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Spine Deformity with Fused Ribs Treated with Proximal Rib- vs. Spine-Based Growing Constructs

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Abstract

Study Design—Retrospective review of prospectively collected data

Objective—To compare the use of spine-based vs. rib-based implants for the treatment of early onset scoliosis (EOS) in the setting of rib fusions.

Summary of Background Data—Treatment for severe early-onset spinal deformity with rib fusions includes growing spine devices with proximal rib or spine anchors. The results of treatment, however, have not been compared between spine-based vs. rib-based proximal anchors.

Methods—169 patients with rib fusions treated with rib-based or spine-based constructs and minimum 2-year follow-up were included. 16 patients were treated with proximal spine-based anchors and 153 with proximal rib-based devices (VEPTRs). 104 of the patients with rib-based fixation underwent thoracoplasty at the index surgery. We evaluated change in T1-T12 and T1-S1 height, coronal Cobb angle, kyphosis and number of lengthening/revision surgeries.

Results—Kyphosis increased a mean of 7 degrees in the rib-based group and decreased a mean of 20 degrees in the spine-based group ($p=0.002$). Major Cobb angle decreased in both groups ($p<0.0001$); however, the spine-based group had greater Cobb angle improvement (24 vs. 11 degrees, $p=0.04$). From implant and lengthening of distraction devices, there was a mean 3.3 cm (22%) increase in T1-T12 height and a mean of 8.0 lengthenings in the rib-based group compared

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to a 5.7 cm increase and 6.3 lengthening surgeries in the spine-based group. Patients with rib-based constructs had a mean of 11 total procedures, whereas spine-based patients had a mean of 8.

Conclusions—Patients underwent a mean of 8 lengthening surgeries prior to final fusion or cessation of lengthening with a modest 2.3 cm increase in T1-T12 height. Compared to proximal rib anchors, proximal spine anchors controlled kyphosis and improved Cobb angle correction for early onset scoliosis with rib fusions.

Keywords

Early onset scoliosis; deformity; rib; VEPTR; spine; growing spine; thoracic; chest wall

Introduction

Early onset scoliosis (EOS) can be associated with severe morbidity due to restricted thoracic volumes and subsequent diminished lung growth¹. A subset of patients with EOS have acquired or congenital rib deformities, further restricting thoracic development and increasing the severity of disease. Campbell *et al.* termed this deformity and the pulmonary sequelae “thoracic insufficiency syndrome” (TIS)². Specific rib-based distraction devices, such as the vertical expandable prosthetic titanium rib (VEPTR), have been designed in order to expand the total chest volumes and correct thoracic deformity as much as possible³. Other surgeons have used hooks according to physician-directed use to provide proximal rib fixation to treat EOS and achieve lung expansion.

Due to its ability to specifically target chest wall growth restriction, rib-based fixation with or without expansion thoracoplasty and rib osteotomies has become a common treatment modality for patients with these deformities. In an animal model of TIS, expansion thoracoplasty has been shown to restore normal architecture to lung parenchyma^{4,5}. The role of proximal rib anchors in patients with scoliosis and fused ribs, however, is still debated, and there is a need for comparison studies on the outcomes of rib-based vs. spine-based constructs to treat patients with EOS and associated rib fusions.

We sought to compare the outcomes achieved in children with early-onset scoliosis with associated rib deformity treated with proximal rib- vs. spine-based growing constructs. Our null hypothesis was that there would be no difference in outcomes based on anchor type. The alternate hypothesis was that there would be improved thoracic height and Cobb angle in patients with congenital fused ribs treated with proximal rib anchors (rib-based growing devices) compared to constructs with proximal spine anchors (spine-based devices).

Materials and Methods

169 patients with early onset scoliosis and associated rib fusions were identified across the member institutions of two multicenter EOS databases. Each patient was treated with either rib-based or spine-based expandable constructs between 1999 and 2014 and had a minimum of 2-year follow-up. 16 patients were treated with proximal spine-based anchors, and 153 with rib-based devices. Proximal spine-based constructs primarily entailed proximal hooks or screws with a 2 level fusion. All patients with rib-based devices had VEPTR devices. 104

patients in the rib-based group had thoracoplasty performed at the time of implant insertion. Of the 153 patients with rib-based devices, 92 patients had a rib-to-rib based distractor as part of the construct. Patients with other types of rib fixation other than VETPR, magnetically controlled growing rods, or with both rib- and spine- based constructs were excluded due to small patient numbers. As early onset scoliosis associated with rib fusions is an uncommon diagnosis and is most commonly treated with proximal rib anchors, there were only 16 patients with 2-year follow-up who had this condition treated with proximal spine anchors.

Study parameters included major coronal and sagittal Cobb angle, T1-T12 and T1-S1 height at initial implantation, change in spinal height during lengthenings, and over the entire course of treatment. Only 41 patients completed final fusion at the time of analysis. Number of lengthening/revision surgeries was also tabulated. Space available for Lung (SAL) was calculated using the information obtained for hemithoracic height. Hemithoracic height was measured from the midpoint of the most cephalad rib (not including the vertebral body) to the center (dome) of the hemidiaphragm on each side. The height of the concave hemithorax was divided by the height of the convex hemithorax and expressed as a percentage⁶. Rib fusion severity was rated from PA and lateral radiographs as: mild – one or two rib fusion, moderate – greater than two rib fusions, or severe – extensive chest wall deformity (Figure 1). If the quality of the radiograph stored in the registry was too poor to evaluate rib fusions, the rating was considered indeterminate (10 cases total).

IRB approval for this study was obtained at each site. This paper constitutes a retrospective review of prospectively collected data from 30 sites. Centralized radiographic measurement was performed by trained personnel for one of the study groups representing 134 patients. Statistical analysis was performed using the Pearson chi-square test for nominal variables and the two-tailed student's t-test for continuous variables. Observed differences that have a less than 5 per cent likelihood of occurring by chance were considered significant.

Results

169 patients (92 females and 77 males) with a mean age of 4.7 years at index surgery met inclusion criteria. In total, 16 patients were treated with spine-based growing devices, while the remaining 153 were treated with rib-based devices. There was no significant difference between the two groups in terms of gender, age at index surgery, time to follow-up, preoperative major Cobb angle, preoperative kyphosis, T1-T12 height or T1-S1 height (Table 1). Of the 169 patients, 165 patients had congenital scoliosis. Four patients, all in the rib-based cohort, had a thoracogenic rib fusion. Two of the 16 (12.5%) patients in the spine-based group had a single rod, while 41% of patients had a single rod in the rib-based group ($p=0.02$).

Mean follow-up between the two groups was not significantly different (6.6 years in the rib-based group vs. 5.9 years in the spine-based group, $p\text{-value} = 0.40$). The group treated with spine-based devices underwent fewer total surgeries (mean, 8.0 vs. 11.3, $p = 0.006$). There was no statistically significant difference detected between the two groups in the number of revision surgeries (1.6 vs. 2.3, $p = 0.18$). Additionally there was no difference in the number

of lengthening surgeries between the two groups (6.3 vs. 8.0, $p = 8.0$). Of the rib-based group there were 30 (19.6%) infections, 30 (19.6%) implant failures, 41 (26.8%) device migrations, and 31 (20.2%) unplanned returns to the OR, compared to 1 (6.3%), 2 (12.5%), 1 (6.3%), 2 (12.5%), respectively, in the spine-based group. Between the two groups, only the difference in device migrations was significant ($p = 0.008$) (Table 3). There was no statistically significant difference in the rate of infection ($p = 0.07$), implant failure ($p = 0.45$), or unplanned return to the OR ($p = 0.41$).

Kyphosis increased in the rib-based group over the study period ($p < 0.0001$), and decreased in the spine-based group ($p < 0.001$). Major Cobb angle decreased in both the spine-based and rib-based group ($p < 0.0001$, $p < 0.0001$), although spine-based patients had lower Cobb at latest follow-up ($p = 0.001$). Changes in T1-T12 and T1-S1 height were measured following index surgery, following the final lengthening procedure, and following final fusion. The spine-based group achieved greater thoracic growth following initial implantation (4.3 vs. 1.1 cm, $p = < 0.0001$), prior to final fusion (5.7 vs. 3.3 cm, $p = 0.04$), and over the total course of treatment (6.0 vs. 3.5 cm, $p = 0.02$). There was no significant difference in total gain in T1-S1 height achieved (8.1 vs. 6.3 cm, $p = 0.18$). Additionally there was no difference in total gain in T1-T12 height achieved during lengthening surgeries (2.1 vs. 2.3 cm, $p = 0.83$). At most recent follow-up, the group treated with spine-based vs. rib-based devices achieved better scoliosis correction as measured by total change in Cobb angle (24° vs. 11° improvement, $p = 0.043$) and improved kyphosis (20° decrease vs. 7° increase in kyphosis, $p = 0.002$) (Table 2).

Discussion

Growing constructs with proximal rib anchors are a targeted treatment modality for patients with congenital or thoracogenic scoliosis with associated rib fusions. Devices with proximal rib anchors preserve spine growth⁷ while correcting coronal plane deformity^{8,9} and have been the preferred treatment for EOS patients with rib fusions. Thus, the majority of patients in this study had proximal rib anchors, and spine-based devices were less common (90.5% vs. 9.5% of the total cohort). The strengths and weaknesses of rib-based vs. spine-based proximal anchors have been studied in some populations, but they have not been directly compared in patients with rib fusions⁹. Although our study had fewer patients with spine anchors, the severity of deformity between the two groups was comparable, and in fact there was more preoperative kyphosis in the proximal spine anchor cohort compared to the rib anchor cohort.

This multicenter study includes a large number of patients with early-onset scoliosis and congenital chest wall abnormalities treated with distraction devices. Previous literature has demonstrated that up to 19% of patients with congenital spinal abnormalities will have corresponding rib or chest-wall abnormalities. Additionally, these rib abnormalities are more likely to occur on the concavity of the thorax, potentially reducing the total space available for lung development¹⁰. As a result, expansion thoracostomy and insertion of growing constructs targeted at rib deformities became an increasingly popular option for addressing these abnormalities and restoring lung volumes.

Our study sought to compare the outcomes of patients with fused ribs treated with rib anchors compared to those treated with traditional spine-based growing constructs, with the hypothesis that rib-based constructs would be more effective at treating chest deformity while offering commensurate treatment of spine deformity. The results of this study demonstrate that spine-based constructs offer better correction of spinal deformity, as measured by correction of Cobb angle and kyphosis, while demonstrating equivalent correction of thoracic and spinal height. However, pulmonary function, quality of life data, thoracic volume, or complete data on space available for the lung data is not available from this cohort.

Previous studies have noted the kyphosing effect of rib-based implants^{10,11,12}. Additionally, while rib- and spine-based distraction rods share many of the same risks and disadvantages, there are some potential complications unique to VEPTR placement, such as brachial plexus injury¹³. On the other hand, proximal anchor failure or migration in rib-based devices hold little risk of spinal cord injury, and avoidance of spine instrumentation may leave the spine more flexible and able to tolerate a greater number of lengthening procedures compared to spine growing rods, which are susceptible to diminishing returns in lengthening^{7,14}.

The role of lengthening surgeries is interesting to consider. A relatively modest amount of T1-T12 growth was achieved through lengthening surgeries - a mean of 2 cm over a mean of 6 growing rod lengthening surgeries and 8 VEPTR lengthening surgeries. Other studies have reported 1.65–2.1cm T1-T12 growth using growing rods^{15,22}, 1.4–3.4 cm growth using magnetically controlled rods^{16,18,19}, and 1.8–3.4 cm T1-T12 growth using rib-based constructs.^{17,20,21} On the other hand, substantial increase in spinal length was obtained over the entire course of treatment (Table 2). Our results suggest that patients treated with growing rods achieved greater improvement to their spine deformity compared to the rib-based cohort, as measured by postoperative kyphosis, and Cobb angle. There was a higher revision rate in the rib-based cohort. This may be due to the higher proportion of patients with a single rod in the VEPTR group compared to the spine anchor group.

Limitations of this study include that this was a retrospective review of prospectively collected data, and patients were not randomized into either treatment modality. A major limitation of our study is that only a limited number of patients were treated with spine-based devices, and there may be undetected differences in patient characteristics between the rib-based and spine-based cohort that appeared statistically insignificant due to small sample size. While statistical analysis shows no significant difference between the patients in each treatment group, it cannot be discounted that treating surgeons may have selected rib-based devices for patients with more significant chest wall abnormalities. Further studies with larger patient numbers in the proximal spine-based group may provide more compelling data in the future. It is unclear what the ideal amount of kyphosis is in the early onset scoliosis population. In typical children, normal kyphosis is reported at 20–40 degrees. In this study, we only measured spine height, not spine length^{23,24}. The benefit of increased kyphosis over 40 degrees (i.e., increased spine length and not height) on pulmonary function and health-related quality of life measures has not been established in the literature; however, it remains possible that total spine length may prove a more important predictor of improved pulmonary function than height alone. In general, our study lacks pulmonary function

testing, validated health-related quality of life outcomes, and other measurements of long-term functional status. Additionally only 3 patients with spine-based growing devices had complete data available for SAL. The data presented for this group is used only to demonstrate that there were no major differences in SAL between the two groups, and should not be interpreted as demonstrating any treatment effect on SAL. Rib-fusions and chest wall deformities are variable and to our knowledge no other classification system exists to grade these deformities on severity. We classified deformity based on the number of rib fusions rather than severity of congenital spine deformity, since these patterns are highly variable. However, preoperative T1-T12 height, age, Cobb angle, kyphosis, and severity of rib fusions were similar between the two groups, so the two cohorts appeared comparable.

In this primarily radiographic study, patients with early onset scoliosis and fused ribs treated with spine-based devices had better correction of kyphosis and scoliosis than those treated with rib-based devices. Patients with rib-based devices underwent more total surgeries than those treated with spine-based anchors. Further studies are necessary to evaluate whether rib-based devices and expansion thoracoplasty provide better health-related quality of life, pulmonary function outcomes, or life expectancy for patients with severe chest wall deformities.

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Key Points

- Patients treated with spine-based devices had better correction of kyphosis and sagittal deformity than those treated with rib-based devices
- Patients with rib-based devices underwent more total surgeries than those treated with spine-based devices

Figure 1.
Rating of rib fusions.

Table 1

Patient Characteristics

	Spine-Based Devices	Rib-Based Devices	P-value
Number	16	153	
Gender (M/F)	5/11	72/81	0.23
Mean Age at Index Surgery	5.9 (1.5–14.9)	4.6 (0.2 – 16.5)	0.21
Preop Mean Major Cobb Angle	61 (25–85)	66 (22–112)	0.32
Mean Preop Kyphosis	53 (15–120)	40 (6–91)	0.13
Mean Preop T1-S1	24.1 (13–32)	23.7 (13–38)	0.80
Mean Preop T1-T12	13.5 (6–20.1)	13.8 (5.9–24)	0.82
# Single Rod	12.5% (2/16)	41.2% (61/148)	0.02
Time to Follow-up	5.9 (1.5–10.1)	6.6 (0.7–15.7)	0.40
Rib Fusion Severity (Mild/Mod/Severe/Indeterminate)	6/6/2/2	58/62/21/12	0.93

Table 2**Treatment Results**

	Spine-Based Devices	Rib-Based Devices	P-value
# Lengthening Surgeries	6.3 (1–14)	8.0 (1–21)	0.11
# All Surgeries	8.0 (2–18)	11.3 (2–30)	0.006
# Revision Surgeries	1.6 (0–7)	2.3 (0–12)	0.18
Initial Implantation Change in T1-T12	2.4 (1.0–5.5)	1.1 (–3.8–6.0)	<0.04
Initial Implantation Change in T1-S1	3.3 (–1.3–7.0)	1.8 (–5.0–10.3)	0.06
Change over Lengthening Procedures T1-T12	2.1 (–6.1–5.7)	2.3 (–4.6–10.0)	0.83
Change over Lengthening Procedures T1-S1	5.3 (0.5–8.8)	4.5 (–6.2–18.2)	0.62
Change T1-T12 excluding Final Fusion	5.7 (–4.5–22.4)	3.3 (–3.3–11.7)	0.04
Change T1-S1 excluding Final Fusion	8.1 (3.7–13.1)	5.9 (–4.1–18.2)	0.13
Final Fusion Change T1-T12	0.4 (–0.7–2.4)	1.5 (–1.6–6.2)	0.25
Final Fusion Change T1-S1	0.6 (–1.3–7)	2.3 (–5–10.3)	0.12
Total Change T1-T12	6.0 (–4.5–22.4)	3.5 (–3.01–11.7)	0.02
Total Change T1-S1	8.1 (3.0–13.1)	6.3 (–4.1–18.2)	0.18
Post-Treatment Cobb	37 (16–62)	58 (11–117)	0.001
Post-Treatment Kyphosis	35 (0–72)	52 (0–108)	0.004
Improvement in Cobb Angle **	24 (–18–66)	11 (–31–88)	0.04
Improvement Kyphosis	20 (–10–62)	6.7 (–63 – 74)	0.002
Final Fusion Change * T1-T12	0.42 (–0.7–2.4)	1.5 (–1.6–6.2)	0.13
Final Fusion Change T1-S1 *	0.58 (–2.2–2.7)	2.3 (–2.7–8.2)	0.10

* Only 41 patients (35 rib-construct and 6 spine-construct) underwent final fusion.

** Improvement in Cobb Angle from pre-op evaluation to final follow-up or final fusion.

Table 3

Breakdown of Complications

	Spine-Based Devices	Rib-Based Devices	P-value
# Infections	1/16 (6.3%)	30/153 (19.6%)	0.07
# Implant Failure	2/16 (12.5%)	30/153 (19.6%)	0.45
# of Device Migrations	1/16 (6.3%)	41/153 (26.8%)	0.008
# Unplanned Return OR	2/16 (12.5%)	31/153 (20.3%)	0.41

Table 4

Space Available for Lungs (SAL)

	Spine-Based Devices	Rib-Based Devices	p-value
Pre-Implant SAL	0.740	0.807	0.11
Post-Index Surgery SAL	0.736	0.829	0.44
Most Recent Follow-up	0.903	0.870	0.17
Total Change SAL	0.121	0.056	0.12