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CASE SERIES

Distraction-based surgeries increase thoracic sagittal spine length after ten lengthening surgeries for patients with idiopathic early-onset scoliosis

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Abstract

Study design Retrospective, comparative, multicenter.

Objectives To determine if the choice of proximal anchor affects thoracic sagittal spine length (SSL) for children with idiopathic early-onset scoliosis (EOS).

Summary Debate exists as to whether spine growth is maintained during treatment for EOS. As rib- (RB) and spine-based (SB) distraction procedures may be kyphogenic, the traditional measurement of spine growth on coronal radiographs may not identify out-of-plane increase in spine length. A measure of SSL, along the spine's sagittal arc of curvature, has been validated to reliably assess the length of the thoracic spine.

Methods Patients with idiopathic EOS treated with distraction-based systems (minimum 5-year follow-up, five lengthening surgeries) with radiographic analysis preoperatively, postimplant (L1), and during lengthening periods (L2–L5, L6–L10) were evaluated with primary outcome of T1–T12 SSL.

Results We identified 34 patients (14 RB, 20 SB) with preoperative age 4.9 years (4.2 RB vs. 5.4 SB), scoliosis 72° (60° RB vs. 77° SB; $p < 0.05$), kyphosis 39° (50° RB vs. 34° SB; $p < 0.05$), and SSL 17.8 cm (15.5 RB vs. 18.5 SB; $p < 0.05$). After initial scoliosis correction from implantation, scoliosis remained constant over time. RB patients had greater kyphosis than SB patients: L1, 46° RB vs. 19° SB ($p < 0.05$); L2–L5, 50° RB vs. 27° SB ($p < 0.05$); L6–L10, 56° RB vs. 26° SB ($p < 0.05$). SSL increased for both groups from preoperative to the tenth lengthening ($p < 0.05$). As compared with RB patients, SB patients had higher SSL preoperatively and maintained this difference to the tenth lengthening ($p < 0.05$). After ten lengthening surgeries, when normalized to preoperative SSL, relative thoracic growth was greater for RB (27%) than for SB patients (19%) ($p < 0.05$).

Conclusion Regardless of proximal anchor choice, thoracic length continued to increase during the distraction phase of treatment for idiopathic EOS.

Level of evidence Level III.

Keywords Spine growth · Early-onset scoliosis · Sagittal spine length (SSL) · Three-dimensional true spine length (3D-TSL)

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Introduction

Early-onset scoliosis is defined as "scoliosis with onset less than the age of 10 years, regardless of etiology" [1]. This is a heterogeneous group of patients that are often challenging to treat. As defined in 2015 by the Scoliosis Research Society's Growing Spine Committee, goals of treatment are to minimize spinal deformity over the life of the patient; maximize thoracic volume and function over the life of the patient; minimize the extent of any final spinal fusion;

maximize motion of chest and spine; to minimize complications, procedures, hospitalizations and burden for the family; and to consider the overall development of the child. Growth-friendly surgical treatments have been developed in an effort to achieve these goals with efforts to preserve chest wall and spine growth while minimizing further spinal deformity [2, 3].

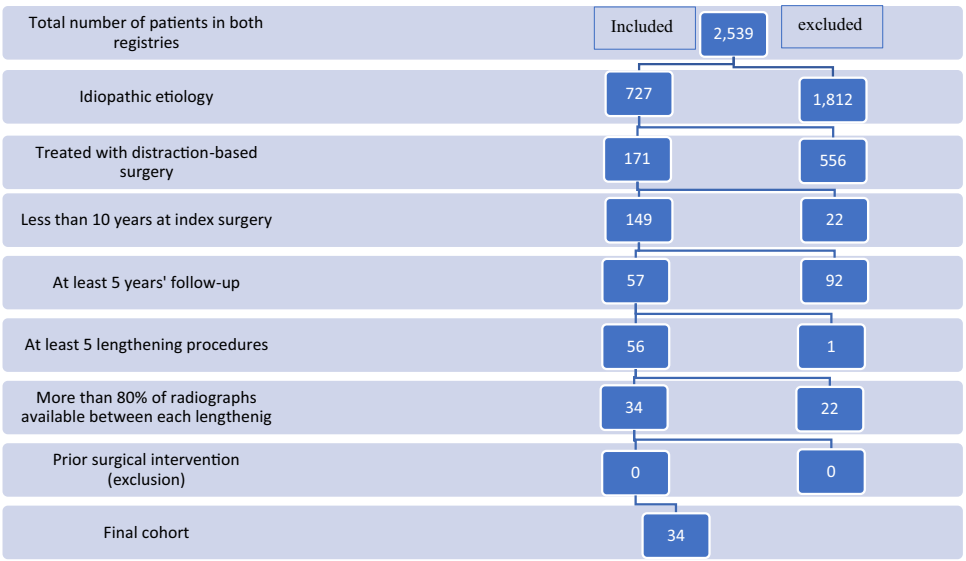
Spine growth has traditionally been measured on the coronal plane with total spine height defined radiographically as the growth per year from the first thoracic vertebrae to the first sacral vertebrae (T1–S1 height). Spinal growth is related to the age of the patient with average rate of T1–S1 growth of approximately 2 cm per year for children younger than 5 years, 0.9 cm per year for children between 5 and 10 years of age, and 1.8 cm per year for children older than 10 years of age until skeletal maturity. Thoracic spine height is defined radiographically as the growth per year from the first thoracic vertebrae to the first lumbar vertebrae (T1–T12 height). Thoracic growth accounts for approximately two-thirds of total spine growth and follows a similar bimodal age distribution [4].

Debate exists as to whether spine growth is maintained during treatment for EOS. As rib-based (RB) and spine-based (SB) distraction procedures may be kyphogenic, the traditional measurement of spine growth on coronal radiographs may not identify out-of-plane increase in spine length. The Halifax method of measuring sagittal spine length (SSL), along the spine's sagittal arc of curvature, has been validated to reliably assess the length of the thoracic spine [5]. Using custom LabView software (National Instruments, Austin, TX), on a lateral radiograph, the center of each superior end plate from T1 to L1 is identified and digitized. The software creates a line of best fit that joins the centers of these endplates following the arc of curvature of the spine, which is called the thoracic SSL. This can similarly be performed from T1 to S1 for total SSL. Recently, this can also be performed using the SA Spine measurement tool on Surgimap software (Nemaris Inc., Globus Medical Inc., Audubon, PA). The custom software can also create a coronal spine length based on a posteroanterior radiograph and can combine the coronal spine length with the SSL to create a three-dimensional true spine length (3D-TSL). Our purpose was to determine if the choice of proximal anchor affects thoracic SSL for children with idiopathic EOS.

Materials and methods

This was a retrospective multicenter review of patients with EOS from two multicenter EOS databases who were treated with SB or RB posterior distraction methods of growth-friendly surgery. Inclusion criteria were idiopathic etiology, diagnosed under the age of 10 years, with a minimum of five lengthening procedures and a minimum of 5-year follow-up. Patients with congenital, neuromuscular, and syndromic etiologies were excluded (Table 1). Radiographs were analyzed at initial implantation and before each subsequent lengthening procedure. Despite this being a multicenter study, the measurements were all performed by a single, unbiased observer at a central location. This was in an effort to decrease interobserver variability of the measurements. Primary outcome was thoracic SSL. Other variables measured were sex, age at each surgical intervention, major curve scoliosis, maximum kyphosis, number of lengthening surgeries, and coronal thoracic spine height (T1–T12). Analysis of variance testing (IBM SPSS Statistics 20; IBM Corp, Armonk, NY) was used to examine the raw data for SSL and for T1–T12 height. Radiographs were obtained and

Table 1. Flow diagram indicating patient selection



analyzed at the following time points: preimplantation surgery, L1 (obtained preoperative first lengthening surgery), L2–L5 (preoperative second lengthening surgery to preoperative fifth lengthening surgery), and L6–L10 (preoperative sixth lengthening surgery to preoperative 10th lengthening surgery). The measurement at time point L1 reflects both the length and height gained from the implantation surgery (biomechanical distraction) plus any growth from the time of implantation to just before the first lengthening surgery. The measurements at lengthening intervals L2–L5 and L6–L10 represent both the length and height gained from each lengthening surgery (biomechanical distraction) plus any growth from the time of each lengthening surgery to just before the subsequent lengthening surgery. These measurements may also include the potential growth stimulation from the effects of mechanical distraction.

Results

We identified 34 patients (14 RB and 20 SB) with a preoperative age of 4.9 years (4.2 years for RB vs. 5.4 years for SB) (Table 1). Preoperative scoliosis for the entire group was 72° (60° RB vs. 77° SB; $p < 0.05$). On average, patients underwent lengthening surgeries every 7.6 months. After initial deformity correction from implantation surgery ($p < 0.05$ from preoperative to L1), scoliosis remained constant during the distraction or growth phase: For all patients, scoliosis was 50° at L1, 48° for L2–L5, and 49° for L6–L10; for RB patients, scoliosis was 54° at L1, 50° for L2–L5, and 52° for L6–L10; and for SB patients, scoliosis was 48° at L1, 46° for L2–L5, and 46° for L6–L10.

Preoperative kyphosis for the entire group was 39° (50° RB vs. 34° SB); postoperatively, kyphosis for the entire group was 31° at L1, 38° for L2–L5, and 41° for L6–L10 ($p < 0.05$). RB patients had greater kyphosis than SB patients at L1, 46° RB vs. 19° SB ($p < 0.05$); L2–L5, 50° RB vs. 27° SB ($p < 0.05$); and L6–L10, 56° RB vs. 26° SB ($p < 0.05$) (Fig. 1).

Preoperative thoracic coronal height for the entire group was 15.3 cm (13.8 cm RB vs. 15.8 cm SB). Postoperatively, thoracic coronal height for the entire group was 17.6 cm at L1, 18.0 cm for L2–L5, and 19.2 cm for L6–L10 ($p < 0.05$). After implantation, L1, there was no difference in thoracic coronal height between RB and SB patients (16.4 cm RB vs. 18.3 cm SB). SB patients had greater coronal thoracic height than RB patients during the L2–L5 (16.1 cm RB vs. 19.3 cm SB; $p < 0.05$) and during the L6–L10 lengthening periods (16.9 cm RB vs. 20.9 cm SB; $p < 0.05$). Coronal thoracic height increased over the lengthening intervals for SB patients ($p < 0.05$) but not for RB patients (Figs. 2, 3, 4).

Preoperative thoracic SSL for the entire group was 17.8 cm (15.5 cm RB vs. 18.5 cm SB; $p < 0.05$). Postoperatively, thoracic SSL for the entire group was at 18.5 cm at

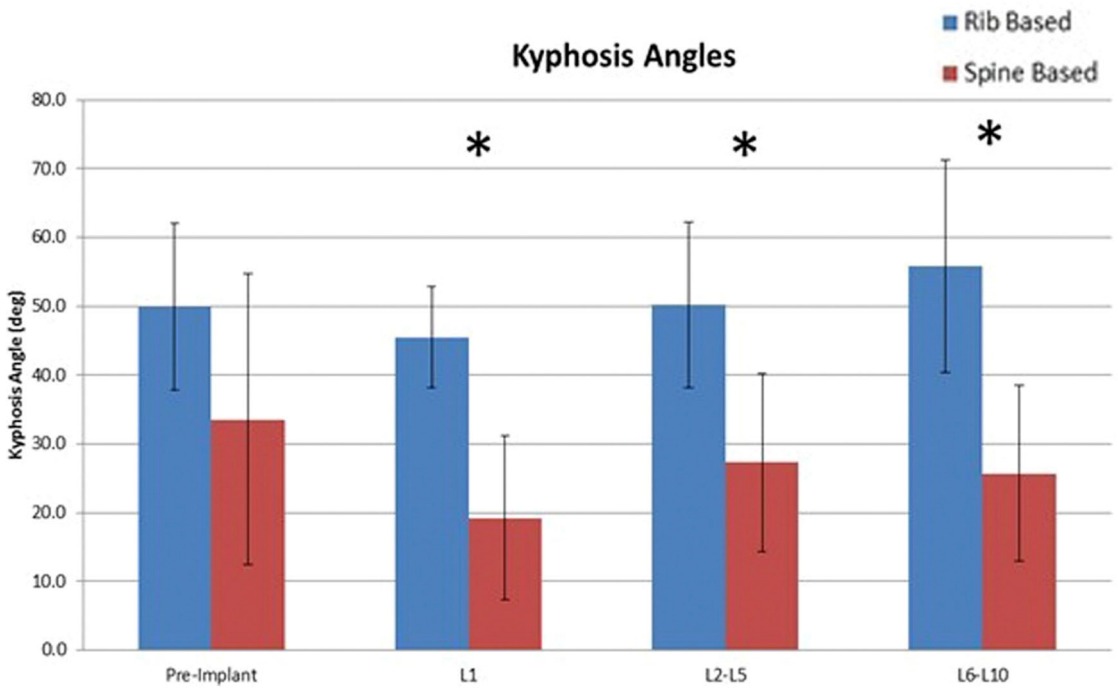


Fig. 1 Preoperative kyphosis was similar between rib-based and spine-based patients; however, postoperatively, rib-based patients had greater kyphosis than spine-based patients (*$p < 0.05$)

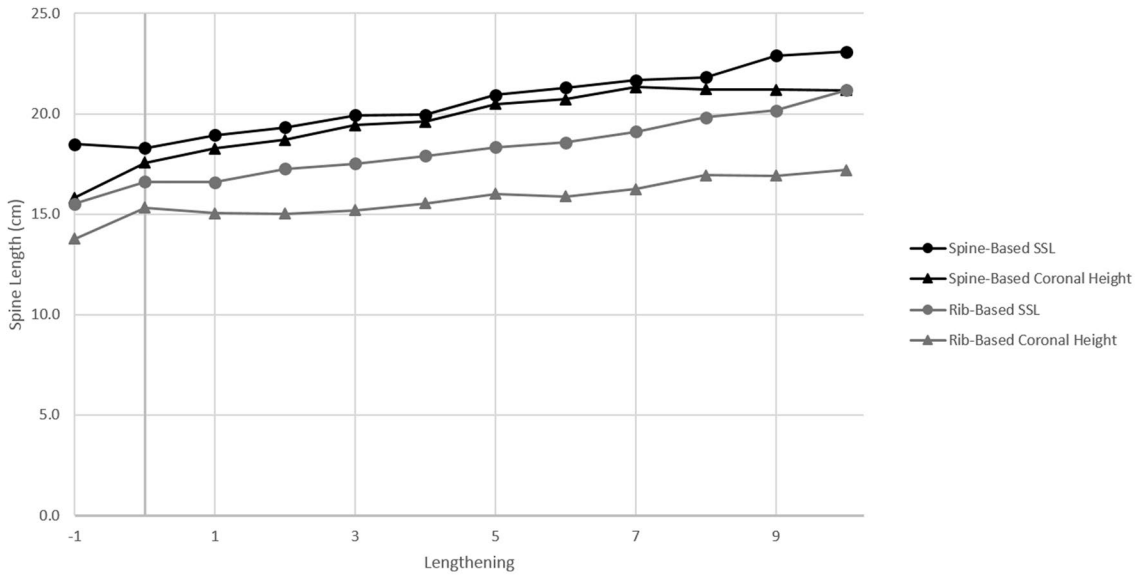


Fig. 2 Coronal vertical T1–T12 height and thoracic SSL for rib-based and spine-based patients versus lengthening

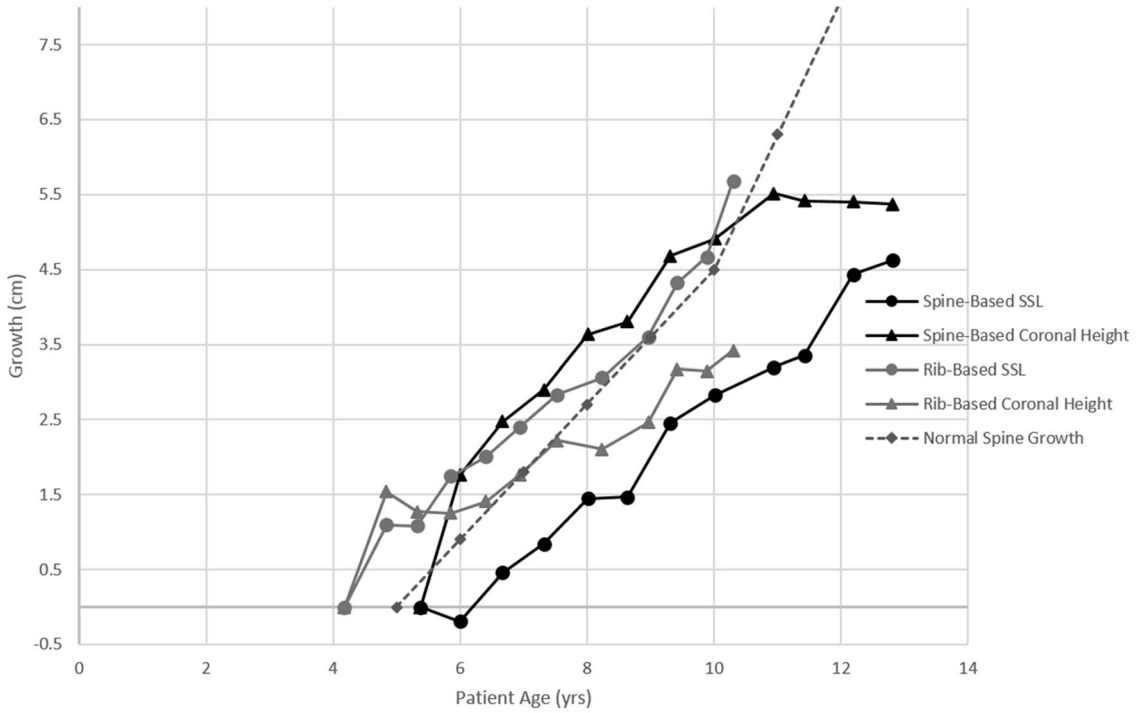


Fig. 3 Coronal vertical T1–T12 growth and thoracic sagittal spine growth for rib-based and spine-based patients versus age. The normal growth rate for children is superimposed [4]

L1, 19.4 cm for L2–L5, and 21.1 cm for L6–L10 ($p < 0.05$). After implantation at L1, there was no difference in thoracic SSL between RB and SB patients (18.4 cm RB vs. 18.7 cm SB). SB patients had greater SSL than RB patients during the L2–L5 (18.5 cm RB vs. 20.0 cm SB; $p < 0.05$) and during the L6–L10 lengthening periods (20.2 cm RB vs. 22.0 cm SB; $p < 0.05$). Thoracic SSL increased over the lengthening intervals for both RB patients and for SB patients ($p < 0.05$). After ten lengthening surgeries, when normalized to preoperative SSL, relative thoracic growth was greater for RB patients than for SB patients (27% RB vs. 19% SB; $p < 0.05$). When evaluating only the growth

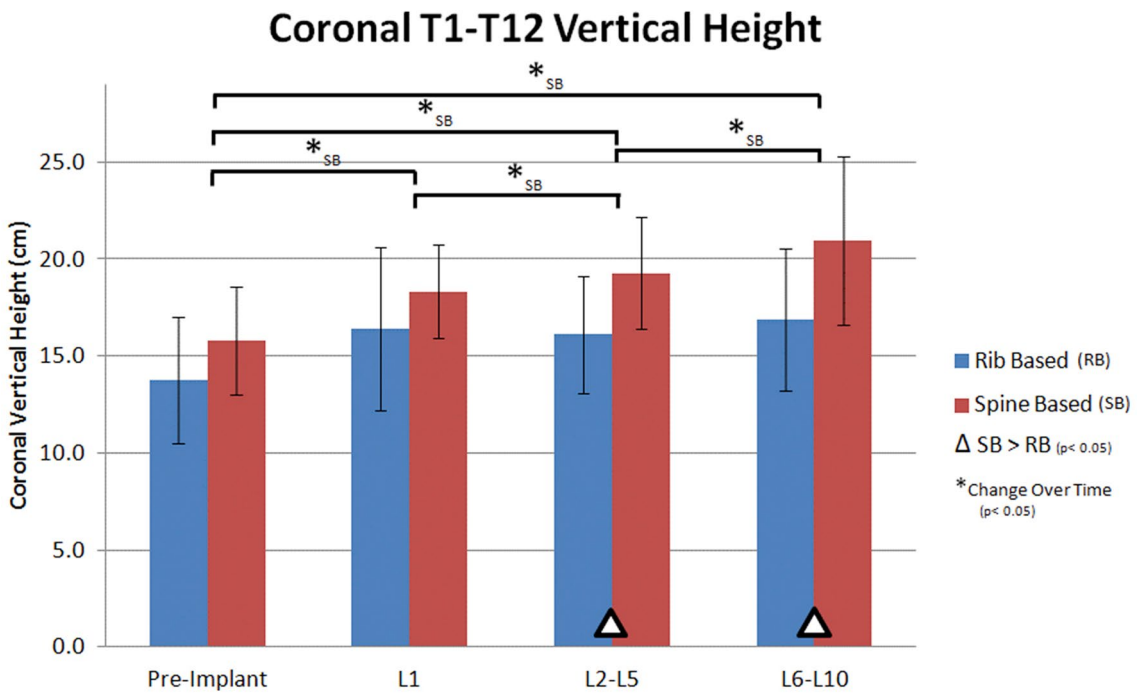


Fig. 4 Coronal vertical T1–T12 heights for rib-based and for spine-based patients over time (*$p < 0.05$)

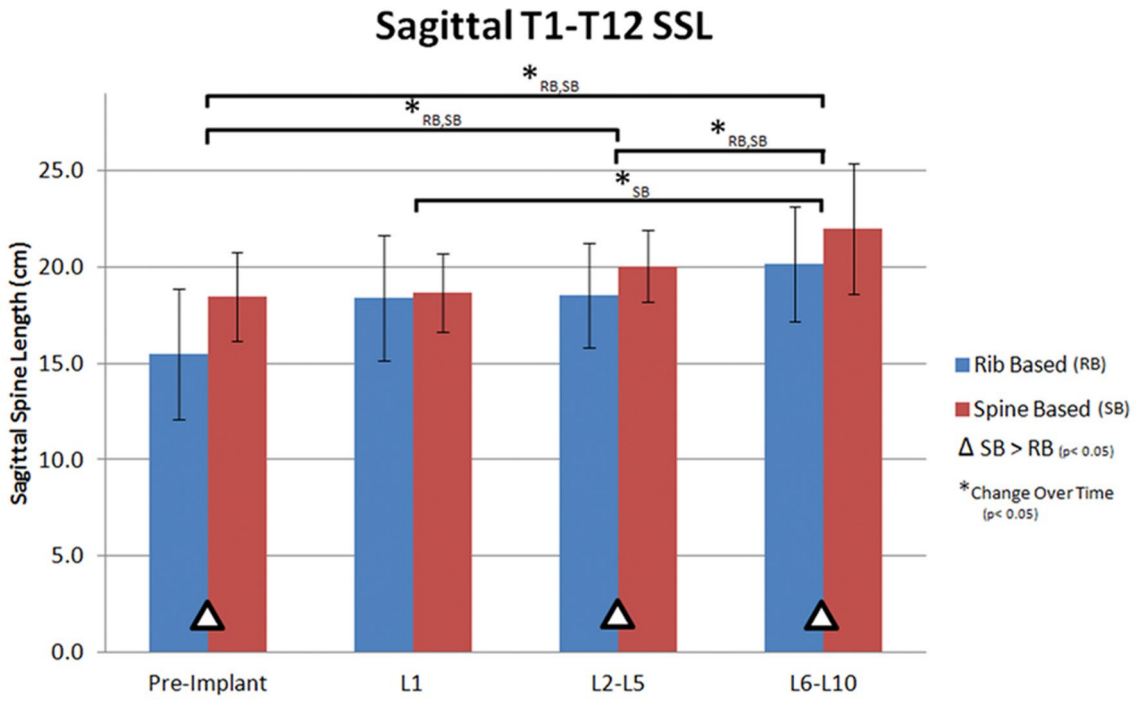


Fig. 5 Thoracic SSL for rib-based and for spine-based patients over time

phase (normalized L1 SSL to final SSL), RB treatment had a 12% increase in SSL versus an 18% increase in SSL for SB (Figs. 2, 3, and 5).

Thirty-one of the 34 patients in this study experienced at least one complication for a complication risk per patient of 91%. There were a total of 70 complications in these

patients for a total complication risk of 206%. For device-related complications, there were 41 complications classified as Smith grade SVI and 23 as SVIIA. There were not any device-related SVIIB, SVIII, or SVIV complications. The most common device-related complications were rod fracture or implant failure ($n=22$), anchor pullout or device migration ($n=13$), wound dehiscence or superficial infection ($n=9$), and proximal junctional kyphosis (PJK; $n=2$). These device-related complications resulted in a total of 57 reoperations. Disease-related complications included pneumonia ($n=2$), seizure, otitis media, metabolic, and death. These complications were classified as SVI ($n=3$), SVII ($n=2$), and SVIV ($n=1$).

Discussion

The purpose of this study was to determine if the location of proximal anchor affects thoracic SSL for children with idiopathic EOS. The Halifax method of measuring SSL, along the spine's sagittal arc of curvature, has been previously validated to reliably assess the spine's length [5]. Our current study has identified that compared with RB patients, SB patients had higher SSL preoperatively and maintained this difference to the tenth lengthening. After ten lengthening surgeries, when normalized to preoperative SSL, relative thoracic growth was greater for RB (27%) than for SB patients (19%).

Limitations of this study include that it is retrospective, multicenter in nature, and the study groups were not matched. However, as EOS is a relatively rare condition, single-center studies would unlikely have sufficient power to determine the effects of presumed growth-friendly surgeries on spine growth. As this study was performed over a 5-year period and there is a high risk of complications with growth-friendly surgery, it is not surprising that this study documented a 91% risk of complication per patient and 57 reoperations. During growth-friendly treatment, changes in spine length are related to three main factors: (1) correction of spinal deformity, (2) patient growth, and (3) biomechanical distraction of the implant. As there were 57 reoperations, it is difficult to assess the effects on spine length from further correction of spinal deformity during these reoperations. We cannot assume that the length gains measured in this study were purely secondary to patient growth and biomechanical distraction of the implant. Another limitation of this study is that the two registries do not record the rationale for the choice of implants that were used. We believe that the choice of RB versus SB anchors was mainly related to surgeon preference and that the VEPTR-based systems (DePuy Synthes Spine, Raynham, MA) were implanted as RB growing rods as opposed to a means of directly increasing thoracic volume via thoracostomy.

EOS is quite heterogeneous, but this study attempted to study the most homogeneous etiology (idiopathic) within the classification of EOS. A weakness of previous studies on SB and on RB distraction has been that the populations studied have included a variety of etiologies. It has been postulated that nonidiopathic patients with EOS may not be expected to have the same growth potential as idiopathic patients. Another strength of this study is its postoperative follow-up of 5 years, which is longer than most previous studies that have evaluated spine growth in this population.

Even when using traditional coronal vertical height to measure spine height, SB surgeries demonstrated significant increase in height up to the tenth lengthening. This is in contrast to previous publications that imply that there are negligible gains in spine height after the seventh lengthening [6]. We believe that the continued growth in our study was related to a longer follow-up period compared with previous studies (5 years minimum), studying a homogenous group of idiopathic EOS patients, and by taking into account the spine growth in the sagittal plane (SSL).

This study confirms previous findings that RB distraction surgeries are kyphogenic [7]. This kyphogenic effect may play a role in the improvement of the pulmonary function of those patients based on the correlation between loss of thoracic kyphosis and decline of pulmonary function in patients with adolescent idiopathic scoliosis [8]; however, the final kyphosis for the RB group was outside of the normal range for EOS. Potentially, this kyphogenic property of RB treatment may be useful to treat patients with preoperative hypokyphosis. This kyphogenic property may also increase the risk of developing PJK. We only identified two episodes of PJK in the databases; however, critical radiographic re-measurement of proximal junctional angle is outside the scope of this project on spine growth. Recently, the two EOS study groups published PJK results from a much larger sample of patients from the databases. In that study of 419 total patients, there was a 20% risk of developing clinically significant PJK for patients treated with RB and SB growth-friendly systems [9]. We expect that the patients in our current study would have a similar risk of developing PJK.

There has been concern that the potential for chest wall scarring with the use of RB anchors may detrimentally affect pulmonary function and may offset any improvements secondary to gains in height and length. We acknowledge that the evaluation of pulmonary function is beyond the scope of our current study. By evaluating the SSL, instead of just coronal vertical heights, any potential gains in length out of the coronal plane will be captured. For both treatment methods, we were able to see greater absolute values for SSL than for coronal plane thoracic height. At the L6–L10 interval, SB SSL was 22.0 cm versus coronal height of 20.9 cm for a difference of 1.1 cm. At the same time interval, RB SSL

was 20.2 cm versus coronal height of 16.9 cm for a difference of 3.3 cm. The greater difference between SSL and coronal height for RB patients than for SB patients can be explained by the effect of kyphosis. At L6–L10, the kyphosis for the RB group was 56° as compared with 26° for the SB group. The larger the angle of kyphosis, the greater the difference between SSL and coronal height. As it has previously been published that this effect is much more substantial for kyphosis greater than 30°, our results are in keeping with the literature [5].

Although using the SSL to account for the effects of sagittal plane deformity is an improvement compared with the traditional coronal plane vertical height measurements, it does not take into account the effect of coronal plane deformity. SB treatment corrected scoliosis by 40% compared with only 13% for RB treatment. At final follow-up, SB treatment had a mean scoliosis of 46° versus 52° for RB. For a more complete representation of spine length, future studies should also consider using a three-dimensional measurement of true spine length (3D-TSL) that takes into account both the arc of curvature in the sagittal plane and the arc of curvature in the coronal plane [10].

The SB treatment group started with greater coronal thoracic height and with greater SSL than the RB group. This may have been related to the older mean age of implant insertion for the SB treatment group (5.4 years vs. 4.2 years for RB). These differences continued through to the tenth lengthening. It was observed that the patients treated with RB fixation continued to have length gains beyond the tenth lengthening procedure. The true reason for this is unknown; however, one hypothesis is related to the law of diminishing returns that has previously been observed for SB anchors, but has not been observed for RB anchors [6, 7]. SB anchor systems are more rigid than RB systems as the upper foundation is fused with the SB systems, whereas the upper foundation is more mobile with RB systems. The increased rigidity may predispose to facet autofusion for SB systems. Despite that difference in absolute height and length, when the SSL was normalized as a percentage increase over preimplantation values, the RB treatment group had a greater percentage increase over time (27% increase vs. 19% increase by the tenth lengthening). However, the SSL change from the implantation procedure was statistically different between the two groups (2.9 cm in RB vs. 0.3 cm in SB). Taking this into account, when evaluating only the growth phase of treatment, normalized increases in SSL were 12% for RB versus 18% for SB.

Both treatment modalities were effective in maintaining gains in height and SSL over time; however, these gains may be at the expense of the well-documented complications of growth-friendly surgery. Regardless of proximal anchor choice, thoracic length continued to increase during the distraction phase of treatment for idiopathic EOS.

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Compliance with ethical standards

Conflict of interest REH (personal fees from DePuy Synthes Spine, Medtronic Spine, and Apifix Ltd; grants from DePuy Synthes Spine and Medtronic Spine; other from Children's Spine Foundation, Pediatric Orthopedic Society of North America, and Scoliosis Research Society; personal fees from Wishbone Medical Inc., outside the submitted work), CKC (none), LEG (none), AJS (none), TSH (none), AMM (none), YEB (none), CEJ (reports other from Medtronics and Elsevier, outside the submitted work), Children's Spine Study Group (grants from DePuy Synthes Spine, grants from NuVasive, outside the submitted work).

IRB approval IWK Health Center Research Ethics Board, file no. 1002256.

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