radiographic parameter including major curve angle, pelvic tilt (PT), and coronal balance (CB) is measured using Cobb’s method and our previously described method on anteroposterior and lateral sitting radiographs of the entire spine. The recorded radiographic parameters from the preoperative, the postoperative, and the 2-year follow-up radiographs were measured and recorded.

To obtain a unified assessment of the apical vertebra rotation across all enrolled patients, the rotation angle of the apex vertebra on the transverse plane was calculated with the method developed by Chi et al. using anterior-posterior X-ray images. The surgical time was defined as the time from skin incision to completion of wound closure. Complications, including stroke, deep vein thrombosis, pulmonary complications, and renal failure, were also recorded.

**Statistical analysis**
Descriptive analysis was performed for each parameter. The Wilcoxon test was used to compare the demographic profile, surgical parameters, and radiological profiles. Pearson correlation coefficient analysis was performed between the amount of correction of each radiological profile and their corresponding correlation coefficient (r). All analyses were performed with the Statistical Package for the Social Sciences (version 19.0, SPSS, Inc, Chicago, IL).

Results

Thirty-three patients enrolled in this study. There were 14 and 19 patients in the sublaminar wire group and the hybrid instrumentation group, respectively. There is no statistical difference between the two groups in terms of demographic data (Table 1) and preoperative radiographic parameters (Table 2). There are 10 (71%) SMA II and 4 (29%) SMA III patients in the sublaminar wire group and 12 (63%) SMA II and 7 (37%) SMA III patients in the hybrid instrumentation group. In hybrid instruments, the sublaminar wires were mostly used in high thoracic vertebra due to the limitation of pedicle size, and the lumbar or thoracolumbar pedicle screws were applied if feasible (Fig. 1).

Table 1
Demographic profiles of patients with SMA receiving instrumentation and fusion.

| Demographic profile          | Sublaminar wire (n = 14) | Hybrid (n = 19) | P value |
|------------------------------|--------------------------|-----------------|---------|
| Male: Female                 | 9:5                      | 10:9            | 0.723   |
| SMA II: III                  | 10:4                     | 12:7            | 0.719   |
| Age (years)                  | 14.5 ± 5.5               | 14.5 ± 9.4      | 0.981   |
| Height (cm)                  | 144.2 ± 11.1             | 151.7 ± 12.4    | 0.101   |
| Weight (kg)                  | 32.5 ± 9.4               | 42.0 ± 14.5     | 0.058   |
| Operation duration (hours)   | 8.3 ± 1.4                | 9.1 ± 1.5       | 0.109   |

All data presented as mean ± standard deviation.
Table 2
Radiographic parameter of patients with SMA receiving instrumentation and fusion.

|                                      | Sublaminar wire (n = 14) | Hybrid (n = 19) | P value |
|--------------------------------------|--------------------------|----------------|---------|
| **Major curve angle**                |                          |                |         |
| Preoperative (°)                     | 58.8 ± 18.6              | 69.8 ± 19.0    | 0.107   |
| Postoperative (°)                    | 22.4 ± 13.4              | 19.3 ± 12.5    | 0.491   |
| Correction (°)                       | 36.4 ± 8.4               | 50.5 ± 11.2    | <0.001* |
| Correction (%)                       | 63.8 ± 11.7              | 74.1 ± 10.5    | 0.013*  |
| 2-year follow-up                     | 31.1 ± 17.3              | 25.9 ± 14.2    | 0.347   |
| Major curve progression (°)          | 8.7 ± 4.8                | 5.1 ± 2.9      | <0.011* |
| **Pelvic tilt (°)**                  |                          |                |         |
| Pre-operative (°)                    | 10.7 ± 6.8               | 15.9 ± 9.6     | 0.092   |
| Postoperative (°)                    | 5.6 ± 3.5                | 6.7 ± 4.4      | 0.432   |
| Correction (°)                       | 5.1 ± 4.3                | 9.2 ± 7.3      | 0.073   |
| Correction (%)                       | 45.2 ± 19.2              | 55.8 ± 20.7    | 0.146   |
| 2-year follow-up                     | 7.0 ± 4.3                | 8.2 ± 5.1      | 0.458   |
| Pelvic tilt progression (°)          | 1.4 ± 1.2                | 1.5 ± 1.2      | 0.833   |
| **Coronal balance (cm)**            |                          |                |         |
| Preoperative (cm)                    | 5.4 ± 2.8                | 5.2 ± 2.2      | 0.897   |
| Postoperative (cm)                   | 2.6 ± 1.8                | 2.2 ± 0.9      | 0.474   |
| Correction (cm)                      | 2.8 ± 1.9                | 3.0 ± 1.7      | 0.674   |
| Correction (%)                       | 50.8 ± 21.6              | 53.0 ± 17.8    | 0.756   |
| 2-year follow-up                     | 3.1 ± 2.0                | 2.9 ± 1.1      | 0.657   |
| Coronal balance progression (cm)     | 0.5 ± 0.3                | 0.7 ± 0.4      | 0.355   |

All data presented as mean ± standard deviation.

The radiographic results are shown in Table 2. The correction of the major curve is significantly better in the hybrid instrumentation group in comparison with the sublaminar wire group (50.5° ± 11.2° vs. 36.4° ± 8.4°, p < 0.001). The correction magnitude of PT is higher for the hybrid construct but not statistically significant (9.2° ± 7.3° vs. 5.1° ± 4.3°, p = 0.073). There is no significant difference in CB correction between the two groups (p = 0.647). The hybrid instrumentations not only provided a better correction to the major curve immediately after surgery but also have a lower rate of scoliosis progression at the 2-year follow-up (5.1° ± 2.9° vs. 8.7° ± 4.8°, p < 0.001). The progression in the PT and the CB were similar in both groups at the 2-year follow-up (p = 0.291 and P = 0.973, respectively).

The most common level of the apical vertebra in both groups is the L1 vertebra, followed by the L2 vertebra (Table 3). No significant difference was noted between the amounts of apical vertebral rotation before the index surgery of the two groups. The hybrid instrumentation could provide a significantly greater magnitude of transverse rotation correction than the
sublaminar wire (10.6° ± 3.9° vs. 4.8° ± 2.6°, p < 0.001) (Table 3). Comparing the surgical outcome of patients who received pedicle screw placement in the apical vertebra (9 cases) and those without apical screw placement (10 cases) in patients within the hybrid instrumentation group, there was no significant difference in the magnitude of correction (e.g. major curve, PT, and CB) between subgroups (data not shown).

| Apical vertebra distribution and apical vertebra rotation angle of patients with SMA receiving instrumentation and fusion. |
|-----------------|-----------------|-----------------|
| **Sublaminar wire** (n = 14) | **Hybrid** (n = 19) | **P value** |
| Apical vertebral location: number of cases | L1:8 | T9:1 |
| | L2:6 | T11:1 |
| | | L1:14 |
| | | L2:3 |
| Apical vertebra rotation angle | Preoperative (°) | 18.1 ± 6.6 | 18.8 ± 5.7 | 0.756 |
| | Postoperative (°) | 13.3 ± 5.6 | 8.2 ± 4.1 | 0.005 |
| | Correction (°) | 4.8 ± 2.6 | 10.6 ± 3.9 | < 0.001 |
| | Correction (%) | 28.1 ± 13.1 | 57.6 ± 17.5 | < 0.001 |
| All data presented as mean ± standard deviation.

Correlations between each of the above-mentioned radiological parameters were performed using the Pearson correlation coefficient analysis. A moderate correlation was observed between the magnitude of correction of apical vertebral rotation angle and major curve (r = 0.528, p = 0.002) (Table 4). A very weak level of correlation was found between other radiological parameters. On the other hand, the correlation coefficient suggests little to no correlation between PT and CB.

| Pearson correlation coefficient analysis between magnitudes of correction of radiological parameters. |
|-----------------|-----------------|-----------------|
| **AVRA** | **Major curve** | **Pelvic tilt** | **Coronary balance** |
| AVRA | 1 | |  |
| Major curve | 0.528 | 1 | |
| Pelvic tilt | 0.119 | 0.330 | 1 |
| Coronary balance | 0.113 | 0.193 | -0.005 | 1 |
| AVRA, apical vertebral rotation angle. |

During the follow-up, one patient developed pseudoarthrosis, and the cephalad wire migration was found in three cases. Iliac bone osteolysis was initially noted in most cases, which might be due to the enlarging procedure for the rod insertion or windshield wiper motion of the rod. Solid bone formation was achieved in all cases in their later follow-ups.

**Discussion**
In this study, we demonstrated that the hybrid constructs can provide a better correction power in major curve angle and apical vertebral rotation, as well as a lower rate of major curve progression at the 2-year follow-up over sublaminar wiring. This could be largely related to the 3 column fixation and increased de-rotation ability of pedicle screws in hybrid instrumentation compared with sublaminar wires, which provided mostly coronal plane correction and the very limited ability of length fixation and de-rotation. To the best of our knowledge, this is the largest cohort study solely focused on patients with SMA that demonstrated the advantage of hybrid instrumentation over sublaminar wiring in SMA scoliosis correction. Although there are limited studies of SMA scoliosis correction, comparative studies between the sublaminar wire and hybrid instrumentation on other neuromuscular scoliosis correction also indicated similar results (Table 5).

| Current study | Wimmer et al. | Watanabe et al. | Arun et al. | Mattila et al. | Albert et al. |
|---------------|---------------|----------------|------------|---------------|--------------|
| NO. cases     | 33            | 52             | 44         | 43            | 66           | 29          |
| Major population (n) | SMA (33)  | CP (17)        | N (N)      | DMD (43)      | CP (31)      | CP (17)     |
| Study design  | Sublaminar wire vs. Hybrid instrumentation | Sublaminar wire vs. Hybrid instrumentation | Wire vs. Hook vs. anterior screw vs. pedicle screw | Sublaminar wire vs. Hybrid vs. pedicle screw | Hybrid instrumentation vs. pedicle screw | Hybrid instrumentation vs. pedicle screws$ |
| Average follow up (months) | 24          | 42.9           | 48         | 56.4          | 33.6         | 29          |
| Surgical outcome on neuromuscular scoliosis. | Better curve correction, less blood loss and less loss of major curve correction in hybrid instrumentation group. | Comparable surgical results and satisfactory questionnaires between these two fixation methods (Luque-Galveston/Isola-Asher system). | Pedicle screws have the greatest correction rate, the smallest loss of correction and the greatest amount of correction of the apical vertebral translation in scoliotic curves greater than 100º. | Comparable results during medium to long term follow up in both methods. More operating time and blood loss in sublaminar wire group. | Pedicle screw group has the shorter operating times, the less blood loss and the better correction of the major curve | Sublaminar bands utilized in a hybrid construct can achieve corrections equivalent to all-pedicle screw constructs |

Abbreviation: SMA, spinal muscular atrophy; CP, Cerebral Palsy; N, not specified; DMD, Duchenne Muscular Dystrophy

$\$, sublaminar band and pedicle screw

The value of hybrid instrumentation with sublaminar wiring in the thoracic spine is unique for pediatric scoliosis correction, especially in the Asian population. Total pedicle screw instrumentation and fusion is a mainstay treatment in scoliosis surgery nowadays. However, Asians have been proven to have a smaller pedicle diameter (T4 vertebral, 2.9 ± 1 mm) in comparison with the Caucasian population (T5 vertebral, 4.7 mm), with the mean pedicle size from T3 to T9 all fall below 4 mm.
Besides, the poor nutritional status in combination with the fact that most neuromuscular scoliosis patients received surgical intervention during their early adolescence further limits the size of the pedicle and increased the difficulty of pedicle screw insertion. Moreover, the low bone mineral density of patients with SMA is another potential risk for breaching the cortical wall during pedicle screw placement 18, which may result in early internal fixation failure and nerve injury 19, 20. On the other hand, hybrid instrumentation has been shown to provide correction power comparable to total pedicle screw instrumentation for other flaccid types of neuromuscular scoliosis 9, 13. Similar results have also been reported between other hybrid instrumentation and total pedicle screw instrumentation in patients with adolescent idiopathic scoliosis 21-23. These results suggested that hybrid instruments could be a possible alternative for ethnic groups with a smaller build if the total pedicle instrumentation and fusion are not feasible.

Throughout our study, all our patients received Galveston procedure for pelvic fixation and the magnitude of PT correction and the correction maintained are similar across different groups. Galveston procedure provided superior resistance to flexion forces and can achieve a fusion rate between 88%-95% 24, 25. Its low instrument profile provides an additional benefit of a lower chance of wound dehiscence24. Recently its application has decreased because of technical difficulty and popularity of other spinopelvic fixation technique e.g. iliac screw and S2-alar-iliac screw. Despite decreased popularity, the Galveston procedure was shown to provide greater pullout strength26 and a similar wound complication rate compared to iliac screws 27, thus remain the method of our choice.

Our results showed a moderate correlation between the correction of major curve angle and the apical vertebra derotation in patients who underwent SMA scoliosis correction. Derotation manoeuvers were found to present with greater major curve correction in adolescent idiopathic scoliosis, but the correlation between coronal correction and apical vertebral derotation has not been reported28, 29. Comparable surgical outcomes are noted between patients who received apical pedicle screw and those who did not in our study. Suggest the apical derotation can be achieved with periapical instrumentation and the apical screws are not necessary to the construct as previous studies demonstrated that concave apical screw shown not related to additional correction power 22, 30.

The limitation of this study including a limited sample size due to the scarcity of the disease itself. Also, some of our medical records dated back to more than two decades ago when computed tomography was not included as the routine imaging modality for scoliosis surgery. This limits our ability to realize the rotational deformity of each vertebral body. The modification and evolution of surgical technique occurred over time that increased the heterogeneity to the correction results. Nevertheless, this study provided a good demonstration of major curve correction and de-rotation achieved with hybrid constructs in scoliosis surgery of patients with SMA.

Conclusion

In this study, we demonstrated that hybrid instrumentation can provide a greater magnitude of major curve correction and better correction maintenance in comparison with sublaminar wire in patients with SMA scoliosis. Given the limitation of pedicle diameter and vertebral rotation, hybrid constructs can serve as an alternative surgical intervention for patients with SMA receiving scoliosis corrective surgeries.

Declarations

Ethics approval and consent to participate

This study was approved by the Institutional Review Board (IRB) of Kaohsiung Medical University Hospital (KMUHIRB-E(I)-20190144). Written informed consents were waivered by the Institutional Review Board (IRB) of Kaohsiung Medical University Hospital (KMUHIRB-E(I)-20190144). All methods were carried out in accordance with relevant guidelines and regulations.
Consent for publication

No identifiable images or details on individuals are reported within the manuscript.

Availability of data and materials

The datasets generated and/or analyzed during the current study are not publicly available due to privacy/ethical restrictions but are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no conflict of interest.

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Authors' contributions

The first two authors (Shih-Hsiang Chou, M.D. and Wen-Wei Li, M.D.) contributed equally to this study

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Category 3

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**Figures**

![Figure 1](image-url)

**Figure 1**

Representative preoperative, postoperative and two-year follow-up anterior-posterior radiography of (A) hybrid construct and (B) total sublaminar wire construct.