Hybrid technique versus traditional dual growing rod technique to treat congenital early-onset scoliosis: a comparative study with more than 3 years of follow-up

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OBJECTIVE The authors’ objective was to compare the intermediate outcomes of patients with severe congenital early-onset scoliosis (CEOS) treated with posterior vertebrectomy/hemivertebrectomy with short fusion and dual growing rods (hybrid technique [HT]) and those treated with traditional dual growing rods (TDGRs).

METHODS A retrospective study of patients who underwent the HT and TDGR technique for CEOS was conducted. The inclusion criteria were CEOS (age < 10 years), Risser stage 0, treatment with HT or TDGR, index surgery performed between 2004 and 2017, and minimum follow-up of 3 years. For patients who completed lengthening procedures, the last lengthening procedure was considered the latest follow-up. Demographic, radiographic, clinical, and patient-reported outcomes and revisions were compared between groups.

RESULTS Sixty-one patients with CEOS were included in this study, with 16 treated with HT and 45 with TDGR technique. There were no differences in age at index surgery, duration of treatment, or number of lengthening procedures. The lengthening interval was longer in the HT group. The preoperative mean ± SD main curve was 81.8° ± 17.1° for the HT group and 63.3° ± 16.9° for the TDGR group (p < 0.05). However, main curve correction was better in the HT group, and no differences in residual curve were found between groups. Although the preoperative apex vertebral translation (AVT) of the HT group was greater, the correction of AVT was better in the HT group (p < 0.05). No differences in T1–S1 and T1–12 height were found between groups at the latest follow-up. The growth of T1–S1 height was less in the HT group (p < 0.05), whereas the growth of T1–12 height was similar between groups. Patients in the HT group had a lower risk of mechanical complications but higher risks of dural tears and neurological complications.

CONCLUSIONS HT may provide better correction and apex control ability than TDGR for EOS patients with severe and rigid deformity at the apex level, and it significantly decreased the risk of mechanical complications with little influence on growth of the thoracic spine. HT may be an option for patients with severe CEOS with large asymmetrical growth potential around the apex of the curve.

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KEYWORDS hybrid technique of vertebrectomy/hemivertebrectomy with short fusion and dual growing rods; traditional dual growing rods; clinical outcome; complications; spinal growth; deformity

CONGENITAL early-onset scoliosis (CEOS), a common etiology of early-onset scoliosis (EOS), is one of the most challenging deformities in spine surgery because most patients with these deformities still have great and important growth potential of the lung, thorax, and spine.1 Beyond the correction of the curve, surgeons must consider the growth of these important structures when treating children with CEOS. For example, short fusion after vertebrectomy/hemivertebrectomy can be used to treat patients with short curves and congenital kyphoscoliosis due to a congenital formation failure.2–4 However, the fusion after vertebrectomy/hemivertebrectomy may be too long (more than 5 segments) to correct a long and severe congenital curve, and thus it should be avoided.
to prevent iatrogenic thoracic insufficiency syndrome in patients with long and severe CEOS. Therefore, many growth-friendly techniques have been introduced for the treatment of EOS. Among these techniques, implantation of traditional growing rods (TGRs) is the most commonly used procedure.

The clinical outcomes of TGR have been promising. However, there are still some challenges with the use of TGR, especially for the treatment of severe and rigid CEOS. Large residual deformity, limited apex control, “the crankshaft phenomenon,” and high risks of mechanical complications have been reported with TGR used for the treatment of CEOS. To address these issues and build on their experience with TGR, Wang et al. described a hybrid technique (HT) for apical vertebrectomy/hemivertebrectomy and a dual growing rod technique for severe and rigid CEOS, aiming to improve correction and apical control of the curve and to decrease complications. They found that this HT could provide good correction of severe and rigid congenital kyphoscoliosis with fewer complications while allowing for spine growth. Their preliminary results were supported in subsequent studies.

However, the advantages and weaknesses of HT need to be compared with those of traditional dual growing rods (TDGRs). The purpose of this study was to compare the intermediate outcomes of patients with CEOS treated with HT and those treated with TDGR.

**Methods**

After obtaining institutional review board approval, we conducted this single-center, retrospective study to evaluate the outcomes related to HT used to treat patients with severe and rigid CEOS and to compare these outcomes with those of a control group of patients with CEOS treated with TDGR. All patients received a minimum of 3 years of follow-up, with complete preoperative, postoperative, and follow-up radiographic data.

In the HT group, all patients were treated with posterior vertebrectomy/hemivertebrectomy with short fusion and dual growing rods. The indications for HT were as follows: a long sweeping curve with pronounced asym-
metrical growth potential around the apex (multiple congenital formation failures or segmentation failures); a severe and angular local congenital curve (usually in the cervicothoracic, upper thoracic, lower lumbar, or lumbosacral region) with a long and structural compensatory curve; and a long coronal curve with pronounced local kyphosis. HT could be performed with the 4-rod technique (Fig. 1) or 6-rod technique (Fig. 2) on the basis of the location of vertebrectomy/hemivertebrectomy. During index surgery, vertebrectomy/hemivertebrectomy and short fusion were performed to correct the local deformity and eliminate some asymmetrical growth potential. The osteotomy gap could be closed bone to bone if the resected vertebra was small. However, if the resected vertebra was large, anterior structural reconstruction was mandatory to avoid overshortening of the spinal cord (Fig. 3). Then, TDGR was applied to correct the whole curve.

In the TDGR group, all patients were treated with the dual growing rod technique described by Akbarnia et al.\textsuperscript{13} The first lengthening procedure was performed 6 months after index surgery in both groups. Then, lengthening could be performed every 6–12 months. The interval could be elongated as the number of lengthening operations increased and growth potential decreased. Spinal cord monitoring was used in all index and lengthening surgical procedures in all patients in both groups.

Data for both groups were obtained at the preoperative, perioperative, first postoperative, and latest follow-up visits within the study period (for those patients in both groups whose lengthening procedures was stopped, the last lengthening procedure was considered the latest follow-up visit). Data included demographic information, radiographic findings, complications, and revision procedures. The Scoliosis Research Society 22-Item Questionnaire (SRS-22) was used to evaluate patient-reported
outcomes. All radiographs were evaluated by well-trained attending spine surgeons.

Chi-square analysis or the t-test, as appropriate, was utilized to evaluate demographic information, radiographic findings, clinical data, and patient-reported outcomes. Between-group comparisons were analyzed at different time points. Analyses were performed using SPSS Statistics for Windows version 22.0 (IBM Corp.). The alpha value was set to 0.05.

Results

Demographic Characteristics

A total of 16 patients were included in the HT group and 45 in the TDGR group. The mean ± SD age at index surgery was 7.0 ± 2.7 years in the HT group and 6.9 ± 2.4 years in the TDGR group. The mean number of lengthening procedures was 4.6 ± 2.7 in the HT group and 5.9 ± 2.8 in the TDGR group. The mean follow-up time was 5.1 ± 2.5 years in the HT group and 4.8 ± 2.4 years in the TDGR group (Table 1).

Correction of Deformity

Significant correction of the main curve was obtained immediately after index surgery and was well maintained at the latest follow-up visit in both the HT (67.7%) and TDGR (52.8%) groups. Compared with the TDGR group, the HT group had a significantly larger preoperative main curve and apex vertebral translation (AVT) and had significantly better correction of the main curve and AVT after index surgery (p < 0.05). There were no between-group differences in residual main curve and AVT at both the postindex surgery and latest follow-up time points (Table 2). Preoperative GK in the HT group was signifi-
TABLE 1. Demographic variables

<table>
<thead>
<tr>
<th>Variable</th>
<th>HT Group</th>
<th>TDGR Group</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of patients</td>
<td>16</td>
<td>45</td>
<td>0.923</td>
</tr>
<tr>
<td>Age at index surgery, yrs</td>
<td>7.0 ± 2.7</td>
<td>6.9 ± 2.4</td>
<td>0.923</td>
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<tr>
<td>Male/female sex</td>
<td>4/12</td>
<td>10/35</td>
<td>&gt;0.99</td>
</tr>
<tr>
<td>No. of lengthening procedures</td>
<td>4.6 ± 2.7</td>
<td>5.9 ± 2.8</td>
<td>0.126</td>
</tr>
<tr>
<td>Lengthening interval, mos</td>
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<td>9.7 ± 2.3</td>
<td>0.192</td>
</tr>
<tr>
<td>Follow-up duration, yrs</td>
<td>5.1 ± 2.5</td>
<td>4.8 ± 2.4</td>
<td>0.624</td>
</tr>
</tbody>
</table>

Values are shown as number or mean ± SD unless indicated otherwise.

Gain of Spine Height

Significant increases in T1–12 height and T1–S1 height were obtained immediately after index surgery. Growth of T1–12 height and T1–S1 height was noted during the lengthening procedures. No significant differences were found in T1–12 height and T1–S1 height at either the postindex surgery or latest follow-up time point. In the HT group, 12 patients had T1–12 heights greater than 18 cm versus 27 in the TDGR group. However, the annual growth of T1–S1 height in the HT group (0.8 cm/yr) was less than that of the TDGR group (1.2 cm/yr) (p < 0.05). The space available for lung (SAL) ratio was smaller in the HT group (p < 0.05). No between-group differences in SAL ratio were found at either the postindex surgery or latest follow-up time point (Table 3).

Complications and Patient-Reported Outcomes

Fifteen complications occurred in 9 patients in the HT group, whereas 69 complications occurred in 30 patients in the TDGR group. No significant between-group differences were found in the number of complications.

Although no between-group differences were found in the number of neurological complications, transient delayed postoperative neurological deficits occurred in 1 patient in the HT group. He was a 5-year-old boy and had severe congenital scoliosis (99°) and kyphosis (80°) in the thoracic spine. His neurological function was normal before the surgery (American Spine Injury Association Impairment Scale [ASIA] grade E). He underwent HT, including T5 posterior vertebral column resection (PVCR) with T3–7 fusion, and TDGR implantation from T3 to L2. Spinal cord monitoring showed normal findings during surgery. However, delayed neurological deficits of both lower limbs (ASIA grade D) occurred 18 hours after surgery. CT was done, and no compression of the cord was found. The neurological deficits improved quickly 3 days after the surgical procedures, and the patient was able to stand and walk. No changes on spinal cord monitoring or neurological deficits occurred after the lengthening procedures. No neurological complications or changes on spinal cord monitoring during the index or lengthening surgical procedures were noted for the other patients in the HT and TDGR groups. There were more mechanical complications and unanticipated revision surgical procedures in the TDGR group than in the HT group (p < 0.05) (Table 4).

Eleven patients in the HT group and 31 in the TDGR group completed the SRS-22 questionnaire at the latest follow-up. The patients in the HT group had higher self-image, satisfaction, and total SRS-22 scores (Table 4).

Discussion

TGR is a distraction-based, growth-friendly method that is widely used in patients with EOS and specific indications. The outcomes of TGR have been well demonstrated.5,13–16 Extra spine and thorax growth are accommodated with multiple rod-lengthening procedures, and curve correction can be maintained.

Although the results of TGR for EOS are promising, there are still some challenges with the use of growing rod techniques, including limited curve control around the apex and a high incidence of complications. Severe and

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rigid CEOS has some unique characteristics, including a large curve at a younger age, rigidity of the curve due to formation or segmentation failure, coronal curve with kyphosis, and great coronal/sagittal asymmetrical growth around the apex, which can lead to poor control of the deformity and resultant complications when TGR is applied. Furthermore, although dual growing rods may provide better correction with more spine growth and fewer complications than single rods, they may be impossible to employ due to large AVT and apex rotation in patients with severe CEOS (Fig. 4). HT was introduced to treat patients with severe CEOS, aiming to improve correction and decrease complications. Previous studies have demonstrated that the early clinical results of HT in patients with CEOS are promising. However, all these studies lacked the control group of patients with CEOS treated with growing rods needed to provide context to the HT findings.

The TGR results for CEOS were not as good as those described for EOS. Wang et al. reported the outcomes of TDGR for CEOS, and the mean correction of main scoliosis was 51.3%; however, in their study, 5 of 30 patients underwent HT. Theoretically, apical vertebrectomy/hemivertebrectomy could improve the correction of rigid curves. Seven patients with severe CEOS were treated with HT in the study by Wang et al., and the mean curve improved from 81.4° to 40.1° after index surgery and was 41.0° at the latest follow-up. Promising outcomes were reported in subsequent studies by Sun et al. and Bas et al. In the current study, the patients in the HT group had a significantly larger main curve than the patients in the TDGR group. More correction was obtained in the HT group than in the TDGR group (67.7% vs 52.8%). No differences in residual curve were found immediately after index surgery or at latest follow-up.

Loss of thorax height, narrowing and rigidity of the convex hemithorax as the axial-plane deformity progresses, and stiffness of the concave hemithorax as the ribs grow progressively more compressed are the main causes of lung dysfunction in patients with EOS. The axial deformity, sometimes referred to as a “windswept thorax,” can be only partially controlled with TGR due to the biomechanical limitations of simple distraction with end vertebral anchors and the possibility of the crankshaft phenomenon occurring even with the use of serial distraction (Fig. 4). Several strategies have been introduced as “apical fusion” or “apical control” to address these issues. The use of apical pedicle screw anchors on the convexity for apical control would allow further concave growth and achieve direct derotation and translation of the apex toward the concave hemithorax. Johnston reported that, with the apical control technique, the growing rod technique can achieve good correction of axial-plane defor-
mility and significantly improve lung volume (by 50% or more). However, this technique is only effective in patients with flexible curves.

Some surgeons have reported a technique that uses convex instrumented hemiepiphysiodesis and concave distraction with a single growing rod. Progressive improvement of the main curve during the lengthening procedures was described; however, mechanical complications occurred in most patients. Apical vertebrectomy/hemivertebrectomy can provide mobility for direct derotation and translation of the apex during HT procedures, even in patients with curves caused by formation or segmentation failures, while eliminating the anterior growth potential that contributes to the crankshaft phenomenon. This procedure may help to improve axial correction and convex thorax volume when used in combination with TDGR. Significant improvement in the SAL ratio and AVT of the curve in CEOS patients treated with HT has been described in previous studies. Wang et al. reported that the SAL ratio improved significantly from 0.86 to 0.96 in their series. In the study by Sun et al., AVT improved from 72.8 mm to 19.6 mm. In the current study, although preoperative AVT was larger in the HT group, AVT improvement was greater in the HT group than the TDGR group. No between-group differences in SAL ratio were found postoperatively and at latest follow-up.

A major concern regarding HT is whether osteotomy and fusion have negative effects on spine growth. Thompson et al. found that apical short segmental fusion within growing rods could exert a negative influence on spine growth. Wang et al. and Sun et al. reported that the growth of T1–S1 height in their cohorts of patients was similar to that reported in previous studies of patients treated with TGR. In the current study, no significant between-group differences were found in T1–S1 and T1–12 heights at the latest follow-up visits. T1–12 height, which can reflect thorax height, was more than 18 cm in most patients.

However, the annual growth of T1–S1 height was less in the HT group. The annual growth of T1–12 height was similar between groups. HT may have negative effects on spine growth compared with TDGR, but this association remains to be evaluated in the future with additional patients and long-term follow-up.

Perioperative neurological complications rarely occurred in patients treated with TDGR. Moreover, aggressive vertebrectomy/hemivertebrectomy procedures could increase the risk of neurological deficits. Thus, spinal cord monitoring should be used during index surgical procedures and lengthening procedures in all patients treated with HT. Delayed neurological deficits occurred in both lower limbs of one of our patients who underwent T5 PVCR and dual growing rod implantation for severe congenital kyphosis. The patient was closely observed after spinal cord compression was ruled out, and he totally recovered 7 days after surgery. No spinal cord monitoring changes or neurological deficits occurred during the following lengthening procedures.

As a fusionless technique, TGR is inherently prone to complications. The complication rate of TGR could be more than 50%, as previously reported. Mechanical failures are the most common complications. Recently, Helenius et al. found that severe EOS was associated with larger residual deformity and more complications than moderate EOS in patients treated with TGR. Rigid congenital vertebral deformities at the apex lead to large resid-

![FIG. 4. Images obtained in a 6-year-old girl who presented with severe congenital scoliosis caused by multiple adjacent butterfly vertebrae in the thoracic spine. AVT was large, and the convex thorax was markedly compressed (A). TDGR was used to treat this severe deformity. Correction seemed good after index surgery (B). The deformity deteriorated with 10 lengthening procedures performed over 8 years because of limited apex control with TDGR and the crankshaft phenomenon. The convex thorax was compressed even more severely (C).](image-url)
ual deformities and have enormous asymmetrical growth potential in patients with severe CEOS, thereby contributing to increased risks of complications for TGR in patients with CEOS. HT was introduced to address these issues. During index surgery with HT, apical vertebrectomy/hemivertebrectomy and short segmental fusion could substantially improve correction of the major curve while eliminating asymmetrical growth potential around the apex. Wang et al. found that correction of the main curve was 50.7%, and no complications occurred. In the study by Sun et al., the mean correction of the main curve was 57.2% after index surgery, and 2 of 13 patients developed 2 complications. In the current study, although the numbers of complications were similar between groups, patients in the HT group had fewer mechanical failures and revision surgical procedures. HT may help to decrease mechanical failures but may increase the risks of dural tears and neurological deficits due to high-grade osteotomy.

Another issue is whether patients treated with growing rods can avoid the final fusion surgical procedures, which may lead to additional complications and revisions. Autofusion occurred in most patients (89%) treated with TGR and made the spine stiff and relatively stable, and some surgeons have tried to avoid final fusion surgical procedures for such patients with acceptable spine alignment. They stopped performing lengthening procedures and instead retained the growing rods. However, this is not an option for patients with large residual curve and poor spine alignment due to limited apex control of growing rods. For patients treated with HT, acceptable spine alignment could be achieved and maintained with vertebrectomy/hemivertebrectomy and dual growing rods. Both short fusion after osteotomy and autofusion of the segments involved with the growing rod can stabilize the spine. Therefore, the final fusion surgical procedures and related complications can be avoided in these patients. In the current study, 10 patients in the HT group had already stopped their lengthening procedures, and we did not perform final fusion because autofusion was noted on CT.

There were several limitations to this study. First, this was a retrospective study. We thought that TGR had some limitations and was not the proper choice for CEOS patients who were candidates for HT, and therefore we could not perform a strict case-matched study. Second, as an aggressive procedure, HT was chosen for only selected patients with severe and rigid CEOS, so the patient sample size of the HT group was relatively small and most patients in both groups were still skeletally immature and still receiving treatment. Larger studies of such patients who complete all lengthening procedures will be critical to fully understanding the strengths, weaknesses, and indications of these procedures. However, understanding the intermediate follow-up period offers valuable information for patients who may be considered candidates for HT.

Conclusions
We recommend HT for EOS patients with a severe and rigid curve at the apex level. HT has the potential to provide better correction of the main curve with fewer mechanical failures than TGR, while providing similar gains in spine height. Aggressive procedures such as vertebrectomy/hemivertebrectomy with HT may place patients at risk for neurological complications, which rarely occur in patients who have undergone TGR.

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References


Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Zhang, Wang, Yiwei Zhao. Acquisition of data: Yiwei Zhao. Drafting the article: Wang, Yiwei Zhao. Critically revising the article: all authors. Reviewed submitted version of manuscript: Zhang, Yiwei Zhao, Yang, Lin, Shen, Yu Zhao, Wu, Zhuang, Du. Approved the final version of the manuscript on behalf of all authors: Zhang. Statistical analysis: Wang, Du. Administrative/technical/material support: Zhang. Study supervision: Zhang.

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